Screening for glucose intolerance and development of a lifestyle education programme for prevention of Type 2 diabetes in a population with intellectual disabilities

NIHR Programme Grant for Applied Research: RP-PG-1209-10057

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Total word count: 69,490 (for main body of report)

Competing Interests

Dr Alison Dunkley – none declared.

Freya Tyrer – none declared.

Rebecca Spong – none declared.

Dr Laura Gray – none declared.

Mike Gillett – has undertaken consultancy work for NHS England and Public Health England, for the National Diabetes Prevention Programme.

Dr Yvonne Doherty – none declared.

Lorraine Martin-Stacey – none declared.

Naina Patel – none declared.

Dr Thomas Yates – has been a member of the National Institute for Health and Clinical Excellence public health guidance on preventing type 2 diabetes.

Professor Sabyasachi Bhaumik – has been a member of the Health Services and Delivery Research (researcher-led) panel for the last three years and before that he was a member of the Community and Psychological Therapies panel of the NIHR for three years. He is the Chair of the Diaspora Committee of the Royal College of Psychiatrists and has been the Chair of the Faculty of Psychiatry of Learning Disability for four years in the past. He is also a co-editor of the only prescribing guidelines in intellectual disability nationally and the 3rd edition of this book “Frith Prescribing Guidelines for People with Intellectual Disability” has been published this year by Wiley.

Thomas Chalk – none declared.

Yogini Chudasama – none declared.

Dr Chloe Thomas – has undertaken consultancy work for NHS England and Public Health England, for the National Diabetes Prevention Programme.

Susannah Sadler – has undertaken consultancy work for NHS England and Public Health England, for the National Diabetes Prevention Programme.

Professor Sally-Ann Cooper – has received grants from the NIHR during the conduct of the study, and grants from the NIHR and from the Scottish Government outside of the submitted work.

Dr Satheesh Kumar Gangadharan – none declared.

Professor Melanie Davies - is a member of the National Institute for Health and Clinical Excellence public health guidance on preventing type 2 diabetes and both are advisers to the UK Department of Health for the NHS Health Checks Programme. She has acted as consultant, advisory board member and speaker for Novo Nordisk, Sanofi-Aventis, Lilly, Merck Sharp & Dohme, Boehringer Ingelheim, AstraZeneca and Janssen and as a speaker for Mitsubishi Tanabe Pharma Corporation. She has received grants in support of investigator and investigator-initiated trials from Novo Nordisk, Sanofi-Aventis and Lilly. She has received grants and support from the NIHR during the conduct of this study.

Professor Kamlesh Khunti (Chair) - is a member of the National Institute for Health and Clinical Excellence public health guidance on preventing type 2 diabetes and adviser to the UK Department of Health for the NHS Health Checks Programme. He has acted as a consultant, served on advisory boards for and speaker for Novartis, Novo Nordisk, Sanofi-Aventis, Lilly, Janssen, Boehringer Ingelheim and Merck Sharp & Dohme. He has received grants in support of investigator and investigator-initiated trials from Novartis, Novo Nordisk, Sanofi-Aventis, Lilly, Roche, Boehringer

Ingelheim and Merck Sharp & Dohme. He has also received grants and support from the NIHR during the conduct of this study.

Keywords:

Intellectual Disabilities	Type 2 Diabetes	Prevention
Impaired Glucose Regulation	Impaired Glucose Tolerance	Screening
High risk of diabetes	Structured Education	Diet
Lifestyle Intervention	Physical Activity	

ABSTRACT

Background

People with intellectual disability (ID) are believed to have higher prevalence of type 2 diabetes (T2DM) and cardiovascular disease (CVD). There is limited research on prevalence and prevention in this population.

Objectives

The objectives for this programme of work were to:

- establish a programme of research conducted in a population with ID that significantly enhances the knowledge and understanding of impaired glucose regulation (IGR) and T2DM in people with ID;
- test strategies for early identification of IGR and T2DM in people with ID;
- develop a lifestyle education programme and educator training protocol to promote behaviour change in a population with ID and IGR (or high risk of T2DM/CVD).

Setting

Leicestershire, UK.

Participants

Adults with ID were recruited from community settings, including residential homes and family homes. Adults with mild to moderate ID with elevated body mass index (BMI ≥25) and/or IGR were invited to take part in the education programme.

Main outcome measures

The primary outcome for the screening programme was prevalence of screen-detected T2DM and IGR. Uptake, feasibility and acceptability of the intervention were assessed.

Data sources

Participants were recruited from general practices, specialist ID services, clinics and through direct contact.

Results

A total of 930 people with ID were recruited to the screening programme: 58% were male, 80% white and 68% overweight or obese. Mean age was 43.3 years (SD 14.2). Bloods were obtained for 675 participants (73%). Prevalence of previously undiagnosed T2DM was 1.3% (95% CI 0.5 to 2%) and IGR was 5% (95% CI 4% to 7%). Abnormal IGR was more common in those of non-white ethnicity, with a first degree family history of diabetes, with increasing weight, waist circumference, BMI, diastolic BP, triglycerides, and lower high density lipoprotein cholesterol.

We developed a lifestyle educational programme for people with ID, informed by findings from qualitative stakeholder interviews (healthcare professionals, n=14; people with ID, n=7) and evidence reviews. Subsequently, 11 people with ID (and carers) participated in pilot education sessions (2 groups) and 5 people attended education for the feasibility stage (1 group). We found it was feasible to collect primary outcome measures physical activity and sedentary behaviour using wrist-worn accelerometers. We found the programme was relatively costly, meaning that large changes in activity or diet (or a reduction in programme costs) were necessary for the programme to be cost effective. We also developed a quality development process for assessing intervention fidelity.

Limitations

We were only able to screen around 30% of the population and only involved a small number in the piloting and feasibility work.

Conclusions

Results from this programme of work have significantly enhanced existing knowledge and understanding of T2DM and IGR in people with ID. We have developed a lifestyle education programme and educator training protocol to promote behaviour change in this population.

Future work: Further work is needed to evaluate the STOP Diabetes intervention to identify cost-effective strategies for its implementation.

Funding: This work was funded through the NIHR Programme for Applied Research scheme. See information on individual contributors for further information.
Word Count: 500

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LIST OF ABBREVIATIONS

ABC	Aberrant Behaviour Checklist
BMI	Body mass index
BP	Blood pressure
CCG	Clinical Commissioning Groups
CI	Confidence intervals
CVD	Cardiovascular disease
DINE	Dietary Instrument for Nutrition Education
DESMOND	Diabetes Education and Self Management for Ongoing and New Diagnosed
DPP-IV	Dipeptidyl peptidase 4 inhibitor
EPIC	European Prospective Investigation into Cancer and Nutrition
EQ-5D	EuroQol-5 Dimensions
GDS	Glasgow Depression Scale
GP	General practitioner
HbA1c	Glycated haemoglobin
HDL	High density lipoprotein
HSE	Health survey for England
HTA	Health Technology Assessment
ICER	Incremental cost-effectiveness ratio
ID	Intellectual disability
IFG	Impaired fasting glucose
IGR	Impaired glucose regulation
IGT	Impaired glucose tolerance
IQ	Intelligence quotient
IPAQ	International Physical Activity Questionnaire
LDL	Low density lipoprotein
LLDR	Leicestershire Learning Disability Register
MeSH	Medical subject heading
MVPA	Moderate to vigorous physical activity
NHS	National Health Service
NICE	National Institute for Health and Care Excellence

OGTT	Oral Glucose Tolerance Test
PAS-ADD	Psychiatric Assessment Schedules for Adults with Developmental Disabilities
PSSRU	Personal Social Services Research Unit
QALY	Quality-adjusted life year
SE	Standard error
SMR	Standardised mortality rates
SPHR	School for Public Health Research
T1DM	Type 1 diabetes mellitus
T2DM	Type 2 diabetes mellitus
UKPDS	UK Prospective Diabetes Study
WHO	World Health Organisation

SCIENTIFIC SUMMARY

Background

Type 2 diabetes (T2DM) is a serious chronic condition that is associated with an increased risk of cardiovascular disease and affects approximately 6% of the UK adult population. Impaired glucose regulation (IGR), whereby blood glucose is elevated above the normal range, is a pre-cursor to T2DM and affects approximately 12% of the UK adult population. T2DM can be prevented through changes to lifestyle, and lifestyle education interventions have been shown to be cost-effective in delaying or preventing the transition to T2DM in people with IGR in the general population.

Intellectual disability (ID), also known as learning disability, is a life-long condition with onset before adulthood, characterised by a reduced ability to understand new or complex information, to learn new skills and a reduced ability to cope independently. People with ID are believed to have higher prevalence of T2DM owing to increased prevalence of a number of risk factors, including obesity and lack of exercise. However, there has been very little research in this area, and the evidence base for detection and prevention of T2DM has not yet been applied in a population with ID.

The focus of this research programme was to conduct a diabetes screening programme among people with ID and to develop a lifestyle multi-component education programme for the prevention of T2DM and cardiovascular disease (CVD), suitable for use in this population.

Objectives

The objectives of the programme were to:

- establish a programme of research conducted in a population with ID that significantly enhances the knowledge and understanding of IGR and T2DM in people with ID;
- test strategies for early identification of IGR and T2DM in people with ID;
- develop a lifestyle education programme and educator training protocol to promote behaviour change in a population with ID and IGR (or high risk of T2DM/CVD based on elevated body mass index (BMI)).

To achieve these objectives, three distinct work packages (WP) were developed.

WP1: development and assessment of the feasibility of a structured screening programme to determine the prevalence and demographic risk factors for T2DM and IGR in people with ID. This work package also included the validation of the Leicester Self-Assessment diabetes risk score in people with ID, cost-effectiveness analysis and establishment of data linkage mechanisms.
WP2: development of a lifestyle education programme for people with ID and IGR (or high risk of T2DM/CVD based on elevated BMI).
WP3: development of an intervention fidelity process for the assessment of educators delivering the intervention.

Service user involvement

Service users were integral to the research programme. People with ID helped to promote the programme, develop study documentation and research processes, recruit and train staff, test procedures and to disseminate the findings.

Methods

WP1: Screening programme

We recruited adults with ID from community settings, including residential homes and family homes. Potential participants were approached through general practices, specialist ID services (using the Leicestershire Learning Disability Register),

specialist ID clinics and through direct contact with the research team. We collected information on demographics, medical and family history, depression, behaviour problems, lifestyle factors and activity levels. We also collected biomedical measures (plasma glucose, glycated haemoglobin (HbA1c), lipids, urea and electrolytes, liver function tests, thyroid function, albumin), anthropometric measures (height, weight, BMI, waist and hip circumference) and blood pressure.

WP1: Physical activity sub-study

Adults who consented to take part in the screening programme and who were able to walk unassisted were asked if they would be willing to wear the ActiGraph waist-worn accelerometer or GENEActiv wrist-worn accelerometer to assess physical activity and sedentary behaviour.

WP1: Validation of the Leicester Self-Assessment risk score

The Leicester Self-Assessment risk score for detecting those at risk of undiagnosed IGR/T2DM was validated using the data from the screening programme. Sensitivity, specificity, positive predictive value and negative predictive value were calculated with 95% confidence interval (CI) for a cut point of greater than or equal to 16 points.

WP1: Cost-effectiveness study

Economic work was undertaken to estimate the cost-effectiveness of the STOP Diabetes lifestyle education programme (see WP2) compared with current routine care in reducing cardio-metabolic co-morbidities among individuals with ID.

WP2: Lifestyle education programme

Adults with mild to moderate ID with BMI ≥25 and/or IGR were invited to take part in the STOP Diabetes lifestyle education programme. This involved initial intervention and curriculum development, two cycles of testing and evaluation and a final

refinement of the programme, and included interviews with adults with ID, carers and health professionals.

Feasibility was assessed by collecting primary outcomes physical activity and sedentary behaviour, and secondary outcomes weight, height, BMI, waist circumference, blood pressure and dietary intake before delivering the education programme and three months after delivering the education programme.

WP3: Intervention fidelity

We conducted preliminary work towards developing an intervention fidelity process and tool specifically tailored to people with ID.

Results

WP1: Screening programme

In total, 930 (29% of those originally approached) took part in the screening programme. Their mean age was 43.3 years. Fifty-eight per cent were men, 80% were white and most were overweight (31%) or obese (37%). Anthropometric measures were available for at least 86% of participants. Bloods were available for 675 participants (73%) to assess the prevalence of IGR/T2DM.

The overall prevalence of screen-detected (undiagnosed) T2DM was 1.3% (95% CI 0.5 to 2%) and IGR was 5% (95% CI 4% to 7%) among people with ID. Abnormal glucose regulation was almost four times more common in those from non-white ethnic groups (OR=3.93; 95% CI 2.10 to 7.33) and more than three times more common among those with first degree history of diabetes (OR=3.35; 95% CI 1.64 to 6.86). Similarly, increasing weight, waist circumference, BMI, diastolic blood pressure, triglycerides and decreasing high density lipoprotein cholesterol, were associated with an increased risk of abnormal glucose regulation.

WP1: Physical activity sub-study

Of 203 people approached, 97 (48%) agreed to wear the waist-worn accelerometer. Valid data (≥8 hours/day for 3 days) were obtained for 55 participants (57%). Similarly, of 76 people approached, 47 (62%) agreed to wear the wrist-worn accelerometer. Valid data were obtained for 39 of these participants (83%). Thus, compliance could be improved by wearing wrist-worn accelerometers.

WP1: Validation of the Leicester Self-Assessment risk score

Of 88% of adults with data available, 82% of people with abnormal glucose regulation were correctly identified as being at high or very high risk (sensitivity). Ninety-eight per cent of participants with low/medium risk scores were correctly identified as being at low risk.

WP1: Cost-effectiveness

Findings from the health economic component of the analysis showed that, in its current form, the STOP Diabetes education programme we developed in WP2 would not be cost-effective at £20,000 cost per quality-adjusted life year (QALY) threshold. However, there were scenarios in which the intervention may be effective if commissioners/payers were willing to fund the intervention up to a threshold of £30,000 per QALY.

WP2: Lifestyle education programme

Interviews carried out at initial curriculum development revealed that people with ID liked to use visual aids to help them to learn. Health professionals also highlighted the importance of allowing for the diverse ability levels of people with ID, such as different attention span and ability levels. Important considerations included the need to use recall and repetition to support learning, ensuring familiarity and consistency and allowing generalised behaviour change goals to allow for different levels of physical ability. For the testing cycles, we found that learning was facilitated by the

group dynamic, recapping main messages, using concrete examples and walking exercises. However, conceptual exercises, abstract examples and giving too many messages did not work so well.

Preliminary findings suggest that it was both acceptable and feasible to collect outcome measures, including physical activity and sedentary behaviour, at baseline and 3-months post intervention delivery for this study. In this small sample (n=5), all anthropometric outcome measures, 80% of blood pressure and 60% (3 out of 4 who agreed at baseline) of accelerometer data were available at three months follow up.

WP3: Intervention fidelity

We completed the first step in developing a tool for assessing intervention fidelity of the education programme. Preliminary findings suggest some variance between educators. The new tool involved focusing on educators' teaching at the group's pace and avoiding abstract concepts, abbreviations and jargon and engaging the learners without asking them to summarise key messages.

Conclusions

This programme of work has significantly enhanced existing knowledge and understanding of T2DM and IGR in people with ID. It has also allowed us to test strategies for early identification of IGR and T2DM in this population. Further work is needed to evaluate the intervention we have developed and to identify cost-effective strategies for its implementation.

Word Count: 1422

PLAIN ENGLISH SUMMARY

Adults with intellectual disabilities (ID) have more health problems than the general population. They are less likely to access help, and more likely to be overweight and not get enough exercise. This may increase their chance of getting diabetes.

Type 2 diabetes (T2DM) is a long-term condition, which can cause damage to blood vessels and nerves. Impaired glucose regulation (IGR) happens when blood sugar levels are higher than normal but not high enough to be T2DM. People with IGR are more likely to develop T2DM, heart disease and strokes, but can make changes to their lifestyle to prevent this.

Our research aimed to:
1) Screen people with ID for T2DM and IGR.
2) Develop a lifestyle education programme to help people with ID stay healthy.

We recruited 930 people, and collected blood samples from 675 to test for diabetes. We found about 1 in 100 people had undiagnosed T2DM and 5 in 100 had IGR. More than two-thirds (68%) were overweight or obese.

We developed a lifestyle education programme. We asked a few small groups of people with ID (and carers) to come to the 8 week programme. Attendance at the education sessions was good. Overall, people felt positive about the education.

To conclude, less people had T2DM or IGR than we expected. However, we found that many people with ID were overweight or obese. We succeeded in developing a lifestyle education programme to help people do more physical activity, eat healthier and lose weight.

Word Count: 245

EASY READ SUMMARY

We want to tell you about the STOP Diabetes Research Study.

A research study is a way we try to find out about the answers to questions.

Our research study was about diabetes.

We want to tell you what we found out.

What is diabetes?

Diabetes is an illness.

People with diabetes have too much sugar (glucose) in their blood.

Their body cannot use sugar properly.

People with diabetes may feel:

- Tired and ill

- Thirsty

- And need to go to the toilet a lot.

Why did we do this research study?

We want people with learning disabilities to be healthy.

We wanted to know if people with learning disabilities have diabetes.

We wanted to know if people with learning disabilities could get diabetes in the future.

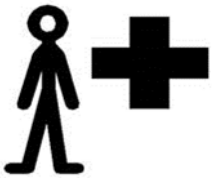
We wanted to know the best way to stop (prevent) diabetes.

Who did the research study?

Professor Khunti

Professor Bhaumik

And a research study team to help them.



- Nurse

- Secretary

- Researcher

How did we do the research study?

We asked people with learning disabilities and their carers about their health.

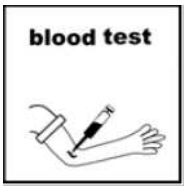
We checked:

- How tall they were.

- How much they weighed.

- What was their blood pressure.

We tested their urine (wee).



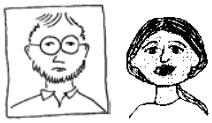
We tested their blood.

We invited some people and their carers to be part of a small group.

This group learned about staying healthy.

What did we find out?

More than 900 people with learning disabilities took part in our research study.



- Slightly more men than women took part.

- 9 people had diabetes.

- 35 people had too much sugar in their blood.

The good news is that not many people had diabetes!

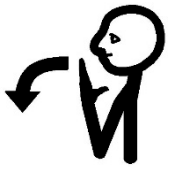
But

We found a lot of people weighed too much.
529 people weighed too much

And others were not very active.

Some of these people learned about staying healthy.

We want to teach more people about staying healthy in the future.



We want to thank everyone who has helped us!

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CHAPTER 1. INTRODUCTION

1.1 Rationale

The focus of this research programme was to estimate the prevalence of type 2 diabetes (T2DM) and impaired glucose regulation (IGR), among people with intellectual disabilities (ID), and to develop and test a lifestyle education programme for the prevention of T2DM, suitable for use in this population.

This research programme was developed to address gaps in the evidence base with regard to determining the prevalence of T2DM and IGR in adults with ID, and lack of suitable prevention programmes specially tailored for people with ID. Since beginning this research, priorities set out in the 2015-2016 National Health Service (NHS) England Business Plan[1] have highlighted the need to improve services for people with ID and to establish a national ID Mortality Review, with both diabetes and obesity identified as health priorities.[2] An additional health priority identified for all patients is the prevention of obesity and T2DM via a national “evidence-based lifestyle management programme”, to support people to make healthy lifestyle changes.[1]

The current evidence-base for screening and successfully managing those at risk of diabetes through diet, exercise and behaviour therapy relates to the general population. It is not currently known whether screening for T2DM and IGR or prevention strategies through lifestyle education can be successful in people with ID.

1.2 Aims and objectives

The aims of the programme were to:

- establish a programme of research conducted in a population with ID that significantly enhances the knowledge and understanding of IGR and T2DM in people with ID;
- test strategies for early identification of IGR and T2DM in people with ID;

- develop a lifestyle education programme and educator training protocol to promote behaviour change in a population with ID and IGR (or high risk of T2DM/CVD based on elevated body mass index (BMI)).

1.3 Overview of the programme of research

To achieve these aims, three distinct work packages were developed (*Figure 1*).

Work package 1 (WP1):

- to develop and assess the feasibility of a diabetes screening programme in a community setting for adults with ID (Chapters 5 and 6);
- to determine the prevalence and demographic risk factors for T2DM and IGR in people with mild to profound ID (Chapters 5 and 6);
- to validate the Leicester Self-Assessment diabetes risk score in people with ID (Chapters 5 and 6);
- to determine the cost-effectiveness of lifestyle intervention (see WP2) compared to current care (Chapter 12);
- to establish data linkage to Hospital Episode Statistics and the Office for National Statistics (Chapters 5 and 6).

Work package 2 (WP2):

- to develop a lifestyle education programme for people with ID and IGR (or high risk of T2DM/CVD based on elevated BMI), (Chapters 8 and 9);
- to assess the feasibility of collecting outcome measures before and 3-months after attendance at lifestyle education (Chapter 10).

Work package 3 (WP3):

- to develop a quality assurance ("intervention fidelity") process for the assessment of educators delivering the education (Chapter 11).

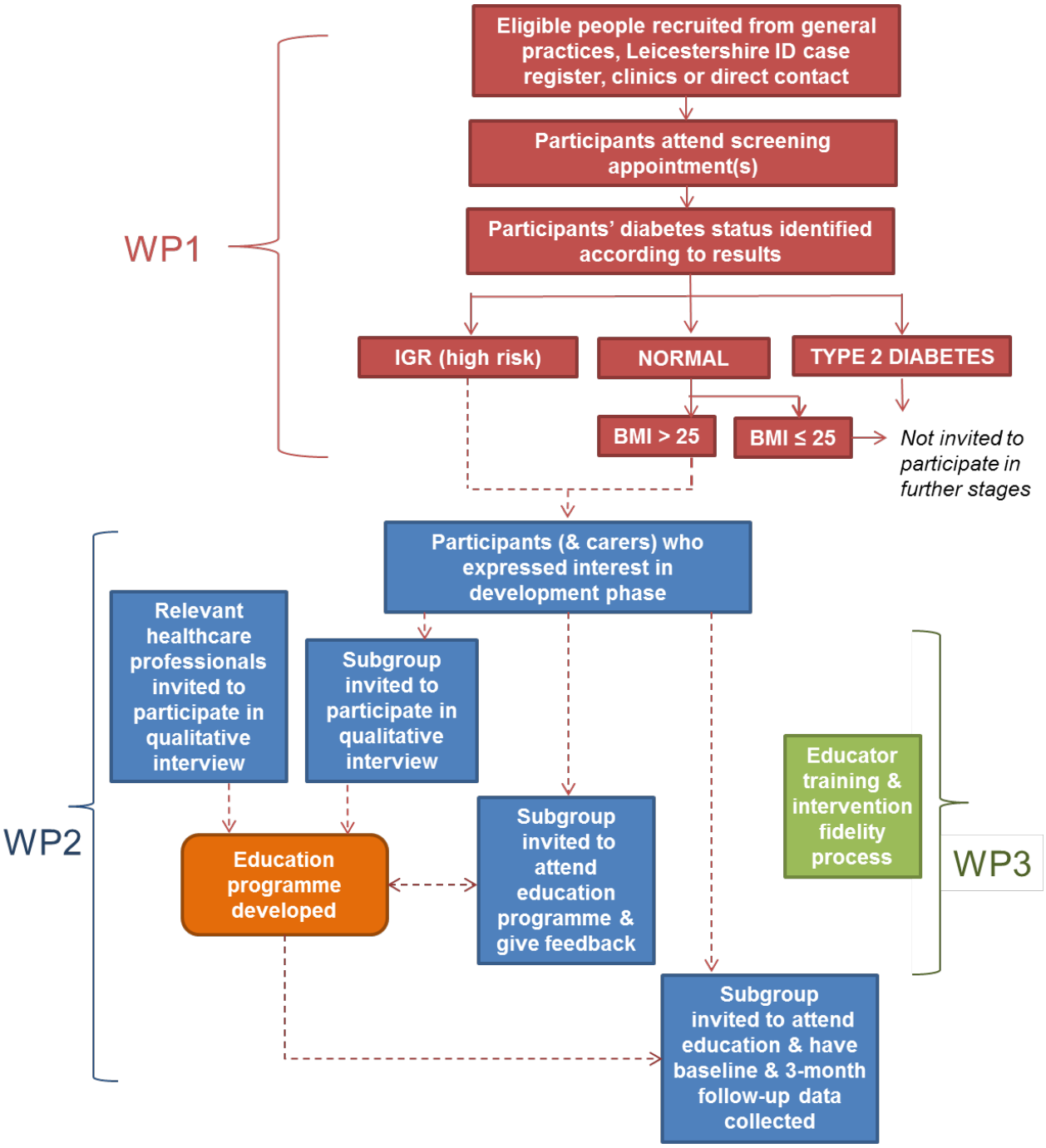


Figure 1: Flow chart programme of work

1.4 Scope of the report

The remainder of this chapter provides a brief overview of the ethics and governance arrangements, and provides the detailed background for this research programme.

Subsequent chapters contain individual summaries, but briefly comprise:

- a systematic review of prevalence/incidence of T2DM in people with ID (Chapter 2);
- a systematic review of multi-component behaviour change interventions in people with ID (Chapter 3);
- details of the involvement of people with ID throughout the programme of research (Chapter 4);
- methods for the screening programme (Chapter 5);
- results from the screening programme (Chapter 6);
- methods and results from a physical activity sub-study (Chapter 7);
- details of the development of the lifestyle education programme (Chapters 8 and 9);
- methods and findings from a feasibility phase collecting pre and post intervention outcome measures (Chapter 10);
- details of the development of the intervention fidelity process (Chapter 11);
- methods and results for the economic analysis undertaken (Chapter 12);
- discussion of findings and conclusions (Chapter 13).

1.5 Ethics and governance

1.5.1 Approvals

The University of Leicester acted as sponsor for the programme of research. NHS research ethics approval was obtained from the East of England - Cambridge Central Research Ethics Committee (reference: 12/EE/0340). Research and development approval was obtained for the research sites from: Leicestershire Partnership NHS Trust; Leicester City Clinical Commissioning Group (CCG), East Leicestershire & Rutland CCG, and West Leicestershire CCG; and University Hospitals of Leicester NHS Trust.

1.5.2 Adherence to mental capacity legislation

Obtaining consent was the largest ethical consideration for this programme. Strict standard operating procedures needed to be established to ensure that valid consent was obtained in accordance with English capacity legislation,[3] whilst taking into account the heterogeneity in capacity of individuals. More details on assessment of capacity and taking consent are contained in the methodology section for the screening programme (see Chapter 4). This included providing people with all information relevant to making the decision on whether to participate in the research, and communicating this in a way that was appropriate to them (such as using simple language and visual aids).

The process for those who lacked capacity involved talking to a ‘consultee’, whose role was to consider the study from the participant’s perspective (see Appendix 1, *Figure 26*). Regardless of whether the person with ID had capacity to decide on participation, the research was discussed with them to help them to understand the project as far as they had the capacity to do so, and to indicate any opinion they had on participation. For example, if a person without decision-making capacity appeared even slightly anxious or reluctant to take part, this would be respected, and they would not be recruited in the study.

1.5.3 Programme steering group

Strategic oversight and direction of the research programme was provided by the programme steering group (see *Figure 2*), which comprised the chief investigator (KK), the lead researcher/project manager (AD), and co-applicants listed in the application, with ad hoc attendance from service users. The meetings were held four times per year and were independently chaired by Dr Colin Greaves, University of Exeter (see *Figure 2*). The meetings involved discussion of contractual issues, staffing, protocol and ethical amendments, public involvement (a rolling agenda item), recruitment progress, economic analysis, education development, anticipated timelines and progress against project aims.

1.5.4 Operational groups

The research team, (researchers, ID research nurses, research administrator) met frequently throughout the programme to plan individual components of the programme and to discuss progress. Details from these meetings were fed back to the steering group.

The education development team, (a multi-disciplinary team of healthcare professionals and researchers, with expertise both in the field of ID and in developing diabetes and CVD prevention programmes) met regularly to oversee and facilitate development of the lifestyle education programme (work package 3). Progress and key decisions were fed back at steering group meetings.

1.5.5 Service user groups

A number of service users were involved in the research programme, but two service user self-advocacy groups were particularly influential. The groups met regularly, facilitated by an experienced supporter, and their comments were fed back to the steering group. More information about these and other service users' involvement is detailed in the fourth chapter of this report.

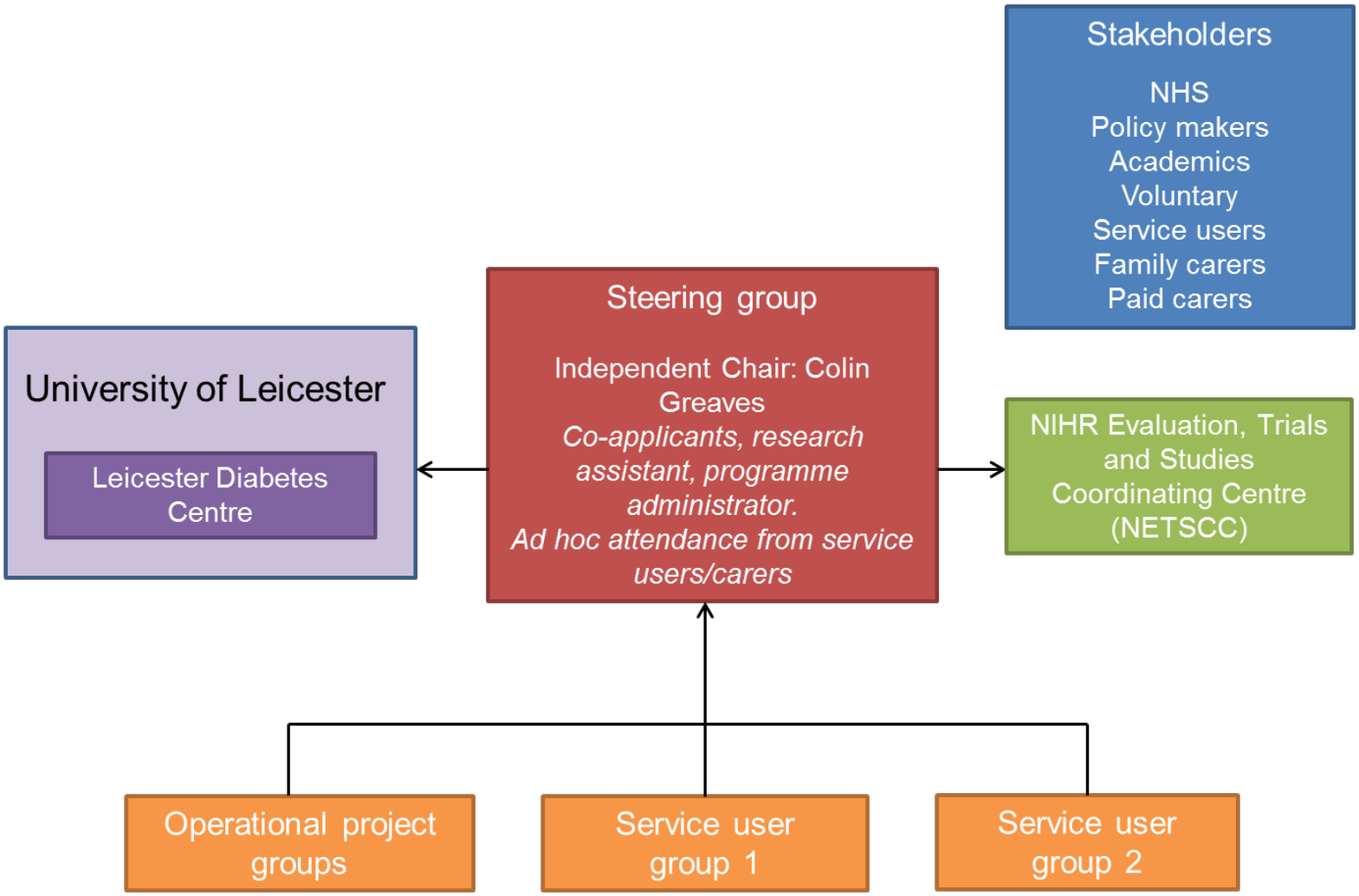


Figure 2: Governance structure of STOP Diabetes programme

1.5.6 Data protection

A six-digit study code was used to identify all study participants. This code was used for all hard and electronic copies of data collected for this programme (including questionnaires, anthropometric data and blood samples), which were retained in a secure setting.

Leicester Clinical Trials Unit (UK Clinical Research Collaboration registration number 43) were responsible for the development of a secure database for the data collected as part of this research programme.

1.6 Background

1.6.1 Definition of intellectual disability and case identification

ID, also known as learning disability, is a life-long condition with onset before adulthood, characterised by a reduced ability to understand new or complex information, to learn new skills and a reduced ability to cope independently.[4] Severity levels for ID are typically categorised by broad intelligence quotient (IQ), alongside the required deficits in independent living skills, into: mild (IQ 50-69), moderate (IQ 35-49), severe (IQ 20-34) and profound (IQ<20) ID.[5] Acknowledging the wide variation that exists between individuals with ID, typical abilities suggested for each category are outlined in *Table 1* (based on World Health Organisation (WHO) ICD-10).[5] More recently in the UK, the Learning Disabilities Public Health Observatory has offered a "working definition" which includes brief practical guidance to improve recognition of ID and assist agencies to target services.[6]

The aetiology of ID can be broadly divided into problems which occur in the antenatal, perinatal or postnatal period, or due to multiple factors. Common causes of ID include: genetic and chromosomal disorders, both non-inherited (e.g. Down's)[7] and inherited (e.g. Fragile X); and non-genetic factors such as infection and environmental factors. However, in the majority of cases no specific cause is found.[8]

A recent meta-analysis of population based studies suggests that overall around 1% of people worldwide have ID, with wide variation dependant on age-group and income of the country (lower, middle and higher); for adults the proportion is around 0.5%.[9] Evidence from existing ID registers and general practice lists in England suggests that the prevalence of ID is approximately 3–5 per 1000 individuals.[10, 11] However, it is thought that the true prevalence could be as high as 2% of the adult population, as people with mild ID are generally under-represented.[11]

For the current research programme, cases were identified via: 1) records held on adults with ID in general practices; and 2) a register of adults attending ID services

owned by the local mental health trust (Leicestershire Partnership NHS Trust), the Leicestershire Learning Disability Register (see section below).

General practices in the UK are now incentivised to maintain a register of people with ID.[12, 13] Locally, for practices within Leicester City, East Leicestershire and Rutland, and West Leicestershire CCGs, the total number of adults (aged 18 and over) on general practice registers with an identified ID is estimated to be around 4,300 (based on figures provided by Leicestershire Partnership NHS Trust).

The Leicestershire Learning Disability Register includes adults with ID (aged 19 years and over) who live in the unitary authorities of Leicester City, Leicestershire and Rutland.[14] The Register was established in 1987 to help facilitate the provision and monitoring of services, and enable the collection of public health data. It is currently a joint venture between Leicestershire Partnership NHS Trust and Leicester City CCG. Enrolment is via a large network of service providers including specialist ID services, social services and primary care. Currently, there are ~3900 people with mild to profound ID on the register. However, as the Leicestershire Learning Disability Register is based on service use, some adults, particularly those with mild ID who have little or no support from services, may not currently be identified. This potentially accounts for some of the differences between the number of people identified on this register and on local general practice registers.

Table 1: Intellectual disability categories and suggested abilities

Severity of ID	Suggested abilities and skills
Mild	• Good verbal communication, and basic reading and writing skills • Usually independent in self-care and practical domestic tasks • Often able to form/maintain good social relationships • May have employment
Moderate	• Limited language • Able to achieve some independence with support, but requirements for support will vary • Usually fully mobile
Severe	• Uses some words and gestures • Activities need to be supervised and ongoing support necessary • May have problems with movement
Profound	• Communication very limited • Support needed for all daily living activities • Mobility usually severely impaired

IQ level

1.6.2 Type 2 diabetes and impaired glucose regulation

T2DM is a serious chronic disease, characterised by prolonged hyperglycaemia.[15] Its symptoms can reduce quality of life and lead to serious health complications, including blindness, renal failure and amputation; 50% of new cases have demonstrable atherosclerosis at diagnosis.[15-17] The prevalence of diabetes in England is estimated to be 6.2%,[18] rising to 8.0% (95% CI 5.7% -11.7%) when including undiagnosed cases.[19] T2DM accounts for around 85-90% of diabetes cases; it creates a huge economic burden on NHS resources, at a cost of £8.8 billion annually (~10% of total NHS expenditure).[20]

IGR is a condition where blood glucose concentrations are elevated above the normal range but do not satisfy the criteria for T2DM.[21, 22] Approximately 12% of the UK adult population have IGR, of which an estimated 5–12% go on to develop T2DM each year. Observational studies show a consistent and continuous association between glycaemia and CVD risk whereby people with IGR have a significantly elevated risk of CVD.[23-25] Given the economic burden associated with this condition and its related co-morbidities, this group represents an important target for preventative strategies.[26] Other commonly used terms to describe IGR include pre-diabetes, non-diabetic hyperglycaemia or high risk of diabetes; throughout the report, this high risk group will be referred to as IGR.

Previously in clinical practice, T2DM and IGR were identified using the “gold standard” oral glucose tolerance test (OGTT).[22] However, since the publication of updated WHO guidance in 2011 and subsequent National Institute for Health and Care Excellence (NICE) guidance in 2012, there has been a shift away from the use of the OGTT to glycated haemoglobin (HbA1C).[27, 28] Potential benefits of HbA1c include it being a non-fasting blood test, less inter-test variability, and the ability to provide an indication of longer term hyperglycaemia (over 6-8 weeks).[29] An HbA1c of ≥48 mmol/l (6.5%) is suggestive of T2DM and 42-47 mmol/l (6.0-6.4%) of IGR or high risk.[27] Further details on the methods used to identify T2DM and IGR for this programme of research are provided in Chapter 5 (see *Section 5.7 and Figure 15*).

1.6.3 Risk factors for type 2 diabetes and cardiovascular disease in people with intellectual disabilities

In the general population, increasing levels of obesity and sedentary lifestyles have been associated with a rise in non-communicable diseases, including T2DM and cardiovascular disease (CVD).[30-33]

Chronic conditions are becoming increasingly important for people with ID as their life expectancy increases.[34] There are a number of risk factors for T2DM that are known to be highly prevalent in people with ID, suggesting that T2DM and CVD may be more prevalent in this group. These include:

- sedentary behaviour;[35-38]
- high prevalence of obesity;[32-34, 39]
- increased antipsychotic drug use for the management of challenging behaviour[40, 41] and psychosis,[42] which are associated with weight gain, hyperglycaemia and worsening of other metabolic CVD risk factors;[43-45]
- genetic conditions associated with obesity (e.g. Prada-Willi syndrome).[46]

Physical inactivity and sedentary behaviour are both common among people with ID, with only a minority (18-33%) achieving the recommended 30 minutes of moderate/vigorous physical activity daily[47, 48] and less than 15% of people with ID complete the recommended 10,000 steps per day.[49] Furthermore, less than 10% of adults with ID who live in supported accommodation have an intake of fruit and vegetables sufficient for a balanced diet.[50] Evidence suggests that paid carers know little about public health recommendations on dietary intake.[50]

However, little is known about T2DM, CVD and associated risk factors in the ID population. UK-based data on the prevalence of T2DM are currently unclear.[32] Current estimates for diabetes prevalence in the UK are based on routinely reported data, rather than population based studies. The suggested prevalence of diagnosed diabetes in people with ID in England is around 6-7% but estimates are unable to distinguish between T2DM and other forms of diabetes.[13, 51] Similarly, the prevalence

of CVD among people with ID is reported to be greater than the general population, but the overall prevalence is unclear.[52]

Further information on the current prevalence of T2DM, CVD and related risks factors in the ID population, is presented in the systematic review in the second chapter of this report.

1.6.4 Diabetes screening

Given the increasing prevalence of diabetes, and the conferred risk of developing cardiovascular disease, early identification and intervention through screening has been shown to be a useful approach in the general population.[53, 54] The value of screening for IGR has also been demonstrated.

It is currently unknown whether screening for asymptomatic glucose disorders is viable within UK populations with ID; there is a lack of evidence on feasibility, acceptability, outcomes and benefits. People with ID have been recommended by NICE as being an important group to consider in terms of diabetes prevention strategies, given their supposed high risk of developing diabetes.[27]

General practitioners (GPs) in England have been incentivised to provide annual health checks to adults with ID since 2008/09 (aged ≥14 years since 2014). Recent data suggest that nationally uptake of checks is around 44%.[55] However, the proportion who additionally have bloods taken as part of the health check, including HbA1c (7%) and cholesterol (30%), is extremely low.[13]

1.6.5 Risk scores for the early identification of impaired glucose regulation and type 2 diabetes

NICE recommend a staged approach to screening for those at risk of diabetes in the general population.[56] This involves using a risk score to pre-screen for individuals at the greatest risk of T2DM followed by a blood test in those at the highest risk. However, this approach has not been tried with populations with ID.

Risk scores are a non-invasive way of stratifying a population for targeted screening. They use information data from non-invasive risk factors to calculate an individual's score; a higher score reflects a higher risk. Risk scores can be applied to: 1) an individual as a questionnaire, (these scores generally only require data from non-invasive risk factors which would be known by members of the public); or 2) a population (for example in primary care, where software is used to calculate the score using routine data from electronic medical records) and screening invitations can then be sent to those at highest risk. A number of diabetes risk scores have been developed and validated for use in the UK general population.[56-60] One such score is the Leicester Self-Assessment Risk Score (see Appendix 2), which allows people to easily assess their own risk of having undiagnosed IGR or T2DM and then self-refer for screening with a healthcare professional.[58] The score contains seven questions, which ask about age, sex, ethnicity, body mass index (BMI), waist circumference, family history of diabetes, and high blood pressure (BP). The score has been validated for use in a multi-ethnic UK population[58, 61] and is specifically recommended for identifying people at risk opportunistically by NICE.[56]

To date, we are not aware of any risk scores that have been specifically assessed for use in ID populations. However, it cannot be assumed that a risk score developed for a specific population will work well in another;[62] for people with ID, there may be different risk factors or weightings for specific risk factors may change, when compared to the general population. Therefore, this programme of work will seek to validate the Leicester Self-Assessment Risk Score in a population with ID (this work is presented as part of the screening study, Chapters 5 and 6).

1.6.6 Diabetes prevention in adults with intellectual disabilities

People with ID experience a disproportionate burden of health inequalities compared with the general population, including poorer mental and physical health and higher rates of mortality.[63-66] Despite their increased health needs, they often find it difficult to access primary care services and participate in health promotion activities.[67-69]

Given the health inequalities among people with ID and possible increased risk of developing diabetes, people with ID have the potential to benefit from lifestyle changes (with appropriate support) that are addressed in lifestyle education programmes. However, the evidence base for diabetes prevention relates to the general adult population; literature focusing on ID is scarce. Details of the key literature on lifestyle behaviour change interventions aimed at modifying risk factors for T2DM and CVD in people with ID, are presented in the systematic review in Chapter three of this report.

Current evidence from studies conducted in the general population suggest that intensive multi-component lifestyle interventions aimed at weight loss, a healthy diet and increased physical activity can successfully reduce the risk of diabetes by 30–60% in those with IGR, and are likely to be cost-effective in the long term.[54, 70]

Increasing physical activity is fundamental to diabetes prevention initiatives as research suggests that inactivity may have more impact than increased body weight in the development of insulin resistance.[71]

NICE recommends that, for both obesity management[72] and prevention of T2DM,[27] lifestyle interventions should be multi-component, involving both dietary and physical activity advice and incorporating behaviour change techniques. However, currently there are no national prevention programmes suitable for people with ID, despite ongoing recommendations to make ‘reasonable adjustments’ to healthcare services to address inequities in provision.[73]

Education, exercise and leisure pursuits are often determined or influenced by carers (paid or family carers) who may have a range of competing time demands and a number of people to provide support for. For people with limited carer support, difficulties in understanding health risks could also influence motivation to change lifestyles. Therefore, there is the potential for this group to benefit from the development of a lifestyle education programme that is targeted at both people with ID and their carers, in order to encourage changes in lifestyle behaviours that could reduce the long-term chances of this high-risk group developing diabetes.

1.7 Concluding remarks

This chapter has provided the rationale and aims for the research programme, and an overview of the programme of work undertaken. The following chapter presents a systematic review conducted to consolidate the evidence on rates of T2DM, CVD and associated risk factors in adults with ID.

CHAPTER 2. SYSTEMATIC REVIEW AND META-ANALYSIS: RATES OF TYPE 2 DIABETES, CARDIOVASCULAR DISEASE AND ASSOCIATED RISK FACTORS IN INTELLECTUAL DISABILITY POPULATIONS

2.1 Overview

In this chapter, we describe the first of two systematic reviews carried out for the research programme. We present the existing evidence in relation to the prevalence of T2DM, CVD and associated risk factors among people with ID. We have used the PRISMA (preferred reporting items for systematic reviews and meta-analyses) checklist[74] as a guide to reporting the methods and findings from the review.

2.2 Rationale

It is recognised in the literature that ID populations may be at increased risk of developing T2DM and subsequent CVD through increased risk factors such as obesity. The global increase in the prevalence of obesity, CVD and T2DM and current discrepancies between studies focusing on prevalence of such conditions in those with ID suggested a need for a systematic review of literature in this area.

Two recent reviews have been conducted that have focused on diabetes prevalence among people with ID.[75, 76] The reviews were unable to distinguish between T2DM and other types of diabetes. Similarly, the prevalence of CVD among people with ID is reported to be greater than the general population, but the overall prevalence is unclear.[52]

The overall aim of this component of the research programme was to consolidate the evidence for current rates of T2DM, CVD, and associated risk factors, restricting to population-based studies of adults with ID. If sufficient data were available, we also intended to conduct a meta-analysis. A secondary aim was to compare these data with the general population, where possible.

2.3 Objectives

The objectives of this review were:

- to establish the prevalence of T2DM in the ID population;
- to establish the prevalence of CVD in the ID population;
- to establish the prevalence of risk factors for T2DM and/or CVD (obesity, adverse lipid profiles, IGR and hypertension) in the ID population.

2.4 Methods

2.4.1 Protocol and registration

This systematic review was registered with the international register of prospective systematic reviews (PROSPERO: CRD42015019048).[77]

2.4.2 Eligibility criteria

The review was guided by the PICOS (population, intervention, comparison, outcome, study designs) model.[78] We defined the population as adults (≥18 years) with ID (whole study population or a defined sub-sample). The items of interest were defined as T2DM, CVD and their associated risk factors. Context was defined as population-based studies. We defined the outcomes as prevalence and/or incidence rates (or data to enable this calculation). Study designs included cross-sectional, retrospective and prospective cohort studies (*Table 2*).

All studies published since 1 January 2000 (until 21 April 2015) and in English language were eligible. We contacted lead authors for further information where inclusion/exclusion could not be determined.

Table 2: Inclusion and exclusion criteria for systematic review of prevalence and risk factors

	Inclusion Criteria	Exclusion criteria
Population	Whole study population or defined sub-sample of adults (≥18 years)*	Restrictively selected cohort based on outcome (e.g. all participants obese at time of data collection)
Items of interest	T2DM / Diabetes CVD (atherosclerotic) Overweight/Obesity Hypertension Hyperlipidaemia Elevated glucose/IGR Metabolic syndrome	
Context	Population-based studies	
Outcomes	Prevalence Incidence	
Study designs	Cross-sectional Retrospective cohort Prospective cohort	

* Because the focus of the review was to obtain prevalence rates generalisable to the adult ID population as a whole, studies with >20% aged under 18 years, >20% disability other than ID and >25% with specific ID were excluded.

We chose to limit studies to those published from the year 2000 so that the current prevalence of T2DM and CVD could be estimated accurately; it is known that the prevalence of both these conditions has increased substantially in recent decades.

2.4.3 Information sources

For this review, we searched databases EMBASE, MEDLINE, and PsychINFO. The last date of the search was 21 April 2015. We also searched the reference lists of relevant articles for possible additional studies.

2.4.4 Search

We combined medical subject headings (MeSH) terms and key words for T2DM, CVD, overweight/obesity, hypertension, hyperlipidaemia, elevated glucose / impaired glucose tolerance (IGT), metabolic syndrome and ID (see *Table 3* for MEDLINE search strategy). The search was limited to English language studies with cohorts of adults ≥18 years of age, depending on the database.

Table 3: Search strategy for MEDLINE electronic database

1. Exp Diabetes Mellitus, Type 2/
2. (diabet* adj3 type adj “2”).ti,ab.
3. T2DM.ti,ab.
4. (diabet* adj3 type adj ii).ti,ab.
5. niddm.ti,ab.
6. (non-insulin-dependent adj2 diabet*).ti,ab.
7. (adult-onset adj2 diabet*).ti,ab.
8. Or/1-7
9. Exp Hypertension/
10. hypertens*.ti,ab.
11. (blood adj pressure adj3 (high or elevated or increased or raised)).ti,ab.
12. Or/9-11
13. Exp Metabolic syndrome x/
14. (metabolic adj syndrome).ti,ab.
15. (cardiometabolic adj syndrome).ti,ab.
16. (Insulin adj resistance adj syndrome).ti,ab.
17. MetSyn.ti,ab.
18. MetS.ti,ab.
19. Or/13-18
20. Exp. Hyperlipidemias/
21. Hyperlipid*.ti,ab.
22. dyslipid*.ti,ab.
23. hypercholes*.ti,ab.
24. hypertriglycer*.ti,ab.
25. (cholesterol* adj2(high or elevated or raised or increased)).ti,ab.
26. (triglcerid* adj2(high or elevated or raised or increased)).ti,ab.
27. (lipid adj profile adj2(adverse or abnormal)).ti,ab.
28. Or/20-27
29. Exp. Glucose intolerance/
30. (impaired adj glucose adj(tolerance or regulation)).ti,ab.
31. (impaired adj fasting adj glucose).ti,ab.
32. IGT.ti,ab.
33. IFG.ti,ab.
34. IGR.ti,ab.
35. Exp Prediabetic state/
36. prediabet*.ti,ab.
37. pre-diabet*.ti,ab.
38. Or/29-37
39. (cardiovascular adj diseas*).ti,ab.
40. CVD.ti,ab.
41. CHD.ti,ab.
42. Exp. Myocardial infarction/
43. (infarct* adj2 myocardial).ti,ab.
44. Exp Coronary disease/
45. (coronary adj2 diseas*).ti,ab.
46. (acute adj coronary adj syndrom*).ti,ab.

47. Exp angina pectoris/
48. angina.ti,ab.
49. Exp myocardial ischemia/
50. (isch* adj2 heart adj2 diseas*).ti,ab.
51. (Myocardial adj2 isch*).ti,ab.
52. Exp. Stroke/
53. strok*.ti,ab.
54. (cerebrovascular adj2 diseas*).ti,ab.
55. (cerebrovascular adj2 accident*).ti,ab.
56. (cerebral adj2 diseas*).ti,ab.
57. (cerebral adj2 accident*).ti,ab.
58. CVA.ti,ab.
59. TIA.ti,ab.
60. (brain adj1 infarc*).ti,ab.
61. (brainstem adj1 infarc*).ti,ab.
62. Exp ischemic attack, transient/
63. (isch* adj2 attac* adj2 transient).ti,ab.
64. Exp Atherosclerosis/
65. atheroscle*.ti,ab.
66. (arteriosclerotic adj vascular adj diseas*).ti,ab.
67. exp Peripheral Arterial Disease/ or exp Peripheral Vascular Diseases/
68. (peripheral adj2 arter* adj2 diseas*).ti,ab.
69. (peripheral adj2 vascular adj2 diseas*).ti,ab.
70. (peripheral adj1 angiopath*).ti,ab.
71. or/39-70
72. exp obesity/
73. obes*.ti,ab.
74. overweight.ti,ab.
75. (body adj weight adj2 (high or elevated or increase*)).ti,ab.
76. (bodyweight adj2 (high or elevated or increase*)).ti,ab.
77. (body adj mass adj3 (high or elevated or increase*)).ti,ab.
78. (waist adj2 (large or elevated or increas*)).ti,ab.
79. Exp body mass index
80. (BMI adj2 (high or elevated or increase*)).ti,ab.
81. or/72-80
82. exp Intellectual disability/
83. (learning adj1 disabilit*).ti,ab.
84. (developmental adj1 disabilit*).ti,ab.
85. (intellectual adj1 disabilit*).ti,ab.
86. (impair* adj2 intellectual adj2 function*).ti,ab.
87. (mental* adj1 impair*).ti,ab.
88. (mental* adj1 handicap*).ti,ab

2.4.6 Study selection

Full texts were identified after titles and abstracts were read separately by two investigators (TC and AD) who discussed discrepancies in selection at a later meeting. Only full length articles were included; review articles were removed after being examined for references. Once we had retrieved the full-text of the articles, they were examined separately (by TC and AD) to check suitability for inclusion.

2.4.7 Data collection process

We designed a data extraction form specifically for this review. Data were extracted by one investigator (TC) and verified for accuracy by another investigator (AD).

2.4.8 Data items

For each study, the first author's name, title of the paper, year of publication, country of the cohort, study type, sampling method, dates of data collection, and inclusion/exclusion criteria were extracted. We also extracted total sample size or sub-population size, mean ages, proportion of male/female, severity of ID and ethnicity. For each of the outcomes, we also extracted how they were defined, how they were measured and the total number and proportion of people for whom they were measured. We extracted data separately for males and females, where reported. When framing the research question and designing the search strategy, we did not consider physical activity/sedentary behaviour, dietary factors or smoking; however, we extracted this information for studies that reported them. We also extracted information on general population data.

2.4.9 Risk of bias in individual studies

We used funnel plots[79] and the Egger's test[80] to examine potential publication bias in the literature for the outcomes T2DM, ischaemic heart disease, obesity, hypertension and undefined CVD.

2.4.10 Summary measures

The main outcome measures for the meta-analysis were the prevalence of T2DM and CVD. Secondary outcome measures were prevalence of:

- overweight/obesity;
- hypertension;
- hyperlipidaemia;
- elevated glucose/IGT;
- metabolic syndrome.

2.4.11 Synthesis of results

Owing to the variation in reporting of outcomes, we extracted descriptions and definitions of each outcome for analytic purposes and sub-categorised for meta-analyses. We sub-categorised circulatory disease outcomes as ischaemic heart disease, cerebrovascular disease, and undefined CVD. We sub-categorised diabetes outcomes as T2DM and pooled diabetes. BMI outcomes were labelled as obese (BMI>30) and overweight (BMI 25-29.9). In some articles, overweight and above (BMI >25) was used as an outcome. We combined papers reporting both obese and overweight data to create an overweight and above outcome. Outcome definitions can be seen in Appendix 3 (*Table 64*).

Owing to the large amount of variability between studies, we used a random effects model to pool the point prevalence for each outcome. We conducted a secondary meta-analysis including data from a sub-set of 10 papers,[81-90] which additionally reported general population comparison data (from the same population and time period). We assessed heterogeneity using the I^2 test.[80]

2.4.12 Additional analysis

After the meta-analysis, meta-regression was used to determine if study characteristics could explain heterogeneity (as measured using the I^2 test). These study characteristics were severity of ID, mean age, and method of data collection

(self/carer reported, researcher collected, retrospective records/database). We conducted all analyses using Stata statistical software, version 14 (StataCorp.). Significance was set at the 5% level ($p<0.05$) and 95% confidence intervals (CIs) are presented throughout.

2.5 Results

2.5.1 Study selection

In total, we identified 4513 articles via the literature searches. After duplicates were removed, 3645 articles remained to be screened. We reviewed the full-text of 158 articles once 7 articles from other sources had been added (*Figure 3*). The authors of seven studies were contacted for information regarding their studies;[91-97] five authors replied and two studies were deemed suitable to be included in the systematic review and meta-analysis.[94, 95] We also included a study from one of the authors who did not reply after re-reading and discussing the article collectively in more depth.[93]

After review, 62 articles[50, 81-90, 93-95, 98-145] were included. Four of these articles reported findings from the same study[90, 102-104] and a further two articles reported findings from the same study,[109, 125] leaving 58 studies remaining for the final systematic review and meta-analysis.

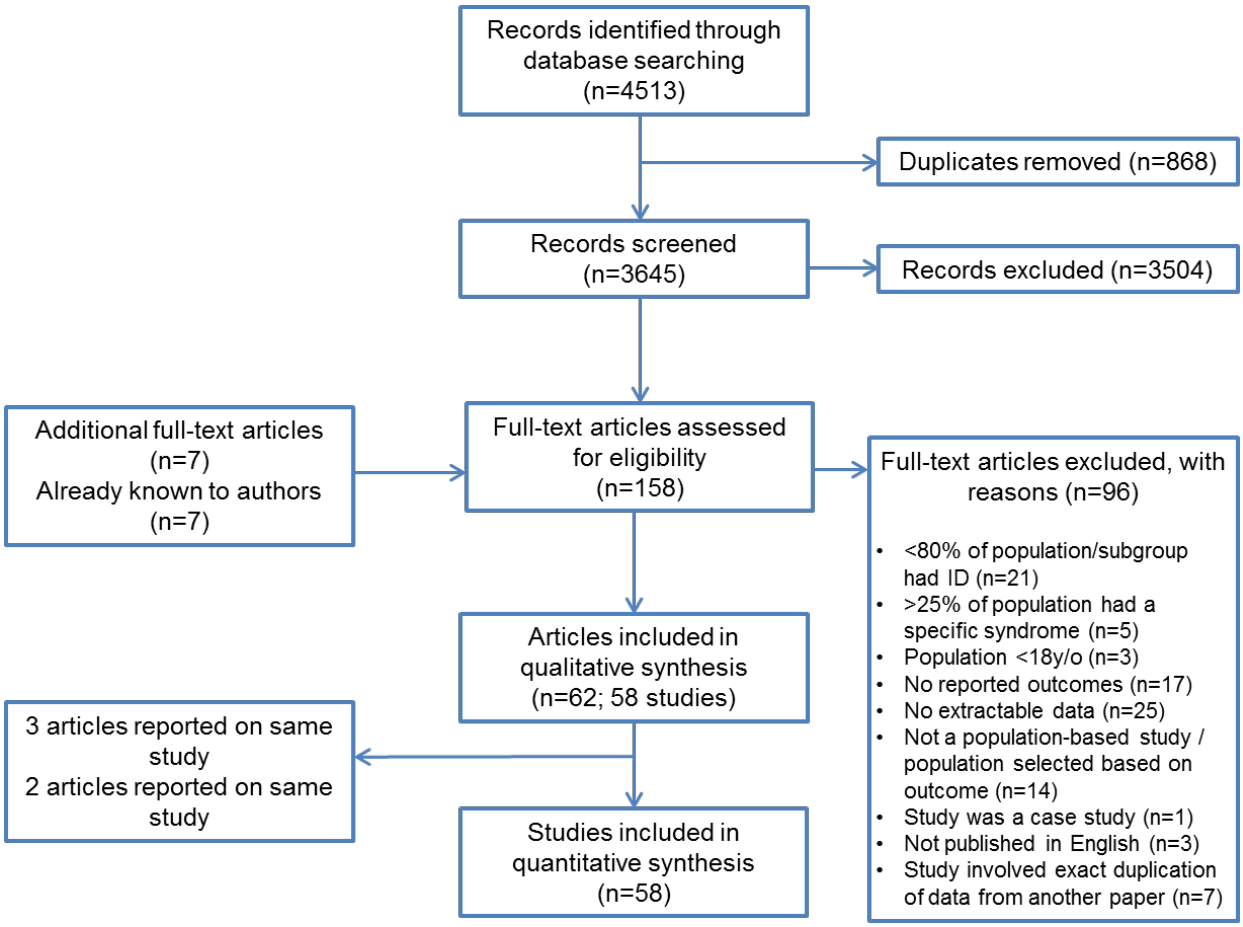


Figure 3: Flow diagram of study selection

2.5.2 Study characteristics

The 58 studies included in the quantitative synthesis presented data on more than 47,000 individuals. The characteristics of each of the studies are presented in *Table 4*.

Ten of the studies included in the systematic review also presented general population comparison data for inclusion in the secondary meta-analysis.

The studies represented 23 countries on five continents. One study covered 14 European countries. Most studies were conducted in the USA/Canada (n = 17). The

remaining studies were conducted in Europe (Netherlands (n = 7); UK (n = 9); France (n = 2); Norway (n = 1); and Ireland (n = 2)), Asia (n = 11), Australia/New Zealand (n = 6) and South Africa (n = 2). Primarily, included studies presented researcher collected data (n = 25). The remaining studies used retrospective database or medical records data (n = 19); self/carer reported questionnaire data (n = 11). Three studies used a combination of the above methods.

All studies were published in the years 2000–2015. The average mean age was 42.8 years, with an average mean range of 23.3-65.5 years old. The average mean percentage of male participants was 52.4%. The number of people included in the studies ranged from 25 to 8911, with a mean of 824.

Table 4: Studies included in the systematic review of prevalence and risk factors

Author/year	Country	ID severity	Data Source/collection method (year collected if retrospective)	Total N	Male %	Mean age	Outcomes reported	Study design
Molteno (2000)[131]	South Africa	MILD 0.3% MOD 18.7% SEV 37.7% PROF 33.5% MISSING DATA	Researcher collected data (CFS)	615	51	NR	Overweight, Obese	Cross-sectional observational
Robertson (2000)[50]	UK	NR	Questionnaire/interview (CFS)	500	60.3	44.4	Overweight, Obese	Cross-sectional observational
Janicki (2002)[114]	USA	MILD 1.3% MOD 50.3% SEV/PROF 47%	Health questionnaire data (CFS)	1373	53.0	53.5	CVD Undiagnosed diabetes Overweight, Obese Hypertension Hyperlipidaemia	Cross-sectional observational
Lewis (2002)[119]	USA	MILD 37.1% MOD 16.4% SEV 14.7% PROF 15.3%	Medical records/information from carers/physical exam by nurse (1997)	353	49.9	35.8	Overweight, Obese Hypertension Hypercholesterolaemia	Cross-sectional retrospective
Marshall (2003)[124]	UK	NR	Health check questionnaire (CFS)	728	NR	NR	Overweight, Obese Hypertension Hypercholesterolaemia	Cross-sectional observational
Havercamp (2004)[83]	USA	MILD 39.4% MOD 26.6% SEV 14.7% PROF10.6%	Health survey interview data	477	56.1	NR	CVD Undefined diabetes	Cross-sectional Retrospective

			(2001-2002)				Overweight, Obese Hypertension	
Hove (2004)[111]	Norway	MILD 39.2% MOD 42.1% SEV 15.5%	Health questionnaire data (CFS)	274	52.0	NR	Overweight, Obese	Cross-sectional observational
Merrick (2004)[129]	Israel	NR	Health questionnaire data (CFS)	2282	51	49.8	Heart disease T2DM Overweight+ Hypertension Hyperlipidaemia	Cross-sectional observational
Moore (2004)[132]	Australia	NR	Researcher collected data (CFS)	93	NR	32.5	Overweight, Obese	Cross-sectional observational
Emerson (2005)[105]	UK	NR	Audit review of the quality of supported accommodation (2000-2002)	1304	54.0	49.3	Overweight, Obese	Cross-sectional observational
Yen (2005)[144]	Taiwan	MILD 22.2% MOD 34.9% SEV 28.1% PROF 14.8%	Postal questionnaire data (2001)	516	NR	NR	Overweight, Obese	Cross-sectional retrospective
Ito (2006)[89]	Japan	NR	Care home periodic medical evaluation data (2002)	526	NR	NR	Overweight, Obese	Cross-sectional retrospective
Lennox (2006)[116]	Australia	NR	Medical history chart/GP examination (CFS)	25	NR	45.0	Overweight, Obese Hypertension	Cross-sectional observational
Levy (2006)[117]	USA	MILD 47.6% MOD 31.1% SEV 14.6% PROF 6.8%	Medical record review (NR)	103	52.4	38.2	Hypertension Hypercholesterolaemia	Cross-sectional retrospective

							Overweight, Obese Undefined diabetes	
McDermott (2006)[86]	USA	NR	Electronic medical records (1990-2003)	618	NR	NR	Ischaemic heart disease Cerebrovascular disease Hypertension Obese T1&T2DM	Cross-sectional retrospective
Rurangirwa (2006)[93]	USA	NR	Study questionnaire data (2004)	173	58.0	23.3	Overweight+	Cross-sectional retrospective
Shah (2006)[135]	UK	NR	Mail questionnaire (CFS)	119	NR	NR	Undefined diabetes	Cross-sectional observational
Van Den Akker (2006)[140]	Netherlands	MILD 11% MOD 53% SEV 28% PROF 8%	Electronic health service provider database (NR)	436	52	NR	Ischaemic heart disease Cerebrovascular disease Hypertension	Cross-sectional retrospective
Levy (2007)[118]	USA	SEV 65.4% PROF 34.6%	Medical record review/retrospective medical billing data (2006-2007)	52	52.0	NR	Overweight+ Hypercholesterolaemia Hypertension Undefined diabetes	Cross-sectional retrospective
McDermott (2007)[87]	USA	NR	Electronic medical records (1990-2003)	585	NR	NR	Undefined diabetes	Cross-sectional retrospective

McGuire (2007)[127]	Ireland	MILD 14.1% MOD 63.5% SEV 12.8% PROF 9%	Postal questionnaire (CFS)	155	53.5	37.0	Overweight, Obese	Cross-sectional observational
Wang (2007)[142]	Taiwan	NR	Health questionnaire data (CFS)	1128	57.6	NR	Heart disease Overweight+	Cross-sectional observational
Bhaumik (2008)[99]	UK	NR	Questionnaire data register (1998-2001)	1119	59.0	NR	Overweight, Obese Heart disease	Cross-sectional retrospective
Henderson (2008)[84]	USA	NR	Medical chart data (2005)	100		NR	T2DM Overweight, Obese Hypertension Dyslipidaemia	Cross-sectional Retrospective
Melville (2008)[128]	UK	MILD 40.9% MOD 25.1% SEV 18.2% PROF 15.8%	Face to face interview/physical examination by nurse (CFS)	945	55.6	NR	Overweight, Obese	Cross-sectional observational
Wallace (2008)[141]	Australia	NR	Medical chart data from GP physical examination (2002-2005)	155	52	NR	CVD Elevated glucose T1&T2DM Overweight, Obese Hypertension Hypercholesterolaemia	Cross-sectional retrospective
De Winter (2009)[81]	Netherlands	MILD 12.1% MOD 33.2% SEV 34.3% PROF 20.4%	GP screened/medical chart/structured	470	NR	NR	Myocardial infarction Cerebrovascular	Cross-sectional observational

			interview (CFS)				disease Hypertension Diabetes Elevated glucose Obese Hypercholesterolaemia	
Gale (2009)[107]	UK	NR	GP survey data collected for study (2007-2009)	1097	58.0	NR	Overweight, Obese	Cross-sectional observational
Henderson (2009)[110]	USA	MILD/MOD 53% SEV/PROF47%	Health questionnaire data (CFS)	1196	53.0	NR	Overweight+	Cross-sectional observational
Maaskant (2009)[123]	Netherlands	NR	Database data from a service care provider (2002-2007)	336	55.1	NR	Overweight, Obese	Cross-sectional retrospective
Moss (2009)[134]	South Africa	NR	Questionnaire/physical examination by nurse (CFS)	100	47	NR	Elevated glucose Overweight+ Hypertension Hypercholesterolaemia	Cross-sectional observational
Sohler (2009)[136]	USA	NR	Medical chart data (2001-2005)	5930	NR	NR	Undefined diabetes Overweight, Obese Hypertension Hypercholesterolaemia	Cross-sectional retrospective
Van de Louw	Netherlands	MILD 10% MOD 38% SEV/PROF 52%	Researcher collected data (CFS)	258	51.6	47	Hypertension	Cross-sectional observational

(2009)[139]								
Shireman (2010)[95]	USA	NR	Medical care database data (2005-2006)	291	52.6	NR	Undefined diabetes	Cross-sectional retrospective
Stedman (2010)[138]	New Zealand	NR	Service user database data collected by doctor/healthy lifestyles coordinator (6 months prior to study)	98	NR	43	Overweight, Obese	Cross-sectional observational
Tyler (2010)[88]	USA	NR	Electronic medical care database (2005-2008)	1267	53.8	38.8	Ischaemic heart disease Undiagnosed diabetes Obese Hypertension Hyperlipidaemia	Cross-sectional retrospective
Chen (2011)[101]	China	NR	Physical exam (2008)	117	NR	NR	Heart disease Diabetes Hypertension Elevated glucose Hypercholesterolaemia	Cross-sectional observational
Frighi (2011)[106]	UK	MILD 48% MOD 30.2% SEV/PROF 21.8%	Care home visitation questionnaire data (CFS)	202	52.0	42.1	Overweight+ T2DM	Cross-sectional observational
Haveman (2011)	14 European	MILD 22.7% MOD 28.2% SEV 20.7% PROF 11.8%	Interview survey data (CFS)	1253	51.0	41.0	Undefined diabetes Hypertension	Cross-sectional observational

POMONA II study[109]	countries						Myocardial infarction Cerebrovascular disease	
Lee (2011)[115]	Australia	MILD 33% MOD 22% SEV 23% PROF 21%	ID database with medical data (2006-2011)	162	52.0	44.0	Ischaemic Heart Disease Overweight, Obese Undefined diabetes Hypertension	Cross-sectional retrospective
Martinez-Leal (2011) POMONA II study[125]	14 European countries	MILD 21.8% MOD 27.7% SEV 19.7% PROF 11.4%	Interview survey data (CFS)	1257	50.5	41.4	Overweight, Obese	Cross-sectional observational
Stancliffe (2011)[137]	USA	NR	Consumer survey interview (2008-2009)	8911	NR	43.5	**Overweight, Obese Overweight+**	Cross-sectional retrospective
Wong (2011)[143]	Hong Kong	MILD 4.9% MOD 41.8% SEV/PROF 51.9%	Survey questionnaire delivered by health professional (CFS)	811	53.3	44	Heart disease Cerebrovascular disease Undefined diabetes Overweight+ Hypertension Hypercholesterolaemia	Cross-sectional observational
Chang (2012)[100]	Taiwan	MILD 65% MOD 16% SEV 9% PROF 10%	Annual health check database (NR)	129	56.6	33.0	Overweight, Obese Heart disease Elevated glucose	Cross-sectional observational

							Hypercholesterolaemia Metabolic syndrome	
De Winter (2012)_1 HA-ID study[103]	Netherlands	MILD 24.8% MOD 48% SEV 16% PROF 8.9%	Medical records/Physical examination (CFS)	945	51.0	61.5	Overweight, Obese	Cross-sectional observational
De Winter (2012)_2 HA-ID study[102]	Netherlands	MILD 24.5% MOD 48.6% SEV 16% PROF 8.7%	Medical records/Physical examination (CFS)	980	51.3	61.5	Hypertension Hypercholesterolaemia Metabolic syndrome Diabetes	Cross-sectional observational
Gazizova (2012)[82]	UK	MILD 61% MOD 24% SEV 15%	Routine health assessment of people within a service (2009)	100	67.0	NR	Overweight, Obese	Cross-sectional observational
Hsu (2012)[113]	Taiwan	MILD/MOD 47% SEV/PROF 53%	Health examination charts (2009)	164	NR	33.0	Overweight+ Metabolic syndrome	Cross-sectional retrospective
Lin, L.P. (2012)[122]	Taiwan	NR	Annual health examination chart (2010)	184	62.5	NR	Hypertension	Cross-sectional retrospective
Morin (2012)[133]	Canada	MILD 32.9% MOD 46.4% SEV 11.2% PROF 5.2%	Mail questionnaire data (CFS)	789	NR	NR	Heart disease Undefined diabetes	Cross-sectional observational
Begarie	France	NR	Questionnaire data	255	NR	NR	Overweight, Obese	Cross-sectional

(2013)[98]			(CFS)					observational
De Winter (2013)[104] **HA-ID study**	Netherlands	MILD 24.9% MOD 53% SEV 13.4% PROF 4.6%	Medical records/Physical examination (CFS)	629	53.6	61.5	Peripheral arterial disease	Cross-sectional observational
Haider (2013)[108]	Australia	NR	Telephone questionnaire (2008-2009)	897	NR	38.4	Heart disease Cerebrovascular disease T2DM Overweight, Obese	Cross-sectional retrospective
Jansen (2013)[85]	Netherlands	MILD 6.9% MOD 37.8% SEV 29% PROF 26.3%	Medical file data (2007)	510	55.7	65.5	Myocardial infarction Cerebrovascular disease	Cross-sectional retrospective
Lin, J.D. (2013)[120]	Taiwan	NR	Annual health examination chart (2010-2012)	215	NR	NR	Hypercholesterolaemia Hypertension Elevated glucose	Cross-sectional retrospective
McCarron (2013)[126]	Ireland	NR	Face to face questionnaire – first wave data for a longitudinal study (CFS)	753	45.0	54.8	Ischaemic heart disease Cerebrovascular disease Hypertension	Cross-sectional observational
Vacek (2013)[94]	USA	NR	Medical care database data (2006-2007)	3079	NR	NR	Hypertension	Cross-sectional retrospective

Hsieh (2014)[112]	USA	MILD 44.9% MOD 23.7% SEV/PROF 8.4%	Longitudinal study baseline data (2012) questionnaire data (CFS)	1450	55.2	37.1	Overweight, Obese	Cross-sectional observational
Mikulovic (2014)[130]	France	NR	Face to face interview questionnaire (2007)	570	NR	38.1	Overweight, Obese	Cross-sectional retrospective
De Winter (2015)[90]	Netherlands	MILD 21.3% MOD 47.6% SEV 16.7% PROF 9.0%	Medical records/Physical examination (CFS)	990	51.3	61.1	Hypertension Hypercholesterolaemia T1DM T2DM Diabetes Peripheral arterial disease Elevated glucose Obese Metabolic syndrome	Cross-sectional observational
Lin, L.P. (2015)[121]	Taiwan	MILD 6.5% MOD 32.6% SEV 34.8% PROF 26.1%	NR (CFS)	67	NR	NR	Overweight, Obese	Cross-sectional observational
Zaal-Schuller (2015)[145]	Netherlands	MILD/MOD 51.1% SEV/PROF 48.9%	Researcher screened (CFS)	407	NR	NR	Peripheral Arterial disease	Cross-sectional observational

ID (intellectual disability); MILD (mild intellectual disability); MOD (moderate intellectual disability); SEV (severe intellectual disability; PROF (profound intellectual disability); NR (not reported); CFS (collected for study); CVD (cardiovascular disease); T2DM (Type 2 diabetes mellitus); T1&T2DM (Type 1 and Type 2 diabetes combined)

Greyed out boxes indicate articles which report findings from the same study

2.5.3 Risk of bias within studies

The funnel plots did not show any obvious asymmetry and Egger's test was not statistically significant for any of the outcome measures (specifically T2DM: t=-0.22; p=0.84; ischaemic heart disease: t=-0.13; p=0.91; cerebrovascular disease: t=0.35; p=0.58) (see Appendix 4 and 5 and 6 (*Figure 27; Figure 28; Figure 29*) for funnel plots).

