The Big Study Teams

Project management Team

Lizzie Chambers and Julia Hodgson
Together for Short Lives

Research Teams

Anne Hunt (Lead Investigator),
Sue Hacking, Melanie Farman
School of Health, University of Central Lancashire
Geography and surveys

Jane Coad, Erica Brown, Chris Owens, Nicky Ashley, Katie May, Jasveer Kaur
Centre for Children and Families Applied Research, University of Coventry
Interviews with children and families

Elizabeth West, Val Chandler, Anna Wik, Helen Magee, David Barron
University of Greenwich
Network analysis and interviews with professionals

Nick Hex, Karin Lowson, Dianne Wright
York Health Economics Consortium, University of York
Economics and costing

Sophie Staniszewska, Lee Gunn
Royal College of Nursing Research Institute, University of Warwick
Inclusion of service users and participation across the study

Acknowledgments

Together for Short Lives would like to thank the following people and organisations for their contributions to the research and development of the study.

- Virginia Barstow, parent researcher.
- Sophie Beale, formerly YHEC.
- Emma Aspinall, Acorns Children's Hospice, Birmingham.
- Dot Gillespie, Donna Louise Trust, Stoke on Trent, Staffordshire.
- Dr Angela Thompson and Susan Davies, Coventry Primary Care Trust.
- Carolyn Bishop, Stephanie Courts and Carol Farrell, The Orchard Centre for Community Health, South Warwickshire NHS Foundation Trust.
- Dr Claire Thomas, Pat Fisher and Satti Kaur, The Gem Centre, Wolverhampton City NHS Primary Care Trust.
- Maureen Bytheway, Sai Medical Centre, Walsall Healthcare NHS Trust.
- Dr Maybelle Wallis, Sandwell General Hospital, Sandwell and West Birmingham Hospitals NHS Trust.
- Kath Jones, Hope House Hospice, Oswestry.
- Caron Eyre, Birmingham Children's Hospital NHS Foundation Trust.
- Sarah Kirk and Rachel Yeates, Heart of England NHS Trust, Birmingham.
- Susan Gallagher, Birmingham Heartlands, Birmingham NHS Trust.
- Dr Helen Underhill and Catherine Norris, The Kite Centre, Wye Valley NHS Trust.
- Amanda Dunne, Zoe's Place, Coventry.
- Rachel Williams, Barbara Hart House, Birmingham Community Healthcare Trust.
- Dr Pankaj Shah, Freshwinds, Birmingham.
- Heather Parr and Helen Baillie, Burton Community Children's Nursing Team, South Staffordshire and Shropshire NHS Foundation Trust.
- Greg Field, See Saw Team, Black Country Partnership Foundations NHS Trust.
- Alison Squires, Stafford Community Children's Nursing Team, South Staffordshire and Shropshire NHS Foundation Trust.
- Louise Leather, Coral House, Shropshire Community Health NHS Trust.
- Rebecca Steele, West Midlands (South) Comprehensive Local Research Network.
- Members of the West Midlands Paediatric Palliative Care Network.
- Members of the Stoke-on-Trent and Staffordshire Paediatric Palliative Care Network.
- Lifeline Language Services, Preston.
- The support of the National Institute for Health Research, through the West Midlands Medicines for Children Research Network
- Gayle Routledge, Andrew Hague, Sharon Reardon and Deborah Linster-Ali and other members of the Big Study Parent and Carer Advisory Group
- Ellie Bailey, Newcastle-under-Lyme, aged 14 at the time of the study for design of the Big study logo.

Contents

The Big Study Report	page
Introduction	5
Strand 1 Understanding the demographic and geographical distribution of children with life limiting conditions and their families University of Central Lancashire	6
Strand 2 Qualitive analysis of families met and unmet needs Coventry University	73
Strand 3 A study of a professional network: Coordination, cooperation and collaboration across the West Midlands Paediatric Palliative Care Network University of Greenwich	134
Strand 4 An economic evaluation of service delivery York Health Economics Consortium	162
Strand 5 User participation in the Big Study The University of Warwick	194

Introduction

Evidence from the Independent Review of Children's Palliative Care Services[1] showed that there was inequity of service provision across England. The Independent Review and the subsequent Labour government strategy for children's palliative care: 'Better Care: Better Lives' called for the development of strong commissioning networks and for a better understanding of local population needs. Evidence from research such as the ACT 'Voices for Change' report highlighted the family and professional perceptions of palliative care provision back in 2003[2].

Children with life-threatening or life-limiting conditions and their families have complex needs that require a range of skills and services provided by a range of different organisations in health, social care, education and the voluntary sector. Care is provided by children's hospices, hospitals, and community services. With this huge range of services involved it is important that services work together to provide care for families in a joined up way and to avoid major gaps in service provision and poor transitions between services. There is also great potential for professional networks to enable better collaboration and partnership working between this multitude of providers.

Against this backdrop of policy and growing evidence, it was felt that the time was right to carry out a major research project to look at service provision as a whole, using one region of the UK to explore this issue in depth. The West Midlands region was chosen as the focus of this study as this is an area with strong networks, a variety of types of services and diverse ethnic communities. We believe that our findings from this region will have the power to inform the provision of services not just in this area but also nationally, and will provide a good baseline to inform future work

The overall aim of the Big Study was to answer the question "How well are the palliative care needs of children with life-limiting conditions and their families met by services in the West Midlands?"

This document is made up of five separate reports, and therefore consists of five distinct sections – one for each 'strand' of the project. Each section is written by the academic partner responsible for that particular strand of research. Related documents and information can be found at www.togetherforshortlives.org.uk/bigstudy and an overview of the research can be found at www.togetherforshortlives.org.uk/professionals/resources.

---

[1] Craft, A. and Killen, S (2007) Palliative Services for Children and Young People in England: An independent Review for the Secretary of State for Health, London Department of Health

2 Hunt, A., Elston, S. and Galloway, J. (2003) Voices for Change: Current Perceptions of Services for Children with Palliative Care Needs and their Families. Bristol: ACT.

Strand 1 Report:

Understanding the demographic and geographical distribution of children with life-limiting conditions and their families in the West Midlands.

Dr Anne Hunt
Dr Sue Hacking
Melanie Farman

Contents

Strand 1

page

1. Introduction 11

1.1 Background 11

2. Aims of Strand 1 Study 12

3. Methods 13

3.1 Recruitment of participating services and families 13

3.2 Questionnaires and Instruments 13
  3.2.1 Parent’s Questionnaire 14
  3.2.2 Bereaved Parent’s Questionnaire 14
  3.2.3 Staff Questionnaire 15
  3.2.4 Service Manager’s Questionnaire 15
  3.2.5 Minimum Data Set 15
3.3 Inclusion of Non-English Speaking Families 15

3.4 Distribution of Questionnaires and Collection of Minimum Data Set 16

3.5 Data Management

3.6 Data Analysis 16
  3.6.1 Management of Missing Data 16
  3.6.2 Statistical Analysis 16
  3.6.3 Categorisation of Children by Disease Group

4. Results 18

4.1 Services 18

4.2 Minimum Data Set 18
  4.2.1 Demographic Information 21
  4.2.2 Gender and Ethnicity 22
  4.2.3 Geographical Distribution of children, young people & families

4.3 Spatial Analysis (Mapping) of the Minimum 23
  4.3.1 Background 23
  4.3.2 Methods 24
  4.3.3 Results 27
  4.3.4 Discussion and Conclusion 42

4.4 Findings from the Questionnaires 43
  4.4.1Findings from the Parent’s Questionnaires 43
    4.4.1.1 Demographics 44
    4.4.1.2 Analysis of the Measure of Processes of Care for Parents (MPOC-56) 46
    4.4.1.3 Families’ Met and Unmet Needs 49
  4.4.2Findings from the Bereaved Parent’s Questionnaire 50
  4.4.3Findings from the Staff Questionnaire 51
    4.4.3.1Demographics 51
    4.4.3.2. Measure of Processes of Care for service Providers (MPOC-SP) 52

4.4.3.3 Staff Perceptions of children's and Families Met and Unmet Needs	55
4.4.4Findings from the service Manager Questionnaire	57
4.4.4.1 Geographical Distribution	57
4.4.4.2 Staffing	58
4.4.4.3 Service Provided	58
4.4.4.4 Limitations of the Services	58
4.4.4.5 Ambitions for the Future	59
4.5 Responses to the Open Ended Question	59
4.5.1Methods	59
4.5.2 Findings	59
4.5.3 Analysis and Discussion	60
4.5.4 Conclusion	64
5. Discussion and Conclusion	66
5.1 Summary of Findings	66
References	68-72

Tables

Table 1: Categorisation of children by disease group

Table 2: Age by disease group for 1180 children with a life limiting condition

Table 3: Age by disease group for 133 children who had died from a life limiting/life threatening condition

Table 4: Gender and Ethnicity of 1180 children living with a life limiting condition

Gender and ethnicity of 133 children who had died from a life limiting condition

Table 6: Ethnic background of children according to PCT cluster area in which they live

Table 7: Distribution of children across the five PCT cluster areas according to disease group

Table 8: Example of PCT amalgamation and use of collapsed categories for deprivation ranks. Bold figures are the final areas used.

Table 9: Distribution and response rates for questionnaires.

Table 10: Disease categories of children whose parents completed the parent questionnaire.

Table 11: Number of families returning parent's questionnaire by PCT cluster area in which they lived

Table 12: Mean score and standard deviation for the five factors of the MPOC-56 by PCT cluster area in which the families lived.

Table 13: MPOC-56 Proportion of families in each PCT cluster area who had an average factor score of less than 4.

Table 14: MPOC domain scores by responses at and above and below the median of 6 on the question 'Do you have a named person to contact if you need help or advice?' 1 is never, 7 is a great extent.

Table 15: Proportion of parents reporting met and unmet needs ordered by largest unmet need.

Table 16: Proportion of families' ratings of unmet needs by PCT cluster area.

Table 17: Disease categories of children who died in the 11 months up to end April 2010.

Table 18: Proportion of families (n=21) for whom the listed needs were met or not sufficiently met.

Table 19: Demographics from the staff questionnaire.

Table 20: MPOC-SP Mean (SD), median and range of scores from 260 staff working with children with life limiting conditions in the West Midlands.

Table 21: Descriptive statistics for the four MPOC-SP domains (n=260 practitioners).

Table 22: Differences in ratings between members of staff who rated the behaviours undertaken.

Table 23: Percentage of practitioners scoring children's and families unmet needs according to geographical distribution by PCT cluster area.

Table 24: Themes identified from qualitative comments from parent, bereaved parent and staff questionnaires.

Table 25: Service Manager's description of current staff employed in the care of children and young people with life limiting conditions and their families.

Figures

Figure 1: Disease groupings of children (n = 1180) with life limiting conditions known to services at end of April 2011.
Figure 2: Disease groupings of 131 children who had died 13 to 24 months previously.
Figure 3: Age Distribution of 1313 children of whom 1180 were living with life limiting conditions (age is calculated at end of April 2011) and 133 who had died in the previous
Figure 4: West Midlands region showing PCT areas and hospice locations.
Figure 5: Numbers of children from the minimum data set living in the West Midlands area.
Figure 6: Numbers of children from the minimum data set living in the West Midlands area by PCT area.
Figure 7: Rates for life limited children compared with the usual population of children in the West Midlands.
Figure 8: Rates of life limiting conditions, per 10,000 children. PCT areas.
Figure 9: Boys, rates with life limiting conditions per 10,000 children. Postcode sectors.
Figure 10: Girls, rates with life limiting conditions per 10,000 children. Postcode sectors.
Figure 11: Boys, rates with life limiting conditions per 10,000 children. PCT areas.
Figure 12: Girls, rates with life limiting conditions per 10,000 children. PCT areas.
Figure 13: PCT areas identified by rank of their deprivation scores.
Figure 14: Average age of life limited children, West Midlands.
Figure 15: Proportion of all children under the age of one year by PCT.
Figure 16: Proportion of children with life limiting conditions under the age of one.
Figure 17: Proportion of life limiting conditions 1-4 years old.
Figure 18: Proportion of life limiting conditions 5-9 years old.
Figure 19: Proportion of life limiting conditions 10-14 years old.
Figure 20: Proportion of life limiting conditions 15-19 years old.
Figure 21: Proportion of life limiting conditions over 20 years.
Figure 22: All children of ethnic minority background by PCT
Figure 23: Children with life limiting conditions and ethnic minority backgrounds
Figure 24: Children with life limiting conditions and ethnic minority South Asian backgrounds.
Figure 25: Distribution of congenital and chromosomal disorders in children
Figure 26: Distribution of CNS progressive disorder in children.
Figure 27: Distribution of cancer in children
Figure 28: Distribution of neuromuscular disorders in children.
Figure 29: Distribution of pulmonary disorders in children
Figure 30: Distribution of static encephalopathy disorders in children
Figure 31: Numbers of children who died during 2010.
Figure 32: Proportion of children with life limiting conditions.
Figure 33: Proportion of children who died during 2010 out of all the children with life limiting: condition as a percentage of all childhood deaths
Figure 34: Buffer zones around hospice facilities-distance to travel

1 Introduction

Understanding the demographic and geographical distribution ofchildren with life-limiting conditions and their families.

This is a report on Strand 1 of the Big Study which explored the demographics, epidemiology and geographical distribution of children and young people with life-limiting conditions and their families and health care services caring for them in the West Midlands. Strand 1 research was conducted by researchers from the School of Health of University of Central Lancashire in Preston.

1.1 Background

In 2008 the then government set out the future direction for children's palliative care in their publication 'Better Care: Better Lives' (DH 2008)[policy now defunct!]. The timing of our research was linked to this strategy, which called for strong commissioning networks and a deeper understanding of local population needs.

The provision of palliative care services for children and young people with life limiting and life threatening conditions continues to undergo changes in policy, commissioning and delivery. The last two decades has brought about significant growth in the provision of palliative care services for children. Such growth will have helped many children and families, however, rapid growth based on a limited information base can result in supportive care that can be fragmented and difficult to obtain, with poor coordination and communication between services, meaning that some families continue to have unmet needs (Hunt, Elston and Galloway 2003; Danvers, Freshwater et al 2003; Carter and Petchey 2006).

In this Chapter we use the term 'Life-limiting Condition/s' to include those children who have a life-threatening condition and are receiving treatment aimed at cure, but whose parents and carers are aware that they may not survive childhood.

Services for children with life limiting conditions are known to be patchy and the need for a UK-wide mapping initiative has been acknowledged (DH 2011; ACT and RCPCH 2009). However, it remains problematic to establish accurate data on those children requiring palliative care and ascertaining where they live. Although, this research is not collecting data from this population UK-wide, the region where the data will be collected from contains both rural and urban areas and it has a diverse ethnic community.

Hatton, Akram, Shah et al (2004) believe that the numbers of children requiring palliative care from minority ethnic communities is rising and will continue to do so. Previous research has also identified that there can be an assumption that the needs of these families are met within their own community, however the evidence suggests that their needs are often unmet (Hendry 2006; Gatrad, Brown and Sheikh 2008).

2 Aims of Strand 1 Study

The aims of this Strand of the Study were to:

1. Identify services and survey their perceptions of their own effectiveness and that of their area in meeting the needs of children and young people with life-limiting conditions and their families. Identify children and families receiving services from the above organisations and survey their perceptions of their met and unmet needs
2. Collect a common minimum data set (MDS) of children with life-limiting conditions known to the services.
3. Map the distribution of the children and the services and examine that distribution in relation to urban and rural areas, areas of deprivation, and distance from children's services, in particular hospices for children.
4. Identify any harder to reach areas where children and families do not appear to be receiving identified services, and explore the extent of unmet need.

3. Methods

3.1 Recruitment of participating services and families

Eligible services were organisations in the West Midlands Strategic Health Authority region that provided support to children with life limiting conditions and their families. Services were identified by a process of snowballing. Prior to the start of the study, consultations were undertaken with children, young people, parents and professionals caring for children with life limiting conditions. Six health professionals with a special interest in children's palliative care were involved in this consultation and these were the starting point for identifying other key individuals providing support for children and young people with a life limiting condition and their families in the West Midlands.

Eligible families were those who had a child or young person with a life-limiting condition who accessed child care services in the West Midlands. Parents should already have been informed that their child's illness was one that could shorten his or her life expectancy, either in childhood or early adulthood. Children would normally be aged from 0-18 years but young people over 18 could be included if they were still in receipt of child health care services, including children's hospice services. Bereaved parents could also be invited to participate if their child had died from 13 months to 24 months prior to the invitation.

3.2 Questionnaires and Instruments

Four questionnaires were designed for distribution to service managers, service staff, parents and bereaved parents.

3.2.1 Parent's Questionnaire:

This requested demographic information about the child and family. It included a slightly adapted version of the 56 item Measures of Processes of Care (MPOC-56) (King, Rosenbaum & King, 2006) and a list of 'Met and Unmet Needs', this list being derived through the consultation which was held prior to the Big Study, from the literature and professional experience of the researchers.

The MPOC-56 (King, Rosenbaum and King 1996) was developed as a measure of parent's perceptions of the extent to which the health service they and their child received is family centred. The MPOC-56, distributed by the *CanChild* Centre for Childhood Disability Research, McMaster University in Canada is widely used in Children's Disability and Rehabilitation Services and has good validity and reliability (King, Rosenbaum & King, 1997). The MPOC-56, with minor changes, was used to evaluate children's disability community services in the UK (McConachie & Logan 2003), and in an evaluation of a Children's Palliative Care service in York (Whitton et al 2003). A shorter version, the MPOC-20 has been developed (King, King & Rosenbaum, 2004).

We chose to use the longer version as there is currently limited data on use of the measures in children's palliative care services. Each of the 56 items is presented under a common question: "To what extent do the people who work with your child..." (for example) "take the time to get to know you and your child?" A 1 – 7 response scale was used. Rather than the 7 headings used by the developers, the 7 points were headed at one end with 7 indicating that the service provider engaged in this behaviour "to a great extent" and 1 = "Never" at the other end. A heading midway indicated "Sometimes". A score of 0 indicated that the item was "not applicable". This slightly different format from the original allowed the questionnaire to be presented in Portrait on A4 paper, rather than in landscape where the number of pages might deter parents from completion.In addition to the above, very minor changes were made to the wording of some of the items in that where items referred to 'treatment' this was, where more appropriate, replaced by 'care'. One item that asked two questions within the one question were split in to two questions, and then the average of the two scores was taken. There is no total score, but analysis of each respondent's data yields five scores, one for each of five factors or scales, these five factors being:

- Enabling and Partnership
- Providing General Information
- Providing Specific Information about the Child
- Coordinated and Comprehensive Care for the Child and Family
- Respectful and Supportive Care

We did not examine the data for alternative factors. A factor score is obtained by computing the average of the items' ratings. The MPOC Manual (http://www.canchild.ca/en/measures/resources/MPOCManual_short.pdf) provides information on which items fall in to which factors and the management of missing item/s within each factor. A score could still be obtained for individual scales / factors where there were no more than a third of items in that scale are missing. If more than 50% of the total items are missing or invalid, that individual's responses are omitted altogether.

3.2.2 Bereaved Parent's Questionnaire:

This was similar to the above but did not include the MPOC measures.

3.2.3 Staff Questionnaire:

This questionnaire included the Measure of Processes of Care for Service Providers (MPOC_SP) (Woodside, Rosenbaum, King, & King, 1998). The MPOC_SP is a well validated tool and has been used in a variety of settings including Australia (Dyke et al., 2006) and Wales (Pickering & Busse, 2010) to help professionals reflect on their own practice. It was used in the UK to evaluate a local service for children with palliative care needs (Whitton et al 2008). The MPOC-SP assesses whether the organisations perceive themselves to be delivering family-centred care on a number of dimensions. Similar changes were made to the headings as for the Parent's version, but no changes were made to the items. Analysis of the data yields four factors, these being:

- Showing interpersonal sensitivity
- Providing general information
- Communicating specific information about the child
- Treating people respectfully

We did not examine the data for alternative factors. In addition to the MPOC-SP, participants were asked to what extent they thought that specific needs of families and children in the service they worked for were being met on a 7 point Likert scale (1 – 7, with 1 being 'not at all' and 7 being 'more than needed').

Free Text: On the final page of the parent's, bereaved parent's and staff's questionnaire there was an opportunity to identify three things they would change about the services received or provided. In addition, parents were asked what might make (or have made) the biggest difference to the quality of their family life.

3.2.4 Service Manager's Questionnaire:

The Service manager's questionnaire asked more generally about the area covered by their service, levels of staffing, their funding, limitations in their service and ambitions for the future.

3.2.5 Minimal Data Set:

The minimal data set was requested from services for children in their care. This consisted of five items: date of birth, diagnosis, partial post-code (omitting last two letters), ethnicity and gender. Age as of end April 2011 was calculated from date of birth. Unless parents had opted out, the child's MDS was forwarded by service providers to the researchers by encrypted file transfer.

3.3 Inclusion of non-English speaking families

Due to the diverse ethnic communities in the West Midlands the research team worked with each participating centre to ascertain the main languages spoken in their area. With support from a well- established language translation service in the UK, a translation box for Gujarati, Urdu, Bengali, Punjabi, Arabic and Mirpuri was provided on the approach letters. This box outlined that if the family required a translator or interpreter they could call the Language Service on a Friday afternoon between 2 pm and 5 pm when someone would be available to help them. This service was made available for four months, throughout the time of the questionnaire distribution.

3.4 Distribution of questionnaires and collection of minimum data set

Lead collaborators at participating centres identified the families by screening the service records to identify potential/eligible children and families for the study. Questionnaire packs were taken to each participating centre then forwarded to eligible families either by post or hand delivered. Each pack forwarded consisted of an approach letter from the service, a parent questionnaire which incorporated the information sheet, a reply slip, an opt-out slip and two reply paid envelopes.

One of the reply slips could be returned to the service distributing the pack if the parents did not want the Service to provide the researchers with their child's minimum data set. Outlined in the approach letter to the family was the information that if the opt-out form was not returned to the service within one month the minimum data set would be provided to the researchers.

The second reply slip could be used by parents to indicate if they were interested in taking part in any other of the study activities, these being parent's interview, child's and young person's interview, children's and young people's art activities, parent advisory group and child and young person advisory group (See Strand 2). This reply slip was returned by parents to the research team in the reply paid envelope along with the completed questionnaire. The slip was then forwarded to Strand 2 research team by encrypted file transfer.

