Children in Scotland requiring Palliative Care: identifying numbers and needs (The ChiSP Study)

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The **Managed Service Network for Children and Young People with Cancer** (MSNCYPC), through **Children's Hospice Association Scotland** (CHAS), commissioned this study. The views expressed are the authors' and do not necessarily reflect those of MSNCYPC and CHAS.

Further copies of this report can be obtained from Children's Hospice Association Scotland (www.chas.org.uk)

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Executive Summary

This report sets out the findings from an investigation into the numbers of children and young people with life-limiting conditions in Scotland, and what current evidence tells us about their, and their families', psychosocial support needs.

The overall purpose of the study was to develop an evidence base to support and inform planning for children's palliative care in Scotland. It is hoped that the evidence generated will be a resource to organisations with responsibility for, or delivering services to, children and young people with life-limiting conditions.

The objectives of the research were as follows:

- to identify the number of children and young people with life-limiting or life-threatening conditions in Scotland;
- to describe this population in terms of their ages, conditions/diagnoses, geographic locations and ethnicity;
- to generate evidence on their psychosocial care needs.

The study comprised two workstreams. First, and achieved by a complex process of linking national administrative and health services datasets, an analysis of population level data in order to describe the numbers and characteristics of children and young people with life-limiting or life-threatening conditions in Scotland. Second, a detailed review of existing international evidence on children and young people's, parents' and siblings' accounts of living with a life-limiting or life-threatening condition, and their perceived psychosocial support needs.

Based on the findings, the research team has made ten recommendations. Five arise from the evidence concerning the numbers of children and young people in Scotland who have a life-limiting condition. The remainder are based on the findings from the review of existing evidence. The recommendations are:

RECOMMENDATION 1: More children and young people of ALL AGES in Scotland with life-limiting conditions should have input from palliative care services.

RECOMMENDATION 2: Children under 1 year of age should be seen as a priority group for input from palliative care services.

RECOMMENDATION 3: Age specific palliative care services for young people (aged 16-25 years) with a life-limiting condition in Scotland should be developed.

RECOMMENDATION 4: Palliative care services should be able to provide culturally competent care to children and young people from ALL ethnic groups.

RECOMMENDATION 5: Future development of palliative care services in Scotland should ensure that access to services for children and young people from areas of high deprivation is prioritised.

RECOMMENDATION 6: Specialist psychological and emotional care should be available to children and young people and all family members. Included in this should be support for couples, and support and guidance to parents as they respond to the emotional needs of their children. Services caring for children and young people with life-limiting conditions should also find ways to provide opportunities for all family members to connect with and spend time with their 'peers'.

RECOMMENDATION 7: Palliative care services should consider incorporating domestic support to families within their portfolio of services. They should also seek ways to extend provision of support to parents in order to reduce, or provide a break from, the caring demands and responsibilities placed on parents.

RECOMMENDATION 8: Palliative care services in Scotland should continue to include specialist residential provision. This provision should be extended in light of evidence of the size of the population of children with life-limiting conditions living in Scotland.

RECOMMENDATION 9: Palliative care services should contribute to, or support activities which seek to, educate and challenge societal perceptions of impairment and disability.

RECOMMENDATION 10: Palliative care services, and other services involved in providing care at end of life, should examine current practices with respect to evidence on parents' psychosocial needs **as parents** at their child's end of life.

Paul Hampton

SECTION 1

Background and Context

Although many of the individual diagnoses are rare, as a group, children and young people with a life-limiting condition[1] are a larger patient population than many other long term conditions in children and young people (e.g. diabetes mellitus (Royal College of Paediatrics and Child Health, 2014)). Previous work has shown that the prevalence of children and young people with a life-limiting condition in England is both rising and higher than previously estimated (Fraser et al., 2011, Fraser et al., 2012).

[1]**Definition of terms**

Life-limiting conditions are those for which there is no reasonable hope of cure and from which children or young people will ultimately die prematurely, e.g., Duchenne muscular dystrophy or neurodegenerative disease.

Life-threatening conditions are those for which curative treatment may be feasible but can fail, e.g. cancer (Together for Short Lives, 2015).

The term, 'life-limiting condition' will be used throughout this report to include both life-limiting and life-threatening conditions.

The ChiSP Study

The ChiSP study (Children in Scotland requiring Palliative Care: identifying numbers and needs) was commissioned in order to provide an evidence base to inform the future planning and design of paediatric palliative care services in Scotland. It focused specifically on two areas of evidence: the numbers of children and young people with life-limiting conditions living in Scotland; and family members' experiences and views regarding their psychosocial support needs.

The study comprised two workstreams. Workstream 1 comprised secondary analysis of routinely collected healthcare and administrative data in order to describe the size of the population, as well as providing more detailed evidence on the characteristics of that population, including the stage of their condition. Workstream 2 was a review of existing research which has explored, using methods such as interviews and focus groups, children and young people's, parents' and siblings' views of their psychosocial support needs. Details of the methods and datasets used by the workstreams are provided in the Appendices.

The results (and their implications) from these two workstreams have resulted in ten recommendations regarding services for children and young people with life-limiting conditions and their families. The recommendations are concerned with both supporting developments in services to ensure all families who may benefit from palliative care have access to such services, and the 'content' of such services – particularly meeting families' psychosocial care and support needs.

The following sections of the report present these recommendations within the context of the data, or findings, which informed them. Section 2 presents the evidence and recommendations arising from the epidemiological work (workstream 1). Section 3 sets out the recommendations regarding psychosocial care and support to families, again nesting these within the evidence from which they were generated.

However, before moving on to present the recommendations, and the evidence underlying them, it is helpful to provide some information about the current context regarding paediatric palliative care in Scotland.

Paediatric Palliative Care in Scotland: the current context

In 2012, the Scottish Children and Young People's Palliative Care Executive (SCYPPEx) published a framework for the delivery of palliative care for children and young people in Scotland. The definition of palliative care used in this report is: "..... an active and total approach to care, from the point of diagnosis or recognition, embracing physical, emotional, social and spiritual elements through to death and beyond. It focuses on enhancement of quality of life for the child/young person and support for the family and includes the management of distressing symptoms, provision of short breaks and care through death and bereavement" (Scottish Children and Young People's Palliative Care Executive Group, 2012 Page 2). This framework aims to ensure that there are recognised pathways for palliative care for every child and young person from the point of diagnosis of a life-limiting condition. The current provision of specialist paediatric palliative care services for children and young people in Scotland is shown in box below[2].

Looking forwards, a report from the Scottish Public Health Network (ScotPHN) – Palliative and End of Life Care in Scotland: The Rationale for a Public Health Approach – will be published in late 2015. Additionally, the findings and recommendations from the Scottish Parliament's enquiry into Palliative Care, as investigated by its Health and Sport Committee, are expected to acknowledge the importance of palliative care for all ages, including babies, children and young people. The Committee was made aware that the ChiSP study will also report in 2015, offering new information on the numbers and specific palliative care needs of individuals 0-25 years, as well as those of their families.

[2]**Specialist paediatric palliative care services and posts in Scotland: the current picture**

Dr Pat Carragher, Medical Director to Children's Hospice Association Scotland

Children's palliative care is part of good medical care, and as such, it is delivered across primary, secondary and tertiary care, as well as in children's hospices, but currently most end of life care is delivered in hospital environments.

In Scotland, within the NHS there are relatively few care professionals who have children's palliative care as a substantive part of their posts or who have undergone specialist post graduate training. Exceptions include NHS Arran and Ayrshire Health Board where there is a WellChild nursing team (with some specific palliative role in children with complex needs) and paediatrician with specialist qualifications and an interest in paediatric palliative care. In addition, 2015 saw a consultant neonatologist commencing work in a joint post between Children's Hospice Association Scotland (CHAS) and NHS Lothian. A second joint consultant post is being developed between NHS Greater Glasgow and Clyde and CHAS, with the post holder having undertaken specialist training in oncology and haematology.

The Managed Service Network for Children and Young People with Cancer (MSNCYPC) employs a Consultant Paediatric Oncologist. This post specifically includes a palliative component (~1 day/week), and includes the remit of seeking to improve paediatric palliative care for children and young people across Scotland (primarily) with oncology and haematology diagnoses. Paediatric Palliative Outreach Nurses work specifically with children and young people with these same diagnoses during diagnosis, active treatment, palliative care and end of life care phases of children's and young people's lives.

Children's hospice services were first introduced in Scotland in 1996 when Rachel House opened. (Before this some parents took their children as far as Martin House in West Yorkshire to obtain specialist palliative care). CHAS now provides hospice services for babies, children and young people across all health board areas in Scotland. Holistic care is provided through its two hospices (Rachel House, and Robin House in Balloch), and via CHAS at Home. It is delivered by a multidisciplinary team including doctors, nurses, social workers, pharmacist, family support workers, as well as a full administrative team in order to provide 24/7 support of its services.

CHAS at Home has teams working from Inverness, Aberdeen, Kinross and Balloch to provide care in families' homes. When required, they can also augment care in hospital. In 2014, a team of Diana Children's Nurses was appointed to work across Scotland, with nurses working within NHS teams but managed and supported by CHAS. The aim of this nursing team is to further develop palliative care services and support for children, as well as their families, and to help improve choices in care. The team has both generic and specialist expertise with one nurse having a specific remit in intensive care and oncology, another in neonatology, and the other in community liaison.

In terms of transition to adult care there are different models across Scotland, but on a pan-Scotland basis, the SCYPPEx Framework states that "individual health boards should develop local pathways for transition of young people with palliative care needs to appropriate adult services" (Scottish Children's and Young People's Palliative Care Executive Group (2012, p23). Some specialities are more developed, as in oncology where the Cancer Plan for Children and Young People in Scotland 2012-15 (MSNCYPC) aims to offer seamless care for individuals up to the age of 25 years. This is based on strong cooperation between children's and adult services, and is aided by the provision of facilities by the Teenage Cancer Trust.

SECTION 2

The numbers and characteristics of children and young people with life-limiting conditions in Scotland: findings and recommendations

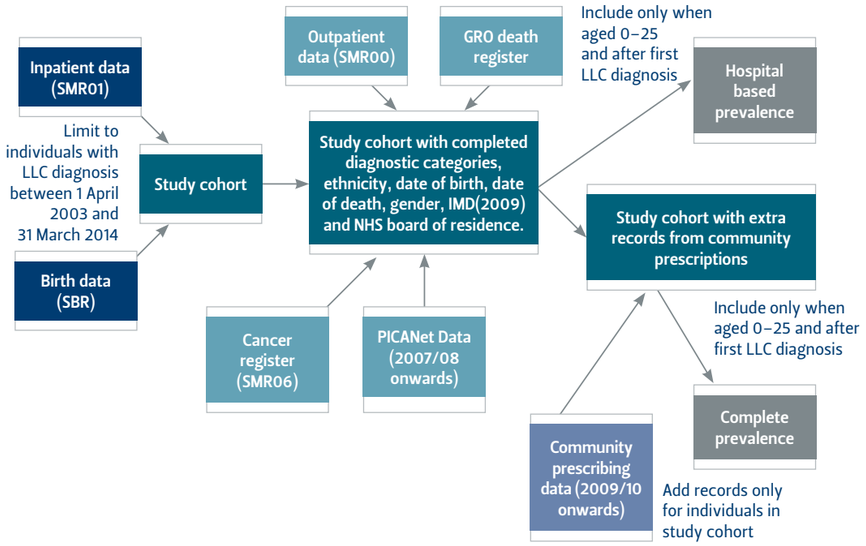
Introduction

This section sets out the recommendations, and supporting evidence, regarding the numbers and characteristics of children and young people with life-limiting conditions living in Scotland. Secondary analysis of routinely collected healthcare and administrative data was used to identify and describe the size of the population, as well as providing more detailed evidence on the characteristics of that population, including the stage of their condition.

Cohort identification

A previously developed (Fraser et al., 2011, Fraser et al., 2012) list of ICD10 (International Classification of Diseases, 10th edition (World Health Organisation, 1992)) codes that constituted the conditions of interest was utilised to identify individuals with a life-limiting condition within routinely collected NHS data (Figure 1). The study cohort was identified by searching the Scottish Morbidity Record Inpatient activity database (SMR01) and Scottish Birth Records (SBR) for individuals resident in Scotland, aged 0-25 years, with any of the included ICD10 codes listed in any record from 1 April 2003 to 30 March 2014. Linkage to the other healthcare data for these individuals was then undertaken.

FIGURE 1: Data sources used in the study. Dark blue boxes indicate sources of records used to identify individuals with a life-limiting condition; light blue box indicates source of records used to indicate presence of identified individuals in Scotland for complete prevalence analyses; light teal boxes indicate sources of demographic and date of death information; dark teal boxes indicate intermediate stages of combined datasets and grey boxes indicate final datasets used to determine prevalence figures..



Prevalence analyses

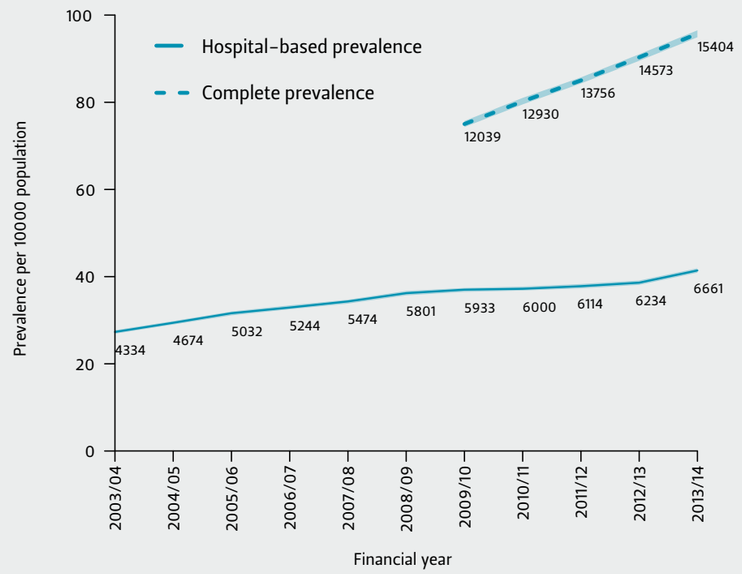
Prevalence is a measure of the number of individuals with a given condition in a population at a given time (each financial year in this study).

$$prevalence = \frac{number\ of\ individuals\ with\ a\ LLC}{population\ at\ risk} \times 10000$$

Two prevalence figures were used in this study. Firstly 'hospital-based prevalence' (Fraser et al., 2012) in which individuals were counted in a given year if they had a life-limiting condition and any inpatient hospital episode (SBR or SMR01) in that year while aged 0-25 years. Under the second criteria (complete prevalence), all individuals counted using the first set of criteria were included and, in addition, individuals were counted if they appeared in the community prescribing records in that year (this indicated that they were still alive and resident in Scotland). As community prescribing data were only available from 2009 onwards, the complete prevalence analyses were limited to financial years 2009/2010 to 2013/2014.

Detailed methodology is available in Appendix 1.

FIGURE 2: Hospital-based and complete prevalence of life-limiting conditions among children and young people (aged 0-25 years) in Scotland for financial years 2003/4 – 2013/14. Shaded regions indicate 95% confidence interval.



Recommendation 1:

More children and young people of ALL AGES in Scotland with life-limiting conditions should have input from palliative care services

The evidence on prevalence

The prevalence of children and young people (aged 0–25 years) with a life-limiting condition in Scotland has risen markedly between 2003/4 and 2013/14 (Figure 2). The hospital-based prevalence had risen from 27.3 (95% CI 26.9–27.7) per 10,000 population in 2003/4 to 41.4 (95% CI 40.9–41.9) per 10,000 population in 2013/14 (Table 1, Figure 2). The complete prevalence estimates also includes children and young people with a life-limiting condition who are still alive and resident in Scotland but who had not received inpatient care in that year. These data also show a rise from a prevalence of from 75.0 (95% CI 74.3–75.7) per 10,000 in 2009/10 to 95.7 (95% CI 94.9–96.5) per 10,000 in 2013/14.

The absolute numbers of children and young people with a life-limiting condition in the hospital based estimates have risen from 4,334 (2003/4) to 6,661(2013/14). The absolute numbers of children and young people with a life-limiting condition in the complete estimates have risen from 12,039 (2009/10) to 15,404 (2013/14).