2.5.4 Results of individual studies and synthesis of results

2.5.4.1 Prevalence of type 2 diabetes

Figure 4 shows the individual studies reporting on the prevalence of T2DM and overall pooled prevalence. Prevalence estimates ranged from 2%[84] to 13%.[90] The pooled prevalence of T2DM was 7.6%. The prevalence of any diabetes was 8.7%; this ranged from 2%[84] to 11%[95, 102, 117] (data not presented).

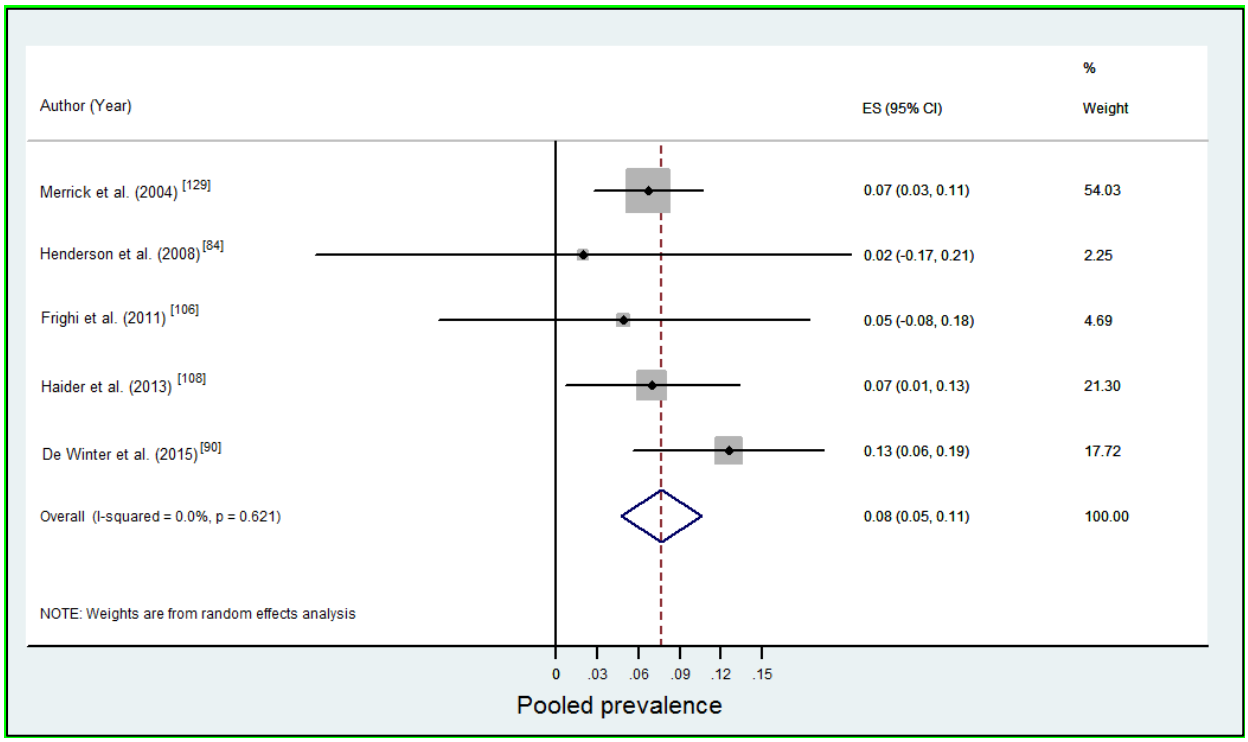


Figure 4: Individual studies and pooled prevalence of type 2 diabetes

2.5.4.2 Prevalence of cardiovascular disease

Figure 5 shows the individual studies reporting on the prevalence of ischaemic heart disease. Prevalence estimates for ischaemic heart disease ranged from 0%[140] to 12%.[126] The pooled prevalence of ischaemic heart disease was 3.7%.

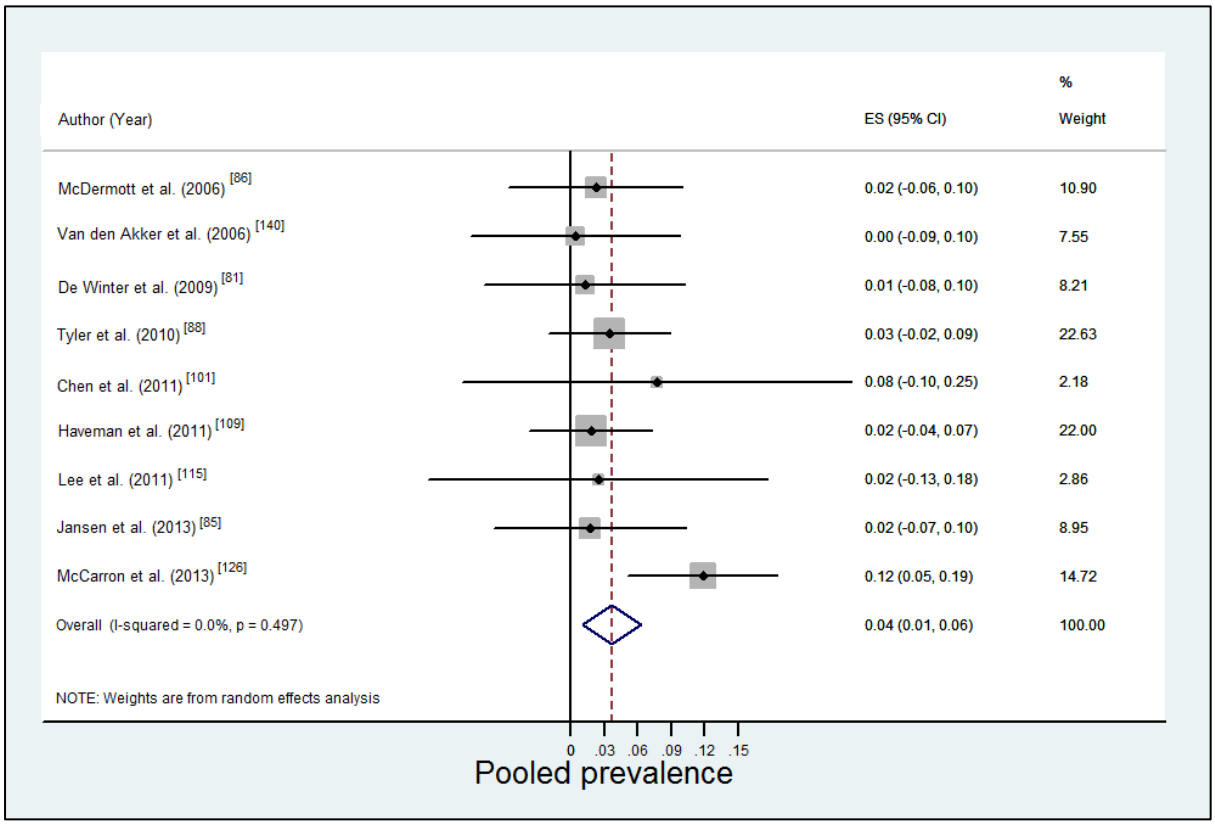


Figure 5: Individual studies and pooled prevalence of ischaemic heart disease

Similarly, *Figure 6* shows the individual studies reporting on the prevalence of cerebrovascular disease. Estimates were fairly consistent in the <1%–4% range. The pooled prevalence of cerebrovascular disease was 2.2%. The pooled prevalence for undefined CVD was 10.6%, but ranged by individual study from 4%[143] to 22%,[114] reflecting the diverse case definitions.

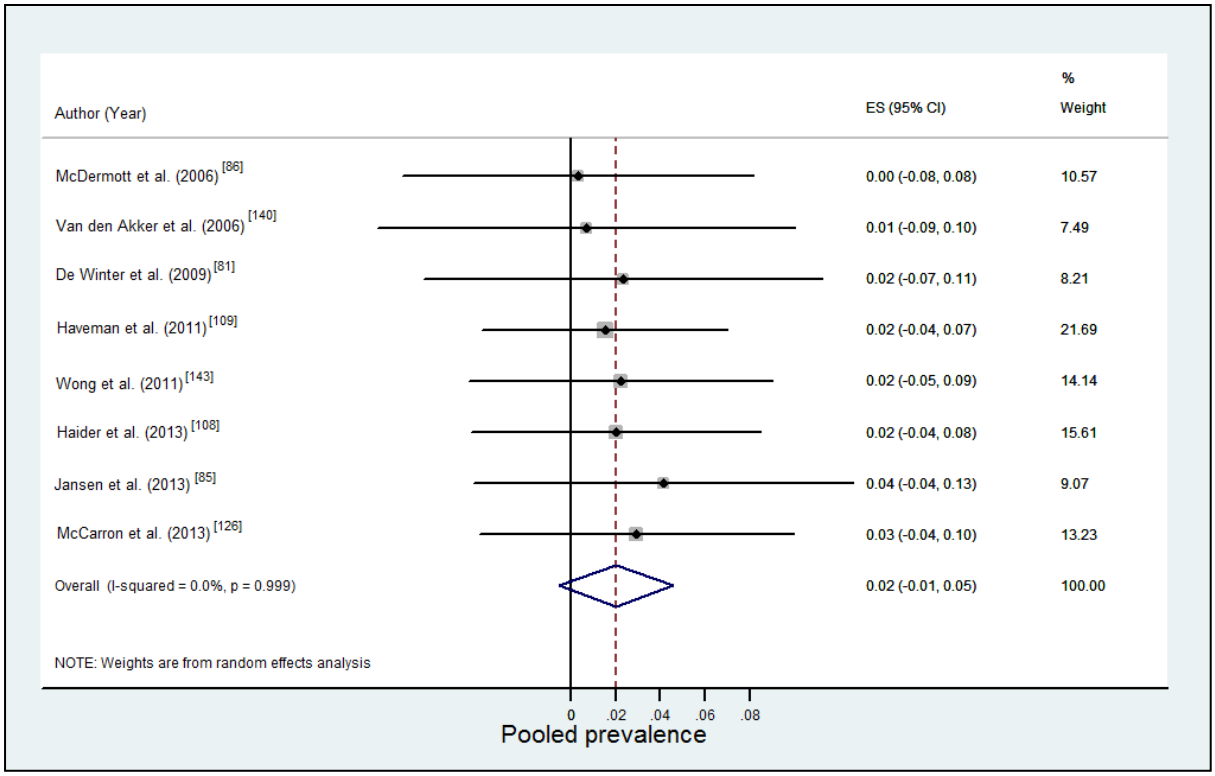


Figure 6: Individual studies and pooled prevalence of cerebrovascular disease

2.5.4.3 Prevalence of other risk factors

Table 5 summarises the findings from the individual meta-analyses. The overall estimated prevalence of hypertension was 18.5%. The estimated prevalence of overweight was 29.2%, obesity was 27.3%, and the prevalence of BMI ≥25 kg/m^2 was 53.4%.

Table 5: Point prevalence for outcome measures in the intellectual disability population

Outcome	Study n	Total n	Total n with outcome	Pooled estimate (95% CI)	I^2
Ischaemic heart disease	9	5586	200	0.04 (0.01, 0.06)	0%
Cerebrovascular disease	8	5748	114	0.02 (0.00, 0.05)	0%
Undefined CVD	8	7773	881	0.10 (0.05, 0.15)	77.5%
T2DM	5	4183	317	0.08 (0.05, 0.11)	0%
Any diabetes	23	19133	1636	0.09 (0.07, 0.10)	0%
Hypertension	28	17161	3008	0.19 (0.13, 0.24)	93.2%
Overweight	32	24923	7434	0.29 (0.26, 0.33)	89.5%
Obese	35	27274	7741	0.27 (0.23, 0.32)	93.6%
Overweight and above	41	31172	16525	0.53 (0.49, 0.58)	96.4%
Hypercholesterolemia	9	3892	491	0.17 (0.08, 0.26)	86.9%
Metabolic syndrome	3	821	287	0.23 (0.00, 0.50)	91.7%

Negative confidence intervals (CIs) have been rounded up to 0.00

On making comparisons with the general population, we found that the ID population had decreased odds of having ischaemic heart disease (OR 0.44 (95% CI 0.34 to 0.58; p<0.01)). No other statistically significant results were found, but we observed high heterogeneity for the other outcomes (see *Table 6*).

Table 6: Findings from the general population comparison meta-analysis

Outcomes	Study n	ID total n	ID total n with outcome	GP total n	GP total n with outcome	Odds ratio (95% CI)	I^2
Ischaemic heart disease	3	2395	67	5441	335	0.44 (0.34, 0.58)*	0%
Any diabetes	6	4014	411	13404	1371	0.96 (0.61, 1.5)	92.2%
Hypertension	6	3588	1097	14262	4598	0.76 (0.58, 0.99)	86.9%
Overweight	4	1487	477	17819	5986	1.31 (0.47, 3.66)	96.5%
Obese	7	3838	1004	23230	6824	1.09 (0.65, 1.82)	95.3%

*p<0.01

2.5.5 Risk of bias across studies

We found high heterogeneity in a number of outcomes when prevalence was pooled (*Table 5*) as well as in a number of outcomes for the general population comparison (*Table 6*). We further explored heterogeneity using meta-regression (see below).

2.5.6 Additional analyses

Meta-regression was used to investigate the effects of mean age, severity of ID and data collection method (researcher collected data, self/carer reported data, or retrospective records data) for prevalence of outcomes (*Table 7*). Mean age was found to affect hypertension only (one unit increase in mean age led to 1% increase in prevalence). Severity of ID had no significant effects on prevalence. Data collection method was found to have an effect on any diabetes when database collected data was compared to self/carer reported data (i.e. database studies tended to report lower prevalence of diabetes). It was also found to have an effect on obesity prevalence when database collected data was compared to researcher

collected data (i.e. database studies tended to report lower prevalence), see *Table 7*. Owing to the small amount of studies and insufficient data, no meta-regression was performed for general population comparison analyses.

Table 7: Results of meta-regression effects analyses for point prevalence

Variable	No. obs	Effect (95% CI)	P-value
Ischaemic heart disease			
Database vs self/carer reported data	9	0.06 (-0.02, 0.14)	0.14
Database vs researcher collected data	9	0.0009 (-0.08, 0.08)	0.98
Mean age	5	0.0001 (-0.01, 0.01)	0.66
% mild/moderate ID	5	0.0005 (-0.01, 0.01)	0.88
Type 2 diabetes			
Database vs self/carer reported data	5	0.05 (-0.21, 0.12)	0.35
Database vs researcher collected data	5	0.06 (-0.39, 0.26)	0.49
Mean age	3	-0.00005 (-0.04, 0.04)	0.99
% mild/moderate ID	*Insufficient observations*		
Any diabetes			
Database vs self/carer reported data	21	-0.05 (-0.08, -0.05)	0.01*
Database vs researcher collected data	21	-0.04 (-0.11, 0.04)	0.33
Mean age	10	0.0006 (0.00, 0.01)	0.70
% mild/moderate ID	11	0.001 (0.00, 0.00)	0.23
Obesity			
Database vs self/carer reported data	35	-0.05 (-0.19, 0.08)	0.44
Database vs researcher collected data	35	-0.12 (-0.21, -0.02)	0.02*
Mean age	18	-0.0009 (-0.01, 0.01)	0.77
% mild/moderate ID	17	0.002 (0.00, 0.01)	0.07
Overweight			
Database vs self/carer reported data	32	0.06 (-0.05, 0.17)	0.26
Database vs researcher collected data	32	0.01 (-0.07, 0.1)	0.8
Mean age	17	0.01 (0.00, 0.01)	0.16
% mild/moderate ID	16	0.002 (-0.00, 0.01)	0.27
Hypertension			
Database vs self/carer reported data	28	-0.08 (-0.21, 0.04)	0.17
Database vs researcher collected data	28	-0.01 (-0.12, 0.11)	0.89
Mean age	13	0.01 (0.00, 0.02)	0.05*
% mild/moderate ID	13	0.0008 (-0.01, 0.01)	0.74

2.6 Discussion

2.6.1 Summary of evidence

In this systematic review, we found that the prevalence of T2DM was 8% and any diabetes was 9%. For CVD, the prevalence of ischaemic heart disease, cerebrovascular disease and undefined CVD was 4%, 2% and 10% respectively.

The current prevalence of T2DM, CVD and associated risk factors in the ID population was found to be similar to that in the general population. However, we found that ischaemic heart disease was significantly lower in the ID population. The meta-regression showed that the method of data collection had minor effects on pooled diabetes and obesity. Mean age had minor effects on hypertension.

2.6.2 Strengths and limitations

A particular strength of this review is that we used robust methods. We wrote to authors to clarify and obtain additional data rather than excluding the articles. To our knowledge, this is the first systematic review and meta-analysis of prevalence of T2DM, CVD and associated risk factors in adults with ID. In addition, it is the first review of its kind to make comparisons with the general population.

However, we had limited data to separate T2DM from other diabetes and were sometimes restricted to unclear or poorly defined outcome measure definitions.
There was also limited data available to make comparisons with the general population. We would have benefited from additional general population data alongside ID population data to make more valid, generalisable comparisons.

2.6.4 Findings in relation to other studies

Two recent reviews have been conducted that have focused on diabetes prevalence among people with ID.[75, 76] The reviews found a mean prevalence of 8.7%[75] and 8.3%[76] for combined gestational, type 1 diabetes (T1DM) and T2DM, but the reviews were unable to report on specific types of diabetes. The overall prevalence of CVD among people with ID is unclear.[52] However, our finding that ischaemic heart disease was significantly lower in the ID population differs somewhat from the literature which suggests that the prevalence of CVD among people with ID is greater than the general population.[52]

2.6.5 Conclusions

Findings from the systematic review and meta-analysis presented in this chapter suggest that T2DM is at least as common in people with ID as in the general population. The findings also identify a gap in knowledge in relation to the prevalence of T2DM as many studies did not report this separately. In addition, none of the studies in our review reported on screen-detected T2DM in the ID population.

CHAPTER 3. SYSTEMATIC REVIEW OF THE EFFECTIVENESS OF MULTI-COMPONENT BEHAVIOUR CHANGE INTERVENTIONS AIMED AT REDUCING MODIFIABLE RISK FACTORS

3.1 Overview

In this chapter, we describe the second of two systematic reviews conducted as part of the research programme. We present the existing evidence in relation to multi-component behaviour change interventions that modify risk factors for T2DM and CVD in people with ID. The PRISMA checklist[74] has been used to guide the reporting of this systematic review.

3.2 Rationale

Non-communicable diseases are on the rise globally and there is increasing demand for lifestyle behaviour change interventions to reduce morbidity, mortality and rising health costs.[146] The suggested mechanisms for this rise are increased availability of energy rich foods and more sedentary lifestyles.[147] T2DM and CVD, and shared associated risk factors are major contributors to morbidity and mortality.[148]

Conditions such as CVD and T2DM share similar risk factors, including dyslipidaemia, hypertension, obesity, and IGR. In the general population, these risk factors can be effectively lowered through interventions focusing on changes in nutrition and physical activity.[149-151] With a suggested increased risk of non-communicable diseases within ID populations, special attention needs to be paid to the efficacy and effectiveness of multi-component behaviour change interventions to reduce this disparity. However, there is a lack of quality evidence on the health and healthcare of people with ID, including effectiveness of health interventions.[152] Previous systematic reviews of lifestyle behaviour change interventions in ID[153-155] have generally been unable to make specific recommendations due to inadequacies

in study design, conduct, lack of theory basis for intervention and/or unclear reporting.

For the current review, we aimed to consolidate the evidence for the reduction of risk of T2DM and/or CVD through the delivery of multi-component behaviour change interventions in the ID population.

3.3 Objectives

The objectives were:

- to establish the effectiveness of multi-component behaviour change interventions in promoting weight loss in the ID population;
- to establish the effectiveness of multi-component behaviour change interventions in reducing other modifiable risk factors for T2DM and/or CVD in the ID population;
- to establish the effectiveness of multi-component behaviour change interventions aimed at primary prevention of T2DM or CVD, or reducing associated risk factors in the ID population.

3.4 Methods

3.4.1 Protocol and registration

The systematic review was registered with the international register of prospective systematic reviews, PROSPERO 2015:CRD42015020758.[156]

3.4.2 Eligibility criteria

The review was guided by the PICOS (population, intervention, comparison, outcome, study designs) model.[78] We defined the population as adults (≥18 years) with ID (whole study population or a defined sub-sample). We defined the intervention as any multi-component lifestyle behaviour change intervention aimed at primary prevention of T2DM or CVD, or a reduction in associated risk factors for

people with ID and/or their carers. We included studies with and without comparison groups. We defined outcome measures as changes in anthropometric measures (weight, BMI, waist circumference), BP, lipid levels, glucose levels, physical activity levels, sedentary behaviour and dietary habits. The study design was defined as an experimental study (before and after study, randomised controlled trial or non-randomised controlled trial) with a follow-up period of at least 24 weeks or 6-months from baseline (to allow for the initiation and maintenance of medium and longer term behaviour change),[157] see *Table 8*.

All studies published since 1 January 2000 (until 21 April 2015) and in English language were eligible. Studies were limited to those published from the year 2000 when most large diabetes prevention trials were first published in the general population.[70]

We contacted lead authors for further information where inclusion/exclusion could not be determined.

Table 8: Inclusion and exclusion criteria of studies included in the systematic review of multi-component lifestyle behaviour change interventions

	Inclusion Criteria	Exclusion criteria
Population	Whole study population or defined sub-sample of adults (≥18 years)*	
Intervention	Multi-component lifestyle behaviour change intervention aimed at primary prevention of T2DM or CVD or a reduction in associated risk factors (weight management, increasing physical activity/reducing sedentary behaviour, dietary improvement)	Interventions involving meal replacements, or were aimed at increasing physical fitness (in isolation) as opposed to changes in levels of physical activity
Comparison	Studies without comparison groups were included	
Outcomes	Changes in anthropometric measures (e.g. weight ,BMI, body fat, waist circumference) Blood pressure Lipid levels Physical activity Sedentary behaviour Dietary habits	
Study designs	Before and after study Randomised controlled trial Non-randomised controlled trial	Follow-up period <24 weeks/<6 months from baseline

* For generalisability to the adult ID population as a whole, studies with >20% aged under 18 years, >20% disability other than ID and >25% with specific ID were excluded.

3.4.3 Information sources

We searched the electronic databases EMBASE, MEDLINE, CINAHL, CENTRAL and PsychINFO for this systematic review. The last date of the search was 21 April 2015. We searched the references lists of relevant systematic reviews and included papers within those for additional studies.

3.4.4 Search

We combined MeSH terms and key words for multi-component lifestyle interventions and outcome measures and ID. The search was limited to English language studies with cohorts of adults ≥18 years of age, depending on the database (see *Table 9* for MEDLINE search strategy).

Table 9: Search strategy for MEDLINE electronic database

1.	(Behav* adj1 (Modif* or therap*)).ti,ab.
2.	Cognitive* therap*.ti,ab.
3.	(Health* adj2 (Educat* or promot* or behav*)).ti,ab.
4.	Educat* adj2 program*.ti,ab.
5.	(Diet* adj2 (Intervention* or modif* or therap*)).ti,ab.
6.	(Health* adj2 Eating).ti,ab.
7.	(Nutrition* adj2 (intervention* or modif* or counsel* or therap*)).ti,ab.
8.	(Exercis* adj2 (intervention* or therap*)).ti,ab.
9.	(Physical adj (education or fitness or activit* or training or exercise)).ti,ab.
10.	(Lifestyle adj2 (advice or guidance or modif* program* or interven*)).ti,ab.
11.	(Weight adj2 (control* or los* or reduc* or maintenance or management)).ti,ab.
12.	Weight adj loss adj program*.ti,ab.
13.	Exercise*.ti,ab.
14.	Sport*.ti,ab.
15.	exp Health Promotion/
16.	exp Nutrition Therapy/
17.	exp Exercise Therapy/
18.	(Sedentary adj (behav* or lifestyle* or individual* or population*)).ti,ab.
19.	or/1-18
20.	exp Intellectual disability/
21.	((learning or development* or intellectua* or mental*) adj1 disabilit*).ti,ab.
22.	(impair* adj2 intellectual adj2 function*).ti,ab.
23.	(mental* adj1 (impair* or handicap*)).ti,ab.
24.	Exp mentally disabled persons/
25.	(mental* adj2 retard*).ti,ab.
26.	Or/20-25
27.	19 and 26
28.	animal/ not (animal/ and human/)
29.	27 not 28
30.	limit 29 to english language
31.	limit 30 to yr=2000-current

3.4.5 Study selection

Full texts were identified after titles and abstracts were read separately by two investigators (TC, AD) who discussed discrepancies in selection at a later meeting. On retrieving the full-text articles, papers were again examined separately by two investigators (TC, AD) to check for suitability for inclusion.

3.4.6 Data collection process

We created a data extraction form for this review. Data were extracted by one investigator (TC) and verified for accuracy by another investigator (AD).

3.4.7 Data items

For each study, we collected the first author's name, title of paper, year of publication, country of the cohort, study design, sampling method, intervention details, and dates of data collection and the intended recipient of the intervention. For the whole study population (and for each group, if applicable), we extracted data on total sample size or sub-population size, mean ages, proportion of males/females, severity of ID, ethnicity, and withdrawals.

For each reported outcome, we extracted information on how outcomes were defined and measured, the total number measured for each outcome, length of follow up, mean baseline and post intervention value, mean between-group change and/or change baseline to follow up along with a measure of variability (SD, SE etc.). We extracted data separately for males and females, where reported.

3.4.8 Quality assessment

The NICE quality appraisal checklist for quantitative intervention studies[158] was used to assess the quality of the selected studies. The checklist included criteria for assessing the internal and external validity of experimental and observational

quantitative studies (randomised controlled trials (RCTs), non-randomised controlled trials, before and after studies) and allowed assignment of an overall quality grade (categories ++, + or -). Studies were assessed by one reviewer (TC) and verified for accuracy by a second reviewer (RS).

3.4.9 Risk of bias in individual studies

Prior to carrying out this systematic review, we anticipated using funnel plots[79] and the Egger's test[80] to examine potential publication bias in the literature for the collected outcomes. However, owing to the small number of studies included in this review resulting in low power to detect bias, these methods were not used.

3.4.10 Data synthesis

Data synthesis for this review involved describing the study characteristics (country, population size, age, %male, ethnicity, severity of ID, eligibility criteria, outcomes report and follow-up period) of included articles. We then described the details and behavioural strategies of each of the multi-component lifestyle behaviour change interventions, including their structure and delivery and the underlying theory behind each of the interventions. Finally, we described the outcome measures and study findings. Given the low number of studies included, a formal evidence synthesis was not undertaken.

3.6 Results

3.6.1 Study selection

The literature searches yielded 3508 articles. After duplicates were removed, 3167 articles remained to be screened. We retrieved and reviewed the full-text of 39 articles for 32 studies (*Figure 7*). Most potentially relevant studies were noted for their small sample size, short follow up, high attrition rates and/or incomplete data for key outcomes. We contacted the authors of four study protocols for further information.[159-162] One of the authors did not reply, and we were informed that the remaining three protocols were still awaiting publication and could not be included in the review. In total, we identified only four studies for inclusion in this review.

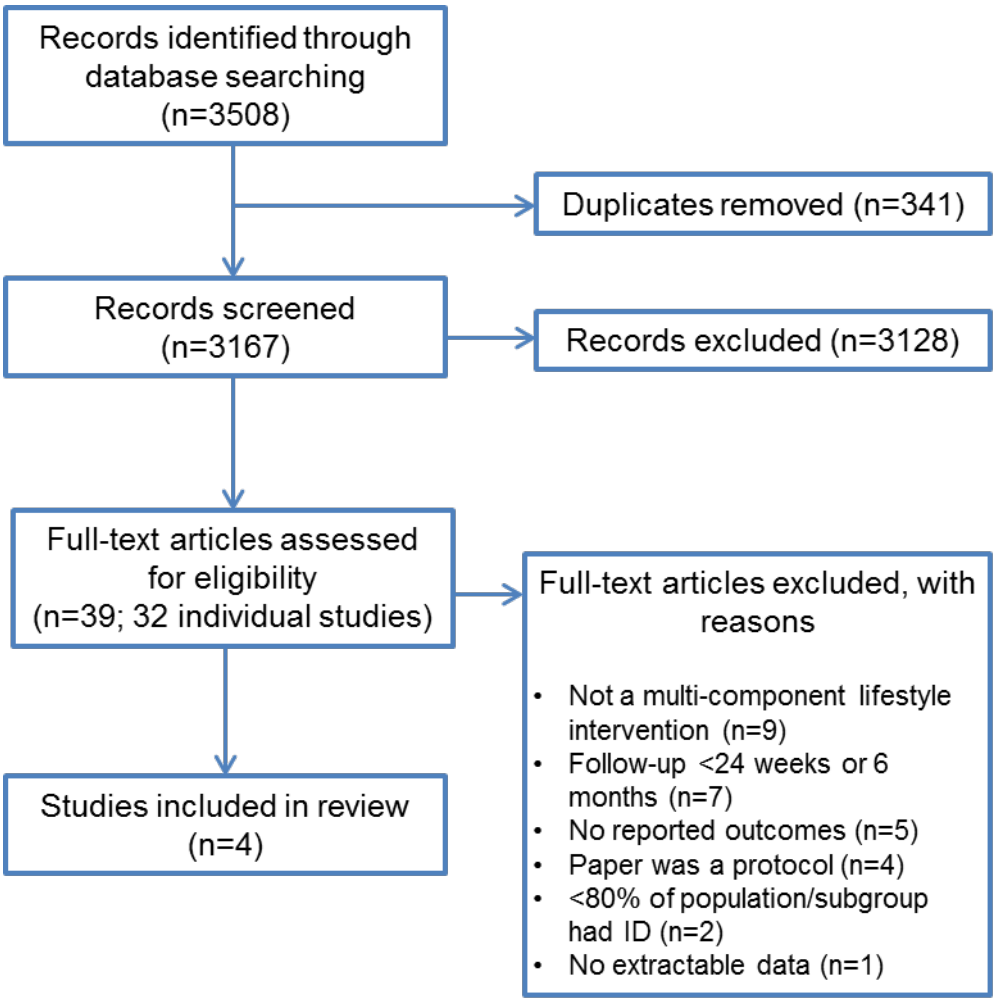


Figure 7: Flow diagram of study selection

3.6.2 Study characteristics

The four studies included in the systematic review presented data on 700 individuals. The characteristics of each of the studies are presented in *Table 10*.

The studies covered three countries (USA, UK and Sweden). Two studies were single arm, and two studies had a control group. All four studies reported data for physical activity and/or sedentary behaviour.

The average mean age was 42.2 years and the average percentage of male participants was 42.9%. The mean average group size was 174 before drop-out and 104 after drop-out. Group sizes ranged from 54 to 443 before drop-out and from 44 to 196 after drop-out. The majority of participants were white (68% where known); approximately one-quarter (26%) were black and the remaining 7% were from other ethnic groups. Only one study provided information on severity of ID, but based on eligibility criteria, the remaining studies were likely to comprise adults in the mild to moderate ID range. Other descriptive information for each study is presented in *Table 10*.

Table 10: Studies included in the systematic review of multi-component behaviour change interventions

Author & year	Country	N (n after drop out)	Mean age (SD)	% Male	Ethnic Group	Mean BMI	Eligibility criteria	Severity of ID	Key outcomes reported	Follow-up period
Bazzano (2009)[163] **"Healthy Lifestyle Change Program (HLCP)"**	USA	68 (44)	44.0	38.6%	White (64%) Black African (21%) Other (16%)	33.3	- Age 18–65 - High-functioning ID - BMI ≥ 25 - Diabetes or risk factors for diabetes (incl.hypertension, hyperlipidaemia, family history, hyperglycaemia, ethnicity (non-white), age >45 years)	NR	BMI, Weight Waist circumference Physical activity (frequency and duration)	7 months
Melville (2011)[164] **"TAKE-5 STUDY"**	UK	54 (47)	48.3	40.7%	White (97%) Pakistani (2%) Other Asian (2%)	40.0	- ≥18 years old - BMI ≥ 30 - Ambulatory Excluded - Prada-Willi syndrome	MILD 31.5% MOD 31.5% SEV 35.2% PROF 1.9%	BMI, Weight Waist circumference Sedentary & Physical activity (mins/day accelerometer)	24 weeks
McDermott (2012)[165] **"Steps To Your Health (STYH)"**	USA	443 (196)	38.8	49.2%	White (42%) Black (57%) Hispanic (1%) Other (1%)	32.4	- Age 18–65 - Voluntary participation - Ambulatory and communicative - Mild to moderate ID - Residence in independent or supported settings. Excluded - Underweight (BMI<18.5)	NR	BMI Moderate to vigorous physical activity (accelerometer)	12 months
Bergstrom (2013)[166]	Sweden	130 (129)	Int. 36.2(10.1) Control 39.4(11.3)	43.1%	NR	Int. 30.0 Control 28.5	- Adults - Mild to moderate ID - ≥3 residents	NR	BMI, Weight Waist circumference Physical activity (steps/day pedometer)	12–16 months

3.6.3 Study quality

A breakdown of study quality is presented in *Table 11*. The studies were generally of high quality; in particular, all of them achieved at least a good quality rating for internal and external validity. However, two of the four studies failed to account for all of the participants when concluding the study, and three of the four studies did not report on whether the studies were sufficiently powered to detect differences.

Table 11: Quality assessment of articles included in the systematic review

	Bazzano[163]	Melville[164]	McDermott[165]	Bergstrom[166]
SECTION 1 – POPULATION				
Source population/area well-described?	+	++	++	++
Eligible population/area representative of source population/area?	++	+	++	++
Selected participants/areas represent eligible population?	+	++	++	+
SECTION 2 – METHOD OF ALLOCATION TO INTERVENTION (OR COMPARISON)				
Allocation to intervention (or comparison). Was selection bias minimised?	NA	NA	++	++
Interventions (and comparisons) well described and appropriate?	++	++	++	++
Was allocation concealed?	NA	NA	NR	++
Participants or investigators blind to exposure and comparison?	NA	NA	NA	NA
Exposure to intervention appropriate?	++	++	++	++
Contamination acceptably low?	NA	NA	++	++
Other interventions similar in both groups?	NA	NA	++	++
Participants accounted for at study conclusion?	-	++	-	++
Did setting reflect usual UK practice?	++	++	++	++
Did intervention or control comparison reflect usual UK practice?	++	+	+	++
SECTION 3 – OUTCOMES				

Outcome measures reliable?	+	+	++	+
All outcome measurements complete?	++	++	+	-
All important outcomes assessed?	++	++	++	++
Outcomes relevant?	++	++	++	+
Similar follow-up times in exposure and comparison groups?	NA	NA	++	+
Follow-up time meaningful?	+	++	++	++
SECTION 4 – ANALYSES				
Exposure and comparison groups similar at baseline? If not, were these adjusted?	NA	NA	NR	++
Intention-to-treat (ITT) analysis conducted?	-	-	++	++
Sufficiently powered to detect an intervention effect (if one exists)	NR	NR	NR	++
Estimates of effect size given or can be calculated?	NR	++	++	+
Analytical methods appropriate?	+	-	++	+
Precision of intervention effects given or able to be calculated? Were they meaningful?	+	++	++	++
SECTION 5 – SUMMARY				
Study results internally valid? (i.e. unbiased)	+	+	++	+
Findings generalisable to the source population? (i.e. externally valid)	++	+	++	+

++ All of the quality assessment checklist criteria were fulfilled; + Some of the quality assessment checklist criteria have been fulfilled; - Few or none of the quality assessment criteria were fulfilled; NR not reported; NA not applicable.

3.6.4 Results of individual studies and descriptive data synthesis

Table 12 summarises the multi-component lifestyle behaviour change interventions evaluated in the individual studies.

Bazzano and colleagues (2009)[163] conducted a single-arm before and after intervention in already overweight or obese individuals (BMI≥25 kg/m^2). The intervention involved peer-mentoring, one-to-one health education, supervised physical activity and clinical support aimed at reducing weight, diet & increasing physical activity.

The study by Melville and colleagues (2011)[164] also conducted a single-arm study in already obese individuals (BMI ≥ 30 kg/m^2) who had been referred to a dietician by their GP. Nine lessons, every 2–3 weeks, were provided for participants and their carers. Lessons were aimed at increasing physical activity and better diet as well as weight loss. Interventions also consisted of personalised diet plans with calorie restrictions (600 kcals per day).

Bergstrom and colleagues (2013)[166] conducted a two-armed trial in community residential homes, targeting both people with ID and their carers. The intervention offered a 'study circle' for carers and an appointed health ambassador at each residential home. An educational health course for the residents was also provided. The community residences in the control arm received the option to take part in the intervention after study completion (wait-list control). The primary outcome for this trial was increasing physical activity and the secondary outcomes were decreasing weight and BMI.

Table 12: Details of the interventions evaluated

Author & year	Key elements of intervention	Structure of intervention (no. sessions & length)	Who delivered intervention	Where intervention delivered	Goal setting	Theory
Bazzano (2009)[163]	**Diet, exercise & behaviour modification** • Interactive health education • Supervised physical activity • Peer mentoring • Clinical support	• Twice weekly, 2 hour sessions (for 7 months) • Each class included 50 mins health education and 1 hour supervised physical activity • Outcomes assessed at baseline and at 7 months	Professionals with ID expertise with assistance from peer mentors	Community organisation	NR	Based on Bandura's social cognitive theory of health behaviour change
Melville (2011)[164]	**Weight loss, diet and exercise** • Energy-deficit diet • Goal setting and self-monitoring to increase physical activity, encourage weight loss and improve diet	• 9 sessions, every 2-3 weeks (40-60 mins each) • 24 week follow up	Two health professionals (dietician & sports medicine graduate) with experience working with individuals with ID	Participant's home	Individual goal setting regarding weight loss, dietary change & physical activity	NR
McDermott (2012)[165]	**Diet, exercise, & stress reduction** • Health promotion intervention focusing on nutrition, exercise, stress management, changing ways of thinking, communication styles, complications of obesity & behaviour management	• 8 weekly sessions (90 mins each) • Data collected at baseline, 9 weeks after completion, 6 months and 1 year	Health educator with experience working with adults with ID	Community venue	NR	Based on Bandura's social cognitive theory of health behaviour change
Bergstrom (2013)[166]	**Diet and exercise modification** Three components: • Health ambassador in each residence • Study circle for caregivers • Health course for residents – learn about health issues, try healthy foods and physical activities	• 12-16 months to complete programme • Health ambassadors: 6 network meetings (3 hrs each) • Study circle: 10 sessions (90 mins each) • Health course: 10 sessions • Outcomes assessed at baseline and at end of intervention (12-16 months from baseline)	• Health ambassador • Member of staff from residence • Course leader from national educational association for adults	Community residential homes	NR	Based on Bandura's social cognitive theory of health behaviour change

Finally, McDermott and colleagues (2013)[165] conducted a two-arm randomised control trial. Intervention participants were assigned to eight weekly lessons in nutrition, exercise, and changing ways of thinking. The lessons focused on stress management, complications of obesity, and behaviour management. The classes emphasised moderate to vigorous physical activity (MVPA), healthy eating and BMI reduction. The control group was assigned to eight weekly lessons on safety and hygiene.

Table 13 summarises the components of the individual behaviour change interventions. All of the interventions used both dietary and exercise components.

Table 13: Individual components of the interventions evaluated

	Bazzano[163]	Melville[164]	McDermott[165]	Bergstrom[166]
DIETARY COMPONENT				
Energy restriction		600 kcal/d		
Weight loss target		5% of initial body weight		
Nutrition advice	✓		✓	
Try healthy foods in session	✓		✓	✓
National recommendations				✓
Healthy dietary habits				✓
Portion sizes		✓		
Individualised diet plan		50% carbs <35% fats <20% protein		
Individualised diet goals		Set one goal per week		
EXERCISE COMPONENT				
Individualised exercise goals		- Walking targets (using pedometer) - Set one goal per week - Minimum 30 minutes mod, intensity physical activity at least 5 days per week		
Advice regarding time and intensity		- Advice on replacing sedentary behaviour for activities in the home, e.g. housework		
Supervised activity in session	- 1 hour during each session - Use of local parks and fitness facilities - Exercise video created by peer mentors		Sessions followed by optional brisk walk	Physical activities in sessions
Information provided regarding local leisure facilities		✓		

Out of the four included studies, the two single-arm studies with a follow up of seven months[163] and 24 weeks[164] respectively, indicated significantly improved outcomes. Reductions in weight, BMI, and waist circumference were demonstrated after the implementation of a behaviour change intervention programme aimed at increasing physical activity and improving diet. Additionally, both studies demonstrated a significant improvement in physical activity outcomes, specifically, for 'minutes per week' and 'frequency of sessions'[163] and a 'reduction in sedentary behaviour'.[164] Both cohorts were overweight to obese when they were enrolled into the study. For the further two studies[165, 166] where the cohort was not recruited based on health status, one of the studies showed significant positive improvements in waist circumference, BMI, and steps per day in those who received the intervention compared with the controls;[166] the second study did not show any significant differences between control and intervention arms (see *Table 14*).[165]

Table 14: Reported data for included studies

Author & year	Body mass index (Kg/m^2)	Weight (Kg)	Waist circumference (cm)	Vegetable intake (Servings per day)	Physical activity/Sedentary behaviour
Bazzano (2009)					**a) Mins per week ǂ** **b) Sessions per week ǂ**
Baseline	33.3 (**n=44**†)	88 (**n=44**†)	104.9 (**n=39**)	2 ((**n=44**)	a) 133 b) 3.2 (**n=44**)
Follow-up (7 months from baseline)	32.8 (**n=44**)	86.8 (**n=44**)	102.6 (**n=39**)	2.2 (**n=44**)	a) 206.4 b) 3.9 (**n=44**)
Intervention group change	-1.5%*	-1.34%*	-2.18%**	+10%	a) +54.89%** b) +21.88%**
Melville (2011)					**a) Sedentary mins per day** **b) Low PA mins per day** **c) MVPA mins per day (accelerometer)**
Baseline	40 [8.03] (**n=47**)	100.6 [26.8] (**n=47**)	122.1 [15.7] (**n=47**)	NR	a) 623.3 [121.5] b) 73.4 [46.8] c) 14.2 [17.5] (**n=45**)
Follow-up (24 weeks from baseline)	39.2 [8.2] (**n=47**)	96.1 [26.9] (**n=47**)	115.8 [16.7] (**n=47**)	NR	a) 581.9 [116.4] b) 81.3 [45.6] c) 17.8 [17.3] (**n=33**)
Intervention group change	-4.45%**	-4.55%**	-5.15%**	NR	a) -6.64%* b) +10.76% c) +25.42%
McDermott (2012)					**MVPA ratio - minutes performed/minutes worn (accelerometer)**
Baseline	32.38 [6.85] (**n=437**)	NR	NR	NR	3.24 [3.93] (**n=401**)

Follow-up (12 months from baseline)	32.13 [6.59] (**n=195**)	NR	NR	NR	4.62 [3.27] (**n=118**)
Intervention group change	-0.78%	NR	NR	NR	-4.18%
Bergstrom (2013)					**Steps per day (pedometer)** ǂ
Baseline	30 [7.6] (**n=126**)	NR	94.5 [16.5] (**n=124**)	1.4 [0.6] (**n=101**)	8042 [5524] ((**n=99**)
Follow-up (12-16 months from baseline)	29.7 (**n=108**)	NR	92.8 (**n=103**)	1.6 (**n=66**)	9650 (**n=69**)
Intervention group change	ǂ-1%	NR	ǂ-1.8%	+14.29%	+19.99%*

Standard deviations in brackets where reported;

†baseline characteristics of 24 people who did not complete the programme were not reported.

ǂself/carer reported data; *significant p<0.05; **significant p<0.01; NR-Not reported; PA-Physical activity; MVPA-Moderate to vigorous physical activity;

3.7 Discussion

3.7.1 Summary of evidence

This review contributes to the existing knowledge on the effectiveness of multi-component lifestyle behaviour change interventions in adults with ID. Three of the interventions included in this review led to some reductions in BMI, weight and waist circumference,[163, 164, 166] but inferences are limited owing to small sample sizes, missing data, selected populations and/or lack of control groups.

3.7.2 Strengths and limitations

To our knowledge, this is the first systematic review and meta-analysis focusing on long-term multi-component behaviour change interventions for people with ID in order to reduce CVD and/or T2DM risk. We used robust methods and sought additional information from authors where relevant. However, only four papers met our inclusion criteria. Significant findings were observed only for the single-arm studies which are known to over-estimate effect sizes.[167] We were unable to test for publication bias or to carry out meta-analytical work to explore combined effects, particularly as the interventions evaluated were so diverse. Similarly, research has shown that improvements in health can be difficult to sustain in the longer term;[168] only two of the included studies had a follow-up period of at least 12-months[165, 166] and even this may not be enough to indicate long-term sustained benefits.

3.7.3 Findings in relation to other studies

In line with previous systematic reviews in this area,[153-155] findings from this systematic review demonstrate a lack of quality evidence on the effectiveness of multi-component behaviour change interventions in people with ID. In 2010, Jinks and colleagues[153] focused a systematic review on qualitative studies of behavioural change approaches in people with ID to aid weight loss and health. The review found 12 papers, of which only one was qualitative. The authors noted an overall lack of

research on behavioural approaches and using qualitative methods. Similarly, in 2013, Spanos and colleagues reviewed 22 papers assessing interventions for weight loss in people with ID.[154] They noted that many of the interventions did not meet the recommended duration in clinical guidelines and were too specific. Brooker and colleagues (2014)[155] also reviewed interventions with a primary focus on physical activity in people with ID. Again, the review noted small sample sizes and invalid measurement tools and recommended further longer term intervention studies.

3.7.4 Implications of findings

This systematic review informed the evidence-base for the development of the STOP diabetes educational programme described in Chapter 8 and Chapter 9. The studies also revealed a high rate of missing follow-up data for participants who completed the multi-component lifestyle behaviour change interventions, which helped to inform further development work on feasibility testing (Chapters 10). The wider implications for research and practice are discussed in Chapter 13.

3.8 Conclusions

The findings from this systematic review have provided some evidence that multi-component behaviour change interventions may be beneficial in modifying risk factors for T2DM and CVD in people with ID. However, there is a paucity of literature on their long-term effects in this population. In keeping with existing recommendations,[154] we highlight the need for robust randomised controlled trials to evaluate the long-term effects of multi-component behaviour change interventions, informed by current guideline recommendations, for people with ID.

CHAPTER 4. SERVICE USER INVOLVEMENT

4.1 Overview

This chapter details the service user involvement throughout the STOP Diabetes research programme. Involvement was integrated into the research from the early stages.

4.2 Introduction

The involvement of service users in research is central to UK policies[169, 170] and is becoming increasingly common both nationally and internationally.[171-174] The benefits of such involvement in health and social care research are manifold. Service users can provide valuable knowledge and insights to research,[175-180] encourage recruitment through publicity,[176, 178, 179] improve quality, relevance and impact of research[181-185] and potentially help to meet recruitment targets.[171] Service users in England contribute financially to public-funded research so arguably have a right to be involved[185, 186] and can personally benefit from their involvement.[182, 184, 187] However, challenges to the successful involvement of service users in research include contrasting priorities,[182, 188, 189] understanding of research methods,[188] use of language and jargon[188] and lack of time and resources.[182, 188]

The involvement of people with ID in research can pose additional challenges to those outlined above and include the need to plan ahead, allow time for effective communication and regular breaks, and ensure that meeting locations are accessible to all.[190-193] Such challenges can be at odds with researchers' own demands and priorities[194] and they often resort to seeking the proxy views of 'sympathetic others', such as parents or carers,[195] which is disappointing, given that people with ID have a lot to say and can improve the quality and relevance of research.[194]

4.3 Involvement prior to submitting the research proposal

Before submitting the research proposal, members of the team visited three local ID partnership boards to discuss the study, invite feedback, discuss how adults with ID could be involved in the research process and advise on reasonable adjustments and practical considerations. The boards comprised a mix of professionals and public members, including councillors, commissioners, clinicians, charity representatives, police officers, family carers, paid carers and people with ID. The boards provided useful advice on tailoring information sheets to service users (for example, using pictures as well as text, using a larger font and modifying the size and colour of the paper for those with visual impairment) and on communication issues (for example, using a staged, step-by-step approach to delivering information). The team also began forming links with two local self-advocacy groups for people with ID: both groups met at least monthly in a central location, and were led by an experienced facilitator whose role was to ensure that members understood what was being discussed, had every opportunity to give their views and contribute to the discussion, and that only one person spoke at a time.

4.4 Involvement during the research programme

4.4.1 Selection of service users for involvement

Service users were approached from different sources to encourage a diverse range of views and to minimise burden. Members from the two self-advocacy groups approached, the *Speaking up for Health Group* and the *Charnwood Action Group*, agreed to help the team with the study. In addition, the manager and residents of a communal care establishment were approached through the lead nurse's contacts and agreed to help us with the study. Service users who entered the poster competition (see next section) were indirectly involved by providing publicity materials for the team (*Figure 8*).

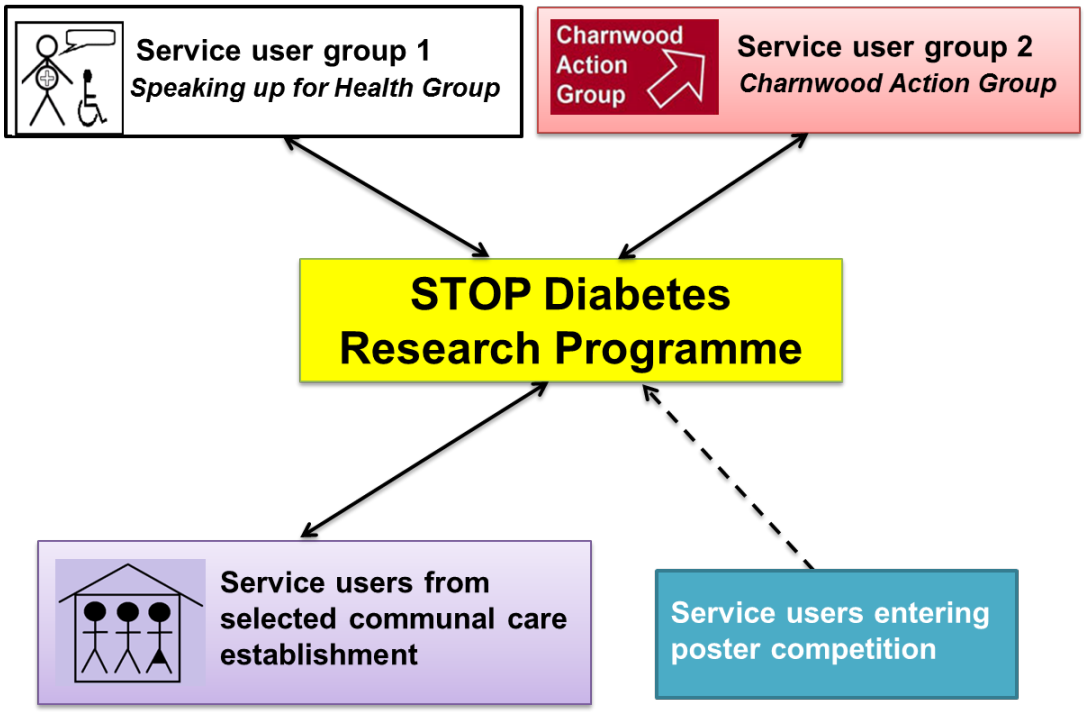


Figure 8: Service users involved in the research programme

4.4.2 Service user involvement in the programme management

A common way of involving service users in research is through representation on steering group meetings[196] and we discussed the potential for this with the service users. We considered tailoring these meetings to make them more accessible, but past experience suggested that they could be lengthy, involving complex discussions about procedures, accelerometer data, health economics and statistical methodologies and often used conference call facilities. We were concerned that the meetings would be isolating for the service user and their supporters, so instead, we agreed to feedback key points from the meetings and that service users could attend on an 'ad hoc' basis.

4.4.3 Service user involvement in promoting the research programme

The study involved a Leicestershire-wide screening programme and it was important to promote the research as widely as possible. Among other considerations, the study logo and publicity materials needed to be suitable and appropriate for the target population. Both of the two service user groups that we approached used "word police" cards, which were shown whenever another member of the group or visitor used an acronym or abbreviation that they did not understand. Therefore, the proposed use of an acronym for the programme was not received favourably and instead, the team opted to call the programme, the "STOP Diabetes study". The creative director subsequently devised four corresponding logos (*Figure 9*) and asked the *Charnwood Action Group* their preference, using a feedback form with a scale and pictures. The preferred logo (see option 4, *Figure 9*) was then shown to the *Speaking up for Health Group* and ways in which the logo and other publicity materials, such as posters and fliers, could be used to publicise the programme were discussed. The service users recommended printing the STOP Diabetes logo on note pads, pens and fridge magnets. When directly asked, they also thought that the logo should be printed on the study documentation, such as information leaflets and consent forms. Members of the group also suggested holding a poster competition as a means of publicising the programme.

All ideas were collated and reported back to the research team for further discussion, and to determine if sufficient resources were available to action them. All of the suggestions were taken up by the research team.

(The fourth picture was the service users' preferred logo)

Figure 9: Proposed logos for the STOP Diabetes programme

Service users were invited to enter the poster competition using brief easy-read information distributed by staff at local day centres, health clinics and other organisations. People who entered the competition were given a certificate and a small award of art and craft materials. Four of the pictures were chosen for the promotional materials (*Figure 10*); this decision was made by both service users and members of the research team.

(The four pictures on the left were used for the publicity materials)

Figure 10: Some of the artwork submitted for the poster competition

The research programme was publicised via the Leicestershire Diabetes Centre website and was also published in the INVOLVE Summer 2013 newsletter,[197] both as means of raising awareness about the programme and sharing our experiences of service user involvement (*Figure 11*). Similarly, having attended one of the programme steering group meetings, one of the co-chairs of the *Charnwood Action Group* contacted the media and was consequently interviewed about the programme (radio and newspaper[198]) which helped with recruitment.

Figure 11: Service users' involvement in promoting the research programme

(Image of article "Volunteers needed for study" reproduced with permission of Leicester Mercury. Copyright © 2016 Local World. All Rights Reserved.);(Image of article "STOP Diabetes study" reproduced with permission of INVOLVE. Copyright © INVOLVE all right reserved 2015.)

4.4.4 Service user involvement in study documentation and process development

It was important that information about the programme be available in simple language, free from jargon and acronyms, and using pictures and symbols, so that potential participants had every opportunity to understand what the research team were doing and to reach an informed decision about whether to take part. Prior to submission to the research ethics committee, the research team, with support from local ID services, drafted an easy-read (symbols and words) information sheet and consent form to partnership boards, local ID services and service user groups for

feedback on the symbols, text and whether additional communication aids might be necessary.

The service users that we asked to read the easy-read documents did not report any difficulties in understanding them, but recommended additional modes of communication, such as flash cards and story cards. A member of the *Speaking up for Health Group* assisted the team by taking photographs to illustrate the information leaflet, flash cards and story cards; three service users modelled for the photographs (*Figure 12*).

Figure 12: Service users' involvement in assisting with study documentation

4.4.5 Service user involvement in staff recruitment

Research nurses were integral to successful recruitment to the study, needing to be patient, sensitive, responsive to participants' needs (e.g. seeing participants outside typical working hours), as well as able to communicate effectively with people with ID. Two members from the *Speaking up for Health Group* offered to help with the interview process for recruiting nurses into the research programme. Supported by their group facilitator, they created two questions to assess how good the nurses

were at communicating with people with ID, and how they might adapt their style of communication if that person did not understand them. On the day that the nurses were interviewed, the service users asked these questions in a separate room, with the facilitator present. They then rated the nurses' responses on a 4-point scale (*Figure 13*). Their input helped to reinforce the panel's decision on who to recruit and was particularly valuable in helping the panel to decide between two similar applicants.

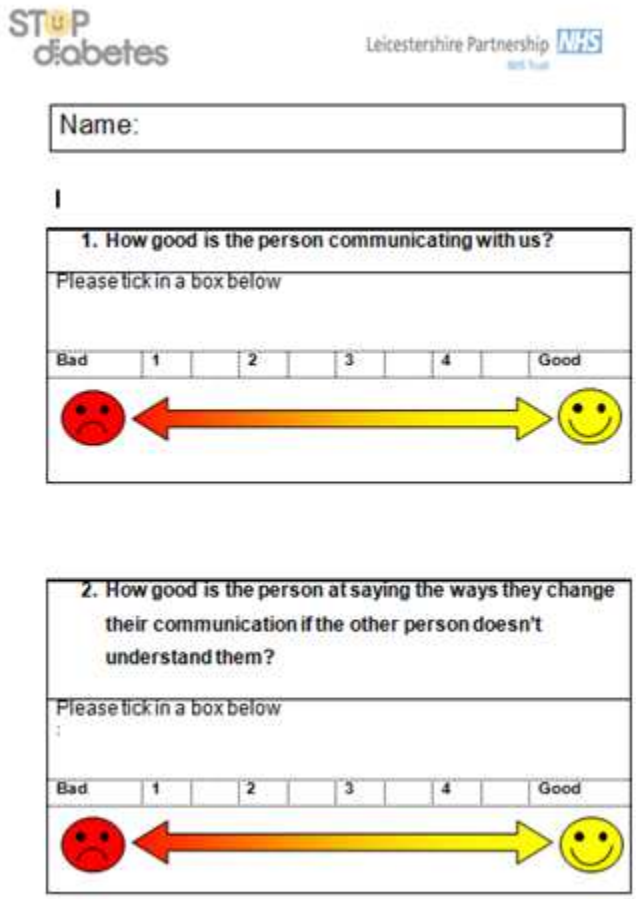


Figure 13: Service users' rating form for recruitment of nurses

4.4.6 Service user involvement in training staff and assessing acceptability of measures

Service users at the participating communal care establishment helped to train staff by allowing them to practise communication-based interactions, consent taking and

measurement collection. Service users from one of the self-advocacy groups were invited to attend a follow-on staff training session so that nurses could put their new skills into practice. They gave feedback on nurses' skills and discussed what they liked and what they did not like, enabling staff to gain confidence and develop competency in various procedures. The mock clinics also helped the research team to determine how long the appointments might take and how many visits may be needed. The service users reported that some of the questionnaires were too lengthy and complex. The team met to discuss the issues raised and made changes to reduce participant burden; these included swapping one of the questionnaires (Psychiatric Assessment Schedules for Adults with Developmental Disabilities (PAS-ADD) mini[199] for the PAS-ADD checklist[199]) and removing two questionnaires (Dietary Instrument for Nutrition Education (DINE)[200] and International Physical Activity Questionnaire (IPAQ)[201]) entirely.

For the research programme and to help with the design of the anticipated future trial, two members of the *Speaking up for Health Group* and one member of the *Charnwood Action Group* wore the activity monitors (both wrist and waist-worn) and provided feedback on their ease of use.

4.5 Service user involvement during final stages of programme

The service user groups were involved in the discussions around disseminating the findings and identifying relevant conferences. During the consent process, participants were asked if they wished to be informed of the findings. As a means of supporting this, and to acknowledge the group homes that had allowed residents to take part in the study, two of the research nurses visited 27 homes to present the findings in an easy-read format. Other participants received an easy-read report posted to them.

4.7 Discussion

This chapter discusses the involvement of service users with ID into our research programme and draws on our own published work arising from this study.[202] In line with previous service user initiatives, the impact of involving service users in the research study is difficult to quantify.[203, 204] We feel that involvement of service users improved the quality of, and recruitment for, our study, but we do not know what would have happened had we not involved them and there are no similar studies in the UK on which to draw comparisons.

We can say with certainty that the team benefited from the involvement, developing a greater understanding of the health and personal issues faced by people with ID. The team also received positive comments from the service users, particularly in relation to being involved in the interview panel process and visiting our study offices. In line with previous research,[205] we found that people with ID valued the opportunity to discuss health issues. Unusually for service user involvement initiatives, our service users were also allowed to take part in the research (because it was a screening programme); the fact that many also chose to be participants in the programme is testament to their commitment.

The service users' involvement in the research programme was collaborative, and not participatory (or 'emancipatory'), which is favoured by many disability academics.[206] The agenda was set by the researchers and final decisions were always made by the lead investigator. Established self-advocacy groups contributed hugely to the success of the involvement, because there was an established group dynamic and all of the service users were keen to discuss health issues and voice their own opinions. As involvement initiatives expand, there is a danger that self-advocacy groups will become inundated with requests for support[207] so we need to ensure that we widen our approach to involvement for future studies. We also encountered problems when we discussed paying the service users for their contribution, because they were concerned about loss to their benefits, and also encountered organisational restrictions. For future work, we aim to consider more

innovative group payments, such as water coolers or coffee machines, with prior organisational approval.

We reiterate the recommendations from INVOLVE that involvement should commence at the early stage of the research process when identifying and prioritising topics for research.[208] We had limited involvement at this stage of the programme and further involvement is likely to have improved the quality of our application and reduced the need to make changes once the study had started. When involving people with ID, it is important to allow extra time for communication and consider their physical and/or psychosocial needs, which may include working outside normal hours, travelling to different locations, making suitable venue arrangements and considering the need for carers, advocates or supporters to be present. We also recommend approaching service users through a number of sources to minimise the burden of their involvement.

CHAPTER 5. SCREENING PROGRAMME: METHODS

5.1 Overview

This chapter describes the methods used for the screening component of the STOP Diabetes research programme included in work package 1. The background and rationale are presented in Chapter 1. The methodology for the cost-effectiveness, which also formed part of this work package, is described in Chapter 12. An additional physical activity sub-study, which was conducted alongside the screening, is described in Chapter 7.

5.2 Aims and objectives

The primary aim of the screening component of the research programme was to evaluate the feasibility and effectiveness of a diabetes screening programme for identifying undiagnosed T2DM and IGR in people with ID.

The specific objectives were:

- to develop and assess the feasibility of a diabetes screening programme in a community setting for adults with ID;
- to determine the prevalence and demographic risk factors for T2DM, IGR and cardiovascular disease in people with mild to profound ID;
- to validate the Leicester Self-Assessment diabetes risk score in people with ID;
- to establish data linkage to Hospital Episode Statistics and the Office for National Statistics.

5.3 Study design

Cross-sectional population-based screening study.

5.4 Study setting

The screening study was conducted between February 2013 and September 2015, in a variety of community locations within the unitary authorities of Leicester city, Leicestershire and Rutland (see Section 1.5.1 on approvals). Based on assumed familiarity and acceptability to service users, the locations initially chosen included day centres, community hospitals, primary care venues and group residential/nursing homes, which were identified through existing service listings. This was subsequently widened to include family homes and independent housing, to maximise recruitment.

5.5 Participants

5.5.1 Inclusion and exclusion criteria

Inclusion Criteria

1. Adults with ID
2. Aged 18 to 74 years inclusive
3. Registered with a general practice in Leicester city, Leicestershire or Rutland
4. Participant and/or carer have sufficient English language skills to enable fully informed consent to be obtained

Exclusion Criteria

1. Previous diagnosis of T2DM or T1DM
2. Disability not confirmed to be ID
3. Malignancy or life-limiting terminal illness
4. Severe systemic disease that may interfere with measurement and interpretation of HbA1c

5.5.2 Participant recruitment process

Eligible participants were invited to take part in the screening programme using a four-pronged approach (summarised in *Figure 14*).

1. Approach via general practice registers
2. Approach via the Leicestershire Learning Disability Register
3. Approach via specialist ID psychiatric service clinics
4. Direct contact with the research team

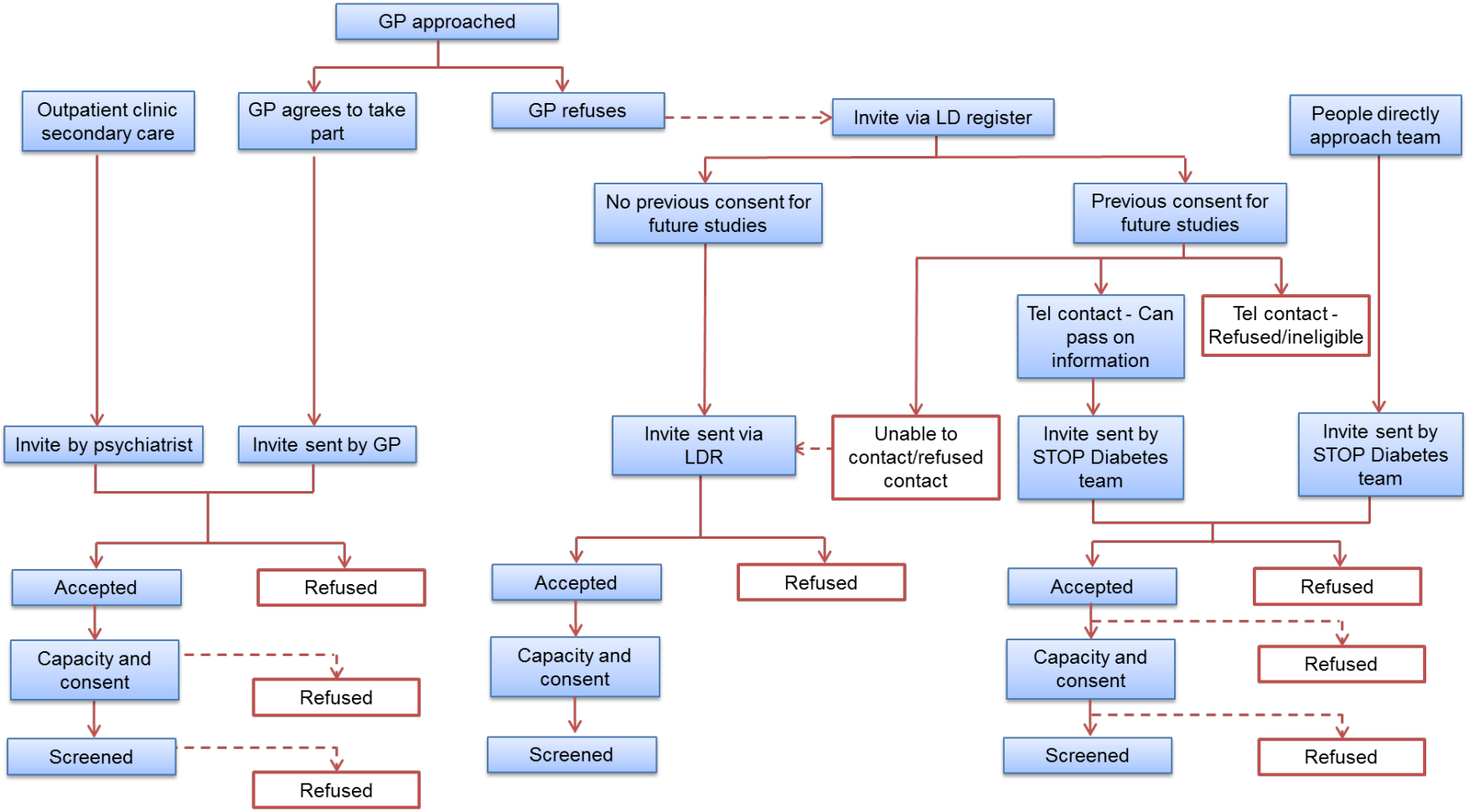


Figure 14: Recruitment pathway to screening programme

5.5.2.1 Approach via general practice registers

All general practices in Leicester city, Leicestershire and Rutland who had patients with ID on their practice register were sent a letter of invitation about the study. This was followed up, if necessary, by a postal reminder and/or telephone call. Practices were asked to return a reply slip to the research team to indicate their willingness to participate. The research team visited interested practices to explain the study in more detail, answer any questions and confirm their willingness to participate. General practice staff were then asked to identify people eligible to take part in the study from their practice ID register, and to send out a postal invitation.

To adhere to the requirements and underlying principles of the Mental Capacity Act,[209] information about the research was provided in stages. First, practices sent potential participants an easy-read invitation letter and a brief easy-read information leaflet, outlining the study. Potential participants were then asked to notify the research team of their willingness to participate (assisted by carers) using an easy-read reply slip and a freepost addressed envelope, or via the telephone. To be equitable to people with ID who could not read, lived alone, or lived with carers who also had reading difficulties, those who did not respond were followed-up with a telephone call. The aim of the call was to check if the invitation had been received, to briefly explain what the information was about and to establish if the person or their carer wished to find out more about the research programme. The telephone calls were initially made by practice staff; however, due to difficulties with practices prioritising the time to undertake them, approval was later sought for an ID research nurse employed on the study to make these telephone calls from the relevant general practice site. See Appendix 7 and 8 and 9 for examples of easy-read documentation used in the research programme.

