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‘It’s really difficult because sometimes you want to be special and different and sometimes you just want to be like everybody else’: Including families of children with chromosomal learning disabilities in a longitudinal qualitative cohort study

Abstract

The UK Economic and Social Research Council (ESRC) is considering developing a longitudinal qualitative cohort study of seldom-heard families. This paper presents findings from a scoping study of best practice in qualitative longitudinal research focused on families with a child with a chromosomal learning disability. We conducted in-depth online interviews with twelve parents and identified their views of research participation in relation to recruitment, retention, data collection and ethics. They identified barriers to, and facilitators of, participation in qualitative longitudinal research for themselves and their children. Their views, analysed using template analysis, were used to co-produce a suite of recommendations for the ESRC. All participants welcomed the proposed research and believed that amplifying the voices of families with experience of learning disability could make a tangible difference to their experience and to public understanding of disability.

Keywords

Qualitative longitudinal research; seldom-heard families; chromosomal learning disabilities; research participation; qualitative methods; inclusion; co-production

Points of Interest

1. The parents of children with chromosomal learning disabilities we interviewed were unanimously supportive of the idea of a longitudinal qualitative cohort study of seldom heard families, including families with experience of learning disability.
2. Participants told us that recruitment should be led by trusted local organisations, that their motivation would be to make life better for others, that they and their children would need to get to know researchers well and that a 'one size fits all' approach to data collection would not work.
3. Participants told us that the most pressing research questions for them related to Education and Healthcare Plans (EHCPs) and social care in adulthood.
4. Participants' views were used to co-produce recommendations for the ESRC who, at the time of the study, were exploring the possibility of commissioning a study of this nature..

Introduction

Designing and delivering evidence-informed and effective education for children and young people with special educational needs and disabilities is dependent on high quality research that includes the voices of these children and young people and those who know them best. However some families, including those with children who have learning disabilities, are known to be seldom-heard in research and this has negative practical implications (Coe et al., 2023; DCP, 2023; Mitchell et al., 2023; NIHR, 2024). Underrepresentation of these families is detrimental to the special educational needs community and to the whole of society because it means that research findings that affect everybody are not as representative as they should be. It is particularly problematic in light of claims that the needs of families with

disabled children are not being met in countries including the UK, the site of the current study, and would benefit from more evidence-informed intervention and support (Aiyegbusi et al., 2023; DCP, 2023). In this paper we explore barriers to participation in research for families of learning disabled children, along with potential facilitators. The aim of the research is to understand how to include families of children and young people with special educational needs in studies that could make a positive difference to their experiences.

Understanding and addressing barriers to participation could lead to these families gaining a more prominent voice in research that can inform policies related to education, as well as health, housing, welfare and social care. It could also lead to an improved socio-cultural understanding of special educational needs and family experience which has the potential to challenge dominant narratives that are often driven by an understanding rooted in a medical model of disability (Lalvani and Polvere, 2013). This is likely to require qualitative longitudinal research rather than a survey-based approach such as that used in studies such as the 1000 Families Study (Hastings et al., 2020) and this presents researchers with particular challenges and opportunities.

While developing a new UK-wide longitudinal cohort study the UK's Economic and Social Research Council (ESRC) have acknowledged that some groups of families, including those with learning disabled children, are less likely to participate and that this has clear implications for the representativeness of national-level data. One solution they are considering to this problem is to develop a longitudinal qualitative study focused specifically on seldom-heard families, including the families of learning disabled children. There is limited literature regarding the methodological challenges of recruiting these particular families and retaining them in longitudinal qualitative research (Torgerson et al., 2024). In order to support their thinking the ESRC commissioned two scoping studies to explore the

benefits of a longitudinal qualitative study of seldom-heard families, including those with learning disabled children, and the optimal research methods for operationalising such a study. The research presented in this paper comes from one of those scoping studies which focused on families of children and young people with chromosomal learning disabilities. The findings are likely to also be relevant to families of children with a much wider range of learning disabilities.

Studies suggest that these families are seldom-heard in research for a variety of reasons, including recruitment challenges such as a lack of willingness among some gatekeepers to grant researchers access to the target population (Williams, 2020). This problem arises from a well-founded desire to protect potentially vulnerable people, sometimes combined with not seeing the research as beneficial. Other reasons include families feeling unable to find time to take part as a result of competing demands (Cox et al., 2021; Gillooly et al., 2022). This challenge is relevant to the recruitment and retention of these families.