Age

Prevalence decreased by age group with the highest prevalence in the under 1 age group with hospital-based prevalence 163.0 (95% CI 157.7–168.3) per 10000 population and complete prevalence 192.1 (95% CI 186.3–197.8) per 10000 population in 2013/14 (Figure 3).

FIGURE 3: Hospital-based and complete prevalence of life-limiting conditions by age group among children and young people in Scotland for financial years 2003/4 – 2013/14. Shaded regions indicate 95% confidence interval.

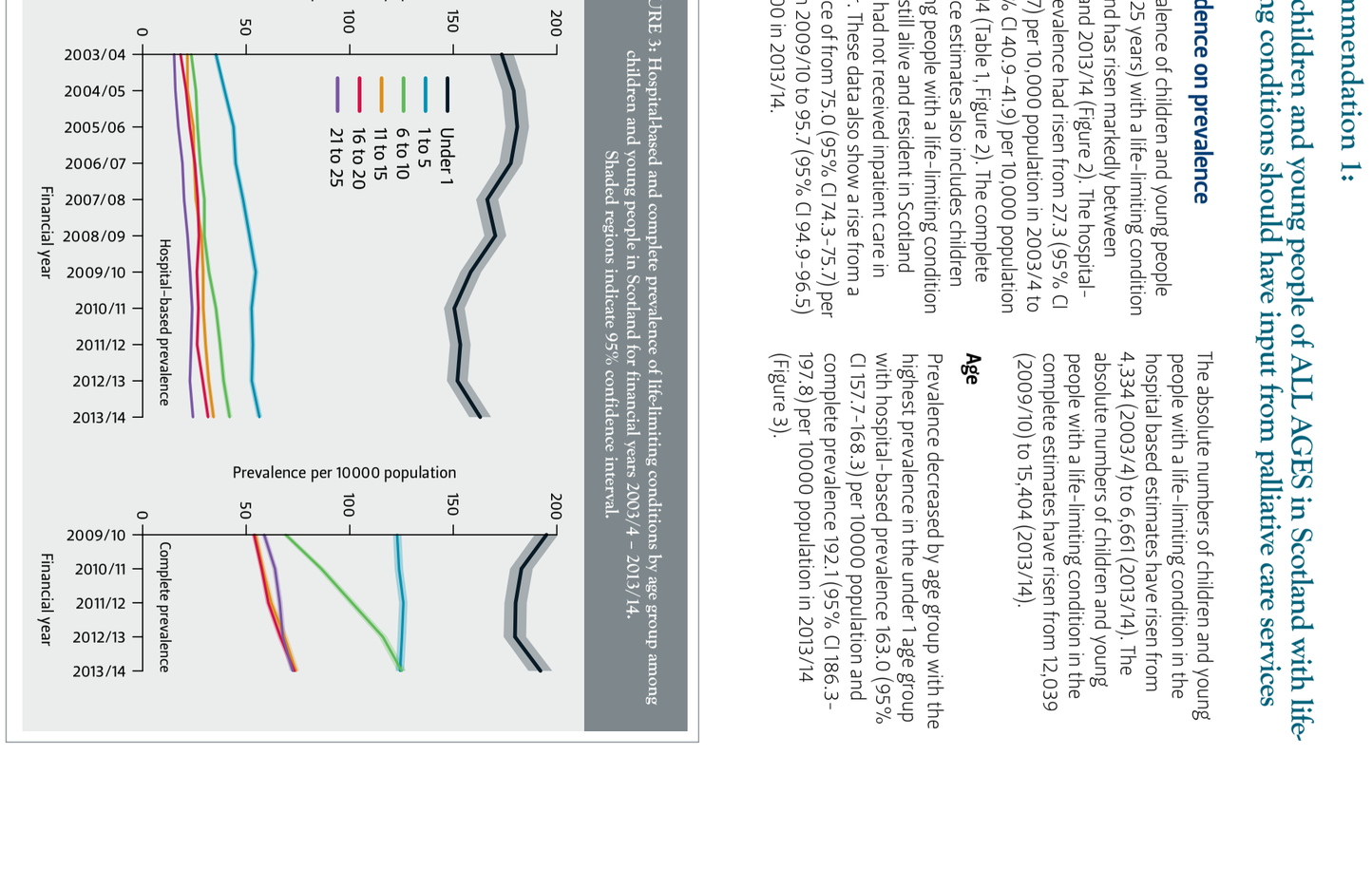


TABLE 1: Overall number of children and young people (aged 0-25) with life-limiting conditions in Scotland and hospital-based and complete prevalence per 10000 population by age group for financial years 2003/2004 – 2013/2014.

FINANCIAL YEAR	NUMBER OF PERSONS AGED 0–25 WITH A LLC	PREVALENCE OF LIFE LIMITING CONDITIONS PER 10000 POPULATION																				
		AGE 0–25	95% CI		AGE < 1	95% CI		AGE 1–5	95% CI		AGE 6–10	95% CI		AGE 11–15	95% CI		AGE 16–20	95% CI		AGE 21–25	95% CI	
HOSPITAL-BASED PREVALENCE																						
2003/2004	4334	27.3	26.9	27.7	173.4	167.7	179.2	35.4	34.2	36.5	23.5	22.6	24.3	21.6	20.8	22.4	18.3	17.6	19.1	15.2	14.5	15.9
2004/2005	4674	29.4	29.0	29.8	179.2	173.5	184.9	39.6	38.3	40.8	25.7	24.8	26.6	21.7	20.9	22.5	21.0	20.2	21.8	15.8	15.2	16.5
2005/2006	5032	31.6	31.1	32.0	180.9	175.2	186.6	43.9	42.6	45.2	26.5	25.6	27.5	24.5	23.6	25.3	22.8	22.0	23.6	17.2	16.5	18.0
2006/2007	5244	32.9	32.5	33.4	177.8	172.2	183.4	44.9	43.6	46.2	27.8	26.8	28.8	24.9	24.0	25.8	25.2	24.3	26.1	19.2	18.5	20.0
2007/2008	5474	34.3	33.8	34.7	166.4	161.0	171.7	48.5	47.1	49.8	29.8	28.8	30.8	25.7	24.8	26.6	26.6	25.7	27.5	20.0	19.3	20.8
2008/2009	5801	36.2	35.7	36.7	170.3	165.0	175.6	51.6	50.3	53.0	29.8	28.7	30.8	28.4	27.4	29.4	27.2	26.3	28.1	21.7	20.9	22.5
2009/2010	5933	37.0	36.5	37.4	158.4	153.3	163.5	54.6	53.2	56.0	32.0	31.0	33.1	29.2	28.2	30.2	26.2	25.3	27.1	22.9	22.1	23.7
2010/2011	6000	37.2	36.8	37.7	150.5	145.5	155.5	52.6	51.2	53.9	35.4	34.3	36.5	29.3	28.3	30.2	26.9	26.0	27.8	23.9	23.1	24.7
2011/2012	6114	37.8	37.3	38.3	153.4	148.4	158.4	53.2	51.8	54.5	37.4	36.3	38.6	30.4	29.4	31.4	26.3	25.4	27.2	23.4	22.6	24.2
2012/2013	6234	38.6	38.1	39.1	152.0	146.9	157.0	52.7	51.4	54.1	39.1	37.9	40.3	31.9	30.8	32.9	29.0	28.1	30.0	22.8	22.1	23.6
2013/2014	6661	41.4	40.9	41.9	163.0	157.7	168.3	56.4	55.0	57.8	42.0	40.8	43.3	34.2	33.1	35.3	31.6	30.6	32.6	24.3	23.5	25.1
COMPLETE PREVALENCE																						
2009/2010	12039	75.0	74.3	75.7	195.0	189.3	200.6	122.9	120.9	125.0	69.1	67.5	70.7	54.5	53.2	55.8	53.8	52.6	55.1	58.7	57.4	60.0
2010/2011	12930	80.2	79.5	81.0	182.9	177.4	188.4	123.8	121.7	125.8	86.3	84.5	88.1	58.0	56.6	59.4	57.5	56.3	58.8	63.9	62.6	65.2
2011/2012	13756	85.0	84.3	85.7	180.1	174.7	185.5	125.9	123.8	127.9	101.1	99.2	103.1	62.1	60.7	63.5	60.7	59.4	62.0	66.2	64.9	67.6
2012/2013	14573	90.3	89.5	91.0	179.8	174.3	185.2	125.2	123.1	127.2	116.0	114.0	118.1	68.5	67.0	70.1	66.5	65.1	67.9	67.7	66.4	69.1
2013/2014	15404	95.7	94.9	96.5	192.1	186.3	197.8	124.3	122.2	126.3	125.1	122.9	127.2	74.0	72.4	75.6	73.3	71.8	74.8	72.5	71.1	73.9

Prevalence in the under 1 age group was steady over the period but prevalence increased for all other age groups over the period from 2003/4 to 2013/14.

Gender

Prevalence was significantly higher for males than for females in all years in both the hospital-based prevalence estimates and the complete prevalence estimates (e.g. in 2013/14 hospital-based prevalence for males was 45.1 (95% CI 44.4-45.8) per 10000 population compared to 37.5 (95% CI 36.8-38.2) for females; complete prevalence was 102.0 (95% CI 100.9-103.1) per 10000 population for males compared to 89.2 (95% CI 88.2-90.3) for females (Figure 4)).

Diagnoses

The diagnostic group with the highest prevalence was congenital anomalies followed by oncology and neurology diagnoses (2013/14 hospital-based prevalence per 10000 population: congenital 15.5 (95% CI 15.2-15.9), oncology 6.3 (95% CI 6.1-6.5), neurology 6.6 (95% CI 6.4-6.8); 2013/14 complete prevalence per 10000 population: congenital 34.3 (95% CI 33.8-34.7), oncology 13.8 (95% CI 13.5-14.1), neurology 12.7 (95% CI 12.4-13.0) (Figure 5)).

Health Board

The prevalence of children and young people with life-limiting conditions is rising in all the Health Boards in Scotland over the period of this study (Figure 6). The findings of this report apply to all Health Board areas however, due to small numbers and therefore fluctuating prevalence, data from the Western Isles, Orkney and Shetland Health Boards could not be displayed in this figure.

The evidence on stage of condition

The four stages of a condition; stable, unstable, deteriorating and dying have been used previously in the independent review of Palliative Care Funding in England (PCFR) (Hughes-Hallet et al., 2011). These stages / phases of illness were the main criteria identified which shaped the palliative care needs of individuals in the PCFR and from which the palliative care currencies were developed. In this study the stages of condition were defined via key transitions in use of health services (Figure 7).

Each year 14-19% of these children and young people with a life-limiting condition were either unstable (12-15%), deteriorating (1.6-2.2%) or dying (1.1-1.6%) (Figure 8).

Over the five year period studied, the proportion of individuals stable for the whole year increased from 80.8% (95% CI 80.5-81.2%; 9729 individuals) to 85.7% (95% CI 85.4-86.0%; 13203 individuals) (Figure 8). The unstable proportion dropped from 15.4% (95% CI 15.1-15.8%; 1857 individuals) to 11.6% (95% CI 11.3%-11.8% 1783 individuals); the deteriorating proportion dropped from 2.2% (95% CI 2.0-2.3%; 262 individuals) to 1.7% (95% CI 1.5-1.8%; 254 individuals); the dying proportion dropped from 1.6% (95% CI 1.5-1.7%; 191 individuals) to 1.1% (95% CI 1.0-1.1%; 164 individuals) (Figure 8).

FIGURE 4: Hospital-based and complete prevalence of life-limiting conditions by gender among children and young people (aged 0-25 years) in Scotland for financial years 2003/4 – 2013/14. Shaded regions indicate 95% confidence interval.

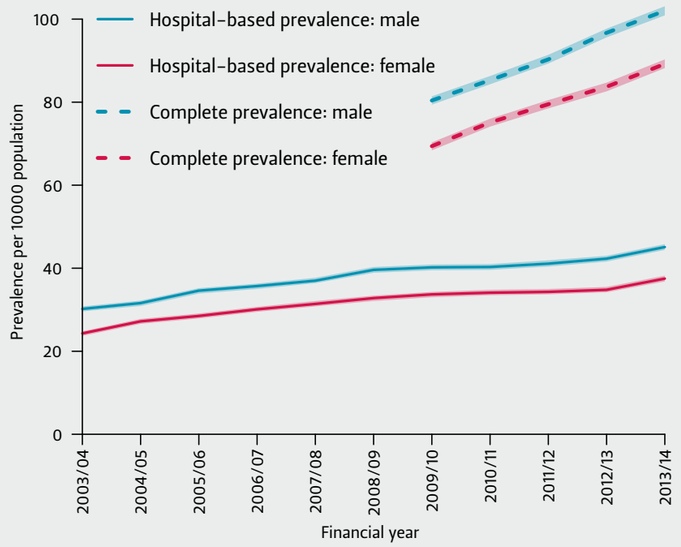
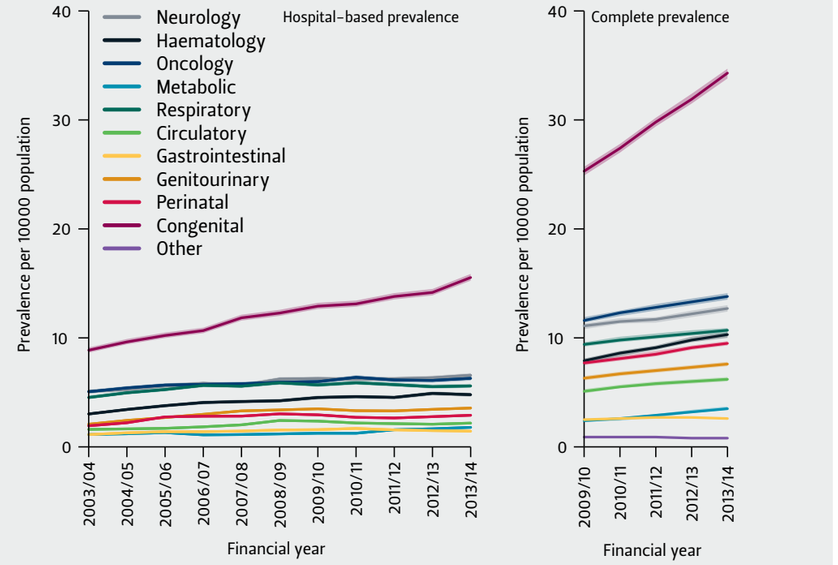


FIGURE 5: Hospital-based and complete prevalence of life-limiting conditions by diagnostic group among children and young people (aged 0-25 years) in Scotland for financial years 2003/4 – 2013/14. Shaded regions indicate 95% confidence interval.



Diagnoses

Those individuals with a perinatal life-limiting condition were most likely to be stable (2013/14: 93.5%), followed by those with congenital (2013/14: 87.6%) and circulatory conditions (2013/14: 89.6%) (Figure 9). Those with gastrointestinal diagnoses were least likely to be stable (2013/14: 69.6%) and were also most likely to be dying (2013/14: 3.4%). Individuals with respiratory conditions were most likely to be deteriorating in most years (2009/10:4.5%).

The evidence on deaths

The average number of deaths in Scotland each year in children and young people with a life-limiting condition was 195 but there was a decline in the number of deaths per year over the study period (from 208 in 2004/5 to 188 in 2012/13). The numbers of deaths per year varied by age group with the largest number in the under 1 year age group (mean 56) followed by the 21-25 year olds (mean 40) and 16-20 years (mean 36) (Figure 10).