Following this initial approach, a member of the research team telephoned interested people to discuss the study further. We anticipated that carers would play an important role in supporting the person with ID with their choice about participation; individuals were encouraged to talk to someone that they trusted about whether or not to participate. Potential participants received verbal explanations about the study,

had the opportunity to ask questions and received a preliminary assessment of their decision-making capacity to consent to participate in the research. Any indication of reluctance or anxiety about taking part was taken as a refusal. Full study information, in an appropriate format, was then sent to volunteers and/or an identified consultee (see *Figure 15*). If a personal consultee (i.e. a person interested in the person's welfare but not doing so for remuneration, such as a parent) could not be identified, a nominated consultee (e.g. a key worker) was identified and consulted. Alternatively, for some people, a face-to-face visit was arranged to facilitate the provision of further/full information, supplemented by additional communication aids/methods. See Appendix 10 and 11 for example of consultee information leaflets used.

5.5.2.2 Approach via the Leicestershire Learning Disability Register

Where general practices declined to take part in the study, potential participants were approached via the Leicestershire Learning Disability Register.[14] Adults who were known to the register were invited to participate following the pathways described in *Figure 14*.

1) The register operates a rolling programme of home interviews[210] and those who agreed to be contacted for research purposes at their most recent interview were contacted by the custodian of the register to confirm that these people were happy to be contacted by the research team. Their contact details were passed on directly to the research team for invitation.

2) People who did not agree to direct contact for research purposes (either due to lack of agreement at previous home interview, or refusal when approached by the custodian of the register, as above) were invited by the Principal Investigator in Leicestershire Partnership NHS Trust (LPT).

For both methods outlined above, potential participants were approached in the same way as for general practices (easy-read invitation letter, brief information

leaflet and reply slip to be returned to the research team). Invitations were cross-checked with those already sent via general practices to avoid duplicate invitations.

Non-responders were also followed-up in a similar manner as previously described for general practices; follow-up telephone calls were made by either the custodian of the Learning Disability Register or ID research nurses working on the research programme. A restricted-access database held on a SQL server was used to record and track whether the individual had received the letter of invitation and whether they would like any more information about the project.

Capacity assessment and provision of full study information, including involvement of carers and/or consultees, also followed the same process as previously described.

The sending of study invitations, via general practices and the Leicestershire Learning Disability Register, commenced in December 2012 and January 2013, respectively. Approval to utilise two further ways was obtained in February 2014 (see Sections 5.5.2.3 and 5.5.2.4 below):

5.5.2.3 Approach via specialist intellectual disability psychiatric service clinics

An additional approach to potential participants was made via specialist ID psychiatric service clinics. For patients attending a planned appointment, the consulting psychiatrist briefly described the research programme and issued an easy-read invitation letter, brief information leaflet and reply slip. Service users were given the opportunity to take the information leaflet and reply slip away with them (to return in the post) or to have their details passed on to the research team. Those who agreed to pass on their details were contacted by a member of the research team to provide further information and make an initial assessment of capacity.

Recruitment and capacity assessment then followed the same procedure as for general practices and the Leicestershire Learning Disability Register. As before, all potential participants were cross-checked against those already invited to ensure that they were not invited to take part in the study more than once.

5.5.2.4 Direct contact with the research team

In some cases, direct contact was made by eligible individuals with ID (and/or their carers) who had heard about the study via publicity materials or through other people who had taken part in the study. The STOP diabetes team provided them with the same initial brief written information, as described for the other recruitment sources. Recruitment, capacity and cross-checking procedures were similarly undertaken.

5.6 Screening process

Following the invitation stage, volunteers were asked to attend an initial screening appointment for consent (see Section 5.6.1) and data collection (see Section 5.6.2). Appointments were arranged by the research team at a time and location that was convenient to the participants (and carers), often early morning or late afternoon/evening in their own homes, but also in day centres, residential homes and primary care settings. The number and length of appointments was flexible to allow for participants' individual needs.

5.6.1 Informed consent

At the participant's first appointment, a final face-to-face capacity assessment was undertaken by a trained ID research nurse and informed consent was obtained; appropriate mental capacity legislation (see Section 1.5.2) was followed. Consent was taken only when it had been established that the person understood the consent form and information sheet and that they had been given the opportunity to ask questions.

People with capacity to consent were asked to sign a consent form. For those who could decide, but were unable to read, the consent form was read to them in the presence of an independent witness. For people who did not have capacity to consent, an appropriate consultee was identified and consulted about the person's potential participation. The consultee was asked to sign a consultee declaration form

confirming that they had been consulted, had their questions answered and had considered the study from the participant's perspective.

The participant and/or personal/nominated consultee (if appropriate) were asked to confirm that:

- they understood the study and were happy with what taking part would mean for them;
- they understood that they could withdraw from the study at any time, without giving a reason (and that this would not affect their care);
- they had been given a chance to discuss their questions with the research team;
- they agreed for their GP to be notified about their participation and of their screening results;
- the research team could access their medical records or records held at their residential home or day centre for additional information, if unable to obtain from the participant or carer;
- relevant sections of their medical notes and/or study data could be looked at by responsible people/regulatory authorities for purposes of auditing the research.

Participants provided their consent for screening to be undertaken (see *Section 5.6.1*and *Table 15*), including a blood test (where the participant was willing). Additional optional consent items that participants could chose to agree to or not included:

- to be contacted to take part in further phases of the study if they screened positive for IGR or high risk of developing T2DM (based on elevated BMI);
- to have an additional blood sample taken for storage and future anonymised genetic analyses;
- to allow access to medical records for long-term follow up;
- for contact details to be stored by the research team so that participants could be informed of the study findings and be contacted about future research studies.

At the end of the appointment, the nurse highlighted the office's telephone number on the participant information sheet to use if the participant decided to withdraw from the study or had any queries.

At the start of any subsequent appointments, participants' retention and understanding of the study was re-confirmed.

Following the final study appointment, a photocopy of the signed consent form/consultee advice form (as appropriate) was sent to both the participant and the general practice; the copy of the form accompanied the screening results, which were subsequently sent (see Section 5.6.3). The original consent and advice forms were retained at the research offices. See Appendix 12 and 13 and 14 for examples of the consent/advice forms.

5.6.2 Data collection

Data collection was usually undertaken over two appointments but could be longer (the maximum was five). The data collection process is summarised in *Table 15*. All data were collected in a standardised way by specially trained research nurses, following study specific standard operating procedures. Full details of the assessment of outcomes are described in Section 5.7.2.

Anthropometric measurements, BP, demographic and lifestyle data were frequently obtained at the first appointment, after consent was obtained, and usually took between one hour and 90 minutes. Questionnaires were completed via interview during the initial screening visit (or at a subsequent appointment), or were given to carers to be completed outside of the appointment, as applicable, see *Table 15*. These typically took between 30 to 60 minutes to complete. Venous blood samples were usually taken during a separate appointment after deciding with participants (and their carers, where relevant) whether a fasting or non-fasting sample would be more appropriate; this decision was based on potential behavioural difficulties and/or cognitive understanding of participants. This appointment lasted about 30 minutes. Medical history and prescribed medication were collected during screening or at a

later date from medical records. Other additional information was extracted by a researcher from the Leicestershire Learning Disability Register, or records held at residential homes or day centres.

5.6.3 Informing of screening results

All participants were informed of their key biomedical screening results in an easy-read format, supplemented by verbal explanations as appropriate. Anthropometric measures and BP readings were presented to participants on the day that they were taken. Results of blood tests taken to determine diabetes status were provided within seven to 10 days.

Participants with normal results were informed by post, with the option to contact the research team and discuss further if they wished. For participants who were screen positive for IGR or T2DM, a research nurse telephoned them to explain their results and answer any questions, prior to a letter being sent in the post. In some cases, this also involved a face-to-face visit by the nurse to support the participant and/or their carer. In accordance with consent taken, these participants were then referred to their general practice for usual care.

As agreed at the time of consent, participants GPs were provided with full details of the screening results, including diabetes status. Additionally, for all participants who were identified as meeting the criteria for IGR or T2DM, a member of the research team contacted their general practice and informed their GP, prior to any results letters being sent.

See Appendix 15 and 16 for example letters used to inform participants and GPs of the results.

Table 15: Summary of data collected during screening

Biomedical measures	
Bloods	**Anthropometric**
Plasma glucose (2.7ml fluoride bottle) [a]	Height (cm)
HbA1c (2.7ml EDTA bottle)	Weight (kg)
Lipids (total cholesterol, LDL, HDL, triglycerides[b]) [c]	BMI kg/m^2
Urea and electrolytes (sodium, potassium, creatinine) [c]	Waist and hip circumference (cm)
Liver function tests (Bili, ALT ALP, GGT) [c]	
Thyroid function (TSH, free T4) [c]	**Blood pressure** (mmHg)
Genetic sample – whole blood (9ml EDTA bottle)[d]	
Albumin: creatinine ratio (urine)	
Questionnaires	
Depression - Glasgow Depression Scale and Carer Supplement	Health related quality of life - EQ-5D
Problem behaviour - Aberrant Behaviour Checklist	Psychiatric disorders - PAS-ADD checklist
Demographic details	
Age; Sex	Ethnic background
Residential circumstances; level of support	Deprivation score
Medical and family history	
Cause of ID; severity of ID	Current medication
Medical history (physical, mental health, ID related)	Smoking status
Family history of diabetes (first degree)	
Lifestyle	
Physical activity	**Diet & nutrition**
Brief questions on mobility, walking, sitting and exercise	Brief questions on eating, food preparation, food groups, portions of fruit and vegetables
Activity levels	
Activity and sedentary behaviour	
Accelerometer – worn for 7 days[e]	

[a] glucose, fasting (8 hours) or non-fasting; [b] triglycerides, only requested if fasting;

[c] 1 bottle (4.9ml serum gel) used for all 4; [d] only if provided optional consent; [e] only for a sub-group.

5.7 Outcomes

5.7.1 Primary and secondary outcomes

The primary outcomes for the screening study were the prevalence of T2DM, IGR and abnormal (T2DM or IGR) blood glucose level.

Diagnosis of T2DM was made following the most recent WHO criteria,[28] more specifically HbA1c ≥48 mmol/L or 6.5%. IGR was defined as impaired fasting glucose (IFG), WHO criteria, or HbA1c 42-47 mmol/L or 6.0-6.4%, see *Figure 15*.

Secondary outcomes included:

- Physical activity levels, including sedentary behaviour, measured by brief questions and accelerometer (for a small sub-group);
- Lipid levels (triglycerides, total cholesterol, high density lipoprotein (HDL) cholesterol, low density lipoprotein (LDL) cholesterol);
- Blood pressure (BP; systolic, diastolic);
- Cardiovascular risk, as measured by the Framingham risk score;[211, 212]
- Health related quality of life, as measured by the EuroQol-5 Dimensions (EQ-5D) questionnaire;[213]
- Dietary/nutritional intake (food groups and fruit and vegetable intake);
- Behavioural disorders, as measured by the Aberrant Behaviour Checklist (ABC);[214, 215]
- Psychiatric disorders, as measured by the PAS-ADD checklist;[216]
- Depression, as measured using the Glasgow Depression Scale (GDS) and carer supplement.[217]

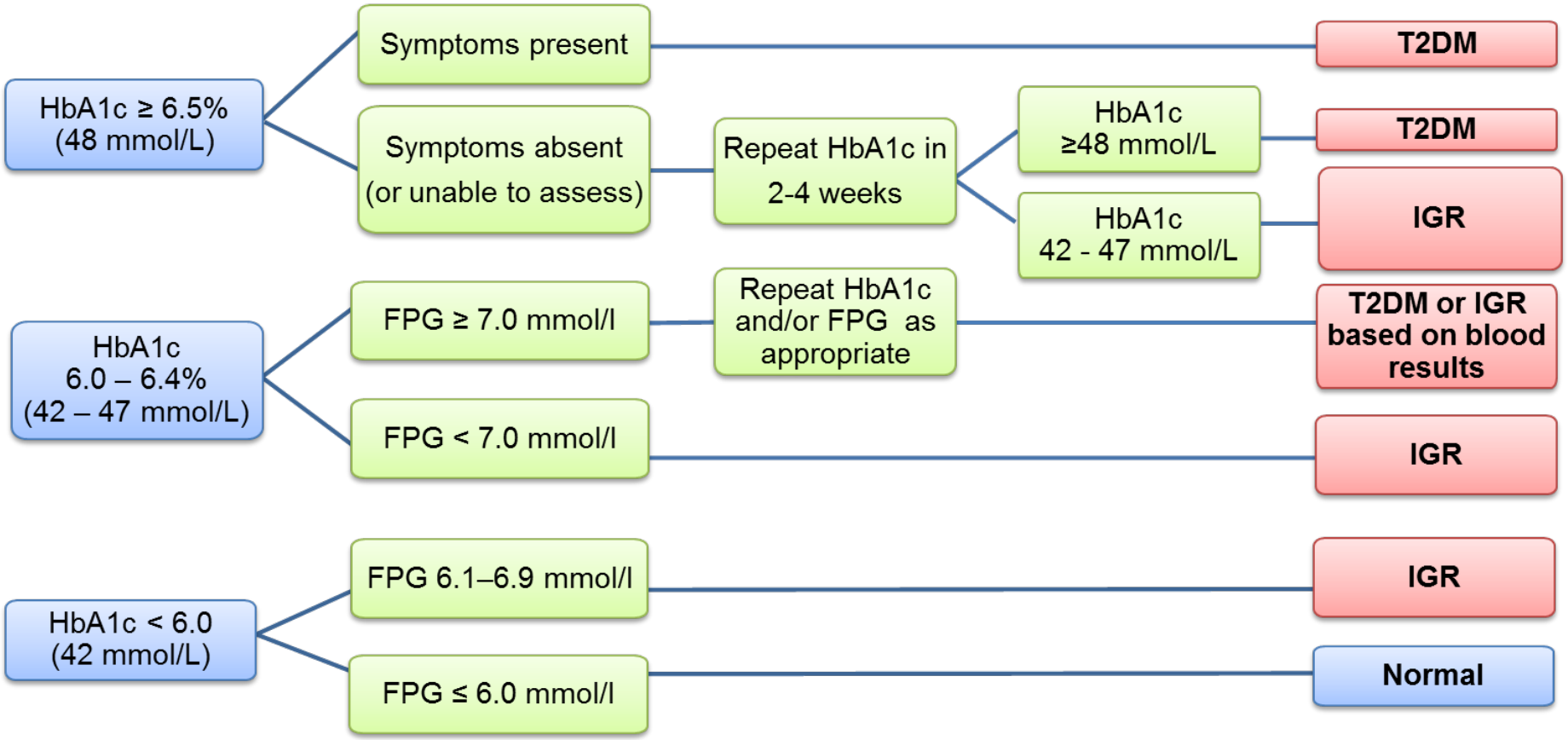


Figure 15: Flow chart to illustrate diagnosis of type 2 diabetes and impaired glucose regulation for participants in the screening programme

5.7.2 Assessment of outcomes

All blood and urine samples were analysed at by the University Hospitals of Leicester, NHS Trust laboratory services, using stable methodology standardised to external quality assurance reference values. HbA1c was measured using an ARKRAY ADAMS HA-8180T analyser (ARKRAY Inc, Kyoto, Japan). Plasma glucose (fasting and non-fasting); serum total cholesterol, HDL cholesterol and triglycerides; and urine albumin and creatinine, were measured using a Siemens Adiva 2400 analyser (Siemens Healthcare Diagnostics, Camberley, UK). The Friedewald equation was used to estimate LDL cholesterol.[218]

Resting BP was assessed in a seated position on the brachial artery, using an Omron M5-I automatic BP monitor (Omron Healthcare UK Ltd); a series of three measurements was recorded with a mean value calculated from the final two. Waist circumference was measured to the nearest mm over minimal clothing, midway between the costal margin and the iliac-crest, and in the mid-axillary line; hip circumference was measured to the nearest mm at the widest point over the buttocks; a soft tape was used for both anthropometric measures (WM02 Body Tape; Chasmors Ltd, UK). Weight was assessed in light clothing and no shoes to the nearest 0.1 kg, using a seca 875 digital floor scale (seca United Kingdom); and height to the nearest cm, using a Leicester portable height measure and with head placed in the Frankfurt plane.

Additional data on health related quality of life (EQ-5D)[213] and depression (GDS and carer supplement),[217] were collected face-to-face via interview administered questionnaires at an appointment. To assess problem behaviour (ABC)[214, 215] and psychiatric disorders (PAS-ADD checklist),[216] questionnaires were taken away by carers and self-completed following the appointment. The validated questionnaires used are described in detail in Appendix 17. Deprivation was assessed according to the 2015 Index of Multiple Deprivation (IMD).[219]

Ambulatory activity and sedentary behaviour were measured for a small sub-sample of participants; full details of the physical activity sub-study undertaken are presented in Chapter 7.

Uptake of screening was measured by recording the: (i) number of invitations sent; (ii) number of people responding and refusing at each stage; and (iii) number of people attending for screening.

If BP, anthropometric measures and/or bloods were unable to be assessed, details of the reason were recorded (refused; physical/behavioural difficulty; equipment error; other). For demographic, lifestyle, medical history and prescribed medication, additional details were recorded on how the data were obtained; for example, from the volunteer, carer/relative or a combination of both, or if personalised records such as a health action plan[4] were used.

5.8 Sample size

We aimed to screen 1,000 adults with ID, which would measure the overall prevalence of T2DM and IGR with 1.49% and 2.01% precision (95% CI) respectively, assuming similar prevalence rates of T2DM and IGR in people with ID as in the general population (6.2% and 12% respectively).[18, 21, 22]

5.9 Data analysis

5.9.1 Feasibility of diabetes screening in adults with intellectual disabilities

The feasibility of conducting a diabetes screening programme in a community setting for adults with ID was assessed using a flow diagram of the screening process and summarising the number of drop outs and those for which data were unobtainable at each step of the screening process. Particular outcomes of interest in terms of the feasibility are (i) the proportion of people invited who complete the screening programme (including the blood tests) (ii) the proportion of people who attended the

screening session but did not have a blood test. We also assessed the completeness of key data items from the CRF and questionnaire to assess the feasibility of data collection for future research projects in this group.

5.9.2 Characteristics

The characteristics of those screened were summarised using means (standard deviations for continuous variables) and n (%) for categorical.

Additional analyses were conducted to compare the representativeness of the STOP Diabetes study cohort to the Leicestershire Learning Disability Register.[14]

5.9.3 Prevalence of type 2 diabetes and impaired glucose regulation

The overall prevalence of IGR, T2DM and any abnormal glucose regulation was calculated with 95% (CI).

5.9.4 Cardiovascular risk

Cardiovascular risk was calculated for participants aged 35-75 years with no previous history of CVD. The Framingham CVD risk score[211, 212] was used to assess risk in White European participants and ETHRISK for South Asians.[220] Participants with incomplete data for key variables (total and HDL cholesterol, systolic BP, smoking status) were unable to be included in analyses. The overall mean risk at 10 years, and level of risk (high, intermediate, low) based on thresholds determined by National Cholesterol Education Program[221], were calculated.

5.9.5 Factors associated with abnormal glucose regulation

Logistic regression was used to assess the association between key biomedical and anthropometric characteristics and the outcome – abnormal glucose regulation. Odds Ratios (OR) and 95% CIs were calculated.

5.9.6 Validation of Leicestershire self-assessment risk score

Our initial analysis plan was to update the Leicester Self-Assessment risk score,[222] described in Chapter 1 (see *Section 1.6.5*), for use in an ID population. This may have included adding or removing risk factors and updating the relative weighting given to risk factors. However, given the low prevalence of IGR/T2DM we found in our screening study (see *Section 6.4*), this was not considered feasible. There are no formal sample size requirements for developing risk scores, although it has been suggested that data sets used to develop risk scores should contain between 10-20 events for each risk factor being assessed.[223, 224] Therefore, our data set would be very underpowered to develop a risk score.

Hence, it was decided that instead of updating the original Leicester Self-Assessment risk score, we would alternatively assess the risk score's performance to detect undiagnosed IGR/T2DM. Although this validation would also be underpowered (studies suggest that external validation data sets should have at least 100 events and 100 non-events),[225] this analysis should provide some preliminary results to suggest if the score is sensitive and specific in our cohort with ID.

The Leicester Self-Assessment risk score contains seven risk factors (age, sex, ethnicity, family history of diabetes, waist circumference, BMI and high BP).[222] To maximise the number of people included in the analysis, the data were analysed in two ways: (i) complete case basis (only including those with all seven risk factors recorded and the outcome); and (ii) imputing missing data for family history of diabetes and high BP. For both a family history of diabetes and high BP, the imputed data set assumed a negative response if these items were missing. In both data sets the sensitivity, specificity, positive predictive value, and negative predictive value were calculated for a cut point of greater than or equal to 16 points. This is the cut point used in the general population for invitation to screening.[222]

All analysis was conducted using Stata version 14 (StataCorp.), statistical significance related to $p<0.05$ and 95% CIs are presented throughout.

5.10 Establish data linkage to Hospital Episode Statistics and the Office for National Statistics

An additional optional consent item that participants were approached about at their initial screening appointment (Section 5.6.1) included consent for follow-up for health issues in the longer term.

5.11 Genetic markers

A supplementary component of work package 1 involved collecting blood samples for future genetic studies in individuals who had provided consent (optional). For this, an extra whole blood sample was taken and stored at -80 degrees centigrade. These samples will be analysed in a batch at the end of the study. Future work will involve extracting DNA and testing biologically plausible interactions between genetic markers and T2DM to determine T2DM susceptibility. The analysis of genetic markers does not form part of the work described in this report.

5.12 Concluding remarks

This chapter has described the methods for the screening component of the STOP Diabetes research programme. The following chapter (Chapter 6) presents the results of the screening study.

CHAPTER 6. SCREENING PROGRAMME: RESULTS

6.1 Overview

This chapter reports the results of the diabetes screening programme undertaken for work package 1. The methods for the screening study were reported in the previous chapter (see Chapter 5). An additional physical activity sub-study, which was conducted alongside the screening, is described in Chapter 7.

6.2 Feasibility of conducting a diabetes screening programme in adults with intellectual disability

6.2.1 Participant recruitment

6.2.1.1 Initial approach

Participants were recruited to the STOP Diabetes screening study between February 2013 and September 2015. In total, 3201 adults with ID were invited to take part via the four different routes (see *Figure 16*).

Fifty one percent (n=73) of all general practices in Leicester city, Leicestershire and Rutland (with adults with ID on their practice list) agreed to be involved with the study. Subsequently, 1736 potentially eligible people were identified and sent an invitation letter by their practice (median n=19, range 3 to 116). People invited via this route accounted for the majority of study invitations (54%).

For practices refusing, 1595 people were identified for possible approach via the Leicestershire Learning Disability Register. Of these, contact details for 418 people (who had previously agreed to be contacted for research purposes) were passed directly to the research team to invite (13% of the overall study total). A further 864 (27%) were invited directly by the principal investigator in LPT.

A much smaller proportion of people, were invited via specialist ID psychiatric service clinics or after making direct contact with the research team, 2% (n=52) and 4% (n=131) respectively.

6.2.1.2 Full stage invitation

From the initial invitation, approximately 30% of people refused, 29% were classed as non-responders, and 40% expressed an interest to participate in the study (*Figure 17*). Following a preliminary assessment of volunteers' decision-making capacity, 1209 individuals (38% of those initially invited) were then provided with full study information (postal invitation or a face-to-face visit). Subsequently, 984 (31%) proceeded to the screening stage.

For people who refused (or agreed) at the initial or full invitation stages, details relating to the method of recruitment and reasons for refusal, are presented in *Table 16*.

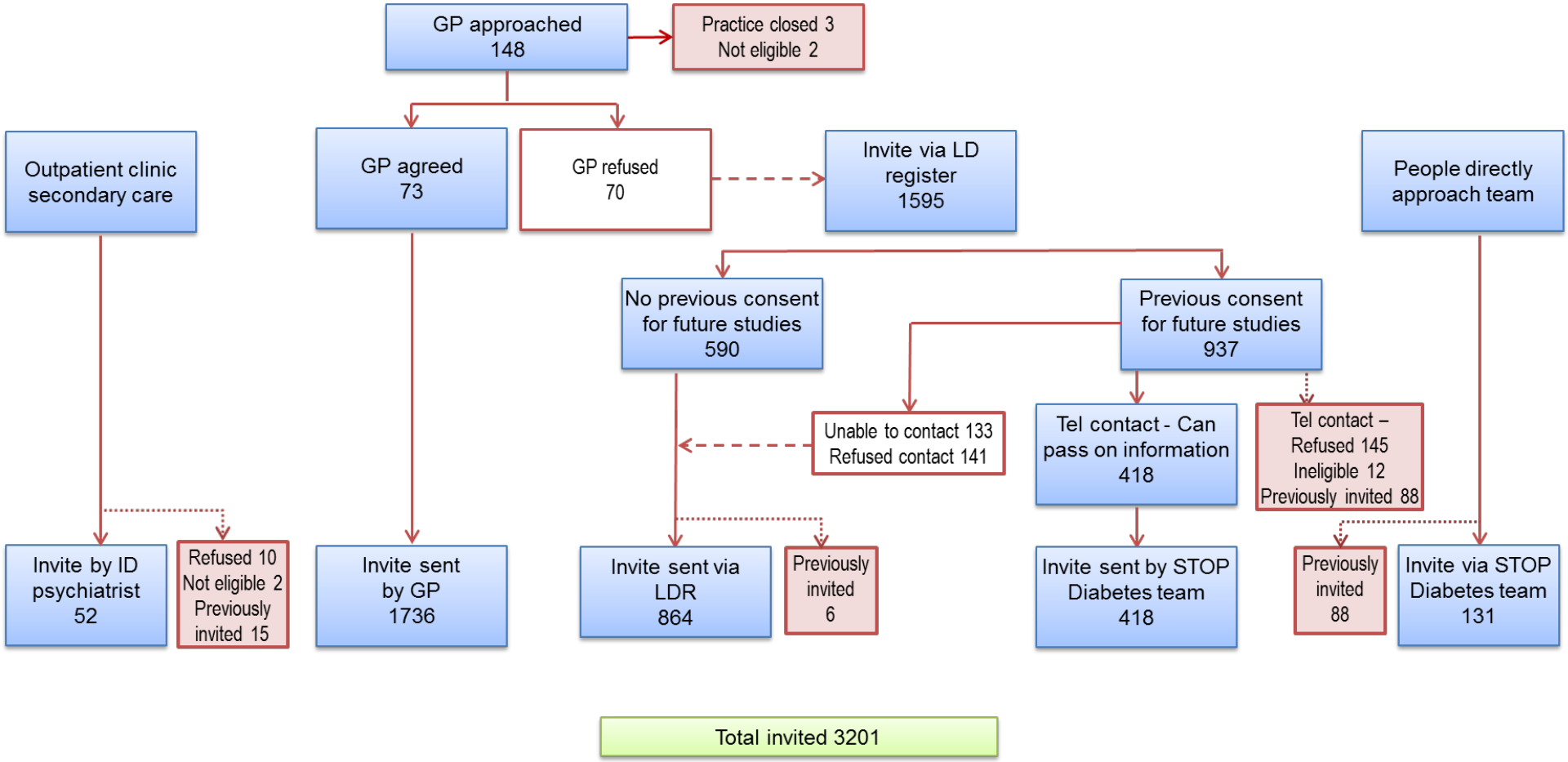


Figure 16: Invitation flow chart

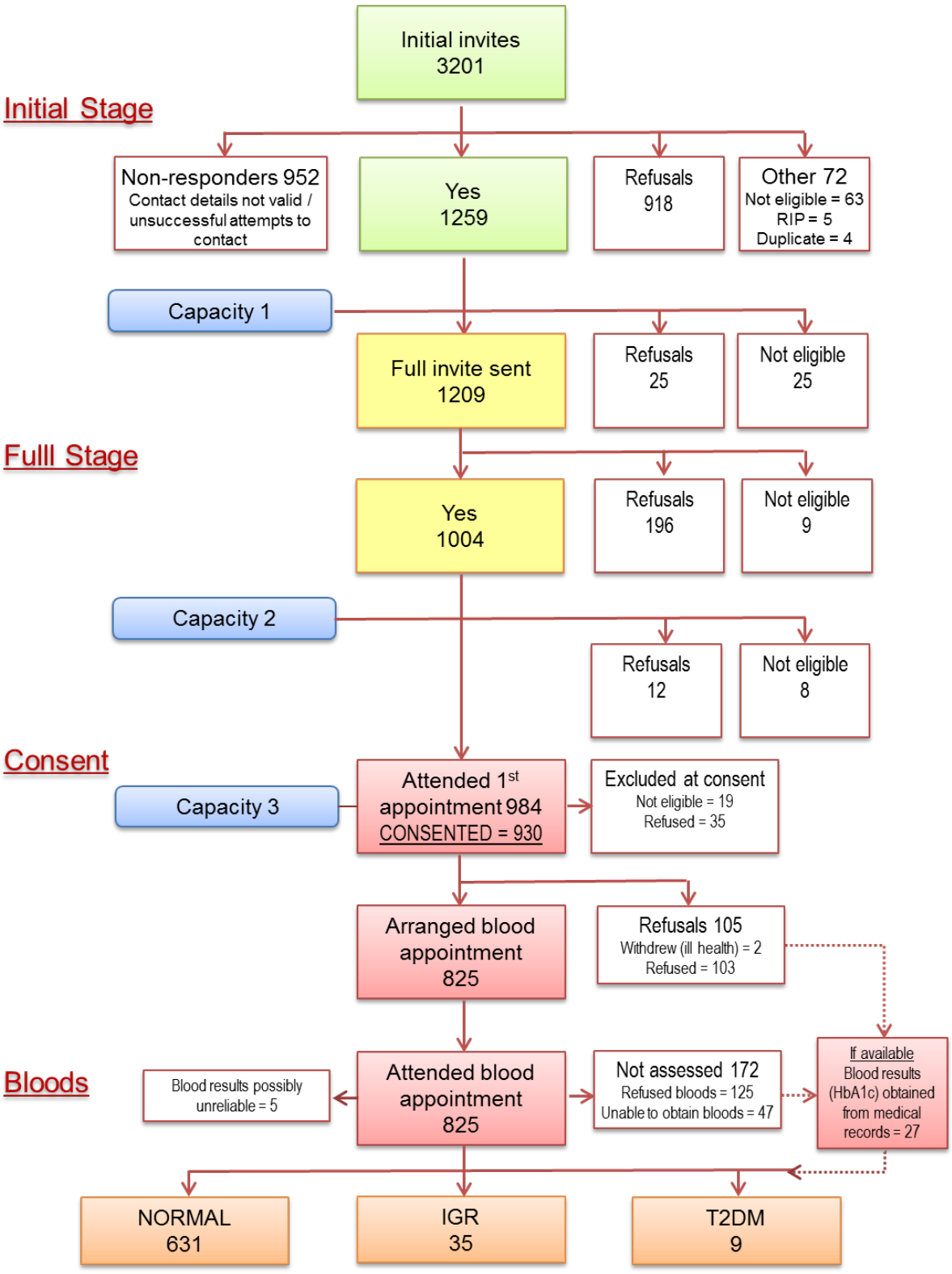


Figure 17: Flow chart of recruitment

Table 16: Responses according to recruitment method at initial and full invitation stage

		Initial invitation (or chasing non-responders)		Full invitation stage (or capacity 1 or 2)	
		Refuse	**Agree**	**Refuse**	**Agree**
Total number		**n =918**	n=1259	**n = 233**	n=984
Male, %		**53%** [a]	58%	**62%** [a]	58%
Age mean, years		**-**	43	**40** [a]	44
Where reside, %	Leicester City	**40%** [a]	41%	**48%** [a]	40%
Recruitment method	GP	**50%**	47%	**67%**	41%
	LD Register	**33%**	18%	**12%**	19%
	Previous consent to research	**15%**	19%	**17%**	20%
	Direct invite	**1%**	12%	**2%**	14%
	Psychiatrist clinic	**1%**	5%	**3%**	6%
Refusal/acceptance method	Reply slip	**28%**	28%	**24%**	28%
	Telephone call	**14%**	20%	**21%**	20%
	Chasing person via tel	**55%**	33%	**55%**	28%
	GP notified team	**2%**	0%	**0%**	0%
	In person	**1%**	18%	**1%**	24%
	Via email	**0%**	1%	**0%**	1%
Reason for refusing	Not Known	**77%**	-	**72%**	-
	Behaviour Issues	**7%**	-	**6%**	-
	Carer would not agree consent	**7%**	-	**5%**	-
	Health issues	**3%**	-	**4%**	-
	Recent Health Check	**3%**	-	**6%**	-
	Too busy	**2%**	-	**2%**	-
	Other	**1%**	-	**6%**	-

[a] estimates provided were appropriate for refusals; percentages rounded so may not add up to 100%

6.2.3 Screening – consent and data collection

At consent, 930 people (29% of those originally approached) agreed to participate and were recruited into the screening study; 54 people either refused (n=19) or were ineligible (n=35), see *Figure 17*. Thirty eight percent of participants (n=350) were able to consent for themselves; the other participants required a nominated (39%) or personal (23%) consultee.

The availability of data for the key screening outcomes is presented in *Table 17*. Full details regarding the availability of data for all study variables are reported in Appendix 18 (*Table 65*). Anthropometric measures and BP were obtained for most participants, approximately 86% and 89% respectively. In the majority of cases, the documented reason for not obtaining anthropometric measures was physical or behavioural difficulties; for BP, the main reason was participant refusal.

A high proportion of participants agreed to attend for a blood appointment (n=825), subsequently 700 (75% of those recruited) proceeded to have a blood test, and bloods to allow screening were successfully obtained for 648 (70%). For a few additional participants, where a blood test was refused or a sample not obtained, recent results were available from their medical records (HbA1c n=27; for other tests the amount varies), see *Figure 17* and *Table 17*. For a further five participants, HbA1c results were not included due to potential unreliability in assessing diabetes status (n=4, due to poor kidney function; n=1, possible Hb variant). Therefore, we were able to assess diabetes status for a total of 675 participants.

Validated questionnaires which were administered via interview, were successfully completed for a large number of participants (EQ-5D ~94%; GDS or carer supplement ~85%). Carer completion of questionnaires outside of the appointment (for the ~80% of participants who had an identified carer) was less successful; approximately 45% of carers completed the ABC and/or PAS-ADD.

Table 17: Data availability for key screening outcomes

Screening outcomes		Outcome measured n (%)	
Anthropometric	Height	800 (86.0)	
	Weight	799 (86.0)	
	Body mass index	782 (84.1)	
	Waist circumference	796 (85.6)	
	Hip circumference	789 (84.8)	
Blood pressure	Diastolic/Systolic	826 (88.8)	
Blood tests	Agreed to blood test	700 (75%)	
	Fasted for test – yes	*491 (70%)*	
	Bloods obtained	648 (70%)	
Blood results available		Taken for study	Obtained from medical records
	HbA1c	648 (69.7);	27 (2.9)
	Plasma glucose – fasting	417 (44.8);	8
	- non-fasting	223 (24.0);	16
	Total cholesterol	614 (66.0)	39
	HDL cholesterol	615 (66.1)	29
	LDL cholesterol	605 (65.1)	26
	Triglycerides*	404 (43.4)	3
Diabetes status assessed			
	Normal, High risk, Abnormal	675 (72.6)	
Validated questionnaires			
	EQ-5D score	872 (93.8)	
	EQ-5D VAS scale	877 (94.3)	
	Glasgow Depression Scale:		
	Volunteers with capacity	317 (34.4)	
	Carer Supplement	464 (50.2)	
	Aberrant Behaviour Checklist	341 (36.7)	
	PAS-ADD Checklist – Section 2	325 (34.9)	

6.3 Characteristics of the screened cohort

Key characteristics for the study population are presented in *Table 18, Table 19, Table 20* and *Table 21*. Full details for all screening variables are reported in Appendix 18 (*Table 65*).

6.3.1 Demographic characteristics

The mean age of those screened was 43.3 years (SD 14.2), 58% were male and the majority were of white ethnicity (80%), see *Table 18.*

Most participants lived either with family (36%) or in a residential/nursing home (38%), with 6% living alone. A high proportion required 24 hour support (71%) and only 7% reported being independent.

The majority of individuals were able to access the community to undertake regular daytime activities. Common activities included attending college (18%), voluntary work (16%) or involvement in service user/advocacy meetings (13%). Around half the participants attended day opportunities/day placements. Only a small number of people were in regular paid employment (8%).

6.3.2 Anthropometric and bio-medical measures

For those screened, the mean waist size was 100.4 cm (SD 16.5), weight 76.4 kg (SD 20.8) and BMI 28.7 kg/m^2 (SD 7.1), see *Table 19*. According to their BMI category, 31% of participants were classed as overweight and 37% obese. Mean values for systolic and diastolic BP were 121.4 mmHg (SD 16.9) and 78.2 (SD 11.1), respectively.

For participants where blood results were available, the mean HbA1c was 35.0 (SD 5.1) mmol/mol (5.3%; SD 1.5), fasting plasma glucose 4.7 mmol/l (SD 0.7) and non-fasting 5.3 mmol/l (SD 1.5). For lipids, mean total cholesterol was 4.9 mmol/l (SD 1.0), HDL cholesterol 1.3 mmol/l (SD 0.4), LDL cholesterol 2.9 mmol/l (SD 0.9) and triglycerides 1.4 mmol/l (SD 0.9).

6.3.3 Current medication and medical history

Where severity of ID was available (n = 816, 88%), similar proportions of participants were classified as mild, moderate or severe (~30% each) and 4% as profound ID, see *Table 20*. Most participants had no confirmed diagnosis or identified cause of their ID (~70%); where it was known, the most common causes were Down's syndrome (n=133, 14%) and cerebral palsy (n=58, 6%).

The overall prevalence of existing cardiovascular disease was 2% (n=19). A history of stroke was reported for 12 (1.3%) people, coronary heart disease for 6 (0.6%), and one person had a history of both conditions.

Congenital heart disease (2%) and other heart problems (2%) were less frequently reported.

Seventy-four participants (8%) had a history of high cholesterol and/or were prescribed a lipid lowering medication, 85 (9%) had a history of previously diagnosed hypertension and/or were prescribed an anti-hypertensive, and 36 (4%) were prescribed an anti-thrombotic. A minority of participants were either current smokers (8%) or ex-smokers (4%).

Where known, approximately one third of participants had a first degree family history of diabetes. Only one participant reported a previous diagnosis of pre-diabetes and one of polycystic ovary syndrome. Nine percent were currently prescribed a steroid medication (the majority were inhaled).

Overall, the most commonly reported diagnosed physical health problems were epilepsy 262 (28%), hypothyroidism 93 (10%) and chronic breathing problems 88 (9%). Thirteen percent of participants had no significant medical history and 19% were currently not prescribed any medication.

For mental health related problems, 152 participants (16%) had a history of a mood spectrum disorder (ICD-10 codes F30-F39), 35 (4%) a psychotic spectrum disorder

(ICD-10 codes F20-F29) and 52 (6%) had a history of both; 143 people (15%) had a neurotic, stress-related or somatoform disorders (ICD-10 codes F40-F48). Additionally, 28% of participants were prescribed antipsychotic medication and 32% depression or anxiety related medication. Other frequently reported problems included autistic spectrum disorders (18%) and a recognised behavioural problem (14%).

When co-morbidities (≥2 diagnosed health problems) were considered, 121 (13%) participants had co-occurring physical health problems, 182 (20%) co-occurring mental health problems, and 286 (31%) multiple physical and/or mental health problems.

6.3.4 Lifestyle and well being

Eighty-five percent of those screened were able to walk independently (without help/support of another person) but including 6% who required a walking aid, see *Table 21.* Data reported directly by participants and/or carers indicated that most people did at least “some” walking on a typical day but only 25% achieved “a lot” of walking. Additionally, around half of the participants reported spending “a lot” or “most/all” of the day sitting.

Sport/exercise or other physical activities that individuals reported undertaking in a typical week included dance (25%), swimming (20%) or walking (21%). Around half the participants reported doing housework (such as dusting/hovering) and ~20% gardening. A small number of people (7%) did regular chair based exercise.

Problems with eating and drinking were reported for some people, 24% had difficulties in chewing or swallowing and 13% needed help to feed themselves (<1% were tube fed). For food shopping and preparation, overall, ~35% of participants did their own food shopping (either independently or with some support), and a similar number were able to prepare at least simple hot and cold food (with or without supervision). Reported daily intake of fruit, vegetables or salad indicated that only around 30% of participants were eating the recommended five or more portions a day.

Where questionnaire data were available, the proportion of participants identified with possible depression (using a cut point of 13) by the GDS or GDS carer supplement was 22% and 16%, respectively.[217] For health related quality of life, the mean EQ-5D descriptive score was 0.8 (SD 0.3) and for the visual analogue scale was 78.1 (SD 19.4). The mean scores for the five problem behaviour sub-scales measured by ABC (for participants with carers) were ~4 for irritability, lethargy, and hyperactivity, and ~1 for stereotyped behaviour and inappropriate speech. The prevalence of mental health problems for organic, affective/neurotic and psychotic disorders (as measured by PAS-AD checklist) was 6%, 9% and 5% respectively.

Table 18: Key demographic characteristics of cohort screened

Demographic	N	Mean (± SD) Unless stated otherwise
Age (years)	930	43.3 (± 14.2)
Sex, Male, n (%)	930	537 (57.7)
Ethnicity, n (%)	930	
White		748 (80.4)
Asian		147 (15.8)
Black		14 (1.5)
Mixed		13 (1.4)
Other		8 (0.9)
Residential circumstances, n (%)	929	
Alone		51 (5.5)
Lives with family		338 (36.4)
Shared house or supported living		157 (16.9)
Shared care		16 (1.7)
Residential home or nursing home		350 (37.7)
Other		17 (1.8)
Level of Support, n (%)	929	
Independent		69 (7.4)
Some Support		205 (22.1)
24 hour support		655 (70.5)
Current status [a], n (%)		
Paid employment	928	71 (7.7)
Voluntary work	927	152 (16.4)
College	925	170 (18.4)
Day opportunities or private day centre	928	431 (46.4)
Shared lives (day placement)	928	19 (2.1)
Attending meetings	926	122 (13.2)
Other	924	385 (41.7)

[a] % will not add to 100 as participants can positively answer >1 category

Table 19: Key bio-medical measures of cohort screened

Biomedical Measurements	N Total (from medical record)	Mean (± SD) Unless stated otherwise
Bloods		
Plasma glucose		
Fasting (mmol/l)	425 (8)	4.7 (± 0.7)
Non-fasting (mmol/l)	239 (16)	5.3 (± 1.5)
Glycated haemoglobin	675 (27)	
HbA1c (mmol/mol)		35.0 (± 5.1)
Derived HbA1c (%)		5.4 (± 0.5)
Lipids		
Total cholesterol (mmol/l)	653	4.9 (± 1.0)
HDL Cholesterol (mmol/l)	644	1.3 (± 0.4)
LDL Cholesterol (mmol/l)	631	2.9 (± 0.9)
Triglycerides (mmol/l) [a]	407	1.4 (± 0.9)
Anthropometric Measurements		
Height (m)	800	1.6 (± 0.1)
Weight (kg)	799	76.4 (± 20.7)
BMI (kg/m2)	782	28.7 (± 7.1)
BMI Categories, n (%)		
Underweight		30 (3.8)
Normal		223 (28.5)
Overweight		241 (30.8)
Obese		288 (36.8)
Waist circumference (cm)	796	100.4 (± 16.5)
Blood Pressure Measurements	826	
Systolic (mmHg)		121.4 (± 16.9)
Diastolic (mmHg)		78.2 (± 11.1)

[a] only if fasted.

BMI, body mass index; HbA1c, glycated haemoglobin; HDL, high density lipoprotein; LDL, low density lipoprotein.

Table 20: Key medical history and current medication of cohort screened

Medical history and current medication	n (%)
Severity of ID, n=865	
Not known	49 (5.7)
Known	816 (84.3)
Mild	260 (30.1)
Moderate	244 (28.2)
Severe	279 (32.3)
Profound	33 (3.8)
Cause of ID, n=866	
Not known	581 (67.1)
Known	285 (32.9)
Downs syndrome	133 (15.4)
Fragile X	8 (0.9)
Cerebral palsy	58 (6.7)
Hydrocephalus	6 (0.7)
Phenylketonuria	5 (0.6)
Prader – Willi syndrome	4 (0.5)
Medical or Health problems, n=929	
None	117 (12.6)
Yes	812 (87.4)
Physical Health	
Stroke	13 (1.4)
Peripheral arterial disease	0
Coronary heart disease	7 (0.8)
Congenital heart disease	19 (2.1)
Other heart problems	15 (1.6)
High blood pressure	63 (6.8)
High cholesterol	62 (6.7)
Hypothyroidism	93 (10.0)
Polycystic ovary syndrome	1 (0.1)
Gestational diabetes	0
Pre-diabetes	1 (0.1)
Chronic breathing problems	88 (9.5)

Sleep apnoea	3 (0.3)
Epilepsy	262 (28.2)
Mental Health	
Dementia	18 (1.9)
Schizophrenia, schizotypal and delusional	35 (3.8)
Mood (affective) disorders	152 (16.4)
Neurotic, stress-related and somatoform	143 (15.4)
Personality disorders	13 (1.4)
Drug / alcohol problems	0
Attention Deficit Hyperactivity Disorder	8 (0.9)
Intellectual Disability related	
Autistic spectrum disorders	165 (17.8)
Behavioural problems	128 (13.8)
Current medication, n (%); n=928	
None	172 (18.5)
Yes	756 (81.5)
Anti-psychotic	240 (25.9)
Depression / Anxiety/ OCD or related	258 (27.8)
For ADHD	4 (0.4)
Anti-epileptic	311 (33.5)
Anti-thrombotic	36 (3.9)
Lipid lowering	74 (8.0)
Statin	72 (7.8)
Fibrate	1 (0.1)
Statin and Fibrate	1 (0.1)
Anti-hypertensive	85 (9.2)
Thyroid medication	93 (10.0)
Steroids	80 (8.6)
Oral	5 (0.5)
Inhaled	62 (6.7)
Topical	9 (1.0)
> 1 type of steroid	3 (0.3)
Not known	1 (0.1)
Anti-obesity	1 (0.1)
Other	571 (61.5)
Smoking status, n=929	

Current	76 (8.2)
Ex	38 (4.1)
Never	815 (87.7)
Family history of diabetes, n=592	180 (30.4)

ADHD, Attention deficit hyperactivity disorder ; OCD, Obsessive compulsive disorder

Table 21: Key lifestyle and well-being characteristics of cohort screened

Lifestyle and well being	n (%)
Physical Activity / Exercise	
Able to walk, n=927	
No	57 (6.2)
Yes (with or without walking stick, aid)	787 (84.9)
Yes, with assistance from person(s)	83 (9.0)
Amount of walking per day, n=927	
None	74 (8.0)
A short distance	259 (27.9)
Some	359 (38.7)
Lots	235 (25.4)
Amount of physical activity per week, n=928	
None	184 (19.8)
1-2 times	360 (38.8)
3-4 times	259 (27.9)
5 or more	125 (13.5)
Time spent sitting per day, n=928	
All / most	180 (19.4)
A lot	252 (27.2)
Sometimes	475 (51.2)
Never	21 (2.3)
Nutrition and diet	
Problems relating to eating and drinking, n (%)	
Difficulties with chewing or swallowing, n=929	227 (24.4)
Needs help or assistance to feed self, n=926	118 (12.7)
Use specialist equipment	95 (10.3)
Fed via an nasogastric tube or gastrostomy	7 (0.8)
Food shopping, n=922	
Independently	89 (9.7)
With support	230 (25.0)
Relative or carer	297 (32.2)

Purchased by residential home	306 (33.2)
Preparing meals, n=921	
Relative or carer	561 (60.9)
With supervision	117 (12.7)
Without supervision	145 (15.7)
Without supervision can prepare variety of meals	98 (10.6)
Daily portions of fruit and vegetables, n=920	
None	33 (3.6)
1 a day	57 (6.2)
2 a day	130 (14.1)
3 a day	230 (25.0)
4 a day	199 (21.6)
5 a day	213 (23.2)
6 a day	36 (3.9)
7 or more	22 (2.4)

6.3.5 Comparison with the Leicestershire Learning Disability Register

Participant demographic characteristics from this study were compared to adults with ID on the Leicester Learning Disability Register. Comparison of age, gender and ethnicity suggests that the STOP Diabetes cohort is a representative sample of the ID population known to services within the Leicester, Leicestershire and Rutland area, see *Table 22*.

Table 22: Characteristics of cohort screened compared to Leicestershire Learning Disability Register

Characteristics	STOP Diabetes (n=930) n(%)	Leicestershire Learning Disability Register <80 years (n=3867) n(%)
Age (years)		
<30	207 (22.3)	1012 (26.2)
30-39	195 (21.0)	856 (22.1)
40-49	211 (22.7)	776 (20.1)
50-59	185 (19.9)	659 (17.0)
60-69	107 (11.5)	416 (10.8)
70-79	25 (2.7)[a]	148 (3.8)
Male	537 (57.7)	2222 (57.5)
Ethnicity		**Of n=3571 known**
White	748 (80.4)	2893 (81.0)
S Asian	147 (15.8)	553 (15.5)
Black/Mixed	27 (2.9)[b]	80 (2.2)
Other	8 (0.9)	45 (1.3)

STOP Diabetes cohort: [a]age screened 18-74 years; [b]ethnicity data collected separately (Black n=14, 1.5%; Mixed n=13, 1.4%)

6.4 Prevalence of type 2 diabetes and impaired glucose regulation

Outcome data to establish prevalence of IGR/T2DM were available for 675 participants. Screening results indicated that, overall, 44 (6.5%) participants had abnormal glucose regulation, prevalence 0.07 (95% CI 0.05, 0.08); 9 participants (1.3%) were found to have undiagnosed T2DM, a prevalence of 0.01 (95% CI 0.005, 0.02); and 35 (5.2%) IGR, a prevalence of 0.05 (95% CI 0.04, 0.07), see *Table 23*.

Table 23: Prevalence of T2DM, IGR and abnormal glucose regulation

	N (%)	Prevalence	95% CI
Outcome:			
Normal glucose	631 (93.5)	0.93	(0.92, 0.95)
IGR	35 (5.2)	0.05	(0.04, 0.07)
T2DM	9 (1.3)	0.01	(0.01, 0.02)
Abnormal glucose	44 (6.5)	0.07	(0.05, 0.08)

6.5 Factors associated with abnormal glucose regulation

Table 24 shows the association of anthropometric and biomedical characteristics with having screen-detected abnormal glucose regulation. Participants of non-white ethnicity were almost four times more likely to have abnormal glucose levels compared to white European participants (OR 3.93 (95% 2.10 to 7.33)); those with a first degree family history of diabetes were over three times more likely (OR 3.35 (95% 1.64, 6.86)). In addition, abnormal glucose tolerance was associated with increasing weight, waist circumference, hip circumference, BMI, diastolic BP and triglycerides, and decreasing HDL cholesterol.

Table 24: Comparison of anthropometric and biomedical characteristics of those with normal and abnormal glucose regulation

	Normal Glucose (n = 631)	Abnormal Glucose (n=44)	OR (95% CI)	P value
Age, years	43.0 (±14.3)	45.4 (±13.5)	1.01 (0.99, 1.03)	0.27
Male	377 (59.8)	28 (63.6)	1.18 (0.63, 2.22)	0.61
Non-white ethnicity	119 (18.9)	21 (47.7)	3.93 (2.10, 7.33)	<0.0001
Weight, kg	76.6 (±20.2)	91.7 (±27.3)	1.03 (1.01, 1.04)	<0.0001
Waist circumference, cm	100.1 (±16.2)	114.0 (±19.0)	1.04 (1.03, 1.07)	<0.0001
Hip circumference, cm	107.4 (±13.5)	115.6 (±19.1)	1.03 (1.01, 1.06)	0.001
Body mass index, kg/m^2	28.6 (±6.9)	34.1 (±10.2)	1.08 (1.04, 1.13)	<0.0001
Current smoker	56 (8.9)	6 (13.6)	1.62 (0.66, 4.00)	0.30
FH of diabetes	132 (29.9)	20 (58.8)	3.35 (1.64, 6.86)	0.001
Systolic BP, mmHg	121.8 (±17.3)	126.5 (±14.4)	1.01 (1.00, 1.03)	0.09
Diastolic BP, mmHg	78.0 (±11.2)	83.7 (±10.0)	1.04 (1.02, 1.07)	0.002
Total cholesterol, mmol/l	4.9 (±1.0)	4.7 (±0.9)	0.78 (0.56, 1.10)	0.15
HDL cholesterol, mmol/l	1.4 (±0.4)	1.2 (±0.3)	0.14 (0.05, 0.43)	0.001
LDL cholesterol, mmol/l	2.9 (±0.9)	2.7 ±(0.8)	0.71 (0.48, 1.07)	0.10
Triglycerides, mmol/l	1.4 (±0.9)	1.9 (±1.0)	1.53 (1.11, 2.11)	0.01

Data given as mean (±SD) for continuous outcomes and n (%) for categorical.

BP, blood pressure; FH, family history; HDL, high density lipoprotein; LDL, low density lipoprotein.

6.7 Validation of the Leicester Self-Assessment risk score

Overall 365 (54%) of the 675 participants with the outcome obtained had complete data for the seven risk factors assessed by the Leicester Self-Assessment risk score. This was increased to 595 (88.1%) when imputing family history and high BP (*Table 25*). Similar percentages of participants fall into the four risk categories based on the complete case and imputed data. In the complete case data, 43.1% would be referred for screening based on their risk score (>=16 points) and 41.4% based on the imputed data.

Table 25: Completeness of the Leicester Self-Assessment risk score variables

Variables	All (n=675)	Complete Case (n=365)	Imputed (n=675)
Age (years)			
49 or younger	445 (65.9)	263 (72.1)	445 (65.9)
50-59	136 (20.2)	69 (18.9)	136 (20.2)
60-69	75 (11.1)	30 (8.2)	75 (11.1)
70 or older	19 (2.8)	3 (0.8)	19 (2.8)
Sex			
Male	405 (60.0)	207 (56.7)	405 (60.0)
Female	270 (40.0)	158 (43.3)	270 (40.0)
Ethnicity			
White European	535 (79.3)	278 (76.2)	535 (79.3)
Other ethnic group	140 (20.7)	87 (23.8)	140 (20.7)
Family History of T2DM			
No	324 (48.0)	248 (68.0)	523 (77.5)
Yes	152 (22.5)	117 (32.1)	152 (22.5)
Unable to assess	199 (29.5)	0	0
Waist Circumference			
Less than 90 cm	153 (22.7)	90 (24.7)	153 (22.7)
90-99.9cm	153 (22.7)	92 (25.2)	153 (22.7)
100-109.9cm	153 (22.7)	83 (22.7)	153 (22.7)

110cm or above	157 (23.3)	101 (27.4)	157 (23.3)
Unable to assess	59 (8.7)	0	59 (8.7)
Body mass index			
Less than 25	188 (27.9)	111 (30.4)	188 (27.9)
25-29	182 (27.0)	116 (31.8)	182 (27.0)
30-34	123 (18.2)	71 (19.5)	123 (18.2)
35 or above	109 (16.2)	67 (18.4)	109 (16.2)
Unable to assess	73 (10.8)	0	73 (10.8)
Antihypertensive medication			
No	525 (77.8)	325 (89.0)	609 (90.2)
Yes	66 (9.8)	40 (11.0)	66 (9.8)
Unable to assess	84 (12.4)	0	0
Total of complete data for			
Leicester Self-Assessment risk score	365	365	595
Final score			
Low (0-6)	-	63 (17.3)	112 (18.8)
Medium (7-15)	-	145 (39.7)	237 (39.8)
High (16-24)	-	121 (33.2)	193 (32.4)
Very high (25-47)	-	36 (9.9)	53 (8.9)

Data given as n (%)
T2DM, type 2 diabetes;

Table 26 presents the validation of the Leicester Self-Assessment risk score in this ID population. The complete case and imputed data have similar results, therefore, only the complete case data is interpreted here. Of the 22 participants with abnormal glucose regulation and full risk score data, 18 are correctly classified as high or very high risk by the risk score. This gives a sensitivity of 81.8%, given the low number of events the 95% CI around this estimate is wide 59.7%, 94.8%. Of the 344 participants with normal glucose regulation, 204 are correctly identified as low or medium risk and therefore would not be referred on for further screening. One hundred and forty participants would be referred for unnecessary screening, i.e. of those with a high or very high risk score only 11.4% have undiagnosed IGR/T2DM. The findings suggest that the score may be useful for ruling out disease; 98.1% of

those with a low or medium risk score are correctly identified and do not have undiagnosed IGR or T2DM.

Table 26: Sensitivity, specificity, PPV, NPV with 95% confidence intervals for the cut-off point ≥16 on the Leicester Self-Assessment risk score for predicting IGR/T2DM

	Sensitivity	Specificity	PPV	NPV
Complete Case Analysis (n=365)	81.8% (59.7%, 94.8%)	59.5% (54.1%, 64.7%)	11.5% (6.9%, 17.5%)	98.1% (95.1%, 99.5%)
Imputed Analysis (n=595)	83.3% (67.2%, 93.6%)	61.4% (57.2%, 65.4%)	12.2% (8.4%, 16.9%)	98.3% (96.3%, 99.4%)

6.8 Cardiovascular risk

Cardiovascular risk, based on Framingham[211, 212] or ETHRISK[220] for participants of South Asian ethnicity, was able to be calculated for 376 (40.4%) participants. The mean risk for CHD in 10 years was 6.1% (SD 5.4%) and for CVD 2.5% (SD 4.4%). Most participants were at low future risk for both CHD (81.9%) and CVD (95.2%), see *Table 27*. However, 18% of participants were of intermediate or high risk of developing CHD in the next 10 years.

Table 27: Ten year risk of CVD event - Framingham risk score

Risk	CHD	CVD
Low (< 10%)	308 (81.9)	358 (95.2)
Intermediate (10-20%)	59 (15.7)	13 (3.5)
High (>20%)	9 (2.4)	5 (1.3)

6.9 Establish data linkage to Hospital Episode Statistics and the Office for National Statistics

Of the 930 people recruited to the main study, 883 participants (95%) gave additional consent for the research team to follow-up their health in the longer term. Preliminary work to establish data linkage is currently being conducted.

6.11 Discussion

6.11.1 Summary of main findings

Utilising a variety of approaches to identify/invite potential volunteers, 930 adults with ID (29% of those approached) participated in the screening programme; 38% were able to consent for themselves, other participants required a consultee. Anthropometric measures (~86%) and BP (89%) were obtained for most participants. A high proportion of participants agreed to attend for a blood test and subsequently, prevalence of T2DM/IGR was assessed for 675 participants (73%).

The mean age of participants was 43.3 years, 58% were male and the majority of white ethnicity (80%). Most lived either with family (36%) or in a residential/nursing home (38%); a high proportion required 24 hour support (71%). Most participants were either overweight or obese; 2% had a history of existing CVD.

Screening results indicated the overall prevalence of undiagnosed T2DM was 1.3% (95% CI 0.5 to 2%) and IGR 5.2% (95% CI 4% to 7%). Participants of non-white ethnicity were almost four times more likely to have abnormal glucose levels compared to white European participants; those with a first degree family history of diabetes were over three times more likely.

6.11.2 Comparison with previous evidence

The prevalence of previously undiagnosed T2DM detected in the screening programme is much lower than previously reported.[90] Combined evidence from other studies, as presented in the meta-analysis in Chapter 2, suggests a prevalence rate of 8% for T2DM in adults with ID. Data to enable comparison of rates for T2DM in UK ID populations is scarce (a suggested 85-90% of diabetes is T2DM[20]). Current estimated prevalence of diabetes (type not specified) in England, based on combined data reported by partnership boards, is 6.8% (range 6.2-8.4%) for people with ID of any age.[51] Based on current data supplied by 40 (55%) of the general practices who took part in the STOP diabetes study, the suggested prevalence of

diagnosed diabetes (type not specified) locally is 9.5% (n=148 of 1553 adults 18-74 years with ID).

The estimates above are based on previously diagnosed diabetes. Our study aimed to screen adults with ID to identify undiagnosed T2DM. The rates suggested by data supplied by local general practices, alongside the higher recorded uptake of health checks locally (57-66% across the three CCGs)[226] compared to the national average (44%),[55] suggests at a local level the lower rate may simply reflect a successful annual health checks programme. In the general population, estimated prevalence of diabetes rises from 6.2% to 8.0% when including undiagnosed cases.[18] However, it is acknowledged that the proportion of adults with ID who currently have bloods checked, including for diabetes, as part of their annual health check is unclear.

6.11.3 Strengths and limitations

To our knowledge, this is the first diabetes screening study conducted in adults aged 18-74 years with mild to severe/profound ID. The successful integration of a multi-disciplinary team, consisting of experienced researchers and ID healthcare professionals, enabled the successful development and conduct of the STOP Diabetes screening programme. This multi-disciplinary approach allowed for sharing of knowledge and best practice, and was complimented by service user involvement, particularly in the early stages of developing and trialling study procedures/processes.

The screening programme developed utilised robust methods. All data were collected by staff who had undertaken study specific training and following standard operating procedures. Minimal exclusion criteria were applied for including people in the study, and reasonable adjustments to facilitate inclusion, such as staged invitation, easy read documents, flexible appointments, and carer involvement, maximised participation. This ensured as many people as possible participated rather than being arbitrarily excluded. Additionally, we applied a staged approach to invitation and made efforts to contact/chase all people where possible.

It is acknowledged that we were unable to establish any contact with approximately 30% of people, who were non-responders. We therefore do not know if they are different in any way to people included in the screening programme; evidence suggests that people with mild ID may be at increased risk due to unhealthier lifestyles but less likely to access services.[11] However, similarities in the demographic characteristics (age, gender ethnicity) of participants in this study, when compared with adults with ID on the Leicester Learning Disability Register, suggests that the STOP Diabetes cohort is a representative sample of the ID population known to services within the Leicester, Leicestershire and Rutland area.

The validation of the Leicester Self-Assessment risk score in the ID population was successful despite the limited number of events and wide 95% CI. Estimates suggest that the Leicester Self-Assessment risk score works as well in ID populations as in the general population, sensitivity 81.8%. Based on this 140 participants would be referred for unnecessary screening. However, the tool is designed for use in a multi stage screening programme and we would rather send more people through the first stage than falsely reassure.

6.11.4 Implications for clinical practice and future research

The screening uptake of 29% of those approached was relatively low but favourable when compared with two previous screening/prevention studies conducted locally in the general population, where 22% and 19% of those invited took part.[227] These relatively low rates of uptake might reflect that participants were invited to screening as part of a research project. If rolled out in clinical practice you would expect to see higher rates, for example uptake to the NHS Health Checks Programme,[228] which is not a research project, has seen uptake rates of double those reported in this and other research screening studies. Future research should focus on increasing uptake to screening in all groups.

Bloods to enable diabetes screening were successfully obtained for a high proportion of participants. However, future research may want to consider allowing for separate consent for blood tests so as to not deter people at the initial recruitment stage. Very

few people directly expressed “the blood test” as a reason for refusal to participate in the screening study; but anecdotal evidence suggests this may have deterred some. Alternatively, a staged approach to screening, involving risk stratification as recommended by NICE might be considered.[27]

Our findings suggest that the Leicester Self-Assessment risk score is statistically effective at identifying people with ID who are at high risk of undetected IGR/T2DM. However, the feasibility of using it in practice with people with ID, given the levels of heterogeneity within the ID population, needs to be considered. It may not be practical or acceptable for people with ID to calculate their own score, with or without added support from carers. Future research could involve developing an easy read version (plus a carer supplement) and additional supportive material/communication aids, such as digital audio/visual materials; qualitative research would be needed to supplement this work. Alternatively, a better way may be to integrate the risk score at practice level and incorporate into the Learning Disability Health Check.

6.12 Concluding remarks

This chapter presented the main results of the screening programme for work package 1. The methods and results of the physical activity sub-study are presented in the following chapter (see Chapter 7).

CHAPTER 7. PHYSICAL ACTIVITY SUB-STUDY

7.1 Overview

This chapter describes the physical activity sub-study, which was conducted alongside the screening component in work package 1. The main methods and results of the screening stage are described in Chapters 5 and 6, respectively.

7.2 Aims and objectives

The aim of this sub-study was to assess the feasibility of collecting physical activity data with the use of a waist worn accelerometer (ActiGraph), see *Figure 18.* However, given the poor uptake to this initial measurement tool, we extended our aim to also include the feasibility of collecting physical activity via a wrist worn device (GENEactiv).

7.3 Methods

7.3.1 Participants

Participants who met the eligibility criteria, as outlined below, were asked to wear an accelerometer as part of the main screening component of work package 1.

7.3.1.1 Inclusion criteria

Inclusion criteria

1. Consented to take part in the main screening component;
2. Able to walk without assistance (stick or similar walking aid permissible).

Figure 18: Waist worn and wrist worn acclerometers

(Waist-worn accelerometer, ActiGraph, Pensacolam FL, USA; wrist form accelerometer, GENEActiv, Activinsights Ltd., Cambridge, UK)

7.3.2 Participant recruitment process

Initial assessment of eligibility to participate in the physical activity sub-study commenced during the capacity assessment process (outline in Chapter 5) and was subsequently confirmed once consent to the main screening study had been obtained. Eligible participants were then approached about wearing an

accelerometer. For most people, this was usually at the end of their first screening appointment.

7.3.3 Data collection

Participants were asked to wear an accelerometer for seven continuous days, not including the appointment day. The procedure for wearing the accelerometer was explained to the participant and/or carer by an ID research nurse. Participants were also provided with a brief accelerometer information leaflet/diary in an easy read format, which explained how to use the accelerometer; this diary was also used to log when participants had worn the accelerometer, with a page for each of the seven days.

After wearing the accelerometer, participants were requested to bring it back to their next appointment. If a participant was not having another planned appointment, a member of the research team would contact them to arrange a convenient time for the accelerometer to be collected. In instances where an accelerometer was not returned or unsuccessfully collected, the research team made repeated attempts (at least 3) to try and retrieve it.

Two different accelerometers were used to collect data. Initially, physical activity data were measured using a waist-worn accelerometer. Later, it was additionally decided to trial a wrist-worn accelerometer, given the poor compliance that was emerging with the waist worn device (see results, Section 7.4) and following discussion with service users (who were assisting with PPI activities). The wrist-worn accelerometer was anticipated to encourage greater compliance since it is waterproof and can also be worn when sleeping; therefore, participants could wear it continuously over the seven day period.

Full details of the assessment of outcomes are described in Section 7.3.6.

7.3.4 Outcomes

Physical activity levels were included as one of the secondary outcomes for the main screening study (see Chapter 5). Other anthropometric (BMI, waist circumference), demographic (ethnicity and social deprivation) and biochemical (fasting blood glucose and HbA1c) outcomes assessed are described in Chapter 5.

7.3.5 Sample size

We initially aimed to include at least 50 participants wearing the waist worn accelerometer. This was updated to include a comparable number with the wrist worn device.

7.3.6 Assessment of physical activity outcomes

Participants attending screening were offered the option of having their physical activity levels assessed by a waist-worn accelerometer (ActiGraph, Pensacolam FL, USA). Once we had achieved our initial aim of at least 50 individuals with data, the remaining cohort were offered an alternative wrist form accelerometer (GENEActiv, Activinsights Ltd., Cambridge, UK). Details of the two accelerometers and analytic methods used are presented below.

7.3.6.1 Waist worn accelerometer

ActiGraph waist-worn triaxle accelerometers were attached to the trunk (placed on the right anterior axillary line) using an elasticated belt. Participants were asked to wear the accelerometer during waking hours for seven days, only taking it off at night when going to bed, or when partaking in water-based activities such as showering or swimming. Participants (and carers) were shown how to re-attach the accelerometer after sleep, and carers were asked to provide reminders. Data were set to record at 100Hz and analysed through a commercially available software package (KineSoft version 3.3.76; Kinesoft, New Brunswick, Canada; www.kinesoft.org). Data were converted to 60 second epochs and count based format. Time spent sedentary, in light-intensity physical activity and in MVPA were gained through applying commonly

used thresholds for adults.[229] Non-wear time was classified as 60 minutes of continuous zero counts.

7.3.6.2 Wrist worn accelerometer

The GENEActiv original wrist-worn triaxle waterproof accelerometer (Activinsights Ltd., Cambridge, UK) was worn continuously on the participants' non-dominant wrist for a minimum of seven days. Data were captured in 100 Hz and processed using two methods.

Data analysis method 1: raw acceleration data were converted to 60-s epochs using the GENEActiv Post-Processing PC Software (version 2.2, GENEActiv; Activinsights Ltd.). Next, the 60-s epoch data files were entered into an open source Excel macro (v2; Activinsights Ltd.) in order to classify activity. Subsequently, time spent in sedentary, light-intensity and MVPA activities was calculated for each participant-day using validated cut-points.[230] Sleep time was estimated using a defined algorithm (Activinsights Ltd) and subtracted from total sedentary time, in order to calculate time spent sedentary whilst awake.

Data analysis method 2: Given standard definitions for physical activity categories are lacking for wrist worn devices, we also included an alternative approach reported in the literature using the Euclidian Norm Method.[231] Data were proceeded in a freely available R-package (GGIR version 1.2-0, http:/cran.r-project.org) using previously described methodology to include time spent in sedentary, light-intensity physical activity and MVPA.[231, 232] In additional, total physical activity levels were reported in m*g*, where *g* = gravity.

7.3.7 Inclusion of physical activity data

Physical activity data were included from each device if there was a minimum of 8 hours wear per day for at least 3 days.

7.3.8 Data analysis

Data are presented as mean (standard deviation). Analysis of covariance models were used to compare differences in levels of assessed physical activity between monitors, adjusted for age, sex, social deprivation and wear time.

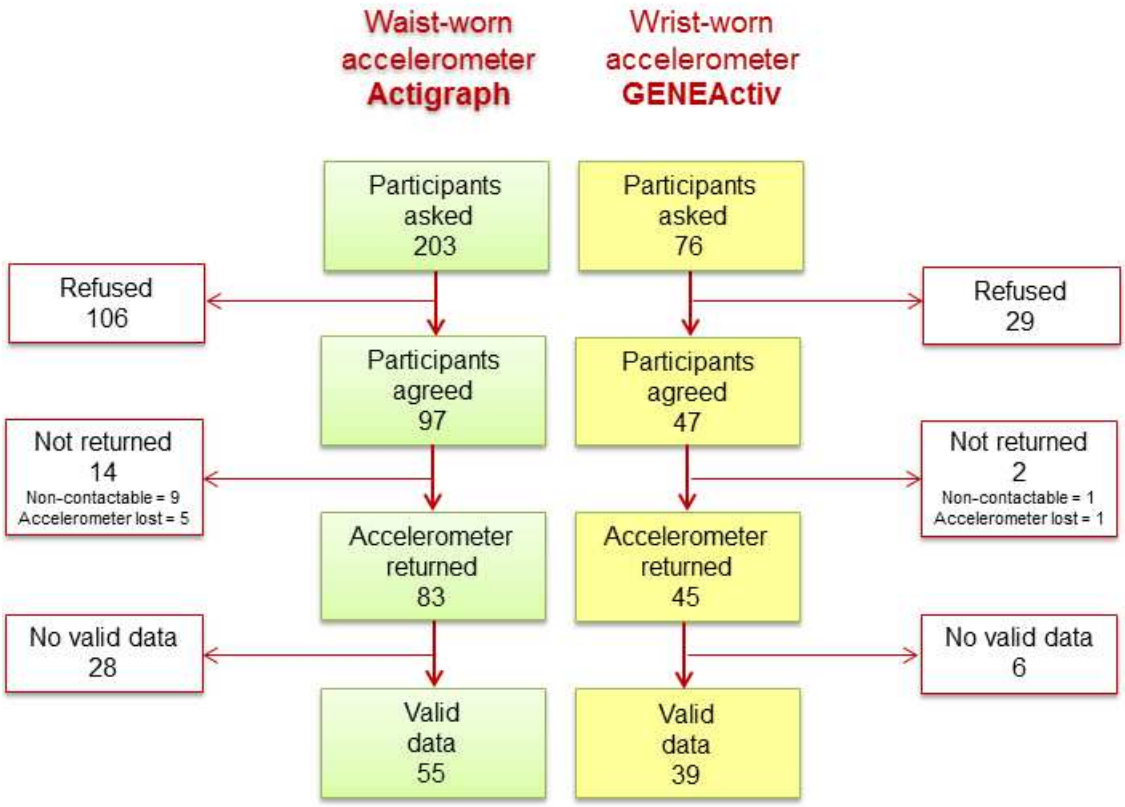
7.4 Results

7.4.1 Feasibility of using accelerometers to assess physical activity in adults with intellectual disabilities

Participants were recruited to take part in the physical activity sub-study between October 2013 and August 2015 (*Figure 19*).