3.5 Data management

Questionnaires returned to the research team were allocated a unique ID number. Each participating centre had been allocated a unique identification code and this code was written on the reply paid envelopes so that it was possible to confirm how many were returned through each participating centre. The data from each questionnaire was then entered into the relevant Access database that had been designed specifically for the study in association with the York Health Economics Consortium (see Strand 4). Relevant data was transferred to the Strand 4 research team by encrypted file transfer.

3.6 Data analysis

3.6.1 Management of missing data

The Staff Questionnaire contained the 27-item Measure of Processes of Care for Service Providers (MPOC-SP) (Woodside et al 2001). Of 264 respondents three who had recorded over 50% of the items as 'not applicable' or over 50% of item responses were missing. These three were removed from the analysis of the MPOC-SP. In all others missing items were recoded using the individual's mean score for the series. We considered that where the response was 'not applicable' this would imply that the behaviour was not taking place and therefore 0 for 'not applicable' was recoded as 1, indicating 'not at all'.

3.6.2 Statistical analysis

Data was transferred for analysis from Access to SPSS vs 19. Averages are reported as mean and standard deviation (sd), and as median when data is skewed. For most purposes MPOC scores are treated as parametric data. Differences between groups are explored using Analysis of Variance and Independent-sample t-tests. Chi-square tests are used when examining differences between observed and expected in proportion by PCT Cluster area.

3.6.3 Categorization of children by disease group.

Children were catagorized by ICD10 and then further in to disease groups that would be meaningful for practitioners. Table 1 provides a summary of the disease groups used in this analysis.

Table 1 Categorization of Children by Disease group

Disease group	Description of conditions included in disease group	ICD10 codes
Congenital and chromosomal	Includes children with chromosomal conditions such as Downs, Pattau, Edwards Syndromes and other less common abnormalities. Also children with congenital abnormalities of the central nervous system such as lissencephaly, hydrocephalus, microcephaly. Also children with congenital heart disease, short bowel, biliary atresia.	Mainly Q codes. (Congenital malformations, deformations and chromosomal abnormalities) Some K codes (Diseases of the digestive system).
CNS Static encephalopathies	Non-progressive CNS disease including cerebral palsy, developmental delay and epilepsy. Brain injury, Birth asphyxia, Hypoxic ischaemic encephalopathy	Mainly G codes (Diseases of the nervous system). Some P codes (Certain conditions originating in the perinatal period) e.g. “Brain injury” / “birth injury” “hypoxic ischaemic encephalopathy”
CNS progressive	Disease often categorized as ‘Progressive Intellectual and Neurological Deterioration (PIND)’ characterized by loss of skills. Includes children with mucopolysaccharidoses (Hurlers, Hunters, Sanfilippo), lipofuscinosis (Juvenile, Late infantile and Infantile Battens), leucodystrophies (Adrenoleucodystrophy, metachromatic leucodystrophy, Krabbes), Retts, Juvenile Huntington’s. Most conditions in this group are inherited as single gene disorders.	Mainly E codes (Endocrine, nutritional and metabolic diseases). Some G codes (Diseases of the nervous system).
Neuromuscular	Duchenne muscular dystrophy, Spinal muscular atrophy, Congenital muscular dystrophy, Ataxia telangiectasia. Friedreich's ataxia. These are also inherited as single gene recessive or x-linked conditions.	Mainly G codes (Disease of the nervous system).
Cancer	Solid tumours, Brain tumours, Cancer of blood and lymphatic systems	C codes (Neoplasms)
Pulmonary / respiratory	Cystic Fibrosis (single recessive gene disorder), Chronic lung disease (sometimes resulting from prematurity)	Cystic fibrosis E 84.9. Chronic lung disease J98.4
Other	Endocrine and renal disorders. Immunodeficiency. Trauma for instance due to road traffic accident	B & D (Immunodeficiency) E (endocrine), K (digestive), M (musculoskeletal), N (genitourinary), S & T (Injury and trauma).

4 Results

4.1 Services

The use of a snowballing technique enabled the research team to identify 18 NHS centres, six Hospices and one complimentary therapy centre to invite to become Participant Identification Centres (PIC) for the study. The NHS centres included both acute and community services. For reporting purposes, three of the hospice services were counted as a single centre, because the distribution of the questionnaires to families was coordinated by the Head Office of that hospice. 2 of the NHS centres identified chose not to take part.

The identified lead collaborators included doctors, nurses and hospice heads of care. There were a higher number of community children's nurses taking the lead collaborator role with only 4 lead collaborators being Community Paediatricians and one being a Medical Director. Each PIC was supported throughout the study by a Research Assistant.

4.2 Minimum Data Set

4.2.1 Demographic Information

1180 children were identified who were living at the time the data was supplied. 131 children (10% of the total of 1313 if the number of deceased children were included) were identified who had died between 13 and 24 months previously. It could be expected that a further 150 children may have died from a life-limiting condition in the year to end April 2011. We were not able due to ethical constraints to obtain information on this group of children. It is expected that services will be following-up families of children who died in the past two years and, therefore, the number of families in their care would be of the order of 1463 - 1500.
Figures 1 and 2 show the populations of living children, and children who died up to end April 2010 by the disease groups allocated by the researchers from the information provided by the services.

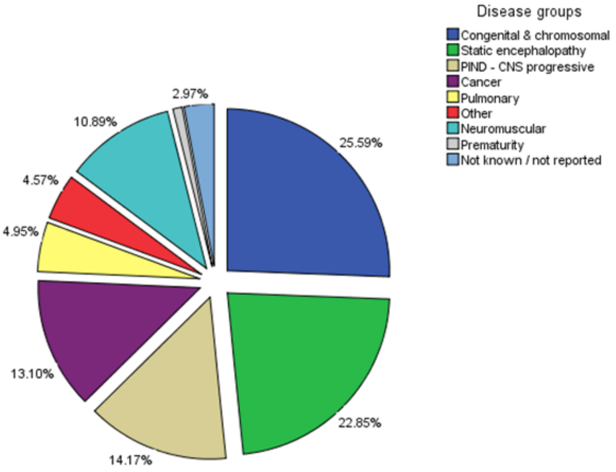


Fig 1. Disease groupings of children (n=1180) with life-limiting conditions known to services at end of April 2011

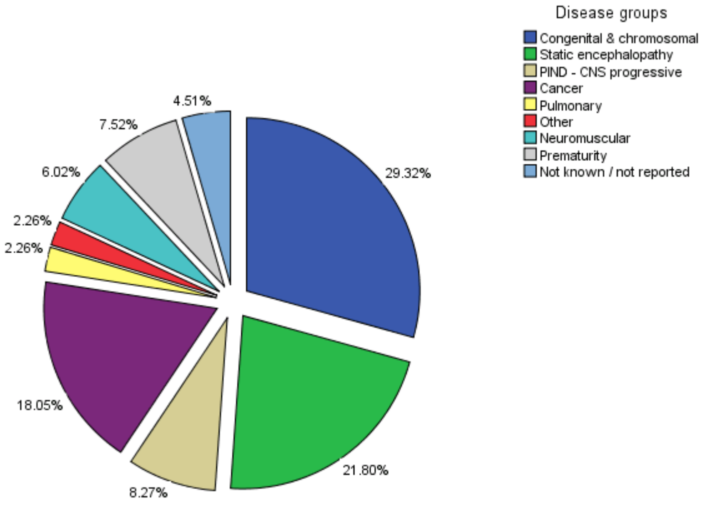


Fig 2. Disease groupings of 131 children who had died 13 to 24 months previously

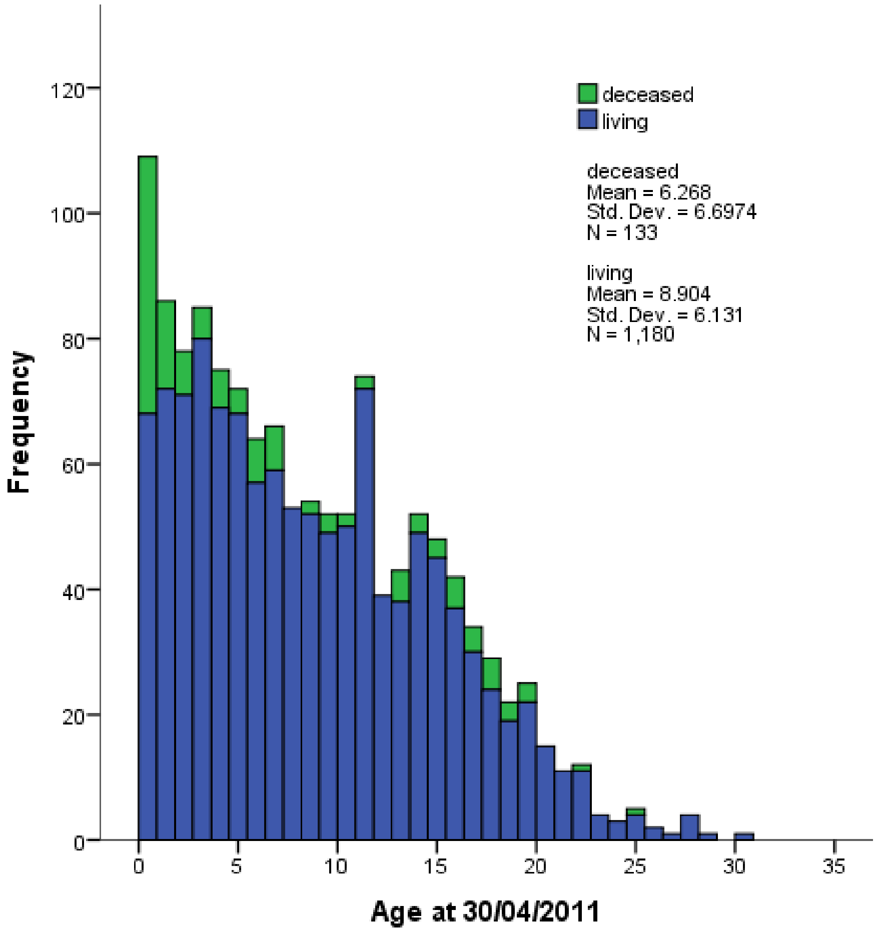


Fig 3. Age distribution of 1313 children of whom 1180 were living with life-limiting conditions(age is calculated at end of April 2011) and 133 who had died in previous year (age calculated at end April 2010).

The mean (SD) age of living children was 8.9 (6.1) years. Median age was 8.0 years. The central 80% of children lay within the 90th percentiles of 1.6 and 17.3 years. The figures suggest that approximately 10% of the children with life-limiting disease may die per year. The mean (SD) age was 6.3 (6.7) years. Median was 3.5 years. The central 80% of children lay within the 90th percentiles of 1.1 and 17.3 years. The ages of the two groups (living and deceased children) are shown by Disease Group in Tables 1 and 2.

Table 2. Age by Disease Group for 1180 children living with a life-limiting condition

Disease groups	n	Mean age (SD) Years	Median age, Years	Age range Years
Congenital & chromosomal	297	7.7 (6.4)	5.7	0 – 26
CNS Static encephalopathy	271	9.2 (5.8)	8.1	0 – 28
CNS progressive	175	9.3 (5.9)	9.0	0 – 27
Cancer	148	8.0 (4.7)	6.9	2 – 22
Neuromuscular	135	13.9 (6.3)	14.0	0 – 30
Pulmonary	62	7.2 (5.2)	6.0	0 – 17
Other	57	6.9 (4.8)	6.9	0 – 18
All	1180	8.9 (6.1)	8.0	0 – 30

Prematurity and Not known groups are included in the total (All) but not the disease group listing.

Table 3. Age by Disease Group for 133 children who died from a life-limiting / life-threatening condition.

Disease Groups	n	Age (years) Mean (SD)	Median Years	Age range Years
Congenital & chromosomal	39	4.1 (6.3)	1.0	0 – 22
CNS Static encephalopathy	29	9.2 (6.0)	2.0	0 – 25
CNS progressive	11	5.0 (6.2)	8.7	0 – 18
Cancer	24	8.4 (6.0)	6.3	0 – 19
Neuromuscular	8	11.2 (9.0)	15.4	0 – 19
Pulmonary	3	4.8 (7.5)	0.5	1 – 13
Prematurity	10	0.4 (0.3)	0.5	0 – 0.7
All	133	6.3 (6.7)	3.5	0 – 25

Other and Not known groups are included in the total (All) but not the disease group listing.

4.2.2 Gender and Ethnicity

Table 4. Gender and ethnicity of 1180 children living with a life-limiting condition

Disease Groups	n	Gender M:F %	White British %	South Asian %	Black	Mixed	Other %	Not known %
Congenital & chromosomal	297	47 : 53	59	29	3	3	2	4
CNS static encephalopathy	271	54 : 45	58	25	4	6	4	3
CNS progressive	175	46 : 53	45	41	2	1	5	6
Cancer	148	61 : 36	69	16	2	2	3	8
Neuromuscular	136	80 : 20	64	24	1	3	4	4
Pulmonary	54	65 : 34	73	16	2	2	3	8
Other	57	63 : 37	47	26	16	7	0	4
Not known	33	58 : 33	58	18	3	0	3	18
All	1180	56 : 43	59	27	3	3	3	5

Table 5: Gender and ethnicity of 133 children who had died from a life-limiting condition

Disease Groups	n	Gender M:F %	White British %	South Asian %	Black %	Mixed %	Other %	Not known %
Congenital & chromosomal	39	64 : 36	62	26	0	5	0	8
CNS static encephalopathy	29	52 : 48	48	41	0	7	0	3
CNS progressive	11	73 : 27	18	64	0	9	0	9
Cancer	24	63 : 29	75	0	0	4	0	21
Neuromuscular	8	63 : 38	64	24	1	3	4	4
Pulmonary	3	33 : 33	100	0	0	0	0	0
Prematurity	10	70 : 30	60	30	10	0	0	0
Other	3	0 : 100	33	67	0	0	0	0
Not known	6	83 : 17	17	33	17	33	0	0
All	133	63 : 35	55	29	2	7	1	8

4.2.3 Geographical distribution of children, young people and families

The table below (Table 6) provides the ethnic background of children according to the PCT Cluster area in which they live.

Table 6 Ethnic background of children according to PCT Cluster area in which they live

	Arden	Birmingham & Solihull	Black Country	Stafford-shire	West Mercia	All
White British and Irish	93	155	113	152	251	764
%	75	33	55	66	89	58
Asian and British Asian	17	222	70	26	17	352
%	14	47	34	11	6	27
Black and Black British	2	33	5	0	0	40
%	2	7	2	0	0	3
Mixed	5	22	12	4	4	47
%	4	5	6	2	1	4
Other	7	21	4	1	6	39
%	6	4	2	0	2	3
Not known	0	17	2	49	3	71
%	0	4	1	21	1	5
Total	124	470	206	232	281	1313

The areas vary considerably in the proportion of families from Black and Minority Ethnic Groups, with the proportion of South Asian families in the Birmingham and Solihull PCT Cluster area being particularly high.

In Table 7 the disease group of children is presented by PCT Cluster in which they live.

Table 7 Distribution of children across the five PCT Cluster areas according to disease group.

	Arden	Birmingham and Solihull	Black Country	Stafford-shire	West Mercia	All
Congenital & chromosomal	28	133	52	49	74	336
%	23	28	25	21	26	26
Static encephalopathy	23	111	44	49	73	300
%	19	24	21	21	26	23
CNS progressive	10	90	29	28	29	186
%	8	19	14	12	10	14
Cancer	25	34	15	48	50	172
%	20	7	7	21	18	13
Neuromuscular	17	40	34	18	34	143
%	14	9	17	8	12	11
Pulmonary	14	15	4	21	11	65
%	11	3	2	9	4	5
Other	3	25	19	17	8	72
%	2	5	9	7	3	5

Not known	4	22	9	2	2	39
%	3	5	4	1	1	3
Total	124	470	206	232	281	1313

The varying proportions of children with cancer across the areas may be due to reporting differences across institutions.

4.3. Spatial analysis (mapping) of the minimum data set provided by services of children with life limiting conditions known to them.

4.3.1 Background

Communities are defined mostly by their geography and most health and social care services are planned within strict geographical boundaries. GIS is increasingly used in community health assessment to describe, identify and explore the needs of communities in terms of comparison of small and large geographic areas (Faraque et al 2003; Foley 2002; Haynes et al, 2006; Cinnamon et al 2008; Dulin et al 2010). GIS are related spatial analytic techniques which have mostly been used to examine delivery and access to healthcare, allowing alternative measures to the constant churning of census data and analysing and comparing a multiplicity of local and national dimensions of social, geographic and environmental factors (Wrigley et al 2002; Langford and Higgs 2006) but sometimes the process can produce impractical interpretations for practice if statistics are too obscure (Higgs 2004). In order to perform this kind of spatial analysis, a GIS computer based relational database programme is used. The GIS tools consist of a database, with linked map features and definitions that anchor the statistics to geographical boundaries on the map. This helps researchers to organise spatial data (database), visualise spatial data (map) and analyse the data (analysis) (Goodman and Wennberg, 1999). GIS allows us to simplify complex data and to compare organised characteristics between samples geographically, analysis alone does not provide this because sometimes it is easier to practically point to an obvious visual clustering and speculate on causes, once the simplification process has been performed that may not have been obvious without the geographical juxtaposition.

This analysis uses three processes:

A simple description of the distribution of numbers of young people with life limiting conditions between and within areas using the minimum data set collected from services for children in the West Midlands area.
A calculation of the estimated prevalence rate of life limiting illness using publically available national statistics (ONS[3]) to represent the usual West Midlands child population compared with population and demographic subgroups of these life limited children in the West Midlands area.

---

[3] ONS statistics – www.nationalstatistics.org.uk by postcode sector (using boundary data from 2007) and postcode points for services (2006). Most statistics were for 2010 except where indicated.

Estimates for ethnic minorities by age (2010)

Indices of Deprivation (2010) by health hierarchy

This analysis also uses publically available mortality and deprivation statistics to situate the context. A calculation of the proportion of the population of life limited children that are represented in certain demographic subgroups (gender, age groups, ethnic group, diagnostic groups).

4.3.2 Methods

Data sources

Data relating to the demographics of children with life limiting illness known to services providing for their care needs were obtained from services agreeing to provide a minimum data set of partial postcode, gender, ethnicity and diagnosis of the child. Services were asked to provide parents with a 'drop out' option if they did not want their details to be included, but few exercised this right. Criteria for which children were included have been discussed previously with the methods and procedures for security. Data about the children who had died in a single year was also collected (2010).

Cleaning procedures and removal of duplicates were performed as already described. A small number of children lived in areas that were just outside the West Midlands boundary and these were removed from the database, this was mostly in the DE postcode sectors (Derbyshire). Calculations were performed and data were prepared using Microsoft excel and then converted for mapping with the GIS computer programme, Mapinfo professional (V.11).

Hospice services for children's palliative care needs in the West Midlands

Figure 4 shows a map of the West Midlands with labels for each area and hospice location represented as red triangles. Twenty two services contributed a minimum data set for the study but only five of these were hospices. These five hospices serve the entire West Midlands population and also some families outside the border will use these services. Of course, families are not limited to the use of services within the health authority, and if they live near to the border they may well use a service within a reasonable distance. We used a 20m radius to define the distance service users would be prepared to travel to a hospice facility. This estimate was provided by our allied service user advisors. Outside the West Midlands border, there are a number of hospices, but only 2 hospices were just within the 20m distance and the others were much further away. These are represented relatively on the map and the distance estimate was used to estimate the population that were not within a reasonable distance of services.

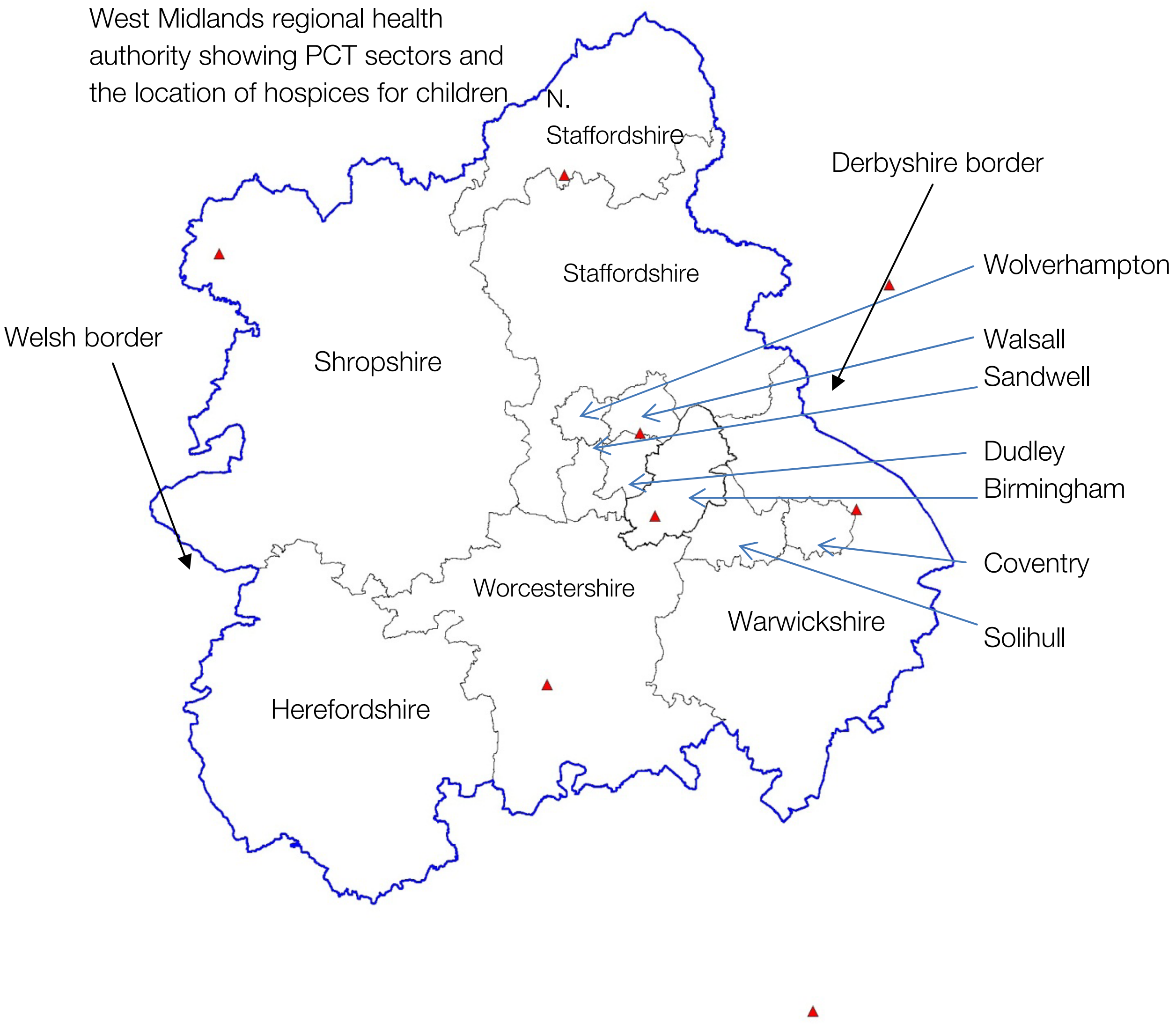


Figure 4. West Midlands region showing PCT areas and hospice locations

Selection of areas for analysis

We used postcode sectors for a small number of maps in order to represent the diversity of rates within small areas, thus it was possible to interpret concentrations of children in a specific section of the map. Rates were the proportion of children identified as having a life limiting illness against all children identified in each area. Some rates were very high because there were few children in the area, such as heavily built up areas. There were no statistics for almost half of the postcode sectors indicating there were no children with LLC in these districts, these were uniformly rural areas and they were left out of the postcode sector analysis because including them would have distorted the information value of the analysis.
The same calculation was performed by PCT to give a more distributed estimate of prevalence.