FIGURE 6: Hospital-based and complete prevalence of life-limiting conditions by NHS Health Board of residence among children and young people (aged 0-25 years) in Scotland for financial years 2003/2004 – 2013/2014. Orkney, Shetland and Western Isles are not shown due to low numbers

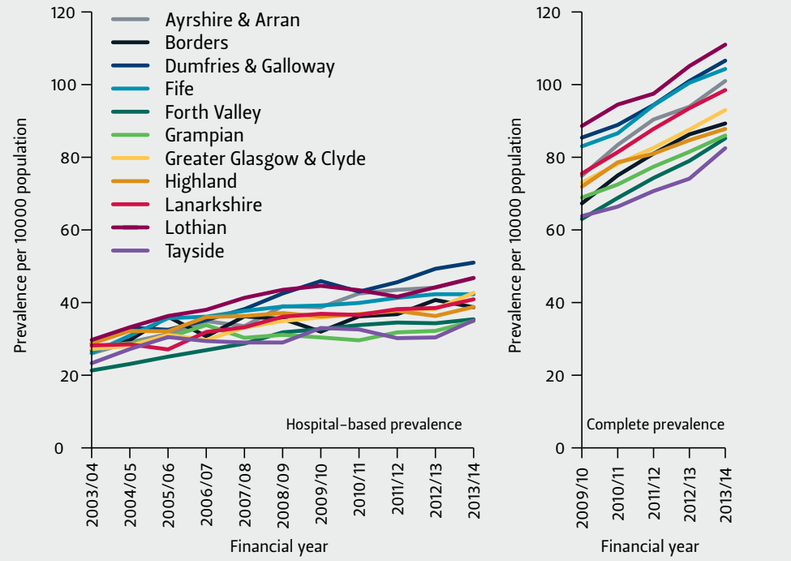


FIGURE 7: Four stages of condition assigned to children and young people with a life-limiting condition and the criteria for transition between stages. Red arrows indicate worsening condition; green arrows indicate improving condition.

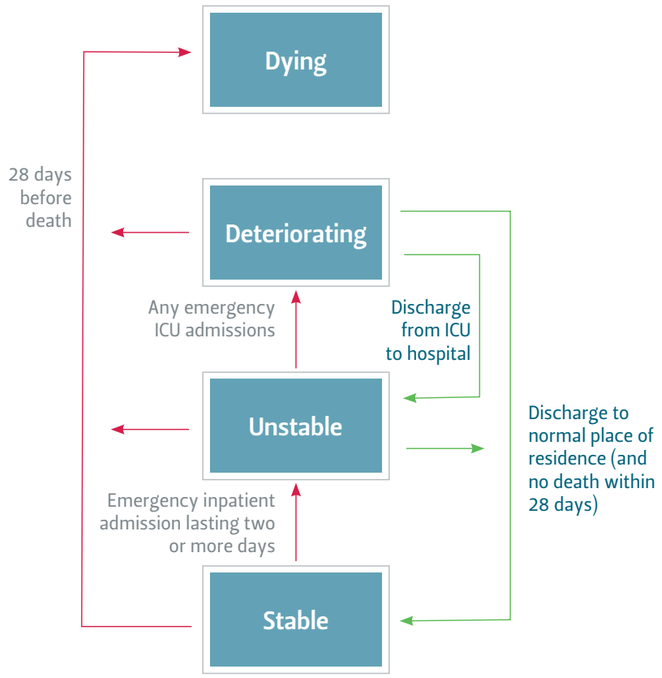
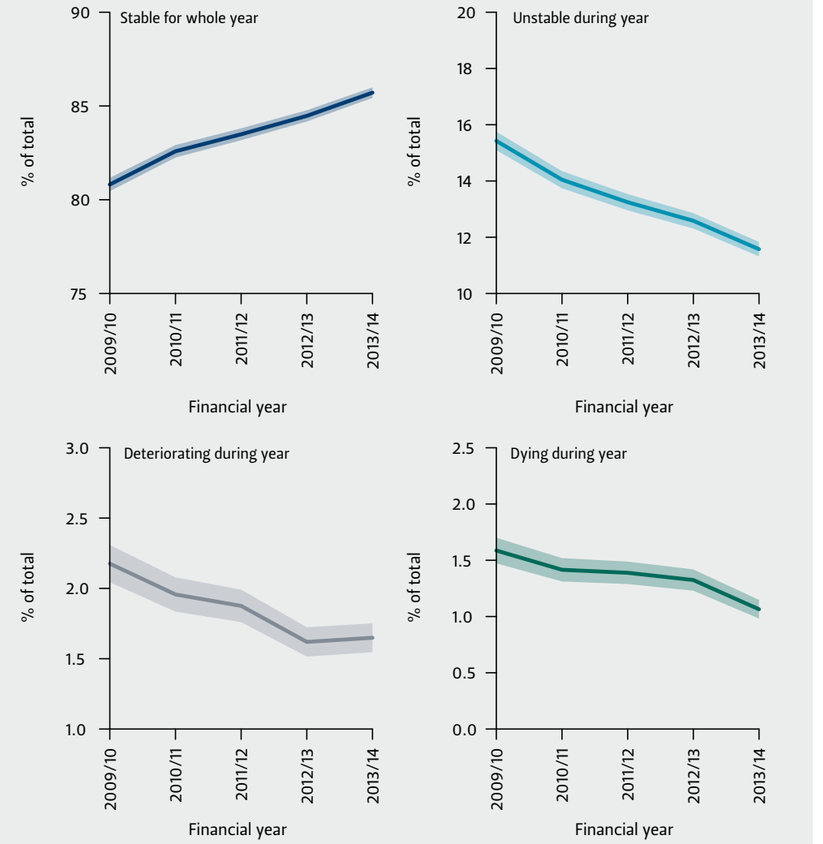


FIGURE 8: Proportions of total children and young people with a life-limiting condition in Scotland at each of four stages of condition in financial years 2009/10 – 2013/14. Shaded regions indicate 95% confidence interval.



Place of death

Overall 73.0 % of deaths were in hospital, 5.6% hospice and 21.4% home. Again variation by age was seen with 90.2% of the under 1 age group dying in hospital (Figure 11).

The increasing prevalence of children and young people with a life-limiting condition in Scotland is similar to the results from previous research from England (Fraser et al., 2012). The lack of increase in the prevalence in the under 1 age group over the period of this study supports the hypothesis that the increase in prevalence is due to increased survival times in these children and young people rather than an increase in incidence. The difference between the hospital-based and complete prevalence estimates highlight that approximately 50% of children and young people with a life-limiting condition in Scotland do not have a hospital admission in each year. It is important to note that the difference between hospital-based and complete estimates is present in all the diagnostic groups and the individuals who do not have a hospital admission varies each year. As the methodology used in this study only required a life-limiting condition to be coded once for each individual it is possible

that these prevalence figures include children and young people who have had a transient life-threatening event but who are no longer life-limited.

The absolute numbers of children and young people (0-25 years) in Scotland with a life-limiting condition in 2013/14 ranged from 6661 (hospital-based estimates) to 15404 (complete estimates). 2201 of these children and young people were unstable/deteriorating or dying during 2013/14. Children's Hospice Association Scotland receives on average around 115 new referrals each year and they care for approximately 380 children and young people and their families each year.

Although paediatric palliative care services should be involved in the care of children and young people with life-limiting conditions early in their disease process, end of life care is an important component of paediatric palliative care services. There were approximately 200 deaths each year in this cohort of children and young people with life-limiting condition. Children's Hospice Association Scotland currently cares for approximately 60 children and young people who die each year.

It is clear that the potential demand for palliative care in the 0-25 year age group outstrips the current provision.

FIGURE 9: Proportions, by diagnostic category, of total children and young people with a life-limiting condition in Scotland at each of four stages of condition in financial years 2009/10 – 2013/14.

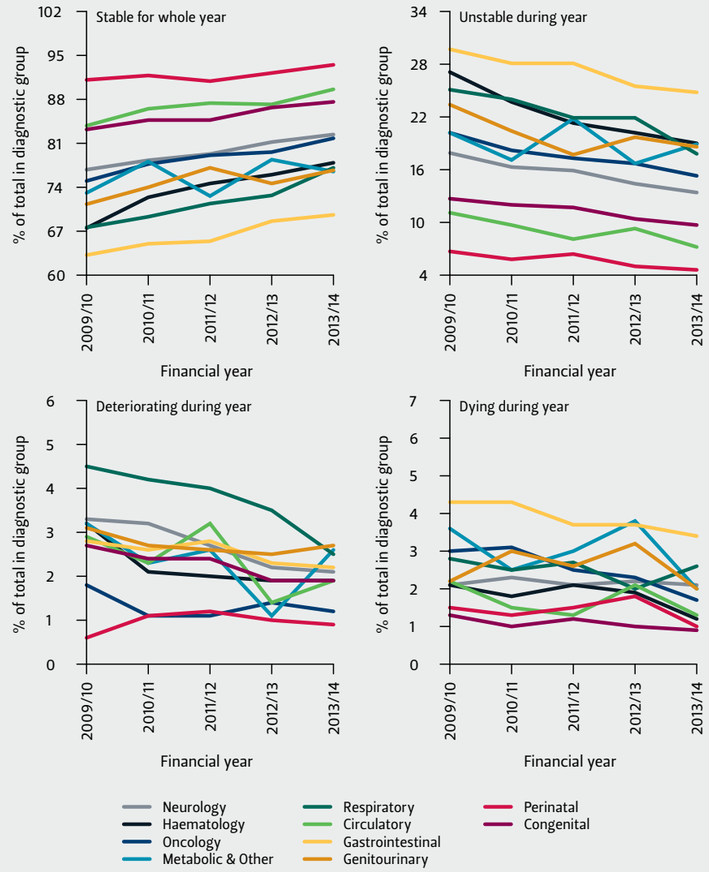


FIGURE 10: Numbers of deaths, by age group, in financial years 2004/5 – 2013/14 among children and young people in Scotland identified as having a life-limiting condition.

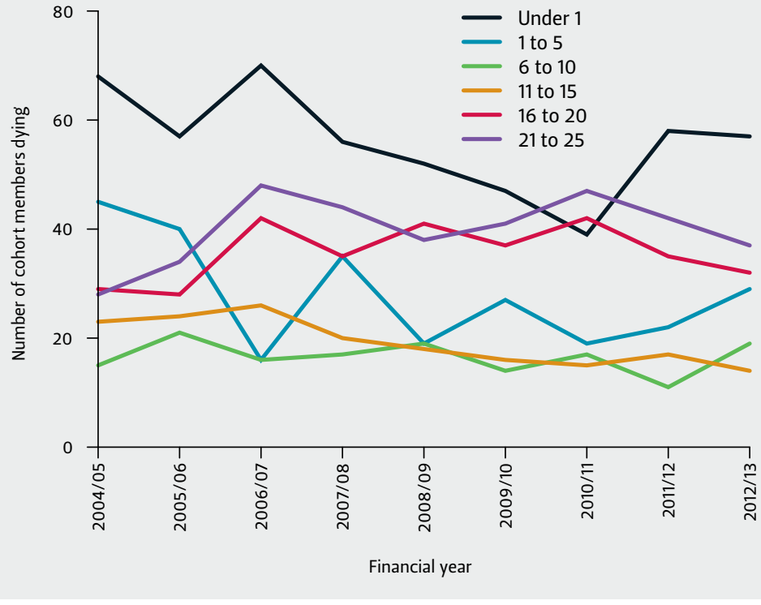
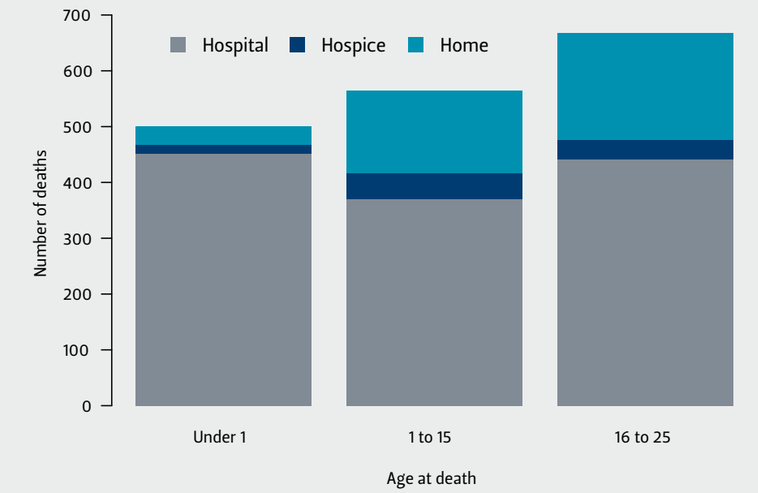


FIGURE 11: Place of death, by age group, for total deaths occurring in financial years 2004/5 – 2013/14 among children and young people in Scotland identified as having a life-limiting condition



Recommendation 2:

Children under 1 year of age should be seen as a priority group for input from palliative care services

The evidence on prevalence

The highest prevalence was in the under 1 age group with hospital-based prevalence 163.0 (95% CI 157.7-168.3) per 10000 population and complete prevalence 192.1 (95% CI 186.3-197.8) per 10000 population in 2013/14 (Table 1, Figure 3). Prevalence in the under 1 age group was steady over the study period.

The evidence on stage of condition

Children aged under 1 year were most likely to be unstable (2013/14: 21.6%), deteriorating (2013/14: 6.7%) or dying (2013/14: 4.5%) and least likely to be stable (2013/14: 67.2%) in this cohort (Figure 12).

The evidence on deaths

The average number of deaths in Scotland each year in children and young people with life-limiting condition was 195 but there was a decline in the number of deaths per year over the period (from 208 in 2004/5 to 188 in 2012/13). The numbers of deaths per year varied by age group with the largest number in the under 1 year age group (mean 56) but this did decline slightly during the time period of the study (Figure 10).

Children under 1 accounted for 28.7% of all deaths in this cohort or 46.8% of the deaths in children and young people with life-limiting condition in Scotland aged <16 years.

Place of Death

90.2% of children under 1 year of age with a life-limiting condition in Scotland who died between 2004/5-2012/13 died in hospital, 6.6% died at home and 3.2% died in a hospice (Figure 11).

The significantly higher prevalence in the under 1 age group is similar to previous results from England (Fraser et al., 2012). The combination of higher prevalence, large percentage of these young children being unstable, deteriorating or dying in a single year and the very high percentage currently dying in hospital makes this age group a priority for input from palliative care services. This input should be a combination of direct provision of palliative care and training and education of professionals working in perinatal, neonatal and paediatric settings.

FIGURE 12: Proportions, by age group, of total children and young people with a life-limiting condition in Scotland at each of four stages of condition in financial years 2009/10 – 2013/14.

Recommendation 3:

Age specific palliative care services for young people (aged 16-25 years) with a life-limiting condition in Scotland should be developed

The evidence on prevalence

The hospital-based prevalence estimates have risen in the 16-20 year age group from 18.3 (95% CI 17.6-19.1) per 10000 population in 2003/4 to 31.6 (95% CI 30.6-32.6) per 10000 in 2013/14 (Figure 3). The complete prevalence estimates for the 16-20 year age group have risen from 53.8 (95%CI 52.6-55.1) per 10000 in 2009/10 to 73.3 (95%CI 71.8-74.8) in 2013/14.

The hospital-based prevalence estimates have risen in the 21-25 year age group from 15.2 (95% CI 14.5-15.9) per 10000 population in 2003/4 to 24.3 (95% CI 23.5-25.1) per 10000 in 2013/14 (Figure 3). The complete prevalence estimates for the 21-25 year age group have risen from 58.7 (95%CI 57.4-60.0) per 10000 in 2009/10 to 72.5 (95%CI 71.1-73.9) in 2013/14.

Diagnoses

Although congenital anomalies had the highest prevalence in all of the younger age groups, oncology diagnoses became the most prevalent diagnosis by 21-25 years (2013/14 hospital based prevalence per 10 000 population: oncology 5.7 (95% CI 5.3-6.1), congenital 4.3 (95% CI 4.0-4.6); 2013/14 complete prevalence per 10000 population: oncology 18.8 (95% CI 18.1-19.5), congenital 13.6 (95% CI 13.0-14.2)).

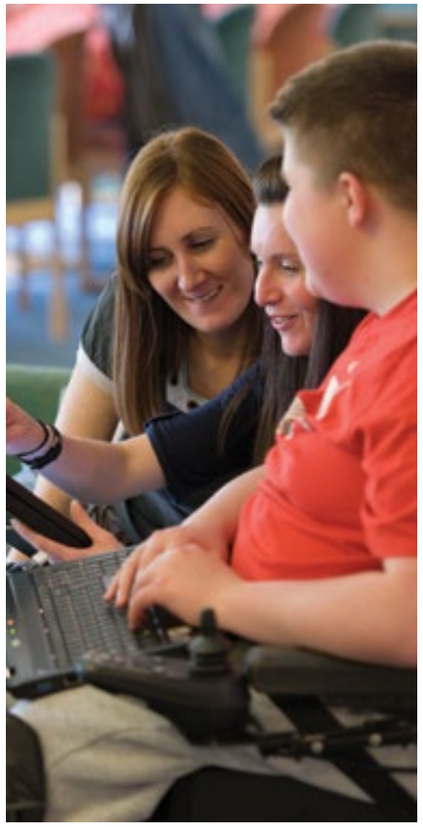
The evidence on stage of condition

Although children with a life-limiting condition in the under 1 age group were most likely to not to be stable in any given year , young people with a life-limiting condition in the 16-20 year and 21-25 year age groups were next most likely to experience instability (Figure 12).