Overall, 203 participants were approached to wear the ActiGraph waist-worn accelerometer. Subsequently, 97 participants (48%) agreed to wear the ActiGraph and valid data (≥8 hours/day for 3 days) were obtained for 55 participants (57%). Reasons for attrition included 14 participants (14%) not returning their accelerometer and 28 participants (29%) not having enough valid days of wear for analysis.

A total of 76 participants were asked to wear the GENEActiv wrist-worn accelerometer and 47 participants (62%) agreed. Valid data were obtained for 39 participants (83%). Two individuals (4%) did not return their accelerometer and six did not have enough valid days of wear (13%).



(Data validity was based on a minimum of 8 hours wear per day for at least 3 days)

Figure 19: Flow chart collection of accelerometer data

7.4.2 Characteristics of participants in physical activity sub-study

The characteristics of those who agreed to wear an accelerometer, either wrist or waist worn, and those with valid physical activity data stratified by accelerometer type are displayed in *Table 28*. Characteristics were similar between those who had valid physical activity data and those who did not. Characteristics were also similar between those who had valid waist worn and wrist worn accelerometer data. Overall, 54% of participants were male, mean age was 39.9 (SD13.0) and 85% were of white ethnicity. Thirteen per cent lived alone, 42% lived in supported living and 46% lived with family; the majority 88% had support from a carer for at least some of the time.

Table 28: Characteristics of participants in physical activity sub-study

Characteristics		Total agreed (n=144)	Total valid (n=94)	Waist agreed (n=97)	Waist valid (n=55)	Wrist agreed (n=47)	Wrist valid (n=39)
Gender, Male, n (%)		78 (54.2)	50 (53.2)	53 (54.6)	30 (54.6)	25 (53.2)	20 (51.3)
Age (years)		39.9 (±13.0)	41.9 (±13.7)	40.8 (±13.6)	43.6 (±14.7)	38.1 (±11.8)	39.3 (±11.8)
Ethnicity, White, n (%)		122 (84.7)	82 (87.2)	82 (84.5)	48 (87.3)	40 (85.1)	34 (87.2)
HbA1c (%)		5.3 (±0.4)	5.3 (±0.3)	5.3 (±0.4)	5.3 (±0.3)	5.4 (±0.3)	5.4 (±0.3)
Fasting plasma glucose (mmol/l)		4.7 (±0.5)	4.6 (±0.5)	4.8 (±0.5)	4.6 (±0.4)	4.6 (±0.5)	4.6 (±0.5)
Waist circumference (cm)		98.9 (±17.1)	98.4 (±16.6)	98.7 (±17.6)	97.9 (±16.2)	99.4 (±16.3)	99.1 (±17.3)
BMI (kg/m^2)		28.4 (±7.5)	28.3 (±7.1)	28.1 (±7.4)	27.7 (±6.2)	29.1 (±7.7)	29.0 (±8.2)
IMD 2015 Rank,		16,280	16,280	16,292	16,456	16,086	15,279
Median (IQR)		(7859.5-24,227.5)	(7734-23,871)	(7546- 24,572)	(7351-24,572)	(8815-23,871)	(7734-21,525)
Accommodation, n (%)	*Alone*	18 (12.5)	10 (10.6)	13 (13.4)	6 (10.9)	5 (10.6)	4 (10.3)
	Lives with family	66 (45.8)	46 (48.9)	44 (45.4)	27 (49.1)	22 (46.8)	19 (48.7)
	Supported environment	60 (41.8)	38 (40.5)	40 (41.3)	22 (40.0)	20 (42.5)	16 (41.0)
Support, n (%)	*Independent*	17 (11.8)	11 (11.7)	10 (10.3)	5 (9.1)	7 (14.9)	5 (12.8)
	Need support	127 (88.2)	83 (88.3)	87 (89.7)	50 (91.0)	40 (85.2)	34 (87.2)
Severity of ID, n (%)	*Mild*	62 (46.6)	39 (44.3)	38 (42.7)	17 (34.0)	24 (54.6)	22 (57.9)
	Moderate	39 (29.3)	27 (30.7)	29 (32.6)	19 (38.0)	10 (22.7)	8 (21.1)
	Severe/Profound	23 (17.3)	16 (18.2)	17 (19.2)	11 (22.0)	6 (13.6)	5 (13.2)
	Not known	9 (6.8)	6 (6.8)	5 (5.6)	3 (6.0)	4 (9.1)	3 (7.9)

Mean (± SD) unless stated otherwise; Data validity was based on ≥8 hours wear per day for ≥3 days;

7.4.3 Main findings

Estimates for time spent in MVPA, light-intensity physical activity and sedentary are presented across the different monitors and physical methods used (see *Table 29*). Estimates for MVPA and sedentary time were significantly higher with the wrist form device, whereas estimates of light-intensity physical activity were lower. Total physical activity volume measured by the wrist worn device was 26.7 (8.7) m*g*.

Table 29: Levels of physical activity and sedentary behaviour as assessed by the waist (Actigraph) and wrist worn monitors (GENEActiv)

Physical activity measures	**Waist worn**	**Wrist worn method 1**	**Wrist worn method 2**	**Wrist method 1 vs waist difference** adjusted for age, sex social deprivation and wear time or estimated waking hours	**Wrist method 2 vs waist difference** adjusted for age, sex social deprivation and wear time or estimated waking hours
Time in MVPA (mins/day)	33.6 (30.8)	136.9 (79.9)	95.8 (51.8)	<0.001	<0.001
Time in light-intensity physical activity (mins/day)	269.1 (72.7)	105.7 (47.1)	195.1 (73.7)	<0.001	<0.001
Time spent sedentary (mins/day)	499.2 (96.7)	632.5 (136.4)	790.8 (116.1)	<0.001	<0.001
Ambulatory activity (steps/day)	6761 (3483)	N/A	N/A		

Data validity: Wrist method 1, based on ≥8 hours wear per day for ≥3 days; Wrist method 2, based on ≥16 hours wear per day for ≥3 days;

7.5 Discussion

The key finding from this sub-study was that the objective measurement of physical activity is likely to be challenging in adults with ID with high levels of non-compliance; however, compliance can be substantially improved and loss of accelerometers reduced with wrist worn monitors. Overall, less than 50% of participants agreed to wear the waist worn device, with valid data only collected for 57% of the sample. In contrast, 62% agreed to wear the wrist worn device with 83% providing valid data.

To our knowledge, this is the first study to access the feasibility of collecting objectively assessed physical activity data in those with ID. However, other studies have reported high levels of missing data when using objectively measured physical activity within their study protocol.[164, 233] These results suggest that studies that include accelerometers may have poor uptake unless participants are allowed to consent separately for this element. These factors will need to be taken into account and considered carefully in future physical activity intervention studies within this population.

To assist with compliance in our study, participants (and carers) were provided with a physical activity diary (instructions) in an easy read format. Service users were involved with the development and initial testing of the diary; however; no formal assessment was conducted to see if the diary increased compliance for participants (and carers). Given the heterogeneity in capacity levels and support needs of individuals, further work is needed to explore possible ways to improve compliance with accelerometer wear in people with ID.

Based on estimates from the waist worn device, our population engaged in more MVPA than several other studies conducted in those with ID. For example, studies from Scotland and the United States have reported between 7 to 14 minutes per day of MVPA.[164, 233] Estimates for MVPA from the waist worn device were also slightly higher than levels reported in a primary care cohort from Leicestershire, UK.[234] Similarly, estimates for total physical activity from the wrist worn device were consistent with those reported for healthy non-obese adults and higher than those

reported for obese or unhealthy populations within the United Kingdom.[235] However, a previous research study in the United Kingdom in those with ID reported similar levels to those found in our study.[236] This suggests that in the United Kingdom, those with ID are not less active than the general population. This is despite institutional barriers that have been hypothesised to inhibit physical activity engagement in those with ID.[237]

An important finding from this sub-study was the difference in activity levels gained from wrist and waist worn devices. Whilst waist worn devices have been widely used in research over the last decade with established methods of categorising collected data which allows for comparisons between studies, wrist worn devices are newer and lack standardised approaches to data analysis. Although the underlying raw acceleration data between waist and wrist worn monitors are likely to be highly correlated, commonly used methods of converting this data into meaningful outputs, such as time spent in MVPA, are likely to be monitor and placement specific. This has important implication for future trials and suggests that intervention effects, standard deviations and population means should be estimated using data gained from the same tool that will employed in the study.

7.6 Concluding remarks

This chapter has described a physical activity sub-study, which formed part of work package 1. The following chapter (Chapter 8) describes the first phase of the education development process that was carried out as part of work package 2, to develop an initial curriculum for a lifestyle education programme for adults with ID.

CHAPTER 8. DEVELOPMENT OF INITIAL CURRICULUM FOR STRUCTURED EDUCATION PROGRAMME

8.1 Chapter overview

This chapter describes the work undertaken for work package 2 to develop an educational programme for a population with ID and IGR or high risk of developing diabetes and/or CVD (based on increased BMI level). A brief overview of the complete development process is presented in Section 8.3. The remainder of this chapter details the work conducted to develop an initial curriculum. Further development work, including two pilot cycles of testing, evaluation and modification, is described in Chapter 9. An additional feasibility phase, which formed part of work package 2, is presented in Chapter 10.

8.2 Aims and objectives

The aim of work package 2 was to develop a structured lifestyle education programme for prevention of T2DM that is suitable for use in an ID population.

Specific objectives were:

- First, to develop a lifestyle education programme for a population with ID who have IGR or are at a high-risk of developing T2DM and/or CVD based on increased BMI level (Chapters 8 and 9);
- Secondly, to assess the feasibility of collecting outcome measures for participants with ID before and 3-months after they attend the education programme (Chapter 10).

8.3 Overview of the development process

A multi-disciplinary team with expertise in ID and in the development of nationally recognised diabetes and CVD prevention programmes developed the intervention. A

systematic approach was employed (see *Figure 20*), based on the current Medical Research Council framework for developing and evaluating complex interventions[238] and intervention mapping.[239] This included reviewing the relevant published evidence from existing programmes and the behaviour change literature. The curriculum was informed by previous prevention programmes that our research group has developed.[240-242] Additional qualitative work was undertaken to further inform the content, process and style of delivery.

Following development of an initial curriculum, two cycles of testing, evaluation, modification and re-testing were conducted during the pilot phase (presented in Chapter 9), prior to the programme being used in a third iteration where the feasibility of collecting before and after data was explored (presented in Chapter 10). This iterative and reflective process, supplemented by qualitative research methodology, is an approach that our group has previously used successfully to adapt patient education modules for different groups.[240, 243, 244]

The core multi-disciplinary team, which included ID nurses, education team members, a qualitative researcher and the lead study researchers, met monthly throughout all stages of the development, supplemented by more frequent meetings at key points in the process. The purpose of these meetings was to decide on the key elements relating to the content, process and style of the initial curriculum, and subsequently reach agreement on any modifications required. This collaborative multi-disciplinary approach allowed the expertise of all members to be used and facilitated the iterative and reflective process.

This development work occurred over a period of approximately 27 months, commencing in October 2012 and ending in January 2015 when the final refinements were made to the curriculum (ready for use in the feasibility phase described in Chapter 10).

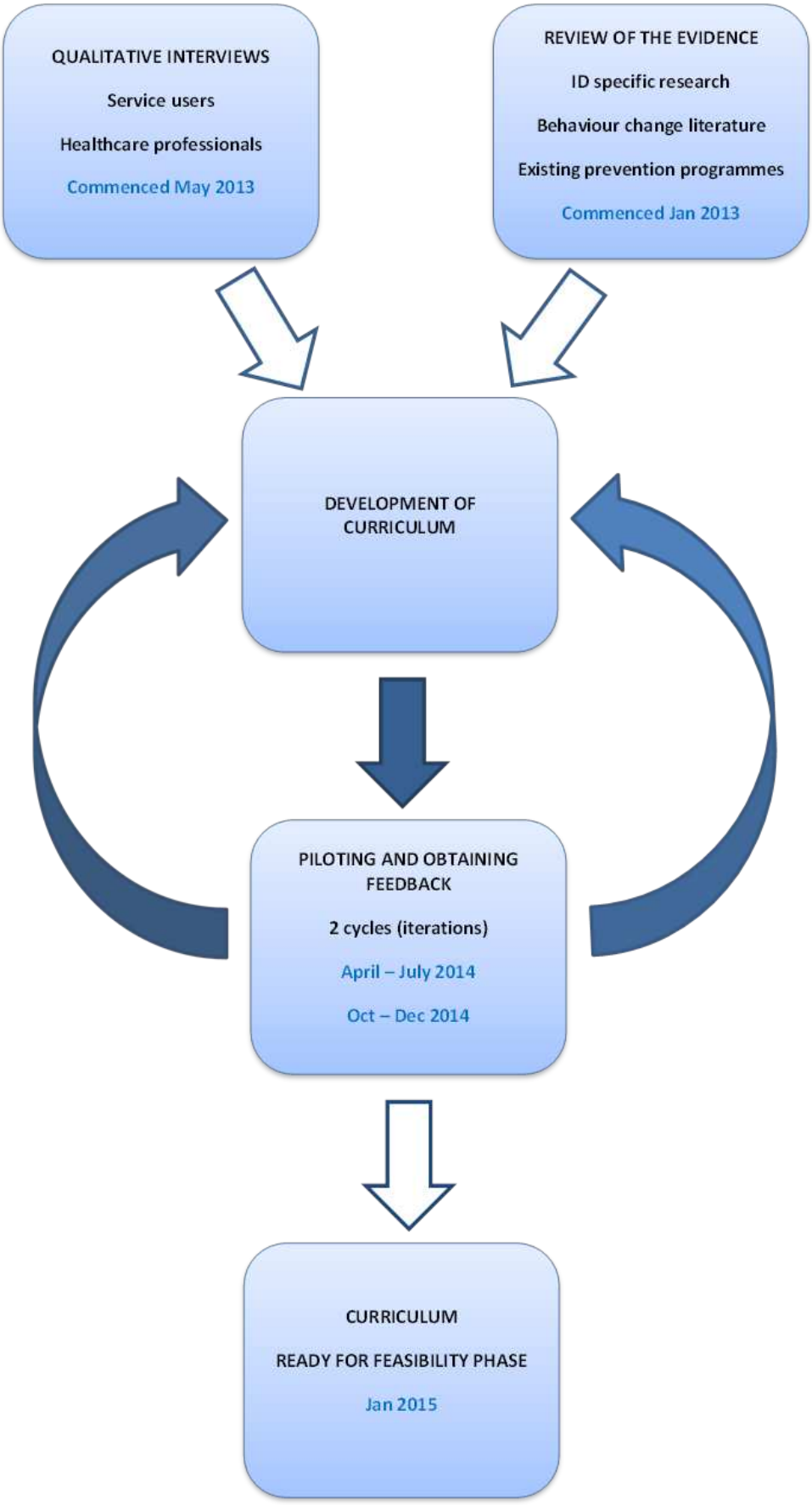


Figure 20: How phases of the development work fit together

8.4 Participants

People invited to engage in work package 2 (qualitative interview, Chapter 8; pilot education sessions, Chapter 9; or feasibility testing, Chapter 10) were service users with mild to moderate ID who had taken part in the screening stage (see Chapter 5) and screened positive for IGR or had a BMI ≥25, and at that time consented to being approached to assist with later phases of the research programme. Carers were also approached. An invitation pack, including easy read documents, was sent directly by the research team. For people volunteering, capacity assessment and consent followed a similar process to previous stages (see Chapter 5).

For people with ID who were invited to assist with the qualitative exploratory interviews (see Section 8.5), no further eligibility criteria applied. Additional inclusion criteria for invitation to attend the pilot education sessions and give feedback (see Chapter 9), or the feasibility phase (see Chapter 10), included:

- able to stand and walk at least short distances;
- able to attend group education sessions;
- not taking part in any other intervention study

ID healthcare professionals (HCPs) were also invited to contribute/assist with development of the initial curriculum by agreeing to a qualitative interview. Further details are provided in the next section (Section 8.5.1).

8.5 Qualitative work to inform development - methods

Semi-structured qualitative exploratory interviews were conducted with service users (and carers) and HCPs providing services to adults with ID, to help inform development of the initial curriculum. This qualitative work was carried out between May 2013 and June 2014.

8.5.1 Recruitment for interviews

A provisional quota was set of conducting up to 25 interviews with the various stakeholders (HCPs and service users with ID), to enable a range of views to be captured.

Recruitment of HCPs commenced in May 2013. A variety of HCPs were identified through ID services at Leicestershire Partnership Trust (LPT). The invited HCPs all had previous experience of working with adults with ID. Purposive sampling was used to ensure inclusion of HCPs who could offer a range of perspectives based on their occupation/professional background. Potential interviewees were sent an invitation pack.

For service users (and carers), recruitment began in January 2014. The eligibility criteria and method of approach are described previously in Section 8.4.

8.5.2 Data collection and recording

Topic guides were developed to ensure relevant issues were captured (see Appendix 19 for service user example). Interviews were semi-structured and based on open questioning to elicit issues surrounding knowledge, understanding and experience of T2DM and modifiable risk factors, relevance of IGR, perceived barriers to behaviour change and support needs for people with ID. Practical aspects in the delivery of an education programme were also explored, for example, whether to develop separate interventions for carers (family members and/or key workers) and people with ID, inclusion of follow-up sessions and length of the programme.

Questions were asked appropriately depending on who was being interviewed. Additional communication tools were used when interviewing service users, such as prompts cards depicting images of various activities (e.g. swimming, bowling, walking), to help with eliciting contributions.

Interviews were conducted with HCPs between June and August 2013. All interviews were conducted by an experienced qualitative researcher at the HCPs normal place of work.

Interviews with service users were conducted from January to June 2014. There was an initial delay in finding service users who were either eligible to be invited or willing to be approached/interviewed. All interviews were conducted by the same qualitative researcher with assistance from an ID research nurse. The researcher had expertise in developing and modifying diabetes prevention programmes for different populations; prior to commencing the interviews the researcher had undertaken additional training within the research team to increase their knowledge and skills in the area of ID. The interviews were conducted in a variety of community settings, including at a participant's family home, a residential/care home, an assisted independent living flat and a community clinic, to suit the needs and preferences of individual participants.

8.5.3 Data analysis

Audio recordings of interviews were transcribed verbatim and thematic analysis was conducted using NVivo version 7, QSR (a qualitative software programme). Subsequently, themes relevant to the development of the intervention were identified.

8.7 Qualitative interviews – findings

8.7.1.1 Characteristics of participants

Service users

Eighteen service users were invited to participate. A total of seven service users were subsequently interviewed (see *Table 30*); in two of the interviews carers were present (one a family carer, one a care worker). Three of the service users who participated were male, the median age was 47 years (range 28-68), and all were of White European ethnicity. Six participants lived in a supported environment with family or carers, and one lived independently. One of the participants was in paid employment (and did voluntary work), two attended college, two others carried out voluntary work, and the remaining participants undertook other activities within the community on a regular basis.

Healthcare professionals

Twenty HCPs were invited to participate. Subsequently, 14 HCPs were interviewed. All HCPs currently worked with adults with ID for all or part of their job role. Professionals included: ID psychiatrists, nurse related roles (community/primary care ID nurse, practice nurse, acute liaison nurse, nursing assistant); allied healthcare professionals (clinical psychologist, occupational therapist, speech and language therapist), and a day centre manager (*Table 31*).

Table 30: Characteristics of service users who were interviewed

	Characteristics service users	n = 7
Age	18 – 39 years	3
	40 – 59 years	3
	60 – 74 years	1
Sex	Male	3
	Female	4
Accommodation	Alone	1
	With family/carers	3
	Residential home	3
Level of support	Independent	2
	Some support	2
	24 hour support	3

Table 31: Characteristics of healthcare professionals interviewed

	Characteristics HCPs	n = 14
Age	20 – 39 years	**5**
	40 – 59 years	**7**
	unknown	**2**
Sex	Male	**2**
	Female	**12**
Profession	ID Psychiatrist	**2**
	Allied healthcare professional	**5**
	Nurse related	**6**
	Other	**1**
Length of time working ID	<5 years	**1**
	6 - 10 years	**2**
	10+ years	**9**
	unknown	**2**

8.7.1.3 Key points from interviews with service users and carers

Interviews conducted with service users ranged between nine to fifteen minutes in duration.

In a few of the interviews it was possible to explore awareness of diabetes. Service users related this to “sugar”, and also spoke about family members who had diabetes and recalled them being on tablets and having injections. When trying to gauge service users’ knowledge about healthy lifestyles, some people were able to describe basic health messages such as eating vegetables, eating a high fibre diet and exercise.

The interviews did yield some useful insights into the lives of service users, for example the types of food they enjoyed, and the degree of choice and control available in relation to foods consumed; discussions about commonly consumed foods ultimately influenced the food images and food models in the dietary sections of the curriculum. For a few participants, the additional use of prompt cards enabled useful discussion around the types of physical activities undertaken; for those who were more independent, walking appeared to be the most preferred and accessible form of physical activity.

It was difficult to explore service users’ preferences towards learning as part of a group or learning on an individual basis. However, the majority of participants spoke about going to some form of group activity sessions, such as sessions held at a local day care centre or at a college; activities included arts and crafts, and learning “life skills” to facilitate independence. Further discussion around participants preferences for photographs or pictorial images (on educational resources), suggested that for most participants they preferred photographs.

More general points arising from the interviews included practical considerations to be taken into account. First, that education sessions needed to be held locally (minimal travelling distance/time for participants), in a setting that was familiar to people, and a venue that was easily accessible via public transport or similar.

Secondly, the importance of including carers within education sessions, to help support participants and make them feel at ease during sessions, and to help facilitate service users with making changes to their diet and physical activity outside of sessions; it was clear from the interviews that both professional and family carers currently fulfilled this role in the daily lives of participants.

8.7.1.4 Key points from interviews with health care professionals

All HCPs were enthusiastic to share their knowledge and experience of working with people with ID. Most HCPs had previous or current experience of promoting positive behaviour change with people with ID, either for behaviour management and/or health promotion.

Pre-assessment

The majority of HCPs stressed the importance of undertaking a pre-assessment prior to embarking on delivery of an education session. Frequently stated reasons relating to carrying out a pre-assessment included:

- to enable cognition matching, which would involve the assessment of preferred communication styles, reading and writing abilities, preferences for working with pictures and/ or written sheets/flip charts;
- to ensure any differences in severity of ID (mild to moderate) between individuals in the group are not too wide;
- to identify and support people who may face challenges or difficulties with verbal communication (for example, some people may only be able to say yes or no);
- to prepare people for taking part in a programme and working in a group setting;
- to gain a measure of the level of insight a person may have about their own health and the perceived relevance of the programme to themselves;
- to assess a participant's ability to identify and engage with their own priorities, and reflect on their own skills for undertaking change or wanting to change;
- to assess how best to support individuals with decision making.

Suggested activities relating to what pre-assessment could involve were:

1. speaking to the person with ID (and their carer(s) as appropriate) and carrying out an assessment via discussion/interview using established tools (questionnaires & checklists) or observations;
2. extracting relevant information from health action plans and core information;
3. eliciting relevant information and knowledge from staff teams involved with the person with ID.

<u>The process and delivery of the programme</u>

1. Preparing the group for learning

Ensuring that participants are in the right frame of mind or in the ***"best place to learn"*** (HCP 07) requires some thought and preparation; one HCP described some of the strategies they used to promote this during a "healthy living" course. These included participants having two to three short breaks over every one hour period, or undertaking physical activity or encouraging them to be physically mobile during the education sessions.

> ***"Because if you get them in the wrong place or they're not in at the right level of arousal or even in the right mood, this can impact on their willingness and their ability to take in information." (HCP 07)***

Other ways in which participants' receptiveness to learn was developed were through watching videos or taking part in practical fun activities.

2. Choosing methods to promote healthy choices

To deliver knowledge and promote healthy food choices, participants described using visual aids including photos and pictures of foods from magazines. They also discussed the importance of undertaking practical activities such as preparing healthy foods. The rationale for these kinds of sessions was to show alternatives in a very literal way. Additionally, to try to convey that too much of a particular food was

bad for your health, you would need to show actual or pictorial images from real life, not cartoon images.

> ***"……rather than saying 'too many biscuits', which are words. You want to show pictures of biscuits and you want to show one pack, plus two pack, plus three pack is this much. Stuff like that. So real pictures, or even better real objects." (HCP 04)***

In relation to the number and type of messages during a session, the advice from HCPs was to keep it simple and not to over load the amount of messages covered in one session.

3. Ways of promoting physical activity

Effective ways to convey messages about increasing levels of physical activity, that were suggested by some HCPs, included giving an opportunity for participants to experience some of the activities during the education sessions (for example, swimming or going for a walk). If this was not possible, another suggestion included using pictorial images to stimulate discussion on how physical activity could be integrated into someone's life. This would need to take into account individual needs, such as restrictive budgets, physical ability and level of independence. For some people, if going for a walk was not possible, alternatives may be skipping or dancing to music.

4. The use of open ended questions

When asked specifically whether the use of open ended questions was appropriate for adults with ID, most HCPs went on to describe using this style of questioning with service users. However, they emphasised the need to follow up this approach with specific and direct questions. The latter helped to ensure questions were not 'too open' and in danger of being misinterpreted. For example, one HCP explained:

> ***"So sometimes open ended questions can be too open. You have to be more specific, like ……. for ground rules - "What is going to keep us all safe amongst ourselves?" - Not talking about slips, trips. Do you***

see what I mean? That you probably do have to tailor it a little bit…" (HCP 03)

Some HCPs also suggested that educators should not assume that commonly used words will always be understood by participants. They emphasised the importance of eliciting understanding on a frequent basis throughout the session and checking for consistency of responses. Other recommended strategies (particularly for those with autism) included, giving two options or choices and changing the order of these to check their selection is based on an informed understanding.

One suggested disadvantage of asking open ended questions was that it could place undue pressure on some individuals, invoking feelings of distress because they may not know the answer; educators may need to use pictures to encourage a response.

5. Retention and recall

To aid retention and recall of messages, the general advice was to use a combination of visual and verbal communication, with opportunities to experientially learn. The need to cater for differences in attention span, types and levels of abilities, and styles of learning, was emphasised. The key message of the interviews was that a flexible approach is needed including: 1) educators gauging understanding at regular intervals and addressing appropriately; and 2) educators using different methods to facilitate delivery to cater for diversity within a group.

6. Health beliefs and behaviours

When specifically asked, the majority of HCPs felt that exploration of health behaviours may be challenging. For adults with ID, their ability to process thoughts and associate them with behaviours, or make causal links at a more complex level, may be lacking in some individuals. The latter, they believed, may partly be influenced by the environment in which people live; service users may have limited or restricted opportunities to be in contact with (or be aware of) other people with a health condition. There may also be a lack of control about dietary choices and/or their association with health conditions. One participant felt, that simple associations

could be made such as *"**too much sugar is not good"**,* and that these have the potential to inform changes in health behaviour. (HCP 04)

A divergent view was that people with ID are not any different to the general population in relation to holding health beliefs. It may just be the communication of their beliefs that is different and necessitates educators taking different approaches, or that their beliefs may be more unusual/idiosyncratic.

7. Understanding the concept of future health risk of developing diabetes and self-reflection

When HCPs were asked whether people with ID are likely to understand the concept of risk, there was a variation in the responses. This appeared to be related to views on the heterogeneous nature of the ID population and also possible perspectives linked to the professional background of HCPs.

One view was that people with mild ID may understand the concept of risk, but generally, people with autism may have difficulties. However, it was additionally felt that would depend on an individual's attitude or motivation.

> ***"You're going to have some people who do understand that things change in the future, and things may deteriorate. Then you may have other people who wouldn't have that concept at all. Particularly you know if you've got somebody with autism and the future doesn't really mean a great deal, because it's not concrete enough for their understanding and perception quite often. So I think it would depend on the level of learning disability and many other conditions that the individual might have." (HCP 06)***

A few HCPs discussed that people with ID may have difficulty with understanding risk, as they may have a cognitive impairment that challenges the ability to conceptualise, including projecting into the future.

> ***"I think those sorts of things are more difficult. A lot of the time we probably are used to working in the here and now. So yes, projecting***

that this might happen to somebody, I think a lot of people find that difficult, don't they, to understand." (HCP 03)

However, this participant acknowledged that there was also evidence that shows that it is:

"…possible for people with a learning disability to be able to handle abstract information, reflect on it, appraise it and therefore bring change, but that is probably best done by people skilled in offering those interventions." (HCP, 04)

Some HCPs suggested ways to explain the concept of future risk, but in amongst these suggestions was a view that it had to be a balancing act between alerting people and not scaring people:

"It's really tricky cos you don't want to scare people, and people can get fixated on something and worry about it, and worry about it. And it could become a bit of an obsession, and they could be really worried and scared about that." (HCP 02)

The potential dangers of the above approach were discussed with another HCP who suggested anxieties could be allayed by discussing the future with ***"positive bits."*** (HCP 06)

One of the suggestions put forward for helping to promote self-reflection included using "DVD clips" to show alternative scenarios and facilitate non-threatening reflection (i.e. it was not about them). Although, even with this approach, it was acknowledged that it would take many weeks of guided discussion and support to help facilitate this process.

8.8 Drafting of initial curriculum

8.8.1 Key points for initial curriculum from the literature

At the start of the curriculum development process described in this chapter, a structured literature review was conducted with a focus on existing lifestyle interventions for adults with ID, aimed at primary prevention of T2DM and/or CVD or modification of risks factors. This was supplemented by reviewing relevant published guidelines, consensus statements, interventions currently in practice and service evaluations.

Later, this was formalised by conducting a systematic review considering evidence on the effectiveness of multi-component lifestyle behaviour change interventions for reducing risk factors for T2DM and/or CVD. The methods and findings of the systematic review are presented in Chapter 3.

Key findings from the literature that directly informed the content, theory and process of the programme are described below. There were only a small number of studies with a focus on people with ID and behaviour change lifestyle interventions. Few of these studies[163-166] provided a description of their theoretical underpinning, although they did recommend the employment of social cognition models such as the Theory of Planned Behavior[245] and Reasoned Action.[246, 247]

The Healthy Lifestyle Change Programme, which was developed by Bazzano et al,[163] was the only published intervention at this initial stage in the development of the STOP programme that outlined a conceptual model. Thus the STOP theoretical conceptual framework, as shown in *Figure 21*, was influenced by this approach.

The STOP framework developed, subsequently informed all aspects of the education programme; the framework highlights the importance of an individual's beliefs about health, ill health and its consequences, specifically the impact on them as individuals and their life. In terms of 'attitude', outcome expectancies were explored, i.e. what would happen if I engage in a particular behaviour and how important is the outcome

for me. Methods of learning, specifically vicarious, observational and concrete kinesthetic were also highlighted in the literature, as was the importance of social support and peer norms.[163, 245-248]

Self-efficacy is a key component of behaviour change,[248] i.e. the person's belief they can perform the behaviour. However, there are many real barriers to behaviour change in this population, such as disability and/or a lack of control over their physical environment, for instance not being the person who buys or cooks the food. Therefore, the concept of actual behavioural control was included in the theoretical framework to ensure that these issues were addressed in the programme. The influence of strong intentions and a detailed action plan was also acknowledged. Intrinsic motivation and the power of reinforcing feedback loops were also highlighted via distal and proximal re-enforcers and the positive impact on quality of life and psychological well-being. These specific components of the programme can be viewed in *Table 32*.

Additional key lessons from the literature were: 1) the need to maximize carer involvement; 2) that people with ID have extremely heterogeneous needs and any intervention would require a multi modal approach; and 3) that pre-group preparation is essential.

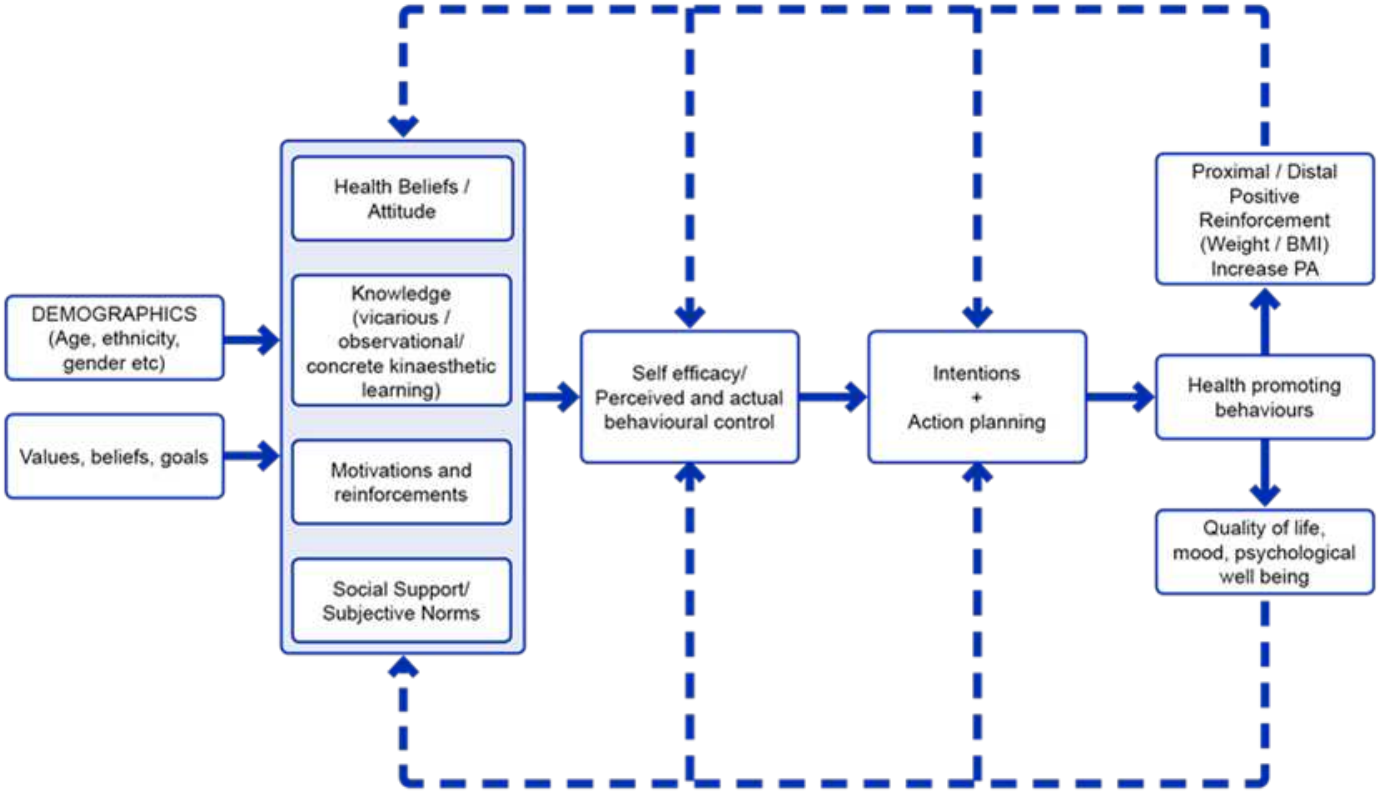


Figure 21: Theoretical framework for the education programme

8.8.2 Key points from multi-disciplinary development group

The overarching framework, content, process and learning methods for the programme were formed at a large multi-disciplinary meeting following a systematic process; this meeting was additional to the regular monthly meetings held throughout the development process. The qualitative findings from HCP interviews, relevant literature and core theoretical constructs were presented, debated and a consensus was formed. This was later supplemented by findings from the qualitative interviews with service users (and carers), once available.

The core multi-disciplinary team, which included ID nurses, education team members, a qualitative researcher and the lead study researchers, met monthly throughout all stages of the development, supplemented by more frequent meetings at key points in the process.

Key points agreed included:

- using a concrete kinaesthetic learning style;
- ensuring that resources developed and methods used to convey messages allowed for tailoring to different levels of intellectual ability;
- the development of a specific carer session to engage and promote involvement;
- ensuring participants were appropriately prepared prior to attendance and at the start of each session;
- reflection on their own levels of risk;
- self-monitoring (diaries and pedometers);
- goal setting and action planning;
- exploration of barriers and individualised solutions.

The specific methods employed to ensure that the themes highlighted above were operationalised are detailed below.

Preparation and grounding

The team agreed that all service users (and carers/family members) would need to meet the educator prior to commencing on the education programme. This would allow the educators to confirm suitability to attend the education sessions, make an assessment of any specific needs that would need to be met, and to briefly describe the sessions and check willingness to attend.

A specific carer session was to be held prior to the participant education sessions.
To provide familiarity and consistency, the sessions would be delivered in a familiar community facility on the same day and time, with the room set out in the same way each week. Additionally, the same core group of educators would carry out the pre-assessment visits and deliver the programme, to ensure continuity and develop rapport.

Style and principles of the education programme

Gaining an understanding of each participant's emotional and physical well-being prior to each session was seen as essential, to allow facilitators the flexibility to meet the needs of individual participants. Therefore, educators were to make time to meet and greet participants and carers before the start of each session. Additionally, educators would establish a set of mutually agreed guidelines for the group at the outset of the education session to support group functioning.

To meet learning styles within the group and tailor the content to meet individual needs, educators would employ multiple methods (to check understanding) and multiple modalities; the additional support of an experienced ID healthcare assistant would be utilised during the sessions.

Regular breaks would be taken as indicated by the groups expressed need or level of engagement. Educators would use the session material to create concrete examples and develop activities that create movement. In addition, the curriculum would be designed to support the use of recall and repetition to support learning;

resources developed by participants such as posters, postcards, cue/prompt cards would support individuals to maintain behavioural and lifestyle changes.

Curriculum content and activities

The main behavioural goals and content (see Section 8.8.4; *Table 32*) were to be drawn from previous prevention studies.[240-242] In addition, abstract concepts like risk and future self were to be developed as activities, games or stories using a concrete kinaesthetic modality. To promote participant engagement in the programme, the educators would use practical and participatory methods such as food models and images, visual memory aids and short walks using a pedometer. The educators would also use reinforcement methods, including certificates of attendance and attendance cards, as a regular activity within the programme. If the participant chose, self-monitoring activities/opportunities would be promoted, such as diaries to record food and physical activity outside of the session; the opportunity to use a pedometer and scales would also be available to monitor weight when attending sessions. The curriculum would include action planning and goal setting opportunities (in most sessions) around activity, food and other behavioural goals, supported by individualised resources. The educators would create opportunities through activities to explore barriers and solutions on an individual basis and in group activities.

8.8.4 Key behavioural goals of the education programme

For the STOP programme, the key behavioural goals and lifestyle messages incorporated into the education sessions were based on those of the Let's Prevent programme (nutritional)[240] and PREPARE programme (physical activity).[241, 242] Specific goals included: losing weight; reducing consumption of total and saturated fat; increasing dietary fibre consumption; and increasing physical activity and/or reducing sedentary behaviour (see *Table 32*). However, the emphasis of the STOP programme was on enabling the individual tailoring of goals based on a participant's needs and abilities, including potential mobility restrictions, level of independence with food shopping and preparation, potential dietary restrictions, opportunities to access the community, cognitive level, and availability and level of carer support required. Therefore, more generalised behavioural goals were emphasized, rather than setting specified targets (see *Table 32*).

Table 32: Key behavior change goals

Specific nutritional and physical activity goals	STOP Diabetes key behavioural goals
Weight reduction Sustained weight reduction of > 5 % body weight	Choose smaller portions Reduce fat intake from all sources Reduce sugary drinks and foods Choose healthier cooking methods Choose healthier snacks and treats Increase physical activity/ reducing sedentary
Reduce total fat consumption Moderate reduction in total fat to < 30% energy intake	Reduce fat from all sources Choose lower fat options Reduce processed foods and ready meals Choose healthier snacks and treats
Low saturated fat intake Reduce saturated fat intake to < 10% energy intake	Reduce fat from all sources Reduce processed and ready meals Choosing healthier snacks and treats
Higher fibre intake Increase fibre intake to >15g per 1000 calories	Increase fruit and vegetable intake to 5 a day minimum Choose healthier snacks and treats
Increase physical activity / reduce sedentary behaviour A minimum recommendation of 30 minutes of moderate intensity physical activity per day	Increase moderate intensity activity by increasing steps or adding extra physical activity Reduce sitting time

8.9 Discussion

This chapter describes the first phase of the development of a lifestyle behaviour change programme for adults with ID. We took a pragmatic approach to intervention development, using the Medical Research Council framework for developing complex interventions[238] to combine existing prevention programme,[240-242] intervention mapping,[239] evidence reviews, stakeholder interviews and expert advice. This systematic process allowed us to make the following underlying assumptions for the programme:

- People with ID have limited knowledge of healthy lifestyle messages.
- People with ID generally have poorer diet and exercise less often than the general population.
- Health beliefs, knowledge, motivation and social support are key in promoting behaviour change among people with ID.
- People with mild and moderate ID need a specially tailored intervention to promote behaviour change. Mainstream interventions are not suitable for this population.

Our qualitative findings largely support the literature [47, 48 49 50] in finding that people with ID had limited knowledge about healthy lifestyle messages and experienced barriers in undertaking physical activity. However, we acknowledge that findings from the qualitative exploratory interviews with service users may be limited due to the short interview length (average 9-10 minutes); although, this length does not include the additional time taken to explain the study, assess capacity, obtain consent and allow for breaks. We recognise that people with ID are not a homogenous group; some people found it difficult to concentrate and for other people several visits to allow trust to be built-up may have been a better approach. Additionally, in some circumstances, carers (personal and care workers) were not able to be present throughout the whole interview or, if there, they did not agree to participate. It would have been beneficial to purposively seek the views of a larger number of carers (both personal and care workers) at the development stage. However, we were able to obtain valuable feedback from carers during the piloting phases (reported in the following chapter, Chapter 9) and modify the programme accordingly.

8.10 Concluding remarks

This chapter has described the first phase of the education development process that was carried out to develop an initial curriculum for a lifestyle education programme for adults with ID. The following chapter (Chapter 9) details a pilot testing and evaluation phase. Chapter 10 outlines a feasibility study conducted following development of the education programme. Chapter 11 describes development of an intervention fidelity process undertaken for work package 3.

CHAPTER 9. PILOT TESTING AND EVALUATION OF AN EDUCATIONAL CURRICULUM FOR PREVENTION OF TYPE 2 DIABETES

9.1 Overview

This chapter describes a pilot testing and evaluation phase, which follows on from work conducted to develop an initial education curriculum (presented in Chapter 8). An additional feasibility phase, which formed part of work package 2, is presented in Chapter 10.

9.2 Aims and objectives

The aim of this further phase of the development work was to conduct two pilot cycles of testing, evaluation and modification of the initial education programme.

9.3 Methods

Following development of an initial curriculum, a pilot phase, which involved two cycles of testing, evaluation, modification and re-testing, was conducted (see Chapter 8, *Figure 20*) The first pilot cycle was conducted between April to July 2014 and the second cycle from October to December 2014.

9.3.1 Participants and recruitment

Inclusion/exclusion criteria for work package 2 have previously been described in Chapter 8 (Section 8.4). People invited to engage in the pilot phase were service users who had taken part in the screening stage (see Chapter 5), screened positive for IGR or had a BMI ≥25, and at that time consented to being approached to assist with later phases of the research programme. Recruitment followed a similar process to the earlier development phase (see Section 8.4). An initial telephone call was

made to potential participants, followed by further information sent in the post or provided at a face-to-face visit.

9.3.2 Delivery of the education

Potential volunteers with ID were approached about attending the education programme, approximately four to six weeks prior to the planned programme start date. Carers were invited to an initial session, held one week before the delivery of the main education sessions. The aim of the carer session was to provide carers with an overview of the education programme, and explore their role in supporting individuals with ID, both within and between the sessions.

Subsequently, the initial curriculum was delivered to a group of individuals with ID. Carers were also invited to attend the sessions to support the service users. Following feedback and refinement of the curriculum (see Section 9.3.4), the modified curriculum was then delivered to a second separate group, which again was followed by feedback and refinement.

Three educators were involved with delivering the programme at each session; a registered ID nurse, a diabetes specialist with an education background, and an additional ID nurse or health care assistant in a supporting role. The educator training process for the study is described in Chapter 11.

9.3.3 Data collection

A range of methods were used to evaluate the education sessions and collect feedback. These included observations recorded during the sessions by an experienced researcher, reflections from the educators leading the programme and qualitative interviews with people who received the programme (those with ID and carers). Additionally, subsequent to the first pilot phase, educators were also interviewed to explore their views about the content and style of delivery, experiences from delivering the programme and perceived practical issues. Feedback and reflection on educator training are described in Chapter 11.

Participants were approached to take part in a feedback interview prior to the last session of the education programme. Interviews were held as soon as possible after the final session. Written consent was obtained immediately prior to the interview. Participant interviews took place in July 2014 for the first cycle and December 2014 for the second cycle.

The purpose of these interviews was to explore participant and carer views about the education sessions; to identify whether the education sessions resulted in changes to participants' diet and physical activity; and to inform changes to the next iteration of education sessions based on participant feedback.

Interviews were conducted and analysed by the same qualitative researcher that carried out the previous interviews (see Section 9.5). Key areas and topics that were explored included experiences of receiving the education programme, ease of understanding, views about the content and style of delivery, usefulness, relevance and practical issues (including duration, provision of support, and suggestions for improvement).

9.3.4 Refinement

At the end of each pilot cycle modifications were made to the curriculum prior to it being used in the next iteration. Modifications and refinements were informed by findings from participant and carer interviews, observations made during the education sessions, and the ongoing reflection and feedback of the educators.

9.4 Findings – first pilot phase

9.4.1 Uptake and attendance at education sessions

The first iteration of the education programme was held in a community resource centre. A total of 21 participants were invited to take part in the education programme. Five participants (four who had carers) initially agreed to attend the programme. Following the carer's session, one person (and their carer) withdrew completely. Subsequently, four participants (and three carers) took part in the main education programme. Overall attendance at the education sessions (seven weeks, one session per week) was very good, with one participant (and carer) attending all seven days and three participants attending six days (see *Figure 22*).

9.4.2 Characteristics of participants

Of the four participants taking part in the first iteration of the education programme, two (50%) were male, the median age was 35 years (range 29-60), three (75%) lived in a supported environment with family or carers, and one lived independently. None of the participants were in paid employment, one attended college and did voluntary work, and all four participated in other activities within the community.

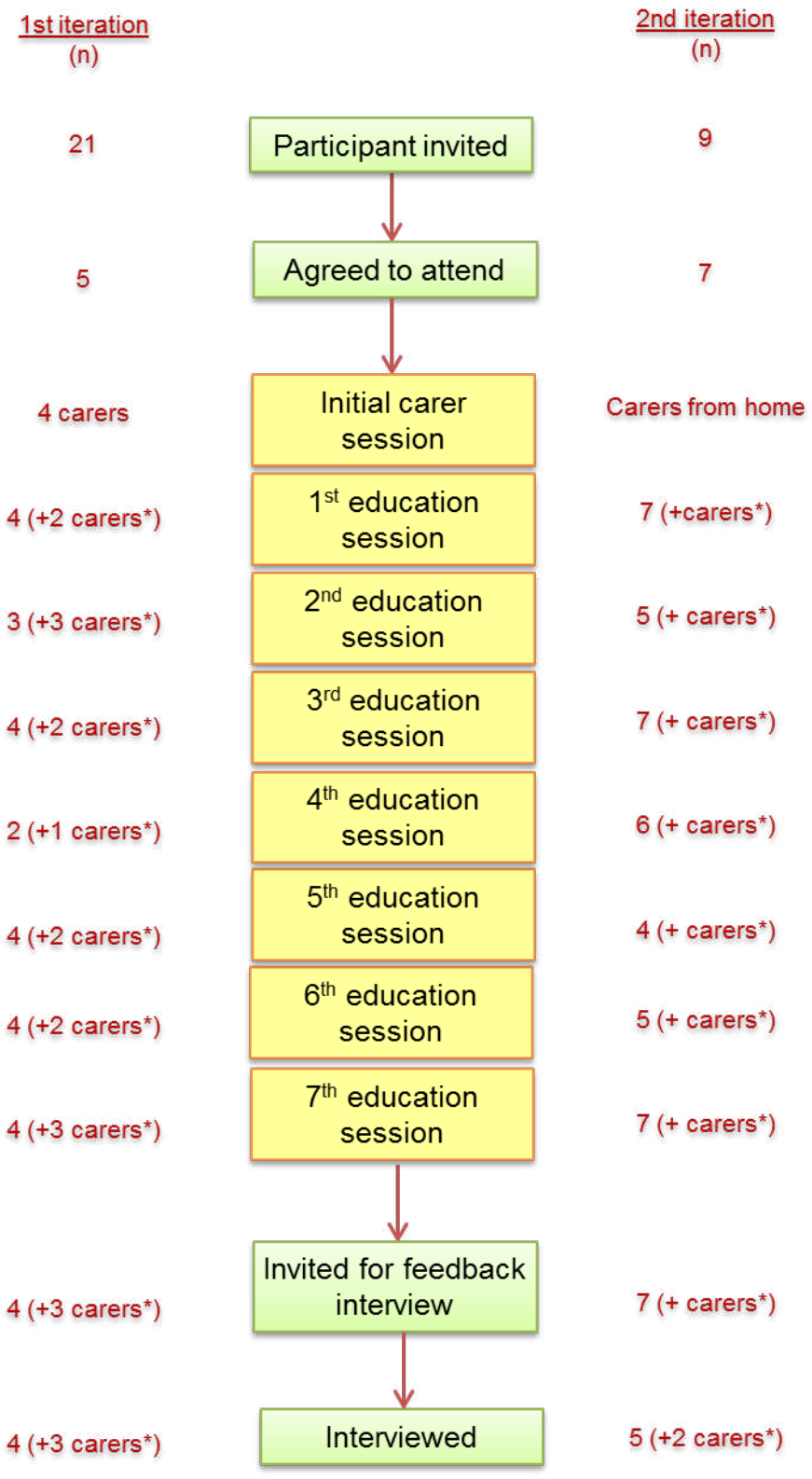


Figure 22: Uptake and attendance at first and second testing phases of pilot cycle

9.4.3 Feedback interviews – first pilot cycle

Following on from the first iteration of the education programme, all participants (n=4) and carers (n=3) agreed to be interviewed. One participant was interviewed independently. For the remaining three interviews, carers and participants were interviewed together; in two of these interviews the carers made major contributions to the interviews.

9.4.3.1 Interviews with participants

Key learning and behaviour changes

For one of the participants, who had previous experience of attending health related courses/groups, initially during the interview, they suggested that they had not learnt anything new. However, through further exploration of the impact of specific activities/games it was possible to identify that the programme had reinforced key health messages, including types of healthy/unhealthy foods, portion sizes and the link between food eaten and body weight.

> ***"Mm, just be careful what…if you do eat any unhealthy [food] not to have so much of it." (Pt2, F)***

> ***"….. yeah, makes you think, doesn't it, er, if you don't control what you eat, you do put…(weight-interviewer)……. Yeah, because they say this country's, don't they, obese?" (Pt2, F)***

For another participant, the education sessions were cited by the carer as helping the participant to become more aware of the changes they needed to make, although the participant was also receiving a form of therapy (hypnotherapy) that they believed was also helping them to make lifestyle changes.

"But since we've been on the course and you've attended the course and they've told you this, you've become much more aware of it, haven't you? (Carer)
"I have, yes." (Pt4, M)

Examples of the dietary changes made by this participant included: reducing the portion size of less healthy foods; swapping/replacing some foods for healthier alternatives; and moderating the amount of alcohol they drank.

"Um, I've also learned that, um, alcohol is, kind of, fattening, but as long as you don't drink it too much…" (Pt 4, M)

Changes to physical activity included doing more walking and going to the gym. Consequently this participant (and carer) reported that he had initially lost 10lbs in weight, which was maintained at 7lbs (immediately following the intervention). However, this participant also reflected that as he grew older he was paying more attention to his lifestyle.

"I just didn't really care so much, to be quite honest, in the past. But now that I'm a lot more older, I'm starting to take things more…more wrong, aren't I, Mumsy? (Pt4, M)

<u>Sustaining changes</u>

The wearing of a pedometer (optional) to measure activity was discussed by one participant; it had helped to quantify her existing level of activity, and to her surprise it was a lot more than she thought. However, there was a sense of despondency that could be observed during the interview about the perceived lack of support or encouragement she had to carry out and sustain lifestyle changes to her diet and activity levels, outside of the sessions/in the future. This issue, coupled with her concerns about her personal safety, featured in the decision not to seek more opportunities to go out walking

"but it's just someone to go with, you know, encouragement. ……sometimes I give up easy with them sort of things. But I've started taking a friend's dog a walk once a week with my other friend and her dog, so round [park], that's nice. I go on the walks with the church and that." (Pt2, F)

"..but I'm not walking to [town] or [town] because it's not safe". (Pt2, F)

She also questioned her ability to make changes. When probed further she identified that if she had encouragement she could make changes.

"But sometimes I can't believe that I can change it." (Pt 2, F)

"I think I could with a bit of encouragement, you know." (Pt 2, F)

For two of the participants who had expressed a desire and commitment to make lifestyle changes, the support and encouragement from carers and other people in their lives assisted them with making changes and possibly helping them to sustain these changes. In the case of one participant, the people in the place he worked were actively trying to help support him to make changes, (specifically changes to his diet). He acknowledged that this support helped keep him on track.

"…and so the girls there are trying to help and support you, aren't they… in every way they possibly can." (Carer for Pt 4)

"It helps a lot, 'cause then if I do it all by myself, I'll be ending up having burgers or something."(Pt 4, M)

Asked if he could sustain the changes he had made to his lifestyle, he responded by saying he felt he could sustain these changes if he focused.

"..if I put my mind to it." (Pt 4, M)

In the case of another participant, family and friends had helped them by taking an interest in the participant's take-home activities and resources (completed outside of the education sessions), and by buying the participant a bike. These were in addition to the dietary and physical activity changes that the family were making. They talked about sustaining these changes even when on holiday.

> ***"And she kept going…I says, no we're walking, and it's very early, but we did it didn't we, [Name]? From the beach right up to the hotel? Yes? Yeah, even though you wanted to sit down! (Carer for Pt 3)***

Symbols and images used to support learning

For some activities, smiley green faces and red sad faces were used to indicate concepts like healthy and less healthy. However, for some participants these images/symbols were unhelpful and confusing. They were open to mis-interpretation or could mean different things than was originally attended in the curriculum. For two of the participants, a red smiley face depicted foods that they did not like as opposed to indicating less healthy food.

> ***"Now, if you gave him a bowl of salad and said, does that go in the red or the green, he'd put it in the red because he don't like it." (Carer for Pt 1)***

Another participant placed an unhealthy food on the green sticker because she liked the food.

Reasons for continuing to attend the education sessions

One participant wanted to continue with the education sessions saying that they had helped him by giving him freedom from work and enjoyment.

> ***"…to actually carry on with it. 'Cause I've got that much out of the…out of the sessions than I would normally do." (Pt 4, M)***

Other reasons given by people for continuing to attend included: meeting other people and learning new activities; enjoying the games/activities; and due to a sense of achievement upon completion

Only one participant discussed that he would have liked a shorter session, about an hour and half, and felt that there was too much information with too much emphasis on food. Another participant felt the venue may deter some people from attending, as it was a venue that cared for a range of people with ID (including people with behavioural difficulties) and they may see people behaving in a challenging way.

9.4.3.2 Interviews with carers

Carer's benefitted from the education sessions in various ways and at different levels. For some carers, the sessions reinforced prior learning, and for others they motivated and encouraged them to make changes. These are described in more detail below:

Support and encouragement

It was evident from contributions made by participants and carers during the education sessions, that many of them had prior knowledge and understanding of key healthy lifestyle messages. These were gained from their attendance at previous health education/promotion courses and from the media.

We were able to explore this issue with one of the carer's who was asked why participation in the education sessions appeared to have made such a difference to their life when there were so many health messages in the media that they could have acted on (for example, in relation to physical activity). The carer attributed the impact of the sessions to the person centered approach, which facilitated their understanding of "how" to make and sustain lifestyle changes.

> ***"Well I think how you explained things to us really. I think that were very helpful, it was.....on a level with me.....you know? And it was as though you were speaking to each individual…….. Not just a great big party or you got a book. ……you were telling us and explaining more to us how to do it. Yeah, I think that's what helped me anyway the most." (Carer for Pt 3)***

When specifically asked whether she had heard about the benefits of walking before coming to the programme, her response confirmed that she had. However, the education sessions contributed by providing encouragement to put into practice her knowledge about walking.

"Oh yes, yes, but we didn't do it. I think you gave us the encouragement to do it." (Carer for Pt 3)

Motivation and focus to make changes

On a different level, another carer felt her levels of knowledge were already raised compared to other people, due to previous attendance at other continuing development sessions as a consequence of being a paid carer. The STOP education sessions had helped to jog and refresh her memory. However, she felt they had little impact on her or the participant making further changes largely because the participant did not really want to make any changes to their lifestyle. Referring to the participant:

"I don't think you've helped [participant], if I'm honest……because (referring to participant) you're a bit stuck in your ways as to what you have and what you don't want, aren't you. For me, yes, it sort of jogged my memory and made me think, oh yeah we'll do this and we'll do that…….. not just for me with [participant], for me with me other service users as well. But I think, yeah, she can do that and what have you. So, although I'm a bit stumped with you (referring to participant)." (Carer for Pt 1)

Another Carer described how the experience of attending the education sessions motivated and focused her efforts to support the participant to make and sustain lifestyle changes. Underlying this motivation was a sense of fear for the participant's future health, that he may need to go on to medication, and a concern about this if she was not around in the future.

"……it's made us focus. You've showed us the little smiley faces. We've had to put the smiley faces on the right things and the wrong things, and we've focussed in with them, you know…. Um, it makes us focus in to what he's doing…" (Carer for Pt 4)

..”because I don’t want him to take medication, because I know that eventually it’ll be insulin, and if I’m not around, goodness, you know…” (Carer for Pt 4)

Additionally, carers identified a number of dietary changes that they had made for the whole family as a result of attending the education sessions, which included reducing sugary foods and fats, and increasing fruit intake.

Carers session

We tried to ascertain the contribution of the carers session to their understanding of what the education sessions would involve prior to them attending. Unfortunately, very little information was gleaned as it was difficult for them to remember. One carer explained that they appreciated the carer session because it helped prepare her and the participant for what the programme would involve.

“So it were a bit, sort of, rather than being just chucked in, I had an idea of what we were going to be doing ……. So that I could explain to [name] what we were going to be doing.” (Carer for Pt 1)

“You see, I suppose really, the carers’ session is for my side of things, and other carers coming in who have to do the meals and have a bit of input.” (Carer for Pt 1)

When asked what they would say to a new group of carers to motivate them to attend the next round of education sessions, one of the carer’s stated:

“I’d stand up and I’d say go for it because you learn an awful lot that you think you know, and you don’t until it’s put down on these. I really would.”(Carer for Pt 3)

9.4.3.3 Interviews with educators

Six educators, who were involved with delivering some or all of the education sessions for the first iteration, were invited to be interviewed. Subsequently, face-to-face semi-structured interviews were conducted with five educators. These included three registered ID nurses, one diabetes specialist with an education background, and one health care assistant. One educator was male. Four of the educators had ≥10 years' experience in their professional area; one had ≤5 years' experience.

Overall, educators felt that the education sessions had been positively received by people with ID and their carers. The key findings regarding the process, curriculum content and delivery of the education sessions, are presented below.

Self and peer reflection

The process of self and peer reflection after delivering the education sessions was reported as invaluable for their role as educators. Discussions with educator colleagues at the end of each education session helped to identify what worked well and areas for improvement in terms of adaptation of resources and identifying any sections of the curriculum or facilitation that required adjustment to meet individual needs on an ongoing basis. This in turn helped to iteratively refine and modify aspects of the curriculum such as, for example, resources and explore different strategies to respond to participant group dynamics.

Venue

Overall views were that a "day centre" was a good environment in which to hold the education sessions because it was familiar to the participants.

Size of the group

Educators felt that the key issue was to balance the need for positive interaction and ensure enough support was given to enable participants to learn. The minimum number of participants suggested was four and avoiding ‘double’ figures.

Views about resources

Educators highlighted that some of the resources needed to be modified to promote greater visibility and accessibility for all participants, as they were often used with the group sitting around a table. Suggested solutions included placing posters on a frame (so they could be displayed upright or flat) or using larger sizes of all images, including photographs. The need to avoid shiny paper/laminating was also advocated.

Educators also discussed in detail which participant resources worked well and some that would need to be adapted. They highlighted the possibility of reducing some resources, so as not to overwhelm participants (and carers). Some key issues identified included:

- attendance certificates, reduce to one card for the whole programme instead of individual sheets for each week;
- the participant folder (handbook of resources) need to be simplified and the overall amount of paperwork reduced;
- sections and inserts need to be differentiated (e.g. by use of colours);
- to aid the use of stickers in participant resources, need clearer labelling and boxes for where to place;
- food diary worked well for some people but may need to consider alternative ways of recording food intake;
- pedometer worked extremely well for some people but not for others;
- when discussing “health checks” in the programme, have actual equipment to promote discussion/illustrate rather than images.

Views about the overall curriculum, style of delivery and group dynamics

Educators contributed a number of things that were perceived to have worked well within the group:

- participants had some prior knowledge which they wanted to apply for themselves; the education sessions contributed towards enabling/supporting this;
- a “happy” and “ keen” group with “characters that complimented each other”;
- the sessions were perceived to be ‘pitched ‘correctly, although it was acknowledged that for group education it may not be possible achieve this for everyone; a flexible approach (altering language, using different resources) and skilled facilitation helped to address this;
- a lot of participant (and carer) interest in food and weight reduction;
- there were visible changes to a participant’s level of confidence over the course of seven weeks;
- the bingo (game/activity) was a useful ‘recap’ tool;
- allowing time to complete ‘homework’ during the first session was perceived to be a better approach; it could have overwhelmed participants if they were required to take something away to complete on their first day;

There was a general perception that the short walking activity within sessions worked really well on several different levels, it:

- helped to break up sessions;
- was energising and helped concentration;
- sent a “massive message” particularly to carers to show how a short walk can result in a lot steps;
- was a huge motivational tool that sparked off discussion.

Educators felt that carer involvement had contributed to a positive learning experience and that “carer” dynamics in the group had worked well. Some of the suggested ways that carers had helped included:

- identifying difficulties or challenges that participants may have at home and could impact affect making lifestyle change;
- help to support challenging behaviours within the group;
- playing a crucial role in supporting and motivating participants to undertake behavioural changes.

What did not work so well

The primary issue underpinning the education sessions was perceived to be keeping the balance between maintaining the motivation of participants to attend each session and not overwhelming them. A few educators felt that there were too many messages within the curriculum and that these could be reduced, with an emphasis in a future iteration on linking and building on messages. Similar views were expressed about the amount of resources used and a recommendation to review the amount and timing of their introduction at different points within the education sessions.

Some specific difficulties raised that related to the second week of the main programme included that there was:

- a lot of repetition (but this may be linked to educators following the curriculum too rigidly);
- too much discussion when pedometers were introduced and participants found it difficult to make the connection about their results.

Other points for consideration included:

- the dominance of one participant highlighted a need to explore different ways of addressing this, should it arise in the future;
- at times there was too much talking, during which some people were lost within the discussions;
- a conceptual exercise/activity called “Big Daddy” did not work well with some participants;
- the pace of sessions was too fast earlier in the programme, although this was adjusted in later sessions and subsequently viewed as working better.

Finally, a few educators recognised that it was difficult to convey the concept of future risk of developing diabetes and recommended that the next iteration should emphasise the importance of providing foundational learning to help motivate and understand healthy eating. Educators' also perceived that future follow-up sessions would be an essential part of the education programme and of particular importance in this population. Future sessions were seen as helping with: retaining focus; reinforcing positive behaviour changes; recap of learning; and identifying progress through practical measures (e.g. weighing scales).

9.5 Findings – second pilot phase

9.5.1 Uptake and attendance at education sessions

The second iteration of the education programme was held in a residential setting. A total of nine participants were invited to take part in the education programme. Several staff (care workers) from the residential home attended the initial carers' session and seven participants agreed to take part in the education programme. In general, attendance at the education sessions was good, with three participants attending all seven days, one attending six and the remainder attending at least four days. Care workers from the residential home also attended at various points during the seven sessions (see *Figure 22*).

9.5.2 Characteristics of participants

Of the seven participants who took part in the second iteration, three (43%) were male, the median age was 43 years (range 29-50), and all seven lived in a residential home supported by carers. One of the participants had paid employment, two did voluntary work and all seven participated in other community activities.

9.5.3 Feedback interviews – second pilot cycle

After the final education session, a total of five participants with ID were interviewed along with two members of staff (carers) who had attended the education sessions. A care support worker who had attended some of the education sessions, provided support to one of the participants during the interview, helping her to feel at ease and assisting her to recall and discuss some of the lifestyle changes she had made. The remaining four participants chose to be interviewed in pairs with their respective partners. This arrangement worked well in terms of facilitating participants recall and support of each other, although it was challenging to ensure both participants contribution to the interview was maximised and balanced.

The presentation of quotations to support the summary of findings includes singular quotations as well as sections of the discussion with the researcher to help contextualise some of the responses to the questions.

9.5.3.1 Interviews with participants

Enjoyable sessions

It was fairly evident that the education sessions had been an enjoyable experience for all the participants. One participant was particularly happy about achieving weight loss and being able to share that achievement with their family.

> ***"The steps I've done, I'm amazed about the certificates. I tell my mum about it, she is very happy…… They're pleased about it, and my weight has gone down with it." (Pt 1, M)***

Other participants talked about the specific things they had enjoyed, such as being part of a team/group, the resources ('stickies') they had used, or for one participant they were enthusiastic about all aspects of the programme.

> ***"Group activities…..and working well as a team. (Pt 4, M)***

> ***"And sticking pictures on the posters, as well."(Pt 5, F)***

> ***"Everything!" (Pt 3, F)***

For three participants, when asked further to expand on what they enjoyed the most, they described sessions and resources relating to physical activity. Their enjoyment appeared to be linked to group walking within sessions, using a pedometer (given to them as part of the programme) to record how many steps they were achieving and recording steps/activity in their physical activity diary in between the education sessions.

"That walking around, kept going and going and going…" (Pt 3, F)

"Plus the pedometers. Count how many steps… Shows how many steps." (Pt 4, M)

"Um, actually writing about my miles." (Pt 5, F)

Key things learnt

In response to a question about what they had learnt from the education sessions two participants cited physical activity and weight loss.

Participant 3, F:	***"I've learnt a lot."***
Researcher:	*What kinds of things have you learnt?*
Participant 3, F:	***"To lose more weight."***
Participant 4, M:	***"And exercise."***
Participant 3, F:	***"And exercise more."***
Researcher:	*And you said exercise, what is it about exercise that you learnt?*
Participant 3, F:	***"It keeps you healthy."***
Participant 4, M:	***"And your heart…"***
Participant 3, F:	***"And your heart beating."***

Establishing if participants associated key dietary and physical activity messages with specific parts of the education sessions proved difficult to elicit as recall about more detailed aspects of individual sessions was low. Therefore, drawing upon observational data that were collected during the seven sessions, the researcher took the opportunity to explore two sessions (storytelling and bingo) which had noticeably demonstrated a high level of engagement and participation, to identify whether participants could link these sessions to specific messages.

Both the bingo session and storytelling session were remembered as enjoyable sessions. One participant's comments also conveyed the key message he took from the storytelling session.

> ***"And the story books were absolutely fantastic…..Yes, I enjoyed it, there's nobody stopping me reading that, because [Name] was showing it, or [name] was showing it, what the whole people were eating, lots of cakes. That's not good, that's bad you know." (Pt 1, M)***

Behaviour changes

All of the participants who were interviewed had discussed during the education sessions that they had lost some weight (ranging from 2-5 kg). A few of the participants were motivated to lose weight for personal goals they had set for themselves. During the interviews participants (or carers) described some of the dietary changes made to help them achieve their goals. These included cutting out fizzy (sugary) drinks, reducing alcohol, smaller portions sizes, replacing chips with jacket potatoes, cutting down on puddings and eating more salad.

> ***"We used to drink loads of fizzy drinks and we don't now." (Pt 3, F)***

> ***"You're not having such big portions ……. you have been trying hard for quite a while to eat better, haven't you? You don't have chips; you have a jacket potato on Friday." (Care Worker, Pt 5)***

Researcher: You went down by two (Kgs)? Your weight went down didn't it?
Participant 2, F: "Yes."
Researcher: [Name], what did you do to change things?
Participant 2, F: "Salad."
Participant 2, F: "Every Monday I don't have puddings."

Feedback also suggested that some participants were consciously focused on undertaking physical activity as a consequence of attending the education sessions.

Pt 1, M: "Not to too many sweets, go for a walk, try to get some more miles down, steps."

Researcher: "So, that's something that's changed for you, you've increased your steps?"

Pt 1, M: "Yes, I'm proving them right, you see."

"I've done…so far I'm going on the walking group and making new friends." (Pt 4, M)

"I was starting riding my bike long time ago….. So I'm starting it again." (Pt 5, F)

Sustaining changes

In response to a question about whether making the changes had been easy or difficult, two participants acknowledged making healthier choices were challenging with respect to reducing portion sizes and to overcome the temptation of sweets which are available in the flat shared with other residents.

"Just getting used to the amount you want and stuff" (Pt 3, F)

"It's hard; it's tempting to have sweets in the flat all the time, that's what tempting." (Pt 1, M)

Care workers appeared to play a key role in helping to motivate and support participants to make and sustain changes to their diet and physical activity. This was illustrated by a participant who had lost a considerable amount of weight and described how staff (and his partner) had helped him with making healthy food choices and eating smaller portions:

"We do (help each other), because my link worker is helping me with my diet. She's got all these healthy eating in my flat, see what I've got in the cupboard. That's like salad sandwich and wraps as well, and coffee and oranges as well, squash. (Pt 1, M)

When asked for examples of how staff had helped him, he responded by describing the following changes:

Participant 1, M: "Eating less, eat salad, eat fresh fruit, coffee, or a sandwich, or something."

Researcher: Is there anyone else who can help you to carry on with the changes?

Participant 1, M: "Link workers"

9.5.3.3 Interviews with carers

Sustaining changes

Care staff had already considered ways to sustain the changes made and the motivation of residents after the education sessions stopped.

> ***"I've said, it's important we keep it up. So while it's still fresh and you can run with it, because the weight loss thing, for their only to be one person who hasn't actually lost weight and even though it's a little bit. ……We very often get the talk about how from your little acorns grow the big trees." (Care worker 1)***

Things currently being discussed by care workers were a healthy living course and a weekly physical activity session. According to staff, following residents' participation in the STOP diabetes study there was a general increased level of interest in "healthy living" and a few residents had recently indicated that they would like to do more exercise.

> ***"Yes, it's [putting on a health living course] come about because you've come here, because of how the residents have responded, but also because I know they've made that request about having more exercise. (Care worker 1)***

Suggestions to facilitate recall of food messages and sustain changes outside of sessions included having images of breakfast alternatives that could be stuck onto a fridge door. This is something that is currently being tried with one of the participants because he has difficulties remembering.