PCT areas: ONS has produced a compilation of publically available data for the boundaries of the 18 PCTs that cover the West Midlands, however most of these have now been collapsed to the 13 larger health areas defined in 2001 and some statistics are not now available for the smaller areas. In 2012 five clusters were defined within the West Midlands regional health authority, but there are vast differences in the size and density of population of these areas;

some clusters contain very economically and socially disparate sectors. In addition, there are no official statistics specifically to fit cluster areas. ONS and other data collated for PCT areas and hierarchies map quite easily on to the clusters but where ranks are offered and standardised corrections have been made (such as for deprivation statistics or mortality statistics), this data becomes difficult to interpret. In order to retain as much information as possible but also to compare distributions effectively and minimise collation problems in fitting boundaries, we decided to use the 13 health areas established in 2001. These map exactly onto the clusters but are small enough to show differences between urban/rural areas (such as between Birmingham urban environment and the Black Country. However, there are difficulties in loss of information, because for three areas out of the thirteen, mortality and deprivation statistics had to be collapsed from smaller areas. Table 1 shows an example of the collapsed categories for the three larger output areas and the geographic codings. The ONS score is an average of all super output area scores within the PCT. It can be seen that although Heart of Birmingham PCT has the highest score in England and the population is therefore ranked the most deprived for that PCT as a whole, the larger area of Birmingham combining the three small PCTs because it includes the rather better off South Birmingham, is now ranked 13, above that of Sandwell.

Table 8. Example of PCT amalgamation and use of collapsed categories for Deprivation ranks. Bold figures are the final areas used.

Index of Deprivation 2010	PCT 2006	Health Hierarchies PCTs 2001	IMD Average Score	Rank of Average Score
Warwickshire PCT	5PM	QEL	14.77	129
South Staffordshire PCT	5PK	QEJ	15.90	120
Solihull PCT	5QW	QEG	16.38	117
Worcestershire PCT	5PL	QEN	16.44	116
Herefordshire PCT	5CN	QED	17.91	103
Shropshire		QEF	20.00	96
Telford & Wrekin PCT	5MK		23.63	74
Shropshire PCT	5M2		16.36	118
Dudley PCT	5PE	QEC	23.22	77
Staffordshire		QEH	26.23	61
Stoke on Trent PCT	5PJ		34.54	18
North Staffordshire PCT	5PH		17.91	104
	5MD	QEA	28.44	43
Coventry Teaching PCT				
Walsall Teaching PCT	5M3	QEK	31.23	28
Wolverhampton City PCT	5MV	QEM	34.41	20
Birmingham		QD9	38.01	13
South Birmingham PCT	5M1		30.55	30
Heart of Birmingham PCT	5MX		45.31	1
East and North PCT	5PG		38.17	9
Sandwell PCT	5PF	QEE	36.97	12

Terms used for our population

LLC	Children with life limiting conditions or life limited children.
All children	estimated population of children with and without LLC in the area.
Ethnic minorities	Black and racial minorities, estimated population.
Bereaved families	families whose child with a life limiting condition had died (2010).

4.3.3 Results

The results for this part of the analysis will be discussed in terms of the questions that were answered. The shading represents the risk point at the top (large numbers or small numbers depending on the variable examined) and the green or lighter colours at the bottom.

Which areas have the most children with a life limiting condition?

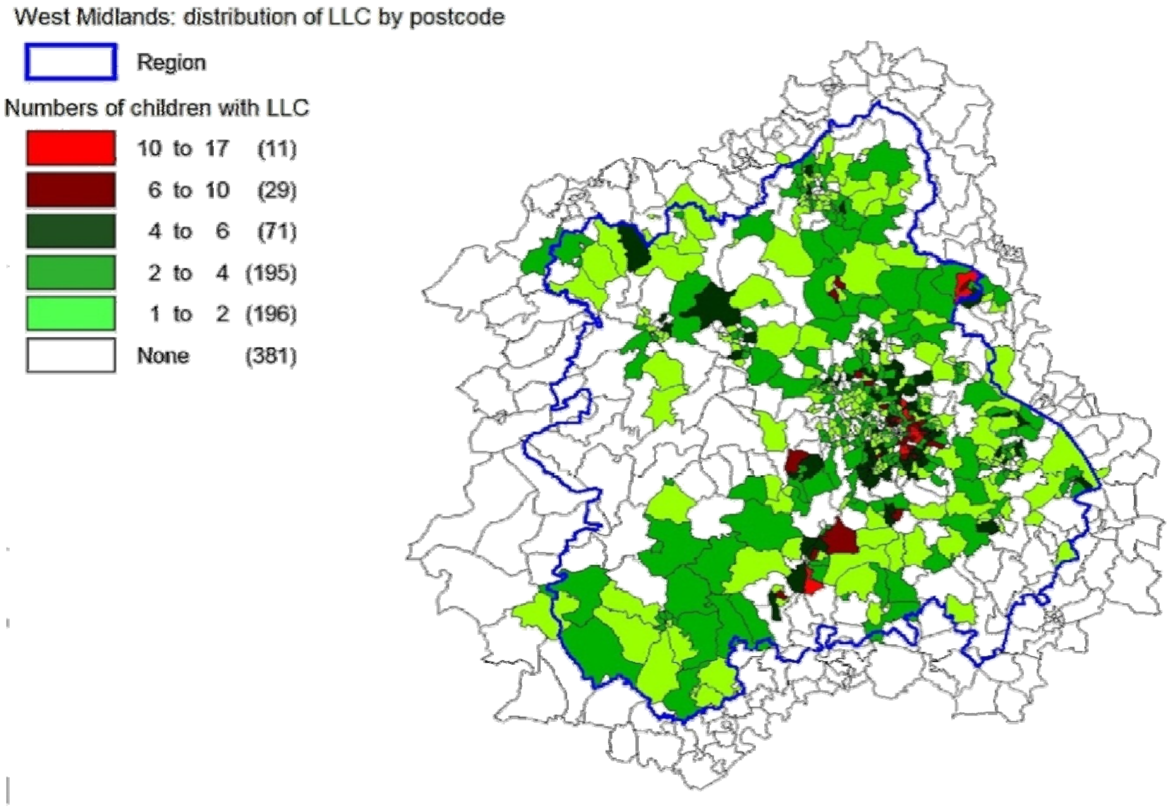


Figure 5. Numbers of children from the minimum data set living in the West Midlands area.

Figure 5 shows the relative distribution of children with LLC in the West Midlands area by postcode sector. It is immediately apparent that there are small pockets of children on the east side of the Birmingham area but fewer on the west. There are clusters around Birmingham and the large centres of population and a puzzling isolated area on the borders of Staffordshire and Shropshire, but also in parts of Herefordshire and borders of Derbyshire. Families in the Derbyshire area (which is represented by the small red ‘hotspot’ on the far right have a distance to travel to a hospice, they might use the Stoke on Trent facility or the facility in Leicester. There are no data for much of Shropshire on the west side, but this is a very rural location. Figure 6 collapses all these small areas into larger areas simplifying the information. The

Birmingham area remains the most populated and a diagonal pattern is easily seen from the Black Country coming down into Worcester. The areas around Birmingham are less populated. However, the small cluster in Shropshire (at the North West) has disappeared.

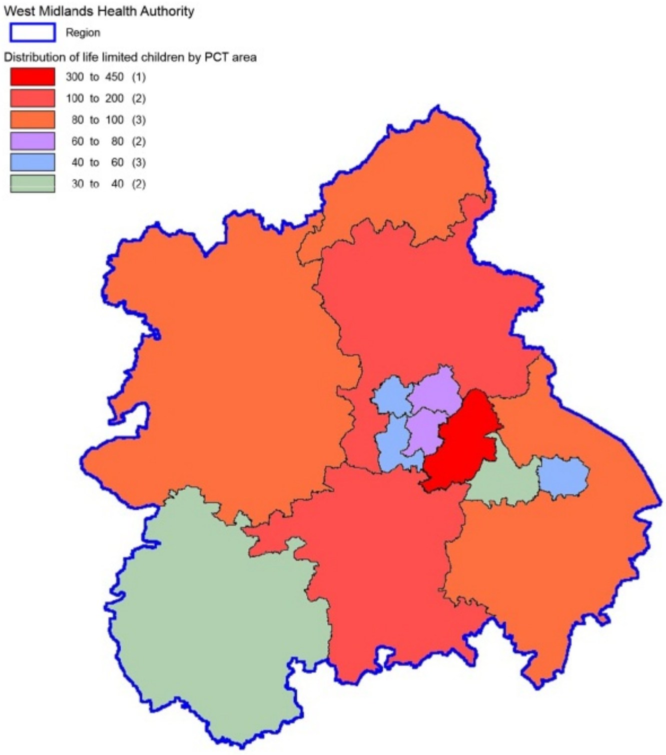


Figure 6. Numbers of children from the minimum data set living in the West Midlands area by PCT area.

There is limited interpretation of this information without comparing whether our figures are consistent with the usual proportion of children living in the area. For this purpose a rate was calculated, using the numbers of life limited children known to services (MDS) compared against estimates from ONS (2010) of the usual proportion of all children and adjusted to reflect the expected number in 10,000.

What is the prevalence of life limiting illness and are conditions distributed proportionately?

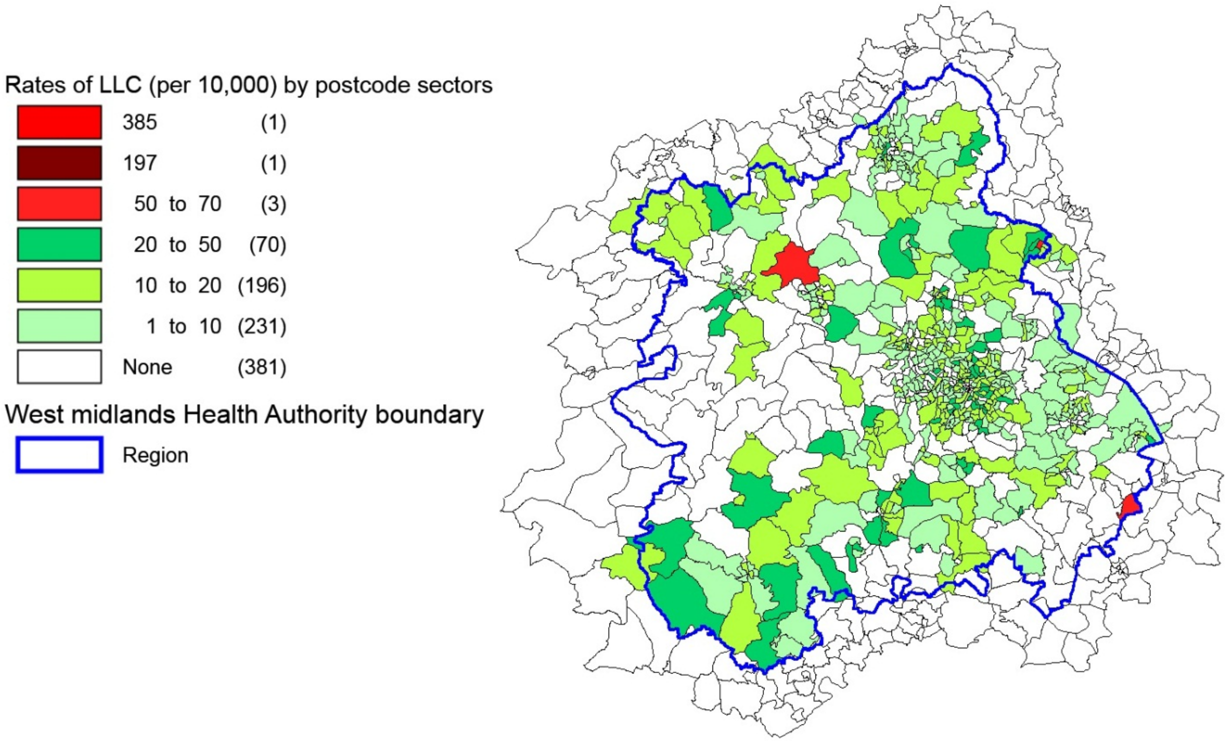


Figure 7. Rates for life limited children compared with the usual population of children in the West Midlands.

If the clusters of life limited children in the figures 5 and 6 simply reflected the variation in numbers of children, this map should be a uniform colour (apart from the areas where there were no identified children). Previous research suggests a rate of 16 in 10,000, but there have been estimates nearer twice the figure (Fraser et al 2012). It was obvious that in rural areas generally, life limiting conditions in children are fewer, the cities contain more children with life limiting conditions relative to the population and there is more diversity in these urban areas. There are a number of very small areas at the top end of the scale within the urban conglomeration. Looking at the table (Table 8) it is apparent that the vast majority of areas have a rate of 1-10 children in 10,000 (which would bring down the average for a larger region), but almost a quarter of areas demonstrate higher rates. Only 75 postcode sectors have a rate higher than 20 children in 10,000. Some of these areas are quite small and others are very rural so they might not have many children resident. Five areas stood out as having a relatively large proportion of children with life limiting conditions compared to the usual population of children. One of these was a small area at the Derbyshire border, another was a larger area at the Worcestershire border and there was a sizeable area at the Shropshire border, which also showed up in figure 5.

For comparison, figures have been collated to PCT area in figure 8, where the diagonal pattern again predominates at the higher levels. However, when collated the rates average out at around 8 to 10 children in 10,000.

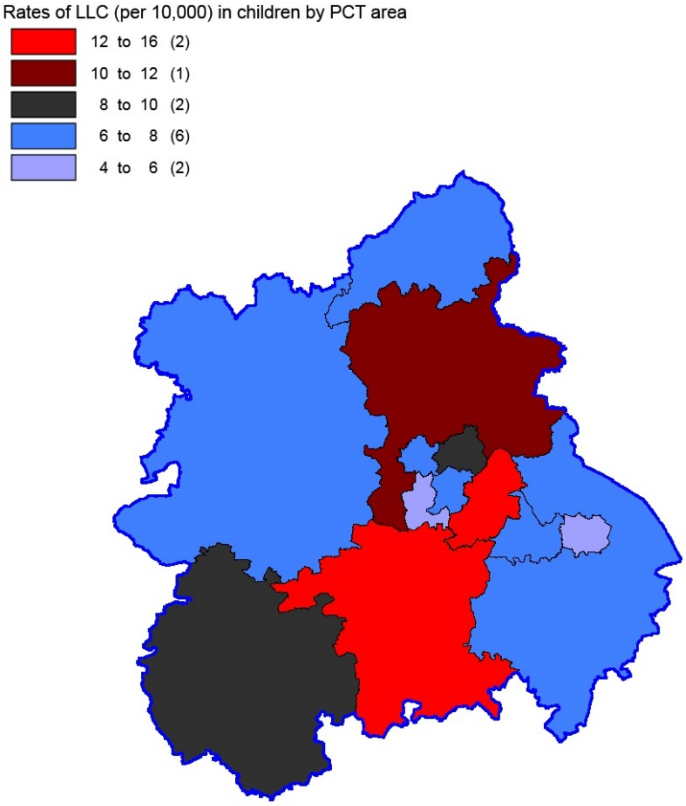
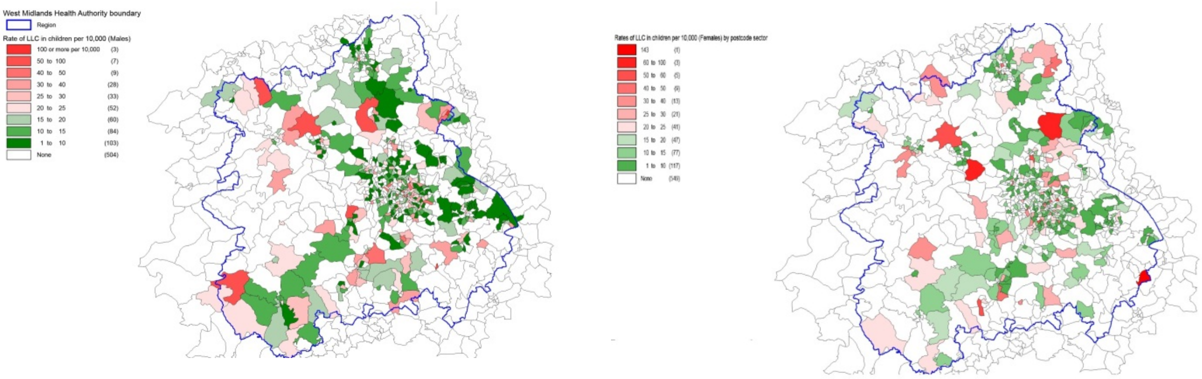
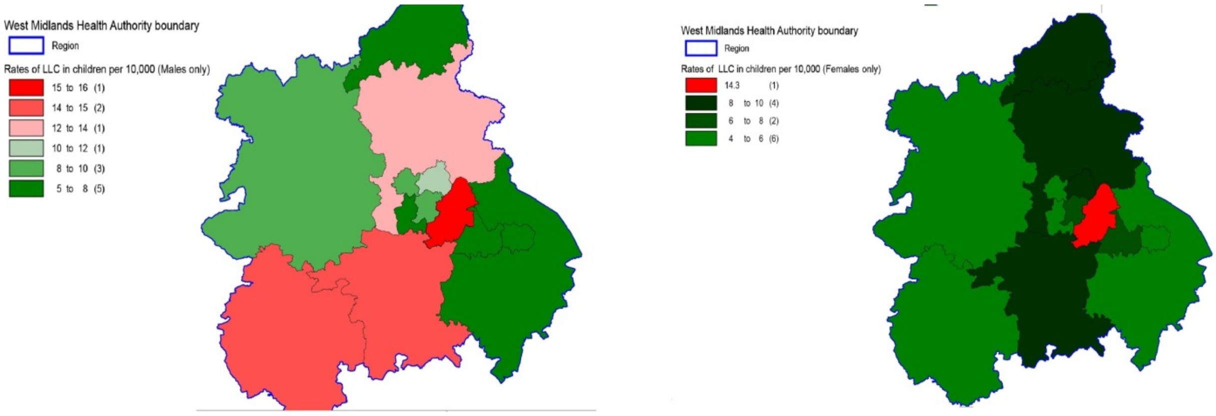


Figure 8. Rates of life limiting conditions, per 10,000 children. PCT areas.

Are there any differences between boys and girls?



Figures 9 and 10, boys and girls, rates of LLC per 10,000 children, postcode sectors.



Figures 11 and 12, boys and girls, rates of LLC per 10,000 children, PCT areas.

Figures 9-12 show the differences in rates for boys and girls with life limiting conditions. The overall average is around 8 -10 per 10,000, the diagonal pattern is shown in both genders but more pronounced in boys as there are more boys overall. A few areas have a large number of boys. Around North Staffordshire some areas have boys but no girls. Herefordshire seems to have a number of boys but fewer girls.

Do areas showing characteristics of deprivation have disproportionate numbers of children with LLC?

Figure 13 shows the rank order for the indices of multiple deprivation applied to PCT areas. There are 151 PCT areas in the UK and the cut points here have been set to indicate quartiles (4 sections: the most deprived quartile being the areas that are ranked 1-38). Birmingham and Coventry appear in the most deprived quartile and certainly the Birmingham area was also identified in every analysis as containing very high numbers of LLC. The areas of Wolverhampton, Sandwell and Walsall however all seem to be similarly identified as deprived whereas they do not stand out as having disproportionately high rates of LLC. The pattern identified in the previous analysis does not contain North Staffordshire, which appears in the second quartile (above average) but it does contain Worcestershire, which is not identified as particularly deprived.