The evidence on deaths

There were a mean 195 deaths per year over the period, but numbers of deaths varied by age group (Figure 10). The mean deaths per year for the 16-20 year olds and 21-25 year olds were 36 and 40 respectively. The overall number of deaths per year declined slightly over the period (from 208 in 2004/5 to 188 in 2012/13) but the deaths in the 16-20 and 21-25 year age groups remained steady.

Over the period from 2004/5 until 2012/13; 66.3% of the deaths in the 16-25 year age group were in hospital, 28.6% at home and 5.1% in a hospice (Figure 11). Although CHAS continues to offer care to young people up to their 21st birthday, at present there is no palliative care provider specifically for young adults in Scotland. Young adults are a unique population with very different needs to children and adults and therefore they should have services designed to meet those needs.



Paul Hampton

Recommendation 4:

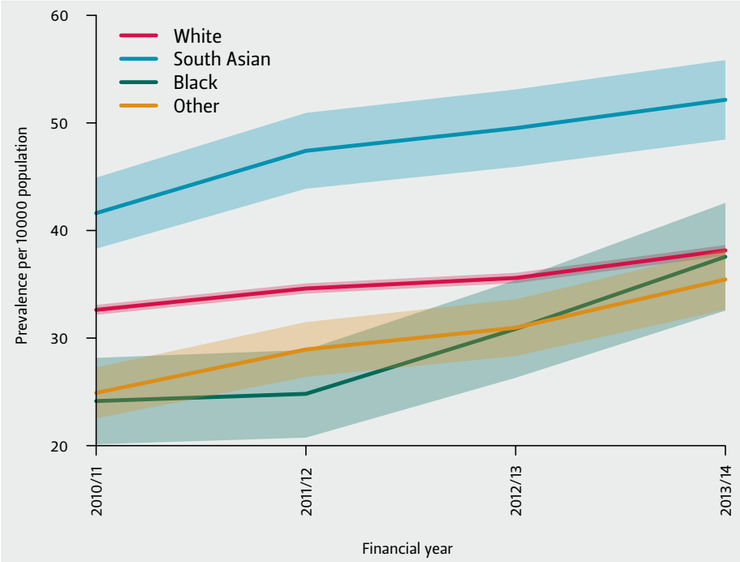
Palliative care services should be able to provide culturally competent care to children and young people from ALL ethnic groups.

The evidence on ethnicity

Prevalence of life-limiting conditions was highest among individuals of South Asian ethnicity (in 2013/2014 hospital-based prevalence was 52.1 (95% CI 48.5-55.8) per 10000 population compared to 38.1 (95% CI 37.6-38.6) for White cohort members, 37.6 (95% CI 32.5-42.6) for Black cohort members and 35.4 (95% CI 32.6-38.3) for cohort members of Other ethnicity (Figure 13).

The significantly higher prevalence of life-limiting conditions in the South Asian population is similar to previous results from England (Fraser et al., 2012). Palliative care services need to be able to offer culturally competent care to children and young people with life-limiting conditions from ALL ethnic groups.

FIGURE 13: Hospital-based prevalence of life-limiting conditions by ethnicity among children and young people in Scotland for financial years 2010/11 – 2013/14.
Shaded regions indicate 95% confidence interval.



Recommendation 5:

Future development of palliative care services in Scotland should ensure that access to services for children and young people from areas of high deprivation is prioritised.

The evidence on deprivation level

There was a linear association with deprivation with 25-26% of children and young people with life-limiting conditions living in areas of highest deprivation and 15-17% of children and young people with life-limiting conditions who living in the least deprived areas (20% of the total population of Scotland live in each deprivation category) (Figure 14).

The evidence on stage of condition

There were small differences between children and young people with life-limiting conditions in the different deprivation categories with the least deprived groups most likely to be stable (2013/14: 86.9% compared to 85.1% for most deprived group) and the most deprived groups were most likely to be unstable (2013/14: 11.8% compared to 10.4% for the least deprived group) (Figure 15).

The higher than expected proportion of children and young people with life-limiting conditions living in areas of high deprivation has been shown previously (Fraser et al., 2012, Taylor et al., 2010) and in other chronic conditions in childhood (Newacheck, 1994). Therefore it is important when developing and delivering services to ensure that access is available for all children and young people with life-limiting conditions in Scotland regardless of where they live.

FIGURE 15: Proportions, by Scottish Index of Multiple Deprivation (2009) category, of total children and young people with a life-limiting condition in Scotland at each of four stages of condition in financial years 2009/10 – 2013/14.

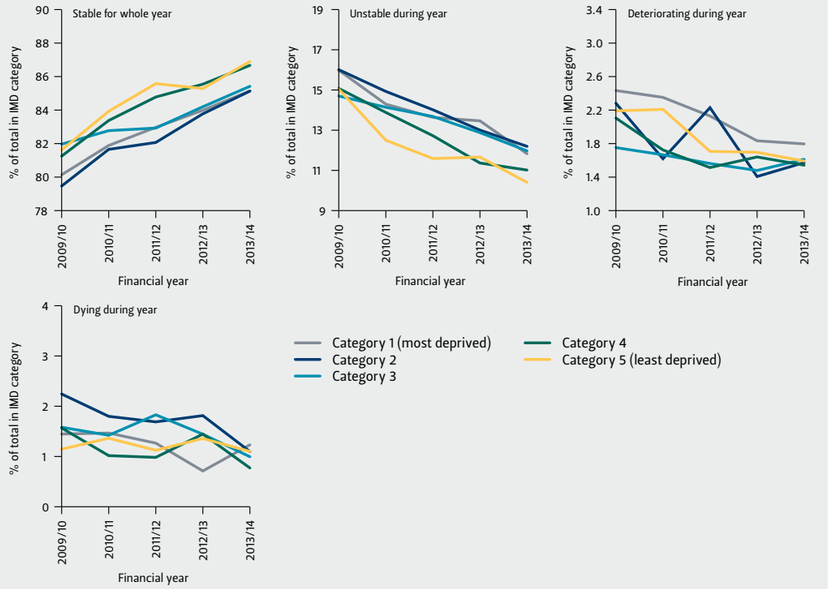
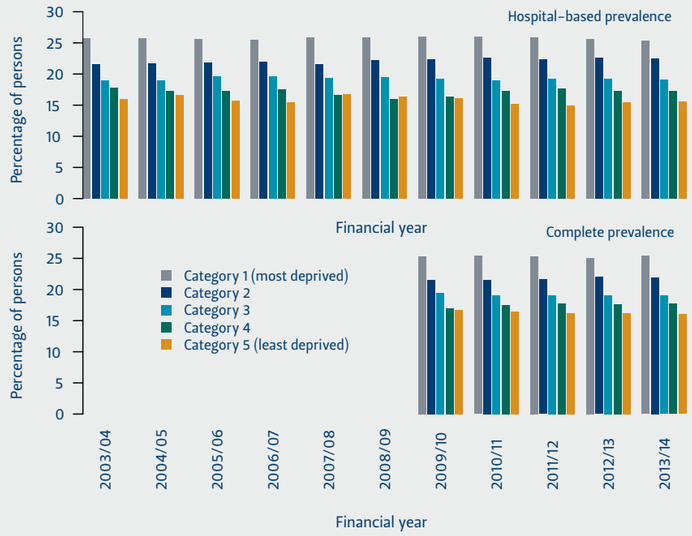


FIGURE 14: Proportions, by Scottish Index of Multiple Deprivation (2009) category, of children and young people (aged 0-25 years) identified as having a life-limiting condition in Scotland in financial years 2003/4 – 2013/14. In the general population equal proportions (20%) are in each of the five categories.



SECTION 3

The psychosocial care and support needs of families: findings and recommendations

Introduction

The second part of this report sets out the recommendations arising from a review of qualitative research with children and young people, parents and siblings. The review comprised two components:

i. A rapid systematic review of studies which had obtained the views and experiences of children and young people with a diagnosis of a life-limiting condition and their families. This review excluded those conditions where, comparatively, it was envisaged that the evidence base would be larger, namely: cancer, cystic fibrosis, HIV, chronic kidney disease, and cerebral palsy.

ii. A review of existing literature reviews which included one or more of the conditions excluded from the rapid systematic review.

An overview of the methods can be found in Appendix 2. Appendices 3 and 4 provide details of the studies included in the review and the recommendation(s) which drew on each study's findings.

The purpose of the review was to identify, describe and synthesise evidence on families' needs and experiences in order to inform recommendations regarding the psychosocial care and support provided by palliative care services in Scotland. Five recommendations are made based on the review's findings. Primarily, and in line with the study objectives, these recommendations concern high level principles regarding what interventions and support should be provided by palliative care services in order to meet families' psychosocial needs, rather than specifics of practice.

Two main, overarching themes emerged from the evidence reviewed: the emotional and psychological impact of living with a life-limiting condition, and the 'extraordinary' roles and responsibilities parents assume. With respect to the latter, meeting the child's care needs, and the impact of that on parents' time to fulfil other roles and responsibilities within the family, emerged as a dominant issue and one where studies consistently report parents feel there is insufficient support.

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Two recommendations arise from these substantial bodies of evidence:

- Recommendation 6: Specialist psychological and emotional care should be available to children and young people and all family members. Included in this should be support for couples, and support and guidance to parents as they respond to the emotional needs of their children. Services caring for children and young people with life-limiting conditions should also find ways to provide opportunities for all family members to connect with and spend time with their 'peers'.

- Recommendation 7: Palliative care services should consider incorporating domestic support to families within their portfolio of services. They should also seek ways to extend provision of support to parents in order to reduce, or provide a break from, the caring demands and responsibilities placed on parents.

Two further, more specific, recommendations arising from the findings of the review are also made. The first concerns the role of residential provision within the wider portfolio of palliative care services:

- Recommendation 8: Palliative care services in Scotland should continue to include specialist residential provision. This provision should be extended in light of evidence of the size of the population of children with life-limiting conditions living in Scotland.

The second arises from evidence of children and young people's, parents' and siblings' experiences of discrimination and social exclusion, and the impact this has on their emotional well-being and the availability of support from informal social networks:

- Recommendation 9: Palliative care services should contribute to or support activities which seek to educate and challenge societal perceptions of disability.

Finally, the review also identified a discrete literature on parents' psychosocial needs during the end of life stage. The topics of decision-making, doctor-parent communication and bereavement support, whilst often appearing as themes or topics within the research reviewed, were not the focus of this study. There are already substantial bodies of evidence on these issues and these have been used to develop existing, and forthcoming, guidance. However, a further set of evidence – consistent across studies – emerged concerning parents' needs **as parents** in the days and hours leading up to their child's death, and the practices which support and enable them to parent their child during this period. This evidence forms the basis of the final recommendation:

- Recommendation 10: Palliative care services, and other services involved in providing care at end of life, should examine current practices with respect to evidence on parents' psychosocial needs **as parents** in the final stages of their child's end of life.

Much of the remainder of this section of the report presents the evidence underpinning these recommendations.

A note about the breadth and quality of the evidence

It is important to stress that these recommendations are derived solely from the available evidence. There are clearly gaps in the current evidence base. Therefore the recommendations do not, and cannot, cover all the potential aspects of psychosocial care and support for children and young people with life-limiting conditions and their families.

Overall, the evidence related to parents' psychosocial support needs was quite robust: a wide range of life-limiting conditions and children's ages were represented and a diverse set of topics or issues explored. The evidence base for children and young people and siblings was much more limited and is dominated by studies of children with particular conditions (cancer, heart conditions) and who do not have cognitive impairments: this constrains the way the evidence can be used when identifying implications for development and design of palliative care services at a more general level. Specific examples of evidence gaps are: the religious and/or spiritual support needs of children and young people; research which has sought to describe the lives of children with profound

impairments beyond using the accounts of parents and other informants; and research which has worked directly with children and young people moving towards end of life in order to identify their needs and experiences.

The issues of access to services and care coordination

Before moving on to discuss each recommendation and the supporting evidence in more detail, it is important to raise the issues of access to services and care coordination. Not unexpectedly, these were both reported and explored in some of the studies included in this review. There was consistent evidence from these studies that difficulties finding out about and accessing services and care coordination add, sometimes quite significantly, to the stresses and demands parents face and their sense of isolation (see overview of evidence for Recommendation 6 below). Furthermore, and in line with wider research on disabled young people, the transition to adult services emerged as a particularly difficult and stressful period, both for parents and young people.

Access to services and care coordination are clearly issues where differences in policy and service organisation are likely to influence families' experiences. None of the research identified and included in this review was carried out in Scotland. It is for this reason we have not developed a specific recommendation regarding these issues.

It is, perhaps, useful to note here that Scottish Government guidance and policy does address the issues of care coordination and supporting families to identify and access services. Getting It Right For Every Child (GIRFEC) – the approach set out to improve services for children and young people – includes the Named Person and Lead Professional roles. The role of the Named Person is to listen, advise and help a child or young person and their family, providing direct support or helping them to access other services. They can help families address their concerns early and prevent situations becoming more serious. Lead Professionals are for those families using services across two or more agencies. The Framework for the Delivery of Palliative Care for Children and Young People in Scotland (Scottish Children and Young People's Executive Group, 2012) identified that the adoption of a GIRFEC approach as a key outcome for palliative care for babies, children and young people in Scotland. Furthermore, GIRFEC underpins the recent Children and Young People (Scotland) Act 2014 which is placing statutory requirements on services regarding the way they support children with additional needs, including those with palliative care needs, and their families.

Recommendation 6:

Specialist psychological and emotional care should be available to children and young people and all family members. Included in this should be support for couples, and support and guidance to parents as they respond to the emotional needs of their children. Services caring for children and young people with life-limiting conditions should also find ways to provide opportunities for all family members to connect with and spend time with their 'peers'.

The emotional experience, and impact on emotional well-being, of living with a life-limiting condition emerged as a significant theme in the review of evidence. It was reported in detail by studies which specifically explored this issue, and was also commonly reported by studies which had focused on other aspects of families' experiences. This indicates the prominence of this aspect of the experience of living with a life-limiting condition.

Many studies report that parents (and siblings) refer to the joy and pleasure they experience in their relationship with their child and their pride in their child's response to the difficulties they encounter.

> **First of all, I see him as my son, whom I am very proud of…He really gives so much happiness.**
> (GRAUNGAARD ET AL., 2011, P121)

> **Yes, my son is amazing. He has taught me everything.**
> (ZIERHUT AND BARTELS, 2012, P51)

Further, the notion of 'positive growth' is something a number of studies identified or investigated in children and young people, parents and/or siblings. 'Positive growth' refers to the changes or gains which individuals regard as positive outcomes of a difficult or negative experience. The following quote from a young adult illustrates this:

> **A lot of people in this world will never experience this, and I'm not saying that I am glad I had cancer, but I kind of am because it's like, it made me stronger, you know…It's educated me. And it is something that I am proud of.**
> (DURAN, 2013, P190)

However, these positive emotional experiences or outcomes occur in parallel with a range of difficult and distressing emotions which, at times, can be felt most acutely:

> **I live in a constant state of desperate happiness…I try to hang on to every happy look and smile, while at the same time I live in fear of her death. It is possible to have sadness, fear and happiness co-existing in you at one time, though. I know, because I'm doing it.**
> (RALLISON AND RAFFIN-BOUCHAL, 2012, P201)

Managing difficult and distressing emotions, and permanently living in a heightened state of emotional strain or stress, take their toll, though this may not be recognised or acknowledged. Furthermore, in addition to managing their own emotional responses, parents want to provide the best emotional care and support to their children. Based on the evidence reviewed, the argument for palliative care services to incorporate psychological and emotional care is strong.

In addition, for children and young people, parents, and siblings, opportunities to share experiences with individuals in similar situations is helpful and comforting.

Peer support consistently emerges as an important source of support.

In seeking ways to implement this recommendation, it would appear useful to: i) investigate how psychology services work in children's cancer, and other specialist, services and the use of clinical psychologists in hospices; and ii) review evidence on effective ways of providing peer support. Evidence on the impact of gender and ethnicity on willingness to reveal or share emotional distress, and ways of coping, should also inform service developments.