Amount of information covered in sessions

In response to being asked about how they found the education sessions, and specifically about the amount of information, two conflicting views were evident.

One carer stated:

> ***"Well, just right, yeah. That was fine for what they were… And because it was the mix of those that could write, could write things, but they have their stickers and their pictures." (Care worker 2)***

Another carer felt based on her observation of one of the sessions that there was too much information that may not help with retention of information:

> ***"I think in one session I was at there was a lot of information being given and maybe just simplify a little bit. Maybe just doing very small steps, and even if it's just one piece of information they learn that session, at least that might have more chance of sticking." (Care worker 1)***

To help with retention this participant felt concentrating on one aspect of diet such as drinks for example, may have helped to focus efforts and facilitate discussion about alternatives.

<u>Carer involvement</u>

In response to a question about what educators needed to consider for any future programmes held in residential homes, the need to allow for variation in care workers attendance at sessions to support participants, due to organisational pressures (including low staff numbers), was highlighted.

The above discussions also elicited the further suggestions for encouraging carer involvement:

- the need to educate staff about what they will need to consider for people who they are responsible for;
- to enthuse and engage staff to help with practical support such as completing the diaries and resetting the pedometer;
- to provide information about alternative (healthier) food choices/options and portion sizes.

9.6 Modifications made to curriculum after the pilot cycles

A number of modifications were made to the programme based on feedback from participants and carers, observations made during the education sessions, and the ongoing reflection and feedback of the educators. Modifications consisted of refinement of resources, together with adaptations to educator facilitation within the sessions.

9.6.1 Revisions made after the first cycle

The main revisions made after the first cycle included:

Modifications to the carer session

- Reducing the amount of information provided at this session.

Modifications to education sessions

Participant resources

- Reducing the amount of worksheets given out at any one time. For the first pilot phase, all activity sheets developed for the seven week programme were given to participants in week one, in a folder format. This caused distraction for some participants and impacted on delivery of the session. Subsequently, this was changed to allow for the work sheets to be provided directly at the point they were required in the programme and for participants‘ to add to their programme folder on a weekly basis.
- Simplifying the physical activity diary to a single sheet of A4, with a table to record date, steps/activity and new goal, from a multi-paged booklet.
- Making the image cards, which are used to facilitate and support learning, recognition, recall and summaries, a much larger size.
- Using realistic images and or photographs in resources. Images were sourced and checked with service user groups prior to being changed.

Session content

- Reducing/simplifying the content of some sessions. Providing too much information led to participants becoming disengaged.
- Changing the symbols used to illustrate healthy and less healthy foods as these were not universally understood by participants. Possibly a menu of symbols to tailor to individual cognitive needs. Ensure educators explain and check understanding when symbols are used.

Maintaining and maximising engagement

- Educators to create opportunities for movement, both within the room to engage in different activities and a short walk during each session, to address participants becoming disengaged when sitting for longer periods. Additionally, to use the walking activity to highlight the number of steps achieved in 5 - 10 minutes of walking.

Communication aids

- Using communication cards (with symbols/pictures) as an aid to manage discussions in the group and facilitate engagement of people who experience difficulty communicating.

9.6.2 Revisions made after the second cycle

<u>Modifications to education sessions</u>

Maintaining and maximising engagement

- Allowing for educator flexibility to adjust the timetable and breaks to suit the needs of individuals, the group dynamic, energy levels and engagement.
- Educators and supporting staff to be aware of the diversity and dynamics in the group, and to arrange the seating and positioning of participants to support engagement and one to one support when required.
- Including more interactive games/activities, such as bingo and board games, to promote engagement.

Participant resources

- Including a menu of options to encorage prompts and motivation towards goals. For example, fridge magnets may not be useful to those in a residential setting if they did not have their own fridge.
- Incorporating photos of participant‘s on to their “health checklist“, as a way of personalising documents, and helping individuals to relate this information to themselves.

9.7 Outline of the STOP education programme

An overview of the final education programme developed, prior to using in the feasibility phase (Chapter 10), is outlined.

First, the initial carer session, which is held prior to the main education programme, is presented (see *Table 33*). Secondly, in *Table 34*, the outline structure of a typical session in the main programme is outlined. Finally, for each individual session (weeks 1-7), the topic areas, the main aims, and the key activities and resources that are designed to support learning and behavioural changes, both within and between sessions, are also presented (see *Table 35, Table 36* and *Table 37*).

Table 33: Outline plan of the initial carer session

Session name	Overview and main aims of activities	Time
Welcome and introductions	To introduce the educators To understand the role of any observers To be aware of the style and aims of the course To ask questions related to the course	15 mins
Outline of education course	To be aware of practical aspects (venue, times, number of sessions) To be aware that carers can attend with participants	10 mins
What is different for people with ID?	To have an opportunity to share their thoughts about learning needs of the person they care for To share their thoughts about supporting people with ID to make lifestyle and behavioural changes	10 minutes
Course content	To be aware of course content and resources To be aware of course activities and support participants may need between sessions to complete	60 mins
What is my role as a carer?	To explore the benefits of attending the course with the participant To explore the potential health benefits for the person they are supporting, and themselves, of attending To be aware of their potential role in supporting the participant who chooses to make lifestyle and behavioural change	15 mins
Questions and concerns	To have an answer to any questions To have concerns explored and addressed	10 mins
		Total 2 hours

Table 34: Outline of the structure of a typical session in the main education programme

Session	Aims and activities	Time
Welcome Welcome and getting to know you - week 1 or Welcome back – weeks 2-7	To ground and settle participants To outline the aims and style of the course To outline the topic areas for the day To reflect on actions from previous sessions- celebrate achievements and identify/explore barriers To develop good working relationship between educator and participants	15 mins
Topic area 1	Explore a different topic area each week	30 - 45 mins
Break	15 minute break allocated within the session Breaks to be taken flexibly according to the expressed needs of the group or as indicated by educators assessment of engagement in the session	15 mins
Topic area 2	New topic area or may build on/consolidate learning from the earlier session	30 - 45 mins
Questions and preparation for next week	To provide an opportunity to express concerns, ask questions relating to the session To provide information and prepare participants and carers for activities between sessions	10 mins
		Total Minimum 100 mins Maximum 130 mins

Table 35: Outline plan for the STOP education programme weeks 1 and 2

Week	Overview of main aims	Activities and resources	Theory
Week 1			
Topic area 1 **What is health? Being healthy and unhealthy**	To explore what the concept of being healthy means to the individual To explore the behaviours linked to health Develop images that represent healthy and unhealthy characters that are used as a learning tool throughout the programme	• Healthy and Unhealthy character poster • Images to prompt recognition and recall • Main message summary cards	SRT TPB
Topic area 2 **What can go wrong with my health?**	To explore the health consequences of lifestyle and behavioural choices To explore lifestyle and behavioural choices that promote health To have an opportunity to express their emotional response to the different lifestyle choices the characters make	• Images to prompt recognition and recall	SCT SRT TPB
Week 2			
Topic area 1 **This is me and Health checks my doctor or nurse will do**	To create an image that represents the individual, their lifestyle and behavioural choices To be aware of the health checks a doctor or nurse will do and be provided with their own biomedical data and risk factors To be aware of which results may be a problem to their health by placing a sticker on profile Plot results on a health profile	• Personal lifestyle and behaviours activity sheet • Images to prompt recognition and recall • Health profile with photograph of individual • Biomedical data • Coloured stickers	SCT SRT TPB
Topic area 2 **What can I do to stay healthy?**	To explore the impact of the biomedical results and risk factors on their own health To express any concerns/emotions relating to their results Recall the consequences of lifestyle and behavioural choices To explore lifestyle or behavioural choices relating to their risk factors Have the opportunity to choose and record lifestyle or behavioural changes on a personal poster Record level of confidence to make this change	• Images to prompt recognition and recall • Confidence activity sheet	SCT SRT TPB
SCT Social Cognition Mode; SRT, Self - regulation Theory; TPB, Theory of Planned Behavior.			

Table 36: Outline plan for the STOP education programme weeks 3 and 4

Week	Overview of main aims	Activities and resources	Theory
Week 3			
Topic area 1 **Being active**	To explore what being active means To be aware of the consequences to health of being inactive To explore the benefits to health of being active, moving more and sitting less To have an experience of using a pedometer to measure steps	• Images to prompt recognition and recall • Physical activity record • Pedometer • Walking activity	SCT SRT TPB
Topic area 2 **Me and my activity**	To have an experience of a short walk and recording steps or activity in a diary To identify ways to increase activity by adding an activity, increasing step count and/or reducing sitting time To record personal confidence to carry out their chosen goal To create a prompt or reminder for their chosen goal To record activity in a diary	• Images to prompt recognition and recall • Confidence activity sheet • Create prompt cards, send a postcard or create a fridge magnet to promote engagement with their goal within the session and between sessions	II SCT SRT TPB
Week 4 **Topic area 1** **How did I do with my activity?**	To reflect on current level of activity Reflect on feelings related to level of activity To explore own and listen to other group members barriers to physical activity To explore strategies for overcoming barriers To experience a short walk to highlight the increase in steps from short periods of activity To identify a new steps or activity goal for the coming week	• Interactive Dice game to explore barriers • Physical activity record • Walking activity (optional)	SCT SRT TPB

Week	Overview of main aims	Activities and resources	Theory
<u>Topic area 2</u> **Changes I can make to be healthy**	Recall the lifestyle and behavioural changes that influence risk factors To be aware of the impact of unhealthy lifestyle and behavioural choices over many years - this is facilitated by using a story book Recall the personal lifestyle and behavioural choices recorded in session 2 Reflect on progress with these choices Participants prepared and facilitated to explore sources of support for recording food, drinks and snacks over the next week	• Personal lifestyle and behaviours activity sheet • Story book • Food diary	II SCT SRT TPB

II, Implementation Intentions; SCT Social Cognition Mode; SRT, Self - regulation Theory; TPB, Theory of Planned Behavior.

Table 37: Outline plan for the STOP education programme weeks 5, 6 & 7

Week	Overview of main aims	Activities and resources	Theory
Week 5			
Topic area 1 **How did I do with my activity?** **Eating well, eating healthy**	To reflect on activity levels over the last week Generate ideas for overcoming barriers Plan a new activity/step goal Walking activity (optional -decision made collaboratively by the group) Recall the main messages relating to health Identify foods that relate to a healthy lifestyle Identify foods that contribute to being unhealthy Have an awareness of the consequences of high fat, sugar and large portions on health Be aware of the consequences of lower fat, sugar and smaller portions on health Sorting activity with food models and images	• Physical activity record • Walking activity (optional) • Food models and images to support recognition and recall • Food sort task • Stickers	II SCT SRT TPB
Topic area 2 **Changes I can make to eat well and eat healthy**	Recall the food messages from the earlier session Record personal confidence to make a change to food choices Identify one or two small changes to make to personal food choices based on their food diary Create personal prompts to behaviour change	• Food models and images • Food diary • Create prompt cards, send a postcard or create a fridge magnet to promote engagement with their goal within the session and between sessions	II SCT SRT TPB
Week 6			
Topic area 1 **Where am I with my activity?**	To reflect on activity levels over the last week Generate ideas for overcoming barriers Plan a new activity/step goal Walking activity (optional -decision made collaboratively by the group) Recall the main food messages related to health and being unhealthy by participating in an interactive game Recall the consequences of food choices	• Physical activity record • Food diary • Food bingo activity • Bingo prize for the winner	II SCT SRT TPB

Week	Overview of main aims	Activities and resources	Theory
Topic area 2 **How am I doing with my eating well, eating healthy?**	Reflect on the food diary Identify successes and barriers to making changes to food choices To explore own and listen to other group members barriers to making changes to food choices To explore strategies for overcoming barriers with an interactive game Explore how to reward personal success Identify sources of support to reach goals Plan a new food goal	• Food diary • Barriers board game • Create or amend prompt cards, send a postcard or create a fridge magnet to promote engagement with their goal within the session and between sessions	II SCT SRT TPB
Week 7 **Topic area 1** **What have I learnt?**	Reflect on activity levels and the food diary over the last week Review the overall programme and raise any outstanding concerns or questions Recall main points of the programme and revisit associated activities Identify successes and barriers to making changes	• Food diary • Activity diary • Healthy and unhealthy character posters • Personal lifestyle and behaviours activity sheet • Images to prompt recognition and recall	II SCT SRT TPB
Topic area 2 **What can help me to keep going with changes to my food and activity levels?**	Record personal changes on activity worksheet Explore possible solutions to barriers Explore strategies to support the maintenance of changes Set new goals and use strategies to help such as writing a postcard to themselves to be sent in 3-months' time or creating fridge magnets Record personal confidence to carry out their chosen goal Explore sources of support to help achieve these goals Celebrate success	• Worksheet to record personal changes that have been made • Postcards, fridge magnets, flashcards and stickers • Prompt card to give to carers to ask for help • Confidence activity sheet • Course attendance certificates	RP SCT SRT TPB

II, Implementation Intentions; RP Relapse Prevention; SCT Social Cognition Mode; SRT, Self - regulation Theory; TPB, Theory of Planned Behavior.

9.8 Concluding remarks

This chapter and the previous chapter (Chapter 8) has described the development and pilot phases (testing, evaluation, modification and re-testing) that were carried out in order to develop a lifestyle education programme for adults with ID.

The STOP programme development benefitted from a systematic process.[238, 239] The theoretical underpinning was developed and expanded upon from the limited evidence in the literature. This informed the content and style of approach, alongside the qualitative findings from people with ID, their carers, and health care professionals with expertise in working with people with ID. The whole programme was then tailored further to the specific needs of this group by more user feedback, and adaptation by a multi-disciplinary team with expertise in ID and the development of education programmes.

From the initial phases the programme has been well received and is acceptable to the people it is trying to support. The initial feedback via qualitative interviews has suggested that some of the elements of treatment receipt initially hypothesised may have been achieved, via reported changes in beliefs and health behaviours.

The following chapter (Chapter 10) details the feasibility phase. Chapter 11 details the intervention fidelity of the education programme

CHAPTER 10. FEASIBILITY STUDY OF STOP DIABETES PROGRAMME

10.1 Overview

This chapter describes a feasibility phase, which follows on from the education development work described in Chapter 8 and Chapter 9, and forms part of work package 2.

10.2 Aims and objectives

The aim of this sub-study was to assess the feasibility of collecting outcome measures for participants with ID before and 3-months after they attend the lifestyle education programme.

10.3 Methods

10.3.1 Study design

Following initial development, testing and refinement of the curriculum (Chapter 8 and Chapter 9), the education programme was delivered to another group of participants to assess the feasibility of collecting pre and post intervention outcome measures.

10.3.2 Study setting

The feasibility phase was conducted between January and June 2015

10.3.3 Participants

10.3.3.1 Inclusion criteria

Eligibility criteria for the feasibility phase were the same as for the two pilot phases described in Chapter 8:

- service users who had participated in the screening stage (Chapter 5);
- screened positive for IGR or had a BMI ≥25;
- indicated a willingness to assist with later phases of the research programme;
- mild to moderate ID;
- able to stand and walk at least short distances;
- able to attend group education sessions;
- not taking part in any other intervention study.

10.3.3.2 Participant recruitment and consent

Participants were recruited following a similar process to earlier phases (see Section 8.4). Potential participants received an initial telephone call to gauge their interest, and this was followed up by further information sent in the post or provided at a face-to-face visit.

Volunteers were invited to attend an initial appointment at a convenient time for them, where information was provided about the study and informed consent was obtained. Consent was obtained following a similar process to that described previously (see Section 5.5.1). People were asked to consent to the collection of baseline data, attendance at the education programme and collection of 3-month follow-up data.

10.3.4 Data collection

10.3.4.1 Baseline data collection

Baseline data were collected using the same schedule as in the screening study (work package 1, see Chapter 5). If participants had taken part in the screening stage within the last three months, and valid measurements had been successfully obtained, then these data were used for baseline values. If a participant took part in the screening stage >3-months ago, then the measurements were repeated.

Data collected included:

- weight
- height
- BMI
- waist circumference
- BP
- dietary intake (fruit, vegetables and salad).

Physical activity (time spent in light, moderate, vigorous activity) and sedentary behaviour (time spent sedentary) were measured using the wrist worn accelerometer (GENEActiv, Activinsights Ltd., Cambridge, UK). The process for measuring activity using this accelerometer has previously been described in Chapter 7.

10.3.4.2 3-month data collection

At 3-months following the end of the education programme, participants were recalled for repeat data collection, as per at baseline.

10.3.5 Outcomes

Particular outcomes of interest in terms of the feasibility included: (i) the proportion of people invited who attend the baseline appointment, education programme,

individual sessions within the programme, and 3-month follow-up; and (ii) the completeness of key data items at baseline and 3-months follow-up.

Problems encountered during data collection appointments and implementing the education were also considered.

10.3.6 Delivery of intervention

Following on from the pilot phase of testing, evaluation and modification described in Chapter 9, the final modified education programme (see Section 9.7) was delivered to another group of adults with ID and their carers, at a local community venue.

10.3.7 Sample size

We aimed to conduct the feasibility study with at least one group of participants (four to eight people with ID, plus carers).

10.3.8 Data analysis

The feasibility of recruiting adults with ID to attend for baseline data collection, education sessions and 3-month follow-up data collection was assessed using a flow diagram to summarise drop outs at each stage. Completeness of outcome data collected at each stage was summarised using counts and proportions.

10.4 Findings

Study timelines for the feasibility phase enabled us to run one iteration of the education programme, with collection of before and after measurements.

10.4.1 Recruitment and consent

A total of 19 participants were invited to take part in the feasibility phase, of which five (26%) agreed to attend an initial appointment at the end of February 2015, where consent and baseline data were obtained. All participants had the capacity to give consent for themselves, without the need to involve a consultee.

10.4.2 Feasibility of collecting baseline data

At baseline, all participants required measurements to be taken as it was >3-months since they had originally taken part in the screening study. Baseline data were obtained for all five participants, with the exception of physical activity data which was only collected for four (80%) participants, as one person refused to wear the accelerometer (see *Table 38*).

Table 38: Availability of data at baseline and 3-months follow-up

	Baseline *n (%)*	**3 months follow-up** *n (%)*
Weight, kg	5 (100)	5 (100)
Height	5 (100)	5 (100)
BMI, kg/m^2	5 (100)	5 (100)
Waist circumference, cm	5 (100)	5 (100)
Systolic BP, mmHg	5 (100)	4 (80)
Diastolic BP, mmHg	5 (100)	4 (80)
Portions of fruit and vegetables	5 (100)	5 (100)
Physical activity and sedentary behavior *(time spent in light, moderate, vigorous intensity activity); (time sedentary)*	4 (80)	3 (60)

10.4.3 Uptake and attendance of education programme

Following the baseline appointment, all five participants took part in the education programme, which was held from March to April 2015. A total of four carers (two paid care workers, two family) attended at least some of the sessions (three regularly). Overall attendance at the education sessions was good, 4 (80%) participants attended ≥ 5 days and 2 participants attended all 7 days (see *Figure 23*). However, one participant only attended 3 days. It is important to note that the most common reason for not attending sessions was due to other existing commitments such as appointments and holidays.

10.4.4 Feasibility of collecting data at 3-months follow-up

All participants agreed to attend a 3-month follow up appointment at the end of June 2015, to obtain repeat measures. Four of the participants (80%) attended the appointment as arranged; one participant needed a second appointment arranging due to non-attendance (this was the same participant who only attended three days of the education programme).

Anthropometric measures (weight, BMI, waist circumference) were obtained for all participants (see *Table 38*). BP readings were successfully obtained for four (80%) participants. Accelerometer data were obtained for three of the four participants who wore one at baseline; four participants initially agreed to wear the accelerometer but one participant later telephoned the research team to inform them he had changed his mind.

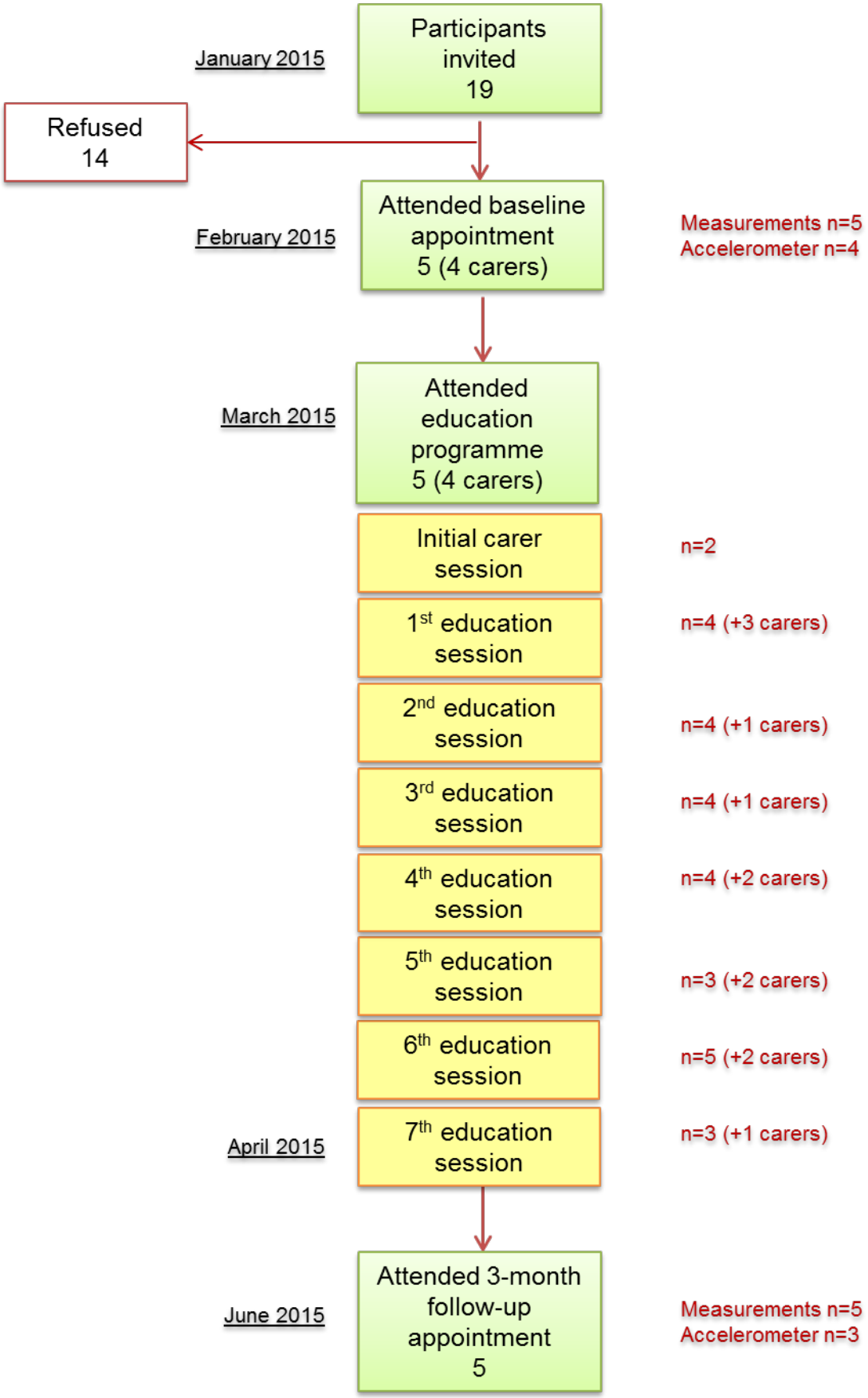


Figure 23: Uptake and attendance for data collection and education sessions

10.4.5 Characteristics of participants

Key demographic characteristics for the five participants' are presented in *Table 39*. Four (80%) participants were male, the median age was 40 years (range 20-51) and all were of white ethnicity. Two participants lived alone, one lived in supported living and two lived in a family setting; the majority (n=4, 80%) had support from a carer for at least some of the time. None of the participants were in paid employment, two did voluntary work on a regular basis, and two attended college and other community related activities,

Table 39: Characteristics of participants

Characteristics	**n = 5**
Age, years (median, range)	40 (20-51)
Sex, male n (%)	80
Ethnicity n (%)	
White	5 (100)
South Asian	0
Other	0
Accommodation n (%)	
Alone	2 (40)
With family/carers	2 (40)
Residential/supported living	1 (20)
Support from carer n (%)	
Yes, at least some of the time	4 (80)
No	1 (20)

10.4.6 Baseline data

Bio-medical and lifestyle characteristics, from data collected at baseline, are presented in *Table 40*. The median values for weight, BMI and waist circumference were 110.2 cm, 36.1 kg/m2 and 114.3 cm, respectively. For BP, the median systolic value was 123 mmHg and diastolic 85 mmHg. Reported daily intake of fruit, vegetables or salad indicated that only two (40%) participants were eating the recommended five or more portions a day (median 5). For physical activity, the median minutes/day for MVPA and light intensity activity were 107.5 and 93.0, respectively; the median time spent sedentary was 555.0 minutes/day.

10.4.7 3-month follow-up data

Data collected at 3-months follow-up are presented in *Table 40*. The main aim of the work conducted was the feasibility of collecting data at two time points, and not looking for change.

However, on an individual basis, three participants lost weight (range 1.0 to 4.2 kg but two participants gained weight. For one participant who had gained a lot of weight (17.6kg), their attendance at the education programme was poor (3 out of 7 sessions); anecdotal evidence at 3-months follow-up suggested that significant changes had occurred in this participants personal circumstances since baseline, including moving out of the family home to live independently in their own flat.

Where data were available, participants showed improvements in physical activity levels and sedentary behaviour at 3-months follow-up when compared to baseline.

Table 40: Baseline and 3 month follow up data – individual level

Participant		Systolic BP (mmHg)	Diastolic BP (mmHg)	Weight (kg)	BMI (kg/m^2)	Waist (cm)	Fruit & vegetables (portions)	Physical activity - time in MVPA (mins/day)	Time in light-intensity physical activity (mins/day)	Sedentary behaviour - time spent sedentary (mins/day)
1	Pre	117	87	133.0	36.1	136.8	7	153.0	126.0	708.0
	Post	115	80	136.4	37.0	137.0	5	51.0	18.0	150.0
2	Pre	101	66	84.4	36.1	112.3	3	106.0	102.0	672.0
	Post	106	68	82.6	35.3	113.5	5	115.0	120.0	708.0
3	Pre	124	83	110.2	36.4	117.5	1	109.0	84.0	438.0
	Post	116	84	106.0	35.0	117.0	5	154.0	96.0	402.0
4	Pre	123	86	112.4	37.6	114.3	6	n/a	n/a	n/a
	Post	n/a	n/a	130.0	43.4	137.4	0	n/a	n/a	n/a
5	Pre	146	85	88.6	28.3	108.1	3	91.0	60.0	366.0
	Post	130	83	87.6	28.0	102.5	3	n/a	n/a	n/a
<u>Overall</u> median (range)	Pre	123 (101-146)	85 (66-87)	110.2 (84.4-133.0)	36.1 (28.3-37.6)	114.3 (108.1-136.8)	3 (1-7)	107.5 (91.0-153.0)	93.0 (60.0-126.0)	555.0 (366.0-708.0)
median (range)	Post	115.5 (106-130)	81.5 (68-84)	106.0 (82.6-136.4)	35.3 (28.0-43.4)	117 (102.5-137.4)	5 (0-5)	115.0 (51.0-154.0)	96.0 (18.0-120.0)	402.0 (150.0-708.0)

10.5 Discussion

Overall, the findings suggest that it is feasible to collect outcome measures at two time points, before and 3-months after attendance at a lifestyle behaviour modification intervention.

Twenty-six per cent (n=5) of people who were invited to take part in the feasibility phase agreed to participate. At baseline, anthropometric measures and BP were obtained for all participants, and accelerometer data for 80%. Attendance at the education programme was overall good, with 80% of participants attending ≥5 days (out of seven sessions for the main programme). At 3-months follow-up repeat data were successfully collected for a high proportion of participants (anthropometric measures 100%; BP 80%; accelerometer data 60%).

It is acknowledged that the feasibility phase only involved a very small number of participants (n=5) and carers. Owing to time restrictions we were only able to conduct one feasibility cycle (delivery of intervention, plus pre and post intervention measures). However, the feasibility phase used robust processes which were informed by the preliminary findings from the screening phase and lessons learnt from the earlier phases of education development and delivery.

It is recognised that the feasibility phase was not aimed at seeing significant findings from baseline to post intervention follow-up. However, at 3-months there were some beneficial changes for most participants.

These preliminary findings are positive but we are unable to assess whether it is possible to collect longer term data, beyond 3-months post intervention, or at repeated time points. The 8-week educational intervention developed as part of work package 2 appears to be both feasible and acceptable to people with ID (and their carers), who are identified as being at high future risk of T2DM and/or CVD. However, the intervention is yet to be tested more robustly via a randomised control trial, including possible follow-up maintenance education sessions. Findings from the

development and feasibility phases provide valuable data to help inform future research.

10.6 Concluding remarks

This chapter has described a feasibility phase that was carried out to assess the feasibility of collecting outcome measures for participants with ID before and 3-months after they attend a lifestyle education programme. Development of the education programme was described in Chapter 8 and Chapter 9. The following chapter (see Chapter 11) details the intervention fidelity of the education programme.

CHAPTER 11. INTERVENTION FIDELITY PROCESS

11.1 Overview

The methods and results of the intervention fidelity process for work package 3 are described below.

11.2 Rationale

Intervention fidelity relates to how an intervention is delivered in practice and whether the delivery of the intervention varies according to the context. It is now recognised as a key component in the evaluation of complex interventions,[238, 249, 250] enabling the assessment of reliability and validity of an intervention and the process factors both advancing the study aims and reducing premature abandonment of future interventions.[251]

The assessment of intervention fidelity is seen as particularly important when interventions are evaluated using multi-centre RCTs, because there is risk of delivering and/or receiving the intervention differently between sites.[250] It was anticipated that findings from the current research programme would inform a future multi-centre RCT.

The Diabetes Education and Self-Management for Ongoing and New Diagnosed (DESMOND) model of structured education,[252] which underpins work conducted in this chapter, draws on theoretical and philosophical perspectives from both health psychology and education;[253-255] patient empowerment is at its centre. DESMOND programmes meet nationally agreed quality criteria[256] for patient education, including delivery by trained and accredited educators, and quality assurance.

One of the key components of quality assurance is intervention fidelity. In practice, assessment of intervention fidelity involves appraising education delivery, with particular emphasis on the assessment of educator behaviours. The DESMOND

education approach purports that individuals in the main are responsible for making their own choices around self-management. Barriers outside of the persons control are acknowledged, but the role of the facilitator or educator is to encourage the participant to explore their motivations for self-management or engaging in health promoting behaviours, rather than telling them what to do; the former set of behaviours can be attributed to being 'DESMOND' and the later are non-DESMOND. Further details of the methods used to assess educator behaviour and the DESMOND and non-DESMOND approach, are outlined in Section 11.5.3 and *Table 42*.

The original programme of work included a pilot RCT to assess the effectiveness of the lifestyle education programme (intervention) developed. However, amendments to the programme of research requested by the NIHR, determined that a feasibility phase was conducted (described in Chapter 10) rather than an RCT. Thus data collection from multiple iterations of the programme and across different educators was not practicable. The amended aims and objectives, as outlined in the next section (Section 12.3), take the above into account.

11.3 Aims and objectives

The primary aim of this component of the research programme was to conduct preliminary work towards developing an intervention fidelity process and tool specifically tailored to this population.

Specific objectives were to:

- develop an outline educator training programme;
- conduct a preliminary assessment of educator behaviour using an existing quality development tool;
- identify key important adaptations to the existing tool for use in a programme developed for the ID population in the future.

11.5 Methods

11.5.1 Educator training

The educators for delivery of the intervention (outlined in Chapter 10) comprised a registered ID nurse and a diabetes specialist with an education background, with support from an ID nurse or health care assistant.

The process of training the educators, first, involved professional development around the DESMOND education programme. This stage took place between January and March 2014, and included observations of DESMOND based programmes and attendance at a Core DESMOND training day. This provided an introduction to the theoretical basis, philosophy and core skills of DESMOND. [252]

Secondly, the educators attended an initial study-specific training day in April 2014, where they were introduced to the theoretical component, content, structure and delivery of the STOP Diabetes education programme (*Table 41*). This was followed by a second study-specific day in June 2014. A staged approach was used for delivering the educator training to take into account any necessary changes to the programme (curriculum content, delivery and structure) based on the delivery of the early sessions.

The training was delivered by two members of the development team, a consultant clinical health psychologist and diabetes specialist nurse with an education background. Training included detailed information on the content, structure and delivery of the programme, and incorporated additional support and considerations around working with people with ID. Guidance around indirect approaches for educating participants (e.g. role-playing behaviour change techniques) and direct approaches (e.g. presentations), were included.

Educators used a specific training curriculum to equip them to support the delivery of education sessions and were encouraged to use personal reflection and peer review

tools to reflect on their delivery. They were also supported with mentorship from trainers attached to the research team.

An outline of the training programme is presented in *Table 41*. The first training day covered the first five sessions of the education programme (the carers' session and weeks 1-4).

Day 2 educator training, which covered the curriculum and resources for the final three sessions (weeks 5 – 7), followed a similar format to the first training day, taking into account feedback relating to the initial training. In addition, educators were able to feedback their experiences on delivery of the first 4 sessions, and to reflect on their own self-monitoring and behaviour change experiences.

Table 41: Outline of the education training programme – Day 1

Session name	Overview and aims	Time
Welcome and introductions	Explore expectations and concerns about training Outline the style and aims of the training	15 mins
What is different about group self-management education for people with ID	Exploring participants experience of delivering education to people with ID Exploring how working with a group may be different to 1:1 Methods to promote learning in this programme	30 mins
Prevention and health messages	Exploring the key messages for CVD and diabetes prevention Explore the key messages for the education programme	30 mins
Key messages for each session	Provide an opportunity to review the curriculum and identify the key messages and activities for each session	30 mins
Development and theoretical underpinning	Recap on the theories that underpin this programme Providing an opportunity to explore their own health behaviours and making a plan.	60 mins
Lunch		
Carer session	Review curriculum and aims of session Explore how to engage carers	45 mins
Sessions: Weeks 1-3	Review curriculum and resources for sessions 1-3 Identify any challenges to delivering these sessions	60 mins
Session: Week 4 and resources	Review curriculum and resources for week 4Be aware of other resources available for use with the programme Identify any challenges to delivering the session and using the resources	40 mins
Overcoming challenges to delivering the sessions	Recall the challenges identified Explore options for overcoming the challenges	50 mins
Reflection and feedback tools	Explore the purpose of the reflection and feedback tools Explore strategies to increase confidence to deliver the programme	15 mins

Session name	Overview and aims	Time
Next Steps	Give out food diaries, pedometers and activity diaries to provide experience of self monitoring and behaviour change. Explore the benefits of experiencing using the tools and self monitoring that participants will use.	15 mins

11.5.2 Evaluation of educator training

Subsequent to completion of their training, educators completed an evaluation form (see Appendix 20) for the STOP diabetes training sessions. Following their first delivery of the education programme, educators involved in delivering the sessions were interviewed by a qualitative researcher, to explore their views about the content and style of delivery, experiences from delivering the programme and perceived practical issues; feedback from these interviews is presented as part of the first pilot phase (see Chapter 9).

11.5.3 Preliminary assessment of educator behaviour

The quality development tool used to conduct preliminary assessment of educator behaviour was based on current assessment tools developed by the DESMOND collaborative;[252] which recognises that training health professionals to deliver education programmes does not always result in appropriate or consistent delivery of the programmes.[257]

The tool was developed in 2015 and consists of 5 'global' categories, each containing specific items to evaluate programme delivery; it is a revision of a previous DESMOND tool.[258] The tool is designed to be used by a separate observer who assesses the educator's behaviours when they teach/deliver education. Each item in the tool represents a discrete DESMOND behaviour which is paired with an 'opposite' item (labelled as a non-DESMOND behaviour), that are coded when observed. The observer is asked to rate which behaviour (i.e. DESMOND or non-DESMOND) is most commonly seen during the training session. For example, the first item requires the observer to assess whether the educator uses open body language to support engagement of participants (*Table 42*). DESMOND behaviours for this item include nodding and smiling at the participants and non-DESMOND behaviours include avoiding eye contact and the educator turning their back on the participants when asking a question. By noting down these behaviours when they occur, the observer can determine which behaviour was most common during the training session.

Prior to using the tool to assess educator behavior for the STOP programme, three members of the education development team reviewed the existing quality development tool, and its application in the STOP diabetes programme. As a result only one of the items in the reflective learning category was changed (see *Table 42*), because it is recognised that people with ID are more amenable to concrete concepts and visual imagery than to more abstract concepts, such as analogies,[165] (see also Chapter 8 and Chapter 9).

Between March and April 2015, a member of the research team attended the final iteration of the education programme employing the tool. The researcher was an experienced member of the research team, had attended specific STOP educator training and was involved in the development of the education programme. The tool was used to describe how the educators interacted with the group, identify differences between educators' behaviours and to assess appropriateness to this client group. The researcher used the quality development tool to conduct assessments during six out of seven of the education sessions. During these sessions the researcher positioned themselves outside of the group and completed the tool separately for each educator.

In addition to noting educator behavior, assessment of participant-educator interaction was undertaken during one observation visit, using a 10 second event coding to estimate the amount of time educators were speaking. When the beep sounded, the coder indicated on a response sheet who was talking at that point in time (whether an educator or a participant), with other activity classed as 'miscellaneous' (indicating silence, laughter or multiple conversations during learning activities). The 10-second event coding is an objective measure and an established method of measuring talk time educator ratio.[259, 260]

Table 42: Assessment items in educator behavior assessment tool

DESMOND behaviour	Non-DESMOND behaviour
Facilitating non-judgemental engagement of all participants	
Uses a range of open body language to support engagement of participants	Tends to use more closed body language behaviours
Uses non-judgemental statements regarding participant verbal utterances	Uses judgemental statements in response to participant verbal utterances
Seeks answers from a number of participants before discussing further, including right and wrong answers	Accepts the first (right) answer and/or immediately providers correct or up-to-date information
Seeks clarification of participants' contribution	Rarely seeks clarification of participants' contribution
Avoids giving general healthy messages	Provides general healthy messages
Avoids giving their own opinion	Gives their own opinion
Eliciting and responding to emotions/feelings (empathetic responding)	
Prompts participants to express and explore their feelings about diabetes	Avoids actively engaging participants in emotional discussion
Acknowledges and/or prompts exploration of participant emotional response	Retreats from/ignores/denied participant emotional response
Facilitating reflective learning	
Uses analogies*	Avoids the use of analogies*
Uses visual tools and resources	Does not use visual tools and resources
Uses and refers to participants' comments and quotes	Uses his/her own words when working through session content
Encourages group to discuss/answer their own questions	Answers most questions asked by the group
Prompts participants to explore misconceptions and gaps in knowledge	Immediately provides correct information to fill apparent gaps in knowledge

DESMOND behaviour	Non-DESMOND behaviour
Notices and prompts discussion of personal health beliefs	Avoids discussion of health beliefs within the group
Prompts all participants to ask questions about issues discussed	Rarely invites (more than once) participants to ask questions
Prompts group to summarise key messages	Tends to summarise key messages
Prompts group to summarise their own understanding	Tends to summarise what she/he thinks is the group's understanding
Prompts 'self talk' about how the key messages from the session applies to them	Does not ask participants to reflect on how the key messages apply to them
Only provides new information after group discussion	Regularly provides new information without group discussion
Behavioural change, planning and goal setting	
Acknowledges when participants decide not to make any future changes to self-care behaviours or beliefs	Expects participants to make necessary changes
Prompts participants to discuss their thoughts about possible changes	Avoids generating discussion about possible changes
Prompts participants to review the impact of possible choices on their future health	Avoids generating discussion about range of options/impact
Prompts participants to talk about what they are going to do as a result of the session	Rarely asks participants what they are going to do as a result of the session
Prompts problem solving of possible barriers to change	Avoids 'active' problem solving support
Prompts participants to reflect on their goals/plans	Avoids reflective discussion regarding goals/plans
Facilitates sharing of stories about positive attempts to manage their health	Avoids use of participant stories of positive success

DESMOND behaviour	Non-DESMOND behaviour
Supports participants to plot their results on the health profile/action plan	Provides little support to assist participants with the completion of their health profile/action plan
Prompts reflection of changes already made	Does not prompt reflection of changes made
Overall group management	
Uses strategies to manage time	Avoids using strategies to manage time
Notices tone/dynamics within the group and uses these to manage the group	Tends to ignore issues within the group
Prompts engagement of quieter participants	Avoids seeking engagement of quieter members of the group
Uses co-educator to support delivery	Appears to work alone despite opportunities that may be assisted by co-educator
Manages group to provide time and space to complete tasks	Avoids managing group to allow time and space
Provides overviews of the sessions/day	Does not provide overviews of the sessions/day
Outlines the style of the sessions	Does not outline style of sessions
Facilitates full engagement in interactive tasks	Tends to facilitate interactive tasks with only a few participants
Engages participants using rapport building skills	Avoids using rapport building skills
* The DESMOND behaviour was modified to "Does not use analogies" and non-DESMOND behaviour to "Does use analogies" for the STOP Diabetes education programme.	

11.5.4 Identifying key important educator behaviours

Once the preliminary assessment had been conducted, members of the research team met to explore the items within the tool, primarily to distinguish between educator behaviors' that were seen to be important versus those that appeared less core or essential when facilitating adult learners with ID.

11.6 Results

11.6.1 Educator training

The STOP Diabetes specific training was attended by two diabetes education specialists, four ID health care professionals and other members of the research team. It is important to note that not all of these members of the team went on to deliver the education programme.

Based on evaluation forms completed by six of the educators following the two training days, the training was universally well evaluated. Using a standard evaluation self-report measure, all of the educators agreed with the statement that they had learned new skills and believed they could apply these skills in practice. The most helpful aspects of training cited by educators included: going through the curriculum and resources; learning about the development and theoretical underpinning of the programme; and group discussions and learning from colleagues.

11.6.2 Preliminary assessment of educator behaviour

Based on findings from one observation visit conducted to assess participant-educator interaction. In general, the observer noted that the diabetes specialist (who was the trained educator) generally displayed more of the facilitation of learning behaviours, such as using open body language, seeking clarification of participants' contributions and providing overviews of the sessions; the ID nurse displayed more engagement behaviours, prompting exploration of feelings, involvement of quieter participants and facilitating full engagement in interactive tasks. The DESMOND behaviour of prompting the group to summarise the key messages was rarely observed.

11.7 Discussion

Based on initial feedback educator training was universally well evaluated. Educators believed they had learnt new skills and could apply them in practice. However, we acknowledge only trained one group of educators.

The preliminary assessment of educator behaviours has identified that different behaviours may be delivered depending on the educator. The small number of noted differences between educators in terms of behaviours may be due to professional background and training. The ID nurse may have had more specialist expertise in engaging people with greater communication and engagement needs. However this difference may have also been due to differing roles in the delivery of the programme as the diabetes educator had been the original developer of the programme and may have been focused on exploring the delivery and the content of the programme further. This work has identified what needs to be focused on for future training in this area.

This work has been a useful first step in the development of a tool that could usefully be employed in this area. Further work is required as the tool thus far has only been used on one programme by one observer. The first two iterations of the development of the STOP diabetes programme could not be assessed using the tool as the programme was still in development. However, despite this the tool allowed for preliminary assessment, it provided a structure for the and even at this early stage found variance between educators and will provide a benchmark for future work.

11.8 Recommendations

The findings from this chapter have provided a positive starting point and highlighted a number of recommendations for future work in this area. The tool is now ready for further adaptation to optimise its relevance for the target population. Possible adaptions include: 1) shortening the tool for simplicity; 2) omission of some items that are not relevant or appropriate for this client group, such as items that relate to abstract concepts and that require participants to remember or summarise the

training points. Items essential to this group have also been identified such as teaching at the group's pace and being flexible. In any future work the tool would also be specifically tailored to follow the structure of the STOP programme. The tool would then need to be tested with a wider group of educators across the delivery of a larger number of programmes.

The health care assistant and the nurses who supported the STOP programme were not included in the intervention fidelity observations. It is suggested they be included in future assessments as they are also key to the delivery of the programme and need to be demonstrating the same set of behaviours. In addition, it is recommended that in the future more than one researcher completes the intervention fidelity tool to increase the reliability of the findings.

Furthermore, the findings from this work can also feed in to recommendations for future educator training, particularly the importance of being aware of quieter participants and encouraging engagement to avoid participants feeling isolated.

CHAPTER 12. ECONOMIC ANALYSIS

12.1 Overview

In this chapter, we describe the economic work undertaken as part of work package 1, in order to estimate the cost-effectiveness of the STOP Diabetes lifestyle intervention. Development of the lifestyle intervention is described in Chapter 8 and Chapter 9.

12.2 Rationale and aims

The overall aim of the economic work undertaken was to estimate the cost-effectiveness of the STOP Diabetes lifestyle intervention, compared with current care, in reducing cardio-metabolic co-morbidities amongst individuals with ID.

The objectives of the economic analysis reflect the revised protocol for the STOP Diabetes study. The economic analysis focused on the purpose of the intervention; that is, to increase physical activity (and to a lesser extent change in dietary behaviours) amongst overweight or obese individuals with ID. The context of the analysis reflects the likely real-world implementation of the intervention, including a screening phase, which could be appended to the established health checks for people with ID which carried out annually within current NHS practice, in order to identify individuals during to identify individuals suitable for the intervention. This screening can therefore be viewed as a potential additional component of the Learning Disability Health Check (see below for more details of the Learning Disability Health Check). If the STOP Diabetes intervention were to be rolled out, it would target only those with mild or moderate ID as the intervention is not suitable for those with severe or profound ID. We therefore restrict the economic evaluation to the subset of individuals in STOP Diabetes with mild/moderate ID.

The economic analysis does not attempt to estimate the cost-effectiveness of screening individuals for IGR and T2DM due to a lack of evidence and poor clarity of

context within current clinical care and the proposed STOP Diabetes intervention. This is explained more fully in Appendix 21.

12.2.1 Context of proposed screening with existing intellectual disability care pathways

12.2.1.1 Learning Disability Health Check

Royal College of General Practitioners' guidelines state that NHS general practices should identify individuals that are a high priority for health checks, stating that mild cases of ID are a lesser priority.[261] However, the current 2015/2016 NHS General Medical Services contract states that all individuals with ID over 14 years of age should be offered a Learning Disability Health Check and we therefore do not exclude those with mild ID from the analysis.

It was assumed that the model starts at the point following attendance of a Learning Disability Health Check by all baseline individuals.

Figure 24: Logic diagram showing how screening for suitability for intervention fits within Learning Disability Health Checks shows how the screening and intervention elements the STOP Diabetes study would fit into the Learning Disability Health Check and the scope of the economic evaluation.

There is no need to explicitly model the Learning Disability Health Check process as the baseline characteristics from the STOP Diabetes study are assumed to reflect characteristics post health check and the resulting baseline risks of CVD and T2DM.

Attending a Learning Disability Health Check obviates the need to participate in the general population “NHS Health Checks” program for those over 40 years of age.

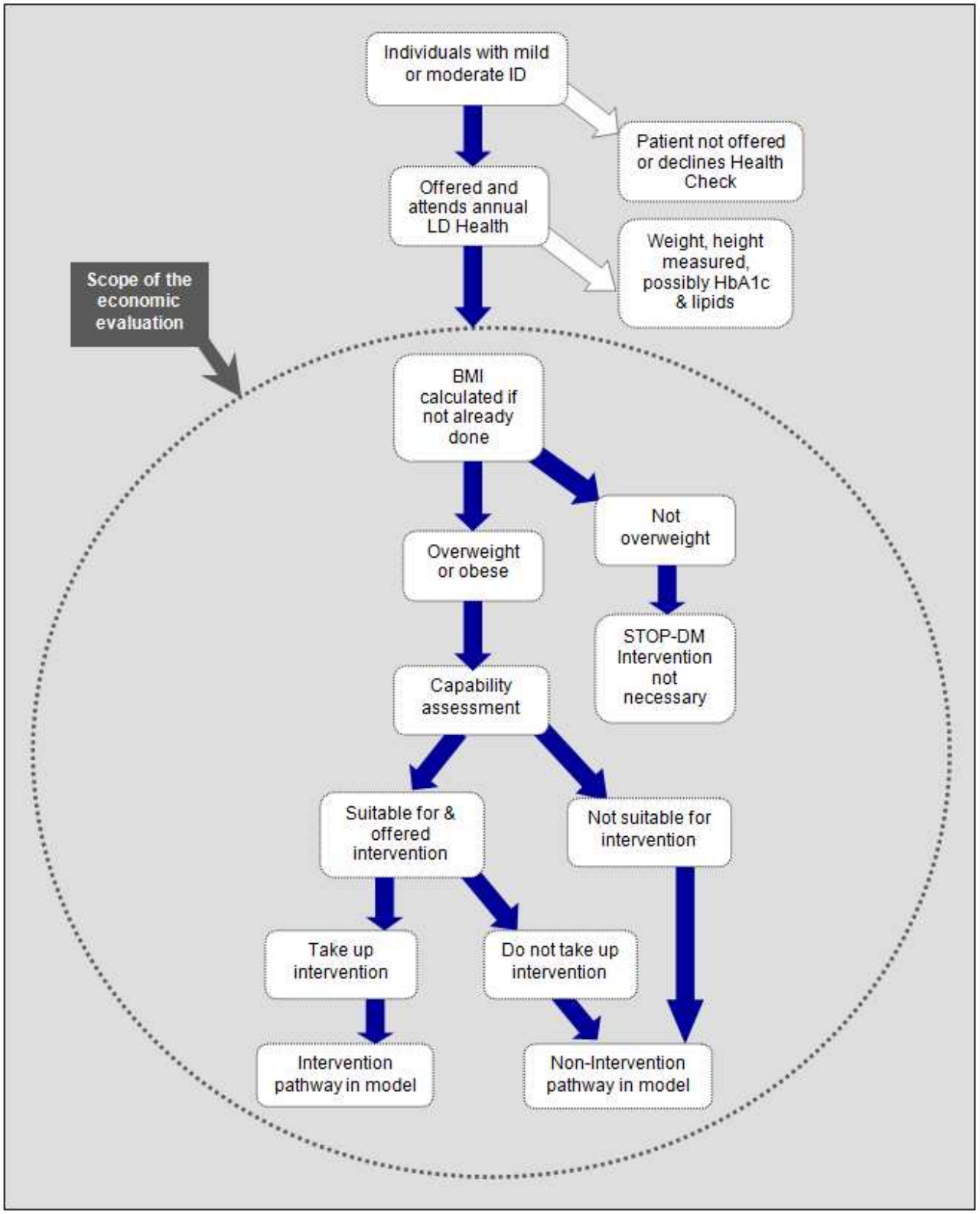


Figure 24: Logic diagram showing how screening for suitability for intervention fits within Learning Disability Health Checks

In 2013/14, nationally 44% of those on an ID register attended a Learning Disability Health Check; this varies greatly by geographical area, ranging from 10% to 60%. Specifically, 29% missed a check, implying an offer rate of 73% and an uptake rate (amongst those offered) of 44%/73% = 60%.[262]

Within the Learning Disability Health Check, BP, weight and height are measured[263] unless not possible because of physical disability (meaning the patient is unable to stand for height and/or weight measurement) or behavioural difficulties, so their BMI should be calculated. Some blood tests may also be carried out during a Learning Disability Health Check (or in advance of their Health Check, or as a follow-up test recommended by patients in their Health Action Plan). There is also scope to screen for hyperglycaemia (and dyslipidaemia) and assess cardiovascular risk as part of routine care, although there is much variation in practice as to whether such blood tests are deemed to be necessary or a priority within a Learning Disability Health Check, or indeed uptake of blood tests by individuals. For the purpose of the modelling, we assume these blood tests would fall within the Learning Disability Health Check.

The model starting point is an individual having been offered and attending for their Learning Disability Health Check; that is, one of the 44% noted above.

12.3 Methods

12.3.1 Overview of approach

This section provides an overview of the economic work before more details are described in subsequent sections.

The first phase of work involved obtaining all necessary parameter inputs and assumptions for the economic model, specifically:

1) Data from the STOP Diabetes study to provide the baseline patient characteristics.
2) Data collected and quotes and input from the study team to provide the details required to calculate the cost per participant of the STOP Diabetes lifestyle intervention.
3) Assumptions on uncertain inputs such as rate of uptake of the intervention and the durability of the benefits of the intervention through discussion with the clinical team.

The modelling itself comprised:

1) Model development - an existing economic model of cardiovascular and diabetes risk, driven by characteristics such as BMI, total cholesterol, HDL cholesterol and HbA1c was adapted to incorporate the relationship between changes in physical activity (steps) and changes in the above risk factors.
2) Modelling the screening process – determining those individuals from the STOP Diabetes study that are suitable for, and take up the intervention. The modelling takes account of individuals' BMI, capacity to participate in the intervention and rates of uptake of the intervention.

Modelling the screening process involved an adaptation of the prevention Sheffield School for Public Health Research (SPHR) Type 2 Diabetes Model. The model is used to simulate the lifetime patient clinical pathways, incidence of complications and associated cost and health utility impacts arising from an intervention compared

to routine care. The assumptions for the modelling are described in detail later in the report, but the key ones are listed here:

1) The STOP Diabetes intervention was estimated to cost £ 1,097 for the initial intervention and eight maintenance sessions, delivered within a one-year timeframe.

2) The benefits of increases in physical activity could be mapped to changes in BMI, systolic BP, total and HDL cholesterol using a relationship identified in the literature.

3) The durability of the intervention effects was uncertain so, for the modelling, two scenarios were adopted with the effects lasting (but decreasing linearly) for three and five years (from the start of the intervention) respectively.

The usual approach to economic evaluation is to estimate the incremental lifetime discounted costs and quality-adjusted life-years (QALYs) of an intervention compared with usual (routine) care. From this, the incremental cost-effectiveness ratio (ICER) can be calculated and compared with usual acceptability thresholds (£ 20,000 - £ 30,000 per QALY). However, because the clinical effectiveness of the STOP Diabetes intervention is not known, the economic modelling was primarily based on threshold analysis. Under this approach, the requisite clinical effect size needed for the intervention to be just cost-effective (given the cost of the intervention) was estimated**.** Because the STOP Diabetes intervention promotes physical activity and dietary change, the output of the threshold analyses was not a single effect size but various permutations of these that would be adequate to make the intervention cost-effective.

Uncertainty around the results was analysed primarily using one-way sensitivity analysis. Due to the computational demands of undertaking threshold analyses, probabilistic sensitivity analysis was restricted to one scenario to give an indication of the degree of uncertainty around such an intervention.

12.3.2 Data sources – STOP Diabetes

12.3.2.1 Baseline Population

The baseline population in the model reflected as far as possible the patient-level baseline data from the STOP Diabetes study during which data from 930 individuals with ID from the Leicester area were gathered. Other risk factors (left ventricular hypertrophy, heart rate, and valve disease) were based on general population prevalence as data were not available from the STOP Diabetes study.

Only individuals with mild/moderate ID would be be targeted with the STOP Diabetes intervention; thus those recorded as having severe/profound ID were removed from the sample, leaving a total of 618 individuals in the modelled cohort. Summary statistics of these individuals are shown in *Table 43*.

The initial Learning Disability Health Check itself is not simulated to prevent modification of baseline characteristics (such as diabetes diagnosis or statin treatment) before initiation of the intervention.

12.3.2.2 Data Imputation

Many individuals were lacking responses to some questions in the baseline questionnaire but had data for others. The SPHR Diabetes Prevention Model uses imputation models based upon Health Survey for England (HSE) 2011 data[264] to impute missing anthropometric and metabolic measures. Full details of imputation models can be found elsewhere in an online discussion document.[265]

12.3.2.3 Clinical effectiveness

The feasibility study was intended to assess the practicality of implementing the intervention in the target group. Sample sizes were, however, too small to quantitatively assess the effectiveness of the intervention (n=4 with before and after accelerometer data reporting step count); therefore, there were no estimates of clinical effectiveness available (see later Section 12.3.15.1 on *Threshold analysis*).

Table 43: Baseline characteristics of individuals with mild or moderate intellectual disabilities in the STOP Diabetes study

Parameter	Number (Total n=618)	Percentage	
Male	337	54.5%	
White	537	86.9%	
IMD 1 (least deprived)	109	17.6%	
IMD 2	107	17.3%	
IMD 3	125	20.2%	
IMD 4	142	23.0%	
IMD 5 (most deprived)	135	21.8%	
Non-smoker	512	82.8%	
Anti-hypertensive treatment	62	10.0%	
Statin treatment	55	8.9%	
Cardiovascular disease	12	1.9%	
Depression/anxiety	171	27.7%	
Congenital heart disease	12	1.9%	
Capable of taking up intervention	484	78.3%	
Eligible for intervention by BMI	458	74.1%	
Eligible for and capable of intervention	384	62.1%	
Parameter	**Mean**	**Standard deviation**	**Median**
Age (years)	43.07	14.15	42.32
BMI (kg/m^2)	29.25	7.36	28.10
Total Cholesterol (mmol/L)	4.91	1.02	4.80
HDL Cholesterol (mmol/L)	1.35	0.49	1.30
HbA1c (%)	5.37	0.49	5.35
Systolic BP (mm Hg)	121.60	17.67	120.00
Diastolic BP (mm Hg)	78.10	11.07	78.00
EQ-5D	0.838	0.219	0.850
Baseline physical activity in mean steps per day (N=46)	6,892	3,556	6,453

*IMD, Index of Multiple Deprivation; BP, Blood Pressure; BMI, Body Mass Index; HDL, High Density Lipoprotein; EQ-5D, EuroQol-5 Dimensions; a BMI>25 **or** BMI>23 and Black or Asian ethnicity*

12.3.3 Assessing suitability for intervention

Process:

All individuals at the start of the model were assumed to be attending the Learning Disability Health Check in the first year and therefore potentially eligible for the intervention. However, not all individuals were deemed to be at sufficient high risk or capable of receiving a lifestyle intervention. Therefore, in the intervention arm of the model, suitable individuals needed to be identified as part of the Health Check process using pragmatic selection and capacity criteria. The selection criteria for clinical need for the intervention was being overweight or obese; that is, BMI>25 (BMI>23 for individuals from black or minority ethnic groups). It was assumed that individuals were capable of taking part in the intervention if they could walk (without aids), if they did not have behaviour problems and if their ID was mild, moderate or unknown. The capabilities assessment itself was assumed to occur in all baseline individuals at the start of the model.

These criteria resulted in 62.1% of individuals with mild or moderate disability being eligible for intervention.

The proportion of individuals receiving the intervention was further reduced by taking account of the willingness of suitable individuals' to participate in the programme of intervention sessions, so the model also incorporates this rate of uptake.

12.3.4 Screening cost

In order to assess the real-world impact of the intervention if it were rolled out at scale, we assume that the above assessments would be carried out at the same time as their routine Learning Disability Health Check, as opposed to a separate appointment that occurred for the screening component of the programme. Therefore, the additional screening-related costs attributable to STOP Diabetes were

simply those to assess: i) an individual's need for the intervention; and ii) an individual's capacity to undertake the intervention (whenever increased activity is recommended). Recruitment costs from the STOP-DM study were excluded as these would be covered by the existing recruitment activity for the Learning Disability Health Check.

It was estimated that the BMI calculation, capabilities assessment, and time taken to explain and potentially gain consent for the intervention would take on average an extra 15 minutes of healthcare assistant time during the NHS Health Check compared with current care (£5.10). This cost is incurred by all individuals in the intervention arm of the model.

The process of risk assessment and any associated screening for diabetes or IGR, and any overall assessment of CVD risk is assumed to fall within the existing remit of their annual Health Check and therefore outside the scope of this economic evaluation.

12.3.5 Intervention form, cost, clinical effectiveness and uptake

12.3.5.1 Form

The costs of the intervention were divided into three phases:

A) Development phase – the costs of setting up the intervention for example the up-front costs of training the educators and equipping them to deliver the intervention

B) Delivering the initial intervention - the key components are:

- Seven sessions for patients plus an additional one for their carers
- Each session lasting 2.5 hours, plus 30 minutes set-up/pack-up time
- Three educators per group, one band 8a, one band 7 and one band 3

C) Maintenance sessions – eight monthly sessions starting after the initial intervention, as it is recognised that for lifestyle interventions to have sustained benefits, some ongoing education is needed to re-enforce behaviour change.

12.3.5.2 Cost

A micro-costing exercise was undertaken by colleagues at Leicester, assisted by the economics team, in order to obtain a cost-per-patient of receiving the intervention. As STOP Diabetes was a feasibility study, not all elements of the full cost of its delivery are known with precision. Furthermore, some cost would be different if incurred in a real-world setting. As the economic analysis is essentially a threshold analysis to inform any further study of the STOP Diabetes intervention within a trial setting, it was decided there was little point in separate costings of the intervention so a single costing was undertaken based on actual resources incurred during the study where available, but modified where appropriate to reflect the costs that would be incurred in a real-world setting and using price quotes obtained by the clinical team for some aspects of the intervention's development.

Within the STOP Diabetes research study, the average number of patients per group was six but for the costing an average number of 8 people per group was assumed as this would be an acceptable maximum number in the 'real world'.

'Research costs', such as recruiting patients to the study and initial development of the intervention, were excluded because the intervention had already been developed and it was assumed that recruitment costs were part of the existing annual health checks process for ID patients.

Costs of the components of the intervention were obtained from several sources. Unit costs of some nurse grades are available from Curtis 2014.[266] Costs for other nurse bands were obtained by combining salary costs provided by the study team with overhead adjustment in line with Curtis 2014. The costs of non-staff items were provided by the study team.

The individual cost elements of the STOP Diabetes intervention are shown in *Table 44.*

Table 44: Cost of the STOP Diabetes intervention

A. One-off costs of intervention	Costs	
Details	**Costs apportioned to**	**Cost**
Trainer costs for educator training (one-off) – initial DESMOND + intervention specific Band 7 x 6 days @ £ 353 Band 8a x 4 days @ £438 In theory, up to max 15 educators possible per group training session	Apportion to 'Educator team set-up'	£ 2,118 + £ 1,752
Trainer time for Quality Development intervention fidelity (i.e. sit in on educator delivering session) - Band 7 trainer 1.5 Days needed per educator x 3 educators @ £ 353 /day	'Educator team set-up'	£ 1,588
Educator costs - attendance at training (time and travel) – initial DESMOND + intervention specific Band 3 x 3 days @ £ 173 Band 7 x 3 days @ £ 353 Band 8a x 3 days @ £ 438 (This mix reflects a group of educators that can deliver a course together)	'Educator team set-up'	£ 519 + £ 1,059 + £ 1,314
Educator costs - prep time to deliver curriculum – each educator time to prepare before deliver their few courses Band 3 x 2 days @ £ 173 Band 7 x 2 days @ £ 353 Band 8a x 2 days @ £ 438 (This mix reflects a group of educators that can deliver a course together)	'Educator team set-up'	£ 346 + £ 706 + £ 876
Educator time for QD intervention fidelity - QD and mentorship visits (1.5 days per educator) Band 3 x 1.5 days @ £ 173 Band 7 x 1.5 days @ £ 353 Band 8a x 1.5 days @ £ 438	'Educator team set-up' Assume this is required over a cycle of 3 years (as in DESMOND)	£ 260 £ 530 £ 657
Developing training package, resources and Intervention Fidelity tools What trainers needed to train up educators. Some elements could be re-used but some could be consumed, e.g. food during training	Apportion to 'Trainer team one-off'	£1,000

Assume 50% of the estimated total £ 2,000 costs could be attributable to a single team of educators (there may be a few training providers around the country so cannot spread the cost over lots and lots of educator teams)		
Delivery materials - education curriculum 3 educators x £ 100	'Educator team set-up'	£300
Delivery materials - education resources and resources/food models per set (initial set £100, more substantive set up to £2,000) Per team of educators – assume non-re-usable	Apportion to 'Educator team set-up'	£1,000
Venue costs for educator training (one-off) - initial DESMOND + intervention specific -£100 per day x 3 days In theory, up to max 15 educators possible per group training session	Apportion to 'Educator team set-up'	£300
B. Initial educational intervention	**Costs**	
	Costs apportioned to	**Costs**
Administrative time and coordinator time (combined) – phone calls to confirm suitability and willingness - 15 mins per person but would not be needed in real world as would be part of nurse assessment; – arrange appointments, send confirmation and follow-up reminder phone call(s): 15 mins per person @ £ 21 per hour (estimated on average £18 per hour administrator, £24 per hour coordinator). – booking/confirming venue (30 mins per group) – coordinating educators and resources (60 mins per session x 8). Assuming £21 per hour average salary costs	per participant per 8-session course per 8-session course	£ 5.25 £ 10.50 £ 168
Delivery 7 sessions to patients + 1 carer session = 8 sessions		

Each session = 2.5 hours delivery + 0.5 hour set-up/pack-up time per session = 3 hours per session[a] Each session run by 2 educators + 1 healthcare assistant (1 x Band 3 + 1 x band 7 + 1 x band 8a). Band 3 x 3 hours @ £ 23 x 8 sessions Band 7 x 3 hours @ £ 47 x 8 sessions Band 8a x 3 hours @ £ 58 x 8 sessions	per 8-session course	 £ 552 £ 1,128 £ 1,392
Participant Handbook	Included under cost of course materials below	
Pedometer (1 per person +50% for carers, at £8 each, assume need 10 per group + 20% for loss) Total per group if 7, allow 13 pedometers	per 8-session course	£ 104
Refreshments £1 per person, plus 50% for carers, for 8 weeks Total per group if 7	per 8-session course	£ 84
Postage/phone calls Total per group = £10	per 8-session course	£ 10
Stationary and reprographics – letters/information/course materials £ 10 per person Total per group	per 8-session course	£ 70
Venue Hire Cost Could be NHS premises (in which case costs absorbed into Curtis rates) Could be non-NHS community (Local Authority) - some would charge Assume 50% of venues not in NHS & incur a charge 3 hours x 8 weeks, £10-20 per hour (assume £15)	per 8-session course	£ 360
Travel costs - staff (45p per mile for 3 educators x 8 sessions, return mileage estimated at 10 to 30 miles depending on distance from base)	per 8-session course	£36.00
Travel costs – In STOP Diabetes, participants (taxi travel or re-imbursement of bus -fare, assuming 50% of people (3-4 per group) need travel costs paying, estimated £10-20 taxi & £2.00-3.00 bus fare per journey)	–	£ 0

In real world, however, it can be assumed that such travel costs me all be accounted for with the patient's free bus pass and/or mobility allowance		
C. Monthly ongoing support sessions	**Costs**	
8 monthly sessions The cost of delivering these sessions is uncertain. They could be delivered one-to-one or in a group. Although group-based delivery would normally be cheaper for lifestyle interventions, for individuals with ID this might not be the case. Delivering maintenance might be achievable though a single educator visiting the patient's home (thereby avoiding venue costs), and provision of a shorter hour long one-to-one session might be sufficient. It was therefore judged to be a conservative approach to allow for maximum potential costs by costing on the assumption of group-based delivery and cost of a maintenance session was assumed to be the same as an initial session		As per section B. above

'Educator team set-up' in the 'Costs apportioned to' column refers to costs to train up a team of 3 educators

a No staff time for travel to courses included as could be on-site or if straight from home, would not be reimbursed if no further than to workplace)

To obtain an overall cost per patient, a sequential process of apportioning 'Educator team set-up' to educators, then educator costs to a cost per course, then to course costs to patients was undertaken, giving a cost per patient of £ 1,097 (combined cost for the initial and maintenance intervention).

Potentially, there might be some scope for either reducing the cost of the intervention or actual costs incurred being lower than estimated above. For example, the mode or frequency of delivery of maintenance sessions could be re-visited, a different mix of grades of educators may be possible in the real world, some individuals that attend

the initial course may not continue to the maintenance sessions, and the method of apportioning overhead costs within Curtis is somewhat arbitrary (appears to load greater overhead costs to more senior staff). Through a cost-specific threshold analysis, we explored to what degree the cost of the intervention would need to be reduced in order to make it cost-effective.

12.3.5.3 Clinical Effectiveness

Due to the small number of individuals in the feasibility study with data on physical activity (as measured using an accelerometer) before and after the programme (n=4), effectiveness estimates were not available from the STOP Diabetes programme itself. Therefore, a threshold analysis approach (see Section 12.3.15.1) was used to estimate the threshold for the effect size needed to make the intervention marginally cost-effective in the £20,000-£ 30,000 cost per QALY range as recommended by NICE.[267] As the intervention includes dietary advice as well as the physical activity element, we report the threshold in terms of possible permutations of the number of steps and the additional diet-attributable BMI and systolic BP changes that would be needed to be able to demonstrate that the intervention is cost-effective.

12.3.5.4 Durability of effect

The initial intervention-related changes in BMI, systolic BP and HDL cholesterol within the year of intervention were subsequently assumed to wear off linearly such that, after three years from the start of the intervention, individuals have returned to the BMI, systolic BP and HDL cholesterol trajectories that they would have followed in the absence of intervention. Alongside the above base case assumption, an additional scenario analysis was carried out to test the response of the results to five-year duration of effect.

12.3.5.5 Intervention uptake

Of individuals that are both eligible for and capable of taking up the intervention, we assumed that 55% do so and thereby incur the full costs of the intervention based on advice for a previous lifestyle intervention evaluation.[268]

12.3.6 Modelling the benefits of changes in physical activity (steps)

The economic model chosen for the modelling (the SPHR Diabetes Prevention model described later) does not include physical activity as a risk factor for CVD or diabetes. However, the primary measure of interest, in terms of informing any future follow-on trial from the STOP Diabetes study, is change in steps per day (and this is a key output of the threshold-based economic analyses, see Section 12.3.15.1, and any subsequent full trial would be based around change in physical activity measured in steps using a pedometer). It was therefore necessary to identify a mechanism to map changes in physical activity to changes in the above risk factors that already exist in the model (BMI, systolic BP, total and HDL cholesterol and glucose-related risk factors), and vice versa.

12.3.6.1 Rationale for surrogate-based model

A surrogate-based approach was chosen because:

1) The SPHR Diabetes Prevention model uses metabolic trajectories to model long-term progression of risk factors and incidence of co-morbidities
2) A meta-analysis was identified which linked changes in physical activity to changes in systolic BP, total and HDL cholesterol
3) We are unaware of any meta-analyses of hazard ratios for the effect of changes in physical activity on incidence of CVD and diabetes
4) Although we were made aware of a study linking physical activity to CVD (Yates 2014; NAVIGATOR Trial[269]), the analysis was undertaken assuming a constant change in steps over a six-year period, whereas the effect of the STOP Diabetes intervention would decline over time. So any hazard-ratio adjustments to the risks of CVD and diabetes would need to have been analysed in a time-dependent way. The behavioural changes in this study were also based on dietary advice but it is unknown how intensive this was compared to the STOP Diabetes

intervention (which contained some dietary advice but no weight loss target). The reported hazard ratio may therefore include significant risk reduction attributable to dietary changes. Additionally, this study recruited a cohort of individuals with IGR at baseline and only reported CVD (not diabetes).

We did, however, believe that it would be useful to compare the predictions of the model (modified to reflect a constant changes in risk factors for six years) with the six-year results reported in Yates 2014. For an increase in activity of 2,000 steps, Yates et al reported a hazard ratio of experiencing a cardiovascular event over the following 6 years of 0.92 (95% CI = 0.86-0.99), that is a risk reduction of 8%. The hazard ratio from our adapted model was 0.95 (5% risk reduction). The details of the method behind this comparison are provided in Appendix 22.