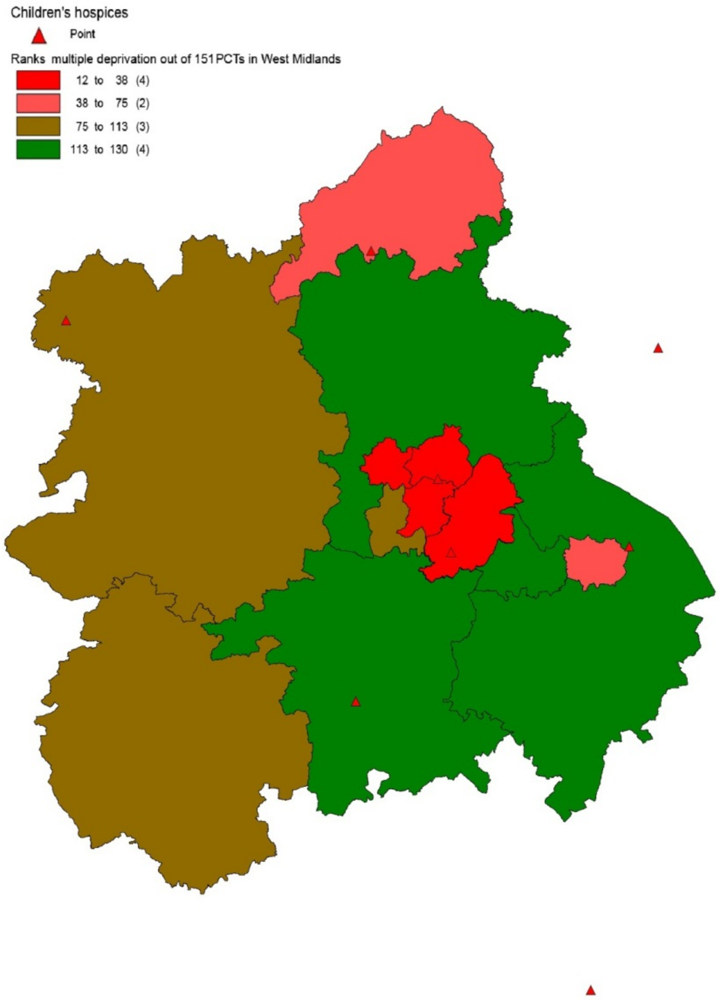


Figure 13. PCT areas identified by rank of their deprivation scores.

Are children with LLC surviving longer in some areas than others?

This is a difficult question to answer because it may be that some areas contain a younger population than others because of migration. Another factor may be that families needing services that are more accessible in the urban environment may relocate to access them. Figure 14 shows that Birmingham has the youngest population of life limited children, closely followed by Staffordshire, Wolverhampton, Sandwell, Solihull and Coventry. Shropshire has more older children on average but because the population is less dense and it is largely rural, there are likely to be fewer in numbers.

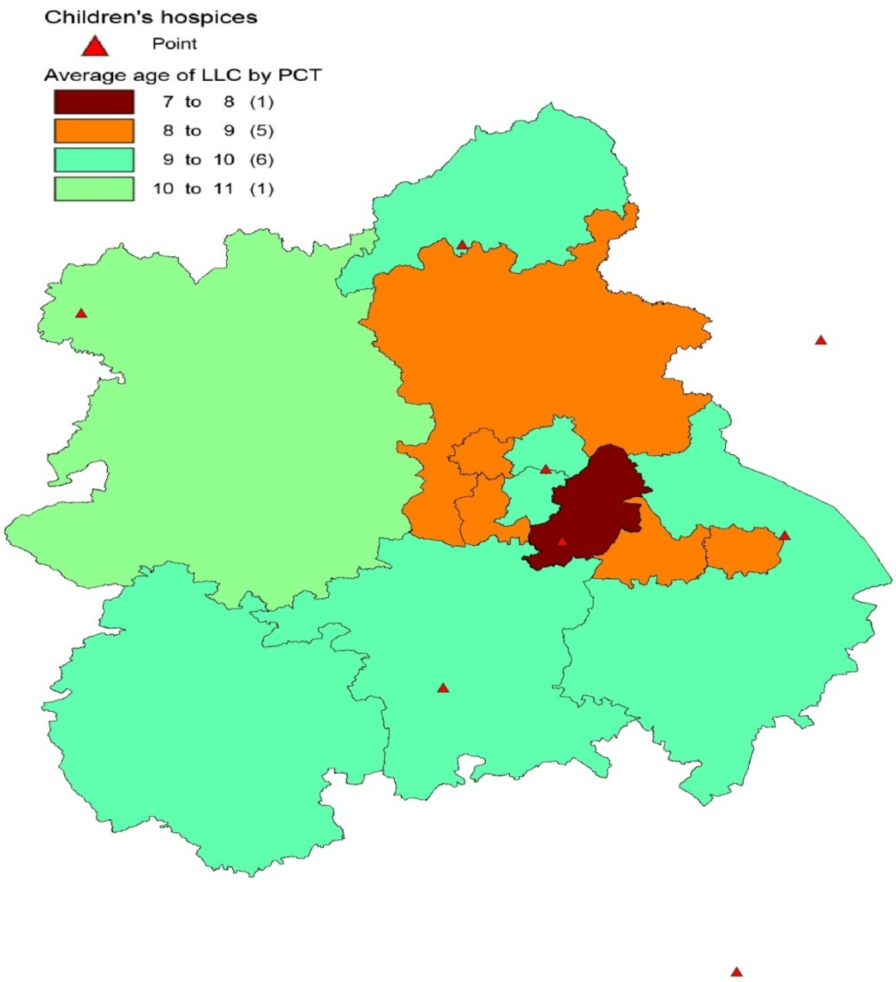


Figure 14 Average age of life limited children, West Midlands.

Figure 15 shows the map of the West Midlands for the proportion of all children under the age of one year. The map should be all one colour if there is no difference between areas; even though numbers may vary, the proportion of babies should be consistent. Looking at the map, there are only very small differences between areas. The small areas around and including Birmingham, and North Staffordshire (which includes the urban city of Stoke on Trent contain proportionately more babies (5 to 6%, against 4 to 5%) than the other areas, although this increase is only slight. When we move on to considering our life limited children, one area stands out above all others, surprisingly, in Walsall, 28 percent of life limited children are babies and Herefordshire and Birmingham 10-15 percent.

The west side of the West Midlands has the most 1-4 year olds (figure 17) with life limiting conditions and stands out among the 10-14 year olds and 15-18 year olds too. Birmingham and Hereford have more 5-9 year old life limited children (figure 18). Only the table for 20 or more years shows a more equitable relationship between areas (figure 21).

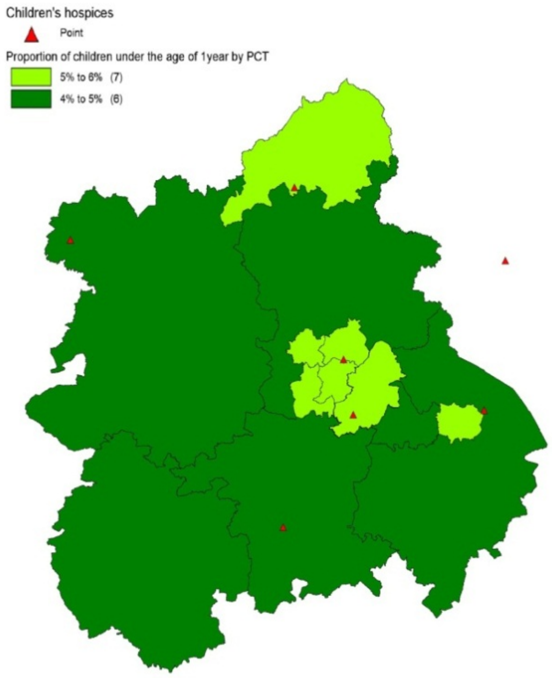


Figure 15. Proportion of all children under the age of one year by PCT

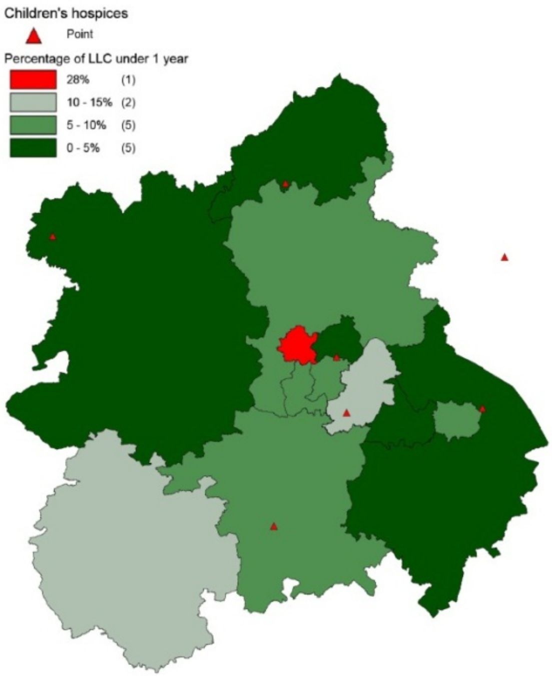


Figure 16. Proportion of children with LLC under the age of one year by PCT

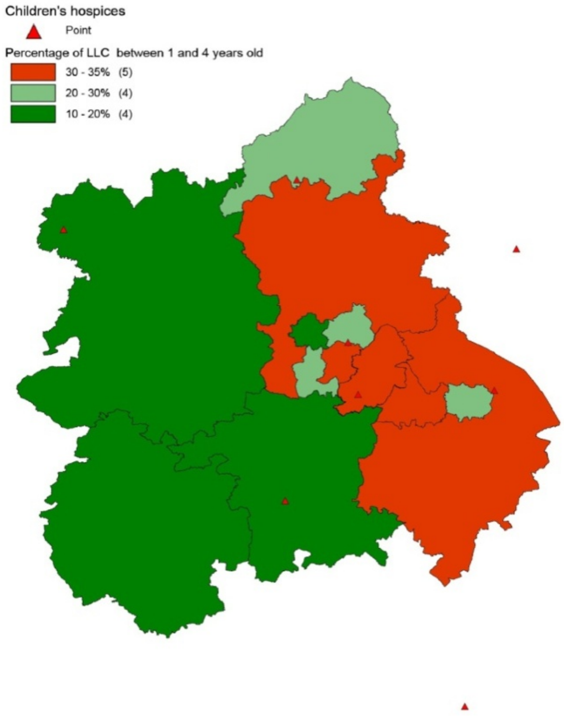


Figure 17. Proportion of LLC 1-4 years old

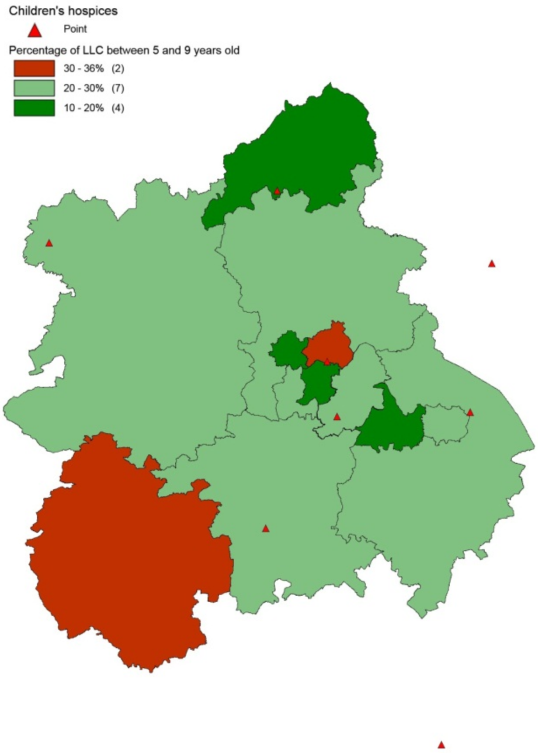


Figure 18. Proportion of LLC 5-9 years old

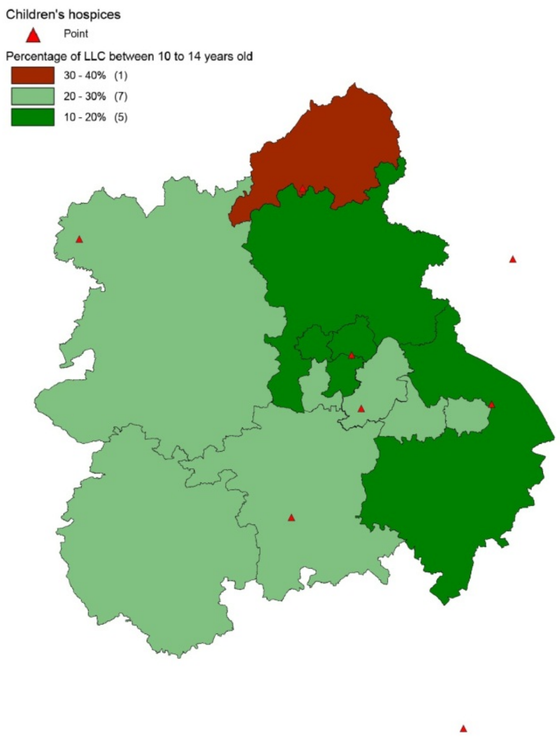


Figure 19. Proportion of LLC 10-14 years old

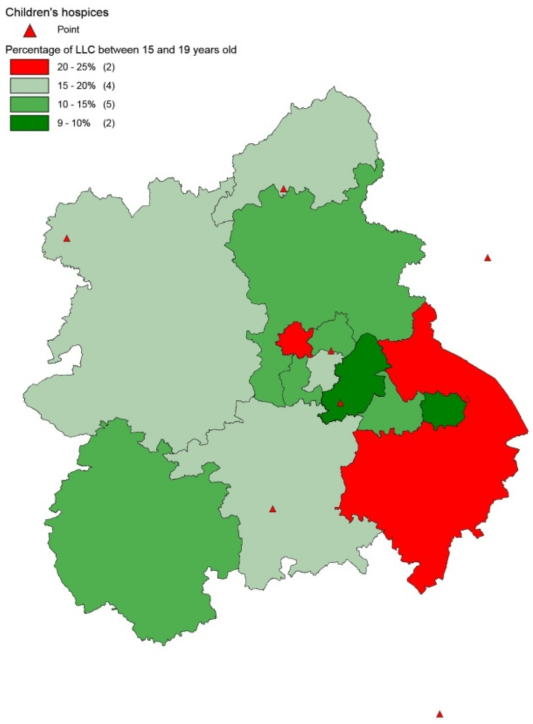
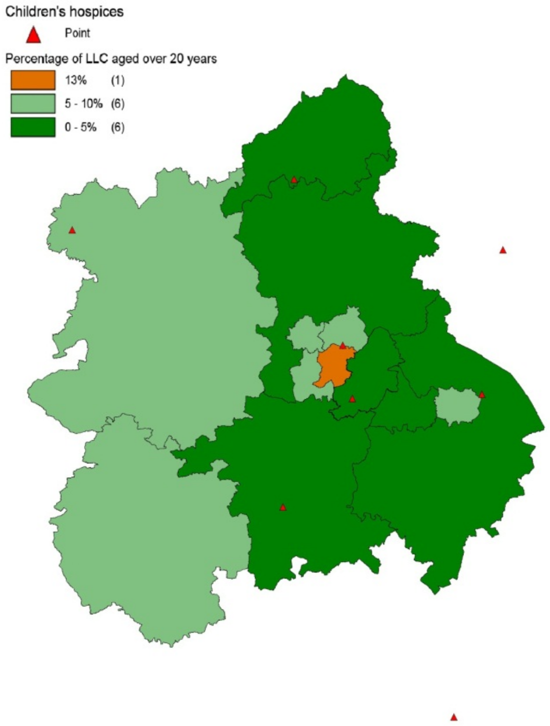


Figure 20. Proportion of LLC 15-19 years old



Are there disproportionate numbers of black and racial minorities amongst life limited children?

Similarly to the previous question, the numbers of ethnic minorities compared between areas should be proportionately distributed, but in practice we find fewer communities with ethnic minority groups in the rural areas. Figure 22 shows the usual population of all children for the West Midlands; Birmingham and environs has the most at around 20 percent and there are fewest in Herefordshire.

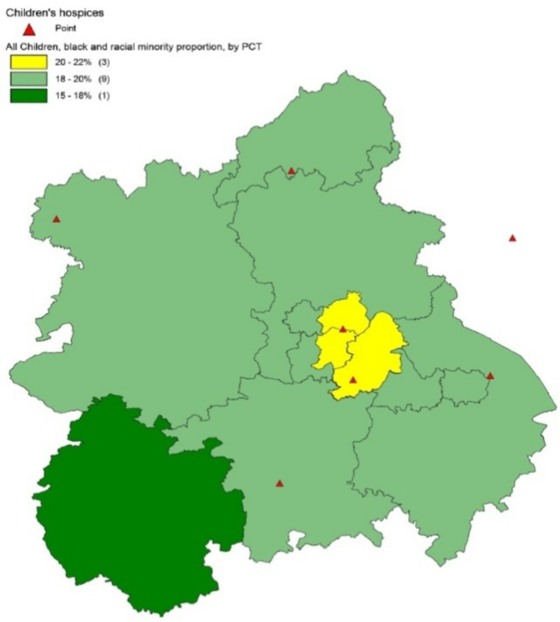


Figure 22. All children of ethnic minority background by PCT Proportion of all children under the age of one year by PCT

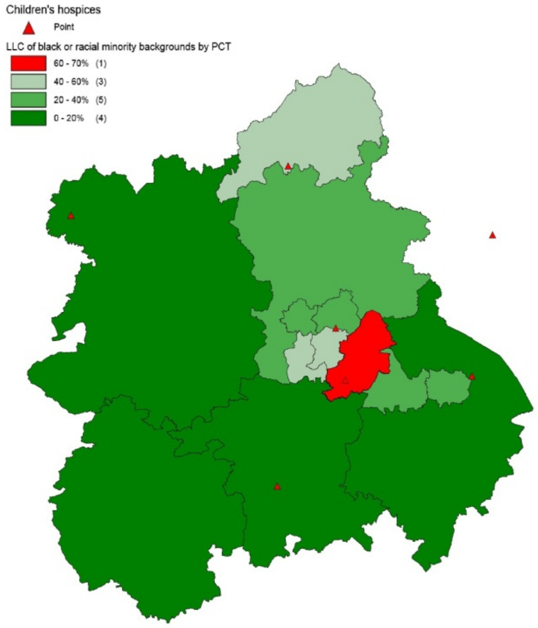


Figure 23. Children with LLC and ethnic minority backgrounds

Comparing this with the children in the study it is immediately apparent that overall, a disproportionate number of children with ethnic minority backgrounds are represented in this group although the distribution is fairly similar to the distribution overall. This predominance of ethnic minorities is well documented and in Birmingham, where the children are most concentrated, around 60 percent are from ethnic minority backgrounds; generally the upper right quadrant of the map shows the highest proportion of children with LCC from ethnic minorities. The largest group in our sample was from South Asian ethnic backgrounds, representing up to 60 percent of the total population of LCC in Birmingham (figure 24).

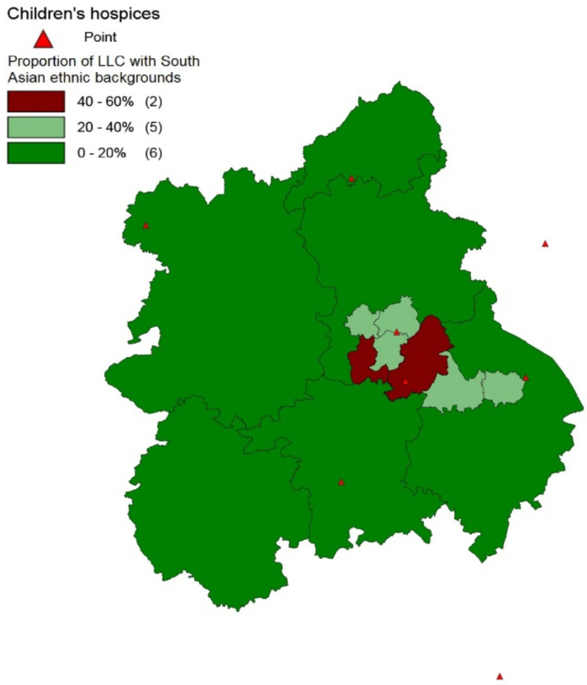


Figure 24. Children with LLC and ethnic minority South Asian backgrounds.

Are there concentrations of any particular diagnosis?

The distribution of conditions is important for the planning of services as different types of disorders and conditions will require different resources. Figure 25shows the prevalence of Congenital and Chromosomal disorders, representing the conditions of 30 percent or more of the children studied in Herefordshire, Sandwell and Dudley.

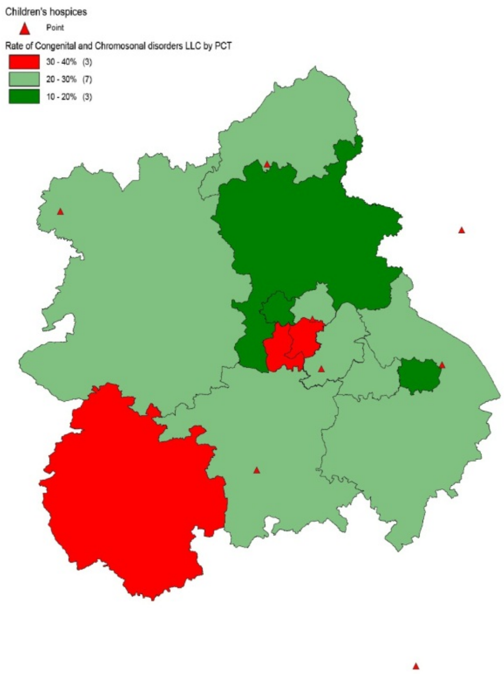


Figure 25. Distribution of Congenital and Chromosonal disorders in children

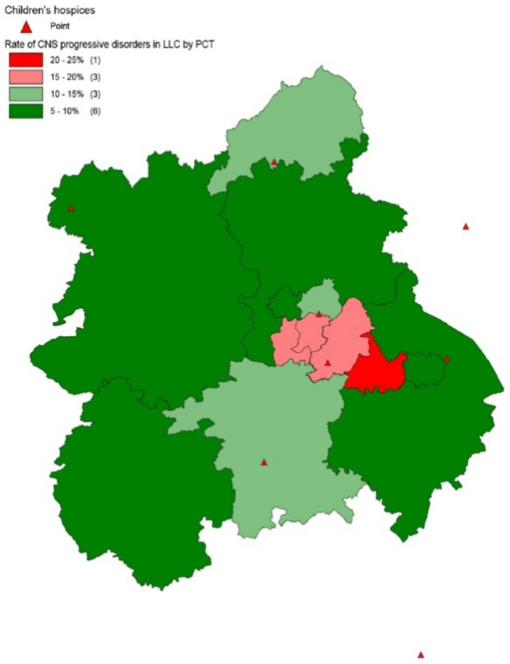


Figure 26. Distribution of CNS Progressive disorder in children

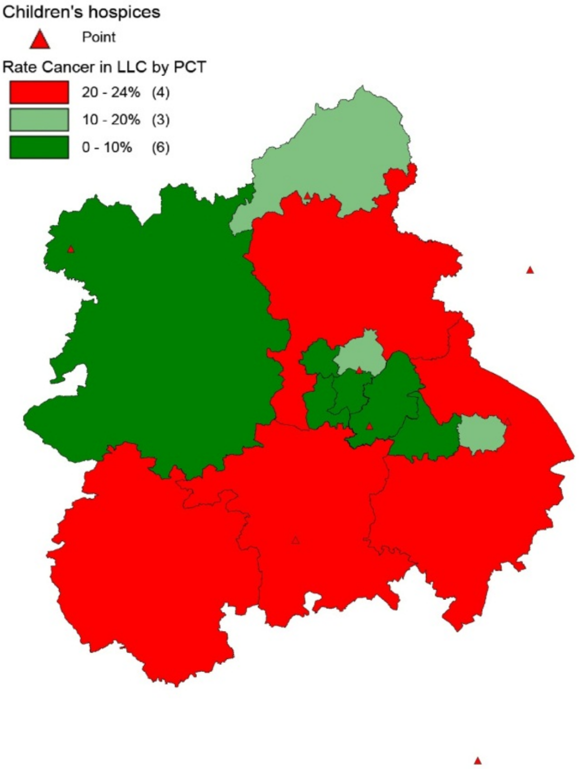


Figure 27. Distribution of Cancer in children.