Attrition is to be expected in qualitative longitudinal research with any population as participants' personal circumstances change over time (Aitken et al., 2003; van Wijk, 2014). Plans to mitigate this need to be built into study design from the outset (Neale, 2021). Helpful strategies when thinking about families of children with learning disabilities are likely to include collaborating with relevant organisations (van Wijk, 2014), employing researchers who understand the group well (Westmarland and Bows, 2018), maintaining contact between data collection waves (Neale, 2021) and ensuring continuity of researchers in order to sustain trusted relationships with participants (Daniluk, 2001; Thomson and Holland, 2003; Shirani, 2010). Studies also suggest that these participants are more likely to be motivated to stay involved if they feel they are making a difference to others, rather than

for financial gain. This is relevant to planning incentives and communication in the context of qualitative longitudinal research (Shirani, 2010). To our knowledge no other study has specifically sought the views of families with learning disabled children on these issues.

Beyond recruitment and retention challenges, a further barrier is the need for data collection to be inclusive, creative and accessible when many data collection tools are not (Gillooly et al., 2022; Lightfoot and Bond, 2013; Williamson, 2019). This is particularly relevant as children develop and it becomes appropriate to include their own voices as well as those of their family members. Researchers in qualitative longitudinal research suggest that a toolbox of data collection approaches can be helpful in addressing this barrier (Plummer, 2001). Interviews are generally viewed as a key data collection strategy but the importance of being flexible about where and how interviews take place is emphasised (Weller, 2012; van Wijk, 2014). Some researchers also recommend arts-based methods as a helpful way of easing hierarchies between researchers and participants (Westmarland and Bows, 2018). Photographs and videos are valued by some as rich and tangible data for tracking change over time (Saldaña, 2003) and supporting interview-based discussions (Bytheway and Bornat, 2012). Diaries are also valued as data generation tools and ways of keeping in touch with participants between interviews. While a toolbox approach, in which these strategies can all be made available, is widely supported it comes with the caveat that all tools need to be evidence-informed and aligned with the needs of the research as well as those of the participants (Neale, 2021). This becomes particularly complex when considering inclusive ways of studying learning disabled children themselves, including those who communicate in non-traditional ways.

As well as challenges related to recruitment, retention and data collection an enhanced ethical consideration is needed for working with learning disabled children and young people and their families (Ballard et al., 2021; Deakin and Jahoda, 2020). Qualitative longitudinal researchers emphasise the importance of taking both proactive and reactive approaches to ethics, and treating ethics as an ongoing process (Neale, 2013). Consent (and assent for the children) needs, for example, to be revisited on a regular basis. In addition, the fact that longitudinal research is reliant on sustaining good relationships means it should be undertaken in a spirit of reciprocity in which researchers do not just 'take' data from participants but also give something in return (Warin, 2011). However, that spirit of reciprocity should not lead to over-dependence on the research team or be to the detriment of participants, researchers or the research itself (Birch and Miller, 2002; Morrow, 2012).

The challenges identified in the literature undoubtedly need careful consideration and planning, but they are unlikely to be insurmountable. One aim of the scoping research presented in this paper was to understand barriers to participation and identify optimal ways of addressing them. That said, in a systematic scoping review undertaken as part of the wider project that this study is part of it was notable that very little evidence was found of longitudinal qualitative research with this population, suggesting the barriers may be having a powerful effect on inclusive research practices (Torgerson et al., 2024).

It is possible that some families of children with chromosomal learning disabilities are interested in participating in longitudinal qualitative studies but do not see the research they are invited to participate in as being particularly impactful or relevant to them (Hoskins et al., 2023). We know, for instance, that some parents are specifically interested in research on their child's unique diagnosis (Gilmore, 2018; Holm et al., 2021) and some are interested in research that will support understanding and meeting their child's needs and supporting their

wellbeing and sense of self (Cox et al., 2021; Deakin and Jahoda, 2020). There is also evidence of interest in participating in research that could change public perceptions of disability (Deakin and Jahoda, 2020). This suggests that a co-produced research agenda that respects the priorities of families of children with special educational needs could potentially increase participation rates. It also suggests that adapting traditional data collection techniques to meet the needs of this group, and clearly communicating the benefits of participation to these families, may be beneficial.

As children grow and become ready to contribute to research themselves it is important that they, and their families, have opportunities to co-produce research methods and approaches that are accessible and acceptable to them (Amann and Sleigh, 2021). This could involve, for example, recommending suitable data collection strategies to encourage recruitment and retention (Gonzalez et al., 2021). In the current study we interviewed parents of children with chromosomal learning disabilities to seek their views on whether they would have wanted to join a longitudinal qualitative cohort study when their child was first born and how that would have worked best for them. We spoke to them about their perceptions of barriers and facilitators to taking part and we worked with them to co-produce recommendations for the ESRC on how to approach recruitment, retention, data collection and ethics sensitively and well if they decide to proceed with a qualitative longitudinal 'seldom heard families' study, including families of learning disabled children. Our research questions were:

5. Do parents of children with chromosomal learning disabilities think a longitudinal qualitative cohort study of 'seldom-heard' families, including families with learning disabled children, is a good idea and why?