There are various reasons why a slightly lower reduction in risk might be expected from the model compared to that observed in the NAVIGATOR trial. First, patients in the NAVIGATOR trial had IGR but were not necessary overweight, whereas the baseline population in the model were mostly normal glucose tolerant but with BMI above defined thresholds. BMI and diabetes both input into cardiovascular risk, but the relative effect this population difference might have on cardiovascular outcomes is unclear. Secondly, the trial participants received both exercise and dietary advice, but potential differences in diet between individuals with different step counts was not accounted for in the trial analysis. It is possible that individuals who follow exercise advice are more likely to follow dietary advice too and so may show a greater effect on metabolic trajectories than accounted for in our analysis based on steps alone. This would result in the model underestimating the results of the trial as is indeed observed. Thirdly, there are some differences in the events picked up in the trial and those accounted for in the model (e.g. transient ischaemic attack and stable angina are in the model but not the trial), although it is unclear what effect restricting outcomes to those used in the trial would have on the hazard ratio. Finally, it cannot be ruled out that exercise may impact positively upon factors not included in the model, which in turn result in reduced cardiovascular risk.

It should be noted that since the Yates 2014 study reports hard outcomes in terms of cardiovascular events but not changes in biomarkers of risk, the results could not be used to parameterise the SPHR model (which estimates a range of outcomes, including CVD, based on cardio-metabolic risk factors and their projected trajectory over time).

12.3.6.2 Review of Relationship between physical activity (steps) and risk factors for cardiovascular disease and diabetes in the model

To identify a suitable mapping between steps and model risk factors, a targeted review of the published literature was carried out using a search of the on-line PubMed database, a subsequent citation search, and advice from clinicians. This evolved into a 3-step process as described below.

Step 1: Search strategy for physical activity studies reporting step as an outcome

The search strategy using the PubMed publications database is detailed in Appendix 23 (*Table 66*). The search yielded 153 results, from which 54 abstracts were selected based on titles. Of these, 19 were relevant, of which full text was available for 14. One of these studies, Stuckey 2011,[270] discussed a meta-analysis undertaken by Bravata and colleagues,[271] which is discussed below.

Step 2: Citation search for review papers citing Bravata 2007

This search identified 22 abstracts for review. Abstract/title sifting led to 7 full text papers being checked. Of these 7, 4 were excluded as narrative reviews only, 1 was a review of reviews but focussed on diet only or diet and physical activity interventions and therefore was not relevant, 1 was excluded because it was a primary trial not a review or meta-analysis. 1 was excluded because it reported only weight and no metabolic outcomes. This search identified two relevant studies of potential use for the modelling, Murphy[272] and Qui 2014,[273] which were reviewed in full text and are discussed below.

Step 3: Conversation with clinical advisors

In discussion with clinicians it was determined that cholesterol changes were also very likely to be observed as a result of the intervention. In this respect, we were referred to a study by Camhi and colleagues[274] which is described below. This conversation also highlighted the Yates 2014 study[269] already discussed earlier.

Description of key studies

Bravata 2007 reviewed and combined the results of studies which used a pedometer to measure physical activity and reported health outcomes (mean duration of studies = 18 weeks). The overall step change induced by intervention studies was 2,491 additional steps per day. Significant reductions were reported in BMI (-0.38 kg/m^2) when 18 studies (n=562) were combined and in systolic BP (-3.8mmHg) when 12 studies (n=468) were combined. There were also non-significant reductions in lipids with, a reduction in total cholesterol of 0.09mmol/l and an increase in HDL cholesterol of 0.06mmol/l.

Murphy 2007[272] was a review of walking interventions on metabolic risk factors, based on minutes per week rather than steps. It reported very similar outcomes to Bravata et al. (188 additional minutes per week was associated with 0.95kg weight loss, 0.28 BMI reduction and 1.54mmHg Diastolic BP reduction).

Qui 2014 was a recent meta-analysis of step-counting and its effects on HbA1c control.[273] Although it found evidence of significant increases in steps it found no strong evidence for changes in HbA1c. This backs up the results of the previous Bravata review which only found significant changes in BMI and BP.

Camhi 2011[274] was a cross-sectional study based on the US NHANES survey (National Health and Nutrition Examination Survey). This study assessed the relationship between activity (steps per day) and cardio-metabolic risk factors, including cholesterol. Data for 1,371 adults were analysed and significant changes were observed in triglycerides, HDL cholesterol and waist circumference for each

1,000 additional steps achieved, but does not report BMI changes or BP. The study reported results as an odds ratio of 0.91 (95% CI 0.81 – 0.96) of having HDL cholesterol above the cut-off of 1.03 mmol/L (men) or 1.2mmol/L (women).

For the purpose of parameterising the model, this study was not preferred over the evidence from the Bravata study because it is only a single study and is cross-sectional whereas Bravata is a meta-analysis of intervention studies.

12.3.6.3 Implementation of mapping steps to risk factors

Physical activity and the mapping are not contained within the simulation model itself. The estimation of the number of steps needed is calculated manually after the model has been run several times and a threshold analysis has identified the degree of BMI change needed for the intervention to be marginally cost-effective (see section 12.3.15.1 on 'Threshold analysis' for more details).

Table 45 shows the mapping between physical activity and risk factors obtained from Bravata.

Table 45: Effectiveness outcomes from Bravata for the mean increase of 2,491 steps per day

Risk factor	Mean change	95% C.I.
BMI (kg/m^2)	-0.38	- 0.05 to - 0.72
Systolic BP (mmHg)	-3.8	- 1.7 to - 5.9
Total cholesterol	-0.09	- 0.32 to 0.15
HDL cholesterol	0.06	- 0.012 to 0.14

12.3.7 Model - Overview and Structure

The SPHR Diabetes Prevention Model is an individual patient simulation model, written in the programming language R, which was built to enable evaluation of a

wide range of different diabetes prevention and weight loss interventions in the general population. The model was originally developed using a new conceptual modelling framework for complex public health models,[275] in collaboration with a project stakeholder group comprising health economists, public health specialists, research collaborators from other SPHR groups, diabetologists, local commissioners and lay members. A review of existing diabetes prevention models was undertaken to inform conceptual model development,[276] resulting in the model including multiple diabetes risk factors (in particular both BMI and IFG) and complications of diabetes and obesity.

The model has been adapted to evaluate the outcomes of an intervention to promote physical activity in high risk subgroups of an ID population. Due to limited data about care pathways, disease risk and utility values in ID populations, much of the model is based on general population data; however, where possible data from ID populations has been used.

The model is based upon individual longitudinal trajectories of metabolic risk factors (BMI, systolic BP, cholesterol and HbA1c). For each individual, yearly changes in these risk factors occur, dependent upon the individuals' baseline characteristics.

Illustrated (see *Figure 25*) is the sequence of updating clinical characteristics and clinical events that are estimated within a cycle of the model. This sequence is repeated for every annual cycle of the model. The first stage of the sequence updates the age of the individual. The second stage estimates how many times the individual attends the GP. The third stage estimates the change in BMI of the individual from the previous period. In the fourth stage, the change in glycaemia is estimated using different statistical models depending upon whether or not they have been diagnosed with diabetes (see below). In stages five and six the individual's BP and cholesterol are updated. In stage seven, the individual may undergo assessment for diabetes, hypertension and dyslipidaemia during a GP consultation. From stage eight onwards the individual may experience cardiovascular outcomes, diabetes related complications, cancer, osteoarthritis or depression. If the individual has a history of cardiovascular disease, they follow a different pathway in stage eight to

those without a history of cardiovascular disease. Individuals with HbA1c greater than 6.5% are assumed to be at risk of diabetes related complications. Individuals who do not have a history of cancer are at risk of cancer diagnosis, whereas those with a diagnosis of cancer are at risk of mortality due to cancer. Individuals without a history of osteoarthritis or depression may develop these conditions. Finally, all individuals are at risk of dying due to causes other than cardiovascular or cancer mortality. Death from renal disease is included in the estimate of other-cause mortality. The time horizon of the model is the lifetime of all baseline individuals.

Cardiovascular events are modelled using the QRISK2 algorithm, more details are provided in Appendix 24 (*Table 67* and *Table 68*). The model uses risk equations from the UK Prospective Diabetes Study (UKPDS) Outcomes model to estimate the occurrence of major events relating to microvascular complications, including renal failure, amputation, foot ulcer, and blindness.[277, 278]

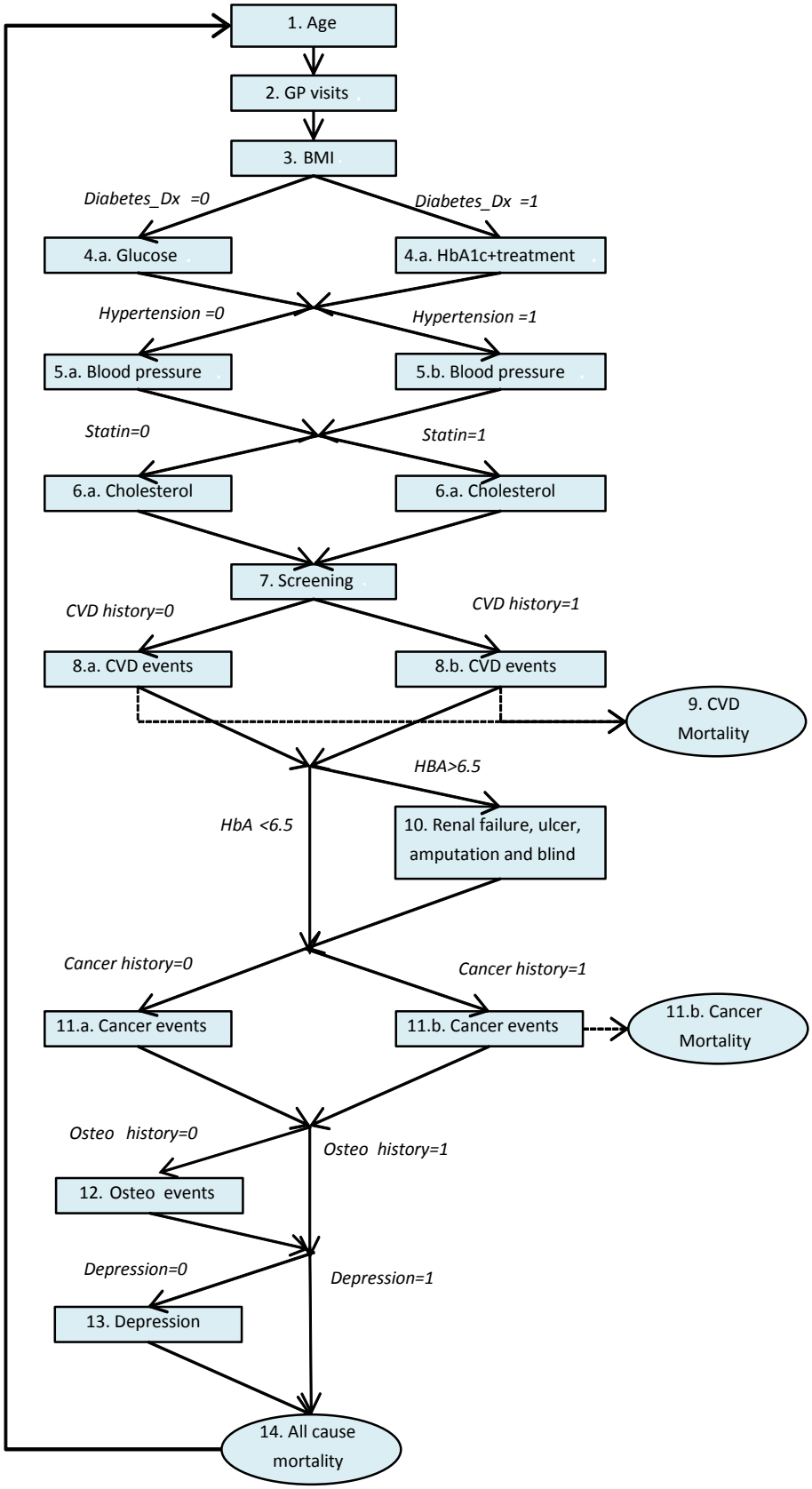


Figure 25: Model Schematic of risk of co-morbidities

12.3.8 Routine care – components of cardiovascular risk reduction

Both intervention and comparator arms of the model need to include any screening for hyperglycaemia and high CVD risk that is carried out routinely in clinical practice. This may be through the Learning Disability Health Check or opportunistic screening.

12.3.8.1 Learning Disability Health Checks

Individuals with ID should be invited to an annual health check where they undergo screening for hypertension, high cardiovascular risk and diabetes amongst other conditions. Although uptake of health checks amongst people with ID is only 44%, it was assumed that at baseline, all individuals had been identified through attending a Learning Disability Health Check and would therefore be very likely to attend future health checks. It was therefore assumed that all eligible individuals would attend annual Health Checks. Individuals that have been diagnosed with diabetes, CVD or if they are taking statins or anti-hypertensives do not continue to receive health checks, as they receive extra GP care specific to their diagnosis.

Not all individuals consent to blood tests as part of their Learning Disability Health Check. It was assumed that 33% of individuals would never consent to blood tests during a health check (based on uptake of blood tests and availability of results for the screening programme, presented in Chapter 5); therefore, they could not be screened for CVD risk or diabetes by this method. However, it was assumed that if such individuals met the criteria for opportunistic diagnosis (see below), they would consent to blood tests and so could be diagnosed through this means. A different cost for a Learning Disability Health Check was used for individuals who do or do not consent to blood tests (see Costs section).

12.3.8.2 General practitioner attendance and opportunistic screening

Frequency of GP visits (separate from NHS health checks) was simulated in the dataset for two reasons; first, to estimate the healthcare utilisation for the ID population without diabetes and CVD and secondly, to predict the likelihood that

individuals participate in opportunistic screening for diabetes and elevated risk factors for CVD. This is important as many individuals in the model cannot be diagnosed through annual health checks, either due to ineligibility or because they do not consent to blood tests as part of their Learning Disability Health Check.

It was assumed that GP attendance in the ID population occurs at the same frequency as in the general population. However, for cost purposes, consultations were assumed to take 40% longer than the general population average (see Costs section). A model of GP attendance conditional on age, sex, BMI, ethnicity, and health outcomes was derived from analysis of wave 1 of the Yorkshire Health Study and is described elsewhere.[265]

12.3.9 Long-term longitudinal trajectories of metabolic factors

The SPHR Diabetes Prevention model[265] is based upon individual longitudinal trajectories of metabolic risk factors (BMI, latent blood glucose, total cholesterol, HDL cholesterol and systolic BP), derived from statistical modelling of the dataset from the Whitehall II cohort study.[279] The statistical modelling uses parallel latent growth modelling to incorporate correlations and associations between risk factors that impact upon long term risk profiles. An advantage of the parallel growth analysis is that it is possible to estimate the effect of growth in BMI on the other metabolic risk factors so, for example, a change in an individual's BMI will result in an indirect change in their HbA1c trajectory. Growth factors are also conditional on several individual characteristics including age, sex, ethnicity, smoking, family history of CVD, and family history of type 2 diabetes. It is also possible to estimate correlation between changes in underlying glycaemia (measured by HbA1c), systolic BP, total cholesterol and HDL cholesterol. Full details of this analysis are currently described in an on-line Discussion Paper[265] with journal publication expected soon.

The characteristics of the Whitehall II cohort (civil servants living in London) are likely to differ significantly from that of the STOP Diabetes ID population. However, there are, to our knowledge, no available longitudinal surveys of ID populations on which to base a similar analysis, and no other analysis of metabolic trajectories takes into

account the correlations between risk factors that make the Whitehall model so powerful. Importantly, the baseline values for the metabolic risk factors do come from the STOP Diabetes ID population, with the Whitehall-based trajectories being used simply to describe the expected changes in metabolic values over time.

If an individual in the model is diagnosed with diabetes, or starts treatment with anti-hypertensives or statins, trajectories alter to reflect the expected changes due to treatment. The criteria for opportunistic screening and diagnosis of diabetes, hypertension and high CVD risk can be found in Appendix 25 (*Table 69*), together with details of changes in metabolic trajectories.

12.3.10 Risks of mortality - raised risk in individuals with intellectual disabilities

In every model cycle individuals within the model are evaluated to determine whether they experience a fatal event or mortality. The evidence for risk of mortality in individuals with ID compared to the general population is described below, analysed by cause, that is, CVD, cancer and other-causes.

Cardiovascular mortality:
CVD mortality is included as an event within the estimated CVD risks calculated by the QRISK2 score as described in the cardiovascular disease section below. There is some evidence for an increased risk of CVD mortality in individuals with ID,[280] but other studies report no difference or even reduced risk compared to the general population (see results of meta-analysis presented in Chapter 2). It is also unclear whether these differences in mortality risk are due to differences in risk factors included in the QRISK or due to other factors. It was assumed for the purposes of the model that any differences in cardiovascular mortality between individuals with ID and individuals in the general population occur simply due to differences in risk factors.

Cancer mortality:

It was assumed that risk of cancer and subsequently cancer mortality would be the same in an ID population as in the general population, taking into account individual differences in risk factors. This is supported by several studies looking at mortality rates from various causes in ID populations.[280, 281]

Other cause mortality:
This describes the risk of death from any cause except cardiovascular disease and cancer. This was derived from all-cause mortality rates by age and sex; extracted from the Office of National Statistics (ONS).[282] The mortality statistics report the number of deaths by ICD codes for 5-year age groups. To obtain other cause mortality, the number of cardiovascular disease, breast and colorectal cancer related deaths were subtracted from the all-cause mortality total.

There is good evidence from various sources that the rate of all-cause mortality is higher in individuals with ID,[280, 283, 284] particularly due to excess deaths from respiratory disorders, neurological diseases, congenital abnormalities and accidents. Standardised mortality ratios (SMR) of 2.28 (95% CI 2.02-2.56) for ID men and 3.24 (95% CI 2.83-3.69) for ID women compared to the general population, reported in a 14 year study of individuals from the Leicester area,[280] were applied to the other cause mortality rates derived from the ONS data. The SMRs were not adjusted upwards to take into account the minimal increase and decrease in cancer and CVD mortality respectively in ID populations as it was unclear how large this adjustment should be and it was expected to make little difference to the outcomes.

The rate of other cause mortality by age and sex was treated as the baseline hazard. An increased risk of mortality was assigned to individuals with diabetes using data from a published meta-analysis.[285] This study used data from 820,900 people from 97 prospective studies to calculate hazard ratios for cause-specific death, according to baseline diabetes status.[285] Cause of death was separated into vascular disease, cancer and other cause mortality. From this study it was estimated that individuals with a diagnosis of diabetes have a fixed increased risk of other cause mortality (Hazard ratio 1.8 (95% CI 1.71-1.9)). The estimates reported in the meta-analysis

include increased risk of death from renal disease; therefore mortality from renal disease was not simulated separately to avoid double counting of benefits.

12.3.11 Comorbid outcomes with no excess risk in individuals with intellectual disabilities

In every model cycle individuals within the model are evaluated to determine whether they have a clinical event. In each case within the simulation, risk equations estimate the probability that an individual has an event, and a random number is drawn to determine whether the event occurred.

12.3.11.1 Cardiovascular Disease

The QRISK2 model was chosen to estimate cardiovascular risk and incidence as it is a validated model based on a UK population.[286] Probability of a first cardiovascular event in the next year (including cardiovascular mortality) is calculated conditional on ethnicity, smoking status, age, BMI, ratio of total/HDL cholesterol, deprivation score, atrial fibrillation, rheumatoid arthritis, renal disease, hypertension, diabetes, and family history of cardiovascular disease. Coefficients for the QRISK2 model can be found in Appendix 24 (*Table 67* and *Table 68*). The QRISK2 assumptions regarding the relationship between diabetes and CVD were modified to reflect observations from the UKPDS and the European Prospective Investigation into Cancer and Nutrition (EPIC) that HbA1c (rather than diabetes) increases the risk of MI and Stroke in a linear manner.[277, 287]

The STOP Diabetes baseline data had no information about atrial fibrillation; however it did have a category representing unspecified other heart conditions, which was recorded for 10 individuals (1.6% of total population). A diagnosis of atrial fibrillation was randomly assigned to 7 of these individuals in line with an audit of patients with ID in North Essex, which found that 1.1% of individuals had atrial fibrillation.[288] The STOP Diabetes baseline data also had no specific questions about rheumatoid arthritis or renal disease. However, one individual was noted to suffer from rheumatoid arthritis using the other health problems variables and no individuals were noted to suffer from renal disease so we assumed no renal disease at baseline.

There is conflicting evidence about whether individuals with ID have a greater risk of CVD than is seen in the general population, and if so, whether this can be accounted for through the risk factors already incorporated into the QRISK2 model. Whilst some studies have found an increase in prevalence of CVD,[83] or CVD mortality[280] in ID individuals, other studies report no difference or reduced risk compared to the general population. (see results of meta-analysis presented in Chapter 2). Lower CVD mortality compared to the general population could partially be explained through competing risks, given that individuals with ID have higher mortality due to other causes, particularly respiratory illnesses, congenital abnormalities, neurological disorders and accidental injury.[280, 281]

The model estimates that individuals recruited into the STOP Diabetes study have a much lower incidence of CVD than unmatched (for age) individuals from the HSE 2011 due to their baseline characteristics (see *Table 46*). This cannot be explained solely by the five year lower mean age of the ID cohort (for example, compare year 10 in the ID cohort with year 5 in the general population cohort). Given the lack of a clear consensus over CVD risk in individuals with ID, it was assumed that the QRISK2 equations were suitable for use in the ID population, and that any differences in CVD risk compared to the general population would be accounted for through the differences in baseline risk factors.

All CVD events modelled using QRISK2 are assigned to a specific diagnosis according to age and sex specific distributions of cardiovascular events reported in a previous Health Technology Assessment (HTA) for statins.[289] Events are also split into fatal versus non-fatal ones.

Table 46: Modelled within-year cardiovascular disease incidence: STOP Diabetes cohort vs. Health Survey for England 2011 (general population) cohort

CVD Incidence (per 10,000)	Year 1	Year 5	Year 10	Year 15	Year 20	Year 25	Year 30	Year 35
ID cohort	37	56	63	83	113	124	146	162
General population cohort	77	91	101	123	144	158	169	195
Relative risk with ID	0.48	0.61	0.62	0.68	0.78	0.78	0.86	0.83

Relationships between risk factors and different types of CVD (e.g. hypertension being more of a risk for stroke) are not incorporated into the model.

12.3.11.2 Congestive Heart Disease

Congestive heart failure was coded as a separate cardiovascular event using the Framingham risk equation[290] because it is not included as an outcome of the QRISK2. The Framingham equation is not ideal as it is based on a US population from the 1990s and there is no evidence for its accuracy in representing risk in an ID population. However, it was thought to be the best option in the absence of data specific to an ID population. The equation includes age, diabetes diagnosis (either formal diagnosis or a HbA1c>6.5), BMI, systolic BP, congenital heart disease, left ventricular hypertrophy, heart rate and valve disease to adjust risk based on individual characteristics. Full details of the equation coefficients can be found in an on-line Discussion paper.[265]

No baseline information was available about three of these risk factors (left ventricular hypertrophy, resting heart rate and valve disease) therefore they could not be included in the model to predict congestive heart disease. The baseline odds of congestive heart disease were adjusted to reflect the expected prevalence of these symptoms; this was done using general population data as data specific to individuals with ID could not be identified. The heart rate for men was assumed to be

63.0bpm and for women 65.6bpm based on data from previous Whitehall II cohort analyses.[291] The proportion of the UK population with left ventricular hypertrophy was assumed to be 5% in line with previous analyses of the Whitehall II cohort.[292] The prevalence of valve disease was estimated from the Echocardiographic Heart of England Screening study.[293] Twelve baseline individuals from the STOP Diabetes study suffered from congenital heart disease (1.9%). This is higher than the prevalence of congenital heart disease in the general population (0.80% of live births),[294] and is unsurprising given the high proportion of individuals with Down's and Williams syndrome who are known to suffer from congenital heart defects.[295] This means that the risk of congestive heart failure is higher in individuals with learning difficulties than in the general population.

12.3.11.3 Microvascular complications of diabetes

UKPDS data, derived from a UK diabetic population,[277, 278] was used to estimate the incidence of major microvascular complications including ulcer, amputation, renal failure and blindness in individuals with HbA1c ≥ 6.5, whether or not they are diagnosed with diabetes. Earlier stages of microvascular complication were not included in the model as they have less of an impact on costs and utilities. It was assumed that risk of microvascular complications would be the same in an ID population as in the general population, taking into account individual differences in risk factors.

The UKPDS outcomes model v2 includes four statistical models to predict foot ulcers, amputation with no prior ulcer, amputation with prior ulcer and a second amputation[277]. In order to simplify the simulation of neuropathy outcomes, the models for first amputation with and without prior ulcer were consolidated into a single equation. UKPDS outcomes model v2 was also used to estimate the incidence of blindness, whereas the UKPDS outcomes model v1 was used to estimate the incidence of renal failure.[278] Early validation analyses identified that the UKPDS v2 model implemented in the SPHR model substantially overestimated the incidence of renal failure. Details of the models used are reported elsewhere in an on-line Discussion Paper.[265]

All equations incorporate a coefficient for age at diabetes diagnosis. This was multiplied by age in the current year if the individual had not been diagnosed with diabetes or by the age at diagnosis if the individual had received a diagnosis. The expected values for the risk factors not included in the SPHR model (heart rate, white blood count, micro-/macroalbuminurea, peripheral vascular disease and atrial fibrillation) were taken from figure 3 of the UKPDS publication in which these are described,[277] with the exception of peripheral vascular disease, which was assumed to affect 2% of the population. The baseline risk was modified to take account of these mean values.

12.3.11.4 Cancer

Breast cancer and colorectal cancer risk are related to BMI and so were included in the SPHR model.

Incidence rates for breast and colorectal cancer in the UK were estimated from the EPIC cohort. This is a large multi-centre cohort study looking at diet and cancer. In 2004 the UK incidence of breast cancer by menopausal status was reported in a paper from this study investigating the relationship between body size and breast cancer.[296] A second paper from EPIC reported the UK incidence of colorectal cancer by gender.[297] Incidence rates in the model for breast and colorectal cancer are shown in *Table 47*.

A large meta-analysis that included 221 prospective observational studies has reported relative risks of cancers per unit increase in BMI, including breast cancer and colorectal cancer.[298] A risk adjustment was included in the model so that individuals with higher BMI have a higher probability of pre-and post-menopausal breast cancer or colon cancer (see *Table 47*). In the simulation, the incidence of breast and colorectal cancer was adjusted by multiplying the linear relative risk by the difference in the individual's BMI and the average BMI reported in the EPIC cohort.

Table 47: UK colorectal/breast cancer incidence

Cancer Type and sub-group	Number of Cases	Person Years	Mean Age	Mean BMI	Incidence rate per person-year
Breast Cancer					
UK pre-	102	103,115	NA	24	0.00099
UK post-	238	842,15	NA	24	0.00283
Colon Cancer					
Male	125	118,468	53.1	25.4	0.00106
Female	145	277,133	47.7	24.5	0.00052

Evidence suggests that mortality from breast and colon cancer occurs at a similar rate in ID populations as in the general population.[280, 281] Cancer mortality rates were obtained from the ONS.[299] The ONS report one and five year net survival rates for various cancer types, by age group and gender. Net survival was an estimate of the probability of survival from the cancer alone. It can be interpreted as the survival of cancer patients after taking into account the background mortality that the patients would have experienced if they had not had cancer.

Table 48: Relative risk of colon/breast cancer per unit increase in body mass index

Cancer Type and sub-group	Mean relative risk	2.5^{th} C.I.	97.5 C.I.
Breast Cancer			
UK pre-menopause	0.89	0.84	0.94
UK post-menopause	1.09	1.04	1.14
Colon Cancer			
UK pre-menopause	1.21	1.18	1.24
UK post-menopause	1.04	1	1.07

CI, Confidence Interval

The age-adjusted 5-year survival rate for breast cancer and colorectal cancer were used to estimate an annual risk of mortality assuming a constant rate of mortality. It was assumed that the mortality rate does not increase due to cancer beyond five years after cancer diagnosis. The five year survival rate for breast cancer is 84.3%, which translated into a 3.37% annual probability of death from breast cancer. The five year survival rate for persons with colorectal cancer is 55.3%, which translated into an 11.16% annual probability of death from colorectal cancer.

12.3.11.5 Osteoarthritis

Osteoarthritis is related to BMI and diabetes, and so was included in the SPHR model. It was assumed that risk of osteoarthritis would be the same in an ID population as in the general population, taking into account individual differences in risk factors. The Bruneck cohort, a longitudinal study of inhabitants of a town in Italy reported diabetes and BMI as independent risk factors for osteoarthritis.[300] The data used to estimate the incidence of osteoarthritis is reported in *Table 49*.

Table 49: Incidence of osteoarthritis and estimated risk factors

Risk factor	Hazard ratio	2.5th C.I.	97.5th C.I.
Diabetes	2.06	1.11	3.84
BMI	1.076	1.023	1.133
73 cases of diabetes, mean BMI 24.8; 13835 person years; incidence rate 0.00053			

12.3.11.6 Depression

The SPHR Diabetes model includes depression as a health state due to its relationship with diabetes, but does not model its severity. Further details are available on-line.[265]

12.3.12 Economic Impact: Utilities

12.3.12.1 Baseline Utility

Baseline utilities for all individuals in the model were extracted from the STOP Diabetes study. The tariffs for the responses to the 3 level EQ-5D were derived from a UK population study.[301] Baseline utility was assumed to decline due to ageing as has been found in general population studies. An absolute decrement of 0.004 per year is applied in the model; this is based on previous HTA modelling in cardiovascular disease.[289]

12.3.12.2 Body mass index and utility

It was assumed that changes in BMI will impact on the utility of an individual with ID in the same way as for an individual in the general population. In a previous modelling of diabetes prevention, weight loss from education interventions was associated with an increase in utility of 0.0025 per kg change in weight.[302] This estimate was derived from weight loss trial data in which all participants were overweight or obese. In the ID population a large proportion of individuals are normal or underweight so it would not be appropriate to extrapolate the effects of weight loss on utility to these individuals. A change in utility due to a change in BMI was added to an individual's EQ-5D if they had a BMI greater than 25. As a consequence, obese individuals who reduce their BMI as a result of the intervention will experience an increase in EQ-5D.

12.3.12.3 Utility Decrements

Utility decrements for long term chronic conditions were applied to the age and BMI adjusted EQ-5D score. It was assumed that a diagnosis of diabetes was not associated with a reduction in EQ-5D independent of the utility decrements associated with complications, comorbidities or depression. Cardiovascular disease, renal failure, amputation, foot ulcers, blindness, cancer, osteoarthritis and depression were all assumed to result in utility decrements. The utility decrements

are measured as a factor which is applied to the individual's age and BMI adjusted baseline. If individuals have multiple chronic conditions the utility decrements are multiplied together to give the individual's overall utility decrement from comorbidities and complications, in line with current NICE guidelines for combining comorbidities.[303]

Due to the number of health states it was not practical to conduct a systematic review to identify utility decrements for all health states. Furthermore, there is very little or no data to inform utility decrements for comorbid conditions specifically in individuals with ID. A pragmatic approach was taken to search for health states within existing HTA for the relevant disease area in the general population or by considering studies used in previous economic models for diabetes prevention. Discussions with experts in health economic modelling were also used to identify prominent sources of data for health state utilities.

Two sources of data were identified for diabetes related complications. A recent study from the UKPDS estimated the impact of changes in health states from a longitudinal cohort.[304] This data was used to estimate the utility decrement for amputation and congestive heart failure. The absolute decrement for amputation was converted into utility decrement factors that could be multiplied by the individuals' current EQ-5D to estimate the relative effect of the complication. Utility decrements for renal failure and foot ulcers were not available from the UKPDS study described above, so were obtained from a different study of 2,048 subjects with T1DM and T2DM.[305]

Utility decrements for the variety of cardiovascular events were taken from an HTA assessing statins to reflect the utility decrements in the general population.[289] A burden of illness study with a broad utility decrement for cancer was identified and used to define utility decrements for breast and colon cancer.[306] A utility decrement for osteoarthritis was taken from a HTA,[307] and a utility decrement for depression was calculated from a trial that had used EQ-5D.[308]

The multiplicative utility factors that are used in the model to describe health utility decrements from comorbid complications are shown in *Table 50*. The mean absolute decrement estimated in each study is reported alongside the baseline utility for each study. The utility factor was estimated by dividing the implied health utility with the comorbidity by the baseline utility.

Economic Impact: Costs

At any given time period of the model individuals can have multiple health complications that incur direct healthcare costs. Some of the health states are mutually exclusive; however an individual can accrue multiple complications within the model. Each health state is associated with an average cost, which is accrued by all individuals for every time period for which the state is indicated. Resource use for each comorbidity is added together and no savings are assumed to be made from the use of the same resources for two or more comorbidities for an individual. An exception to this is an assumed adjustment to the utilisation of GP services for individuals with chronic diseases. In the majority of cases it is assumed that the unit costs of healthcare for someone with ID would be the same as the unit costs for an individual in the general population.

Table 50: Utility decrement factors

Event / Co-morbidity	Mean Absolute decrement	Standard error for absolute decrement	Baseline Utility	Multiplicative Utility Factor	Source
Foot ulcer	-0.099	0.013	0.689	0.856	Coffey[305]
Amputation	-0.172	0.045	0.807	0.787	UKPDS[304]
Blind	0.033	0.027	0.807	1.041	UKPDS[304]
Renal failure	-0.078	0.026	0.689	0.887	Coffey[305]
Stable Angina				0.801	Ward HTA[289]
Unstable Angina y1				0.770	Ward HTA[289]
Unstable Angina y2				0.770	Ward HTA[289]
Myocardial Infarction y1				0.760	Ward HTA[289]
Myocardial Infarction y2				0.760	Ward HTA[289]
Transient Ischaemic Attack				1.000	Ward HTA[289]
Stroke y1				0.629	Ward HTA[289]
Stroke y2				0.629	Ward HTA[289]
Breast Cancer	-0.060		0.800	0.913	Yabroff[306]
Colorectal Cancer	-0.060		0.800	0.913	Yabroff[306]
Osteoarthritis	-0.101				Black HTA[307]
Depression	-0.116		0.7905	0.875	Benedict[308]
Congestive Heart Failure	-0.101	0.032		0.875	UKPDS[304]

UKPDS baseline utility 0.807; HSE baseline 0.7905

12.3.13 Economic Impact: Costs

At any given time period of the model individuals can have multiple health complications that incur direct healthcare costs. Some of the health states are mutually exclusive; however an individual can accrue multiple complications within the model. Each health state is associated with an average cost, which is accrued by all individuals for every time period for which the state is indicated. Resource use for each comorbidity is added together and no savings are assumed to be made from the use of the same resources for two or more comorbidities for an individual. An exception to this is an assumed adjustment to the utilisation of GP services for individuals with chronic diseases. In the majority of cases it is assumed that the unit costs of healthcare for someone with ID would be the same as the unit costs for an individual in the general population.

The exception was cost for a GP appointment, which was expected to be 40% higher than in the general population due to increased length of consultation. All costs were inflated to 2014/15 values using the retail price index where necessary, from the Personal Social Services Research Unit (PSSRU) sources of information[266].

Currently the following costs are incorporated are:

- Costs of GP appointments;
- Costs of hypertension/dyslipidaemia/diabetes diagnosis and treatment with statins and anti-hypertensives. Statins have a 65% uptake rate;
- Diabetes costs (three stage treatment regimen incorporating Metformin monotherapy (HbA1c>6.5%), Metformin plus a dipeptidyl peptidase 4 inhibitor (DPP-IV), (when HbA1c>7.4%), and insulin plus antidiabetics (HbA1c>8.5%), together with associated costs such as blood tests, extra GP visits);
- CVD and Heart Failure Costs (including hospital and primary care costs, medications and ongoing care costs for people with Stroke);
- Microvascular Costs (including renal dialysis and transplant, treatment costs for amputation, ulcer and blindness);
- Cancer Costs (including screening and treatment by cancer stage);

- Osteoarthritis costs (including extra primary care, medications and joint replacement);
- Depression costs (including nurse costs, medication and emergency care).

A summary of all the unit costs used in the model and their sources is shown in *Table 51*.[266, 309-328]

12.3.13.1 Diabetes costs

A three stage diabetes treatment regimen is applied in the model as a trade-off between model simplicity and capturing key cost differences between the interventions. At diagnosis all patients are prescribed low cost treatments, represented by metformin (weighted average of standard and modified release) to describe the average cost of these medications. If HbA1c increases above a threshold the individual is prescribed one of the more expensive DPP-IV inhibitors in addition to metformin. The individual continues to receive metformin plus DPP-IV inhibitor for a period of time until they require insulin. The cost of diabetes in the year of diagnosis is assumed to be greater than subsequent years because the individual will receive more contact time whilst their diabetes is being controlled.

Simulated individuals experience an annual increase in HbA1c. It is assumed that individuals switch to dual treatment in the first annual cycle in which HbA1c increases above 7.4%, based on a recent HTA.[329] For costing purposes the second drug to be added to metformin was assumed to be Sitagliptin. The second major treatment change is assumed to be initiation of insulin. Within the model the individual is switched to insulin in the first annual cycle at which HbA1c exceeds 8.5%.[329] The insulin Glargine was chosen to represent insulin treatment in the UK.

Table 51: Summary of all drug, treatment, care and resource costs included in the model

Drug, Treatment, Care and Resource Costs	Cost per year/ incident in 2014/15 prices	Source
STOP Diabetes intervention per person	£ 1,097	Micro-costing
Screening and Intervention costs		
Standard Learning Disability Health Check	£43.48	Dept. of Health[316]
Learning Disability Health Check without Blood Tests	£18.67	
Health Check capabilities assessment and explanation of intervention (10 mins Health Care assistant time)	£3.40	PSSRU[266]
First line diabetes treatment - low cost diabetes monotherapy		
Ongoing costs of diabetes monotherapy – made up of…	£79.06	
Metformin 500 mg *bid* standard (85% of patients) or modified release (15%) tablets	£18.83	BNF[315]
Nurse at GP (consultation)	£25.52	PSSRU[266]
Health care assistant (10 mins)	£3.40	PSSRU[266]
Urine sample	£1.00	[326]
Eye screening	£24.31	[320]
Lab tests – made up of…	£6.00	
HbA1c test	£3.00	[326]
Lipids test	£1.00	[326]
Liver function test	£1.00	[326]
B12 test	£1.00	[326]
Additional first year costs of diabetes monotherapy – made up of…	£103	
Nurse at GP (2 x consultations)	£51.03	PSSRU[266]
Health care assistant (2 x 10 mins)	£6.80	PSSRU[266]
Urine sample (x2)	£2.00	[326]

Lab tests as above (x2)	£12.00	[326]
Smoking cessation (central estimate of cost of nicotine replacement therapy) taken up by 50% of the assumed 20% of population who smoke	£30.90	PSSRU[266]
Second line diabetes treatment - Metformin and a DPP-IV inhibitor – made up of…	£529	
Sitagliptin 100 mg daily	£434	BNF[315]
Metformin 500 mg *bid* standard (85% of patients) or modified release (15%) tablets	£85	BNF[315]
Self-monitoring strips (82 per annum) [322]	£16.36	BNF[315]
Nurse at GP (consultation)	£25.52	[266]
Health care assistant (10 mins)	£3.40	[266]
Urine sample	£1.00	[326]
Eye screening	£24.31	[320]
Lab tests as for first line treatment	£6.00	[326]
Third line diabetes treatment - Insulin and oral anti-diabetics – made up of…	£1,503	
Nurse at GP (3 x consultations)	£76.55	PSSRU[266]
Health care assistant (3 x 10 mins)	£10.21	PSSRU[266]
Urine sample (x3)	£3.00	[326]
Eye screening	£24.31	[320]
Lab tests as for first line treatment (x3)	£18.00	[326]
Insulin treatment costs – made up of…	£1,376	
Glargine	£830.83	*[312]*
Oral anti-diabetics	£57.75	*[312]*
Reagent test strips	£292.74	*[312]*
Hypoglycaemic rescue	£30.98	*[312]*
Pen delivery devices	£72.44	*[312]*
Sharps	£90.98	*[312]*
Other primary care costs		
GP visit (17 minutes)	£68.38	PSSRU[266]
Diagnosis of hypertension (including ambulatory BP monitoring)	£56.51	[327]
Annual treatment with statins (simvastatin 20 mg *bid*)	£26.59	BNF[315]
Annual treatment with anti-hypertensives	£195.94	[321]

Cardiovascular disease costs		
Unstable Angina year 1: Secondary care costs: 100% hospitalisation, 50% revascularisation procedure, three outpatient appointments). Primary care costs (three GP visits) and medications	£4,674	324
Myocardial infarction year 1 Secondary care costs: 100% hospitalisation, 50% revascularisation procedure, three outpatient appointments) Primary care costs (three GP visits) and medications.	£4,813	324
Subsequent ACS care costs Secondary care costs (one outpatient appointment). Primary care costs (three GP visits) and medications.	£410	324
Stroke year 1 (NHS costs) Costs of acute events reported in Youman et al. [57] weighted by the distribution of severity of stroke [21].	£9,716	309
Social care costs of stroke in subsequent years The costs of ongoing care at home or in an institution weighted by the distribution of severity of stroke and discharge locations.	£2,730	324
Fatal coronary heart disease Assumed that 50% of fatalities incurred cost.	£713	313
Fatal non cardiac vascular event Assumed that 50% of fatalities incurred cost.	£4,443	309
Congestive heart failure	£3,091	UKPDS[318]
Other complications of diabetes costs		
Renal failure – weighted composite of…	£25,046	
Haemodialysis with overheads	£42,049	*323*
Automated peritoneal dialysis	£27,217	*323*
Continuous ambulatory peritoneal dialysis	£19,742	*323*
Transplant (year 1)	£23,660	*310*

Immunosuppressant (10 years)	£6,959	[310]
Foot ulcers	£216	[317]
Amputation first year	£10,101	UKPDS[325]
Amputation subsequent years	£1,896	UKPDS[325]
Blindness first year	£1,434	UKPDS[325]
Blindness subsequent years	£479	UKPDS[325]
Breast cancer	£13,818	[314]
Colorectal cancer	£18,729	[311]
Osteoarthritis	£962	[328]
Depression - made up of…	£137	[319]
Practice nurse at surgery	£13.70	
Practice nurse at home visit	£0.54	
Practice nurse telephone	£0.99	
Health visitor	£1.94	
District nurse	£0.38	
Other nurse	£1.17	
HCA phlebotomist	£1.05	
Other primary care	£4.85	
Out of hours	£6.18	
NHS direct	£2.28	
Walk-in centre	£8.15	
Prescribed medications	£74	
Secondary care	£21	

Assumed 20% smoking prevalence and 50% uptake of smoking cessation services

SANG Stable angina; UANG unstable angina; MI myocardial infarction; TIA transient ischemic attack; CHD congestive heart failure; ACS acute Coronary Syndrome; UKPDS United Kingdom prospective Diabetes Study; BNF British National Formulary; DPP-IV, dipeptidyl peptidase 4 inhibitor

12.3.13.2 Health checks

The cost of a health check in the ID population was derived from the Department of Health Economic modelling for vascular checks.[316] This study estimated the cost of a health check in the general population to be £23.70 in 2009 including blood tests, healthcare professional time, follow up and administration costs. For individuals with ID, it was assumed that all staff costs would double as the health check would take twice as long to perform (based on personal communication from Dr Kamlesh Khunti). All other costs were assumed to stay the same. Costs were inflated to 2014/15 values prices, giving a final value of £43.48 for a full Learning Disability Health Check.

Some individuals refuse to have blood taken as part of the health check. For these individuals a modified health check cost was derived, removing the cost of blood tests (consumables and laboratory costs) and the cost of nurse follow-up from the total. After inflation this came to £18.67.

12.3.13.3 Other primary care costs

Individuals with ID are assumed to visit their GP with the same frequency as individuals in the general population; however, each consultation is estimated to take 40% longer than the average (based on personal communication from Prof K Khunti, Diabetes Research Centre, University of Leicester, UK; October 2015). PSSRU unit costs were used to estimate the cost of a 17.2 minute consultation at £67,[266] which was then inflated to 2014/15 prices. Individuals who are prescribed statins receive a daily dose of 40mg of generic Simvastatin. The individual remains on statins for the rest of their life. A unit cost of anti-hypertensives was obtained from a 2004 study[321] and inflated to 2014/15 prices. Due to the number of different anti-hypertensive treatments available and possibilities for combination therapies, using the cost from this study of prescriptions was preferred to using costs directly from the BNF.

12.3.13.4 Cardiovascular costs

Costs for cardiovascular disease were obtained from a 2009 HTA for high dose lipid-lowering therapy.[324] The costs included are shown in *Table 51*. The costs of fatal stroke and MI were obtained from two separate studies,[309, 313] and it was assumed that 50% of individuals would incur these costs. The costs of congestive heart failure were estimated from the UKPDS costing study for complications related to diabetes.[318]

12.3.13.5 Costs of other comorbidities

More details of the costs of microvascular complications of diabetes, cancers, osteoarthritis and depression are available on-line.[265]

12.3.14 Other model inputs

- Perspective: the model adopts an NHS and social care perspective. Societal costs are not included.
- Horizon: The time horizon of the model is the lifetime of all baseline individuals.
- Discount Rates: Costs and QALYs are discounted at 1.5% per annum in line with NICE guidance for economic evaluation of public health interventions[158].

12.3.15 Reporting outcomes of the economic modelling

The model compares the outcomes of an identical baseline population undergoing the screening (and possible intervention) with those if current care were followed. The model allows a variety of different clinical outcomes to be gathered, as well as costs and QALYs. The model also allows a range of other incremental outcomes to be collected including life years saved and diabetes and cardiovascular cases prevented.

12.3.15.1 Use of threshold analysis

The usual output of an economic evaluation, for a pre-specified intervention with known clinical effectiveness and cost per patient, is the incremental cost-

effectiveness ratio, the ICER (see below for formulae) which can then be compared with the £ 20,000 to £ 30,000 cost per QALY acceptability threshold set out by NICE.

However, as the clinical effectiveness of the STOP Diabetes intervention was not tested as part of the current programme of research (only its feasibility), a different approach for this analysis was needed. The output was the change in effectiveness needed for the intervention to be marginally cost-effective; that is, the ICER equals the cost per QALY acceptability threshold. As the primary clinical outcome of interest to the STOP Diabetes study investigators is change in steps per day, the economic analysis deals with the change in physical activity (steps) and associated risk factors (BMI, SBP, total and HDL cholesterol) needed for the intervention to be marginally cost-effective. The STOP Diabetes intervention also contains dietary advice (but no specific weight goal) so the threshold analysis needs to take account of benefits attributable to physical activity and diet. The results tables for the threshold analyses therefore show alternative permutations of step changes together with the additional benefit from dietary change that would be necessary for the intervention to be cost-effective overall.

12.3.16 Analyses, scenarios and sensitivity analyses undertaken

All statements herein about an increase in the number of steps refer to the increase in steps per day.

12.3.16.1 Exploratory analysis

An initial analysis was undertaken to assess how cost-effective the STOP Diabetes intervention would be if it achieved the average 2,491 change in daily steps reported in the Bravata meta-analysis. Initially, it was assumed that the intervention would increase mean daily step count by 2,491 steps as detailed in Bravata et al, leading to:

- mean reduction in BMI of 0.38
- mean reduction in systolic BP of 3.8mmHg
- mean reduction in total cholesterol of 0.09mmol/L

- mean increase in HDL cholesterol of 0.06 mmol/L

The intervention effect was assumed to decline linearly such that by three years (from the start of the intervention), the risk factors have reverted back to their trajectory had there been no intervention.

The ICER is obtained using the incremental costs and QALYs gained from implementing the intervention rather than current care, calculated using the following formulae:
Incremental costs (£) = Total costs Intervention (£) – Total costs Comparator (£)
Incremental QALYs = Total QALYs Intervention (£) – Total QALYs Comparator (£)
ICER (£/QALY) = Incremental Costs (£) / Incremental QALYs

Although total costs and QALYs can be assessed at any year in the model, allowing estimation of both short-term and long-term cost-effectiveness, we report the long-term cost-effectiveness as this is what regulatory bodies are primarily interested in.

In addition, a simple budget impact was calculated, as follows:

1. The number of adults in England with moderate to critical needs using social care was taken from 2015 estimates by PHE's learning disabilities observatory for adults (546,489)[330]
2. The percentage of the above that have IGR was based on the percentage in the STOP screening study found to be IGR after screening, recruitment and blood testing (5%)
3. The percentage likely to take up an intervention was based on the percentage of those in the feasibility study invited to take part on the STOP programme who actually attended sessions (26%)
4. The resulting number of likely STOP users for the whole of England (7,104) was multiplied by the intervention cost per user to give the total budget impact of implementing the STOP programme.

12.3.16.2 Scenarios for duration of effect

For all analyses undertaken, we model two fundamental alternative scenarios to test the sensitivity of the cost-effectiveness to two alternative durations of effect of the intervention, that is, after starting to reduce after year 1, by which time-point have the benefits of intervention worn off. The first scenario, assuming a three-year duration, is considered most likely given that the proposed maintenance sessions finish at the end of the first year and the alternative five-year scenario is presented as a what-if scenario.

12.3.16.3 Deterministic one-way sensitivity analysis

Previous analyses using the model and of economic evaluations of lifestyle interventions suggested which parameters are likely to have the largest effect on model results, so the following were considered for one-way sensitivity analysis:

i) Increased clinical effects

There is significant uncertainty around the relationship reported in Bravata, so we explored the impact of a more beneficial impact by taking the 65th percentile for the possible magnitude of beneficial change in BMI, systolic BP, total cholesterol and HDL cholesterol from the CIs reported in Bravata (assuming the distributions are normally distributed). In this case, an increase in mean daily step count of 2,491 steps as detailed in Bravata is estimated to lead to:

- mean reduction in BMI of 0.45 kg/m^2
- mean reduction in systolic BP of 4.21 mmHg
- mean reduction in total cholesterol of 0.14 mmol/L
- mean increase in HDL cholesterol of 0.07 mmol/L

This alternative mapping was explored in the context of both the three and five-year duration of effect scenarios above.

ii) Uptake rates:

These were considered not to be a key driver because individuals that do not uptake the intervention incur no costs other than the very small cost of screening relative to the cost of the intervention.

iii) Discount rates:

Alternative rates were considered but testing out lower rates than 1.5% seems implausible and higher rates would not have altered the conclusions.

12.3.17 Subgroup analyses

As initial analyses suggested that the intervention would be unlikely to be cost-effective in the overall ID population, further work was set out in order to identify the most beneficial subgroups. Three sub-groups were identified to explore if the cost-effectiveness of the intervention might be improved if screening were more targeted:

1) By age band

Based on the distribution of age in the STOP Diabetes study, the age bands were chosen: <35, ≥35 and <40, ≥40 and < 45, ≥45 and < 50, ≥50. Selecting individuals aged 35+ would include 65% of the STOP Diabetes cohort; aged 40+ 45+ and 50+ would include 55%, 45% and 35% respectively. Age cut-offs of 55+ and 60+ would have only covered 24% and 14% of the cohort only respectively.

2) BMI

We carried out a subgroup analysis in which everyone was screened but only obese individuals were eligible for intervention.

3) Baseline cardiovascular risk

We calculated the baseline 10-year CVD risk using the QRISK score and excluded any individuals with a risk <5%. This is a low cut-off but using a higher cut-off would have meant fewer than 25% of the cohort being screened, and a very low proportion actually receiving and benefiting from intervention.

12.3.18 Probabilistic sensitivity analysis

Due to the exploratory nature of the analysis (described in Section 12.3.16.1), full probabilistic sensitivity analysis (PSA) was carried out on only one of the threshold scenarios, with the aim of illustrating the extent of non-linearity in the model (i.e. by comparing the results of the PSA with the corresponding deterministic results). PSA,

which describes the uncertainty in model parameter inputs, is not suitable for describing the decision uncertainty in this analysis i.e. the current research was leading to the stage at which the intervention could be considered for implementation into clinical practice. Instead, analyses were more exploratory to inform potential future research and intervention refinement.

In addition, the true uncertainty around the effectiveness estimates is much wider than that around the parameters available from the Bravata-based relationship. There is also uncertainty around the effectiveness of the planned intervention in increasing physical activity and uncertainty around whether increasing the number of steps increases metabolic benefits in a linear way. This uncertainty cannot be accurately quantified (although it could potentially be estimated through a time-consuming expert elicitation, which is outside the scope of this investigation), but PSA analysis without it would vastly underestimate the uncertainty in the cost-effectiveness estimates.

For the single PSA completed, a suitable distribution was selected for each parameter, based upon its mean and standard error, and within the simulations, random sampling across all input parameter distributions was undertaken. 2000 different random samples of parameter values were selected, and each was applied to a different random cohort of 5,000 individuals randomly sampled with replacement from the baseline STOP Diabetes population. For each probabilistic sensitivity analysis sample, the model was run and results compiled.

More details of the distributions around key model parameters are shown in Appendix 26 (*Table 70* and *Table 71*).

12.4 Results

This section presents a series of results for the scenarios and sensitivity analyses described earlier. First is an analysis of how cost-effective the STOP Diabetes intervention would be if it achieved the average change for a physical activity intervention, 2,491 steps, as reported in the Bravata meta-analysis. Next, results of the threshold analyses for the necessary risk factor changes needed to achieve cost-effectiveness are presented under a variety of scenarios and sub-groups. Lastly, an alternative threshold analysis explores what the maximum budget for the intervention would be given certain changes in risk factors.

Whenever results are stated as the 'base case', these reflect the base case assumption for the effects a change in steps has on risk factors for CVD (BMI, systolic BP, total and HDL cholesterol). Most analyses present results for both 3 and 5 years durations of intervention effect, but if not specified, the base case of 3 years applies.

It should be noted that effects lasting to year 3 means that they have worn off 2 years from the end of year 1 (which is close to when the last monthly maintenance session occurs). Similarly, effects that have worn off by year 5 effectively last for 4 years from the end of the maintenance sessions.

12.4.1 Cost-per-QALY results based on Bravata step count

Before the threshold analyses were undertaken, an exploratory analysis was undertaken to see how cost-effective the STOP Diabetes intervention would be, assuming an increase in steps in line with that calculated in the meta-analysis by Bravata. This analysis assumes no dietary intervention.

The estimated incremental cost-effectiveness ratio (cost per QALY gained) under the base case is £275,000 compared to a usual acceptability threshold (what funders are willing to pay) in the range £ 20,000 to £ 30,000 per QALY. Savings in lifetime costs of CVD and primary care, and savings in treating diabetes and its complications are

far outweighed by the £ 1,097 intervention cost per person. A much greater intervention effect in terms of either physical activity, diet or both, or a reduction in intervention cost would be required to make the intervention cost effective.

The estimated budget impact for delivering the STOP programme to 7,104 adults with ID and IGR across England was estimated at £7.8m. If the programme were taken up by **all** ID adults with IGR in England (over 27,000) then the total cost could be as high as £30.0m. There is uncertainty around the true prevalence of IGR amongst adults with ID. If this is actually 10% (rather than 5%), the above budget impacts would rise to £15.6m and £60.0m respectively.

12.4.1.1 Probabilistic sensitivity analysis

Probabilistic sensitivity analysis was carried out for the above Bravata-based analysis (an increase of 2,491 steps), giving a probabilistic central estimate of the ICER of £253,000 which is lower than the deterministic estimate by about 6%, demonstrating a small degree of non-linearity in the model. Given how high the deterministic ICER was compared with the acceptability threshold of £ 30,000/QALY, this is clearly a negligible difference for the overall conclusion about the intervention.

The probability that such an intervention is cost-effective compared to current care at a threshold of £ 30,000 per QALY is almost zero at 0.15%. However, as described in the methods section, the uncertainty around intervention effectiveness is much higher than the uncertainty described in the Bravata study and used in the PSA, meaning that the PSA will underestimate the total uncertainty.

The cost-effectiveness plane for the intervention compared to current care at £ 30,000 per QALY is shown in Appendix 27 (*Figure 30*).

12.4.2 Threshold analyses for effect sizes needed

A series of model simulations were performed in order to determine the thresholds required for the intervention to be cost-effective at acceptability thresholds of

£20,000 and £ 30,000 per QALY. The presented thresholds are rounded to the nearest 500 steps.

At the calculated thresholds, the ICER for the intervention is £20,000 (or £ 30,000) per QALY, and, the greater savings from CVD and diabetes treatment and primary care costs, together with the value of the health gain (QALYs) are just enough to outweigh the additional cost of the intervention.

Ascertainment of the thresholds relies upon the assumption of a linear relationship between change in steps and risk factors (that observed in Bravata) being maintained over the wide range of steps inherent within the calculation of the thresholds.

It is recognised that under many, if not most scenarios, the magnitude of the additional diet-related changes in BMI and systolic BP (that are necessary to attain cost-effectiveness) are implausible in terms of their achievability. They are nevertheless genuine estimates from the threshold approach fundamental to this economic analysis.

12.4.2.1 Summary of key results from threshold analyses

In subsequent Section 12.4.2.5, detailed results are presented for a number of alternative levels of increase in steps per day, up to 15,000. There are however many tables, each containing many permutations of the magnitude of physical activity-related and dietary related BMI change that could achieve cost-effectiveness as illustrated in *Table 56*. To aid digestion of the results, in this section, the results are summarised for some mid-range levels of change in steps, 3000, 5000 and 7000 per day.

In
Table 52, the necessary diet-related effects that would need to be achieved, in addition to the effects arising from an increase of 5000 steps, are shown across

various scenarios and sub-groups (see Section 12.3.15.1. for the rationale of the threshold analyses as presented).

As an example, consider the base case row of
Table 52, assuming an acceptability threshold of £30,000 per QALY and 3 year duration of effects. BMI and systolic BP reductions of 2.9kg/m^2 and 29mmHg respectively would be needed, in addition to the BMI and systolic BP benefits of 5000 additional steps, in order for the intervention to be cost-effective. An increase of 5000 steps corresponds to a BMI reduction of 0.76kgm^2, an systolic BP reduction of 7.6mmHg and a reduction in the lipid ratio of 0.22 (all purely through physical activity without diet). So the overall (steps plus dietary) BMI and systolic BP reductions needed for a cost-effective intervention would be 3.66kg/m^2 and 36.6mmHg, which are clearly unachievable in practice.

Table 52: Summary of dietary effects needed in addition to 5000 steps

Population group	**£20,000 per QALY**		**£30,000 per QALY**	
	3 year	**5 year**	**3 year**	**5 year**
	Dietary contributions only			
	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)
Base case	-4.2 (-42)	-2.6 (-26)	-2.9 (-29)	-1.5 (-15)
Increased effectiveness intervention	-4.0 (-40)	-2.0 (-20)	-2.4 (-24)	-1.2 (-12)
Obese subgroup	-4.0 (-40)	-2.0 (-20)	-2.6 (-26)	-1.3 (-13)
45 to 49-year-old subgroup	-1.8 (-18)	-1.1 (-11)	-1.4 (-14)	-0.9 (-9)
50+ age group	-3.4 (-34)	-2.0 (-20)	-1.8 (-18)	-0.9 (-9)
High CVD risk group (≥5% 10-year risk)	-2.7 (-27)	-1.4 (-14)	-1.5 (-15)	-0.8 (-8)

Table 53 and *Table 54* present similar analyses for a change in steps of 3000 and 7000 respectively.

Table 53: Summary for 3000 steps

Population group	£20,000 per QALY		£30,000 per QALY	
	3 year	**5 year**	**3 year**	**5 year**
	Dietary contributions only			
	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)
Base case	-4.5 (-45)	-2.9 (-29)	-3.2 (-32)	-1.8 (-18)
Increased effectiveness intervention	-4.3 (-43)	-2.3 (-23)	-2.7 (-27)	-1.5 (-15)
Obese subgroup	-4.3 (-43)	-2.3 (-23)	-2.9 (-29)	-1.6 (-16)
45 to 49-year-old subgroup	-2.1 (-21)	-1.4 (-14)	-1.7 (-17)	-1.2 (-12)
50+ age group	-3.7 (-37)	-2.3 (-23)	-2.1 (-21)	-1.2 (-12)
High CVD risk group (≥5% 10-year risk)	-3.1 (-31)	-1.7 (-17)	-1.8 (-18)	-1.1 (-11)

Table 54: Summary for 7000 steps

Population group	£20,000 per QALY		£30,000 per QALY	
	3 year	**5 year**	**3 year**	**5 year**
	Dietary contributions only			
	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)
Base case	-3.9 (-39)	-2.3 (-23)	-2.6 (-26)	-1.2 (-12)
Increased effectiveness intervention	-3.7 (-37)	-1.7 (-17)	-2.1 (-21)	-0.9 (-9)
Obese subgroup	-3.7 (-37)	-1.7 (-17)	-2.3 (-23)	-1.0 (-10)
45 to 49-year-old subgroup	-1.5 (-15)	-0.8 (-8)	-1.1 (-11)	-0.6 (-6)
50+ age group	-3.1 (-31)	-1.7 (-17)	-1.5 (-15)	-0.6 (-6)
High CVD risk group	-2.4 (-24)	-1.1 (-11)	-1.2 (-12)	-0.5 (-5)

(≥5% 10-year risk)

12.4.2.2 Intervention cost threshold analysis

As an alternative type of threshold analysis, we explored the maximum intervention cost that could be afforded for a given increase in steps per day. At a value of £30,000 per QALY, for the subgroup of individuals aged over 50, with an assumed duration of effect of 3 years, the maximum intervention budget (combined initial plus maintenance cost) to achieve cost-effectiveness is around £280 for a 4,000 step per day increase (0.61 BMI reduction and 6mmHg systolic BP reduction) and around £420 for a 6,000 step per day increase (0.91 BMI reduction and 9mmHg systolic BP reduction). For the subgroup of individuals with increased CVD risk, the maximum budget for a cost-effective intervention is around £500 for a 4,000 step per day increase and around £700 for a 6,000 step per day increase.

12.4.2.3 Interpretation of threshold analysis results

First, we summarise the implications of aiming to achieve cost-effectiveness through step changes alone. For the overall STOP Diabetes cohort, to achieve cost-effectiveness at a value of £20,000 per QALY, the results suggest that in excess of 30,000 additional steps per day would be required (around 5 kg/m^2 reduction in BMI and 50mmHg reduction in systolic BP), which is biologically implausible. At £30,000 per QALY the threshold was around 24,000 additional steps per day (3.7 kg/m^{2} - reduction in BMI and 37mmHG reduction in systolic BP). If the intervention effect is assumed to last (decreasing linearly) until year 5, the threshold reduces to around 22,000 steps (2.4 kg/m^2 reduction in BMI and 34mmHG reduction in systolic BP) at £20,000 per QALY and 15,000 steps (2.3 kg/m^2 reduction in BMI and 23mmHG reduction in systolic BP) at £30,000 per QALY. Note that as all these values represent effect sizes more than five times that achieved in the source study (Bravata et al.) and as such they are reliant on extrapolations of the Bravata relationship well outside the range in which it can be reliably done so.

Cost-effective effect sizes could alternatively be achieved through a combination of risk factor changes through physical activity and risk factor changes through dietary intervention. Clearly there are many permutations of the magnitude of physical activity-related and dietary related changes that could achieve cost-effectiveness. A necessary increase of 13,000 steps under an assumption of increased effectiveness and 5 years duration (at £30k per QALY) equates to a BMI change of -2.75kg/m^2 based on Bravata et al. This overall BMI change could be achieved, for example, through the effect of an additional 7,000 steps per day together with further BMI reduction achieved through dietary intervention of -1.68kg /m^2.

12.4.2.4 Alternative body mass index and systolic blood pressure equivalents

A limitation of the threshold results presented above is that the necessary BMI and systolic BP changes attributed to diet exclusively use the Bravata study in their calculation. This results in the small combined (steps plus diet) BMI changes relative to the systolic BP changes reflecting the ratio of benefits that could be expected from a physical activity rather than a dietary intervention.

To aim to address this, as an exploratory analysis, the table below shows an alternative set of permutations, the difference being that the diet-related BMI and systolic BP changes are now ‘re-aligned’ to reflect more realistically the relative ratio of BMI and systolic BP changes likely through dietary change. The calculations are underpinned by the BMI and systolic BP hazard ratios in the QRISK score for CVD, and thereby the analysis relies on the assumption that most of the economic benefits of intervention accrue through CVD risk reduction. We estimated that a 1mmHg systolic BP reduction gives approximately the same benefit as 0.6kg/m^2 BMI reduction.

The analysis was undertaken just in the context of a change of 5000 steps, that is, a re-working of results presented earlier in
Table 52.

For the high CVD risk groups, if the effects could be maintained such that the effect is not completely lost until year 5, then the effect sizes needed are becoming closer to those achievable in practice.

Table 55: Body mass index/systolic blood pressure equivalents for the diet-attributable benefits needed to be cost-effective

Population group	£20k per QALY		£30k per QALY	
	3 year	5 year	3 year	5 year
	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)	BMI (systolic BP)
Base case	-11.8 (-29)	-7.3 (-18)	-8.1 (-20)	-4.2 (-11)
Increased effectiveness intervention	-11.2 (-28)	-5.6 (-14)	-6.7 (-17)	-3.4 (-8)
Obese subgroup	-11.2 (-28)	-5.6 (-14)	-7.3 (-18)	-3.6 (-9)
45 to 49-year-old subgroup	-5 (-13)	-3.1 (-8)	-3.9 (-10)	-2.5 (-6)
50+ age group	-9.5 (-24)	-5.6 (-14)	-5 (-13)	-2.5 (-6)
High CVD risk group (≥5% 10-year risk)	-7.6 (-19)	-3.9 (-10)	-4.2 (-11)	-2.2 (-6)

12.4.2.5 Full threshold analysis results tables

Here we present the full set of model results for a wide range of change in step count. For disadvantaged groups, there is a higher likelihood than normal of NICE recommending a treatment at the upper end of the usual £ 20,000 - £ 30,000 per QALY bracket so the results presented here are for a willingness to pay of £30,000 per QALY. The diet-related improvements in BMI, systolic BP and cholesterol that would be needed at £ 20,000 per QALY would be even more challenging (or implausible) than those presented here so the results for £20,000 per QALY are presented in Appendix 28 (*Table 72, Table 73, Table 74, Table 75, Table 76, Table 77*).

Base case results

The systolic BP and cholesterol ratio effects shown in the third and fourth columns of the tables are the total (step-related) effects for these parameters, that is, effects mediated indirectly through BMI reduction and direct effects of physical activity.

Table 56: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for base case intervention effects, at £30,000 per QALY assuming all risk factors change together.

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-3.7	-37	-1.54	-2.3	-23	-1.08
1000	-0.2	-2	-0.09	-3.5	-35	-1.49	-2.1	-21	-1.03
3000	-0.5	-5	-0.26	-3.2	-32	-1.40	-1.8	-18	-0.90
5000	-0.8	-8	-0.42	-2.9	-29	-1.30	-1.5	-15	-0.78
7000	-1.1	-11	-0.57	-2.6	-26	-1.19	-1.2	-12	-0.64
9000	-1.4	-14	-0.71	-2.3	-23	-1.08	-0.9	-9	-0.50
11000	-1.7	-17	-0.84	-2.0	-20	-0.97	-0.6	-6	-0.34
13000	-2.0	-20	-0.97	-1.7	-17	-0.84	-0.3	-3	-0.18
15000	-2.3	-23	-1.08	-1.4	-14	-0.71	0.0	0	0.00

a obtained from model runs

b obtained from manual calculation after model runs

A more detailed breakdown of the base case events, costs and QALYs is shown in *Table 57*. The mean QALY gain per person screened equates to an average of less than 1 day of additional life (in full health).

Table 57: Detailed breakdown of results for 2,491 steps

Incremental Outcomes per person	Base Case effectiveness		Increased effectiveness	
	3 year duration	5 year duration	3 year duration	5 year duration
Total costs	£329	£326	£328	£322
Total QALYs	0.0012	0.0018	0.0014	0.0021
ICER	£273,000	£ 183,000	£ 231,000	£ 154,000
Cardiovascular cases (per 1m)	-130	-187	-153	-239
STOP Diabetes Intervention cost	£336	£336	£336	£336
Diabetes Treatment Costs	£ 0	£ 1	£0	£0
Cardiovascular Costs	-£5	-£6	-£5	-£7
Costs other Diabetes Complications	-£6	-£8	-£7	-£10

Sensitivity Analyses – increased clinical effects

In this analysis, it was assumed that effects on BMI, systolic BP and the lipid ratio of a given increase in steps would be greater than the base case, as described in the methods section.

Under this modified assumption and assuming 3-year duration of effects, the estimated incremental cost per QALY gained for an increase of 2,491 steps (with no dietary intervention) was £228,000.

To achieve cost-effectiveness at a value of £20,000 per QALY, threshold analysis suggests that in excess of 30,000 additional steps per day would be required (some 4.7 kg/m^2 reduction in BMI and 47 mmHg reduction in systolic BP), which is still biologically implausible. At £30,000 per QALY the threshold is around 21,000 additional steps per day (3.2 kg/m^2 reduction in BMI and 32mmHG reduction in systolic BP). If the intervention effect is assumed to last (decreasing linearly) until year 5, the threshold reduces to around 18,000 steps (2.8 kg/m^2 reduction in BMI and 27mmHG reduction in systolic BP) at £20,000 per QALY and 13,000 steps (2.0 kg/m^2 reduction in BMI and 20mmHG reduction in systolic BP) at £30,000 per QALY. The combination of step change and additional dietary change needed to reach cost-effectiveness (assuming all risk factors change together) is shown in *Table 58* (see Appendix 28, *Table 73* for £ 20,000 per QALY acceptability)

Table 58: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for an increased effectiveness intervention at £30,000 per QALY assuming all risk factors change together.

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-3.2	-32	-1.40	-2.0	-20	-0.97
1000	-0.2	-2	-0.09	-3.1	-31	-1.35	-1.8	-18	-0.90
3000	-0.5	-5	-0.26	-2.7	-27	-1.25	-1.5	-15	-0.78
5000	-0.8	-8	-0.42	-2.4	-24	-1.14	-1.2	-12	-0.64
7000	-1.1	-11	-0.57	-2.1	-21	-1.03	-0.9	-9	-0.50
9000	-1.4	-14	-0.71	-1.8	-18	-0.90	-0.6	-6	-0.34
11000	-1.7	-17	-0.84	-1.5	-15	-0.78	-0.3	-3	-0.18
13000	-2.0	-20	-0.97	-1.2	-12	-0.64	0.0	0	0.00
15000	-2.3	-23	-1.08	-0.9	-9	-0.50	0.0	0	0.00

Sub-group analysis: obese

Separate results are reported for a subgroup of the population only (using the base case clinical effects) who were defined as obese.

The estimated incremental cost per QALY gained for a 2,491 increase in steps under this scenario was £276,000.

To achieve cost-effectiveness at a value of £20,000 per QALY, threshold analyses suggest that in excess of 30,000 additional steps per day would be required and at £30,000 per QALY the threshold is around 22,000 additional steps per day. If the intervention effect is assumed to last (decreasing linearly) until year 5, the threshold reduces to around 18,000 steps at £20,000 per QALY and 13,500 steps at £30,000 per QALY. These values are very similar to the whole population results.

Table 59: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for an obese subgroup in a base case intervention at £30,000 per QALY assuming all risk factors change together.

Initial increase in steps needed	**Change attributable to the increase in steps**			**Additional change needed to be generated through diet**			**Additional change needed to be generated through diet**		
				Base case (3 year durability)			**5 year durability**		
	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-3.4	-34	-1.45	-2.1	-21	-1.00
1000	-0.2	-2	-0.09	-3.2	-32	-1.40	-1.9	-19	-0.94
3000	-0.5	-5	-0.26	-2.9	-29	-1.30	-1.6	-16	-0.81
5000	-0.8	-8	-0.42	-2.6	-26	-1.19	-1.3	-13	-0.68
7000	-1.1	-11	-0.57	-2.3	-23	-1.08	-1.0	-10	-0.53
9000	-1.4	-14	-0.71	-2.0	-20	-0.97	-0.7	-7	-0.38
11000	-1.7	-17	-0.84	-1.7	-17	-0.84	-0.4	-4	-0.22
13000	-2.0	-20	-0.97	-1.4	-14	-0.71	-0.1	-1	-0.05
15000	-2.3	-23	-1.08	-1.1	-11	-0.57	0.0	0	0.00

Subgroup analysis: age subgroups

The results in *Table 60* show the effect sizes needed for subgroups by age (for both the base case clinical effects and increased effects sensitivity assumption).