Overview of evidence: children and young people

Studies of children able to participate in verbal interviews revealed the different aspects of having a life-limiting condition which can arouse negative emotions and/or threaten emotional well-being.

First are the anxieties associated with health and prognosis: these were present even when understanding of the prognosis did not include the life-shortening nature of their condition.

> **The thing I worry about most is the, um, dying bit. That's what I don't like.**
> (GAAB ET AL., 2013B, 190)

Also, there were the worries and concerns experienced by any child or young person but which may be exacerbated by having a life-limiting condition including: friendship difficulties, returning to school after an absence, wider relationships with peers, schoolwork and future issues, such as leaving school, opportunities for intimate or sexual relationships and fertility.

Dependency on others, or being aware that their condition imposed restrictions or demands on other family members, caused a range of emotions, particularly guilt or frustration.

> **I felt like s***....I felt like my brothers and sisters missed out on time with my mum.**
> (GAAB ET AL., 2013B, P189)

In addition, some studies reported children and young people's sadness when observing the distress of other family members.

> **As she [mother] cries, tears also flow in my eyes...and I feel very sorry for her.**
> (LEE AND KIM, 2012, P401)

Witnessing peers enjoy physical health, activities or experiences, or their growth towards independence, could prompt feelings of anger and jealousy and a 'longing' to participate. This latter concept crossed the boundary with feelings of loss and grief which were also reported by some studies.

> **It sucks to have it and sometimes makes me mad, 'cause when I see my friends like ride bikes off jumps and stuff...it's hard to watch them do that.**
> (PEHLER AND CRAFT-ROSENBERG, 2009, P 485)

Studies including children and young people with visible physical impairments, or who used mobility aids or medical technologies outside of the home, reported that feelings of 'difference', embarrassment and social discomfort were part of their emotional experiences.

> **When I'm around people other than the ones I know well, I feel uncomfortable and embarrassed at myself because of my physical disability.**
> (GAAB ET AL., 2013B, P189)

Researchers noted that fatigue and ill-health affected children and young people's resilience or ability to deal with, or manage these emotions. In terms of day to day experiences, night-time was often the time of day when fears, anxieties and sadness were most acutely experienced.

> **When I'm about to fall asleep I usually think about it...what if something happens?**
> (BRATT ET AL., 2012, P 532)

In addition, certain times or situations were identified as posing a threat to emotional well-being. These included:

- the time around diagnosis;
- periods of ill-health or evidence of deterioration;
- experiencing loss of participation in usual occupations and activities due to extended period of hospitalisation or ill-health;
- experiencing significant changes to the body / appearance (e.g. hair loss; scars; changes in weight);
- the death of a peer;
- changes to, or changing services;
- gaining a fuller understanding of their prognosis;
- feeling un- or under-informed.

Some of these events were defined by researchers using words such as 'traumatic': a term which efficiently captures the emotional and mental work which may be required to recover from, or adapt to, the experience.

Finally, some studies described the tensions that can arise between children and young people and their parents. These can arise for a number of reasons: managing compliance with treatment regimes, perceived parental 'over-protectiveness', returning to higher levels of dependency on parents due to ill-health, and the transition to greater independence during the teenage years and into young adulthood.

Overview of evidence: parents

Key conclusions from studies of parents' experiences are set out below. It is important to note that these experiences may well follow the highly traumatic period of the diagnosis or the birth of the child. Thus, parents may move into a chronic experience of emotional distress and dis-ease in an already fragile or depleted emotional state.

Parents' emotional worlds:

- Parents can live in a permanent state of raised anxiety, dread, and/or sorrow.
- In addition, parents experience periods of intense, and sometimes very mixed, emotions.
- Sources of distress and/or anxiety identified include:
  - living with uncertainty;
  - living with the possibility of death;
  - witnessing their child's physical and/or emotional suffering, including suffering caused by treatment regimens/protocols;
  - living with loss – loss of 'what could have been' – and anticipating the loss of their child;
  - the reactions and responses of family, friends and wider society to their child or their situation as parents of a child with a life-limiting condition.
- Being responsible for (often complex and life-affecting) treatment protocols can be a significant source of emotional strain, particularly in the early days of taking on this responsibility.
- Dealings with services often aroused feelings of frustration, loneliness and anger.
- Parents who work may experience conflicting emotions around needing to work but wanting to spend more time with child and family.
- Ostensibly positive events (for example, completing cancer treatment) may be associated with heightened levels of anxiety, uncertainty and/or a sense of isolation.

Parents' responses to, or the impact of, emotional strain and distress:

- Some parents' accounts reveal very significant levels of emotional distress and fragility.
- Distress and anxiety contributes to parents' experiences of poor quality or interrupted sleep.
- Parents may not recognise, or choose not to face, their emotional experiences and well-being.

- Hiding anxiety and distress or venting stress and frustration can hinder couples' abilities to support each other and may threaten their relationship.
- Mothers and fathers may respond differently to the emotional demands and strains they face: this can be a source of friction, isolation and another source of distress in itself.

> **"It's an emotional roller coaster. It has been in all different levels since the day [our child] was diagnosed."**
> (CARROLL ET AL., 2012, P6)
>
> **"The anxiety I constantly feel never subsides. It's firmly rooted...is unrelenting."**
> (EATOUGH ET AL., 2013, P1045)
>
> **"It's like a ticking time bomb. You think about it every day. You know it's going to happen: you just don't know when."**
> (ZIERHUT AND BARTELS, 2013, P48)
>
> **"Some days we noticed he did not want to play, and was more downbeat, more sensitive, more introverted, did not want to interact... and that distressed us much: because we did not know what to do to help him."**
> (MONTOYA-JUAREZ ET AL., 2013, P722)
>
> **"[Transition] was one of the worst years in my life. I found it really stressful and frustrating... it's nearly beaten me. It just really, really broke me."**
> (KIRK AND FRASER, 2014, P345)
>
> **"If I've had a really bad day, my instinct when he [partner] comes in is to attack: I need to scream at somebody and he knows.. he does. But it does not make for a good relationship."**
> (RODRIGUEZ AND KING, 2009, P9)

Overview of evidence: siblings

Evidence from studies which have explored the experiences of siblings also reveals the potential for a wide-ranging emotional impact. Those reported in the studies reviewed are summarised in Figure 16.

In addition, some studies highlighted the potential impact on emotional well-being caused by loss, or disruption, of normal routines, medical crises, and changes in the primary caregiver (e.g. father or grandparent replacing mother).

A number of researchers commented on the conflicting nature of siblings' emotional responses to having a brother or sister with a life-limiting condition and what that entails. For example, feeling angry at the same time as feeling guilty about that anger.

Figure 16 sets out what might be regarded as negative emotions. In addition, however, some studies explored possible positive experiences and outcomes for siblings specifically associated with having a brother or sister with a life-limiting condition. For example, caring for, and having particular responsibilities for care or household tasks, could engender a sense of pleasure or pride. Studies also report that the particularly close relationships within families could be a source of joy.

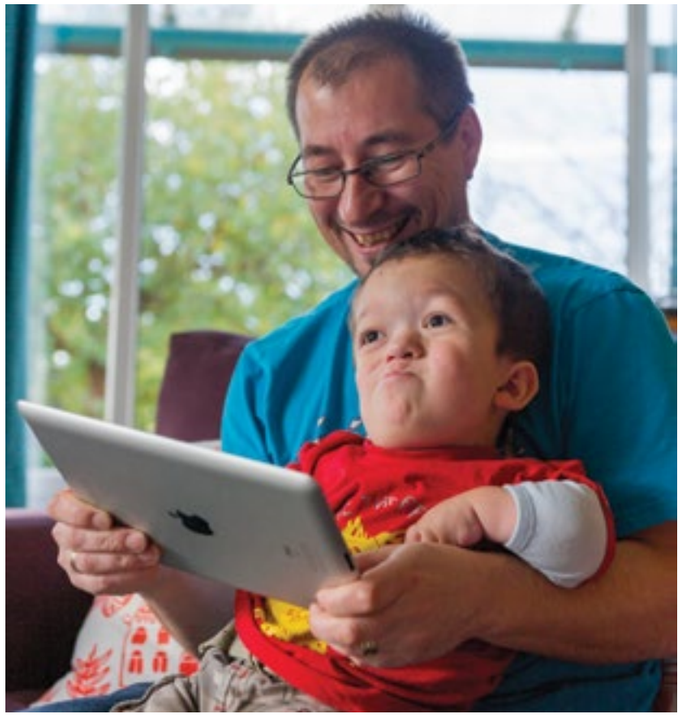
Finally, however, it is important to note that some studies reported that siblings may hide their emotional responses in order to avoid distressing their parents.

> **"I try not to make them upset. I silence bits of my life. "**
> (BRENNAN ET AL., 2013, P818)

FIGURE 16: Siblings' experiences of negative emotions and possible causes

TYPE OF EMOTION	SOURCE OR CAUSE
Guilt	Sibling's own healthiness Negative feeling towards brother or sister
Anger	Restrictions on life Responsibilities associated with caring for brother or sister Other people's reactions to their brother or sister
Anxiety	Brother or sister's health and prognosis Own health and mortality
Sadness	Brother or sister's health and prognosis Anticipating death of brother or sister
Lonely	Parents pre-occupied with brother or sister
Resentment	Responsibilities associated with caring for brother or sister

> **"When I'm at school I'm different. I don't have to worry about things, you know. Well, I do, but not as much: I can be myself and everything."**
> (BRENNAN ET AL., 2013, P819)
>
> **"Alone in my room most of the day. Sad I couldn't help [child]. Most of my normal days are like this."**
> (GAAB ET AL., 2013B, P189)
>
> **"I'm afraid that she is going to get sick and nobody will be there."**
> (GAAB ET AL., 2013B,P189)
>
> **"I feel sad when we are not altogether at home. When [child] is at hospital: I don't know if she is really sick and if she might die."**
> (GAAB ET AL., 2013B, P189)



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Recommendation 7:

Palliative care services should consider incorporating domestic support to families within their portfolio of services. They should also seek ways to extend provision of support to parents in order to reduce, or provide a break from, the caring demands and responsibilities placed on parents.

Three of the themes identified in the review of evidence provide support for this recommendation:

- parents' experiences of the caring role;
- fulfilling domestic tasks;
- parents' accounts of physical exhaustion.

Evidence on the way care needs can make significant demands on parents' time, attention and physical resources is extensive and persuasive. Importantly, all the studies reviewed which explored this issue also reported that parents felt they did not receive enough support with meeting their child's care needs. We also draw on two further topics or themes in making the case for this recommendation. First, a small number of studies revealed the impact of caring responsibilities on parents' abilities to complete domestic tasks, something which some parents report finding hard to neglect even when at the point of utter tiredness. Second, conclusions around the importance of assisting parents with caring and domestic tasks are further supported by parents' accounts of physical exhaustion.

Parents' experiences of caring responsibilities

A number of studies reviewed explored parents' descriptions of the caring role. Caring responsibilities included feeding, bathing, toileting, administering treatments and medications, comforting, monitoring physical health and symptoms, physiotherapy, washing, entertaining or keeping occupied, watching over to ensure safety in and outside of the home, supporting or enabling mobility and communication.

The notion of caring being a continuous responsibility was a common theme. This concept was used to describe the day by day, hour by hour, minute by minute experience of caring; it was also used to capture its long-term, or permanent, nature. A number of studies reported that parents directly linked caring responsibilities to their experiences of a lack of time for themselves and/or other members of the family, though some studies also reported parents contextualised their experiences as being those of any parent.

Often co-occurring with the concept of the continuous nature of caring was a commentary on the pervasiveness of caring. Researchers used this and similar terms to represent the range of care needs a child may have and the way that meeting these care needs draws on parents' physical, mental and emotional faculties.

> **"She follows me everywhere."**
> (BRUNS AND SCHREY, 2012, P166)

> **"Sometimes the days with him are particularly intense since the morning I wash, dress, prepare his food and feed him, I have to keep up with him 24 hours to 24. I cannot think of me anymore, it's impossible."**
> (EATOUGH ET AL., 2013, P1044)

> **"Because there have been long period where he is crying every waking minute and I feel that I simply can't stand that for a very long time."**
> (GRAUNGAARD ET AL., 2011, P127)

A few studies usefully highlighted how parents' sense of caring responsibility may continue even when someone else was looking after their child; something that studies of parents' experiences of using short break/hospices also report.

> **"You never really get a break, you never get a break from the weight of the responsibility... It is there and it remains present whether you are with your child or not. And to not acknowledge that is to deny part of yourself."**
> (RALLISON AND RAFFIN-BOUCHAL, 2013, P200)

Research which looked at experiences of some sort of short-break service were very rich in parents' accounts of the pervasiveness of care demands, with the break from these demands highlighting to parents the 'extraordinary' nature of their lives as a parent of a child with a life-limiting condition.

> **"Just taking care of her takes a lot. Just travelling with her, with the wheelchair, with the pump, dealing with all this stuff. Not to do that: it's respite in itself."**
> (CHAMPAGNE AND MONGEAU, 2012, P247)

> **"That's what's important when he's in respite: I don't have to stick to a schedule."**
> (CHAMPAGNE AND MONGEAU, 2012, P247)

The concept of the pervasiveness of caring was also used when describing the impact of meeting these care needs on other aspects of the parent's and family's life including parents' physical and mental health, parents' ability to fulfil other roles and spend time with other family members and the ability of families to enjoy 'everyday' family experiences and activities.

> **"When she went [to a short break facility], I had a chance to catch up a bit more on my sleep. I went back to bed, and I went back to sleep, and got some of my strength back."**
> (CHAMPAGNE AND MONGEAU, 2012, P247)

Keeping up with domestic and other household tasks

The issue of managing household tasks was explored and reported on in a small number of the studies reviewed. The multiple demands on parents' time was identified as impacting on their ability to keep up with domestic and other household tasks. Importantly, this experience was reported by mothers and fathers; something which, it could be argued, serves to highlight the range of tasks required in looking after and maintaining the home.

A couple of studies (of mothers only) suggest parents may respond differently to this. Both report some mothers accepted the situation and adjusted their expectations down to what they felt was acceptable.

> **"The house isn't always clean but everyone is fed and comfortable."**
> (BRUNS AND SCHREY, 2012, P166)

For some, however, keeping up with household chores was important to parents' sense of control, or an untidy or unclean (by their standards) home environment was, in itself, an additional source of stress. Indeed, it is interesting that studies which have explored parents' experiences of short-term care have observed that some mothers use the break from caring to catch up on housework. The final source of evidence on this theme comes from studies of siblings where a number of studies report that helping out with domestic duties was one of the roles siblings may assume.

Parents' accounts of physical exhaustion

Physical exhaustion was ascribed to the demands of fulfilling significant levels of caring responsibilities **alongside** other roles and responsibilities, such as domestic and other household management tasks, as well as desire to make time for siblings and wider family life:

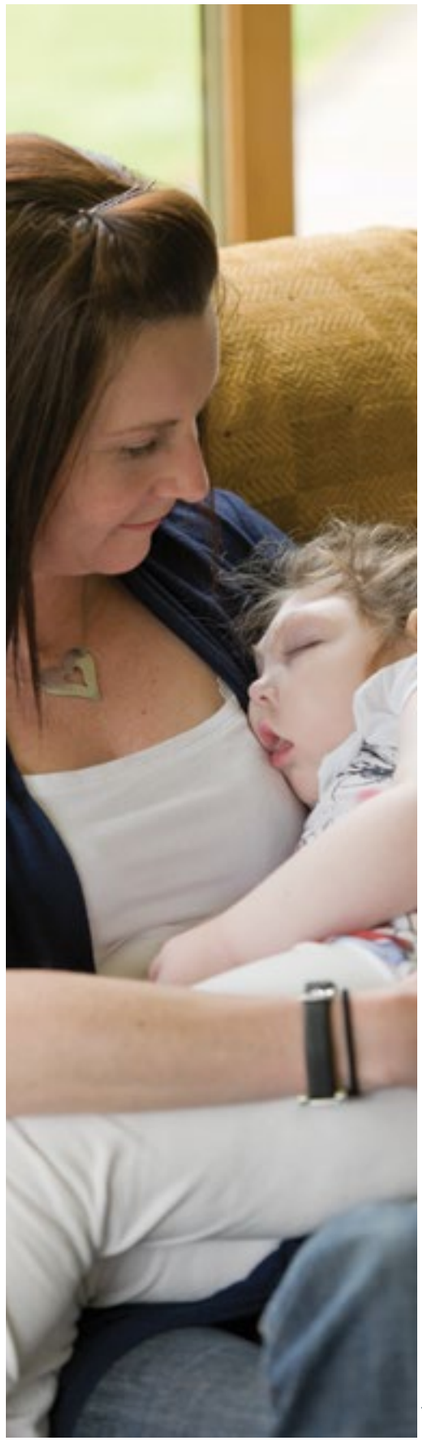
> **"... at the moment it's mere survival ... it's a big problem that we are always so tired..."**
> (GRAUNGAARD ET AL., 2011, P122)
> **"And you keep going, but your body is giving you signals and you don't listen to it. But I don't have it in me anymore. I do it because I have no choice. But it is hard, you know. You get older and you don't have that energy anymore."**
> (DAVIES ET AL., 2011, P37)

There is also evidence that there are, potentially, also multiple causes of insufficient, disturbed sleep or poor quality sleep. Night-time care requirements and a sub-conscious 'listening out' for the child or equipment alarms were both potential sources for parents' sleep to be affected. For parents whose sleep was interrupted or disturbed by caring responsibilities, an opportunity to catch up on sleep was consistently identified as a benefit of short-term breaks.