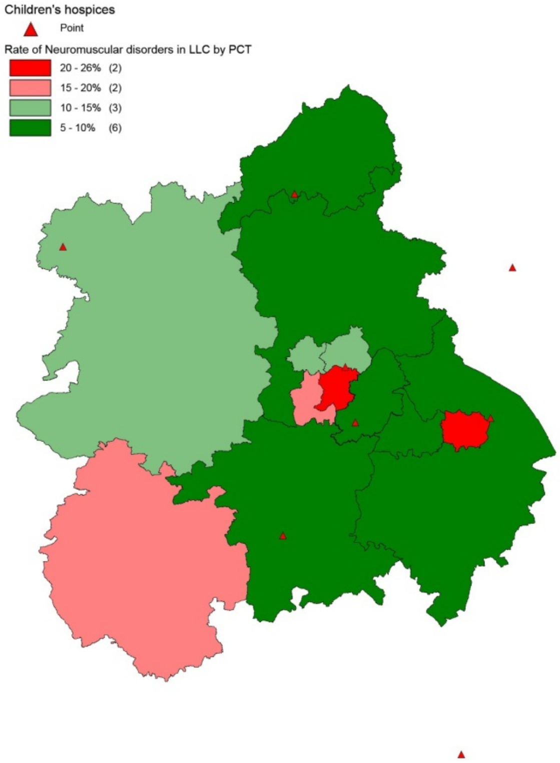


Figure 28. Distribution of Neuromuscular disorders

Birmingham and environs, particularly Coventry also has a greater incidence of CNS Progressive disorder (figure 26), but is lower in Cancer (figure 27), Neuromuscular disorders (figure 28) and Pulmonary disorders (figure 29) than other areas. Warwickshire and North Staffordshire have a greater incidence of Pulmonary disorders and Shropshire a greater incidence of Static Encephalopathy.

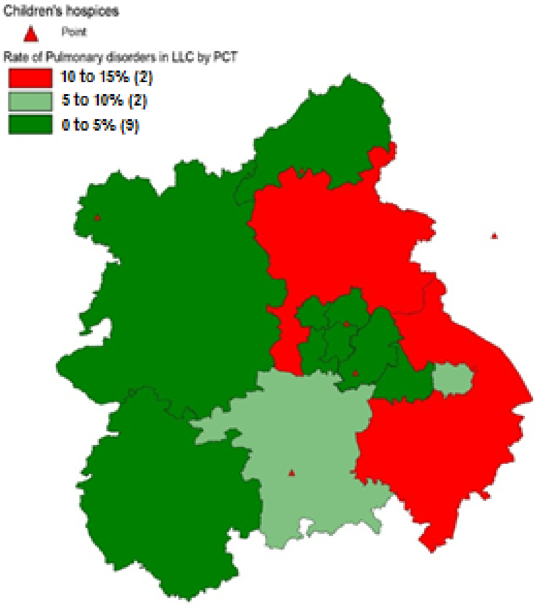


Figure 29. Distribution of Pulmonary disorders in children

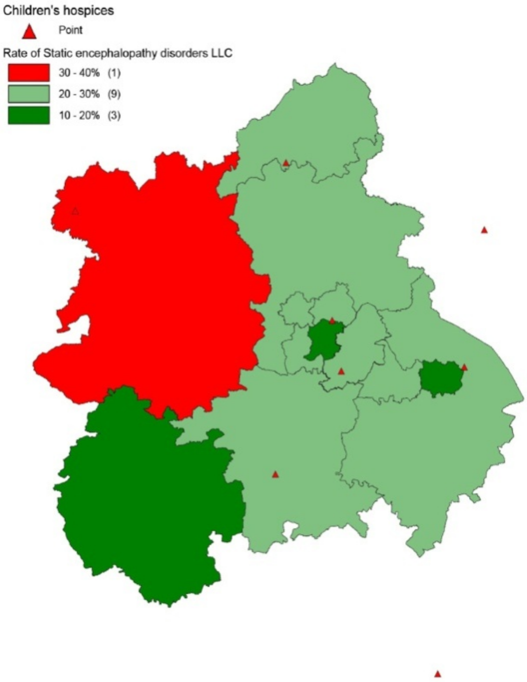


Figure 30. Distribution of Static encephalopathy disorders in children

What proportions of children are surviving their LLC and are there any differences between areas?

This data was an addition to the minimum data set of a single year's information on children who had died. Again Birmingham and Wolverhampton being large metropolitan areas with dedicated hospitals had the greatest number of children who had died during the year, and Shropshire followed (figure 31).

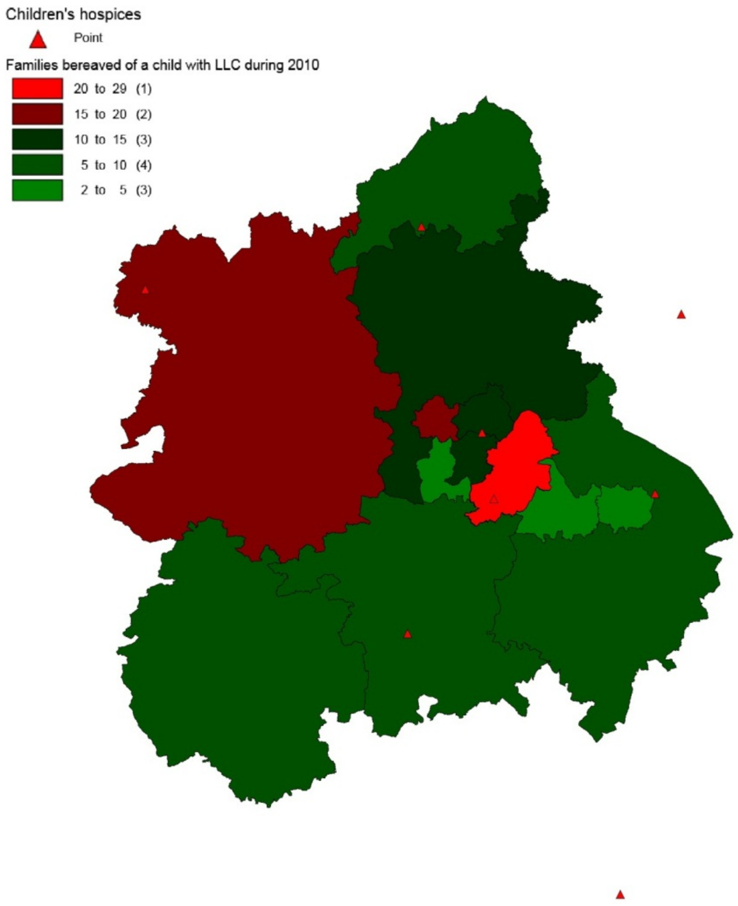


Figure 31. Numbers of children died during 2010.

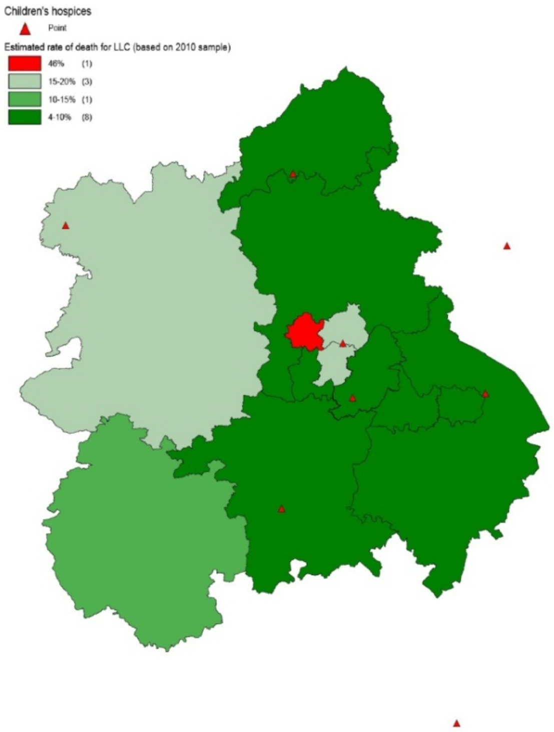


Figure 32. Proportion of children with LLC died during 2010 out of all children with LLC

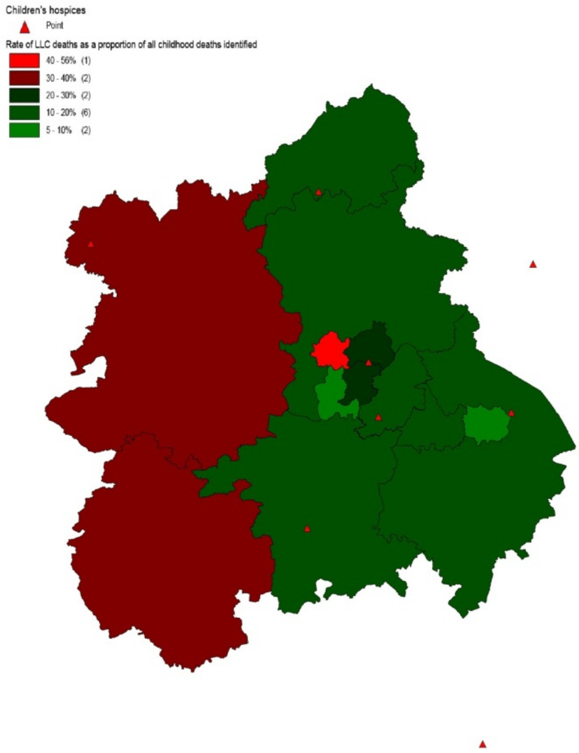


Figure 33. Proportion of children with LLC as a percentage of all childhood deaths

Figure 32 shows the proportion of the sample children who had died during 2010, which gives an estimate of rate of death. In Walsall, 46 percent of children with LLC died against all the surrounding areas which averaged 4–10 percent. Figures were obtained for all childhood deaths in Birmingham known to paediatric care services. In Walsall, it appears that 40–56 percent of children who died would have the life limiting conditions identified in this study, whereas in Coventry only 5 to 10 percent were similarly identified.

How many children live in areas where it is difficult to access services for children's palliative care?

The last analysis was to define 'buffer' zones of 20 miles radius from a hospice facility (figure 34). The 20 miles radius was a figure offered by parent advisors when asked how far away they would be prepared to travel to such a facility. The GIS program calculates the numbers of children included in the 20m radius (which includes the facilities near but outside the borderlines).

There were forty five postcode sectors outside the buffer zone containing 24 children who lived further than 20 miles from a hospice facility. It can be seen from the map that most of these lived in the Hereford and Shropshire area.

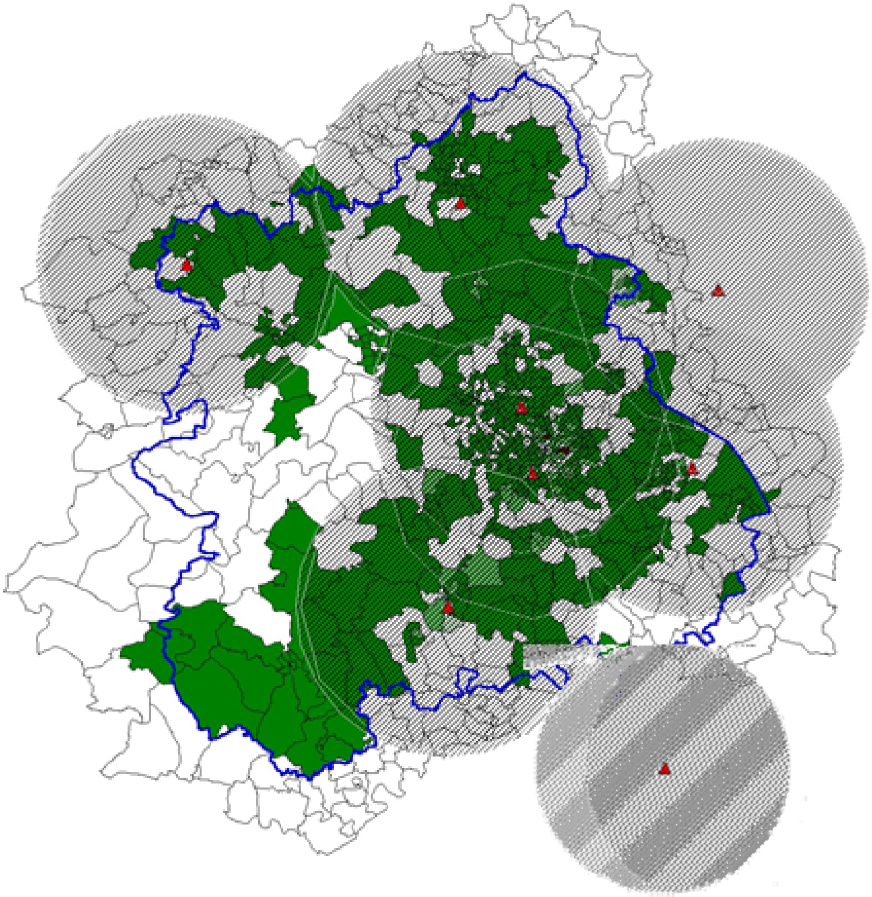


Figure 34. Buffer zones around hospice facilities – distance to travel.

4.3.4 Discussion and conclusion

We have undertaken an analysis of the demographic characteristics of young people requiring palliative care services in the West Midlands area. We have used supplementary data to provide context information and estimated accessibility to the most important service at the end of a child's life or when they are particularly vulnerable. We have identified a number of issues of interest to commissioners and providers of services.

The east side of the region is better provided with hospices and children's services, this is probably because of the swathe of urbanisation through the centre of the region and the density of population. Adjusting the analysis by rate to account for differences in population did not eliminate all the disparities. We are not in a position to explain or speculate about the causes or solutions to the issues described but there are high percentages of children with LLC and a disproportionate number of young children under one year.

The middle section of the West Midlands area contains more of the population of interest and it is particularly high around the denser city areas of Birmingham and the northern suburbs. Birmingham and Staffordshire have fewer older children with LLC and more younger children based on average age. It is tempting to conclude that children are living longer on the east side of the region because it is less industrialised and more rural. However, whilst there are no particular identifiable patterns amongst the other age groups, Birmingham also has fewer of the oldest children of 20 years or more and contained a large proportion of the figures for children who died in 2010. The bottom of the region was less likely to contain these children. There is probably a wider association that is not linked to area effects but further study would help to confirm this.

A possible explanation for these results may be because the conditions defined here are more likely to occur in black and racial minority populations and the figures here represented, support this. The South Asian population was particularly vulnerable and most of this population lived in Birmingham. Several studies have found links between deprivation and life limiting illness, but for this study, there was little relation between areas in the first quartile (more deprived) and areas of concentration of LLC.

Some disorders were more likely to occur in geographical patterns, it is not clear why this is so, but it may be that they were being picked up by the services more effectively. This data would help services to plan or provide specialist facilities in some areas or research as to the causes of the apparent greater incidence and supporting information from service user groups.

Generally, the big message is that the west side of the region is less accessible, less well provided with care facilities, seems to have more disparity between small areas and there is less information about children in these areas and their needs. There may truly be fewer children in the west side of the region but equally there might be more families in need of particular help to access services or a system of informal provision that may benefit from statutory support.

This part of the research is a preliminary and speculative description subject to caveats and limitations most of which concern the representativeness of the data. We focused on areas rather than individuals. The study covers as far as we can determine, the whole population known to services but it is limited because we are unable to practically explain the patterns and in particular to speculate on the causes of disparity.

GIS can be used to analyse a wide range of healthcare service issues and this analysis points to a geographical inequity in provision and need. We suggest there is scope for further work to support or inform this analysis and that this would be considerably easier if services were able to share their individual information and contribute to a wider collation. Service planning can not be effective if the basic information regarding the demographics of the service users is not easily accessible.

4.4 Findings from the Questionnaires

Table 9 provides the numbers of questionnaires distributed from services and the response rates. A qualitative analysis of the free text comments from the surveys is included in Section 4.5.

Table 9. Distribution and response rates for questionnaires

Questionnaire	Distributed	Returned	Response Rate	Free text comments
	n	n	%	n
Parents	1532	192	12.3	146
Bereaved Parents	180	23	12.8	17
Staff	504	264	52.4	211
Service Managers	58	21	36.2	n/a

4.4.1 Findings from the Parent's Questionnaire

The overall response rate from parents was 12.5%.

4.4.1.1 Demographics

The diagnoses of children they described and their ages are listed in Table 10.

Table 10. Disease Categories of children whose parents completed the Parent Survey.

Disease category	N	%
CNS - Static Encephalopathy	46	24.0
Congenital and Chromosomal	41	21.4
Neuromuscular	32	16.7
Cancer	28	14.6
CNS Progressive	21	10.9
Other	6	3.1
Diagnosis not known	2	1.0
Missing or unable to classify	16	8.3
Total	192	100

Children and young people were aged from 0 to 24 years (median age 10 years). Mean age was 9.8 (SD 5.9). 154 (80.2%) were White British or Irish, 32 (16.7%) were South Asian or British Asian, 3 (1.6) were Black or Black British. Data was missing on 3 (1.6%).
The median household income lay between £15,000 and £30,000. There were from 0 to 5 other children in the household (median 0).

The PCT Cluster areas in which families lived are listed in Table 11.

Table 11. Number of families returning Parent's Survey by PCT Cluster area in which they lived.

PCT Cluster area	N	%
Arden	22	11.5
Birmingham and Solihull	48	25.0
Black Country	35	18.2
Staffordshire	38	19.8
West Mercia	37	19.3
Information missing or out of area	12	6.2
Total	192	100

4.4.1.2 Analysis of the Measure of Processes of Care for Parents (MPOC-56)

Parents completed the 56 item MPOC which had been slightly adapted for this population. Because the numbers of returned questionnaires were relatively small in relation to the number of items, this analysis does not explore the data for alternative factors but uses the five factors identified by King, Rosenbaum & King, 1997 and used in a number of other studies. The scores for the factors are listed in Table 12 by the PCT Cluster areas in which the families lived.

Table 12. Mean score and standard deviation for the five factors of the MPOC-56 by PCT Cluster area in which families lived.

	Arden	Birmingham and Solihull	Black Country	Stafford -shire	West Mercia	ANOVA *P*
n=	19 - 20	46 – 48	33 - 35	34 - 36	33 - 36	
Enabling and Partnership (n=174)	5.4 (1.4)	5.1 (1.4)	4.7 (1.3)	5.1 (1.3)	4.9 (1.4)	0.277
Providing General Information (n=169)	4.6 (1.3)	4.4 (1.5)	3.8 (1.5)	4.3 (1.4)	3.8 (1.8)	0.146
Providing Specific Information about the Child (n=169)	5.1 (1.3)	5.2 (1.3)	4.6 (1.4)	5.4 (1.4)	4.9 (1.4)	0.087
Coordinated and Comprehensive Care for the Child and Family (n=172)	5.5 (1.2)	4.9 (1.4)	4.4 (1.4)	5.0 (1.4)	4.7 (1.5)	0.068
Respectful and Supportive Care (n=175)	6.0 (1.2)	5.3 (1.4)	4.9 (1.3)	5.4 (1.4)	5.3 (1.5)	0.076

Although there were no statistically significant differences in Factor scores between areas there is a trend towards lower scores in different areas which vary by the Factors. Lowest scores overall tend to be for the Factor “Providing General Information”. Table 13 examines the data by the proportion of respondents who scored less than 4 (4 indicates “Sometimes” experiencing the behaviour in the item).

Table 13. MPOC-56. Proportion of families in each PCT Cluster area who had an average factor score of less than 4

	PCT Cluster					
	Arden	Birmingham and Solihull	Black Country	Staffordshire	West Mercia	Chi Sq P
N	19 - 20	46 - 48	33 - 35	34 - 36	33 - 36	
	%	%	%	%	%	
Enabling and Partnership (n=174)	10.5	22.9	31.4	22.2	30.6	0.438
Providing General Information (n=169)	42.0	37.5	57.1	32.4	60.6	0.071
Providing Specific Information about the Child (n=169)	15.0	19.6	24.2	11.4	22.9	0.653
Coordinated and Comprehensive Care for the Child and Family (n=172)	15.0	25.0	37.1	28.6	35.3	0.399
Respectful and Supportive Care (n=175)	5.0	20.8	25.7	13.9	24.1	0.359

The order across areas varies to some extent by Factor. However, the proportion of families scoring below 4 especially in relation to “Providing General Information” where two areas score over 50% families, is higher than would be hoped for and supports the more general findings in this and other studies that families find it difficult to access general information for instance about services and financial benefits. A Chi Square test was used to explore differences between areas in proportion of respondents whose average score on the factor was less than 4, or equal to and above 4. Again the differences between areas are not statistically significant. See Discussion for comparison with other studies.

Care Coordination

A question that was asked within the MPOC questions but was not part of the scale and was analysed separately was whether parents had a named person to contact if they needed help and advice. This question was phrased to elicit whether the family had a care-coordinator, key worker or similar. The question was scored similarly to the MPOC questions from 1 to 7 (‘never’ to a ‘great extent’).