The estimated incremental cost-effectiveness ratio (cost per QALY gained) in the base case across the whole baseline population was £276,000. For age-based sub-groups, this ICER varied from £172,000 (ages 50+) to £482,000 (ages 35-39). In the base case with 5 year durability, the ICER varied from £107,000 (ages 50+) to £301,000 (aged less than 35). In the increased effectiveness scenario with 5 year durability, the ICER ranged from £92,000 (ages 50+) to £262,000 (aged less than 35).

Table 60: Change in body mass index / systolic blood pressure required to achieve a cost-effective outcome for individual age bands

Population group	Base case clinical effects				Increased clinical effects			
	£20k per QALY		£30k per QALY		£20k per QALY		£30k per QALY	
	Base case (3 yrs)	5 yrs	Base case (3 yrs)	5 yrs	Base case (3 yrs)	5 yrs	Base case (3 yrs)	5 yrs
All ages	-5.8 / -58	-3.4 / -34	-3.8 / -38	-2.3 / -23	-5.6 / -56	-2.9 / -29	-3.5 / -35	-1.9 / -19
Age <35	-18.3 / -183	-4.7 / -47	-7.9 / -79	-3.4 / -34	-13 / -130	-4 / -40	-6.7 / -67	-2.8 / -28
Age 35-39	-4.3 / -43	-2.7 / -27	-3.6 / -36	-2.2 / -22	-2.8 / -28	-2.4 / -24	-2.4 / -24	-2 / -20
Age 40-44	-5.3 / -53	-3.1 / -31	-3.7 / -37	-2.2 / -22	-5.2 / -52	-2.7 / -27	-3.4 / -34	-1.9 / -19
Age 45-49	-2.6 / -26	-1.9 / -19	-2.1 / -21	-1.7 / -17	-2.8 / -28	-2.1 / -21	-2.2 / -22	-1.6 / -16
Age 50+	-4.1 / -41	-2.7 / -27	-2.6 / -26	-1.7 / -17	-4.6 / -46	-2.1 / -21	-2.5 / -25	-1.3 / -13
Only BMI and Systolic BP effects are shown but corresponding changes in the lipid ratio in line with Bravata would also be needed								

To achieve cost-effectiveness at a value of £30,000 per QALY in the overall ID cohort, threshold analyses suggest that 25,000 additional steps per day would be required. For age sub-groups, this threshold varied from 14,000 additional steps per day (ages 45-49) to 52,000 (ages less than 35). If the durability were extended to 5 years the threshold ranges from 11,000 additional steps (ages 45+) to 22,000 additional steps (ages less than 35) and if effectiveness is increased the threshold ranges from 8,500 additional steps (ages 50+) to 18,500 steps (ages less than 35). The combinations of steps and additional dietary changes needed to reach cost effectiveness for people aged 45-49 and aged more than 50 at £30,000 (£20,000) per QALY are shown in *Table 61* and *Table 62* (and Appendix 28, *Table 75* and *Table 76*).

Table 61: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for a 45 to 49-year-old subgroup in a base case intervention at £30,000 per QALY assuming all risk factors change together

Initial increase in steps needed	**Change attributable to the increase in steps**			**Additional change needed to be generated through diet**			**Additional change needed to be generated through diet**		
				Base case (3 year durability)			**5 year durability**		
	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-2.1	-21	-1.03	-1.7	-16.8	-0.8
1000	-0.2	-2	-0.09	-2.0	-20	-0.97	-1.5	-15.3	-0.8
3000	-0.5	-5	-0.26	-1.7	-17	-0.84	-1.2	-12.2	-0.6
5000	-0.8	-8	-0.42	-1.4	-14	-0.71	-0.9	-9.2	-0.5
7000	-1.1	-11	-0.57	-1.1	-11	-0.57	-0.6	-6.1	-0.3
9000	-1.4	-14	-0.71	-0.8	-8	-0.42	-0.3	-3.1	-0.2
11000	-1.7	-17	-0.84	-0.5	-5	-0.26	0.0	0.0	0.0
13000	-2.0	-20	-0.97	-0.2	-2	-0.09	0.0	0	0.00
15000	-2.3	-23	-1.08	0.0	0	0.00	0.0	0	0.00

Table 62: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for a greater-than-50-year-old subgroup in a base case intervention at £30,000 per QALY assuming all risk factors change together

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-2.6	-26	-1.19	-1.7	-16.8	-0.8
1000	-0.2	-2	-0.09	-2.4	-24	-1.14	-1.5	-15.3	-0.8
3000	-0.5	-5	-0.26	-2.1	-21	-1.03	-1.2	-12.2	-0.6
5000	-0.8	-8	-0.42	-1.8	-18	-0.90	-0.9	-9.2	-0.5
7000	-1.1	-11	-0.57	-1.5	-15	-0.78	-0.6	-6.1	-0.3
9000	-1.4	-14	-0.71	-1.2	-12	-0.64	-0.3	-3.1	-0.2
11000	-1.7	-17	-0.84	-0.9	-9	-0.50	0.0	0.0	0.0
13000	-2.0	-20	-0.97	-0.6	-6	-0.34	0.0	0.0	0.0
15000	-2.3	-23	-1.08	-0.3	-3	-0.18	0.0	0.0	0.0

Subgroup analysis: high cardiovascular risk subgroup

We ran an additional analysis looking at effectiveness of intervening in a subgroup of the population with 10-year CVD risk of at least 5% using the base case assumptions about clinical effects. A 5% cut-off was chosen because if the cut-off had been ≥ 10%, this would have resulted in around only 10% of the population being screened (before factoring in eligibility, suitability and willingness so probably less than 5% would have actually received the intervention).

The estimated incremental cost effectiveness ratio (cost per QALY gained in the base case across the whole baseline population was £177,000. With 5 year durability of effects, the ICER falls to £133,000.

To achieve cost-effectiveness in the base case at a value of £30,000 per QALY, threshold analyses suggest that 15,000 additional steps per day would be required with 3 year durability and 10,000 if the durability were extended to 5 years. The combinations of steps and additional dietary changes needed to reach cost effectiveness for people with high CVD risk at £30,000 per QALY are shown in *Table 63* (see Appendix 28, *Table 77* for £20,000 per QALY).

Table 63: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for a high cardiovascular risk subgroup in a base case intervention at £30,000 per QALY assuming all risk factors change together.

Initial increase in steps needed	Change attributable to the increase in steps			**Additional change needed to be generated through diet***			**Additional change needed to be generated through diet**		
				Base case (3 year durability)			**5 year durability**		
	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	Systolic BP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-2.3	-23	-1.08	-1.5	-15	-0.78
1000	-0.2	-2	-0.09	-2.1	-21	-1.03	-1.4	-14	-0.71
3000	-0.5	-5	-0.26	-1.8	-18	-0.90	-1.1	-11	-0.57
5000	-0.8	-8	-0.42	-1.5	-15	-0.78	-0.8	-8	-0.42
7000	-1.1	-11	-0.57	-1.2	-12	-0.64	-0.5	-5	-0.26
9000	-1.4	-14	-0.71	-0.9	-9	-0.50	-0.2	-2	-0.09
11000	-1.7	-17	-0.84	-0.6	-6	-0.34	0.0	0	0.00
13000	-2.0	-20	-0.97	-0.3	-3	-0.18	0.0	0	0.00
15000	-2.3	-23	-1.08	0.0	0	0.00	0.0	0	0.00

12.5 DISCUSSION

12.5.1 Statement of principal findings

Using a threshold analysis approach, the base case results indicate that the STOP Diabetes intervention that we have evaluated, costing £ 1,097 per patient, would need to result in a very large overall increase in steps, systolic BP, BMI and cholesterol for it to be cost-effective at a threshold in the £ 20,000 to £ 30,000 cost per QALY range usually adopted by NICE. These increases are much more than could be expected to be achievable in practice, even for the general population. Specifically for steps, an increase of 3,000 to 5,000 per day appears to be much more commonly reported for an intervention.

If we adopt two very favourable assumptions: 1) that the benefits of the intervention would not be fully lost until 4 years after the intervention (5 years from the start); and 2) that commissioners/payers would be willing to fund the intervention up to a threshold of £ 30,000 per QALY, then some of the scenarios begin to show more favorable results to some extent.

Targeting screening at either individuals over age 45, or at those obese, or those at relatively high baseline risk of CVD improves the cost-effectiveness of the intervention but it is still not cost-effective at an readily achievable combinations of steps and diet-attributable changes in risk factors (unless the cost of the intervention could be reduced).

12.5.2 Strengths and limitations

A strength of the analysis is that it was based on a relatively large set of baseline data for an ID cohort, so the baseline risks of the cohort were well-evidenced. However, there are large uncertainties around the intervention cost, and the precise relationship between changes in physical activity and cardiovascular risk factors. ‘Number of steps’ per day was the primary outcome of interest from the modelling based on the fact that it is the primary measure of interest to the STOP-DM study

investigators. The available evidence linking steps to biomarkers is somewhat limited, and comes from very diverse studies. Each intervention is unique and therefore estimates of effectiveness are unlikely to predict exactly the outcomes of the intervention in question. In addition, identified studies were undertaken in the general population not an ID population, and it is unknown whether differences in either behavioural or physiological response to intervention vary in the ID community.

Despite the fact that most diabetes prevention interventions combine dietary and physical activity elements, in order to model the threshold level of the primary measure of interest (steps) we therefore had to use steps as the key variable in our estimates, with its effects mediated through changes in BMI, systolic BP and cholesterol. This complicates interpretation because it then becomes necessary to show what permutations of combined diet and activity intervention may achieve the necessary magnitude of changes to BMI, systolic BP and cholesterol. The fact that the intervention is not cost-effective with 2,491 additional steps (in line with the effect size observed by Bravata)[271] means that, in order to determine the cost-effective threshold, extrapolation is required. The extrapolation makes an assumption of a linear relationship between step increase and change in biomarkers, however, the true relationship may be non-linear.

The Bravata study was considered to provide the most suitable data for the mapping between steps and risk factors. This study reported a seemingly large change in systolic BP (-3.8 mmHg) relative to the change in BMI (-0.38) for an increase of 2,491 steps. Further evidence confirming this relationship is desirable. If this ratio of systolic BP change relative to BMI is overstated, then the true results may be less favourable than presented. A further limitation is the inevitable disparity between the STOP Diabetes intervention and the heterogeneous mix of interventions (in terms of number of sessions, delivery period, end-point timing) that were pooled in the Bravata meta-analysis. The effects of uncertainty around the effectiveness and other parameter estimates were assessed using an illustrative PSA, however, it is not possible to incorporate the full uncertainty around the applicability of pooled estimates from Bravata et al. to this specific intervention in the target population.

Further uncertainty analysis would be required once the clinical effectiveness of the STOP programme has been quantitatively assessed.

There is a lack of good evidence on the durability of effects of an intervention such as STOP Diabetes especially when the maintenance sessions all take place within the same year as the initial intervention. Additionally, the STOP Diabetes cohort does not overall appear to be a particularly unhealthy one at baseline, perhaps due to recruitment criteria or self-selection, or just purely due to the average age of individuals recruited. There may be some obscure mechanisms that are not capture within the model structure and that drive the reported reduced life-expectancy for individuals with ID. If such mechanisms exist and can be modified by lifestyle intervention, then the results will not capture the economic impact of such benefits.

12.5.3 Comparison with related studies

There are several factors concerning the form of the intervention that lead to a high intervention cost per patient compared to other preventive lifestyle interventions:

1) Relative small group size of eight that the clinical team considered most appropriate for educating those with LD
2) Longer sessions for those with ID compared to the general population – 2 hours versus 1 hour 15 minutes for the NICE prevention modelling[302]
3) The need for 3 educators rather than two, and more advanced at a higher grade
4) The need for maintenance sessions to be spaced fairly close together, such as monthly, for information to be retained by individuals with ID. This compares with less frequent sessions, every four months in years two to four in the case of the modelling undertaken for the NICE diabetes prevention guidance.[302]

12.5.4 Implications

The purpose of the economic analysis was to provide a reasonable estimate of the cost-effectiveness of the STOP Diabetes intervention with a view to a subsequent full trial to assess effectiveness. The results of the current analysis suggest that, in the likely range of effectiveness achievable, the STOP Diabetes intervention in its

current form would not be cost-effective at a £ 20,000 to £ 30,000 cost/QALY threshold.

Risk profile of STOP Diabetes and ID population in general:
The large effect sizes needed for the intervention to be cost-effective are partly a result of the low risk profile of the STOP Diabetes cohort, in particular the average age at being 43.

The intervention primarily reduces risks of CVD and cancer. The evidence suggests that excess risks in ID cohorts compared to the general population are attributable primarily due to respiratory disorders, neurological diseases, congenital abnormalities and accidents. These risks are unlikely to be reduced through an intervention such as STOP Diabetes. The mortality aspects of this risks may be captured through the increased other-cause mortality for individuals with ID (see methods chapter), but other-cause mortality is not linked to the risk factors modified by the STOP Diabetes intervention (BMI, systolic BP, total and HDL cholesterol). It is therefore rational that an intervention targeting CVD risks, costing £ 1,097 (3-4 times the cost of diabetes prevention lifestyle interventions), will necessitate our reported very large reductions in risk factors in order to fall within usual NICE thresholds for cost-effectiveness.

The low risk profile is reflected in the mean QALY gain per person screened (with a subsequent increase in steps of 2,491 for suitable individuals) equating to an average of less than 1 day of additional life (in full health).

Equity is a factor that decision-makers take into account when deciding whether or not to recommend an intervention. Given that the ID population is a disadvantaged group, decision-makers might be prepared to pay more per QALY than for the general population [267] In practice this means that there is a greater likelihood of recommending an intervention at the upper end of the usual £ 20,000 to £ 30,000 per QALY range than for the general population. £30,000 per QALY tends to be the upper limit except in the context of end of life.

Although the average person with ID is overweight (and nearly obese), the 10-year CVD risk using cohort averages (mild/moderate severity only) is around only 2%. This is because risk factors seem to be well controlled in STOP Diabetes – average systolic BP was 121mmHg, and the lipid ratio was a healthy 3.63 – and it was a relatively young cohort.

A limitation of the STOP Diabetes intervention is that the reduction in CVD risk is likely to be confined to a few years following intervention. Combined with the average baseline age of 43 and associated low average CVD risk, this explains why the intervention has such as high cost per QALY for the overall group. An alternative more cost-effective structuring of the intervention sessions may be possible such that a smaller initial benefit is achieved but sustained for more years, possibly through additional maintenance sessions beyond the first year.

A minority, though significant, proportion of individuals identified as suitable for intervention may not make the desired progress towards reducing their risk factors for CVD. This could be due either to an individual's physiological response to the intervention or the numbers of intervention sessions that they actually attend, or a combination of both factors. For such patients, continuing with the maintenance session may be reducing the overall effectiveness of the intervention that might be more cost-effective, potentially at favourable cost-per-QALY levels, in those who achieve a good initial response.

12.5.5 Unanswered questions and further research

Further subgroup analyses could be undertaken in groups at 10%, 15% or 20% 10-year risk of CVD (diabetes risk could also be factored in), although such sub-groups would result in a very small proportion of the STOP Diabetes cohort actually receiving the intervention.

Further research is needed to identify the optimal mix of initial and maintenance sessions for physical activity programs, together with a better understanding of how long benefits are likely to last. Modelling may also help to inform primary research

into the optimal trade-off between investment in initial intervention and maintenance sessions.

Weight loss can be difficult to maintain even with ongoing maintenance as seen in the Finnish DPS. However, physical activity can help to maintain weight loss so it is unknown if benefits of physical activity can be sustained for longer than dietary-induced weight loss (hence why we explored 3 and 5 years as scenarios for durability of effect).

Further analysis may help to identify optimal permutations of the magnitude/intensity of the physical activity and dietary advice components to maximise the cost-effectiveness

CHAPTER 13. DISCUSSION AND CONCLUSIONS

13.1 Overview

In this chapter, we summarise and discuss the research programme's findings against each of its objectives. We also summarise the findings in terms of outputs, implications for practice, make research recommendations and discuss dissemination activities and plans for the research programme.

13.2 Development and assessment of the feasibility of a diabetes screening programme in adults with intellectual disabilities

13.2.1 Main findings

Adults with ID for the screening programme were identified through general practices, specialist ID services (through the Leicestershire Learning Disability Register), specialist ID clinics and through direct contact with the research team. In total, 930 (29% of those originally approached) took part in the screening programme; 38% were able to consent for themselves, other participants required a consultee. There were slightly more men than women in those screened (58%), participants were relatively young (mean age 43.3 years), mainly of white ethnicity (80%) and most were overweight (31%) or obese (37%). We were able to collect data on anthropometric measures for most participants (~86%), BP for (89%), and outcome data for 675 participants (73%) to assess the prevalence of IGR/T2DM.

13.2.2 Physical activity sub-study

We found that the objective measurement of physical activity is likely to be challenging in adults with ID, given that there are high levels of non-compliance. However, compliance could be substantially improved using wrist-worn monitors. Of 203 people approached, less than half (n=97; 48%) consented to wear the waist-

worn device, compared with 62% (47 of 76 approached) of those consenting to wear the wrist-worn device. Similarly, valid data were obtained from 57% (n=55) of the sample who wore the waist-worn devices, compared with 83% (n=39) of those wearing the wrist-worn devices.

Other studies among adults with ID have found a high proportion of missing data when using objectively measured physical activity data.[164, 233] However, to our knowledge, this is the first time the feasibility of collecting objectively measured physical activity data in those with ID has been formally assessed. The results suggest that poor compliance needs to be considered when conducting studies of physical activity interventions in this population. Researchers may also need to explore the potential for allowing separate consent in their study design for proposed accelerometer components.

Another somewhat unexpected finding was the high level of physical activity observed in our study population. We found that adults with ID engaged in similar amounts of physical activity as the general population, whereas most,[164, 233] but not all,[236] studies have found that people with ID generally engage less. This might reflect current policy drives to improve health and fitness in this population but may also indicate selection bias (i.e. active people preferentially choosing to wear the monitors) or behaviour change as a result of accelerometer wear.

13.3 Prevalence and demographic risk factors for type 2 diabetes and impaired glucose regulation in people with intellectual disabilities

The overall prevalence of screen detected (previously undiagnosed) T2DM was 1.3% (95% CI 0.5% to 2%) and IGR was 5% (95% CI 4% to 7%) among people with ID, which is lower than previously reported. Our systematic review (Chapter 2), found that the prevalence of diagnosed T2DM was approximately 8% (95% CI 5% to 11%), similar to that found in the general population. None of the studies in the review reported on screen-detected T2DM (they included prevalent known cases, we excluded) so it is not possible to make direct comparisons. Our lower than expected

rates of T2DM may simply reflect a successful annual health check programme, at least in the study's geographical location, and the younger age of participants.

Abnormal glucose levels were associated with non-white ethnicity (OR=3.93; 95% CI 2.10 to 7.33), a first degree family history of diabetes (OR=3.35; 95% CI 1.64 to 6.86), increasing weight, waist circumference, BMI, diastolic BP, triglycerides and decreasing HDL cholesterol.

13.4 Validation of the Leicester self-assessment diabetes risk score in people with intellectual disabilities

When the seven risk factors in the Leicester Self-Assessment risk score were used to explore risk of having undiagnosed IGR/T2DM among people with ID (with data available), the risk score achieved a sensitivity of 82% in identifying those with abnormal glucose regulation. High sensitivity is generally considered most important for screening tools because the priority is to 'rule out' the disease without missing true cases. Ninety-eight per cent of participants with a low/medium risk score were correctly identified as being at low risk. Our findings suggest that the Leicester Self-Assessment risk score is statistically effective at identifying people with ID who are at risk of undiagnosed IGR/T2DM and does not require modification, if it was integrated at practice level. However, it may not be practical or acceptable for people with ID to calculate their own score; development of an easy read version (plus a carer supplement) and additional supportive material would need to be explored.

13.5 Cost-effectiveness

Findings from the health economic analysis showed that, in its current form, the STOP diabetes multi-component intervention would need to result in a very large overall increase in steps, systolic BP, BMI and lipids, for it to be cost-effective at a threshold in the £ 20,000 to £ 30,000 cost per QALY range usually adopted by NICE.

The results would be favourable under the assumptions that:

1) the benefits of the intervention would not be fully lost until 4 years after the intervention (5 years from the start);
2) commissioners/payers would be willing to fund the intervention up to a threshold of £ 30,000 per QALY.

The cost-effectiveness of the intervention would be improved by targeting screening at the following groups, either:

- individuals aged over 45 years;
- individuals with BMI in the obese range;
- individuals with relatively high baseline risk of CVD.

However, it is still not cost-effective at readily achievable levels of change in steps and diet-attributable risk factors, unless the cost of the intervention could be reduced.

The relatively high cost of the STOP diabetes intervention compared with other similar multi-component behaviour change interventions is due to a number of factors:

- small samples needed for each group session;
- longer sessions;
- the need for three educators rather than two;
- the need for more experienced educators; and
- the need for regular, monthly refresher sessions.

Many of these factors were identified in advance as being important for the interventions to be appropriate and relevant for people with ID. It is known that the high support needs of this population, including co-existing challenging behaviour,[64] psychiatric disorders,[331] physical health problems[32] and communication difficulties make this a challenging group for behavioural interventions. We aim to explore other ways in which the intervention may be adapted to minimise resources, such as the potential to target carers under certain circumstances.

Finally, the findings also revealed a lack of good quality evidence for the durability of effects of multi-component behaviour change interventions, such as that developed for the STOP diabetes programme.

13.6 Data linkage to Hospital Episode Statistics and the Office for National Statistics

In total, 883 (95%) participants gave consent for the research team to follow-up their health in the longer term via data linkage.

13.7 Development of a lifestyle education programme for people with intellectual disabilities and impaired glucose regulation

The research involved the development of a structured lifestyle education programme for a population with ID with IGR or at high-risk of developing T2DM and/or CVD based on a high BMI. This was a complex process encompassing initial curriculum development, two cycles of testing, evaluation, modification and re-testing, prior to final refinement of the programme.

The STOP programme development benefitted from a systematic process.[238, 239] The theoretical underpinning was developed and expanded upon from the limited evidence in the literature. This informed the content and style of approach, alongside the qualitative findings from people with ID, their carers, and health care professionals with expertise in working with people with ID. The whole programme was then tailored further to the specific needs of this group by more user feedback, and adaptation by a multi-disciplinary team with expertise in ID and the development of education programmes (with psychological underpinning).

From the initial phases the programme has been well received and is acceptable to the people it is trying to support. The initial feedback via qualitative interviews has suggested that some of the elements of treatment receipt initially hypothesised may have been achieved, via reported changes in beliefs and health behaviours.

The research also involved an assessment of feasibility of collecting outcome measures from participants with ID before and 3-months after delivering the intervention programme. For this component, our findings suggest that it is both acceptable and feasible to collect outcome measures for weight, height, BMI, waist

circumference, BP and dietary intake (portions of fruit and vegetables), and objective measures for physical activity and sedentary behaviour, using wrist-worn accelerometers, both before and after (3-months) attending the programme. At baseline, anthropometric measures and BP were obtained for all participants, and accelerometer data for 80%. Attendance at the education programme was overall good, with 80% of participants attending ≥5 days (out of seven sessions for the main programme). At 3-months follow-up repeat data were successfully collected for a high proportion of participants (anthropometric measures 100%; BP 80%; accelerometer data 60%). Owing to time restrictions, we were only able to conduct one feasibility cycle and were also unable to assess whether it is possible to collect longer term data, but these preliminary findings are overall positive.

Only four of the five participants who took part in the intervention agreed to wear the wrist-worn accelerometers at baseline, and this suggests that an assessment of willingness to wear the accelerometer is an important component of any future evaluation work. Furthermore, the feasibility component of our work suggested that lifestyle circumstances could play an important role in adhering to the education programme and this needs to be considered for future work.

13.8 Development of an intervention fidelity process for the assessment of educators delivering the intervention

As part of this research, we successfully completed the first step in developing a tool for assessing intervention fidelity of the STOP diabetes educational programme. Preliminary findings using the tool already suggest some variance between educators, which will provide a benchmark for future work. One of the key considerations for this component of the research involved reconsidering existing learning methods that are known to be effective in the general population to meet the needs of people with ID. This included removing abstract concepts, avoiding abbreviations and jargon, teaching at the group's pace and, above all, avoiding isolating the learners by 'putting them on the spot' to summarise key messages,

13.9 Main findings and outputs

The main findings and outputs arising from this extensive research programme are summarised below:

- We developed and assessed the feasibility of a diabetes screening programme for adults with ID.
- In total, 930 (29% of those originally approached) people with ID took part in the screening programme; 58% were men and the average (mean) age of participants was 43 years old.
- Most people who took part in the screening programme (68%) were overweight or obese.
- We were able to collect blood samples from 73% of participants and anthropometric measures on more than 85% of participants.
- We found that wrist-worn accelerometers were more acceptable to participants with ID than waist-worn accelerometers to measure physical activity.
- We found that 1.3% of people with ID had undiagnosed T2DM and 5% of people with ID had IGR (screen detected).
- We found that abnormal glucose tolerance was associated with non-white ethnicity, first degree family history of diabetes, increasing weight, waist circumference, hip circumference, BMI, diastolic BP, triglycerides and lower HDL cholesterol.
- We developed a lifestyle intervention programme for a population with ID with IGR or at high-risk of developing T2DM and/or CVD based on a high BMI.
- Using concrete messages and visual aids facilitated learning in this group; abstract and conceptual examples tended to be less well received.
- We found that the collection of outcome measures prior to and after (3-months) delivering the intervention was both acceptable and feasible.
- We identified that for the intervention to be cost-effective (£20,000 to £ 30,000 cost per QALY range), the required change in steps and diet-attributable risk factors may be more than is achievable in practice

- We found that if commissioners were willing to fund the intervention up to a higher threshold, cost-effectiveness may improve by targeting specific individuals (>45 years; obese, high CVD risk).
- We developed a preliminary quality development tool to assess intervention fidelity of the educational programme for people with ID.

13.10 Limitations

We have found that conducting a programme of research to enhance the knowledge and understanding of IGR and T2DM in people with ID, including development of a lifestyle education programme, is feasible but not without challenges. We acknowledge the following limitations:

- With regard to the systematic review of the evidence in relation to prevalence of T2DM and IGR, we acknowledge that limited data were available on T2DM in people with ID and that reported outcomes were sometimes poorly defined or unclear. We would also have benefited from more general population data for comparison.
- Similarly, for the systematic review of long-term multicomponent behaviour change interventions for the prevention of CVD and T2DM in people with ID, we acknowledge that only four papers met our inclusion criteria which limited our ability to draw meaningful conclusions. However, our findings do highlight the lack of work in this area and the need for robust interventions, such as that developed for this programme of work.
- Despite highlighting a number of achievements in involving service users in our research programme, we acknowledge that we could have done more to involve them in the design and dissemination phases of our programme.
- We acknowledge that the recruitment approach utilised for the screening study may not be transferable to other geographical areas in England. Recruitment was facilitated by the Leicestershire Learning Disability Register[14] (either via direct invitation from the register, or for people previously agreeing to be approached about future research) which accounted for 40% of people invited (~39% of participants). The register is only one of three adult ID case registers in England and has a strong research tradition. However, we

only approached people via this route for general practices that declined to take part in the study and we feel that approaches such as direct invitation and invitation via ID psychiatric service clinics could be replicated in other areas.

- We acknowledge some difficulties in recruiting services users to the development phase (work package 2) despite using a direct approach to people who had already participated in the screening phase. For the qualitative development interviews low recruitment was largely due to an initial lack of people who were either ineligible (based on severity of ID, and/or BMI ≥25 or IGR) or unwilling. Additionally, for the later phases, where participation involved attending a course of education sessions held over several weeks (with little flexibility in scheduling), reported barriers were largely linked to the regular daytime commitments (social activities/work/education) of service users which they were either unwilling or unable to change. The "busy schedules" of potential participants has previously been identified as a barrier to recruitment for people with ID.[332] Unfortunately, within the constraints of this research study there was no flexibility to offer alternative dates to attend the programme. However, for the second pilot education cycle, which was held in a residential setting, the day and timing of sessions were arranged as much as possible to suit the needs of both service users and care workers, and recruitment levels were much higher.
- For the economic evaluation, we acknowledge the exploratory nature of the work, given that data on clinical effectiveness for the STOP Diabetes programme were not available. In particular, the analysis involved extrapolating data outcomes, which assumed a linear relationship between step increase and changes in biomarkers (BMI, systolic BP and cholesterol), which may not reflect their true relationship.
- We further acknowledge limitations with using the EQ-5D for the economic evaluation since this has not been validated in people with ID. We look forward to the outputs from current work to validate the EQ-5D in this population.[159]

13.11 Implications for practice

We have found that, at least in Leicester/Leicestershire, there is a low prevalence of previously undiagnosed (screen detected) IGR/T2DM. However, we also found that a significant proportion of people with ID are overweight or obese and are likely to be at risk of developing T2DM and/or CVD in the future. Our non-invasive risk score might also help to identify people at risk of undiagnosed IGR/T2DM. The development of the STOP diabetes educational programme is the first stage in identifying preventative strategies for future research.

13.12 Research recommendations

We make the following recommendations for further research:

- The recruitment rate for the screening study was relatively low (29%). In some cases the use of gatekeepers, including general practitioners, residential home managers and family carers presented a barrier to recruitment. We recommend utilising a multi-pronged/multi-layered approach, actively engaging with both intermediaries and service users, and following up all potential participants to ensure people are given an equitable chance to participate.
- In order to be truly inclusive, we highlight the importance of making reasonable adjustments, including offering appointments whenever and wherever is most appropriate for the person, minimising disruption to their routine and ensuring that appropriate support is in place. Given limited resources, it is likely that researchers and funders need to lower the threshold for an ‘acceptable’ response in this population, so that adults with ID are not excluded altogether from taking part in research.
- We recommend a staggered consent process when recruiting people with ID into research to enable them to opt out of some components, such as having blood tests or wearing accelerometers.

- We have demonstrated that adults with ID can be meaningfully involved in the research process; we recommend exploring further ways in which people with ID can be involved in research and be recompensed for their time.
- We recommend further work to explore ways in which compliance with accelerometer wear can be improved in people with ID.
- We recommend ongoing monitoring of the participants in our study to identify longer term health and mortality outcomes.
- Finally, we have found preliminary evidence that the STOP Diabetes education programme is acceptable and feasible. We recommend further work to evaluate its clinical and cost effectiveness in a randomised controlled trial informed by the Medical Research Council framework for evaluating complex interventions,[238] with a view to integrating the programme into national preventive strategies and reducing health inequalities among people with ID.

13.13 Dissemination activities and plans

During the consent process, participants were asked if they wished to be informed of the findings. Between September and December 2015 we disseminated the results to participants (and carers). Two of the ID research nurses visited 57 homes (group homes, supported living, residential and nursing homes) to present the findings to participants in easy-read format supplemented by verbal explanations/presentations. Other participants received a brief easy-read report sent in the post. We have begun to disseminate the findings to healthcare professionals locally, both in primary care and within ID services.

The work from the service user involvement component of this research has been published in one of the NIHR INVOLVE newsletters[197] and in the academic literature.[202] The initial education development work has previously been presented at the Diabetes UK Professional Conference in March 2015.[333] Similarly, the two systematic reviews for this programme and the screening study were presented at the 2016 Diabetes UK Professional Conference.[334-336]

The next steps will involve writing up and submitting academic articles in relation to the individual components of the research programme. This will include the screening study, risk score validation, cost-effectiveness component, intervention development and updated versions of the two systematic reviews. We will continue to present the findings both locally, through existing collaborations with NIHR CLAHRC East Midlands and the East Midlands Academic Health Services Network (AHSN), and nationally. We have been invited to present our work at a meeting of the Royal Society for Medicine Intellectual Disability Forum (Managing diabetes in people with intellectual disabilities: recent advances) in November 2016.

13.14 Summary

Results from this programme of work have significantly enhanced existing knowledge and understanding of T2DM and IGR in people with ID, and have enabled us to test strategies for early identification of IGR and T2DM in people with ID. This is the first large diabetes screening study in people with ID in the UK and to our knowledge the largest screening study globally. We have also developed a lifestyle education programme and educator training protocol to promote behaviour change in a population with ID at risk of developing T2DM. Further work is now needed to evaluate the intervention we have developed and to identify cost-effective strategies for its implementation.

ACKNOWLEDGEMENTS

This project was funded by the NIHR Programme Grants for Applied Research and will be published in full in *Programme Grants for Applied Research*; Vol. X, No. X. See the (PGfAR programme website) for further project information. The work by TC was supported by a PhD studentship funded jointly by the University of Leicester and Leicestershire Partnership NHS Trust.

The authors gratefully acknowledge the people with ID, their families and health professionals who took part in the STOP diabetes study. We would especially like to thank the contribution from service users and facilitators from the 'Speaking up for Health' and the 'Charnwood Action' self-advocacy groups who helped us throughout the research process.

We acknowledge help from key people throughout the STOP diabetes research programme: Colin Greaves for his excellent chairing of the steering group meetings; research nurses, Ella Bailey, Clare Makepeace, Kay Massey, Elaine Perkins and Paul Underwood; administrative expertise from Yvette Walters; educational programme support from Marian Carey, Director of the DESMOND programme and support from Lesley Green (Leicestershire Learning Disability Register).

We also thank the following people who provided useful help and support at various stages of the programme: Navneet Aujla, Kiran Bains, Michael Bonar, Lesley Bryan, Mandy Clarkson, Heather Daly, Karen Davis, Charlotte Edwardson, Jules Galbraith, Panna Mandalia-Wilson, Jacqui Troughton and Shelley Winterton.

Data sharing – all available data can be obtained from the corresponding author.

AUTHOR CONTRIBUTIONS

Dr Alison Dunkley (Research Associate in Nursing) was the lead researcher/project manager for the programme and was responsible for its design and conduct. She also authored chapters of the manuscript and sat on the steering group.

Freya Tyrer (Research Fellow in Epidemiology) led on recruitment from the Leicestershire Learning Disability Register, authored a chapter of the manuscript and sat on the steering group.

Rebecca Spong (Research Assistant) contributed to all components of the programme and contributed to chapters of the manuscript.

Dr Laura Gray (Senior Lecturer of Population and Public Health Sciences) designed and analysed the quantitative results and oversaw their reporting and interpretation. She also authored a chapter of the manuscript and sat on the steering group.

Mike Gillett (Research Fellow) designed and led on the health economics component of the programme, authored a chapter of the manuscript and sat on the steering group.

Dr Yvonne Doherty (Consultant Clinical Psychologist) co-led on the development of the education programme, contributed to chapters of the manuscript and sat on the steering group.

Lorraine Martin-Stacey (Senior Research Associate) co-led on the development of the education programme and contributed to chapters of the manuscript.

Naina Patel (Research Associate) conducted the interviews with health care professionals, service users and carers for the research programme, analysed the qualitative results, contributed to chapters of the manuscript and sat on the steering group.

Dr Tom Yates (Reader in Physical Activity, Sedentary Behaviour and Health) contributed to the initial grant application, advised on evidence relating to physical activity and cardiovascular risk for the health economics chapter and authored a chapter of the manuscript.

Professor Sabyasachi Bhaumik (Consultant Psychiatrist) was the Principal Investigator from Leicestershire Partnership NHS Trust, contributed to the initial grant application, facilitated recruitment from specialist ID clinics, sat on the steering group and reviewed the final manuscript.

Thomas Chalk (PhD student) led on the two systematic reviews for the programme and reviewed the final manuscript.

Yogini Chudasama (Research Assistant in Medical Statistics) analysed the quantitative results and led on their reporting and interpretation under the supervision of Laura Gray. She reviewed the final manuscript.

Chloe Thomas (Research Assistant) made a substantial contribution to the health economics component of the programme under the supervision of Mike Gillett and contributed to a chapter of the manuscript.

Susannah Sadler (Research Associate) made a substantial contribution to the health economics component of the programme under the supervision of Mike Gillett and contributed to a chapter of the manuscript.

Professor Sally-Ann Cooper (Professor of Learning Disabilities) contributed to the initial grant application, sat on the steering group and reviewed the final manuscript.

Dr Satheesh Kumar Gangadharan (Medical Director of Leicestershire Partnership NHS Trust) contributed to the initial grant application, sat on the steering group and reviewed the final manuscript.

Professor Melanie Davies (Professor of Diabetes Medicine) contributed methodological and practical advice to the research programme and reviewed the final manuscript.

Professor Kamlesh Khunti (Professor of Primary Care Diabetes & Vascular Medicine) was the Chief Investigator for the study and conceived the idea for the study, contributed methodological and practical advice to all components of the research programme and publications arising from the research.

PUBLICATIONS

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1. Dunkley A, Clarkson M, Tyrer F. *STOP Diabetes Study. INVOLVE Summer 2013 Newsletter*; 2013.

2. Martin-Stacey L, Doherty Y, Makepeace C, Patel N, Spong R, Dunkley AJ. The systematic development and theoretical framework of a lifestyle educational programme for prevention of Type 2 diabetes in a population with intellectual disabilities: P338. *Diabetic Medicine* 2015;32 (Supplement 1):132

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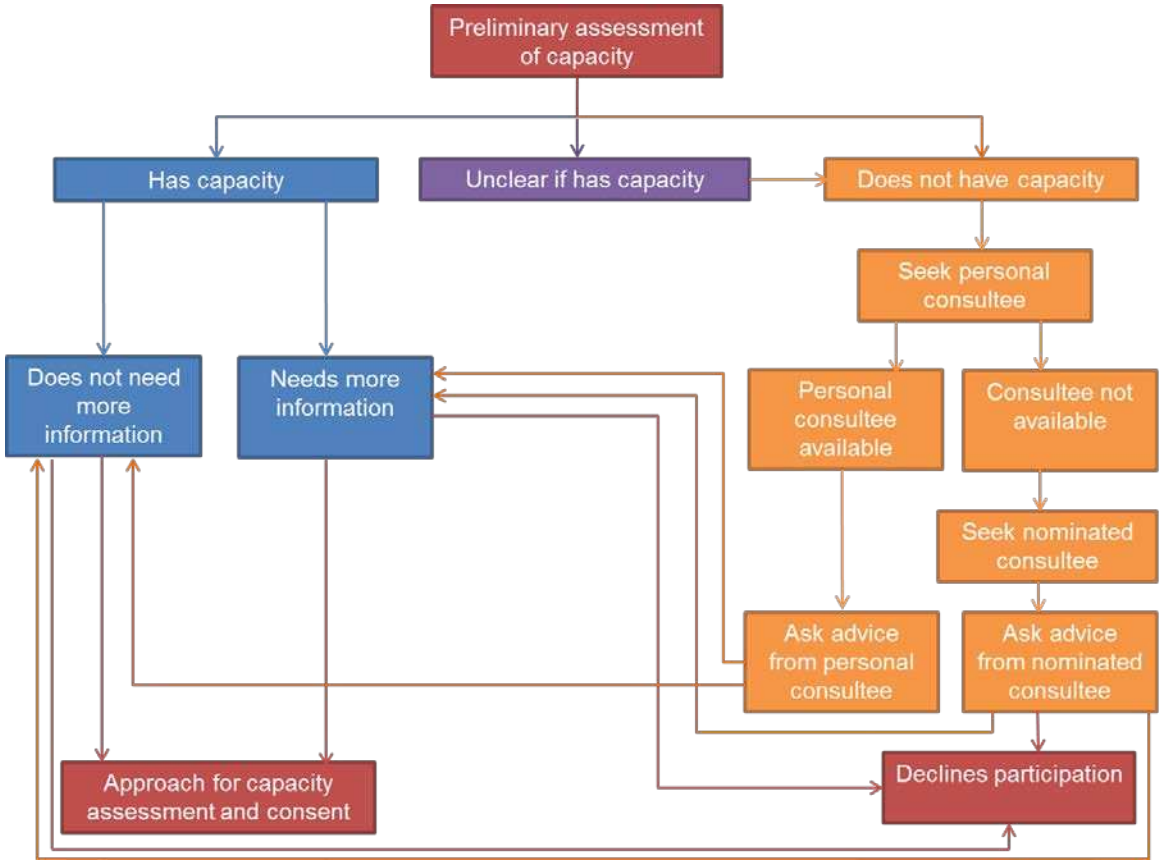


Figure 26: Diagram showing how capacity and consent were assessed in the study

(flowchart adapted from Dixon-Woods and Angell)[337]

Appendix 2: Example from Leicester Self-Assessment Risk Score

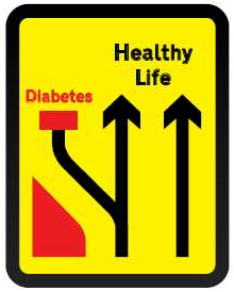
COULD **YOU** HAVE TYPE 2 DIABETES?

Type 2 diabetes develops when the body cannot control the amount of sugar in the blood. Type 2 diabetes can develop over a number of years without any symptoms. You can use this questionnaire to work out your own 'risk' of Type 2 diabetes.

What does 'risk' mean?

'Risk' of Type 2 diabetes means how high is the chance of you having Type 2 diabetes now, or getting it in the future. Things which increase your risk of having Type 2 diabetes are called risk factors. Example risk factors are: being older, or having diabetes in your family.

This questionnaire asks you about your risk factors to give you a 'risk score'.



How will knowing your risk score help you?

Knowing your risk score will tell you what you need to do next. For some people, this may mean talking to their GP.

If you find out that you already have Type 2 diabetes, the good news is that being diagnosed sooner rather than later may help to prevent or delay future problems. You could reduce your chances of having heart attacks and strokes, and serious problems with your eyes, feet and kidneys.

If you find out that you have a higher risk of getting Type 2 diabetes in the future, there is some good news for you too. Making a few small changes (for example, to what you eat) can prevent or delay diabetes.

QUESTIONNAIRE: Do you want to know your risk of Type 2 diabetes?

For each question, tick one white box (✔).

1. Which age group are you in?					
49 years and younger		0	60-69 years		9
50-59 years		5	70 years and/or older		13
2. Are you male or female?					
Male		1	Female		0
3. How would you describe your ethnicity?					
White European		0	Any other ethnic group		6
4. Do you have a parent, brother, sister and/or child with Type 1 or Type 2 diabetes? (Do not count step-relatives)					
Yes		5	No		0
5. Which waist size group are you in? (See instructions on page 5)					
Less than 90 cm (35 inches)		0	100 -109 cm (39-42 inches)		6
90-99 cm (35-38 inches)		4	110 cm (43 inches) and above		9
6. Which Body Mass Index (BMI) group are you in? (See explanation and instructions on pages 6 and 7)					
Less than 25		0	30-34		5
25-29		3	35+		8
7. Have you ever been told by a doctor or nurse that you have high blood pressure?					
Yes		5	No		0
To get your risk score, add up the numbers in the blue boxes next to the seven boxes that you have ticked. Write the total number here – **This is your risk score:** To find out what this means go to pages 8 and 9					

Appendix 3: Outcome definitions for T2DM and CVD prevalence and risk factors

(see main report for reference list)

Table 64: Outcome definitions for articles included in the systematic review of T2DM and CVD prevalence and risk factors

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
Molteno (2000)			**Obese** BMI >30 **Overweight** BMI 25–<30				MILD 0.3% MOD 18.7% SEV 37.7% PROF 33.5% **MISSING DATA**
Robertson (2000)			**Obese** BMI >30 **Overweight** BMI 25.1–30				
Janicki (2002)	**Cardiovascular disease** ǂ NR	**Diabetes** ǂ Adult onset	**Obese** ǂ BMI >27 **Overweight**ǂ BMI 22–27	**Hypertension** ǂ NR	**Hyperlipidaemia** ǂ NR		MILD 1.3% MOD 50.3% SEV/PROF 47%
Lewis (2002)			**Obese** BMI ≥30 **Overweight** BMI 25–29.9	**Elevated BP** SBP >140mmHg or DBP >90mmHg	**Hypercholesterolemia** Total cholesterol ≥ 13.3mmol/L		MILD 37.1% MOD 16.4% SEV 14.7% PROF 15.3%
Marshall (2003)			**Obese**	**Hypertension**	**Elevated**		

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
			BMI ≥31 **Overweight** BMI 26–30	SBP >140mmHg	**Cholesterol** Definition NR		
Havercamp (2004)	**Cardiovascular disease** ǂ Definition NR	**Diabetes** ǂ Definition NR	**Obese** definition NR – BMI data were collected **Overweight** definition NR – BMI data were collected	**Elevated BP** ǂ Definition NR			MILD 39.4% MOD 26.6% SEV 14.7% PROF 10.6 %
Hove (2004)			**Obese** BMI ≥30 **Overweight** BMI 25–29.9				MILD 39.2% MOD 42.1% SEV 15.5%
Merrick (2004)	**Heart disease** ǂ Definition NR	**Type 2 diabetes** ǂ Definition NR	**Overweight and above** ǂ BMI >27	**Hypertension** ǂ Definition NR	**Hyperlipidaemia** ǂ Definition NR		
Moore (2004)			**Obese** BMI ≥30 **Overweight** BMI 25 –<30				
Emerson (2004)			**Obese *** BMI > 30 **Overweight *** BMI 25.1–30				
Yen (2005)			**Obese** ǂ BMI ≥27				MILD 22.2% MOD 34.9%

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
			Overweight[ǂ] BMI 24–<27				SEV 28.1% PROF 14.8%
Ito (2006)			**Obese** BMI >30 **Overweight** BMI 25–30				
Lennox (2006)			**Obese** BMI >30 **Overweight** BMI 25–30	**Elevated BP** SBP>140mmHg			
Levy (2006)		**Diabetes*** Definition NR	**Obese *** BMI ≥30 **Overweight** BMI 25–29.9 **Obese/overweight** ≥25	**Elevated BP*** **Definition NR**	**Hypercholesterolemia*** Definition NR		MILD 47.6% MOD 31.1% SEV 14.6% PROF 6.8%
McDermott (2006)	**Coronary artery disease*** ICD-9-codes **Transient ischemic attack*** ICD-9- codes	**Type 1 & Type 2 Diabetes*** ICD-9- codes	**Obese*** NR	**Hypertension & Elevated BP*** ICD-9- codes			
Rurangirwa (2006)			**Overweight/obese**[ŧ] ≥ 25				
Shah (2006)		**Diabetes**[ŧ]					

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
		Definition NR					
Van Den Akker (2006)	**Coronary heart disease*** ICD-10-codes **Cerebrovascular disease*** ICD-10-codes			**Hypertension*** ICD-10-codes			MILD 11% MOD 53% SEV 28% PROF 8%
Levy (2007)		**Diabetes*** Definition NR	**Overweight and above** BMI ≥25	**Elevated BP*** Definition NR	**Hypercholesterolemia*** Definition NR		SEV 65.4% PROF 34.6%
McDermott (2007)		**Diabetes*** Although a detailed description is given, it is not possible to define the type of diabetes is used as an outcome.					
McGuire (2007)			**Obese** ǂ BMI >30 **Overweight** ǂ BMI >25				MILD 14.1% MOD 63.5% SEV 12.8% PROF 9%
Wang (2007)	**Heart disease**ǂ ICD-9-codes Specific codes in manual for the Rochester health status survey (includes		**Overweight and above**ǂ				

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
	some non-CVD codes)						
Bhaumik (2008)			**Obese *** BMI ≥ 30 **Overweight** BMI 25.1– <30	Hypertension * SBP ≥140 mmHg and/or DBP ≥ 90mmHg			
Henderson (2008)		**Type 2 diabetes*** Derived from medical problem lists	**Obese*** BMI >30 **Overweight*** BMI ≥ 25 ≤ 30	**Hypertension*** Derived from medical problem lists	**Dyslipidaemia*** Derived from medical problem lists		
Melville (2008)			**Obese** BMI ≥30 **Overweight** BMI 25–<30				MILD 40.9% MOD 25.1% SEV 18.2% PROF 15.8%
Wallace (2008)	**Cardiovascular disease *** History of: Peripheral vascular disease, stroke, or coronary heart disease.	**Elevated glucose *** >6.1mmol/L (fasting and non-fasting tests grouped together in results) **Type 1 & 2 diabetes**	**Obese *** BMI ≥ 30 **Overweight** BMI 25-29.9	**Hypertension *** SBP >140mmHg	**Elevated cholesterol *** >5.5mmol/L (fasting and non-fasting tests grouped together in results)		
De Winter (2009)	**Cerebrovascular disease*** Diagnosed by CT scan **Myocardial infarction*** Diagnosed by ECG	**Diabetes** glucose ≥ 7.0 mmol/L or use of anti-diabetic drugs.	**Obese** BMI ≥ 30	**Hypertension** SBP ≥ 140mmHg or use of drugs	**Hypercholesterolemia** Total cholesterol >5.1mmol/L to ≥6.5 mmol/L (depending on		MILD 12.1% MOD 33.2% SEV 34.3% PROF 20.4%

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
	changes				laboratory reference values) or use of cholesterol lowering drugs **Elevated LDL** ≥3.5 mmol/L		
Gale (2009)			**Obese** BMI 30–<40 **Severely Obese** ≥ 40 **Overweight** BMI 25–<30				
Henderson (2009)			**Overweight or above** ǂ BMI ≥25				MILD/MOD 53% SEV/PROF 47%
Maaskant (2009)			**Obese** BMI ≥ 30 **Overweight** BMI 25–<30				
Moss (2009)		**Elevated glucose** Non-fasting test – definition NR	**Overweight and above** BMI >25	**Hypertension** Definition NR	**Elevated total cholesterol** Non-fasting test – definition NR		
Sohler (2009)		**Diabetes***	**Obese***	**Hypertension***	**Hypercholestero**		

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
		Definition NR	BMI ≥ 30 **Overweight*** BMI 25–29.9	Definition NR	**lemia*** Total cholesterol >13.3mmol/L		
Van De Louw (2009)				**Hypertension** SBP >140mmHg			MILD 10% MOD 38% SEV/PROF 52%
Shireman (2010)		**Diabetes*** ICD-9-codes					
Stedman (2010)			**Obese*** BMI ≥ 30 **Overweight*** BMI 25–29.9				
Tyler (2010)	**Coronary heart disease*** ICD-9-codes	**Diabetes*** ICD-9-codes	**Obese*** ICD-9-codes	**Hypertension*** ICD-9-codes	**Hyperlipidaemia*** ICD-9-codes		
Chen (2011)	**Heart disease** Such as cardiac arrhythmias and coronary atherosclerosis. Diagnoses based on clinical manifestations or ECG findings.	**Elevated blood glucose** exceeding normal range 3.9–6.1 mmol/L **Diabetes** FPG ≥ 7mmol/L or 2h plasma glucose ≥ 11.1 mmol/L or OGTT 2h >11.1mmol/L		**Hypertension** SBP ≥ 140mmHg or DBP ≥ 90mmHg	**Elevated total cholesterol** ≥ 6.2 mmol/L **Elevated triglycerides** ≥2.26 mmol/L		
Frighi (2011)		Type 2 diabetes	**Overweight or**				MILD 48%

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
		NR	**above** definition NR – BMI data & WC were collected				MOD 30.2% SEV/PROF 21.8%
POMONA II study Haveman (2011) + Martinez-Leal (2011) (Obesity data)	**Heart attack** ǂ Definition NR **Cerebrovascular disease** ǂ Definition NR	**Diabetes**ǂ Definition NR	**Obese** definition NR – BMI data were collected **Overweight** definition NR – BMI data were collected	**Hypertension** ǂ Definition NR			Haveman MILD 22.7% MOD 28.2% SEV 20.7% PROF 11.8% Martinez-leal MILD 21.8% MOD 27.7% SEV 19.7% PROF 11.4%
Hsu (2011)			**Overweight or above *** BMI ≥ 24			3/5 criteria NCEP-ATPII	MILD/MOD 47% SEV/PROF 53%
Lee (2011)	**Cardiac illness*** History of coronary heart disease or congestive cardiac failure	**Diabetes*** implied by prescription of hypoglycaemic drugs	**Obese*** BMI ≥ 31 **Overweight*** BMI 26–30	**Hypertension*** Definition NR			MILD 33% MOD 22% SEV 23% PROF 21%
Stancliffe (2011)			**Obese** BMI $\geq$ 30 **Overweight**				

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
			BMI ≥ 25–<30 **Overweight and above** BMI ≥25				
Wong (2011)	**Heart disease**[ǂ] Definition NR **Cerebrovascular disease**[ǂ] Definition NR	**Diabetes**[ǂ] Definition NR	**Overweight and above**[ǂ] BMI >23	**Hypertension**[ǂ] Definition NR	**Hypercholesterolemia**[ǂ] Definition NR		MILD 4.9% MOD 41.8% SEV/PROF 51.9%
Chang (2012)		**Elevated blood sugar** FPG ≥ 5.6 mmol/L or use of drugs	**Obesity** BMI (definition NR) **Overweight** BMI (definition NR) **Central overweight** FWC ≥ 80cm/MWC ≥ 90cm	**Hypertensive SBP** ≥ 130mmHg or use of drugs **Hypertensive DBP** ≥ 85mmHg or use of drugs	**Elevated triglycerides** ≥8.3 mmol/L (or use of drug) **Reduced HDL** HDL Male < 2.2 mmol/L, Female < 2.8mmol/L (or use of drugs)	3/5 criteria NCEP-ATPIII and MetS criteria for Taiwanese people	MILD 65% MOD 16% SEV 9% PROF 10%
De Winter (2012)_1 HA-ID study			**Obesity** BMI ≥ 30 **Overweight** BMI ≥ 25 **Central obese** FWHR ≥ 88cm/MWHR ≥				MILD 24.8% MOD 48% SEV 16% PROF8.9%

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
			102cm **Central overweight** FWHR ≥ 80cm/MWHR ≥ 94cm				
De Winter (2012)_2 HA-ID study		**Diabetes** FSG ≥ 6.1 mmol/L or use of drugs		**Hypertension** SBP ≥ 140mmHg Or DBP ≥ 90mmHg and/or medication	**Hypercholesterolemia** Fasting serum total cholesterol >6.5 mmol/L or use of drugs	**Defined separately by:** 3/5 criteria (joint interim statement) and 3/5 criteria NCEP-ATPIII	MILD 24.5% MOD 48.6% SEV 16% PROF 8.7%
Gazizova (2012)			**Obese** BMI >30 **Overweight** BMI 25.1–30				MILD 61% MOD 24% SEV 15%
Lin, L.P. (2012)				**Hypertension** SBP ≥ 140mmHg or DBP ≥ 90mmHg			
Morin (2012)	**Heart disease** [+] ICD-10-codes	**Diabetes** [+] ICD-10-codes					MILD 32.9% MOD 46.4% SEV 11.2% PROF 5.2%
Begarie (2013)			**Obese**				

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
			BMI ≥ 30 **Overweight** BMI ≥ 25–<30				
De Winter (2013) HA-ID study	**Peripheral arterial disease** Ankle-Brachial-Index ≤ 0.9 (measured only in subjects with >1 CVD risk)						MILD 24.9% MOD 53% SEV 13.4% PROF 4.6%
Haider (2013)	**Heart disease** ǂ ever diagnosed by a doctor/relevant healthcare professional **Stroke** ǂ ever diagnosed by a doctor/relevant healthcare professional	**Type 2 diabetes** ǂ In the paper it groups type 1 and 2 together, but in a separate report outcomes are available separately, it also says if been told by doctor	**Obese** ǂ BMI >30 **Overweight** ǂ 25–<30				
Jansen (2013)	**Cerebrovascular accident*** acute disruption of cerebral circulation with focal neurological symptoms ≥24hr **Myocardial infarction*** clinical signs & ECG diagnosis and/or lab						MILD 6.9% MOD 37.8% SEV 29% PROF 26.3%

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
	results						
Lin, J.D. (2013)		**Hyperglycaemia *** FPG ≥ 7mmol/L		**Hypertension *** SBP ≥ 140mmHg or DBP ≥ 90mmHg or use of drugs	**Hyperlipidaemia *** Triglyceride ≥ 11.1mmol/L or Total cholesterol ≥ 13.3mmol/L		
McCarron (2013)	**Heart disease** ǂ History of Angina, heart attack, coronary heart failure, open heart surgery (ever diagnosed by a doctor/relevant healthcare professional) **Stroke/TIA** ǂ ever diagnosed by a doctor/relevant healthcare professional			**Hypertension** ǂ ever diagnosed by a doctor/relevant healthcare professional			
Vacek (2013)				**Hypertension*** ICD-9-codes			
Hsieh (2014)			**Obese** ǂ BMI ≥30 **Overweight** BMI 25 – <30				MILD 44.9% MOD 23.7% SEV/PROF 8.4%
Mikulovic (2014)			**Obese** BM I>30				

Author/year	CVD outcomes	Diabetes/blood sugar outcomes	Obesity/Overweight outcomes	Blood pressure outcomes	Lipid outcomes	Metabolic syndrome	Split by ID severity
			Overweight BMI ≥25				
De Winter (2015)		**T1DM** **T2DM** **Diabetes** FSG ≥ 6.1 mmol/L or use of drugs	**Central obese** FWHR ≥ 88cm/MWHR ≥ 102cm **Central overweight** FWHR ≥ 80cm/MWHR ≥ 94cm	**Hypertension** SBP ≥ 140mmHg Or DBP ≥ 90mmHg and/or medication	**Hypercholesterolemia** Fasting serum total cholesterol >6.5 mmol/L or use of drugs	**Defined separately by:** 3/5 criteria (joint interim statement) and 3/5 criteria NCEP-ATPIII	MILD 24.5% MOD 48.6% SEV 16% PROF 8.7%
Lin, L.P. (2015)			**Obese** BMI ≥27 **Overweight** BMI 24–26.9				MILD 6.5% MOD 32.6% SEV 34.8% PROF 26.1%
Zaal-Schuller (2015)	**Peripheral arterial disease** Ankle-Brachial-Index <0.9						MILD/MOD 51.1% SEV/PROF 48.9%

*retrospective data extracted from database/medical records, or, ǂ data self-reported or reported by carer; NR (not reported); SBP (systolic blood pressure); DBP (diastolic blood pressure); HDL (high density lipoprotein); LDL (low density lipoprotein); BMI (body mass index); FPG (fasting plasma glucose); MWC (male waist circumference); FWC (female waist circumference); MWHP (male waist-to-hip ratio); WWHP (female waist-to-hip ratio).

Definitions

Ischaemic heart disease: defined as ischaemic heart disease, myocardial infarction, heart attack, coronary atherosclerosis and/or coronary artery disease.
Cerebrovascular disease: defined as cerebrovascular disease, stroke and/or transient ischaemic attacks
Undefined CVD: defined as undefined heart disease, undefined CVD, or a combined CVD outcome where the majority is undefined.
T2DM: defined as T2DM only

Appendix 4: Funnel plot for T2DM

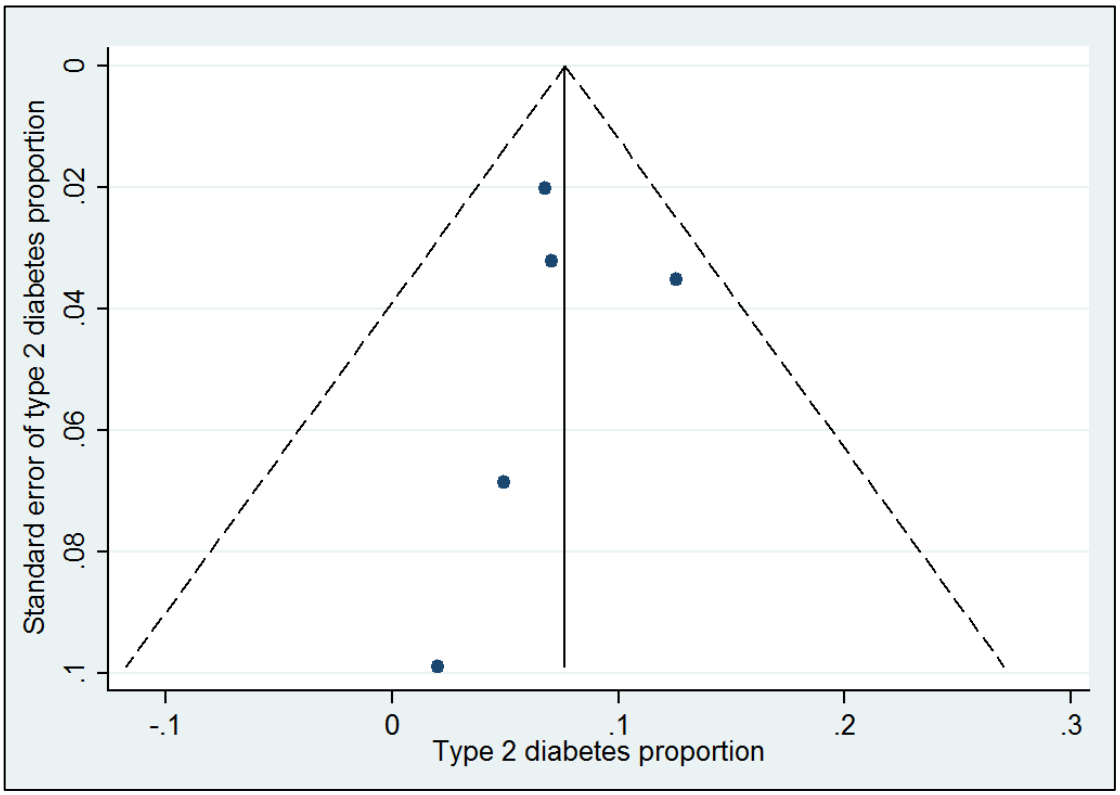


Figure 27: Funnel plot with pseudo 95% confidence limits for T2DM

Appendix 5: Funnel plot for ischaemic heart disease

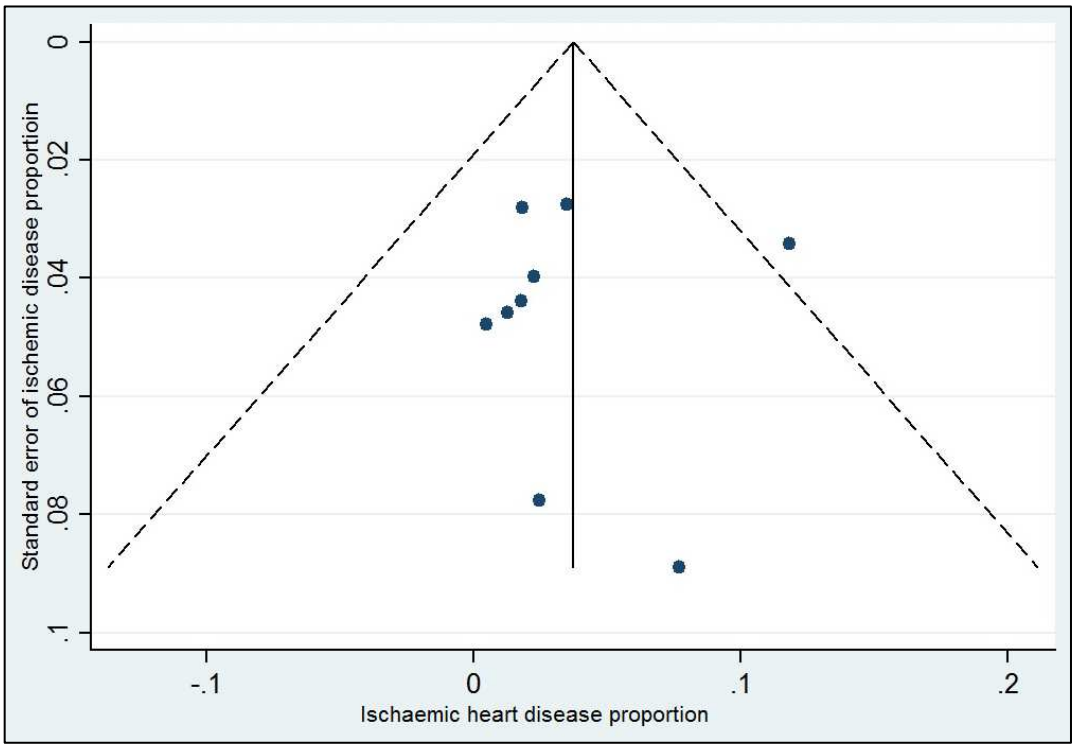


Figure 28: Funnel plot with pseudo 95% confidence limits for ischaemic heart disease

Appendix 6: Funnel plot for cerebrovascular disease

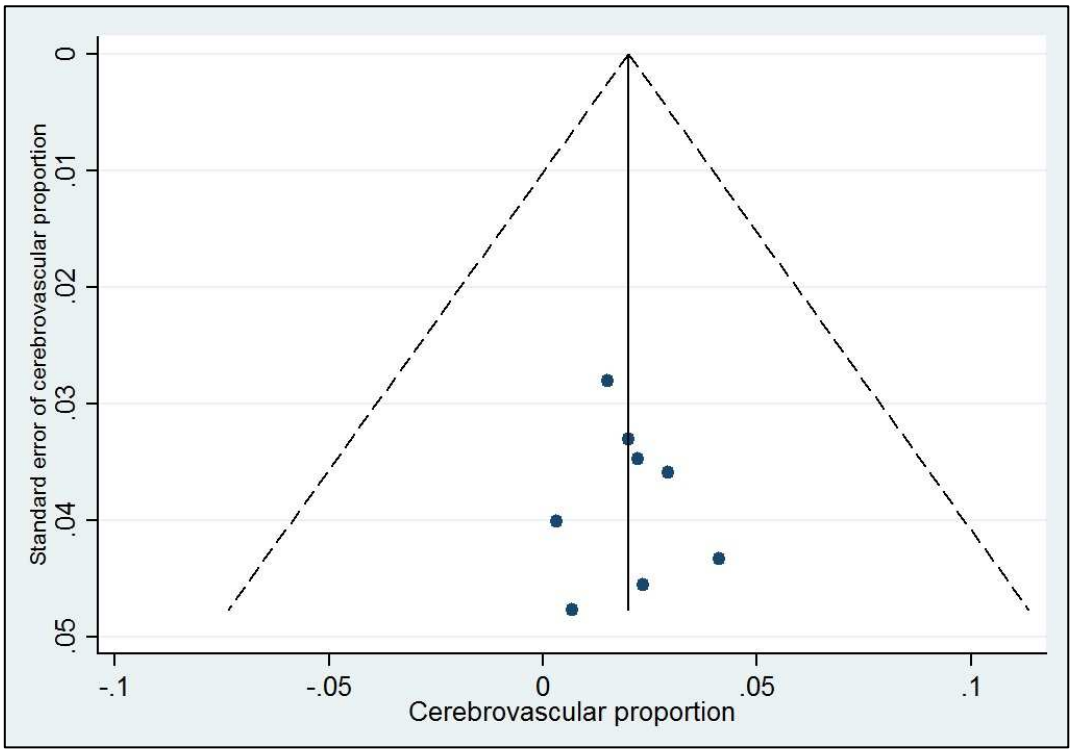


Figure 29: Funnel plot with pseudo 95% confidence limits for cerebrovascular disease

Appendix 7: Example easy-read invitation letter

< INSERT GP ADDRESS >

<Name>
<Address 1>
<Address 2>
<Address 3>
<Address 4>

Date

Initial Invitation Letter

Dear <PARTICIPANT NAME (DOB)>

STOP diabetes

Would you like to take part in the STOP Diabetes research study?

The study involves a Diabetes Health Check.

Do you want to learn more about the study? The information leaflet with this letter will tell you more about the study.

Yours sincerely

<INSERT GP SIGNATURE HERE >

<INSERT GP NAME>

Symbols taken from Change and Somerset Total Communication Through the Maze Widget and People First.

WP1_Easy read initial letter of invitation GP_V1_01.05.12.doc

Appendix 8: Full easy-read information sheet

Leicestershire Partnership NHS
NHS Trust

University of Leicester

Full Information Leaflet

STOP diabetes

STOP Diabetes Research Study

Please read this information leaflet.

Talk to a relative, carer or someone that you trust about it.

What is a research study?

A research study is a way we try to find out the answers to questions.
This research study is about diabetes.

You are invited to take part in the STOP Diabetes research study.

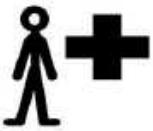
You will be a volunteer.
A volunteer is someone who helps with some work but does not get paid.

Who is doing this research study?

Professor Khunti

Professor Bhaumik

A research study team will help them:



- Nurse

- Secretary

- Academic

3

What is diabetes?

Diabetes is an illness.

People with diabetes have too much sugar (glucose) in their blood.
Their body cannot use sugar properly.

People with diabetes may feel

- Tired and ill

- Thirsty

- And need to go to the toilet a lot.

If you have diabetes you might get other health problems.

Why are we doing this research study?

We think some people with a learning disability might have diabetes.

We think some people with a learning disability could get diabetes in the future.

We want to find out if this is true.

We also want to find out the best way to stop (prevent) diabetes.

Why have you been invited?

It is important to keep people healthy.

Some people who have a learning disability find it hard to tell their doctor or nurse when they are poorly.

A Diabetes Health Check will help you find out if you have diabetes.

It is good to know if you have diabetes.

This means your doctor can give you the right treatment to make your health better.

Do I have to take part?

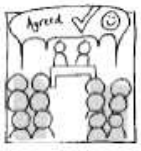
You can choose.

You can say yes

or no

If you say yes to take part in the research study

We will ask you to sign a piece of paper called a consent form.



This is a form that says you are happy to take part in the research study.

We will give you a copy of the consent form to keep.

It is okay to change your mind.

If you say no to taking part in the research study.

- Your rights will not change

- Your health care will not change

What will happen during the Diabetes Health Check?

If you say yes

You will be asked to come to 2 appointments.

At both appointments you will be in a small group.

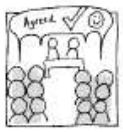
You can bring a parent, carer or friend with you.
It is good to bring somebody who knows you well.

What will happen during appointment 1?

This appointment will be for about 2 hours.

First we will explain about the research study.

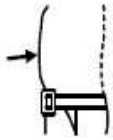
We will ask you to sign a piece of paper called a consent form.



This is a form that says you are happy to take part in the research study

We will ask you questions about your family and your life and your health.

We will measure how tall you are and how much you weigh.



We will also measure around your middle and your hips.

We will measure your blood pressure to see how well your heart is working.

We will ask you to come again for appointment 2.

We might ask you to fast.

Fasting means you are not allowed to eat or drink.

You can still have water to drink.

We will also give you some forms with questions on to take away with you and fill in.

Ask a relative, carer or someone you trust to help you with the questions and fill in the forms.

Please bring the forms you have filled in back to appointment 2.

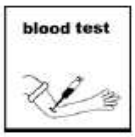
What will happen during appointment 2?

This appointment will last for about 1 hour.

We will ask if you have fasted from 12.00 midnight before appointment 2

At the appointment

We will test your urine (wee).



We will ask you to have a blood test.
The blood will be collected into bottles.

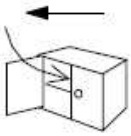
The blood test might hurt a little.

The nurses who will take your blood test are friendly.
They will look after you.

Some of the blood will be tested later that day.
This blood will be tested for diabetes and other health problems.

We will tell your GP what we find out.

If you agree.



1 bottle with your blood in will be kept (stored) before it is tested.

This blood will be tested for things called genetic markers. We want to see if some genetic markers make people have diabetes.

You can choose if you want to have this blood stored.

If you have any worries,

you can talk to the research study team.

After the Diabetes Health Check?

We will tell your GP what we find out.

We send you a letter to tell you what we have found out.

If we find out you have diabetes or think that you could get diabetes in the future.

We will ask you to go and see your GP.
Your GP can help you.