6. What do they see as the optimal methods of recruitment, retention, data collection and ethics for a study of families of children with special educational needs?

Methods

Taking a critical realist approach, the study aimed to elicit experiences and perceptions of research from parents of children with chromosomal learning disabilities.

Participants

Parents or carers of a child or young person with a chromosomal learning disability such as Down Syndrome, Edwards' Syndrome or Patau's Syndrome were eligible to participate. We included parents of children and young people up to the age of 19 as the majority of these young people complete their school-based education at 19 rather than 18 in the UK.

Sampling and Recruitment

Recruitment initially focused on social media advertising and 48 respondents consented to participate within a very short time. However, on further investigation, it became clear that most of these respondents did not meet the study's eligibility criteria, with some clearly feigning their status as parents of children with a chromosomal learning disability. This problem has recently been noted elsewhere in studies that offer cash or vouchers to participants and appears to be a growing problem (Pellicano et al., 2024). A rigorous checking process suggested that only 2 of the 48 respondents were actually eligible to take part. A further recruitment drive using purposive sampling via closed special interest groups proved more successful and enabled the study to reach the intended population. This led to 15 new participants consenting to take part in the study. Three of the 17 eligible participants

eventually could not take part (due to other commitments) and two did not reply to the invitation leaving a final sample of *n*=12 (10 female). Table 1 describes the most relevant characteristics of participants' children.

Table 1. Characteristics of the children

Diagnosis	**Age**	**Gender**
Down syndrome	2	Female
	5	Male
	7	Male
	11	Female
	13	Male
	13	Female
	19	Female
5q duplication	Age 8	Female
3 genes missing (SWAN)	Age 16	Female
Edwards Syndrome	Age 8	Female
	Age 12	Female
Phelan McDermid Syndrome	Age 11	Male

Measures and Procedure

In-depth semi-structured interviews probed parents' perspectives of barriers and enablers to taking part in research. The interview schedule (OSF link) explored participants' ideas about the best ways of identifying and recruiting parents of children with learning disabilities; how

they felt about different types of qualitative data collection; what would help them to remain engaged with a longitudinal qualitative study; their thoughts about research ethics; and finally, thinking ahead, about the optimal ways of including the voices of children with special educational needs in such a study.

Parents were invited to take part in a recorded online interview at a time that was convenient for them. Interviews were conducted by the first author and were audio recorded and transcribed by an approved transcription service. Participants were offered the opportunity to review their transcript prior to full anonymisation and one chose to do so, and was happy for their data to be included in the study.

Analysis

Interview data were analysed thematically using Template Analysis (Pandey, 2016; King, 2004). Both authors familiarised themselves with the data prior to coding and analysis. An initial template was then developed using *a priori* themes based on the main areas represented in the interview schedule. Both authors then independently coded three transcripts using this template and added potential codes to each of the themes. Similarities and differences in coding, and thoughts on how well the *a priori* themes represented the data were then discussed and a new version of the template was developed to reflect these discussions. This iterative process continued as the first author coded the remaining transcripts, meeting with the second author to discuss new coding or altered coding and to update the template. The cycle of coding transcripts, discussion, and making revisions to the template continued until all transcripts had been coded. In total there were six iterations of the template before the final version was agreed. Regular team discussions ensured reflexivity and the development of codes and themes which described the data well (King, 2024). The stages of template development can be viewed here (OSF link).

Ethical Considerations

The Ethics Committee of the researchers' university department approved the study. It was considered possible that parents may become distressed when talking about why it could be difficult for them and their child to participate in longitudinal qualitative research. To address this, questions were framed sensitively and were tightly focused on the research aim of understanding potential barriers to, and facilitators of, participation. The interviewer was experienced in interviewing parents and working with children with special educational needs and disabilities and their families.

Results

Theme 1: Perceived benefits of taking part in longitudinal qualitative research

Participating parents of children and young people with special educational needs were unanimously enthusiastic about qualitative longitudinal research focused on families with experience of learning disabilities, describing it as something necessary that would: allow them to make a difference to others; generate evidence tailored to the challenges they and their children face; and benefit them and their children.

Why we need this

Participants acknowledged that studies that are not tailored to learning disabled children, can feel irrelevant and leave them feeling excluded and upset. One participant described how: 'especially in very structured surveys … you're kind of going, "Yeah, but you know, which one to tick because actually that's not us, that's not us"?' (P57).