> **"I could switch off and not sleep with one eye open all night. I could get a good night's sleep. Because he is on a ventilator machine your mind is always on it and you don't want to get into a deep sleep because you are worried that you might not hear it [alarm], so it is not a proper sleep I get at night times."**
> (KIRK AND PRITCHARD, 2012, P35)

In addition, parents may choose to reduce the time they sleep because of a desire to ensure domestic and other tasks were properly attended to – something which some parents reported finding helpful in terms of a sense of control and feeling they were fulfilling their role, or desire, to care for and nurture the whole family.

> **"I found that if I want to get things done, my only real option is to sleep less. I have to stay up late or get up early."**
> (BRUNS AND SCHREY, 2012, P166)



Paul Hampton

Recommendation 8:

Palliative care services in Scotland should continue to include specialist residential provision. This provision should be extended in light of evidence of the size of the population of children with life-limiting conditions living in Scotland.

Evidence from the studies which explored children and young people's, parents' and siblings' experiences of using residential short-break services suggests that these are positive and valued aspects of families' lives and an important element of the care and support families receive.

The findings of these studies, together with evidence from research which has explored the experiences of living with a life-limiting condition more widely, highlight the **particular** benefits residential provision can offer.

First, it provides opportunities for peer support between children and young people, parents and siblings. Of the studies exploring this issue, the chance to spend time with others going through the same situation and experiences was consistently reported as being valued and desired by children and young people and parents.

> **"It's a break and you get to see each other. With our short life expectancy we don't see each other enough, so they are really precious to me."**
> (KIRK AND PRITCHARD, 2012, P36)

Second, for all family members, particularly children and young people, residential services are an environment where illness and impairment is the 'majority experience', thus offering a time of feeling 'normal' rather than 'different'. Across the studies reviewed, experiences of discrimination, negative or unhelpful attitudes from members of the public were reported by children and young people, parents and siblings. Furthermore, as reported earlier, some children and young people described feeling uncomfortable or embarrassed when out and about.

> **"I hate being out in public with my wheelchair because I see people that I know and sometimes they treat me differently and they have no idea what I'm going through."**
> (GAAB ET AL., 2013B, 190)

Third, for children and young people, a well-designed residential service means moving around is less effortful and restrictive. This can be dramatically different to their experiences at home.

> **"...two wheelchairs can race down the corridor side-by-side! And it's easy for me to move from room to room."**
> (SWALLOW ET AL., 2012, P263)

Fourth, whether or not it is a whole family visit, residential services can allow parents and siblings to have time together. This serves to address parents' concerns regarding the time they have available for other children, and the ways family life can be restricted because of the care and other needs of the child. The outcome of this is less emotional 'dis-ease' for parents regarding siblings' well-being and quality of life.

> **"It's good to know we can enjoy ourselves like a family, but he can enjoy himself too. I think that's the biggest thing."**
> (CHAMPAGNE AND MONGEAU, 2012, P 249)

Fifth, for parents, it provides a break from care and caring responsibility. Our earlier discussion of the evidence informing Recommendation 7 clearly supports the argument that this is a much-needed function of residential palliative care services. Importantly, confidence in staff is essential to parents' using, or gaining the most from, this provision. The health and

nursing care skills and expertise of staff were critical to parents' feeling confident about handing over the care of their child. In addition was a desire that the child also benefited from the change in carer.

> **"They pay more attention to new medications, hours, diets; everything is very, very well monitored"**
> (CHAMPAGNE AND MONGEAU, 2012,P249)

> **"It is reassuring for us that he is happy, and that he is enjoying himself as much as we are. That has an enormous effect on our state of mind."**
> (CHAMPAGNE AND MONGEAU, 2012,P249)

Finally, residential care provides parents with an opportunity for sleep and physical recuperation. Again, as reported in the evidence presented to support Recommendation 7, physical exhaustion and sleep deprivation are two outcomes of the 'extraordinary' roles and responsibilities they may assume as a parent of child with a life-limiting condition.

Paul Hampton

Recommendation 9:

Palliative care services should contribute to or support activities which seek to educate and challenge societal perceptions of impairment and disability.

Many children with life-limiting conditions have physical, learning and/or sensory impairments. At a community or societal level, therefore, there is the potential for children's palliative care services to contribute in some way to educating and challenging societal perceptions of impairment and disability.

This recommendation arises from the findings across a number of studies that children and young people, parents and siblings experience social exclusion, discrimination and negative attitudes because of their impairments, or fear such experiences happening if they divulge their diagnosis. Findings from the studies reviewed indicate that these experiences may be encountered at school, in local communities and within the extended family.

> **"I don't think others fully understand: my brain is the same as other people. It's just my physical body is different... At school they say mean things."**
> (GAAB ET AL., 2013B, P190)
>
> **"I get angry 'cause people look at [brother/sister] differently. I get annoyed when classmates make fun of disabled people."**
> (GAAB ET AL., 2013B, P190)
>
> **"Everyone starts pulling away from you too because they are afraid."**
> (EATOUGH ET AL., 2013, P1045)

Such experiences were consistently associated with emotional distress and a sense of exclusion and social isolation. They added to the emotional load of living with a life-limiting condition. For parents and siblings, observing the child or young person with a life-limiting condition experience exclusion and negative attitudes was an additional source of distress and angst.

Recommendation 10:

Palliative care services, and other services involved in providing care at end of life, should examine current practices with respect to evidence on parents' psychosocial needs as parents at their child's end of life.

This final recommendation draws on findings from studies which explored parents' experiences of their child's death. Whilst only a few studies were identified which explored this topic, they do represent experiences of end of life across the entire age range, from perinatal death forwards. In addition the findings across studies are consistent. A key theme emerging from the findings of these studies, and which has informed the above recommendation, was parents' need to **continue** to be parents to their child during the final days and hours of their child's life. Yet medical crises and intensive care settings can restrict this. It would be fair to argue that supporting and enabling mothers and fathers to be parents during this period will also yield positive outcomes for the child.

The evidence which informed this recommendation draws on the research with bereaved parents. It is worth noting that in these studies parents made a connection between their satisfaction with the opportunities they had to continue to be their child's parent, with their views about the quality of their child's death and their subsequent adjustment and acceptance of their child's death.

The wider context for parents' specific needs at end of life is the evidence that parents have a very strong, sometimes consuming, desire to provide the very best for their child. This motivates and energises parents on a day to day basis, and these feelings are likely to become more acute when parents become aware their child's life is drawing to a close.

> **"I do not want to be thinking after he has gone, "Well, I wish I had done this and I wish I had done that"...because you cannot change things then. ...So as long as I know that I have given him the best quality of life he could have possibly had, then I have nothing to reproach myself for later."**
> (RODRIGUEZ AND KING, 2009, P9)

Finally, as noted earlier, other parental roles during the end of life stage – particularly decision-making – were not within the scope of the evidence review.

Parents' desires to parent during the end of life phase: overview of evidence

i. Parents reported wanting choice and control over the extent to which they maintained their parental role in terms of meeting the child's care needs through to the point of death. Bereaved parents' experiences in neo-natal units also stress the importance of this experience, even though their role had been transitory. Appropriately supported, the final act of caregiving could be a helpful experience.

> **"The nurse helped us bathe her and dress her ...we will remember that time fondly. It is the only chance we got to really feel like her parents."**
> (BRANCHETT AND STRETTON, 2012, P42)
>
> **"Sometimes I was told not to touch her and this was very painful. Before I could change her and cuddle her – the typical things you do with a baby. Here [PICU] I felt a distance between me and her to avoid medical complications."**
> (LAMIANI ET AL., 2013, P1337)

ii. Parents wanted to be able to touch and hold their child in order to offer emotional comfort to their child and/or to comfort themselves. This was regarded as a fundamental element of the role, and right, as a mother or father. It extended beyond the moment of death.

> **"The fact that they called us when [child] was already dead was horrible....not being able to be there in that moment was devastating ....holding him and saying 'goodbye'. That time I was almost angry. I understand that you [doctors] have to do everything possible, but the son is mine, isn't he?"**
> (LAMIANI ET AL., 2013, P1340)
>
> **"I think the most important thing to me was that I got to hold him and sit with him in a private room and I wasn't rushed into anything."**
> (BRANCHETT AND STRETTON, 2012, P42)

iii. The opportunity for private times with their child was a third aspect of what parents desired during their child's end of life. Parents described a desire for time as a family, for intimacy and quiet. This was not just with respect to the moments around their child's death but as a wider feature of the days and hours leading up to that moment.

iv. Finally, three of the studies which contributed to this recommendation offered evidence on ethnic or cultural influences on parents' wishes and desires during this period. Three issues emerge:

- There is evidence that there may be differences between ethnic groups in terms of the extent to which treatments and interventions are pursued. The researchers note that the transition from active management to palliative care may be particularly challenging for some ethnic groups.
- Members of some minority groups may be more likely to feel dis-empowered to express their wishes and desires.
- Inadequate provision of interpreters risks parents' wishes and desires not being understood or fulfilled.

Concluding Comments

The purpose of the ChiSP study was to generate evidence to support the planning and delivery of paediatric palliative care in Scotland. It sought to identify the number of children and young people with a life-limiting condition living in Scotland, and to describe this population in terms of age, diagnosis, geographic location and ethnicity. A further aim was to describe this population in terms of the stage of their condition; that is stable, unstable, deteriorating or dying. Its final objective was to identify and describe the psychosocial care and support needs of these children and young people, and of their families. In terms of service planning, these are all key areas of evidence.

A mixed-method study design, comprising two workstreams, was used to achieve these aims. First, analysis of existing administrative and health service datasets was used to generate detailed information about the population of children and young people with life-limiting conditions in Scotland. The findings from this work are presented in Section 2 of this report. The second element of the study was a review of research with children and young people with a life-limiting condition, their parents and/or siblings which explored or identified their psychosocial support needs. The findings from this evidence review are set out in Section 3.

On the basis of the findings of these two workstreams, ten high level recommendations have been made. The first five recommendations arise from the epidemiological work (workstream 1) and draw implications from the evidence regarding the nature of the population of children and young people with life-limiting conditions living in Scotland. The remainder of the recommendations are based on the findings of the review of evidence on psychosocial care and support needs (workstream 2). They are therefore concerned with the types of psychosocial care and support which should fall within a portfolio of palliative care services for children and young people and their families.

It is important to note that these recommendations do not address all the evidence needs of those involved in planning paediatric palliative care in Scotland. As noted in Appendices 1 and 2, and discussed in the earlier sections of this report, there were limitations to the data available for both workstreams. This inevitably limits the detail of, and the topics covered in, the recommendations'. Nevertheless, we hope this report will prove a useful resource to all organisations in Scotland who have a responsibility for children and young people with life-limiting conditions, and/or are involved in the planning and delivery of paediatric palliative care.

Finally, as is typically the case at the end of a study, a number of topics or issues emerge as potential areas for future research. In terms of the specific recommendations made in this report, certainly monitoring and robust evaluation of any changes to the provision, design or content of services would be very useful. To this end, it would be helpful to introduce systems by which this can be achieved from the outset which should include high quality data collection. At a broader level, a prospective cohort study of children and young people with life-limiting conditions and their families could provide valuable evidence of their use of healthcare, experience of transition(s) and much needed evidence on the physical and mental health of parents and carers.

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APPENDIX 1

Methods for the quantitative analyses of linked healthcare data workstream

Definition of life-limiting conditions

A previously developed (Fraser et al., 2011, Fraser et al., 2012) list of ICD10 (International Classification of Diseases, 10th edition (World Health Organisation, 1992)) codes that constituted the conditions of interest was utilised to identify individuals with a life-limiting condition. Further refinement of this coding framework was undertaken; three ICD10 diagnostic codes (Q74.8, Q44.5 and F80.3) used to identify individuals with a life-limiting condition were omitted as no individuals with these codes were known to have died during the study period (Table 2).

Population data

Population at risk was determined from mid-year estimates by age and gender derived from census data (National Records of Scotland, 2015). These estimates were not available split by ethnic group therefore the 2011 census population was used to determine prevalence by ethnicity.

Datasets used

Routinely collected NHS data were used (Figure 1). The study cohort was identified by searching the Scottish Morbidity Record Inpatient activity database (SMR01) and Scottish Birth Records (SBR) for individuals resident in Scotland, aged 0-25 years, with any of the life-limiting condition indicative ICD10 codes listed in any record from 1 April 2003 to 30 March 2014 (financial years 2003/4 to 2013/14). Extracts were then made for the identified individuals from the following data sources:

- Scottish Birth Records (SBR)
- Scottish Inpatients Episodes (SMR01)
- Scottish Outpatient Dataset (SMR00)
- Scottish Cancer Registry (SMR06)
- GRO Death registration data
- Scottish Community Prescribing data (available from 2009/10)
- UK Paediatric Intensive Care Audit Network (PICANet) (available from 2007/8)

The SBR contained records of births in Scotland and associated neonatal care episodes with diagnoses. SMR01 contained all inpatient episodes occurring in hospitals in Scotland, with patient demographics and diagnoses. The PICANet data contained demographic and clinical information on episodes involving an admission to a PICU in the UK (Paediatric Intensive Care Audit, 2015). Community prescribing data contained records of prescriptions dispensed outside of hospitals in Scotland and patient demographics. The cancer registry contained date(s) of diagnosis with cancer, patient demographics and, if occurring, deaths. Death registration data contained date of death and diagnostic codes. SBR, SMR01 and the community prescribing data were used to determine prevalence and the other datasets were used to complete demographics and to determine date of death (Figure 1).

Access to the NSS Scotland data was given by the Privacy Advisory Committee (ref number: XRB14010) and the linkage of the PICANet data to the NSS Scotland data was approved by the Health Research Authority. All analyses took place within the NHS National Services Scotland eDRIS Safe Haven secure environment (Information Services Division, 2015). The results presented here underwent disclosure control before release (Information Services Division, 2012).

The numbers of referrals and deaths per year to Children's Hospice Association Scotland were provided by Children's Hospice Association Scotland.

TABLE 2: ICD-10 diagnostic coding framework used to identify children and young people in Scotland with a life-limiting condition. Conditions were categorised in eleven diagnostic groups.