Other things you need to know

If your Diabetes Health Check shows that we think you could get diabetes in the future:

- We might invite you and a carer, friend or relative to talk to us.

- We might invite you to a small group. The group of people will learn how to stay healthy

If you say yes we can send you an invite.
We will choose some people to invite back.

After the STOP Diabetes research study

We would like to use information we have found out about you in the research study.

This could be in:

- Reports
- Presentations

We will not use your name.

If you say yes, we will send you a report about the STOP Diabetes research study when it is finished.

We would like to keep your contact details.

If you say yes, we might invite you to help with other research studies in the future.

Will I be paid for taking part?

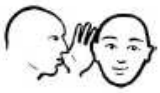
You will not be paid for taking part in the research study.

We will pay for your travel.

If you do not get free travel or mobility allowance

Will my information be kept private?

We will keep your information safe.



We will not tell anyone it is about you.

We will tell you what we find out.

We will tell your doctor what we find out.

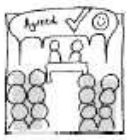
We will ask you if you will let us look at your medical notes to help us with this research study.
We will also ask if you will let us look at this information in the future.

Who is paying for this research study?

The government (the Department of Health) is paying the money to do this research study.

Who has checked this research study?

Before we do a research study, it is checked by a Research Ethics Committee.



A Research Ethics Committee is a group of people who make sure:

- It is ok to do the research study.
- The research study is being done in a good way

This research study has been checked by an ethics committee.

What should I do if I want to take part in the STOP Diabetes research study?

You can choose if you want to take part.

Please think about it.

It is a good idea to talk to a relative or someone that you trust.

Would you please fill out the reply form?

Please post it back to us in the envelope provided.

The envelope does not need a stamp.

Thank you

Who can I contact to find out more about the research study?

If you have any questions please contact:

STOP Diabetes Research Team
Leicester Diabetes Centre
Leicester General Hospital

You can phone on
0116 258 4251

Symbols taken from Change and Somerset Total Communication Through the Maze,Widget & People First.

Appendix 9: Full easy-read reply form

STOP diabetes

Leicestershire Partnership NHS NHS Trust

University of Leicester

Full Reply Form

STOP Diabetes Research Study

Please tick 1 of the boxes below.

☑ ☐ **Yes**. I want to take part in the STOP Diabetes research study.

☒ ☐ **No**. I do not want to take part in the STOP Diabetes research study.

Please tick ☑ this box if you have completed this form ☐

Please tick ☑ this box if your carer has completed this form ☐

1

WP1_Easy read full reply form_V1_01 06 12 - Copy.docx

My name is:

My address is:

My telephone number is:

Signed by you

Name of carer:

Relationship:

Address:

Telephone:

Are you happy to support this person in the research study?

☐ **Yes.** I am happy to provide my support.

☐ **No.** I do not want to provide my support.

Has the person decided to take part in the STOP Diabetes research study?

☐ **Yes** ☐ **No**

Or have you decided for them in their best interests?

☐ **Yes** ☐ **No**

Signature ☐

Thank you for filling in this reply form.

Please put this form in the envelope we have sent to you

The envelope does not need a stamp.
Please post this envelope back to us.
OR
Ask a parent or carer to post it back.

Appendix 10: Personal consultee information leaflet

PERSONAL CONSULTEE INFORMATION SHEET

STOP Diabetes

(Diabetes Screening Study for People with Learning Disabilities)

Introduction
We are inviting you to act as a 'personal consultee' for someone who is unable to make a decision for themselves. You are being asked to advise the researchers about this person's wishes and feelings as to whether they themselves would have wished to join this research. Before you decide, it is important for you to understand what it means to be a consultee. You also need to understand why the research is being done and what it will involve. Please take the time to read this information sheet carefully and talk to others about the study if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide if you wish to be a consultee.

What does it mean to be a personal consultee?
A consultee is someone who knows a person with a mental incapacity well and is willing and able to offer an opinion as to what the incapacitated person's wishes would have been did they not have a mental incapacity. We are intending to recruit participants to this project who may not have the capacity to consent to their participation. This means that they may not be able to judge for themselves whether they should like to take part or refuse. You do not have to act as a consultee if you do not want to. If you decide to act as a consultee, you will be asked to sign a Personal Consultee Advice Form.

The project has been approved by an NHS Research Ethics Committee. We shall make sure that the project is safe for each participant and does not cause them unnecessary distress. To help with this, the researchers need information from people who have known the participant for some time.

Why have I been approached?
As a partner, friend or relative of a (potential) participant in the study, you will have an interest in the person's well-being and welfare. You may have been given a Lasting Power of Attorney to make personal welfare decisions on their behalf when they can't. You may be a deputy appointed by the Court of Protection.

Researchers in the project would like to discuss with you whether you think that your partner, friend or relative would like to take part. As you have known them for some time, you may be aware of any views they may have about taking part in such a project or whether they have made an 'Advance Decision'. If your partner, friend or relative has made an 'Advance Decision' this is important as it shows that they have ready made decisions for themselves. The researchers would like to respect the person's wishes.

Secondly, if you think that your partner, friend or relative may be interested in taking part in the project, you may be able to tell us about any possible difficulties they may have. You also may be able to tell us how they may communicate that they wanted to stop being involved. When thinking about the wishes and interests of your partner, relative or friend, it is important that you should set aside any of your own personal views about the project.

What is the study about?
Diabetes is an illness. People with type-2 diabetes have too much sugar (glucose) in their blood. This happens because the body can't use sugar (glucose) properly. People with type-2 diabetes may feel tired, thirsty and need to go to the toilet a lot. Some people have no symptoms at all. Type-2 diabetes can lead to health problems such as heart disease, stroke, and kidney disease. Lots of people have type 2 diabetes for many years before they are diagnosed and some of these people may have already developed health problems.

Why should people be screened (checked) for type 2 diabetes?
It is important to keep people healthy. Some people with learning disabilities find it hard to tell their doctor or nurse when they are poorly. A diabetes health check is a good way to identify people who have type-2 diabetes. If a person is found to have diabetes they can be given treatment to stop them feeling poorly and to help keep them healthy.

We can also identify people who may be at 'high risk' of developing diabetes in the future. This is called pre-diabetes. If people with pre-diabetes eat healthier foods and do more exercise this may help them not to get diabetes.

Why is this research being done?
Many people with learning disabilities have more health problems than the general population. We also know that health problems are more likely to go undiagnosed and untreated in people with learning disabilities.

We think people with learning disabilities may be more likely to get type-2 diabetes than some other people. We would like to find out if this is true and what the risk factors (things that make something more likely) are for type 2 diabetes in people with learning disabilities.

Does the individual have to take part?
No! Individuals can choose if they want to say yes and volunteer to take part or if they want to say no. Where individuals with a learning disability are unable to decide, we are asking advice from other people. If you volunteer to give advice, we will record your advice on a Personal Consultee Advice Form. You will be given a copy of your signed form for you to keep. Even if you say yes now, you (or the individual) can change your mind and stop at any time. The decision you both make will not affect the quality of the health care the individual receives.

What will be involved?
If you agree to help with the research, you will both be asked to attend 2 appointments for this study. The appointments will be held at a location that is convenient to yourself (e.g. day centre, general practice, community venue). You will both have the chance to meet the members of our team and to ask any questions you both might have.

Appointment 1
We expect that appointment 1 will last about 2 hours. During the appointment we would like to take some measurements (blood pressure, height, weight, waist, hips). We will also ask some questions about general health; such as any illnesses the person with learning disabilities has and any medications (tablets) they take. We will also give you some questionnaires to take away with you and bring back to the second appointment.

Appointment 2
We expect that appointment 2 will last about 1 hour. During the appointment we would like to test the person with learning disabilities urine (wee) and take a blood test. The blood will be taken from their arm using a needle and syringe. The blood will be tested for diabetes. We will check their liver and kidney function and measure how much cholesterol there is in the blood. We would also like to take some extra blood (optional) to be stored and tested at a later date. The total amount of blood we would like to take is about 10mls (2 teaspoonfuls). If

you agree that extra blood can be taken to be stored and tested in the future this will be about 20mls (4 teaspoonfuls).

On the evening before the second appointment, if appropriate, we may ask the person with learning disabilities to fast. This means having NO food or drink after 12 midnight and NO breakfast. They are allowed to have water to drink.

If you have any worries or questions about what will happen at the appointments please contact a member of the team to discuss how we can make it easier for you both to take part in the study. You can take a break at any time. If you prefer, some people may be allowed to have all the tests and measurements done together in 1 appointment but we would prefer you both to have 2 appointments.

Will I be asked to do anything else?
When you sign the Personal Consultee Advice Form there are some extra things we will ask you to think about. If the person with learning disabilities is found to be at high risk of getting diabetes we may want to invite you to take part in the next stage of our research. This is the development of a lifestyle education programme for people with learning disabilities. We are only asking if we can send you both the extra information, it does not mean you have to take part.

We would also like to take an extra sample of blood that will be stored for genetic analysis at the end of the study. You can both still take part in the diabetes screening study without agreeing to these extra things.

Might anything about the research be upsetting?
Individuals may suffer slight discomfort while the blood samples are being taken from their arm and some people do experience bruising after blood samples have been taken.

What are the possible benefits of taking part?
We might find out that the individual has diabetes or is at high risk of getting diabetes. This will mean that they can be given the right treatment by their GP (doctor). This may help prevent future health problems (keep them healthy).

The study will also help us understand if some people with learning disabilities are more likely to get diabetes and this may help us develop ways to prevent diabetes.

Will the persons GP be informed of the results?
Yes, their GP (doctor) will be informed of all the results of the tests and measurements taken as part of this study.

Will people be paid for taking part in the study?
If one or both of you have to pay for travel, we will pay you the money back up to the value of £15. Please keep all your receipts.

Will information that is given be kept confidential (private)?
Yes. All information that is collected from you both during the research study will be kept confidential (private). We will also ask your advice for permission to look at the persons medical records for information related to the research study (essential), and long-term follow-up (optional). Any information about them which leaves the hospital/surgery will have their name and address removed so that they cannot be recognised from it.

Some of the data collected as part of the study may be looked at by authorised people to check that the study is being carried out correctly.

What will happen to the results of the research study?

The results of the study may be published in a medical journal or presented at research seminars or conferences. People taking part will not be identified in any report or publication.

Who is doing this research?
The people who are doing the research study are:
Professor Khunti at the University of Leicester, and Dr Bhaumik at Leicestershire Partnership Trust.

Who is funding this research?
The funding (money) to do the research study has come from the Department of Health.

Who has reviewed this study?
Before a research study goes ahead it has to be checked by a group of people called a Research Ethics Committee. These people make sure that the research is ok to do. This study has been checked by an NHS Ethics Committee.

What do I have to do now?
If you think that your partner, friend or relative would be interested in taking part, please complete the reply form and return to the Diabetes Research Team. We have given you a pre-paid envelope with our address. The envelope does not need a stamp. The Diabetes Research Team will then contact you to arrange a time for a discussion.

If you are unsure or do not want to advise the researcher then please suggest who else we could ask. If you think that your friend, partner or relative would **NOT** be interested in taking part, please still complete and return the form.

If you would like to ask any questions before deciding, please feel free to contact the STOP Diabetes Research Team:

Prof Kamlesh Khunti
Professor of Primary Care Diabetes & Vascular Medicine
University of Leicester

Prof Sabyasachi Bhaumik
Medical Director,
Leicestershire Partnership Trust

Tel: 0116 258 4251

E-mail: stopdiabetes@le.ac.uk

Or you can write to us:
STOP Diabetes Research Team
Leicester Diabetes Centre
Leicester General Hospital
LE5 4PW

Appendix 11: Nominated consultee information leaflet

NOMINATED CONSULTEE INFORMATION SHEET

STOP Diabetes study

(Diabetes Screening Study for People with Learning Disabilities)

Introduction
You are being invited to act as a 'nominated consultee' for someone who is unable to make a decision for themselves. You are being asked to advise the researcher about this person's wishes and feelings as to whether they themselves would have wished to join this research. Before you decide, it is important for you to understand what it means to be a consultee, as well as why the research is being done and what it will involve. Please take time to read this information sheet carefully and talk to others about the study if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide if you wish to be a consultee.

What does it mean to be a nominated consultee?
A 'nominated consultee' is someone who knows a person with a mental incapacity well and is willing and able to offer an opinion as to what the incapacitated person's wishes would have been did they not have a mental incapacity. We are intending to recruit participants to this project who may not have the capacity to consent to their participation. This means that they may not be able to judge for themselves whether they should like to take part or refuse. You do not have to act as a consultee if you do not want to. If you decide to act as a consultee, you will be asked to sign a Nominated Consultee Advice Form.

The project has been approved by an NHS Research Ethics Committee. We shall make sure that the project is safe for each participant and does not cause them unnecessary distress. To help with this, the researchers need information from people who have known the participant for some time.

Why have I been approached?
You may be someone who already knows the prospective participant, working with them as a paid carer or in a professional capacity, such as a doctor or a solicitor. Alternatively, you may already have been approached by a Leicestershire Partnership Trust and agreed to act as a Nominated Consultee.

Researchers in the project would like to discuss with you whether you think that the prospective participant would like to take part. If you have known them for some time, you may be aware of any views they may have about taking part in such a project or whether they have made an 'Advance Decision'. If the potential participant has made an 'Advance Decision' this is important as it shows that they have ready made decisions for themselves. The researchers would like to respect the person's wishes.

Secondly, if you think the potential participant may be interested in taking part in the project, you may be able to tell us about any possible difficulties they may have. You also may be able to tell us how they may communicate that they wanted to stop being involved. When thinking about the wishes and interests of the prospective participant, it is important that you should set aside any of your own personal views about the project.

What is the study about?
Diabetes is an illness. People with type-2 diabetes have too much sugar (glucose) in their blood. This happens because the body can't use sugar (glucose) properly. People with type-2 diabetes may feel tired, thirsty and need to go to the toilet a lot. Some people have no symptoms at all. Type-2 diabetes can lead to health problems such as heart disease, stroke, and kidney disease. Lots of people have type 2 diabetes for many years before they are diagnosed and some of these people may have already developed health problems.

Why should people be screened for type 2 diabetes?
It is important to keep people healthy. Some people with learning disabilities find it hard to tell their doctor or nurse when they are poorly. A diabetes health check is a good way to identify people who have type-2 diabetes. If a person is found to have diabetes they can be given treatment to stop them feeling poorly and to help keep them healthy.

We can also identify people who may be at 'high risk' of developing diabetes in the future. This is called pre-diabetes. If people with pre-diabetes eat healthier foods and do more exercise this may help them not to get diabetes.

Why is this research being done?
Many people with learning disabilities have more health problems than the general population. We also know that health problems are more likely to go undiagnosed and untreated in people with learning disabilities.

We think people with learning disabilities may be more likely to get type-2 diabetes than some other people. We would like to find out if this is true and what the risk factors (things that make something more likely) are for type 2 diabetes in people with learning disabilities.

Does the individual have to take part?
No! Individuals can choose if they want to say yes and volunteer to take part or if they want to say no. Where individuals with a learning disability are unable to decide, we are asking advice from other people. If you volunteer to give advice, we will record your advice on a Nominated Consultee Advice Form. You will be given a copy of your signed form for you to keep. Even if you say yes now, you (or the individual) can change your mind and stop at any time. The decision you both make will not affect the quality of the health care the individual receives.

What will be involved?
If you agree to help with the research, you will both be asked to attend 2 appointments for this study. The appointments will be held at a location that is convenient to yourself (e.g. day centre, general practice, community venue). You will both have the chance to meet the members of our team and to ask any questions you both might have.

<u>Appointment 1</u>
We expect that appointment 1 will last about 2 hours. During the appointment we would like to take some measurements (blood pressure, height, weight, waist, hips). We will also ask some questions about general health; such as any illnesses the person with learning disabilities has and any medications (tablets) they take. We will also give you some questionnaires to take away with you and bring back to the second appointment.

<u>Appointment 2</u>
We expect that appointment 2 will last about 1 hour. During the appointment we would like to test the person with learning disabilities urine (wee) and take a blood test. The blood will be taken from their arm using a needle and syringe. The blood will be tested for diabetes. We will check their liver and kidney function and measure how much cholesterol there is in the blood. We would also like to take some extra blood (optional) to be stored and tested at a

later date. The total amount of blood we would like to take is about 10mls (2 teaspoonfuls). If you agree that extra blood can be taken to be stored and tested in the future this will be about 20mls (4 teaspoonfuls).

On the evening before the second appointment, if appropriate, we may ask the person with learning disabilities to fast. This means having NO food or drink after 12 midnight and NO breakfast. They are allowed to have water to drink.

If you have any worries or questions about what will happen at the appointments please contact a member of the team to discuss how we can make it easier for you both to take part in the study. You can take a break at any time. If you prefer, some people may be allowed to have all the tests and measurements done together in 1 appointment but we would prefer you both to have 2 appointments.

Will I be asked to do anything else?
When you sign the Nominated Consultee Advice Form there are some extra things we will ask you to think about. If the person with learning disabilities is found to be at high risk of getting diabetes we may want to invite you to take part in the next stage of our research. This is the development of a lifestyle education programme for people with learning disabilities. We are only asking if we can send you both the extra information, it does not mean you have to take part.

We would also like to take an extra sample of blood that will be stored for genetic analysis at the end of the study. You can both still take part in the diabetes screening study without agreeing to these extra things.

Might anything about the research be upsetting?
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What are the possible benefits of taking part?
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The study will also help us understand if some people with learning disabilities are more likely to get diabetes and this may help us develop ways to prevent diabetes

Will the persons GP be informed of the results?
Yes, their GP (doctor) will be informed of all the results of the tests and measurements taken as part of this study.

Will people be paid for taking part in the study?
If one or both of you have to pay for travel, we will pay you the money back up to the value of £15. Please keep all your receipts.

Will information that is given be kept confidential (private)?
Yes. All information that is collected from you both during the research study will be kept confidential (private). We will also ask your advice for permission to look at the persons medical records for information related to the research study (essential), and long-term follow-up (optional). Any information about them which leaves the hospital/surgery will have their name and address removed so that they cannot be recognised from it.

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Before a research study goes ahead it has to be checked by a group of people called a Research Ethics Committee. These people make sure that the research is ok to do. This study has been checked by an NHS Ethics Committee.

What do I have to do now?
If you think that the prospective participant would be interested in taking part, please complete the reply form and return to the Diabetes Research Team. We have given you a pre-paid envelope with our address. The envelope does not need a stamp. The Diabetes Research Team will then contact you to arrange a time for a discussion.

If you are unsure or do not want to advise the researcher then please suggest who else we could ask. If you think that the prospective participant would **NOT** be interested in taking part, please still complete and return the form.

If you would like to ask any questions before deciding, please feel free to contact the STOP Diabetes Research Team:

Prof Kamlesh Khunti
Professor of Primary Care Diabetes & Vascular Medicine
University of Leicester

Prof Sabyasachi Bhaumik
Medical Director,
Leicestershire Partnership Trust

Tel: 0116 258 4251

E-mail: stopdiabetes@le.ac.uk

Or you can write to us:
STOP Diabetes Research Team
Leicester Diabetes Centre
Leicester General Hospital
LE5 4PW

Appendix 12: Easy-read consent form

STOP diabetes | Leicestershire Partnership NHS Trust | University of Leicester

Consent Form
(Version 2, 1st September 2012)

STOP Diabetes Research Study

<INSERT STUDY ID NO>

This form is a way for us to make sure that you don't mind taking part in the STOP Diabetes Research study

You will need to put a tick in each box.

1. The research team has explained the STOP Diabetes research study to me. **Yes** 1

2. I understand that I will have a Diabetes Health Check. **Yes** 2

3. I have asked all the questions I want. **Yes** 3

4. My questions were answered. **Yes** 4

5. It is my choice to take part.

Yes **5**

6. I understand it's okay to say no.
Saying no won't affect my rights, services or support.

Yes **6**

7. If I say yes, I know I can still change my mind later on.

Yes **7**

8. I understand that my doctor will be told I am taking part. My doctor will also be told if I have diabetes or not.

Yes **8**

9. I understand that my information will be kept private.
My information will only be seen by:
- The research study team and
- People who check the research study is being done properly.

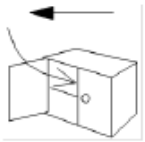
Yes **9**

10. The research team can use information about me:
- In reports about the STOP Diabetes research study
- In presentations about STOP Diabetes research study

Yes **10**

11. The research team can look at medical information about me that is kept at my doctor's surgery, home or day centre.

Yes **11**



12. I agree for some of my blood to be kept and tested for things called genetic markers when the research study ends.

You can choose to say yes or no

Yes 12

No 12

13. I would like to take part in the STOP Diabetes research study and have a Diabetes Health Check.

Yes 13

You need to sign this part of the form. When this part is signed it means you have said yes to taking part in the STOP Diabetes research study and having a Diabetes Health Check.

My name is

Am happy with taking part in the STOP Diabetes research study and have a Diabetes Health Check.

Today's date is:

Signed (or mark) by you

Name of researcher (PRINT NAME)

Signed by researcher

Date:

**Please Note: If participant is unable to sign their name, this should be witnessed.*

Name of witness (PRINT NAME)

Signed by witness

Date:

Appendix 13: Personal consultee advice form

PERSONAL CONSULTEE ADVICE FORM
(Version 1, 1st June, 2012) INSERT STUDY ID:

Please give your opinion on what the past and present wishes and feelings the person who lacks capacity would have been about taking part in the above study.

Please initial in each box

Please note that as a personal consultee you must not be connected to the above research project, or be under any influence by a member of the research team.

1. I confirm that I have read and understand the information sheet (Version 1, dated: 01/06/2012) for the STOP Diabetes study, and understand what it means to be a personal consultee. I have had the opportunity to consider the information, ask questions, and have had these answered to my liking.
2. I understand that I am free to change my opinion on what the participant would have wished for and felt about this study at any time, without the participant's care or rights being affected.
3. In my opinion, the participant would have provided consent for their GP (doctor) to be informed of their participation in the STOP Diabetes study and be sent copies of their biomedical results collected as part of this study.
4. In my opinion, the participant would have provided consent for relevant sections of their medical notes and/or data collected during the study to be looked at by individuals from the study team, the sponsor, the NHS Trust or from regulatory authorities where it is relevant to their taking part in this research.
5. In my opinion, the participant would have provided consent for researchers from the STOP Diabetes study to have access to their NHS medical records for additional data collection that is relevant to this current research study.
6. In my opinion, the participant would have provided consent for researchers from the STOP Diabetes study to have access to any health records held on them by their residential home, day centre or other care establishment, for additional data collection that is relevant to this current research study.
7. I understand that any information collected during the study may be used in future reports, articles or presentations by the research team and that names will not appear anywhere.
8. In my opinion, the participant would like to take part in the STOP Diabetes study.

---

9. In my opinion, the participant would consent for an extra blood sample to be taken for genetic testing. I understand that the blood will be stored and tested at the end of the study. **(optional)**
10. In my opinion, the participant would consent to being approached with information about the next stage of the STOP Diabetes study if they are found to be at high risk of developing diabetes. I understand that this involves the development and testing of a lifestyle education programme. **(optional)**
11. In my opinion, the participant would give permission for the Diabetes Research Team to have access to their NHS medical records for long-term follow-up data collection in the future. **(optional)**
12. In my opinion, the participant would like to receive a summary of the results of the study and agree to them being posted to the address on the participant pack. **(optional)**
13. In my opinion, the participant would give permission for retention of their contact details for contact at a later stage for invitation to participate in follow-up or related studies. **(optional)**

Name of research participant .....

Relationship to participant .....

Name of Consultee ..... **Signature**..... **Date** .....

Name of Researcher ..... **Signature**..... **Date** .....

(3 copies; 1 researcher, 1 general practice, 1 personal consultee)

WP1_Personal Consultee advice form v1_01.06.12.docx

Appendix 14: Nominated consultee advice form

STOP diabetes

NOMINATED CONSULTEE ADVICE FORM
(Version 1, 1st June, 2012) INSERT STUDY ID:

Please give your opinion on what the past and present wishes and feelings the person who lacks capacity would have been about taking part in the above study.

Please initial in each box

Please note that as a nominated consultee you must not be connected to the above research project, or be under any influence by a member of the research team.

☐ 1. I confirm that I have read and understand the information sheet (Version 1, dated: 01/06/2012) for the STOP Diabetes study, and understand what it means to be a nominated consultee. I have had the opportunity to consider the information, ask questions, and have had these answered to my liking.

☐ 2. I understand that I am free to change my opinion on what the participant would have wished for and felt about this study at any time, without the participant's care or rights being affected.

☐ 3. In my opinion, the participant would have provided consent for their GP (doctor) to be informed of their participation in the STOP Diabetes study and be sent copies of their biomedical results collected as part of this study.

☐ 4. In my opinion, the participant would have provided consent for relevant sections of their medical notes and/or data collected during the study to be looked at by individuals from the study team, the sponsor, the NHS Trust or from regulatory authorities where it is relevant to their taking part in this research.

☐ 5. In my opinion, the participant would have provided consent for researchers from the STOP Diabetes study to have access to their NHS medical records for additional data collection that is relevant to this current research study.

☐ 6. In my opinion, the participant would have provided consent for researchers from the STOP Diabetes study to have access to any health records held on them by their residential home, day centre or other care establishment, for additional data collection that is relevant to this current research study.

☐ 7. I understand that any information collected during the study may be used in future reports, articles or presentations by the research team and that names will not appear anywhere.

☐ 8. In my opinion, the participant would like to take part in the STOP Diabetes study.

☐ 9. In my opinion, the participant would consent for an extra blood sample to be taken for genetic testing. I understand that the blood will be stored and tested at the end of the study. **(optional)**

☐ 10. In my opinion, the participant would consent to being approached with information about the next stage of the STOP Diabetes study if they are found to be at high risk of developing diabetes. I understand that this involves the development and testing of a lifestyle education programme. **(optional)**

☐ 11. In my opinion, the participant would give permission for the Diabetes Research Team to have access to their NHS medical records for long-term follow-up data collection in the future. **(optional)**

☐ 12. In my opinion, the participant would like to receive a summary of the results of the study and agree to them being posted to the address on the participant pack. **(optional)**

☐ 13. In my opinion, the participant would give permission for retention of their contact details for contact at a later stage for invitation to participate in follow-up or related studies. **(optional)**

Name of research participant .....

Relationship to participant .....

Name of Consultee ..... **Signature**..... **Date** .....

Name of Researcher ..... **Signature**..... **Date** .....

(3 copies; 1 researcher, 1 general practice, 1 nominated consultee)

WP1_Nominated Consultee advice form v1_01.06.12.docx

Appendix 15: Example of letter to inform participants of results

Leicestershire Partnership NHS Trust

STOP diabetes

University of Leicester

Leicester Diabetes Centre
Leicester General Hospital
Leicester
LE5 4PW

«Patient_Name»
«Patient_Street_Name»
«Patient_Town_Village»
«Patient_City_County»
«Patient_Postcode»

«study_ID»

Date: 06 October 2015

Results Letter
STOP Diabetes Research Study

Dear «Patient_Name» - «Patient_DOB»

Thank you for taking part in the STOP Diabetes research study.

We said we would let you know what we found out at your Diabetes Health Check.

You had a blood test at the Diabetes Health Check.

The blood test told us that your blood sugar was normal.

This means you do not have diabetes.

WP1_Easy read results letter Normal_V1_01 06 12.docx

Appendix 16: Example letter to inform general practice of results

Leicestershire Partnership NHS Trust

STOP diabetes

University of Leicester

STOP Diabetes Research Team
Leicester Diabetes Centre
Leicester General Hospital
LE5 4PW

Date;06/10/15

«GP_Name»
«Practice_Name»
«GP_Street_Name»
«GP_Town_Village»
«GP_CityCounty»
«GP_Postcode»

Dear **«GP_Name»** Study ID N[2];**«study_ID»**

Patient: **«Patient_Name», «Patient_Street_Name», «Patient_Town_Village», «Patient_City_County», «Patient_Postcode»**
Date of birth: **«Patient_DOB»**

The above named patient is participating in the STOP Diabetes study and attended a screening appointment on **«Date_of_Appointment»**.The results of the diabetes test and general health screen are listed below:

Measurements		**Blood glucose**	
Height (m)	**«Height_m»**	Fasting plasma	**«Fasting_glucose_mmoll»**
Weight (kg)	**«Weight_kg»**	Random plasma	**«Random_glucose_mmoll»**
BMI (kg/m^2)	**«BMI_kgm2»**	HbA1c (%)	**«HbA1c_»**
Waist	**«Waist_cm»**	HbA1c (mmol/l)	**«Hba1c_mmoll»**
Hip circumference	**«Hip_cm»**	**Lipids**	
Systolic BP	**«Systolic_BP_mmHg»**	Total cholesterol	**«Total_Chol»**
Diastolic BP	**«Diastolic_BP_mmHg»**	LDL cholesterol	**«LDL_Chol»**
Urea &		HDL cholesterol	**«HDL_Chol»**
Sodium (mmol/l)	**«Sodium»**	Triglycerides	**«Trilycerides»**
Potassium (mmol/l)	**«Potassium»**	**Liver function**	
Urea (mmol/l)	**«Urea»**	Alanine	**«ALT»**
Creatinine (mmol/l)	**«Creatinine»**	Alkaline	**«ALP»**
eGFR (ml/min)	**«eGFR»**	Total bilirubin	**«Billirubin»**
Urine		Gamma-GT (iu/L)	**«GGT»**
Albumin-creatinine	**«Urine_ACR»**	**Thyroid function**	
Smoking status		TSH	**«TSH»**
Current Smoker:	**«Current_Smoker»**	Free T4	**«Free_T4»**

We would like to advise you that the results of these tests indicate that they **do NOT** have diabetes.

Your patient will not be called back for any further tests by the research team. If you feel you need any further information from us please do not hesitate to contact us on: 0116 258 4251.

Yours sincerely

Prof Kamlesh Khunti
(Professor Primary Care Diabetes & Vascular Medicine)
Principal Investigator

Appendix 17: Questionnaires used in the research programme

EQ-5D: a generic instrument for the measurement of health related quality of life.[213] It provides a simple descriptive profile in five dimensions (mobility, self-care, usual activities, pain/discomfort and anxiety/depression), each with three levels. This instrument can be used in the clinical and economic evaluation of health care and can be used to analyse changes in the health status of individuals or groups of individuals over time.

Aberrant Behaviour Checklist: an informant-based problem behaviour rating scale which assesses a wide range of behavioural disorders and has been shown to be a reliable and valid behaviour rating instrument.[215] The questionnaire consists of 58 items, scored on a 4-point scale.[214] The sub-categories are: (i) irritability, agitation, crying; (ii) lethargy, social withdrawal; (iii) stereotypic behaviour; (iv) hyperactivity, noncompliance; and (v) inappropriate speech.

PAS-ADD Checklist: a 25-item questionnaire and can be used to make an initial assessment for mental illness/psychiatric disorders in people with ID.[216] The instrument generates threshold scores which are then used as a measure to indicate the likely absence or presence of possible psychiatric problems. The scores produced relate to: i) affective or neurotic disorder; ii) possible organic condition (including dementia); iii) psychotic disorder.

Glasgow Depression Scale: an established measure of depression among people with ID.[217] The Glasgow Depression Scale for people with learning disability (GDS-LD) differentiates depression and non-depression groups, correlates with the Beck Depression Inventory II (r=0.88), has good test-retest reliability (r=0.97) and internal consistency (Cronbach's a=0.90), and a cut-off score of 13 yielded 96% sensitivity and 90% specificity. The Carer Supplement is also reliable (r=0.98; a=0.88), correlating with the GDS-LD (r=0.93).

Appendix 18: Summary of baseline characteristics

Table 65: Baseline characteristics of participants in the screening study

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Biomedical Measurements		
Plasma glucose		
Fasting (mmol/l)	425 (mr: 8)	4.7 (± 0.7)
Non-fasting (mmol/l)	239 (mr: 16)	5.3 (± 1.5)
Glycated haemoglobin	675 (mr: 27)	
HbA1c (mmol/mol)		35.0 (± 5.1)
Derived HbA1c (%)		5.4 (± 0.5)
Lipids		
Total cholesterol (mmol/l)	653	4.9 (± 1.0)
HDL Cholesterol (mmol/l)	644	1.3 (± 0.4)
LDL Cholesterol (mmol/l)	631	2.9 (± 0.9)
Triglycerides (only if fasted) (mmol/l)	407	1.4 (± 0.9)
Urea and electrolytes		
Sodium (mmol/l)	713 (mr: 84)	139.6 (± 3.1)
Potassium (mmol/l)	701 (mr: 80)	4.3 (± 0.5)
Urea (mmol/l)	712 (mr: 83)	5.4 (± 1.9)
Creatinine (mmol/l)	714 (mr: 84)	69.0 (± 22.7)
eGFR (mL/min), n (%)	603 (mr: 80)	
≥90		476 (78.9)
60-89		110 (18.2)
45-59		10 (1.7)
30-44		4 (0.7)
≤29		3 (0.5)
Liver function tests		
Bilirubin (umol/l)	683 (mr: 52)	9.6 (± 5.9)
Alanine transaminase (iu/l)	691 (mr: 61)	24.8 (± 15.8)
Alkaline phosphatise (iu/l)	694 (mr: 67)	86.8 (± 27.6)
Gamma GT (iu/l)	621 (mr: 3)	32.5 (± 32.2)
Thyroid function		
Thyroid stimulating hormone (mui/l)	637 (mr: 22)	2.6 (± 2.1)
Free thyroxine (T4) (pmol/l)	621 (mr: 10)	14.0 (± 2.4)

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Urine ACR		
Urine albumin creatinine ratio (mg/mmol)	569 (mr: 1)	2.5 (± 12.5)
Anthropometric Measurements		
Height (m)	800	1.6 (± 0.1)
Weight (kg)	799	76.4 (± 20.7)
BMI (kg/m2)	782	28.7 (± 7.1)
BMI Categories, n (%)		
Underweight		30 (3.8)
Normal		223 (28.5)
Overweight		241 (30.8)
Obese		288 (36.8)
Waist circumference (cm)	796	100.4 (± 16.5)
Hip circumference (cm)	789	107.6 (± 14.0)
Blood Pressure Measurements	826	
Systolic (mmHg)		121.4 (± 16.9)
Diastolic (mmHg)		78.2 (± 11.1)
Demographic and Lifestyle		
Age (years)	930	43.3 (± 14.2)
Sex, Male, n (%)	930	537 (57.7)
Ethnicity, n (%)	930	748 (80.4)
White		147 (15.8)
Asian		14 (1.5)
Black		
Mixed		13 (1.4)
Other		8 (0.9)
Residential circumstances, n (%)	929	
Alone		51 (5.5)
Lives with family		338 (36.4)
Shared house or supported living		157 (16.9)
Shared care		16 (1.7)
Residential home or nursing home		350 (37.7)
Other		17 (1.8)
Level of Support, n (%)	929	
Independent		69 (7.4)
Some Support		205 (22.1)
24 hour support		655 (70.5)

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Current status*, n (%)		
Paid employment	928	71 (7.7)
Voluntary work	927	152 (16.4)
College	925	170 (18.4)
Day opportunities or private day centre	928	431 (46.4)
Shared lives (day placement)	928	19 (2.1)
Attending meetings	926	122 (13.2)
Other	924	385 (41.7)
Deprivation (IMD 2015)**, Median (IQR)	930	16,353 (7351-23,606)
Medical History		
Severity of ID, n (%)	865	
Not known		49 (5.7)
Known		816 (84.3)
Mild		260 (30.1)
Moderate		244 (28.2)
Severe		279 (32.3)
Profound		33 (3.8)
Cause of ID, n (%)	866	
Not known		581 (67.1)
Known		285 (32.9)
Downs syndrome		133 (15.4)
Fragile X		8 (0.9)
Cerebral palsy		58 (6.7)
Angelman syndrome		4 (0.5)
Cytomegalovirus		1 (0.1)
Foetal Alcohol syndrome		0
Homocystinuria		0
Hydrocephalus		6 (0.7)
Hurler syndrome		0
Klinefelter's syndrome		3 (0.4)
Lesch – Nyan syndrome		0
Neurofibromatosis		2 (0.2)
Phenylketonuria		5 (0.6)
Prader – Willi syndrome		4 (0.5)
Rett syndrome		1 (0.1)
Sturge – Weber syndrome		1 (0.1)
Tay – Sachs disease		0
Triple X syndrome		0
Trisomy 13		0
Trisomy 18		0

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Tuberous sclerosis		2 (0.2)
Turner syndrome		0
Other cause		57 (6.6)
Medical or Health problems, n (%)	929	
None		117 (12.6)
Yes		812 (87.4)
Physical Health		13 (1.4)
Stroke		0
Peripheral arterial disease		
Coronary heart disease		7 (0.8)
Congenital heart disease		19 (2.1)
Other heart problems		15 (1.6)
High blood pressure		63 (6.8)
High cholesterol		62 (6.7)
Hypothyroidism		93 (10.0)
Polycystic ovary syndrome		1 (0.1)
Gestational diabetes		0
Pre-diabetes		1 (0.1)
Chronic breathing problems		88 (9.5)
Sleep apnoea		3 (0.3)
Epilepsy		262 (28.2)
Mental Health		
Dementia		18 (1.9)
Schizophrenia, schizotypal and delusional		35 (3.8)
Mood (affective) disorders		152 (16.4)
Neurotic, stress-related and somatoform		143 (15.4)
≥2 disorders		52 (5.6)
Personality disorders		13 (1.4)
Drug / alcohol problems		0
Attention Deficit Hyperactivity Disorder		8 (0.9)
Intellectual Disability		
Autistic spectrum disorders		165 (17.8)
Behavioural problems		128 (13.8)
Current medication, n (%)	928	
None		172 (18.5)
Yes		756 (81.5)
Anti-psychotic		240 (25.9)
≥2 medication		24 (2.6)
Depression/Anxiety/OCD or related		258 (27.8)
≥2 medication		43 (4.6)
For ADHD		4 (0.4)
Anti-epileptic		311 (33.5)
Anti-thrombotic		36 (3.9)

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Lipid lowering		74 (8.0)
Statin		72 (7.8)
Fibrate		1 (0.1)
Statin and Fibrate		1 (0.1)
Anti-hypertensive		85 (9.2)
Thyroid medication		93 (10.0)
Steroids		80 (8.6)
Oral		5 (0.5)
Inhaled		62 (6.7)
Topical		9 (1.0)
More than 1 type of steroid medication		3 (0.3)
Not known		1 (0.1)
Anti-obesity		1 (0.1)
Other		571 (61.5)
Smoking status, n (%)	929	
Current		76 (8.2)
Ex		38 (4.1)
Never		815 (87.7)
Family history of diabetes, n (%)	592	180 (30.4)
Physical Activity / Exercise		
Able to stand, n (%)	929	
No		58 (6.2)
Yes		871 (93.8)
Able to walk, n (%)	927	
No		57 (6.2)
Yes (with or without walking stick, aid)		787 (84.9)
Yes, with assistance from person(s)		83 (9.0)
Mobility aids, n (%)	928	
No		703 (75.8)
Yes		225 (24.3)
Uses a walking aid		52 (5.6)
Uses a wheelchair, all or most		81 (8.7)
Uses a wheelchair, some		78 (8.4)
Other		12 (1.3)
Not known		2 (0.2)
Amount of walking per day, n (%)	927	
None		74 (8.0)
A short distance		259 (27.9)
Some		359 (38.7)

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Lots		235 (25.4)
Speed of normal walking (if can walk), n (%)	850	
Slow		301 (35.4)
Steady		373 (43.9)
Brisk or fast		176 (20.7)
Activities*, n (%)		
Keep fit/aerobics	928	83 (8.9)
Walking		197 (21.2)
Running/jogging	929	39 (4.2)
Swimming		190 (20.5)
Dance		233 (25.1)
Bowling		155 (16.7)
Gym		92 (9.9)
Horse riding		32 (3.4)
Cycling		62 (6.7)
Gardening		179 (19.3)
Housework	927	489 (52.8)
Chair based exercise	863	68 (7.9)
Other	925	131 (14.2)
Amount of physical activity per week, n (%)	928	
None		184 (19.8)
1-2 times		360 (38.8)
3-4 times		259 (27.9)
5 or more		125 (13.5)
Time spent sitting per day, n (%)	928	
All / most		180 (19.4)
A lot		252 (27.2)
Sometimes		475 (51.2)
Never		21 (2.3)
Nutrition and diet		
Problems relating to eating and drinking, n (%)		
Difficulties with chewing or swallowing	929	227 (24.4)
Needs help or assistance to feed self	926	118 (12.7)
Use specialist equipment		95 (10.3)
Fed via an ng-tube or a gastrostomy		7 (0.8)

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Only included if not fed via tube	922	
Food shopping, n (%)		
Independently		89 (9.7)
With support		230 (25.0)
Relative or carer		297 (32.2)
Purchased by residential home		306 (33.2)
Prepare meals, n (%)	921	
Relative or carer		561 (60.9)
With supervision		117 (12.7)
Without supervision		145 (15.7)
Without supervision & prepare variety of meals		98 (10.6)
Types of food daily eaten, n (%)		
Starch	919	916 (99.7)
Fruit / vegetables	921	864 (93.8)
Milk / yoghurt	920	896 (97.4)
Meat, fish, eggs/ other vegetarian alternative	919	898 (97.7)
Daily proportion of fruit, vegetable, n (%)	920	
None		33 (3.6)
1 a day		57 (6.2)
2 a day		130 (14.1)
3 a day		230 (25.0)
4 a day		199 (21.6)
5 a day		213 (23.2)
6 a day		36 (3.9)
7 or more		22 (2.4)
Questionnaires Administered Via Interview	930	
Health Related Quality of Life		
EQ-5D Score	872	0.8 (± 0.3)
EQ-5D Scale	877	78.1 (± 19.4)
Depression		
GDS-LD	317	7.5 (± 6.7)
Number Depressed, n (%)		67 (21.1)
GDS-LD Carer Supplement	464	5.5 (± 5.8)
Number Depressed, n (%)		71 (15.3)
Carer Completed Outside Appointment	930	

Characteristic	N (medical record)	Mean (± SD) Unless stated otherwise
Behaviour Problem		
Aberrant Behaviour Checklist	341	
1) Irritability, Agitation, Crying		4.3 (± 6.7)
2) Lethargy, Social Withdrawal		3.5 (± 5.5)
3) Stereotypic Behaviour		1.2 (± 2.6)
4) Hyperactivity, Noncompliance		3.9 (± 6.0)
5) Inappropriate Speech		1.3 (± 2.2)
Total score		14.0 (± 19.5)
Psychiatric Disorders		
PAS-ADD Checklist Section 1	930	
No events		207 (22.3)
Death of a first degree relative		34 (3.7)
Death of a close friend, carer or relative		36 (3.9)
Serious illness or injury		21 (2.3)
Retirement form work		1 (0.1)
Serious illness of relative, carer or friend		28 (3.0)
Move of house or residence		45 (4.8)
Break up of steady relationship		10 (1.1)
Separation or divorce		1 (0.1)
Alcohol problem		1 (0.1)
Drug problem		1 (0.1)
Serious problem with relative, carer/friend		11 (1.2)
Unemployed/seeking work		4 (0.4)
Breakdown of relationship with parent(s)		4 (0.4)
Laid off or sacked from work		0
Something valuable lost or stolen		4 (0.4)
Problems with police or other authority		7 (0.8)
Major financial crisis		1 (0.1)
Sexual problem		2 (0.2)
Other event		38 (4.1)
PAS-ADD Checklist Section 2	325	
Possible organic condition		1.0 (± 1.7)
≥Threshold score, n (%)		20 (6.2)
Affective or neurotic disorder		1.4 (± 3.2)
≥Threshold score, n (%)		28 (8.6)
Psychotic disorder		0.2 (± 0.6)
≥Threshold score, n (%)		16 (4.9)

Appendix 19: Example topic guide for service users interviews – education development stage

Topic guide

I'm not going to be testing anything you want to say is going to be fine.

If you don't understand what I'm asking please say that you don't understand and I will try and explain the question in another way.

You can say as little as you like or as much as you like.

Pre-Warm

Can you tell me a bit what you like doing during the day?

What kinds of things do you like eating?

Do you have any favourite TV programmes you like watching?

Do you go out with your key worker/carer, or in a group?

A. Being Healthy

I am going to ask some questions about being healthy? Is that ok?

1. What sort of things can people do to be healthy?

   Prompt: use cards/photos: walking, gym, swimming, eating a cake, eating salad etc.

2. What can you do to be healthy?

3. Are you healthy now? (if answered yes) probe following:

   - Did you get any help to get you healthy? Probe: what did you do?

4. If answered no the above probe the following:

   - What could you do to make that happen? Probe: for example, what foods could you eat and/or exercise?

5. Who helps you to stay healthy? Probe: doctor, nurse, carer, friend, parent

   - What do they do that helps you?

1

B. **Diabetes**

I am going to ask you some questions about diabetes now, is that ok?

6. What is diabetes?

7. How do people get diabetes?

8. Do you know how you can stop diabetes?

C. **Group learning**

I am going to ask you about learning in a group now, is that ok?

9. Have you ever worked in a group before to learn something? If yes probe the following:

    - Did you enjoy it? Probe what they enjoyed
    - What did you learn?
    - Did they have:

        - Pictures
        - Photographs
        - Games
        - Someone standing at the front and teaching you or sitting with you
        - Talking with other people in a group

10. If answered no the above, would you like to learn with people in a group? Probe: responses

D. Carers

11. Would you like carers to be there with you?

12. Would you like carers to be in a separate group?

Tips

- Take time in between questions and allow time for them to respond.
- If the question hasn't been understood, then do not re-word the question but simply repeat the same question.

Appendix 20: Example form for educator training

STOP Diabetes: Educator Training Evaluation Day 1

STOP diabetes

Date of attendance: April 3rd 2014

Thank you for attending our STOP Diabetes: Educator Training. We would welcome your opinion and reflection on whether the training has met your expectations, fulfilled your training needs and what aspects you have found to be enjoyable.

Please rate how useful you found the following components by circling the relevant score:

Session	Not at all useful					Very useful
Participants arrive and Coffee	1	2	3	4	5	6
Welcome and Introductions	1	2	3	4	5	6
What is different about group self-management education for people with ID	1	2	3	4	5	6
Prevention Messages	1	2	3	4	5	6
What's the take home message?	1	2	3	4	5	6
Development and theoretical underpinning	1	2	3	4	5	6
LD STOP Carer Session	1	2	3	4	5	6
LD STOP Participant sessions 1-4 and resources	1	2	3	4	5	6
Feedback: Sessions 1-4	1	2	3	4	5	6
Challenges and solutions	1	2	3	4	5	6
Preparing to deliver STOP.	1	2	3	4	5	6
What are the next steps and My Action Plan	1	2	3	4	5	6

Please circle one answer on the questions below. Feel free to add comments about specific issues.

The mixture of familiar and unfamiliar information was:

Far too little information | Too little information | About right | Too much new information | Much too much new information

I have learnt new skills

Strongly agree | Agree | Neither | Disagree | Strongly disagree

I believe I can apply the skills

Strongly agree | Agree | Neither | Disagree | Strongly disagree

I would recommend this training to other people?

Strongly agree | Agree | Neither | Disagree | Strongly disagree

STOP Diabetes: Educator
Training Evaluation Day 1

Which were the three most helpful aspects of the training to you? i ii ii
Which were the three least helpful aspects of the training to you? i ii ii
Identify at least one way that the training could be improved?
Identify which part of the training you found most enjoyable:
Identify at least one aspect of this training that you could implement in your practice:
Any other comments:

Venue Evaluation:

Please circle as appropriate	Very unsatisfactory				Satisfactory
Meeting Room	1	2	3	4	5
Catering	1	2	3	4	5
Accessibility	1	2	3	4	5
Any other comments:					

Thank you for completing this evaluation sheet

Appendix 21: Scope of the economic evaluation

The reasons for not attempting to estimate the cost-effectiveness of screening people with ID for diabetres (including T2DM)/IGR and overweight/obese are listed below:

1. Lack of evidence

There is a dearth of good quality evidence in relation to the costs and effects of diabetes prevention interventions in people with ID.

2. Number of pathways/screening strategies

The economic model needs to take account of all permutations of screening for diabetes only, screening for diabetes and IGR, and screening for overweight/obese. Since screening cannot be considered in isolation (i.e. it depends upon interventions), the economic model would need to take into account of how standard prevention interventions and the STOP diabetes education programme would be implemented for people with ID. It is also unclear how such screening would fit into existing policy in relation to Learning Disabiltiy Health Checks.

3. Evaluation of screening outside of the UK

Evaluating screening outside the UK in people with ID would lead to unreliable conclusions because:

- we do not have estimates for the prevalence of undiagnosed diabetes and IGR, and the rates are likely to be different in other countries (even those within Europe);
- there are different thresholds for HbA1C for diagnosing IGR;
- we do not know how effective prevention interventions would be; and
- we would need to model different countries' diagnostic and care pathways, use country-specific costs, and different thresholds for 'willingness-to-pay'.

Appendix 22: Comparison of surrogate-based physical activity approach against Yates et al. 2014

When using biomarkers (e.g. changes in physical activity through BMI and SBP) to predict clinical events, it is important where possible to undertake validation against a study reporting hard clinical outcomes. Potentially surrogate-based modelling could overlook some other mechanism of reduction in risk of CVD. To the extent that any such other mechanisms are correlated with changes in BMI and systolic blood pressure, these mechanisms would be captured within our mapping. It was decided to compare the model's predicted impact on CVD outcomes with another study.

In consultation with clinical experts we were directed to the NAVIGATOR trial results (Yates et al. 2014[269]), which could be used for the validation. In this study, all groups participated in a lifestyle modification programme that was designed to help them achieve and maintain a 5% weight loss, reduce the amount of saturated and total fats in their diet and increase physical activity to 150 minutes per week. The study reported the relationship between activity (steps) and CVD outcomes (events) in a cohort of 9,306 people. The analysis controlled for changes in BMI.

For the validation, a model adaptation was created which mimicked the NAVIGATOR trial by assuming changes in daily step counts continued without declining for a period of 6 years (the study followed participants for 6 years, but was not an intervention trial so we assumed that steps/day was stationary rather than declining). For an increase in activity of 2,000 steps, Yates and colleagues reported a hazard ratio of experiencing a cardiovascular event over the following 6 years of 0.92 (95% CI = 0.86-0.99); that is, a risk reduction of 8%. The hazard ratio from our adapted model was 0.95 (5% risk reduction).

Appendix 23: Database search terms for health economic analysis

Table 66: PubMed database search terms for physical activity studies

("activity"[title] OR "sedentary"[title] OR "exercise"[title])
AND
("weight"[title] OR "diabetes"[title] OR "BMI"[title] or "cardio-metabolic"[title] or "glucose"[title])
AND
("steps"[All Fields] OR "step-counter"[All Fields] OR "accelerometer"[All Fields])
AND ("weight"[All Fields] OR "diabetes"[All Fields] OR "BMI"[All Fields])
AND ("blood glucose"[All Fields] OR "hba1c"[All Fields] OR "cholesterol"[All Fields] OR "BMI"[All Fields] OR "weight"[All Fields] OR "waist"[All Fields] OR "hip"[All Fields] OR "blood pressure"[All Fields] OR "glycated haemoglobin"[All Fields] OR "blood sugar"[All Fields])
NOT ("school"[title] OR "child"[title] OR "children"[title] OR "childhood"[title])

(Date of search: 23/10/2015)

Appendix 24: Modelling cardiovascular events

The QRISK2 risk equation can be used to calculate the probability of a cardiovascular event, including coronary heart disease (angina or myocardial infarction), stroke, transient ischaemic attacks and fatality due to CVD.

The QRISK assumptions regarding the relationship between IGR, diabetes and cardiovascular disease were modified for the model and are outlined below:

1) It was assumed that individuals with HbA1c>6.5 have an increased risk of cardiovascular disease even if they have not received a formal diagnosis.
2) Risk of cardiovascular disease was assumed to increase with HbA1c for test results greater than 6.5 to reflect observations from the UK Prospective Diabetes Study (UKPDS) that HbA1c increases the risk of myocardial infarction and stroke.[277]
3) Prior to T2DM (HbA1c>6.5), HbA1c was assumed to be linearly associated with CVD. A study from the EPIC Cohort found that a unit increase in HbA1c increases the risk of coronary heart disease by a hazard ratio of 1.25, after adjustment for other risk factors[287] Individuals with an HbA1c greater than the mean HbA1c observed in the Health Survey for England (HSE) 2011 cohort were at greater risk of CVD than those with an HbA1c lower than the HSE mean.[264]

The QRISK algorithm identifies which individuals experience a cardiovascular event but does not specify the nature of that event. The nature of the cardiovascular event was determined independently. A targeted search of recent Health Technology appraisals of cardiovascular disease was performed to identify a model for the progression of cardiovascular disease following a first event (Table 67).

Table 67: Coefficients from the 2012 QRISK2 risk equation and estimated standard errors

Covariates	Estimated coefficients adjusting for individual characteristics								
	Women		Men			Women		Men	
	Mean	Standard error	Mean	Standard error	Interaction terms	Mean	Standard error	Mean	Standard error
White	0.0000	0.0000	0.0000	0.0000	Age1*former smoker	0.1774	0.035	-3.881	0.776
Indian	0.2163	0.0537	0.3163	0.0425	Age1*light smoker	-0.3277	0.066	-16.703	3.341
Pakistani	0.6905	0.0698	0.6092	0.0547	Age1*moderate smoker	-1.1533	0.231	-15.374	3.075
Bangladeshi	0.3423	0.1073	0.5958	0.0727	Age1*Heavy smoker	-1.5397	0.308	-17.645	3.529
Other Asian	0.0731	0.1071	0.1142	0.0845	Age1*AF	-4.6084	0.922	-7.028	1.406
Caribbean	-0.0989	0.0619	-0.3489	0.0641	Age1*renal disease	-2.6401	0.528	-17.015	3.403
Black African	-0.2352	0.1275	-0.3604	0.1094	Age1*hypertension	-2.2480	0.450	33.963	6.793
Chinese	-0.2956	0.1721	-0.2666	0.1538	Age1*Diabetes	-1.8452	0.369	12.789	2.558
Other	-0.1010	0.0793	-0.1208	0.0734	Age1*BMI	-3.0851	0.617	3.268	0.654
Non-smoker	0.0000	0.0000	0.0000	0.0000	Age1*family history CVD	-0.2481	0.050	-17.922	3.584
Former smoker	0.2033	0.0152	0.2684	0.0108	Age1*SBP	-0.0132	0.003	-0.151	0.030
Light smoker	0.4820	0.0220	0.5005	0.0166	Age1*Townsend	-0.0369	0.007	-2.550	0.510
Moderate smoker	0.6126	0.0178	0.6375	0.0148	Age2*former smoker	-0.0051	0.001	7.971	1.594
Heavy smoker	0.7481	0.0194	0.7424	0.0143	Age2*light smoker	-0.0005	0.000	23.686	4.737
Age 1[a]	5.0327		47.3164		Age2*moderate smoker	0.0105	0.002	23.137	4.627
Age 2[a]	-0.0108		-101.2362		Age2*Heavy smoker	0.0155	0.003	26.867	5.373
BMI[a]	-0.4724	0.0423	0.5425	0.0299	Age2*AF	0.0507	0.010	14.452	2.890
Ratio Total /HDL cholesterol	0.1326	0.0044	0.1443	0.0022	Age2*renal disease	0.0343	0.007	28.270	5.654
SBP	0.0106	0.0045	0.0081	0.0046	Age2*hypertension	0.0258	0.005	-18.817	3.763

Townsend	0.0597	0.0068	0.0365	0.0048	Age2*Diabetes	0.0180	0.004	0.963	0.193
AF	1.3261	0.0310	0.7547	0.1018	Age2*BMI	0.0345	0.007	10.551	2.110
Rheumatoid arthritis	0.3626	0.0319	0.3089	0.0445	Age2*family history CVD	-0.0062	0.001	26.605	5.321
Renal disease	0.7636	0.0639	0.7441	0.0702	Age2*SBP	0.0000	0.000	0.291	0.058
Hypertension	0.5421	0.0115	0.4978	0.0112	Age2*Townsend	-0.0011	0.000	3.007	0.601
Diabetes	0.8940	0.0199	0.7776	0.0175					
Family history of CVD	0.5997	0.0122	0.6965	0.0111					

AF: Atrial Fibrillation; CVD: Cardiovascular disease; SBP: systolic blood pressure

[a] covariates transformed with fractional polynomials

All QRISK events are assigned to a specific diagnosis according to age- and sex- specific distributions of cardiovascular events used in a previous Health Technology Assessment (HTA).[289] The probability of cardiovascular outcomes by age and gender is shown in Table 68.

Table 68: The probability distribution of cardiovascular events by age and gender

Sex	Age	Stable angina	Unstable angina	MI rate	Fatal CHD	TIA	Stroke	Fatal CVD
Men	45-54	0.307	0.107	0.295	0.071	0.060	0.129	0.030
	55-64	0.328	0.071	0.172	0.086	0.089	0.206	0.048
	65-74	0.214	0.083	0.173	0.097	0.100	0.270	0.063
	75-84	0.191	0.081	0.161	0.063	0.080	0.343	0.080
	85+	0.214	0.096	0.186	0.055	0.016	0.351	0.082
Women	45-54	0.325	0.117	0.080	0.037	0.160	0.229	0.054
	55-64	0.346	0.073	0.092	0.039	0.095	0.288	0.067
	65-74	0.202	0.052	0.121	0.081	0.073	0.382	0.090
	75-84	0.149	0.034	0.102	0.043	0.098	0.464	0.109
	85+	0.136	0.029	0.100	0.030	0.087	0.501	0.117

MI: myocardial infarction; CHD: coronary heart disease; TIA: transient ischaemic attack; CVD: cardiovascular disease

After an individual has experienced a cardiovascular event, it is not possible to predict the transition to subsequent cardiovascular events using QRISK2. Instead, as with assigning first CVD events, the statin HTA reports the probability of future events, conditional on the nature of the previous event.[289] More details on the probabilities within a year of transitioning from stable angina, unstable angina, myocardial infarction (MI), transient ischemic attack (TIA) or stroke for individuals in different age groups can be found in an on-line Discussion Paper.[265]

Appendix 25: Assumptions made for diagnosis and treatment of diabetes, hypertension and CVD risk for health economic analysis

Table 69: Assumptions made for diagnosis and treatment of diabetes hypertension and high CVD risk

Diabetes	Hypertension	High CVD risk
DIAGNOSIS		
- At baseline, individuals are assigned an HbA1C threshold above which diabetes is detected opportunistically. - Individuals with HbA1C levels above their individual threshold will attend the GP to be diagnosed with diabetes.	- Assumed that people eligible for anti-hypertensive treatment will be identified through opportunistic screening if they meet certain criteria and see the GP at least once during the simulation period.	- Assumed that people eligible for statins will be identified through opportunistic screening if they meet certain criteria and see the GP at least once during the simulation period.
TREATMENT Assumed that there are three, non-mutually exclusive outcomes from the vascular checks and opportunistic screening		
Patient's blood glucose test indicates T2DM as measured by HbA1c > 6.5mmol/L (assumed that FPG and 2-hr glucose test not used for diabetes diagnosis, but future adaptations of the model could include these criteria). A 3-stage treatment regime assumed (as a trade-	Patient has high blood pressure and should be treated with anti-hypertensive medication: - Anti-hypertensive treatment initiated if systolic BP>160. - Anti-hypertensive treatment initiated if systolic BP>140 and individual also has a history of CVD, diabetes, or CVD risk >20%.[338]	Patient receives statins to reduce cardiovascular risk: - Statins initiated if >20% 10-year CVD risk estimated from the QRISK[286] 2012 algorithm.[339]

off between model simplicity and capturing key cost differences between interventions): (a) At diagnosis patients are prescribed low cost treatments, represented by Metformin 500mg/day. (b) If HbA1c level rises above 7.4%, individual is prescribed more expensive DPP-IV inhibitor + Metformin. (c) Individual is assumed to continue DPP-IV inhibitor + Metformin until HbA1c level rises above 8.5% whereby they are assumed to require insulin.		

More details are available on-line.[265]

Appendix 26: Distributions for key parameters within the probabilistic sensitivity analysis

Given the very large numbers of parameters in the model, many of which belong to complex forms of statistical modelling, it would not be helpful to present all of them in this report. Below (Table 70) we present distributions for parameters related to the intervention, the relationship between physical activity (steps)[271] and BMI and other risk factors. Details of distributions for the other model parameters are reported elsewhere.[265]

Table 70: Uncertainty around Bravata-based intervention effect size (assuming 2,491 steps)

Parameter Description	Distribution	Parameter 1	Parameter 2	Central estimate	Source
BMI	Normal	-0.38	0.171	-0.38	Bravata
SBP	Normal	-3.8	1.071	-3.8	Bravata
Total Cholesterol	Normal	-0.09	0.120	-0.09	Bravata
HDL Cholesterol	Normal	0.06	0.039	0.06	Bravata

No uncertainty is included around uptake rates. As duration of effect is explored through scenario analyses, no uncertainty is included around this parameter.

Mortality

Mortality rates from other causes by age were assumed to be constant in the probabilistic sensitivity analysis (PSA).[282] The parameter distributions for the hazard ratio for other cause mortality with diabetes and for the standardised mortality ratios for other cause mortality in males and females with ID are reported in Table 71. The table shows the probability distribution for each model parameter and the mean value (central estimate). Parameter 1 and parameter 2 are arguments for the specific forms of statistical model, such as lognormal.

Table 71: Input parameters for mortality hazard ratio for diabetes and standardised mortality ratios (SMR) for intellectual disability

Parameter Description	Distribution	Distribution Parameter 1	Distribution Parameter 2	Central estimate
Mortality hazard ratio for diabetes	Lognormal	0.588	0.186	1.80
SMR for intellectual disability in males	Normal	3.24	0.219	3.24
SMR for intellectual disability in females	Normal	2.28	0.138	2.28

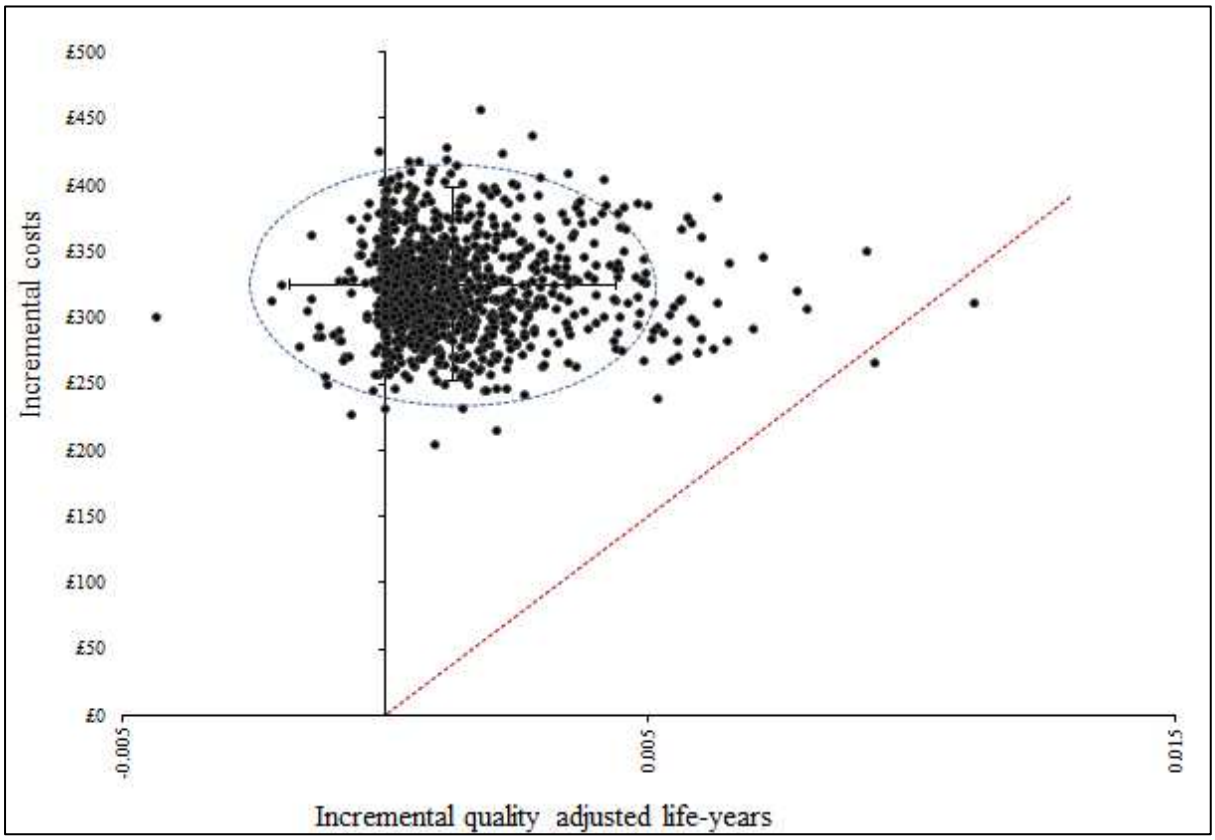


Figure 30: Cost-effectiveness plane for an increase of 2,491 steps at £30,000 per QALY

QALY: Quality-adjusted life year

In Figure 30, each black dot represents a result from a sample run of the PSA. The red line represents the cost-effectiveness frontier; points below this line represent sample results from the PSA that lie in the cost-effective region. The spread of the points gives an indication, for this type of intervention, of how much uncertainty there is around the reported mean incremental costs and QALYs (quality-adjusted life years).

Appendix 28: Detailed threshold analysis results tables at £20,000 per QALY

Table 72: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome (base case intervention £20,000 per QALY assuming all risk factors change together)

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-5.0	-50	-1.88	-3.4	-34	-1.45
1000	-0.2	-2	-0.09	-4.8	-48	-1.85	-3.2	-32	-1.40
3000	-0.5	-5	-0.26	-4.5	-45	-1.77	-2.9	-29	-1.30
5000	-0.8	-8	-0.42	-4.2	-42	-1.69	-2.6	-26	-1.19
7000	-1.1	-11	-0.57	-3.9	-39	-1.60	-2.3	-23	-1.08
9000	-1.4	-14	-0.71	-3.6	-36	-1.52	-2.0	-20	-0.97
11000	-1.7	-17	-0.84	-3.3	-33	-1.42	-1.7	-17	-0.84
13000	-2.0	-20	-0.97	-3.0	-30	-1.32	-1.4	-14	-0.71
15000	-2.3	-23	-1.08	-2.7	-27	-1.22	-1.1	-11	-0.57

Table 73: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome (increased effectiveness intervention at £20,000 per QALY assuming all risk factors change together)

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-4.7	-47	-1.83	-2.7	-27	-1.25
1000	-0.2	-2	-0.09	-4.6	-46	-1.79	-2.6	-26	-1.19
3000	-0.5	-5	-0.26	-4.3	-43	-1.71	-2.3	-23	-1.08
5000	-0.8	-8	-0.42	-4.0	-40	-1.63	-2.0	-20	-0.97
7000	-1.1	-11	-0.57	-3.7	-37	-1.54	-1.7	-17	-0.84
9000	-1.4	-14	-0.71	-3.4	-34	-1.45	-1.4	-14	-0.71
11000	-1.7	-17	-0.84	-3.1	-31	-1.35	-1.1	-11	-0.57
13000	-2.0	-20	-0.97	-2.7	-27	-1.25	-0.8	-8	-0.42
15000	-2.3	-23	-1.08	-2.4	-24	-1.14	-0.5	-5	-0.26

Table 74: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for an obese subgroup (base case intervention at £20,000 per QALY assuming all risk factors change together)

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-4.7	-47	-1.83	-2.7	-27	-1.25
1000	-0.2	-2	-0.09	-4.6	-46	-1.79	-2.6	-26	-1.19
3000	-0.5	-5	-0.26	-4.3	-43	-1.71	-2.3	-23	-1.08
5000	-0.8	-8	-0.42	-4.0	-40	-1.63	-2.0	-20	-0.97
7000	-1.1	-11	-0.57	-3.7	-37	-1.54	-1.7	-17	-0.84
9000	-1.4	-14	-0.71	-3.4	-34	-1.45	-1.4	-14	-0.71
11000	-1.7	-17	-0.84	-3.1	-31	-1.35	-1.1	-11	-0.57
13000	-2.0	-20	-0.97	-2.7	-27	-1.25	-0.8	-8	-0.42
15000	-2.3	-23	-1.08	-2.4	-24	-1.14	-0.5	-5	-0.26

Table 75: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for a 45 to 49-year-old subgroup (base case intervention at £20,000 per QALY assuming all risk factors change together)

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-2.6	-26	-1.19	-1.9	-19	-0.94
1000	-0.2	-2	-0.09	-2.4	-24	-1.14	-1.8	-18	-0.87
3000	-0.5	-5	-0.26	-2.1	-21	-1.03	-1.4	-14	-0.74
5000	-0.8	-8	-0.42	-1.8	-18	-0.90	-1.1	-11	-0.61
7000	-1.1	-11	-0.57	-1.5	-15	-0.78	-0.8	-8	-0.46
9000	-1.4	-14	-0.71	-1.2	-12	-0.64	-0.5	-5	-0.30
11000	-1.7	-17	-0.84	-0.9	-9	-0.50	-0.2	-2	-0.13
13000	-2.0	-20	-0.97	-0.6	-6	-0.34	0.0	0	0.00
15000	-2.3	-23	-1.08	-0.3	-3	-0.18	0.0	0	0.00

Table 76: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for a ≥50-year-old subgroup (base case intervention at £20,000 per QALY assuming all risk factors change together)

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-4.1	-41	-1.67	-2.7	-27.5	-1.2
1000	-0.2	-2	-0.09	-4.0	-40	-1.63	-2.6	-25.9	-1.2
3000	-0.5	-5	-0.26	-3.7	-37	-1.54	-2.3	-22.9	-1.1
5000	-0.8	-8	-0.42	-3.4	-34	-1.45	-2.0	-19.8	-1.0
7000	-1.1	-11	-0.57	-3.1	-31	-1.35	-1.7	-16.8	-0.8
9000	-1.4	-14	-0.71	-2.7	-27	-1.25	-1.4	-13.7	-0.7
11000	-1.7	-17	-0.84	-2.4	-24	-1.14	-1.1	-10.7	-0.6
13000	-2.0	-20	-0.97	-2.1	-21	-1.03	-0.8	-7.6	-0.4
15000	-2.3	-23	-1.08	-1.8	-18	-0.90	-0.5	-4.6	-0.3

Table 77: Combinations of daily step increases and additional dietary changes required to achieve a cost-effective outcome for a high CVD risk subgroup (base case intervention at £20,000 per QALY assuming all risk factors change together)

Initial increase in steps needed	Change attributable to the increase in steps			Additional change needed to be generated through diet			Additional change needed to be generated through diet		
				Base case (3 year durability)			5 year durability		
	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio	BMI (kg/m^2)	SBP (mmHg)	Total:HDL cholesterol ratio
0	0.0	0	0.00	-3.5	-35	-1.49	-2.1	-21	-1.03
1000	-0.2	-2	-0.09	-3.4	-34	-1.45	-2.0	-20	-0.97
3000	-0.5	-5	-0.26	-3.1	-31	-1.35	-1.7	-17	-0.84
5000	-0.8	-8	-0.42	-2.7	-27	-1.25	-1.4	-14	-0.71
7000	-1.1	-11	-0.57	-2.4	-24	-1.14	-1.1	-11	-0.57
9000	-1.4	-14	-0.71	-2.1	-21	-1.03	-0.8	-8	-0.42
11000	-1.7	-17	-0.84	-1.8	-18	-0.90	-0.5	-5	-0.26
13000	-2.0	-20	-0.97	-1.5	-15	-0.78	-0.2	-2	-0.09
15000	-2.3	-23	-1.08	-1.2	-12	-0.64	0.0	0	0.00

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