They were supportive of taking a qualitative approach, feeling that this would make it easier for them to participate in a meaningful way. Participants discussed how a qualitative approach could capture the nuances of their family lives and their children's experiences more effectively than quantitative measures, and that this was important as they perceived a need for family life with a learning-disabled child to be represented realistically in society. They saw this as enhancing understanding among decision-makers and providers of support such as schools, but also among the public: 'I think there's just a lack of knowledge generally' (P10).

They also suggested that we simply don't know enough about children with Special Educational Needs (SENDs), and how to support them, and described a desire to build a more substantial evidence-base for policy-makers and education and healthcare professionals to draw upon in order to meet their children's needs.

> I think when we got [name]'s diagnosis I assumed that a lot of the medical people that would deal with him would know a lot about Down Syndrome and all the people that would look after him in his life and support him at school would know a lot about Down Syndrome but actually I seem to know a lot more about it now than a lot of the people that I come into contact with. (P56)

In short, participants were unanimous in their view that a qualitative longitudinal study of families of children with chromosomal learning disabilities would be beneficial and they described the negative impact of an absence of evidence in their own lives.

We want to make a difference

Altruism was one of participants' primary motives for supporting qualitative longitudinal research into families of children with special educational needs. Almost all described a strong desire to smooth the path for families coming up behind them, so that they could be spared some of the challenges they themselves had faced: 'I think we were willing to

participate in that kind of research because while it might not help us necessarily, it may help somebody else in the future*'* (P59). This was the first benefit identified by almost every participant.

What we care about

Participants were clear on the biggest challenges they face, and on group-specific topics a longitudinal qualitative study could and should address. Two particular issues of interest related to education and social care.

Participants were interested in research that could shed light on and improve Education and Healthcare Plan (EHCP) processes and outcomes (the EHCP is the document that gives children and young people a statutory entitlement to support to meet their needs in the UK). As one parent put it: 'It's a very horrific system' (P10). In the context of education they were also keen to focus on the mental health needs of learning disabled children and young people.

The other dominant issue raised by participants related to the availability and quality of social care from age 19 onwards: 'I think and I hope … it will inform … social care, which seems to drop off a cliff when children grow up and children get to 18*'* (P10). This was seen as a core priority for research.

What's in it for us?

Participants identified a lot of benefits to themselves of taking part in a study like this. Most notably, they saw it as an opportunity to be heard and the process of being listened to as cathartic: 'it helps process everything that's going on*'* (P51). They also talked about a range of positive emotions they perceived as likely outcomes of being involved, including enjoyment, pride, relief and belonging. For example, P10 said:

> but then I also think like... I think it could be an amazing study and I'd kind of want to be like, "Oh, look, I took part in this". Like not bragging rights, honestly … but like I want it to have an impact. I want it to change things. (P10)

Participants also described a belief that being part of a study like this would give them access to knowledge and information that they might otherwise not find that could support them in giving their children the best possible opportunities.

Theme 2: Perceived risks involved in taking part in longitudinal qualitative research

Overwhelmingly, participants saw the proposed research as being low risk. However, parents did acknowledge that talking about their families and their lives could make them feel exposed and vulnerable.

Talking about our lives can make us vulnerable

Parents felt it was important to take part in research into families with children with chromosomal learning disabilities because it could provide *'*a real snapshot of what's happening*'* (P10). However, they also acknowledged a tension here as *'*you won't want to always show the reality of what's happening or talk about it*'* (P10). This suggests a need for careful thought around how participants can be supported in ethical and meaningful ways, including co-production of dissemination and data management plans.

Some parents talked about how they were particularly vulnerable in the early years, while still processing their child's diagnosis. They described how talking about their lives can amplify their children's differences and how this can be *'*really hard because it highlights every time you do it everything she can't do*'* (P51).

Parents also expressed their concern about being identified more widely, even with anonymisation, and to potentially ask others to consider how they might feel if they self-identified their comments in research outputs:

> I suppose, you know, no matter how much of a pseudonym you throw in there, there are…you're talking about your circumstances, talking about your child, you might well, there might only be one child in the world …that ticks those particulars (P57)

Assumptions of representation

Participants provided good examples of things to consider when planning qualitative studies of families of learning disabled children. They suggested researchers be alert to the possibility that one parent's perspective may not be representative of the views of all family members, including those of the learning disabled child themself. They also advised that researchers should not assume too much similarity between children and young people, and their families, on the basis of a shared diagnosis, particularly if putting families in the study in touch with each other.

Finally, researchers were advised to ensure the explanation of the study is not stigmatising: 'it's really difficult because sometimes you want to be special and different and sometimes you just want to be like everybody else'. (P62)

In summary, participants predominantly felt that a qualitative longitudinal cohort study of families of children with learning disabilities would be low risk. However, from previous research encounters, participants identified approaches they felt would improve the positivity and inclusivity of any new qualitative research design.