The responses were split in to two groups, below and at or above the median score of 6 for the question. Differences in MPOC-56 domain scores between these two groups were examined using Independent-Sample t-tests. Differences were highly significant for all domains (Table 14).

Table 14. MPOC Domain scores by responses at and above and below the median of 6 on the question 'Do you have a named person to contact if you need help or advice?' I is 'never', 7 is 'to a great extent'.

	Score	N	Mean	Std. Dev	P
Enabling and partnership	below 6	87	4.00	0.95	< 0001
	6 and above	98	5.54	0.97	
Providing general information	below 6	85	3.25	1.13	< 0001
	6 and above	95	4.99	1.51	
Providing specific information about child	below 6	84	4.14	1.09	< 0001
	6 and above	97	5.78	1.05	
Coordinated and comprehensive care	below 6	86	3.84	0.98	< 0001
	6 and above	96	5.78	1.02	
Respectful and supportive care	below 6	86	4.23	1.11	< 0001
	6 and above	99	6.20	0.90	

4.4.1.3 Families' met and unmet needs.

Parents were asked a series of questions about the extent to which the child's and families' needs were met.

Table 15. Proportion of parents reporting met and unmet needs ordered by largest unmet needs.

Question number		No need or need is met	Need not sufficiently met
19	Do you have opportunities to plan future care for child e.g. transition to adult services	20.9	67.5
4	Does your family need special provision for family holidays?	34.1	63.4
20	Do you have opportunities to plan care for child around time of death	24.1	55.5
18	Do you have continuity of care across services?	40.3	54.0
11	Do you need information about services and how to obtain them	51.9	45.5
9	Do you need help to be able to take up work as desired?	53.3	40.7
2	Does your child need special play facilities?	57.6	40.3
15	Do you need financial advice and support?	61.3	36.6
3	Does your child's condition need adaptations to your home?	61.2	35.6
7	Do you need help with domestic chores around the house?	63.3	35.6
17	Do you need a key worker to help you organize your child's care services?	62.3	33.5
12	Do you need access to psychological / emotional support for yourselves?	65.9	33.0
6	Do you need help to be able to go out for a short time from home?	64.9	32.4
1	Does your child need help with mobility e.g. buggy, wheelchair, adapted car?	69.6	28.8
22	Relief of other troublesome symptoms?	68.6	27.3
14	Does your child need access to psychological / emotional support?	75.9	21.9
5	Does your child need short breaks away from home (not	76.9	21.5

	with family)?		
10	Do you need information about your child's condition?	78.0	20.9
13	Do your child's brothers and/or sisters need access to psychological / emotional support?	75.9	20.9
8	Does your child receive appropriate education?	79.6	16.2
21	Does your child need relief from pain and discomfort?	80.6	15.6
16	Do family members need spiritual support?	88.5	10.4

The earlier finding relating to care-coordination was followed up by an analysis of a similar question under the Needs Section – Do you need a key worker to help you organize your child's care services?". When the MPOC Domains were investigated for differences on whether this need was met ('no' (need) or 'this need is met'; n=124 – 128)) or not met ('we pay for this ourselves' or 'yes, but not enough support'; n=56 - 59) differences remained highly significant (P=0.014 to <0.001).

Table 16. Proportion of families ratings unmet needs by PCT Cluster area

		PCT Cluster Area					
		Arden	Birmingham & Solihull	Black Country	Stafford -shire	West Mercia	Chi Sq P
	N=	19 - 22	46 - 48	33 - 35	43 - 46	33 - 37	
	Parents reporting unmet needs	%	%	%	%	%	
1	Does your child need help with mobility e.g. buggy, wheelchair, adapted car	27.3	29.2	45.8	17.8	37.8	NS
2	Does your child need special play facilities?	18.2	50.0a	29.4	48.9	41.7	NS
3	Does your child's condition need adaptations to your home?	28.6	42.2	32.4	28.9	47.2	NS
4	Does your family need special provision for family holidays?	38.1	68.9a	74.3a	67.4a	62.9a	NS
5	Does your child need short breaks away from home (not with family)	9.1	31.1	11.4	26.7	18.9	NS
6	Do you need help to be able to go out for a short time from home	9.1	38.6	45.7	27.3	37.8	0.043
7	Do you need help with domestic chores around the house?	27.3	50.0a	32.4	26.1	43.2	NS
8	Does your child receive appropriate education?	40.0	22.7	45.5	40.0	42.9	NS
9	Do you need help to be able to take up work as desired?	40.0	47.6	45.5	40.0	42.9	NS

10	Do you need information about your child's condition	13.6	34.8	22.9	13.3	16.2	NS
11	Do you need information about services and how to obtain them	36.4	60.0a	48.6	40.9	41.7	NS
12	Do you need access to psychological / emotional support for yourselves	36.4	43.5	31.4	28.9	27.0	NS
13	Do your child's brothers and/or sisters need access to psychological / emotional support?	36.4	33.3	15.2	8.9	23.5	0.023
14	Does your child need access to psychological / emotional support?	31.8	21.7	26.5	15.6	19.4	NS
15	Do you need financial advice and support?	40.9	55.6a	41.7	22.2	27.8	0.013
16	Do family members need spiritual support?	4.5	17.4	8.6	10.9	5.6	NS
17	Do you need a key worker to help you organize your child's care services?	15.0	40.9	32.4	38.6	37.8	NS
18	Do you have continuity of care across services?	52.4a	65.9a	51.4a	53.5a	63.9a	NS
19	Do you have opportunities to plan future care for child e.g. transition to adult services	77.8a	79.1a	75.9a	67.4a	81.3a	NS
20	Do you have opportunities to plan care for child around time of death	75.0a	82.1a	63.0a	59.5a	48.7	NS
21	Does your child need relief from pain and discomfort	4.8	24.4	14.7	13.6	16.7	NS
22	Relief of other troublesome symptoms	15.0	37.2	31.4	20.5	27.0	NS

We have highlighted in the table where the more than 50% parents reported insufficiently met needs, suggesting that these may be areas that might be the focus for further service improvement or investment. Again, the areas of coordination between services, communication and information giving are to the fore. The needs of families in Birmingham and Solihull are particularly of note.

4.4.2. Findings from Bereaved Parent's Survey.

Overall response rate 12.8% with one centre having a 38.9% response rate. Four centres chose not to distribute the bereaved parent's questionnaires and 7 centres generated a zero response.

Twenty three questionnaires were returned. These were completed by the deceased child's mother in all but one case. Ethnic origin of children was white British or Irish for 78%, South Asian for 17%.

The median age of children when they died was 2.3 years (range 0 to 17 years). Disease categories are listed in Table 17.

Table 17. Disease categories of children who died in the 11 months up to end April 2010.

Disease category	N	%
CNS - Static Encephalopathy	6	26.1
Congenital and Chromosomal	6	26.1
Cancer	5	21.7
Perinatal / prematurity	3	13.0
Chronic lung disease	1	4.3
Not known or unable to classify	2	8.7
Total	23	100

Parents were asked to rate to what extent particular needs were met. These are listed in Table 18.

Table 18. Proportion of families (n=21) for whom the listed needs were met or not sufficiently met.

Question number		No need or need met	Need not sufficiently met	Missing
		%	%	%
5	Did you receive financial advice and support?	47.8	52.2	
2	Were you offered opportunities to discuss and plan end-of-life care for your child?	52.2	47.8	
9	Did your child and the family receive sufficient psychological, emotional and spiritual support?	52.2	47.8	
14	Did you continue to receive support from your child's health care team or others after your child's death?	56.5	43.5	
15	Have you / the family received sufficient financial advice and support?	52.2	43.5	4.3
1	Did you have a key worker to help you organize your child's care services?	69.6	26.1	4.3
6	Did you receive sufficient respite or short breaks	73.9	26.1	
12	Did you have opportunities to build memories of your child e.g. time with your child after death, photographs, hand prints.	73.9	26.1	
11	Did you have opportunities to talk about your child's illness and cause of death with your child's doctor?	78.3	21.7	

13	Did you have the help you needed to plan your child's funeral and was it as wished?	78.3	21.7	
7	Did your child and family receive sufficient support from medical professionals?	82.6	17.4	
8	Did your child and family receive sufficient support from nurses and carers?	82.6	17.4	
4	Did your child have relief from other troublesome symptoms e.g. difficulty in breathing, seizures?	82.6	13.0	4.3
3	Did your child have relief from pain and discomfort?	91.3	8.7	

Most families appeared to report a high level of support from medical and nursing staff and good pain and symptom management for their child. Areas where needs were less well met were the need for financial advice and support and the need for making plans around their child's death. After their child's death parents' and families' reported that their need for on-going psychological, emotional and spiritual support was poorly met. The need for financial advice and support remained unmet.

4.4.3. Findings from the Staff Survey.

The staff questionnaire was returned by 264 (52.4%) individuals.

4.4.3.1. Demographics

The staff are believed to be predominantly female (we neglected to ask). Age group, ethnicity and professional role are listed in Table 19 below.

Table 19. Demographics from Staff Questionnaire

Staff age	n	%
< 25 years	5	1.9
25 – 40	102	38.6
41 – 55	125	38.6
55 and over	28	10.6
Missing	4	1.5
All	264	100
Ethnicity	n	%
White British and Irish	238	90.2
South Asian and British Asian	11	4.2

Black Caribbean & Black African	6	2.3
Mixed	3	1.1
Missing	6	2.3
All	264	100
Professional role	n	%
Nurses	160	60.6
Family support workers / health care assistants	45	17.0
Therapists & Social workers	36	13.6
Doctors	7	2.7
Missing	16	6.1
All	264	100
Highest academic qualification	n	%
PhD or MD	3	1.1
Post Graduate MSc, MA	27	10.2
University degree	93	35.2
A'Level or DipHE	106	40.1
O'Level / GCSE	20	7.7
No formal qualifications	2	0.8
Missing	13	4.9
All	264	100

Forty four percent of practitioners reported that they were a named care co-ordinator (or key-worker) who works across services to help manage a child's care.

4.4.3.2. Measure of Processes of Care for Service Providers (MPOC-SP).

After removal from the analysis of four individuals who had more than 50 percent of the MPOC items missing, 260 individuals remained who rated their practice on the MPOC-SP. Four domains have been previously reported by the developers of the instrument (ref) and others (refs), these domains being 'Treating people respectfully' (TPR), Showing interpersonal sensitivity' (SIS), Communicating specific information about the child' (CSI) and 'Providing general information' (PGI). We explored whether these domains also describe the work of practitioners caring for children with life-limiting conditions.

Mean, standard deviation, median and range of scores for each item in the scale are given in Table 20.

Table 20. MPOC-SP. Mean (SD), median and range of scores from 260 staff working with children with life-limiting conditions in the West Midlands.

	In the past year, to what extent did you...	Domain	Mean (SD)	Median	Percent scoring < 4
15	Provide parents with written information about their child's condition, progress, or treatment?	CSI	3.9 (2.1)	4	41.7
27	Have general information available about different concerns (e.g., financial costs or assistance, genetic counselling, respite care, dating and sexuality)?	PGI	3.9 (1.9)	4	40.9
14	Tell parents about the results from tests and/or assessments?	CSI	4.2 (2.3)	5	37.1
23	Promote family-to-family "connections" for social, informational or shared experiences?	PGI	4.2 (2.0)	4	34.8
25	Provide advice on how to get information or to contact other parents (e.g., through a community's resource library, support groups, or the Internet)?	PGI	4.2 (1.8)	4	33.0
16	Tell parents details about their child's services, such as the types, reasons for, and durations of treatment/ management?	CSI	4.4 (2.0)	5	29.5
26	Provide opportunities for the entire family, including siblings, to obtain information?	PGI	4.4 (1.8)	5	29.5
24	Provide support to help families cope with the impact of their child's condition (e.g., informing parents of assistance programs, or counselling how to work with other service providers)?	PGI	4.7 (1.7)	5	22.7
12	Help each family to secure a stable relationship with at least one service provider who works with the child and parents over a long period of time?	SIS	5.1 (1.8)	5	18.9
5	Tell parents about options for services or treatments for their child (e.g. equipment, school, therapy)?	SIS	5.0 (1.7)	5	17.8
4	Discuss expectations for each child with other service providers, to ensure consistency of thought and action?	SIS	5.1 (1.6)	5	14.4
11	Let parents choose when to receive information and the type of information they wanted?	SIS	5.2 (1.5)	5	13.3
9	Anticipate parents' concerns by offering information even before they ask?	SIS	5.0 (1.3)	5	12.1
1	Suggest treatment/ management activities that fit with each family's needs and lifestyle?	SIS	5.1 (1.6)	5	11.7
19	Make sure parents had opportunities to explain their treatment goals and needs (e.g., for services or equipment)?	TPR	5.4 (1.5)	6	11.4
8	Discuss/explore each family's feelings about having a child with a life limiting condition (e.g., their worries about their child's health or function)?	SIS	5.2 (1.4)	5	10.2
2	Offer parents and children positive feedback or encouragement (e.g., in carrying out a home programme)?	SIS	5.5 (1.5)	6	9.8

18	Treat parents as equals rather than just as the parent of a patient (e.g. not referring to them as "Mum" or "Dad")?	TPR	5.9 (1.3)	6	5.3
13	Answer parents' questions completely?	TPR	5.7 (1.1)	6	3.8
10	Make sure parents had a chance to say what was important to them?	TPR	6.2 (1.0)	6	2.3
20	Help parents feel like partners in their child's care?	TPR	6.2 (1.0)	6	2.3
3	Take the time to establish rapport with parents and children?	SIS	6.5 (0.9)	7	1.9
21	Help parents to feel competent in their roles as parents?	SIS	6.2 (0.9)	6.5	1.5
22	Treat children and their families as people rather than as a "cases" (e.g., by not referring to the child and families by diagnosis)?	TPR	6.6 (0.8)	7	0.8
6	Accept parents and their family in a non-judgmental way?	TPR	6.6 (0.6)	7	0.4
7	Trust parents as the "experts" on their child?	TPR	6.3 (0.8)	6	0.4
17	Treat each parent as an individual rather than as a "typical" parent of a child with a "problem"?	TPR	6.5 (0.8)	7	0.4

MPOC-SP Domains. TPR = Treating people respectfully; SIS = Showing interpersonal sensitivity;
CSI = Communicating specific information about the child; PGI = Providing general information.

Most of the items have a median score of 5 or more indicating that the participants used these practices more often than 'sometimes', however, a few items score lower particularly those relating to provision of information to families; items 15, 23, 25 and 27 where the median score is 4, indicating that for 50% the practice was less usual.
The mean scores for the four domains introduced above are shown in Table 21 below.

Table 21. Descriptive statistics for the four MPOC-SP domains (n=260 practitioners).

	Mean	Std. Dev	Minimum	Maximum	Proportion scoring < 4
Treating people respectfully (TPR)	6.14	0.63	4.11	7.00	0.4
Showing interpersonal sensitivity (SIS)	5.41	0.91	2.90	7.00	8.0
Providing general information (PGI)	4.27	1.51	1.00	7.00	39.8
Communicating specific information (CSI)	4.14	1.92	1.00	7.00	41.3

Of the four domains those concerned with providing either general information or information specific to the child scored considerably lower than those behaviours concerned with treating the person with respect and sensitivity. Although not ideal given the families' needs for information, this is commonly the case in other studies and is discussed later in relation to findings from other studies. It is possible here that staff underestimate families' needs for both general and specific information.

Analysis of Variance was used to explore differences in MPOC-SP domain scores of staff /practitioners according to their professional training and whether they reported themselves to be a care co-ordinator / key worker for child's care. Several areas of difference were identified. Those who reported that they were care coordinators had significantly higher scores on each of the Domains. Several Health Care Assistants (HCA) and Family Support Workers (FSW) had reported items in these domains to be not applicable to their role, and hence scored 1 on these items, and HCAs and FSWs tended to score lower than others on Communicating Specific Information and Providing General Information.

We used Independent Sample T-Tests to explore whether staff who were Care-coordinators rated the service provided to families higher on the MPOC-SP domains than those who were not care coordinators. Staff who were care-coordinators scored significantly higher than those who were not for each domain (Table 22).This difference in ratings occurred independently of discipline / training so that, for instance, FSWs also scored higher if they acted as Care Coordinators.

Table 22. Differences in ratings between members of staff who rated the behaviours undertaken

MPOC-SP Domains	Care-coordination role	N	Mean (SD)	P=
Showing interpersonal sensitivity (SIS)	Not care coordinator	148	5.04 (1.04)	< 0.001
	Care Coordinator	116	5.78 (0.77)	
Treating people respectfully (TPR)	Not care coordinator	148	6.03 (0.73)	0.012
	Care Coordinator	116	6.24 (0.61)	
Communicating specific information (CSI)	Not care coordinator	148	3.60 (1.94)	< 0.001
	Care Coordinator	116	4.76 (1.72)	
Providing general information (PGI)	Not care coordinator	148	3.92 (1.57)	< 0.001
	Care Coordinator	116	4.66 (1.39)	

Analysis of variance was used to examine the extent to which behaviours differed by the PCT Clusters in which their organizations were geographically situated. The PCT Clusters are Arden, Birmingham, Black Country, Staffordshire and West Mercia. No significant differences in the MPOC-SP domain scores were found between geographical areas.

4.4.3.3. Staff perceptions of children's and families met and unmet needs.

The practitioners were asked to rate from 1('Not at all') to 7('More than needed') the extent to which they thought specific needs of families and children were being met in the services they work for. Proportion (percent) of practitioners who rated the needs as poorly met (scores from 1 to 3) are listed in Table 23 below by the PCT Cluster area in which their service lay. Chi Square test was used to investigate where there were differences in proportion between the areas.

Table 23.Percent of practitioners scoring children's and families' unmet needs according to geographical distribution by PCT Cluster area. Items are listed with needs perceived as least met at top.

Q no	N=	ALL	Arden	B'ham & Solihull	Black Country	Stafford -shire	West Mercia	Chi square
Q no	N=	239 - 255	42-46	64-69	27-30	21-24	81-86	P
		%	%	%	%	%	%	
9	Domestic help with chores around the house	86.6	89.1	89.4	79.3	91.3	84.1	NS
10	Parents able to work as desired / as appropriate – Part time	82.5	69.8	86.2	79.3	95.2	84.1	NS
11	Parents able to work as desired / as appropriate – Full time	74.5	65.1	76.6	67.9	95.2	74.7	NS
8	Access to short breaks at home	57.9	36.4	74.2	56.7	56.5	57.1	0.004
18	Spiritual support for family members	57.2	60.5	43.5	65.5	83.3	56.5	0.011
5	Necessary adaptations to house	52.2	40.9	59.4	70.0	65.2	42.2	0.015
6	Access to short breaks away from home with family / holidays	48.0	42.2	65.2	48.3	45.8	37.3	0.013
7	Access to short breaks for child away from home (not with family)	44.8	41.4	53.6	41.4	45.8	40.5	NS
15	Access to psychological / emotional support for siblings of affected child	44.7	52.2	53.6	50.0	41.7	32.6	NS
16	Access to psychological / emotional support for affected child	42.3	43.5	47.8	56.7	39.1	32.9	NS
22	Opportunity to plan future care for child e.g. transition to adult services	40.7	37.0	54.4	20.0	54.2	35.3	0.008
17	Financial advice and support	39.0	41.3	34.3	46.7	70.8	29.8	0.005
4	Access to appropriate play facilities	38.6	20.0	54.4	56.7	21.7	33.7	< 0.001
14	Access to psychological / emotional support for parents	37.4	47.8	35.3	53.3	37.5	27.9	NS
20	Continuity of care across services	33.2	19.6	44.9	25.0	25.0	36.1	0.04
1	Help with mobility e.g. buggy, wheelchair, adapted car	32.8	31.8	35.3	26.7	58.3	26.2	0.05
19	Key worker to help organize the child's care services	29.2	28.3	33.8	13.3	25.0	32.9	NS
2	Access to appropriate	28.2	19.6	27.5	30.0	36.4	30.9	NS

1	education for affected child							
24	Access to bereavement support	23.4	30.4	27.5	32.1	12.5	16.5	NS
23	Opportunity to plan care for child around time of death	18.7	17.4	26.1	17.2	16.7	14.3	NS
13	Information about services and how to obtain them	15.1	13.0	14.7	10.3	25.0	15.3	NS
12	Information about the child's condition	13.4	8.9	18.8	10.0	12.5	12.8	NS
3	Relief of other troublesome symptoms	8.4	2.2	11.8	14.8	8.3	7.0	NS
2	Relief of pain and discomfort	6.4	0.0	10.1	7.1	4.3	7.1	NS

Whilst both parents and staff tend to agree that pain and symptoms are well managed, parents have tended to perceive items relating to planning for the future and provision of information as less well met than staff.

4.4.4 Findings from Service Manager Questionnaire

The overall response rate for the service manager's questionnaire was 36.2%. Information was provided by 19 of the 21 services.

4.4.4.1 Geographical distribution

Each service manager was asked to outline the geographical area their service covered. The areas identified were: Staffordshire, including Burton, Uttoxeter and the surrounding area, a small number of surgeries in South Derbyshire, Shropshire, Stoke, Birmingham, Coventry and Warwickshire, Walsall, Sandwell, Wolverhampton, Dudley, Telford, Warwickshire, Solihull, Cannock, Rugeley, Codsall, Stone, Worcester, Redditch and Kidderminster. The list demonstrates the geographical diversity of the West Midlands from urban central areas of conurbation to the rural western counties of Shropshire and Herefordshire which border Wales. As far as we are aware the whole West Midlands area is covered by the 21 services, though there may be gaps within. The West Midlands covers 902 square kilometres (Evi 2012).

4.4.4.2 Staffing

The managers were asked to identify those staff in their service whose role was specific to children with life limiting conditions or their families. The types of staff members listed illustrate the range of staff and roles that are required due to the complex care needs of children and young people with life limiting conditions and their families. (Table 25).

Table 25: Service Managers' description of current staff employed in the care of children and young people with life-limiting conditions and their families.