NEUROLOGY	HAEMATOLOGY	ONCOLOGY	METABOLIC	RESPIRATORY	CIRCULATORY	GASTROINTESTINAL	GENITOURINARY	PERINATAL	CONGENITAL		OTHER
A17	B20-B24	C00-C97	E31.0	E84	I21	K55.0	N17	P10.1	Q00.0	Q44.2	H11.1
A81.0	D56.1	D33	E34.8	J84.1	I27.0	K55.9	N18	P11.2	Q01	Q44.7	H49.8
A81.1	D61.0	D43	E70.2	J96	I42	K72	N19	P21.0	Q03.1	Q60.1	H35.5
F84.2	D61.9	D44.4	E71	J98.4	I61.3	K74	N25.8	P28.5	Q03.9	Q60.6	M31.3
G10	D70	D48	E72		I81	K76.5		P29.0	Q04.0	Q61.4	M32.1
G11.1	D76.1		E74			K86.8		P29.3	Q04.2	Q61.9	M89.5
G11.3	D81		E75					P35.0	Q04.3	Q64.2	T86.0
G12	D82.1		E76					P35.1	Q04.4	Q74.3	T86.2
G20	D83		E77					P35.8	Q04.6	Q75.0	Z51.5
G23.0	D89.1		E79.1					P37.1	Q04.9	Q77.2	
G23.8			E83.0					P52.4	Q07.0	Q77.3	
G31.8			E88.0					P52.5	Q20.0	Q77.4	
G31.9			E88.1					P52.9	Q20.3	Q78.0	
G35								P83.2	Q20.4	Q78.5	
G40.4								P91.2	Q20.6	Q79.2	
G40.5								P91.6	Q20.8	Q79.3	
G60.0								P96.0	Q21.3	Q80.4	
G60.1									Q23.2	Q81	
G70.2									Q21.8	Q82.1	
G70.9									Q22.0	Q82.4	
G71.0									Q22.1	Q85.8	
G71.1									Q22.4	Q86.0	
G71.2									Q22.5	Q87.0	
G71.3									Q22.6	Q87.1	
G80.0									Q23.0	Q87.2	
G80.8									Q23.4	Q87.8	
G82.3									Q23.9	Q91	
G82.4									Q25.4	Q92.0	
G82.5									Q25.6	Q92.1	
G93.4									Q26.2	Q92.4	
G93.6									Q26.4	Q92.7	
G93.7									Q26.8	Q92.8	
									Q28.2	Q93.2	
									Q32.1	Q93.3	
									Q33.6	Q93.4	
									Q39.6	Q93.5	
									Q41.0	Q93.8	
									Q41.9	Q95.2	
									Q43.7		

Age

Date of birth was assigned as the most commonly recorded date. Each individual was assigned an age in each financial year based on age at the start of the first episode (in SMR01 and SBR) in that financial year. If there was no SMR01 or SBR episode then age at the date of the first community prescription in the year was used.

Gender

Gender was recorded as male, female or not known. The most commonly reported gender (excluding not known) was assigned to each individual.

Diagnoses

Diagnostic fields in SMR01 and SBR were used – there were 6 diagnostic fields in SMR01 and 32 in SBR. Diagnoses were categorised into 11 groups based on ICD10 chapters: neurology, haematology, oncology, metabolic, respiratory, circulatory, gastrointestinal, genitourinary, perinatal, congenital and other (Table 2). No attempt was made to prioritise diagnoses for an individual and therefore individuals may have a life-limiting condition in more than one category.

Ethnicity

Ethnicity was determined first by collapsing the various ethnicities recorded to four main groups: White; South Asian (Indian, Pakistani and Bangladeshi); Black; Other (including mixed ethnicity). The most commonly recorded ethnic group (excluding "not known") was assigned to each individual.

Deprivation

Deprivation category (SIMD2009) was assigned based on datazone of residence. The five categories were population weighted to be equal in size – i.e. 20% of Scotland's population were in each category. Individuals were assigned the first deprivation category recorded each year.

Date and place of death

Dates of death were contained in death registration data, SMR01, the SMR06 and PICANet. Dates of death up to one day before the beginning of the last known SBR or SMR01 record were retained to allow for errors or delays in registering the beginning of an episode; any dates of death earlier than this were considered invalid and removed. Dates of death were completed first using the death registration data and the other sources were used to replace only invalid or missing dates of death. Place of death was determined from the death registration data, as hospital, hospice, home, other or unknown. The hospice category included the children's hospices operated by CHAS and self-contained adult hospices (hospice units within hospitals could not be separately identified and were recorded in the hospital category).

Analyses

Prevalence

Prevalence figures were estimated using two criteria. For "hospital-based prevalence" (Fraser et al., 2012) individuals were counted in a given year if they had a life-limiting condition and any inpatient hospital episode (SBR or SMR01) in that year while aged 0-25 years. Under the second criteria ("complete prevalence"), all individuals counted using the first set of criteria were included and, in addition, individuals were counted if they appeared in the community prescribing records in that year (this indicated that they were still alive and resident in Scotland). As community prescribing data were only available from 2009 onwards, the complete prevalence analyses were limited to financial years 2009/10 to 2013/14.

Prevalence and 95% confidence intervals (CIs) per 10 000 population was calculated for each year overall, by age group, gender, IMD category, ethnicity and diagnostic group. Due to poor recording of ethnicity in earlier years and in the community prescription records, analyses of prevalence by ethnicity were limited to 2010/11 onwards and to only hospital based prevalence.

Number of Deaths

The number of deaths within the cohort each financial year was also calculated and split by age group. Data on deaths for the first (2003/4) and last years (2013/14) were excluded due to cohort edge effects.

Stage of Condition

Four stages of condition were defined: stable, unstable, deteriorating and dying (Hughes-Hallet et al., 2011).

Transitions between stages were defined a priority (Figure 7).

Individuals were stable when present and not in one of the other three stages. Status was analysed by financial year, with the most severe stage of condition being recorded.

The percentage of the total population of children and young people with a life-limiting condition in each category per year was calculated overall, by age group, diagnostic group and deprivation category. The denominator for these analyses was the number of individuals in the corresponding complete prevalence calculations. The numbers of cohort members of non-White ethnicity was too small for any analyses of stage of condition by ethnicity to be disclosed.

Strengths and limitations

This study used a transparent and repeatable methodology utilising high quality administrative data. This study has built upon previous methodology and refined the ICD10 coding framework.

There are some limitations. Importantly, although data were available from CHAS on the numbers of referrals and deaths, this was aggregate data and therefore could not be linked to the other datasets. Also no data was available from the other providers of paediatric palliative care in Scotland. Disclosure control rules limited the release of some data.

As an individual only required to have a life-limiting condition ICD 10 code recorded once to be included, this methodology may be including individuals who have had a life-threatening event, particularly around the time of birth, but this event may have been transient and they may no longer be life-threatened or life-limited.

The stage of conditions definitions were based upon clinical knowledge and availability of data.

APPENDIX 2

Methods used for the review of qualitative evidence

The overarching objective of the review was to synthesise evidence on children and young people's, parents' and siblings experiences of living with a life-limiting condition in order to identify and describe the psychosocial support needs of families who may benefit from support from palliative care services. The findings of the review were then used to develop recommendations regarding the design and future development of palliative care service for children and young people in Scotland. The focus of the review was the psychosocial care and support needs of families excluding the time of diagnosis (where there is extensive evidence and guidance on supporting families at this time) and after the child's death. Papers were therefore excluded if (a) they only reported data on care and support needs at the time of diagnosis; (b) they only reported on bereavement support

There were two elements to the review:

- a rapid systematic review of primary research on all life-limiting conditions (just under 300 specific diagnoses) with the exception of a small number of conditions where there is a high volume of research on families' experiences of living with a life-limiting condition, namely: cancer, cystic fibrosis, cerebral palsy, HIV/AIDS, and renal disease;
- a 'review of reviews' of research representing cancer, cystic fibrosis, cerebral palsy, HIV/AIDS, and chronic kidney disease.

Searches

All searches were restricted to papers published in the English language, from OECD countries, over the past five years (2009-14). These restrictions sought to ensure that the findings reported in the papers would be relevant and broadly transferrable to the situation in Scotland. A full report of the searches is available (Arber, 2014).

The searches involved a comprehensive and systematic search of published studies indexed by three databases: Medline, CIAHNL, and Psychinfo. For the rapid systematic review, the search string consisted of 'diagnosis' + 'family member' + 'qualitative research'. The diagnoses included in the research were drawn from a list of life-limiting conditions which was used to create the ICD coding framework used in workstream 1 (Fraser et al., 2011).A total of 14,193 papers were identified. For the 'review of reviews' the search string was adapted to 'condition' + 'family' + 'qualitative' + 'review'. A total of 3,747 papers were identified

Screening and selection

Rapid systematic review

Titles were screened against inclusion and exclusion criteria, at which point 13,372 papers were eliminated. The abstracts of the remaining 821 papers were then screened independently by two researchers and a further 693 papers were excluded. Full texts of the remaining 128 papers were retrieved and read in detail, at which point a further 74 papers were excluded. A total of 54 papers met the inclusion criteria and were included in the review (see Figure 17). Details of the papers are set out in Appendix 3.

Exclusion criteria

- <50% of the sample had a life-limiting condition;
- <50% of the sample was diagnosed at age 18 years or younger;
- <50% of the sample was aged 25 years or under;
- study only included children with cancer in remission;
- study only investigated experiences of diagnostic process, bereavement support and/or decision-making
- study only collected quantitative data

FIGURE 17: Screening process for the primary research papers

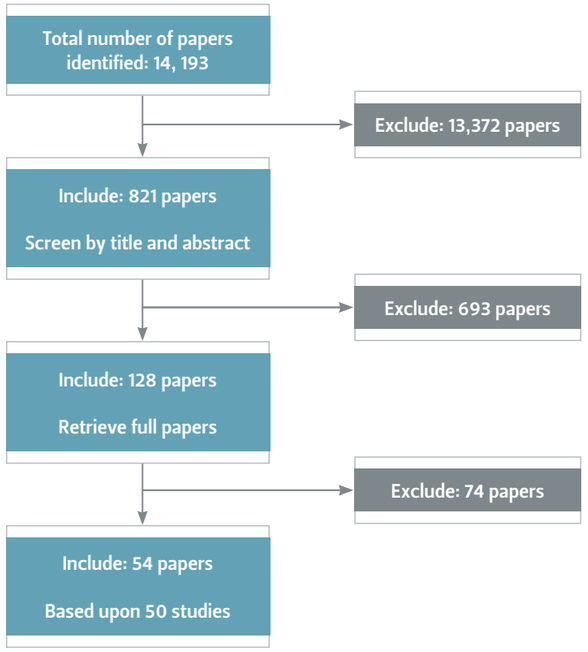
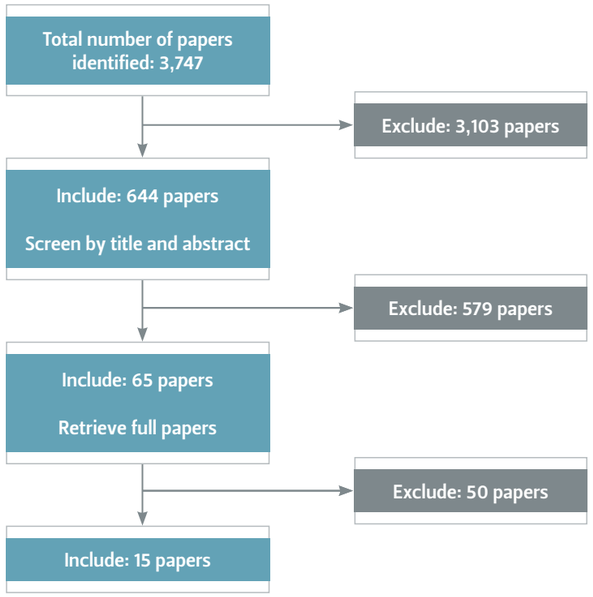


FIGURE 18: Screening process for the review papers



The review of reviews

The screening process followed that outlined above for the rapid systematic review. Papers were screened against the search terms reported, and were included only if: (a) they included studies using qualitative research methods; (b) information was provided about the studies included in the review; and (c) method(s) of analysis were reported. A total of 15 review papers were included in our review (see Figure 18). Details of the included papers are set out in Appendix 4.

Rapid systematic review: research participants

The total number of participants across the included studies was: 90 children and young people with life-limiting conditions, 599 mothers (of which, 208 were bereaved), 233 fathers (63 bereaved), 106 'parents' (46 bereaved), 49 brothers (12 bereaved), 60 sisters (13 bereaved), 2 gender unspecified siblings, and 40 other family members (13 bereaved) (see Table 3 below). In total, 1,179 research participants took part in the studies included in the review. Of those, 355 were bereaved. 835 children and young people from 827 families were represented.

Of the 54 papers, 3 reported research conducted solely with children and young people, 33 with parents, 2 with siblings, 4 with children/young people and their parents, 1 with children/young people and their siblings, one with parents and siblings, 3 with children/young people, parents and siblings, 6 with parents and other family members (grandparents, aunts, cousins), and 1 conducted solely with grandparents (see Table 4).

TABLE 3: Total number of research participants

PARTICIPANT GROUP	TOTAL NUMBER	OF WHICH, BEREAVED
Children and young people with life-limiting conditions	90	-
Mothers	599	208
Fathers	233	63
'Parents'	106	46
Brothers	49	12
Sisters	60	13
'Siblings'	2	-
Other family members	40	13
TOTAL	1,179	355

TABLE 4: Number of papers reporting research conducted with each group

POPULATION	NO. OF PAPERS REPORTING RESEARCH CONDUCTED WITH THIS POPULATION
Children/young people with a life-limiting condition	3
Parents	33
Siblings	2
Children/young people and parents	4
Children/young people and siblings	1
Parents and siblings	1
Children/young people, parents and siblings	3
Parents and grandparents, cousins, aunts, etc	6
Grandparents	1
TOTAL NO. OF PAPERS	54

Thus, parents were participants in 47 of the 54 papers, children and young people in 11 papers, and siblings in 7 papers. Consequently, the parent data was the richest, most wide-ranging, and diverse with respect to the types of life-limiting conditions represented. Children and young people with a life-limiting condition represented the smallest number of research participants. Those who took part in research studies were predominantly older children and teenagers, those who were not 'medically frail', and those without significant cognitive impairments. For further information about the ages and diagnoses of participants see Appendix 3.

Rapid systematic review: countries represented

The 54 papers were based on 50 studies. 16 of the papers (from 15 studies) were based

on research conducted in the USA, 12 from England, 7 from Canada, 4 (from 3 studies) from New Zealand, 3 (from 2 studies) from Sweden, and one paper from each of Australia, Brazil, Denmark, Iceland, Italy, Korea (South), the Netherlands, Spain, and Switzerland. In addition, two papers (from one study) were based on research conducted in the USA, Canada, England and Scotland, and one paper was based on research conducted in the Netherlands, Italy, Poland and Sweden.

Rapid systematic review: quality of the evidence

The quality of the papers included in the review of primary evidence was appraised using the methodology checklist for qualitative studies published by the National Institute for Health and Care Excellence (NICE, 2012). In line with the checklist, papers were assessed in terms of their execution and reporting of the characteristics of the population in the sample, the methods, and analysis. Each paper was assessed against a range of criteria which were then grouped within a sub-category and graded 'good', 'mixed', or 'poor'. A grade of overall quality was then produced: 'good' if all 3 sub-groups (sample, methods, analysis) scored good, 'good/mixed' if the paper scored good in two sub-groups and mixed in the third, 'mixed' if at least two of the sub-groups were scored as mixed, and 'poor' if two or more of the sub-groups had been graded as poor. Of the 54 primary research papers, 26 were rated as good, 16 good/mixed, 11 mixed, and 1 poor. Studies were not excluded or weighted on the basis of the quality assessment.

Analysis

A thematic approach to the analysis was taken. Each paper was initially read by two members of the research team (BB and NM) to identify themes or 'topic areas'. These were informed by the research objectives. The identification of themes was an iterative process as discussion within the team was ongoing. Themes/topic areas on which research evidence could be extracted was agreed within the team. Each paper was then reviewed for evidence on a theme-by-theme basis. Relevant evidence, including study information and verbatim quotes, were extracted into distinct 'topic documents'. Each paper was then reviewed again to check that all relevant evidence on all identified topics had been extracted. Discussion and review of the topic areas and the conceptual framework in which the evidence and review of that evidence was located was ongoing within the team. Topic documents were reviewed as a whole, across each of the populations (parents, children and young people, siblings) to identify higher level concepts. Analysis of the higher level concepts, and the evidence underpinning them, was used to identify and inform the recommendations contained in this report.

Strengths and limitations

A key strength of the review of primary evidence is that it synthesises evidence relevant to understanding families' psychosocial care and support needs from a large number of life-limiting conditions, many of which are rare and have only been included in one, or a handful of, studies. The review also represented a good range of children's ages. The review is a rapid systematic review as there were not the resources for two researchers to extract data from each paper. The quality of studies was reasonable, though weaknesses in terms of selection bias and insufficient detail about the data collection and/or analytical process are to be noted.