Theme 3: How to recruit families of children with special educational needs for longitudinal qualitative research

Who we trust to recruit us

Participants expressed strong views on who an invitation to participate in research should come from and there was strong agreement that recruitment via special interest groups was likely to be the most effective. Overall, there was a preference for national organisations (e.g. the Down Syndrome Association, SOFT UK, Unique as some of the specific UK examples participants mentioned) to endorse a study and for the opportunity to be cascaded to families via local branches of that national organisation or other, related local organisations.

Participants also explained that endorsements from health professionals increased trust and could help with early recruitment of the most marginalised families. One participant said:

> Yes, portage, straightaway, particularly for the younger age group … And I'm also thinking you could also link in with the health visitors even before portage because they're referring families on to things …. That would be very beneficial (P51)

It was also suggested that advertising the study in GP surgeries and hospital settings may help to include families of learning disabled children and to increase the visibility of, and trust in, the study.

Finally, there was a discussion of the pros and cons of recruiting via social media. There was some openness to this, particularly in the context of a local special interest or support group with a Facebook page or a WhatsApp group. However, challenges with social media recruitment were also identified, in particular, the risk of missing some of the most seldom-heard families.

> I hope you will reach out for families who are not easy to reach, because there are a lot of these families who are not on Facebook, so for them, it's probably easier with younger children, it's easier to reach through medical professionals, like physio, occupational therapist or specialist teaching team at the council. (P52)

Participants strongly suggested that practitioners on the ground would be best placed to identify these more seldom heard families.

When to recruit us

Participants made the important point that it is not as straightforward as recruiting a cohort of infants with chromosomal learning disabilities at birth. This is for many reasons, including the fact that not all chromosomal learning disabilities are identified straightaway and, in some cases, they are either not evident until later or diagnosis takes a very long time, something that is true for many special educational needs and disabilities.

> So, you wouldn't have identified [name] … because it wasn't really until about 18 months that we recognised that she wasn't growing, so she had a growth hormone deficiency, and that she wasn't babbling. So, she wasn't making any speech production sounds. I thought I was blessed. I thought I had quite a quiet child. So, yeah, you wouldn't have identified us. (P14)

Most participants raised the point that families can also take a long time to process their child's diagnosis and may not be ready to engage for some time.

> I don't think I would have been able to, purely because I was quite traumatised by the diagnosis … so I was grieving when he was a baby. I don't think I would have been able to engage and talk about it at all, if I'm being honest. (P10)

This suggests a need for flexibility in the timing of the recruitment with families able to join any longitudinal qualitative cohort study at different times rather than solely from the first year of life.

The resources we need

Participants told us about the importance of clarity in recruitment materials, specifically the importance of using plain English, of preparing recruitment materials in multiple languages,

and of being accessible and non-intimidating: ‘you know, universities are approaching people, it just feels quite intimidating, I think, when you’ve got Professor this, and Dr that and whatever’ (P57)

It was also important for participants that research teams should make clear that their needs would be understood, for example acknowledging explicitly that their child could be with them during data collection if needed.

Theme 4: How to collect data from these families

It was explained to parents that qualitative data could be collected in diverse ways including interviews, home visits, diaries, photographs, videos or other creative methods.

The need for flexibility

Parents unanimously agreed that a single method would not be appropriate for all parents and that a toolbox approach in which participants could choose how they felt able to tell their stories, along with logistical flexibility to accommodate their needs, would be necessary to facilitate ongoing participation. As one participant put it: ‘the first question would be, “Right, well what would work for you?”’ (P59)

This need for flexibility also applied to the frequency of data collection with participants reporting mixed views on how often they would feel willing and able to give data.

> I think... my gut says annually. I think certainly no more (P59)
>
> I would say probably once about every six months feels about right. (P62)
>
> I'd rather have more often and less to do than a lot in one go because I just don't have those chunks of time. (P58)

This suggests that there may be a benefit to developing a data collection system in which participants are strongly encouraged to participate in a main wave of data collection each year but also have the opportunity to participate in other optional activities throughout the year.

Challenges

Participants were clear that the research team would need to ensure the demands of the study should not be too high. For instance, in discussing their view of diary methods one participant said:

> It would depend on the level of detail and how much I'm expected to write and how much time, to be honest. If it was like you need a lot of them, like every day, it might be hard for me to do. (P10)

They also felt that some methods may act as emotional triggers. When discussing photo elicitation, for example, one participant said:

> I suppose you could get emotional if you kind of picked a particular photo that kind of set off things … I think that would be potentially the thing that I would think might be tricky for some. (P57)

Finally, parents explained the challenges of keeping their children - many of whom experience health challenges, infection free and asked that researchers take safety precautions prior to working with them, e.g. testing for COVID-19.