Division	Type of Staff Member
Medical	GP, Medical Consultant, Senior Medical Officer, Medical Officer
Nursing	Registered Children's, Adults and Learning Disabilities Nurses, Community Children's Nurses, Senior Nurse Managers, Senior Nurses, Nurse Prescriber, Hospice at home staff nurses, specialist nurses in epilepsy, cystic fibrosis, palliative care, clinical educators, school nurses, caseload managers, palliative care coordinator, palliative care team leader
Support	Support Workers, Nursery Nurses, Care Assistants, Health Care Assistants, Sibling Support Workers, Community Team Workers, Play Therapists, Play Leaders, Activity Workers, Transition workers, palliative care play therapist, advice worker
Allied Health Professionals	Physiotherapist
Social Care	Social Workers, Asian Liaison Officers,
Therapists	Counsellors, Music Therapists, Complimentary Therapists, Complimentary Therapy Coordinator
Administration	Administrators, clerical assistants
Housekeeping	Maintenance worker, catering, housekeeper

In the hospices, registered nursing staff made up 104.5 whole time equivalents (WTE), whereas in the NHS the registered nursing staff made up 50 WTE. The complimentary therapy centre had4 WTE but no registered nurses on staff.
Only one hospice reported that they had vacancies that were hard to fill during the past year.. These were primarily children's nursing posts. However, within the NHS, four services reported unfilled vacancies. Again these were children's nursing posts, and a social worker post. The need for experienced staff to apply for vacancies was also expressed.

4.4.4.3 Services provided

All the hospices were providing 24/7 care. Some NHS services were providing 24/7 cover. Others were trying to address this deficit by offering 24 hour telephone cover and 24/7 cover at end of life. This cover was often 'as required' and ad hoc usually for end of life and emergency care only. Cover was provided through the good will of staff, who could take time back if able. Both hospices and NHS services catered for children from 0 to 19 years of age. A variety of services was provided, including short breaks at night and during the day, counselling and emotional support, end-of-life care, parent support groups and befriending services and bereavement services including sibling groups.

4.4.4.4 Limitations of the current services

Service providers were asked whether there are any groups of children, young people and families who they believed might be in need of services but are difficult to provide for currently.

Hospice managers reported that they had difficulty reaching certain ethnic minority groups, for instance they had few referrals of Afro-Caribbean, Jewish, or travelling families. In addition, they did not have many referrals of neonates for hospice care. Involvement with children with neoplastic disease tended to come at a very late stage in the child's illness. They were limited in their ability to provide 24/7 hospice care in the child's home.

NHS service managers also indicated that some families had needs it was difficult for them to meet. These were, for instance, families whose children had complex care and social support needs, those experiencing financial difficulties and families from Black and Minority Ethnic communities. They were limited in their ability to meet the needs of families whose child might at the point of diagnosis not have a nursing need, but families might still require psychological support and contact regarding their child's disease progression.

The service manager from the complimentary therapy centre outlined there was a need for expansion and extension of their services. Again, they would like to reach more black and ethnic minority children and families, as well as families experiencing financial difficulties who were currently unable to access the service.

4.4.4.5 Ambitions for the future

The hospices had a variety of ambitions for the future which included counselling, end of life care, physiotherapy, neonatal services, more community service, youth worker, music therapy, physiotherapy, a specially designed befriending service for parents/carers, youth adult outreach service, improvements in sibling support team, activities team, adolescent team and setting up a hospice at home service in Birmingham and developing the transition service.

The NHS services ambitions were many and similar. To highlight a few, they endeavoured to increase the workforce with the aim of providing an equitable service. They planned to teach clinical skills to extended family members in order to provide further respite for parents. They recognized a need to forge better links with their referrers

4.5. Responses to open ended questions.

4.5.1 Methods

These expressions were processed by inductively generating themes from the responses to the question "What three things would you change about the services or the help you have had that would make the biggest difference to the quality of your family life or to other families? Please use this space to tell us anything else you feel the research team should know about." After reading each comment carefully, each comment was described / named and these names / themes were entered onto three separate Microsoft Excel spread sheets; one for the parent's responses, one for the bereaved parents responses and one for the staff responses. The service manager's questionnaire did not capture free text responses. The themes were continuously sorted so that they lay next to like themes. The frequencies of similar statements were tallied then totalled so that important and overarching themes could be identified.

4.5.2 Findings

From the 476 parents, bereaved parents and staff questionnaires that were returned, 374 of these had taken the opportunity to express themselves in the open ended question at the end of the questionnaire (Table 9). The top four themes from each questionnaire are outlined in Table 24 with theme 1 receiving the most tallies and theme 4 the least. There were other themes identified, however, these were not reported as often. The top four themes for parents were: better communication and coordination, more financial support, more respite and more well-being support. The top four themes for bereaved parents were; practical help, better communication and coordination, more well-being support and more financial support. The top four themes for staff were; better communication and coordination, more respite for families, more well-being support for families and staff and better transition.

Table 24: Themes identified from qualitative comments from parent, bereaved parent and staff questionnaires

Free text Responses	Theme 1	Theme 2	Theme 3	Theme 4
Parent n=146	More financial support n = 59	Better communication and coordination support n = 32	More respite n = 14	More well-being support n = 13
Bereaved Parent n=17	Practical help n = 9	Better communication and coordination n = 6	More well-being support n = 5	More financial support n = 4
Staff n=211	Better communication and coordination n = 61	More well-being support for families and staff n = 51	More respite n = 39	Better transition n = 27

4.5.3. Analysis and Discussion

In order to obtain a range of views, free text responses were sought (Steiner and Norman 2008), thereby uncovering parents, bereaved parents and staff opinions of what is working well and what could be better. There were some positive expressions from families about the services they receive. However, the question seeking free text response was phrased in a way to generate responses about what could be better. Free text responses from all three questionnaires revealed four themes where all three interfaced at some point; better communication and coordination, more financial support, the need for more well-being support and more respite. Each of these four themes will now be discussed separately in relation to what knowledge already exists and what the families and staff are reporting.

Better Communication and Coordination

The need for good communication, with timely flow of both clinical and social information in both directions is essential for the coordination and continuity of care (ACT and RCPCH 2009), and was identified as an unmet need in this study with this finding also reported by Steinhauser, Christakis, Clipp et al 2000). Steinhauser et al (2000) also documented that the needs of families for more timely information of the right level may be under acknowledged by health care professionals. The results from this research however, found that staff were aware of this need. Together for Short Lives (2012) who support children and families with life limiting conditions outline how important it is that families receive the right information from the moment of diagnosis so that every child and family can make choices about the care they receive.
The <u>most often mentioned</u> unmet need by parent (n = 32), bereaved parents (n = 6) and staff (n = 61) was better communication and coordination.what about financial support? Included in this are responses related to information needs.

'If we had a coordinated, integrated care package that was planned around our child and followed wherever our child went with a shared team that know our child well, then I wouldn't have to continually manage peaks and troughs of crisis. The system does not support us at present I am dragged into bureaucracy and administration and I feel a sense of hopelessness that I am not able to provide for any of my children properly. I just want to be mum and wife to my family, not super woman' (Parent, 1911).

'(1) More joined up and connected services; (2) Packages of information for parents/clients to research about conditions/help available; (3) More training of staff to enable conversations to be handled correctly.' (Staff, 215)

'I think the biggest difference would be if services would tell you what they can offer, as a lot of people in our situation do not know what they are entitled to unless they are told' (Parent, 101).

'Services should know exactly what area they cover in relation to providing a service. We have a postcode of one area but technically live in a different county. We have the services we need now, but early on it seemed no-one wanted to take responsibility for our child. That was not a very nice feeling when we were waiting to be released from hospital when our child was born' (Parent, 213)

'Co-ordinate all the different teams so that care plans do not have to keep being repeated' (Parent, 232).

It is even more vital that effective communication and coordination occurs during end of life care, death and bereavement phase of the families' journey. When families have the chance to prepare and have access to the relevant services, then their needs can be met more effectively which then means that the time following bereavement although painful is free from feelings of regret. The bereaved parents who responded to this questionnaire drew attention to their need for: improvement in information provision, support groups for bereaved families, a key worker to liaise with families and professionals to make sure each family has the support in place for when it is needed before and after death of their child. For professionals to treat each child and family as individual recognising that one service/method does not fit all (D'Argostino, Berlin-Ronalis, Jovcevska and Barrera 2008). There are many important decisions that a family need to make during this process and therefore it is of vital import that families receive high quality care at this painful time (Warr 2007).

'Professionals did not communicate with each other' (Bereaved Parent, 501).

'I think medical professionals need to establish what level of information parents want at an early stage. We had a situation where doctors were not telling us everything until we stated we wanted to know' (Bereaved Parent, 801).

'When a child is very, very ill in Hospital be aware what they say around child and visitors. One Doctor discussed letting our child go in front of our child, and 8 year old cousin and we had not even been told his condition was life-limiting at the time' (Parent and Bereaved Parent, 222).

More Financial Support

The largest unmet need that parents (n = 59) expressed and was also mentioned by bereaved parents (n = 4) although not prioritised as highly was the need for more financial support. Staff (n = 22) did document that they felt families needed more financial support, however, this was not mentioned as often. The need for financial support came as the fifth most reported theme by staff.

It appears that many families are paying for things themselves and have increased costs at a time when they have a reduction in income/ability to work as a result of caring for a child with a life limiting condition. The work by Glendinning and Kirk (2000) supports this finding as they documented that parents have to give up work at a time when their expenditure is increased by extra laundry, heating and caring costs. Previous research found that parents often experience difficulties in combining caring with paid employment (Heaton et al 2005; MacDonald and Callery 2007). A recent report by Contact a Family (2012) also highlights that many parents who are not in work due to their caring responsibilities are going without meals and cannot afford to heat their homes.

'I am in a lot of debt and I have borrowed a lot of money from friends and family' (Parent, 249).

'Cost is exceptional and we are in major debt' (Parent, 258).

'Single parents have to sometimes give up careers and job to care for their child, is this fair? This has a big effect on your future when your child leaves or dies you have to start your life again but with very little but you have given a lot' (Parent, 266).

'My partner and I both had to give up full time employment to care for our child. After our child passed away I was able to find employment but my partner has only just been able to gain employment three years later. During this time no financial support has been available and some would have been welcome in this stressful time' (Bereaved Parent, 1301).

'Money is instantly stopped for DLA and my job were looking after my child, all the nurses involved stop phoning and, you are just expected to get on with life. If my son was in care the Government would pay but when you love your child and care for him for their lifetime and they die you get nothing' (Bereaved Parent, 202).

Not being able to work and having a child who is very gifted has meant enormous transport costs and further fragmentation to family time. Slowly debt is beginning to build up and we are looking to move and I am seeking work-not sure if PCT will provide care. Do not want to lose our home' (Parent, 201).

More Well-Being Support

The need for well-being support such as emotional and psychological support was reported to be needed by parents (n = 13), bereaved parents (n = 5) and staff (n = 51). This incorporated a wide range of support mechanisms, such as counselling, support groups and the need for staff that have cared for the child to stay in touch as they have been part of the family sometimes for years. The Department of Health and Department of Skills and Education (2004) in their National Services Framework for Children, young People and Maternity Services highlighted the need for families caring for a child with a life limiting condition need more well-being support to reduce their levels of stress.

'For medical staff to stay in touch after child passes away. They were part of our family for three and half years……losing a child is the worst thing anyone can experience so protecting the

family afterwards is paramount to helping them still be members of society but, there is no support out there for families' (Bereaved Parent 202).

'Parents should be told how difficult it is going to be watching your child take their last breath, how horrible the noise sounds, nothing can prepare you for it' (Bereaved Parent, 503).

'More support psychological & bereavement support' (Staff, 413).

'More local bereavement service for the whole family with home visiting support if required' (Staff, 604).

'Counselling provided to help us deal with having a child that is disabled or who has died. Counselling for everyone' (Parent and Bereaved Parent, 209).

'Provide on-going counselling for the family especially at diagnosis but on-going' (Parent, 232).

It is important to consider not only the child and family well-being but also staff as one staff member commented –

'Our team works very hard to keep families together and works extremely hard to maintain the packages of care. However, we have lives and families of our own and the off duty system to keep these packages in place is often awful and leaves staff drained, exhausted and ill. How can we be of the best services consistently and put the needs of our lovely families and children first when we often don't have enough staff and the ones we do have are off sick.' (Staff, 602).

'We are very fortunate in our area to have a very supportive consultant who makes herself available 24 hours a day during end of life care that allows families and ourselves access to advice and reassurance at critical times to give the children and their families the best possible death. This has enabled a lot of the families to cope with the tragic loss of their child and reflect positively when life must feel so negative. It would be nice to think this is the case in all areas of the country' (Staff, 701).

More Respite

Respite was found to be lacking by parents (n = 14) and staff (n = 39). This included respite in the home and away from home. MacDonald and Callery (2007) support these findings as parents in their study reported family life revolves around routines of care and that parents often suffer fatigue, depression, sleep disruption and isolation from undertaking this multi-faceted role of caring for their child. Exhaustion, financial strain, changes to usual activities and continuous care giving has been found to contribute to significant mental health issues such as anxiety and depression (Cochrane, Goering and Rogers 1997).

'Everything was a constant battle even got refused a disable badge at first. Had to fight to get respite and only got it the last six months of his life' (Bereaved Parent, 503).

'Increased home care support; Increased respite at home, especially for families who live long distances away from hospices; Better "joined-up" care' (Staff, 114).

‘More short breaks for children; more meeting up of families; Easier to get what a family needs without having to go through lots of red tape’ (Staff, 219).

Additional Themes

The staff reported the need for better transition service (n = 27). This was mentioned by parents but it was not reported as frequently as being an unmet need. Themes that came out almost as high to that of transition from staff opinion were: the need for play specialists and increase in play provision/activities, training and education and joined up working. For parents those themes that were deemed to be important but not reported as frequently as the top four were; poor social services support, wishing that equipment and adaptations could be facilitated more quickly and travel and transport issues.

The bereaved parents reported their top theme as the need for more practical help in relation to end of life, death and bereavement issues. The work of Rabow, Hauser and Adams (2004) supports the emerging themes that were identified in this research in regards what families need and want when caring for their child at end of life and throughout the bereavement process. Rabow et al (2004) describe five burdens of family care-giving (time and logistics, physical tasks, financial costs, emotional burdens and mental health risks and physical health risks). Although children receiving care at the end of life may be supported by complex, intensive medical care it is typically the parents that sustain the child at this time (Rabow et al, 2004). Families play a central role in the practical and well-being aspects of care and decision making at end of life. However, this role may result in the family carrying significant burdens.

4.5.4 Conclusion

When considering these identified themes in relation to this strand’s question ‘under what local conditions are the children’s and families’ care needs better met?’ from listening and reporting on what the families are telling us then the way forward is clear. In order to meet the often complex physical, medical and the well-being needs of children with life limiting and life threatening conditions and their families is for local care teams to respond in a coordinated way. Families report that if they had a coordinated, integrated care package that was planned around their child and followed them wherever they were, be this at school, hospice or home, with a shared team that knows each child well then this would meet their needs in a more sustainable way and provide a better quality of life. It appears that local conditions do not support families fully at present and families express being dragged into bureaucracy and administration, leaving them with a sense of hopelessness as all they want to do is care for their children properly, but they cannot do this alone.

The two main themes identified from the free text responses could not be put more clearly, than as one parent put it:

‘More Communication, More Cash!’ (Parent, 203)

Caring for children with life limiting/life threatening conditions and their families involves services in meeting complex needs for the child’s biological, psychological, developmental and social needs. The comments provided by staff for the purposes of this research indicate their awareness of families’ needs but there are still areas that need improvement. All the partners involved in caring for children with life limited and life threatening conditions and their families aim to work together to improve their health, well-being and experiences. The only way every child and young person unlikely to reach adulthood, and their family will have access to sustainable, holistic, family centred and high quality care and support whenever and wherever they need it, is by the family, professionals and organisations working in partnership. This is a journey that they all take together, walking side by side, step by step. How people die is remembered by their loved ones who live on (Saunders 1989). This makes it even more essential then that all those involved in caring for children with a life limiting or life threatening condition and their families support them to make this profound experience one that is not filled with regret but one of dignity and a memory that their child lived well and died well.

5. Discussion and Conclusions

5.1 Summary of Findings

In the MDS we identified 1300 families including 133 bereaved families across the region. Over fifty per cent of the children suffered from either chromosomal congenital disorders or static encephalopathy for example severe cerebral palsy and this figure was also reflected in the children that had died. Whilst the median age of living children was 8 years the median age of children who had died was 3.5 years. However, the central eighty per cent of children both living and deceased ranged from 1 to 17 years. The proportion of young people over eighteen is relatively small.

Overall approximately twenty eight per cent of living and deceased children were of South Asian background. In contrast the proportion of staff who were from black and ethnic minority populations was 6.5 per cent. The highest proportion of these is living in the Birmingham and Solihull PCT cluster area. Mapping demonstrated eleven postcode sectors where 10-17 children and young people with life limiting conditions and their families were resident. The vast majority of areas have a rates of 1-10 children and young people with a life limiting condition in 10,000 but almost a quarter of areas demonstrate a higher rate. Only seventy five [is this correct?]Postcode sectors have a higher rate of 20 children on 10,000. Five areas stood out as having a higher rate of children and young people with life limiting conditions compared to the usual rate of children. One of these was at the Derbyshire border another was a larger area at the Worcester border and a sizeable area at the Shropshire border.

Buffer zones of 20 miles radius were drawn around each of the children’s hospice facilities including two that were outside the West Midlands area (Loughborough and Oxford). Only forty five postcode sectors in the West Midlands were outside the buffer zones. Twenty four families were living further than 20 miles from children’s hospice facilities, most of these were living in the Shropshire and Hereford area. It is possible that within the twenty mile buffer zone there are more children than could be catered for by individual hospices. This is being investigated further.

Strand 1 found that MPOC were useful instruments for the evaluation of family-centred care for children and young people with life-limiting conditions and their families. It was demonstrated that whilst families perceived their services to be respectful and enabling, the provision of information was less highly rated. This finding is often the case; however parents greatly value the provision of information specific to their child and more general information such as financial advice and availability of services. We found that where parents had a named person to contact if they needed help and advice (care coordinator) their rating of services was higher across all the five domains. We identified items of need where more than fifty per cent reported needs that were not sufficiently met, suggesting that these may be areas that might be the focus for further service improvement or investment. There appears a risk that provision of information may fall through the net unless the role is more clearly defined. Possibly further examination of communication and information provision together with appropriate training.

Again the areas of coordination between services, communication and information giving came to the fore. The needs of families in Birmingham and Solihull were of particular note.

References

Association for Children with Life-threatening or Terminal conditions and their Families (ACT) and Royal College of Paediatrics and Child Health) (RCPCH) (2009) *A Guide to the Development of Children's Palliative Care Services*. 3rd Edition. Bristol: ACT and RCPCH.

Carter, Y. and Petchey, R.(2006) *Evaluation of the BIG Lottery Fund Palliative Care Initiative,*[Internet]Available from:

<http://www.biglotteryfund.org.uk/er_eval_palliative_care_final_report.doc>

[Accessed 26 June 2012].

Cinnamon, J., Schuuman, N., Crooks, V. (2008) A method to determine spatial access to specialized palliative care services using GIS. *BMC Health Services Research*, 8:140-11.

Cochrane, J.J., Goering, P.N. and Rogers, J.M. (1997) The mental health of informal caregivers in Ontario: an epidemiological survey. *American journal of Public Health* 87, pp. 2002-2007.

Contact a Family (2012) Counting the Costs: the Financial Reality for Families with Disabled Children across the UK. London: Contact a Family.

Craft, A. and Killen, S. (2007) Palliative Care Services for Children and Young People in England: An independent Review for the Secretary of State for Health, London: Department of Health.

Cotterill, S. et al. (2008) *West Midlands Key Health Data.* University of Birmingham.
Danvers, L., Freshwater, D., Cheater, F. and Wilson, A. (2003) Providing a seamless service for children with life limiting illnesses: experiences and recommendations of professional staff at Diana Princess of Wales Children's Community Service, *Journal of Clinical Nursing* 12(3), pp. 351-359.

Davies, R. (1999) The Diana community nursing team and children's palliative care, *British Journal of Nursing,* 8(8), pp.506-511.

Davies, R. (2003) Establishing need for palliative care services for children/young people *British Journal of Nursing* 12(4), pp.224-232.

Department of Health and Department of Skills and Education (2004) *National Services Framework for Children, Young People and Maternity Services*, London: Department of Health and Department of Skills and Education.

Department of Health (2007) *Palliative Care Statistics for Children and Young People*, London: Department of Health

Department of Health (2008) Better Care: Better Lives- improving Outcomes and Experiences for Children, Young People and their Families Living with Life-Limiting and Life-Threatening Conditions, London: Department of Health.

Department of Health (2011) Palliative Care Funding Review: Funding the Right Care for Everyone: An Independent Review for the Secretary of State for Health, London: Department of Health.

D'Argostino, N.M., Berlin-Romalis, D., Jovcevska, V. and Barrera, M. (2008) Bereaved parents' perspectives on their needs, *Palliative and Supportive Care* 6 pp. 33-41.
Dominca, F. (2003) Douglas House: a 'respice' for young people, *European Journal of Cancer* 39(18), pp. 2669-2670.

Dulin, M., Ludden, T., Tapp, H., Blackwell, M., Urquita de Hernandez, B., Smith, H. Furuseth, O. (2010) Using geographic information systems (GIS) to understand a community's primary care needs, *Journal of the American Board Family Medicine*, 23(1): 13-21.

Dyke, P., Buttigieg, P,. Blackmore, A.M. and Ghose, A. (2006) Use of the measure of process of care for families (MPOC-56) to evaluate family-centred services in a paediatric setting. *Child Care, Health and Development,* 32(2), pp 167-176.

Evi (2012) Facts about the West Midlands [Internet], Available from:
<http://www.evi.com/q/facts_about__west_midlands_2> [Accessed 13 August 2012].
Faraque, F., Lofton, S., Doddato, T., Mangum, C. (2003) Utilizing geographic information systems in community assessment and nursing research, *Journal of Community Health Nursing,* 20(3), pp. 179-181.

Field, M.J. and Behrman, R. E. (2003) Committee on Palliative and End-of-Life Care for Children and Their FamiliesWhen children die: improving palliative and end-of-life care for children and their families,Washington: National Academy Press.