In terms of the review of reviews, cancer is over-represented and the low representation of cystic fibrosis, HIV and cerebral palsy in the studies taken forward into the review is a limitation. However, based on our knowledge of UK children's hospices, it is our judgement that children with cerebral palsy were represented in studies included in the review of primary evidence in studies where participants were simply described as having a 'life-limiting condition' and were recruited from children's hospices. Finally, not all studies were systematic reviews, perhaps reflecting the relatively recent implementation of this methodology with qualitative data.

Given the resources available, conducting a review of primary evidence and a 'review of reviews' to achieve the study objectives appears to have proved a useful approach.

However, whilst there is a high volume of qualitative research on children and young people with HIV and cystic fibrosis, it would appear there have not been many attempts to review and synthesise this evidence in terms of psychosocial needs: this was unexpected.

Overall, the evidence reviewed is dominated by parents' accounts. Furthermore, the children and young people who took part in the studies had neither cognitive nor communication impairments. Whilst we did extract and use data on parents' accounts of their views of their child's life for this review, this does highlight a need for qualitative research which captures and describes the worlds of children and young people with more severe impairments. Similarly, the evidence base for siblings of children with life-limiting conditions was relatively sparse compared to that for parents' views and experiences. Finally, sampling bias is particularly an issue with respect to the studies which included children and young people or siblings.

Description of the papers included in the review of primary evidence

AUTHOR(S) & YEAR	COUNTRY	AGES OF C/YP WITH LLC[1]	CONDITION(S)[2]	PARTICIPANTS	METHODS	RECOMM. INFORMED BY THE STUDY[3]
Bousso et al. 2010	Brazil	Children	LLC (n/s)	9 mothers, 1 father, 1 grandmother (rep. 9 families)	Oral history	6
Branchett & Stretton 2012	England	Neonates	n/s	54 bereaved mothers, 3 bereaved fathers (rep. 57 families)	Open-ended electronic survey	10
Bratt et al. 2011 (=Bratt 2012)	Sweden	5-16 years	Hypertrophic cardiomyopathy	7 mothers, 5 fathers (rep. 10 families)	Interview	6
Bratt et al. 2012 (=Bratt 2011)	Sweden	8-18 years	Hypertrophic cardiomyopathy	13 Children (rep. 13 families)	Interview	6
Brennan et al. 2013	England	n/s	LLC (n/s)	18 brothers (8 bereaved), 13 sisters (5 bereaved) (aged 5-15) (rep. 21 families)	Interview, photo-elicitation	6, 8
Brooten et al. 2013	USA	0-9 years	Congenital anomalies, prematurity, head trauma, chromosomal abnormalities.	44 bereaved mothers, 19 bereaved fathers (rep. 47 families)	Interview	10
Bruns & Foerster 2011 (=Bruns 2012)	USA, Canada, England, Scotland	3-30 years	Rare trisomy conditions (9, 13, 18)	18 mothers, 2 fathers (rep. 20 families)	Open-ended electronic survey, interview	6
Bruns & Schrey 2012 (=Bruns 2011)	USA, Canada, England, Scotland	3-30 years	Rare trisomy conditions (9, 13, 18)	18 mothers, 2 fathers (rep. 20 families) [Same participants as in Bruns & Foerster 2011]	Open-ended electronic survey	6, 7
Carroll et al. 2012	USA	0-24 years	Neuromuscular, respiratory, metabolic, congenital, malignancy	9 mothers, 7 fathers (rep. 15 families)	Interview	6
Champagne & Mongeau 2012	Canada	11 months-17 years	Degenerative, severe disabilities, cancer	25 mothers, 8 fathers (rep. 27 children from 25 families)	Interview	6, 7, 8
Contro et al. 2010 (=Davies 2010)	USA	n/s	LLCs (n/s)	19 bereaved mothers, 7 bereaved fathers, 12 other bereaved relatives (grandmother, brother, sister, aunt, cousin, brother-in-law) (rep. 21 families)	Interview	10
Cote-Arsenault & Denney-Koelsch 2011	USA	In utero-1 month old	Lethal Foetal Diagnosis: Trisomy 18, hypoplastic left heart, renal agenesis and potter's sequence, multiple GU malformation	5 mothers, 3 fathers (rep. 5 families)	Interview	6, 10
Davies et al. 2010 (=Contro 2010)	USA	1 day-20 years	Oncologic/ hematologic, cardiac, preterm, congenital abnormalities, other	Additional 6 bereaved mothers and 4 bereaved fathers (rep. additional 9 families) [additional to those included in the paper by Contro et al 2010]	Interview	10
Davies et al. 2011	Canada	18-21 years	Complex chronic neurological condition and intellectual impairment	10 mothers, 5 father, 2 other family members (grandmother, foster mother) (rep. 11 families)	Interview	6, 7
Eatough et al. 2013	Holland, Italy, Poland, Sweden.	9-24 years	Juvenile Huntington's Disease	13 mothers, 1 father (rep. 14 families)	Interview	6, 7, 8, 9
Einarsdottir 2009	Iceland	n/s	Preterm <1000g	28 mothers, 25 fathers (rep. 29 families)	Interview	
Gaab et al. 2014	New Zealand	3-19 years	Cancer, heart conditions, dystrophies	12 sisters (7 bereaved), 6 brothers (4 bereaved), (siblings aged 9-22 years) (rep. 9 families)	Interview	7
Gaab et al. 2013a (=Gaab 2013c)	New Zealand	3-18 years	Cancer, heart conditions, dystrophies	12 mothers (5 bereaved), 5 fathers (3 bereaved), 2 other caregivers (1 bereaved) (rep. 11 families)	Interview	6, 9
Gaab et al. 2013b	New Zealand	9-18 years	Cancer, heart conditions, dystrophies	7 CYP, 3 brothers, 6 sisters (all aged 9-18 years) (rep. 8 families)	Audio and written diaries	6, 8, 9
Gaab et al. 2013c (=Gaab 2013a)	New Zealand	3-18 years	Cancer, heart conditions, dystrophies	12 mothers (5 bereaved), 5 fathers (3 bereaved), 2 other caregivers (1 bereaved) (rep. 11 families) [Same participants as in Gaab et al 2013a]	Interview	6
Gilmer et al. 2013	USA	0-14 years	Cardiac, congenital defects, neonatal-specific, infectious disease, cancer	14 bereaved mothers, 1 bereaved father (rep. 15 children)	Telephone interview	10
Goldstein & Rimer 2013	USA	1 month-11 years	SIDS, prematurity, cardiac, neurological, oncological, metabolic/genetic, GI, Other	16 bereaved parents (rep. 16 children)	Interview	6

AUTHOR(S) & YEAR	COUNTRY	AGES OF C/YP WITH LLC[1]	CONDITION(S)[2]	PARTICIPANTS	METHODS	RECOMM. INFORMED BY THE STUDY[3]
Graungaard et al. 2011	Denmark	1-27 months	Infantile spasms, lissencephali, lumbar meningo-myelocele, Wolf-Hirschhorn syndrome, Down's syndrome, unknown	8 mothers, 8 fathers (dyads) (rep. 8 children)	Interview	6, 7
Grinyer et al. 2010	England	10 months-27 years	Kidney disease, Phenoketoneuria, Miller-Dieker disorder, Rett's syndrome, chromosome deletion 6, Nieman-Pick Type C, Foetal Alcohol Syndrome, Dravet's syndrome, neurological condition n/s, chromosomal disorder, chromosomal abnormality, cerebral palsy	3 young people (aged 8-26 years), 3 sisters (1 bereaved), 2 brothers (age of siblings n/s), 11 mothers (1 bereaved), 4 fathers (1 bereaved), 2 grandmothers, I paid carer. (rep. 11 families)	Interview	7, 8
Haley 2011	USA	2-18 years	Neuro-musculoskeletal (medically fragile and technology dependent)	6 mothers (rep. 6 children)	Interview	
Heringhaus et al. 2013	Sweden	5 months-4 years	Birth asphyxia	15 mothers, 11 fathers (rep. 16 families)	Interview	6
Hexem et al. 2011	USA	0-24 years	Neuromuscular, respiratory, GI, metabolic, congenital, malignancy	37 mothers, 24 fathers, 3 other family members (aunt, grandmother, step-parent) (rep. 41 children)	Interview	
Inglin et al. 2011	Switzerland	0-18 years	Cancer, neurological disorders, other (e.g. heart diseases, immune-deficiencies)	15 mothers (6 bereaved) (rep. 15 children)	Interview	6, 7
Kirk & Fraser 2014	England	16-31 years	DMD, other nervous system, spinal muscular atrophy, congenital, Down's syndrome, metabolic	16 young people, 12 mothers, 4 fathers (rep. 12 families)	Interview	6
Kirk & Pritchard 2012	England	6-22 years	LLC (n/s in interview; mainly defined as central nervous system conditions and cerebral palsy in survey)	Questionnaire completed by 108 families (37 bereaved). Interviews with 12 parents (5 bereaved), 7 young people (aged 9-22 years) (rep. 12 families)	Postal survey, interview	7, 8
Lamiani et al. 2013	Italy	1 month-12 years	Down Syndrome with complex congenital heart defect, preterm ELBW, Chronic renal failure, meningitis, tetraplegia, cerebrohepatorenal syndrome, pontocerebellar hypoplasia	7 bereaved mothers, 5 bereaved fathers (rep. 8 children)	Interview	10
Lee & Rempel 2011	Canada	2-60 months	Hypoplastic left heart syndrome	9 mothers, 7 fathers (rep. 9 children)	Secondary analysis of interview data	6

AUTHOR(S) & YEAR	COUNTRY	AGES OF C/YP WITH LLC[1]	CONDITION(S)[2]	PARTICIPANTS	METHODS	RECOMM. INFORMED BY THE STUDY[3]
Lee & Kim 2012	Korea (South)	14-22 years	Congenital heart disease	10 young peopled (aged 14-22) (rep. 10 families)	Interview	6, 9
Liem et al. 2011	USA	5-23 years	Thalassemia (transfusion dependent)	7 mothers, 7 fathers (rep. 16 children from 14 families	Interview	6, 9
Limbo & Lathrop 2014	USA	In utero-infant (n/s)	Three trisomy disorders, anencephaly, holoprosencephaly, renal agenesis, osteogenesis imperfect type II, urorectal condition	15 bereaved mothers (rep. 16 infants)	Secondary analysis of interview data	6, 10
Maddison & Beresford 2012	England	14-21 years	Degenerative, undiagnosed	14 mothers, 5 fathers (rep. 14 families)	Interview	6, 7
Menezes 2010	England	2-18 years	LLCs (n/s)	11 children/YP (aged 2-18 years), 3 brothers, 8 sisters (age of siblings n/s), 10 mothers, 7 fathers (rep. 10 families)	Case study: Participant observation, artwork, interviews	6
Montoya-Juajrez et al. 2013	Spain	1-14 years	Terminal illness (n/s)	10 mothers, 3 fathers (rep. 13 families)	Interview	6
Moro et al. 2011	USA	23-24 weeks gestation	Preterm <25 weeks	5 bereaved mothers (rep. 5 families)	Interview, review of medical charts	10
Packman et al. 2012	USA	16-27 years	Maple syrup urine disease (MSUD)	8 young adults (aged 16-27), 8 parents (dyads) (rep. 8 families)	Interview, case study	6, 9
Pehler & Craft-Rosenberg 2009	USA	12-17 years	DMD	9 young people aged 12-17	Interview	6, 9
Rallison & Raffin-Bouchal 2013	Canada	<17 years (n/s)	Progressive neurodegenerative illness	Interviewed: 8 mothers, 4 fathers, 1 child, 2 siblings, 2 other caregivers. Observed: 6 children, 2 siblings (rep. 6 families)	Interview, observation	6, 7
Ravindran & Rempel 2011	Canada	Pre-school	Hypoplastic left heart syndrome	10 grandmothers, 5 grandfathers of healthy siblings aged 18 months-6 years (rep. 6 children)	Interview	

AUTHOR(S) & YEAR	COUNTRY	AGES OF C/YP WITH LLC[1]	CONDITION(S)[2]	PARTICIPANTS	METHODS	RECOMM. INFORMED BY THE STUDY[3]
Read et al. 2011	England	5-22 years	DMD	18 sisters, 17 brothers (siblings aged 11-18 years), 27 mothers, 2 fathers (rep. 29 families)	Interview	6
Rempel et al. 2009	Canada	2-60 months	Hypoplastic left heart syndrome	9 mothers, 7 fathers (rep. 9 families)	Interview	6
Rodriguez & King 2009	England	2-12 years	LLCs (n/s)	10 mothers (rep. 10 families)	Interview	6, 10
Samson et al. 2009	Canada	7-17 years	DMD	7 mothers (1 bereaved), 5 fathers (1 bereaved) (rep. 10 children from 9 families)	Interview	6, 7
Sullivan et al. 2014	Australia	3 months-12 years	Cancer, spina bifida, brain damage, cerebral palsy, neurological disorder, neuromuscular disorder, muscle disorder, cardiac abnormality, chromosomal abnormality, metabolic disease	25 bereaved parents (rep. 21 children)	Interview	
Swallow et al. 2012	England	14-18 years	LLCs (n/s)	15 mothers, 5 fathers, 5 young people (rep. 16 young people from 15 families)	Interviews and focus groups with parents, focus groups with young people	6, 8
Weidner et al. 2011	USA	Children, infants, new-borns	Malignancy, premature birth, cardiac illness, neurological illness, GI illness	20 bereaved mothers, 9 bereaved fathers (rep. 20 families)	Interview, focus groups	10
Whiting 2013	England	2-17 years	Respiratory, neurological, severe cerebral palsy, metabolic disorder, neuromuscular, cancer	32 mothers, 7 fathers (rep. 34 children from 33 families)	Interview, mind maps	6, 7, 8, 9
Wray et al. 2013	England	n/s	LLC/LLI (n/s)	45 parents (rep. 45 families)	Interview (face to face, telephone), focus groups, online survey	6
Yuen et al. 2012	Netherlands	Birth-32 months	Lethal Epidermolysis Bullosa	11 bereaved mothers, 10 bereaved fathers (rep. 16 families)	Telephone interviews	6, 10
Zierhut & Bartels 2012	USA	4-21 years	Fanconi Anemia	6 mothers, 3 fathers (rep. 7 children)	Interview (face to face, telephone)	6, 9

APPENDIX 4

Description of the review papers

AUTHOR & YEAR	LIFE-LIMITING CONDITION(S) REPRESENTED IN THE REVIEW	PERIOD COVERED	AUTHOR'S DESCRIPTION OF THE REVIEW	NO. OF PAPERS INCLUDED IN THE REVIEW	RECOMM. INFORMED BY THE REVIEW
Alderfer et al. 2010	Cancer	1997-2008	Systematic review	65	6,7
da Silva et al. 2010	Cancer	1997-2009	Integrative review	14	6,7,8
Duran 2013	Cancer	1975-2010	Narrative synthesis	35	6
Gibbins et al. 2012	Cancer	1998-2010	Systematic review	28	6,7,8,9
Jones et al. 2010	Cancer	1984-2007	Integrative review	53	6,7,8
Lee et al. 2012	Cancer	2000-2010	Metasynthesis	8	6,9
Long & Marsland 2011	Cancer	1996-2009	Systematic review	71	6,7
Marris et al. 2011	Cancer	1996-2008	Review	18	6,8
Muhammad et al. 2012	Chronic Kidney Disease	1957-2011	Literature review	63	6
Pini et al. 2012	Cancer	1991-2011	Systematic review	22	6,8,9
Pousada et al. 2013	Cerebral Palsy	1996-2011	Systematic review	46	6
Santer et al. 2014[1]	HIV, Cystic Fibrosis	1996-2011	Systematic review	19	6,9
Tjaden et al. 2012	Chronic Kidney Disease	1971-2011	Systematic review	17	6,9
Tong et al. 2013[2]	Chronic Kidney Disease	1991-2009	Systematic review	14	6,8
Wakefield et al. 2011	Cancer	1979-2009	Review	15	6

1 Review also included studies representing children with asthma, diabetes, juvenile arthritis.

2 Review also included studies representing children with diabetes.

1 Papers were only included in the study if more than 50% of the sample were aged 25 years or younger

2 Papers were only included in the study if more than 50% of the sample had a life-limiting condition.

3 Not all the evidence reviewed was taken forward into informing the recommendations set out in this report, hence some cells in this column are not populated. This was because, for a small number of themes emerging from the analysis, there was not sufficient or high enough quality evidence to be taken forward to inform a recommendation(s).

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