Our gut reactions to each data collection approach

Participants preferred there to be a range of collection methods, as a 'one size fits all' approach would not be appropriate. This becomes particularly important when thinking about data collection from children and young people with special educational needs or from parents who may have special educational needs themselves. However, all participants could see the benefits of interviews as the main data collection strategy.

There was diversity in where participants felt interviews should take place, with some preferring home, some online and some a university campus, but all were happy to participate in interviews if their choice could be accommodated. Although many parents liked the idea of meeting face to face, they acknowledged 'it's also the hardest thing to organise and it takes the most amount of effort from people as well' (P56).

Beyond interviews, participants showed openness to a broad array of methods. Diary approaches were generally popular as long as participants weren't expected to update their diary on a daily basis: 'If it was like a weekly or like what's been the highs of the week, what's been the lows of the week, then yeah, I could do that.' (P51). However, in spite of this openness there was a significant pushback against diary entries being published, something that also applied (even more strongly) to photo and video-based approaches. 'I'm comfortable with it as long as like my diary entries and photos are not all published in the report or whatever, that would be embarrassing.' (P10)

Participants were open to guided diaries in which they could respond to questions sent by the research team: 'it's always easier to have a sense of what people want to hear from you rather than just a blank sheet of paper, isn't it, let's face it?' (P61). They were also open to digital diaries that both participant and research team could access at any time. 'If it was online perhaps you could access it on your phone so maybe you'd just be sat on a train somewhere and you could just put a few notes down in it' (P56)

Some participants indicated that audio diaries might work better for them.

> I think I'm quite a fan of sort of little video notes and audio notes, that sort of thing. I think finding a pen in this house half the time is difficult … I think embracing modern technology to do it would be a great way forward. (P59)

Most participants indicated that they would be happy to complete diary entries or photo elicitation tasks in between interviews.

Although a handful of parents were also open to the idea of videoing interactions between them and their child as a form of data collection, others disliked 'the video thing' (P10).

Finally, participants reported being open to arts based approaches, noting that this was a new and novel concept for them 'because no-one's ever offered it as an option, I guess' (P61). One of the benefits parents foresaw from this type of approach was being able to keep their child's artwork, sometimes for reasons specifically related to their diagnosis.

> with Edwards' syndrome, people are always very conscious of wanting to have physical memories in case things go wrong, and I think people are... very up for having footprints and fingerprints and any kind of art that involves something to do with the child as a part of ... the tapestry of their journey. (P62)

However, for some parents, creative, artwork approaches were 'probably not for me' (P59).

> If you said to me, "Would you like to do some art work with your child?" I would say, "How the hell am I going to fit that in?" And actually, the clearing up afterwards would be another three hours. So, no, thank you very much. I would prefer not to do that. (P14)

Again, this attests to the importance of choice in data collection methods.

Theme 5: How to motivate families of learning disabled children to stay in a longitudinal qualitative study

Understanding what would keep parents interested and motivated to stay in a longitudinal study was a key aspect of the current research.

Belonging

A key motivator for parents taking part in a longitudinal qualitative study was to feel a sense of belonging. Participants reported that developing a strong and positive relationship with the

research team is: ‘really, really important’ (P14). They suggested a range of ways to develop rapport, including: ‘just that little friendliness beforehand’ (P56)

Participants acknowledged that it would be impractical to assume they could speak to the same researcher for every interview and were happy to ‘build a relationship with a cohort of researchers’ (P14) as long as changes were kept to a minimum and they could be introduced to all potential interviewers through a closed social media group.

There was also some interest in forming a community of participants who could share experiences of navigating their children’s special educational needs and disabilities. There was openness to this group meeting online but also some interest in occasionally meeting in person.

> I think it's a mixture of both. I think some people will have the time to go and do the social eventy bits and it will bind them into the study, and others just won't have the time or the inclination to. (P14)

Whatever the extent of their involvement in a participant community, participants felt it would enhance their sense of belonging to the study and that this would benefit retention.

Incentives

Parents reported that receiving regular updates on the difference the research was making would be a highly motivating incentive to stay in the study over time. This could take the form of regular newsletters or social media updates. Indeed, knowing they were making a difference, and ‘the feeling of being part of something bigger’ (P14) was considered to be the most motivating incentive.

> I think what would keep me the most motivated is to know that there is some output happening and... or that there is going to be a good ending ... and I’d just like to be kept updated about that, about, you know, what it’s achieving and how it can be used.

> And I'd really like it to inform like government policy ideally, but I know that's just wishful thinking (P10)

Some participants also liked the idea of communications such as birthday cards for their children, to remind them that they were also part of something important. Some also hoped that researchers would be able to *'*signpost you to more local support*'* (P14) or set up events that would offer access to specialist support. For example:

> Nearly all parents that I know that have a child with Down Syndrome, the biggest need they struggle for is speech and language help, speech and language therapists … I think that would be a massive incentive to people, especially if you were going to organise perhaps a coffee morning and you were trying to get, you know, certain groups together. You know, "Come along for two hours, there's going to be play for your children, it's a relaxed environment, there's coffee, there's some cake, and there'll be a speech and language therapist to offer some professional advice," that I think would really get people motivated because they're getting then a benefit for their child that they can't access easily (P56)

As mentioned previously, some parents also felt incentivised by the idea that sharing data - especially if the team shared it back with them - was an opportunity to create a meaningful memento of their child's development over time.

> You know, that kind of like we're going to create a memory book for you and that's going to include your pictures and your artwork … that kind of idea would be the real kind of attraction for me taking part in it long term, that I was going to get some kind of, yeah, some kind of book at the end of it … That would be the most precious … thing. (P62)

Financial Incentives

Participants were most strongly motivated to continue participation by the knowledge that they would be making a difference. However, they were also open to financial incentives and, for some, these were very important: *'*it's that nod and acknowledgement that your time is precious*'* (P57). Parents also acknowledged that incentives could encourage participants to respond promptly

> For me it's a nice to have and I think if you're trying to encourage people to do it, the way I'd probably do it is you would get an incentive if you submitted it by a certain time. So, you got the response back when you wanted it. (P51)

Some acknowledged the impact of the current cost of living crisis, rendering payments more important for some than others. Some participants sought reassurance that accepting small financial or other research incentives would not impact their payments and emphasised the need to be clear about this.

Parents were also asked about the best ways of compensating their child for taking part in the research which, over time, would seek to include their independent voices. Parents suggested that compensation should be meaningful, immediate and tailored to their child's needs and developmental stages. Suggested ideas ranged from chocolate and sweets to vouchers for takeout food or social activities. Including children and young people's voices is a key priority for this research and is being explored in a separate strand of this study.

Theme 6: Ethics and data management

Parents were asked how they felt about research ethics, including open research practices and data management. Participants reported that they would trust the original research team to protect their data because of the ethical processes some of them knew to be in place in research institutions.

> I would assume if you've got ethical approval to run it that it would be quite firmly bolted down and what you could and couldn't do would be quite rigid, so I'd probably be reassured as a lay person that that was all going to happen. But personally, I'd be reassured if you'd got sort of university oversight that that was going to happen anyway. (P62)

However, parents were more worried about open research practices and what they would mean for them and their children.

How we feel about Open Science

Concerns about Open Science, which were widespread in our sample, often related to the qualitative nature of the data being collected.

> I just think it's really personal data. It's not numbers, it's not stats. It's personal data and I think it should be protected a little bit more than just being open. (P10)

Participants were keen to maintain control of how their family's data would be used, and by whom. For example, they expressed concerns about their data being used for commercial purposes and being used internationally without their explicit consent: 'It would depend on what they want to use it for' (P51).

Some participants were reasonably relaxed about some data being shared openly.

> if it's just, you know, 'Participant A says this and feels like this and Child A has developed like this' then it's totally anonymous and that's fine. And I feel if you're going to go to all the trouble to take part in this research then the more people it can benefit, the better. You know, if it can benefit people in a different country then let's help as many people as we can. So yeah, I would be happy for it to be in an open research, it just would need to be kind of managed in the right way (P56)

It was when talking about potentially identifying data that their concerns kicked in. Participants were particularly against visual data, such as photos or videos being made openly available: 'I don't want my photos going all over the place thank you very much' (P14). This was largely driven by fears around the challenge involved in making them truly anonymous: 'The more personal the data, obviously the harder it is to de-identify the people in it' (P62).

Some participants concluded that they would want to have the opportunity to provide consent for their data to be used in any study other than the one they signed up for.

> I think I might want to be given the opportunity to give my permission separately for any kind of secondary research, just because there are some purposes that I might not be as comfortable with, so I'd probably just want to know what the study was for and

> have the opportunity to give permission ... I think it feels more respectful to be able to give my permission again, so yeah (P62).

Overall, the general feeling was that fully open research practices felt too risky for a study such as this.

Concerns about our data

One of the main concerns parents reported was their data not being handled or stored correctly, which could lead to data breaches. Again, they showed particular concerns around identifying visual data: 'I would only be concerned if people had images of [name] and perhaps they could fall into the wrong hands' (P56).

Further concerns were raised about the long-term consent implications of keeping data about children and what this would mean for them. This is a particularly sensitive issue with learning disabled children who may have a different understanding of consent/assent than their parents, which may also differ from the understanding of non-learning disabled children: 'I guess the other thing is just around privacy information … I'm not so bothered about me, but sort of long term from my child's perspective' (P61)

In summary, most parents had no problem with sharing data with the original research team, but raised concerns about a fully open approach to research, due to the sensitive nature of their data and the vulnerability of their children, particularly if the data was made up of photographs and videos.

How to support us through the process

Participants discussed several issues around informed consent and how to ensure the process was a reliable one. They argued that gaining informed consent has to involve more than

ticking a box to say yes to something and that it should be a continual process in a longitudinal study.

> the consent taking process isn't necessarily just about signing a form, it's about talking through the form and taking the consent…And maybe revisiting that consent as well, you know, through the course of the study, because studies do change (P57)

Another aspect of discussion was around the children themselves taking part in the research. Participants questioned how researchers would know that their children truly understood what was being asked of them:

> Just I guess the obvious ones that they have to have an awareness of what they're talking about and I'm not sure they do. [name] doesn't, so I don't know how the researchers will go about gaining consent from them, if that makes sense, because I don't even know how you'd do that (P10)

Participants did not identify answers to this but were concerned about it as an ethical issue.

Discussion

Our twelve parents of children and young people with chromosomal learning disabilities were unanimously enthusiastic about the prospect of a qualitative longitudinal study of seldom-heard families, including families with experience of special educational needs and disabilities. They were able to clearly articulate the benefit of their voices, and those of their children, being amplified, including the opportunity to contribute to their children's special educational needs being more effectively met by schools and other services.

Participants' responses strongly endorsed the methodological guidance that already exists in the qualitative longitudinal research literature (Neale, 2021). In relation to recruitment and retention, for instance, they expressed a preference for being recruited through trusted local groups (van Wijk, 2014) and for researcher stability over time (e.g. (Shirani, 2010). It seems clear that a good understanding of local organisations and the recruitment of researchers with

a commitment to long service, as far as that is possible, is integral to doing this well. They also told us that their key motivation for joining and staying in a study like this would be to make things better for those coming up behind them (Shirani, 2010) but that financial incentives would be important to some families too. Other types of reciprocity, such as access to expert advice or the sharing back of data collected over time, were also important and the latter may be particularly important to families of children with life limiting conditions.

It is clear from our data that participants have strong views on some of the questions they would like research to address, and this supports the importance of co-producing a research agenda so it meets the needs of participating families as well as society more broadly. EHCPs and the transition out of education were particular priorities. The importance of co-production, and understanding participants' needs and constraints, was also clear in their discussion of data collection approaches. They expressed clear support for a toolbox approach, as suggested by the literature (e.g. (Plummer, 2001), on the basis that a 'one size fits all' approach would not work for them or, eventually, for their children. They also echoed the literature in emphasising the importance of viewing ethics and consent as an ongoing process (Neale, 2013).

All of this fed into a series of recommendations to the ESRC, on how to design and deliver a successful longitudinal qualitative cohort study of seldom-heard families, including families with learning disabled children. These recommendations were also informed by other strands of the project and can be seen in Table 2.

Table 2: Recommendations to the ESRC for the design and delivery of a qualitative longitudinal cohort study of seldom-heard families, including those with experience of learning disability

1. Take a local, community focused approach (drawing on advice around recruitment through trusted groups and developing sustainable relationships)
2. Recruit diverse types of seldom-heard family, paying particular attention to intersectionality and levels of marginalisation
3. Develop and maintain a communications and impact strategy that will support retention (addressing participants' desire to know they are making a difference)
4. Take a new approach to the recruitment, retention and development of researchers (to support the development of sustainable and trusted relationships)
5. Take a flexible, personalised, rigorous and ethical approach to data collection
6. Co-produce a clear and explicitly articulated plan for data sharing and safeguarding (to address concerns around open research practices)
7. Develop a rigorous plan for ensuring and supporting both consent and assent (including for children who communicate in non-traditional ways)

In order to conduct research like this well it will also be important to focus on developing inclusive and evidence-informed approaches to the learning-disabled children themselves, including those who communicate in non-traditional ways, and to learning disabled adults so that their voices are also clearly heard in research that affects them.

The most significant limitation of the research presented here is that it is unlikely to have captured the most seldom-heard families of children with chromosomal learning disabilities. It is also potentially a limitation that we have used a cross-sectional design to make recommendations for longitudinal research. It would be interesting to return to these participants at a later date to gain a more experiential perspective on participating in longitudinal qualitative research.

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