Foley, R. (2002) Assessing the applicability of GIS in a health and social care setting: planning services for informal carers in East Sussex, England. *Social Science and Medicine,* 55, pp.79-96.

Frager, G. (1996) Pediatric palliative care: building the model, bridging the gaps. *Journal of Pediatric Palliative Care*, 12, pp. 9–12.

Fraser, L., Miller, M., Hain, R., Norman, P., Aldridge, J., McKinney, P.A. and Parslow, R.C. (2012) Rising national prevalence of Life-limiting conditions in children in England, *Pediatrics.* [Internet], 129(4); e923-e929. Available from:
http://pediatrics.aapublications.org/content/129/4/e923.full.html [Accessed 2 May 2012].
Gatrad, R., Brown, E. and Sheikh, (2008) *Palliative Care for South Asians-Muslims, Hindus and Sikhs*, London: Quay Books.

Glendinning, C. and Kirk, S. (2000) High- tec care: high-skilled parents, *Paediatric Nursing*, 12(6), pp. 25-27.

Goodman, D. Wennberg, J. (1999) Maps and health: the challenges of interpretation. Public Health Management Practice, 4: 10-15.

Hatton, C., Akram, Y., Shah, R., Robetson, J. and Emerson, E. (2004) *Supporting South Asian Families with a Child with Severe Disabilities.* London: Jessica Kingsley.

Haynes, R., Jones, A., Sauerzapf, V., Hongxin, Z. (2006) Validation of travel times to hospital estimated by GIS, International Journal of Health Geographics, 5:40-8.

Heaton, J., Noyes, J., Sloper, P. and Shah, R. (2005) Families experiences of caring for technology dependent children: a temporal perspective. *Health and Social Care in the Community,* 13(5) 441-450

Hendry, J. (2006) An Introduction to Social Anthropology: Other People's Worlds. London: Macmillan.

Hewitt-Taylor, J. (2005) Caring for children with complex needs: staff education and training, *Journal of Child Health Care,*9(1), pp. 72-86.

Higgs, G. (2004) A literature review of the use of GIS measures of access to health care services, Health Services and Outcomes Research Methodology, 5: 119-139.

Hunt, A., Elston, S. and Galloway, J. (2003) Voices for Change: Current Perceptions of Services for Children with Palliative Care Needs and their Families. Bristol: ACT.

Hynson, J., Gillis, J., Collins, J., Irving, H. and Trethewie, S. (2003) The dying child: how care is different? *Medical Journal of Australia*, 179(6S) pp. 20-22.

King, S. King, G, and Rosenbaum, P. (2004) Evaluating health service delivery to children with chronic conditions and their families: development of a refined measure of processes of care (MPOC-20). *Children's Health Care* 33 pp.35-37.

King, S., Rosenbaum, P. and King, G. (1996) Parents' perception of care giving: development and validation of a measure of processes, *Developmental Medicine and Child Neurology* 38 pp. 757-772.

King, G.A., Rosenbaum, P.P., and King, S.M. (1997) Evaluating family-centred service using a measure of parents' perceptions. *Child: Care, Health and Development* 23 (1) pp. 47-52.

Kirk, S. (2007) *Young People's Experience of Living with Medical Technology.* Manchester: The University of Manchester, School of Nursing, Midwifery and Social Work.

Langford, M. Higgs, G. (2006) Measuring potential access to primary healthcare services: the influence of alternative spatial representations of population, The Professional Geographer, 58:294-306.

MacConachie, H. and Logan, S. (2003) Validation of the measure of processes of care for use when there is no Child Development Centre, *Child Care, Health and Development*, 29(1), pp 35-45.

MacDonald, H., & Callery, P. (2007) *Parenting Children Requiring Complex Care: A Journey Through Time*, 34(2) pp. 207-213,London: Blackwell Publishing Ltd.

Mash, E. and Lloyd-Williams, M. (2006) A survey of the services provided by children's hospices in the United Kingdom, *Support Care Cancer* 14, pp. 1169-1172
National Hospice and Palliative Care Organization (2000) *Compendium of Pediatric Palliative Care.* Alexandria:National Hospice and Palliative Care Organization.

Pickering, D. And Busse, M. (2010) An audit of disabled children's services – what value is MPOC-SP?, *Clinical Audit,* 2, pp. 13-22.

Rabow, M.W., Hauser, J.M. and Adams, J. (2004) Supporting family caregivers at the end of life: 'They don't know what they don't know' *Journal of American Medical Association* 291(4) pp. 483- 491.

Sahler, S.J, Frager, G., Levetown,M., Cohn, F.M. and Lipson, M.A. (2000) Medical education about end-of-life care in the pediatric setting: principles, challenges, and opportunities*Pediatrics,* 105 (2000), pp. 575–584

Saunders, C. (1989) Pain and impending death. In. Wall, P.D. and Melzack, R. (1989) eds. *Textbook of Pain*. New York: Churchill Livingstone.

Sidhu, K. (2008) West Midlands Key Health Data 2007/8 [Internet] Available from:

<www.pcpoh.bham.ac.uk/publichealth/publications/key_health_data/index.html> [Accessed 27 July 2012].

Sourkes, B., Frankel, L., Brown, M., Contro, N., Benitz, W., Case, C., Good, J., Jones, L., Komejan, J., Modderman-Marshall, J., Reichard, W., Sentivany-Collins, S. and Sunde, C. (2005) Food, toys and love; pediatric palliative care, *Current Problems in Pediatric and Adolescent Health Care*, 35(9) pp. 350-386.

Steele, R. and Davies, B. (2006).Impact on parents when a child has a progressive, life-threatening illness*, International Journal of Palliative Nursing*, Vol. 12, No. 12, pp. 576-585.
Steiner, D.I. and Norman, G.R. (2008) *Health Measurement Scales*, Fourth Edition, Oxford: Oxford University Press.

Steinhauser, K.E., Christakis, N.A., Clipp. E.A., McNeilly, M., McIntyre, L. and Tulsky, J.A. (2000) Factors considered important at the end of life by patients, families, physicians and other care providers *Journal of American Medical Association* 284 pp. 2476-2482.
Together for Short Lives (2012) Together for Short Lives Strategic Plan 2012-2015 [Internet] Available from: <http://tinyurl.com/tfslstrategy>[Accessed 26 June 2012].

Warr, B. (2007) Administrative and practical requirements when a child dies. In Brown, E. with Warr, B. ed. *Supporting the Child and the Family in Children's Palliative Care,* London: Jessica Kingsley Publishers, pp. 67-75.

Whitton, C., Williams, C., Wright, B., Jardine, J. And Hunt, A (2008) The role of evaluation in the development of a service for children with life limiting conditions in the community. *Child Care, Health and Development,* 34(5), pp 576-583.

Wood, D., Clark, D. Gatrell, A. (2004) Equity of access to adult hospice inpatient care within north-west England. Palliative Medicine, 18: 543-549.

Woodside, J.M., Rosenbaum, P.L., King, S. and King, G. (2001) Family-centred service: developing and validating a self-assessment tool for pediatric service providers*, Children's Health Care,* 30(3), pp. 237-252.

World Health Organization (WHO) (1998) Definition of Palliative Care for Children[Internet] Available from: <http://www.who.int/cancer/palliative/definition/en/> [Accessed 1 June 2012].

Worswick, J. (2000) *A House Called Helen: the Development of Hospice Care for Children.* Second Edition. Oxford: Oxford University Press.

Wrigley, M., Barnett, S., Roderick, P. (2002) Increasing the sophistication of access measurement in a rural healthcare study, Health and Place, 8:3-13

Strand 2 Report:

Qualitive Analysis of families' met and unmet needs

Strand 2 TEAM

Centre for Children and Families Applied
Research (CCFAR)
Coventry University

Professor Jane Coad (Strand lead),
Professor in Children and Family Nursing,
Director – CCFAR

Erica Brown
Research Fellow, CCFAR

Dr Christopher Owens
Research Assistant, CCFAR

Jasveer Kaur
Research Assistant, CCFAR

June 2012

Children and Family Unmet and Met service needs

Acknowledgements

Thanks are extended to children, families and key stakeholders who gave consent and time to contribute to Strand 2. Thank you to Sean Graham, Learning Technologist Centre of Excellence in Learning Enhancement (CELE) for technology support.

> We wouldn't swop [child's name] for the world – he has taught us more about life than anything else. ..He has enriched our lives beyond words but it is a very different life... There is nobody in the wider family that this hasn't impacted on
> Participant 3, Mother, Black Country

For further information on Strand 2, please contact:

Professor Jane Coad; Professor in Children and Family Nursing, Coventry University,
Faculty of Health and Life Sciences, Richard Crossman Building
Coventry.
Tel: 024 7779 3802 Email jane.coad@coventry.ac.uk

Contents

Strand 2	page
1. Introduction	76
2. Literature Review	77
2.1 Context	77
2.2 Background	77
3. Aims	82
4. Methods and Methodology	83
4.1 Recruitment of Participants	83
4.2 Data Analysis	84
5. Findings	86
5.1 Demographics	86
5.2 Where service needs are met	90
5.2.1 Family perspectives	90
5.2.2 Beneficial services	91
5.2.3 Professional staff issues	94
5.2.4 The importance of good communication	96
5.3 Where service needs were unmet	99
5.3.1 Service visions for the future	99
5.3.2 Discussion	102
5.3.3 Professional staff issues	108
5.3.4 Communication difficulties	109
5.4 Service visions for the future	113
5.4.1 Meeting needs for the whole family	113
5.4.2 Responsive services	113
5.4.3 Professional Staff	116
5.4.4 Effective communication	116
6. Discussion	118
6.1 Impact on the family	118
6.2 Implications for the family	120
6.3 Professional staff issues	121
6.4 The need for effective communication	122
6.5 Limitations	124
6.6 Recommendations	125
6.7 Conclusion	128
References	129 - 134

1. Introduction

Paediatric palliative care embraces a philosophy that attends to the psychological, physical, spiritual and social needs of the child/young person and their family. The joint ACT/RCPCH Report (2003) recommends that palliative care should be an integral part of clinical practice, available to all children and young people. Such a philosophy presents a major challenge both clinically and culturally, demanding that all providers work together to ensure seamless care that will meet the needs of medically vulnerable children and young people with life-threatening and life-limiting illness and their families.

This Strand 2 report focuses on the project work commissioned to Coad and team at Coventry University which overall aimed to explore met and unmet needs of children and young people with life-threatening and life-limiting conditions and their families.[4]

---

[4] Throughout Strand 2 report children and young people with life-threatening and life-limiting conditions and their families is used consistently.

2. Literature Review

2.1. Context

There is paucity of research in the United Kingdom regarding holistic care for families with a life-threatened or life-limited child. Graham et al (2006) postulated that research in paediatric palliative care had significantly lagged behind that of adult palliative care. Some individual service-providers have undertaken internal evaluations and, although findings may be insightful for the organisations concerned, in general, there is no robust evidence on which to base care in the community (Knapp and Contro, 2009). Furthermore, figures for the numbers of children and young people who have complex health, including those who have a diagnosis of a life-threatening or life-limiting illness vary enormously. Therefore, limited confidence has been expressed in the figures available (Glendinning et al 2001, Noyes et al, 2006).

During Strand 2 a number of searches were undertaken of published literature. Electronic databases including MEDLINE, CINAHL, EMBASE/MEDLINE and ClinPSYC were used. The results were supplemented by policy documents known to the researchers and cited references from relevant papers. The search was limited to papers and policy documents published in English 2000-2012. Rather than being a systematic review, literature was chosen relevant to each strand. For Strand 2 this resulted in searches using the following key words:

- Set 1 Child/children/young person/paediatric/pediatric/families
- Set 2 Life-limiting and life-threatening illness/conditions/end of life care/complex care needs/palliative care
- Set 3 Service provision, met needs, unmet needs
- Set 4 Participation/user involvement/decision-making/service provision

Inclusion and exclusion criteria were established and each paper was scrutinised in relation to the keywords. The search was limited to papers published in English from 1996-2011 and where an abstract was available for review. Reference Manager software was used to collate, summarise, categorise, store and retrieve the relevant search results. Many papers appeared in more than one database and the search terms identified a large number of references. All were scanned for relevance using the set inclusion and exclusion criteria based on the information available in the title and abstract. The papers included at this stage were read in full and descriptively summarised using a data extraction sheet in relation to their aim, health context, methods, participants, setting and results. Following this additional review a number of the papers were regarded as not being directly applicable to the questions being addressed; only a minority of papers met the selection criteria.

The search yielded 274 publications of which 70 were considered relevant to the review of Strand 2. A summary is included of the review.

2.2. Background - Perspectives on Paediatric Palliative Care

Palliative care for children and young people is widely perceived as a multi-disciplinary total approach to care, from the point of diagnosis or recognition of life-threatening and life-limiting conditions (ACT and RCPCH, 2003). Although each family is unique, many families have needs in common (Lenton et al, 2001). Families require a broad range of services across various disciplines and agencies. There is evidence from The Independent Review of Children's Palliative Care Services (Craft and Killen, 2007) that there is inequity of service provision across England. For example there is variation in availability of Children's Community Nursing Services and access to respite care. Furthermore, Hewitt-Taylor's (2009) small-scale study of parents with a child with complex medical needs concluded there remains significant inequity of provision and access to recreational activities, social opportunities and transport.

Since 2003 national and local governments in England have embarked on reforms aimed at improving and integrating health, education and social services for children and their families. Effective partnership working has been a core principle running through a number of key policies (Department for Education and Skills and Department of Health (DoH), 2004, DoH, 2007, HM Treasury and Department for Education and Skills, 2007, Department for Children, Schools and Families, 2007).

It is recognised that a coherent, holistic approach to service commissioning often breaks down because of narrow and short term preoccupation with budget restrictions applying to a single aspect of service need. Poor co-ordination of services has been deemed to have severe consequences for families (Bachmann et al, 2009). 'Better Care, Better Lives'(DoH, 2008) stated vision was that: 'Every child and young person with a life-limiting or life-threatening condition will have equitable access to high-quality, family centred, sustainable care and support, with services provided in a setting of choice, according to the child and family's wishes'. The document goes on to outline that service provision should be:

- Responsive to and respectful of the diverse needs of children, their families and carers; and
- Built around a philosophy of 'children first' i.e. adopting a needs led, problem solving approach rather than relying on diagnostic labels, enabling to pursue ordinary lives and see their children achieve their full potential.

Assessment of Need/Diagnosis

When a child or young person is diagnosed with a life-threatening or life-limiting condition, families enter a world previously unknown to them (Kirk, 2001). Heaton et al's (2005) qualitative small-scale study of parents caring for a technology dependent child or young person found that while the numbers of technology dependent children is increasing, access to education and social activities was likely to be compromised and that caring for a technology dependent child at home place considerable demands on families.

The importance of assessing children within the context of the family has been highlighted in the literature (DoH, 2004, Kerr, 2001). The Act Charter (ACT, 2005) suggests that each child/young person and their family should receive an individual care plan which is reviewed at regular intervals and that, where possible, the child and the family should be active participants in deciding on the Care Plan. Sloper and Bereford's study (2006) found that, although needs were often recognized by service providers, they were often unmet, largely because of financial constraints. The needs of children and young people with life-threatening and life-limiting conditions are on a constant cycle of change through periods of remission from the illness to medical crisis. At each trajectory of the illness family members will have to develop coping strategies. Most families do succeed at adapting, but this is dependent on well co-ordinated service provision that is easily accessible (DoH, 2004).

Advancement in medicine and nursing means that children with complex needs are surviving longer. While et al's (2004) regional study found that the incidence of technology dependent young people living at home is increasing. A large proportion of the children studied had a physical disability in addition to their diagnosed life-threatening and life-limiting condition. Technology dependent children require a high cost package of support and there remains wide variation in the amount of paid care available to families (Noyes 2006).

Respite/Short-break Care

Respite care has been recognised as helping reduce stress and fatigue in parents caring for a child with a disability (Craig, 2006). Eaton's (2008) survey of parents with a life-limited child called for more overt referral systems for families and a regular assessment of need. However, as Doig et al's (2009) findings suggest, most caregivers say that there is a need for greater flexibility and co-ordination of respite care services. This is endorsed by Grinyer et al's (2010) mixed-methods evaluation of respite care offered by a children's hospice in North of England which demonstrated tensions as a direct result of power relationships between the staff who delivered the resources and the families concerned.

Transition

Effective transitioning of life-threatened or life-limited young people from paediatric to adult services is poorly documented and supported by limited evidence (Doug et al, 2012, Knapp et al, 2008). Most of the published studies are small and may not be relevant to the general U.K. population. Families and young people report this phase of their lives as particularly stressful (ACT, 2007, Brown and White, 2008, Tuchman et al, 2008). However, there is evidence that the role of the family in facilitating transition is central to a successful outcome (Kelly et al, 2002). The Independent Review of Children's Palliative Care (Craft and Killen, 2007) acknowledged there was poor co-ordination of services between children's and adult services the result of which was unmet needs among the growing numbers of young people living with a life threatening or life-limiting condition. Since the Independent Review there has been increasing discussion about how best to meet the unique needs of this population of young people. In 2007, the ACT Transition Care Pathway set out six sentinel standards that should be developed as a minimum, with the aim of achieving of services for all young people and families. The newly announced 'Transition Taskforce' (2012-2015) commissioned by Together for Short Lives and led by the Chief Executive of Acorns Children's Hospices Trust, will develop and implement an integrated system for the care and support of life-limited and life-threatened young people making the transition from children's services to adult services.

Professionals and Interdisciplinary Dialogue

Family-centred care demands that all those involved in supporting the child or young person and the family work with, and on their behalf. Carers are likely to come from a wide range of services and each will have their preferred way of working, organizational structures, salary scales and professional development programmes. This can result in what Jassal and Sims (2006) term as ‘different ideologies cultures and attitudes’. P.154. Furthermore Taylor (2000) identified that differences may create barriers and inhibits meaningful discussion with children or young people and families.

Key workers have a pivotal role in meeting holistic family needs (ACT 2005, DfES, 2005) and negotiating access to a multi-disciplinary children’s palliative care team with knowledge about the whole range of relevant support services. Danvers et al’s regional study (2003) which evaluated the provision of the Diana Princess of Wales Children’s Service, suggested that children and young people with life-threatening and life-limiting conditions and their families benefit from an effective seamless service.

The child and family perspective

For many parents bringing up children/young people with life-threatening or life-limiting conditions is a life-long responsibility. Craig (2006); Brykczynska and Simons (2011) and Coad et al (2012) have all highlighted that caring for a child or young person with a life- life-threatening and life-limiting illness places an immense burden on families. Radriguez and King’s (2009) small-scale study of the impact on parental mental health highlights the continuous adjustment by parents throughout the trajectory of their child’s illness.

Although the dying child is often referred to as ‘the victim of illness’ (Brown, 2007), brothers, sisters and entire families are partners in the same experience. The findings of a study by Brown and Arens (2005), suggest that the duration and type of the sibling’s illness, and how well siblings are included in the family situation, are important factors in how brothers and sisters cope. Often the impact of a life-threatening and life-limiting condition is greatest for families with the lowest income. To obtain benefits families need to be proactive in discovering what is available to them. According to research by Simons (2002) parents frequently glean information in a piecemeal way from voluntary organisations or well-informed friends.

The need for good communication

The need for good communication that prepares parents or legal guardians for the eventuality that their child might or will die has been identified as a core issue (ACT 2004, Avis and Reardon, 2008, Department of Health, Social Services and Public Safety, 2003, Midson and Carter 2010). It is recommended that written information is used to support face-to-face discussions (ACT 2004, Department of Health 2004). Communication between families and health care providers has received considerable attention in the literature. The importance of good communication within families has also been highlighted as a significant factor in how well they cope with a life-threatened or life-limited child (Brown, 2007, Down and Simons 2006). Therefore it is important to assess the needs of family members both individually and as a unit (Elston, 2003)

The rights of children to be involved in decisions made on their behalf have been recognised (ACT, 2003, UNESCO, 1991). Adults may however wish to protect their child from information about their illness (Lester et al, 2002). However, Brown’s study (2007) revealed that children may be very knowledgeable about their illness and, providing they are supported by understanding adults, they are likely to voice preferences about the care they receive.

The quality of the relationship between staff caring for a child and the child's parents has been well documented (Hsiao et al, 2007; Zwaanswijk et al, 2007; Coad and Houston, 2007; Coad and Shaw, 2008; Lyon et al, 2009; Enskar et al, 2000; Zwaanswijk et al, 2011; Brykczynska and Simons, 2011; Coad et al, 2012). Swallow and Jacoby's, 2001 study revealed good communication is often the criteria against which families make judgments about the quality of the care they receive. Arguably, when a child is ill and parent(s) seek healthcare advice, the child may not be visible within the consultation, as health professionals interactions are often orientated towards the parent (Hsiao et al, 2007; Ranmal et al. 2009). In addition, ensuring all members of the family are included and heard, irrespective of age or abilities, can be challenging for health professionals. Open communication about illness and treatment is regarded as the best policy for children, young people and parents as it leads to improved knowledge and understanding of the illness and its probable consequences and better engagement in decision making (Coad and Shaw 2008). Despite the wealth of evidence regarding professional-patient/family communication, and more specifically communication with sick children and young people, effective communication is often perceived to be inadequate.

Understanding and responding to children's, young peoples and parental/carer body language and verbal expression requires advanced communication skills. Undoubtedly, health professionals' experiences are invaluable in the iterative process of understanding children, young people and family communication patterns. However this needs to be implemented with sound knowledge based on robust evidence. Interestingly, Ranmal et al (2009) published a comprehensive Cochrane review in this area that highlighted that health professionals frequently use their subjective judgment rather than considering the most effective communication strategy during patient-professional interactions.

The need for education

All children have a right to education (UNESCO, 1991). However, many parents/legal guardians report that they encounter challenges so that their child is able to enjoy equal opportunities in schooling. In some cases, barriers are likely to prevent their child realising their full potential (Brown, 2012, Hewitt-Taylor, 2008). Factors which influence educational opportunities include negative attitudes, lack of staff confidence and skills, and confusion over funding responsibility.   It is reported that the transition of young people to post school education and vocational opportunities, so they are able to achieve the goals to which they aspire is problematic, particularly in respect of government policy (Knapp et al 2009)

3. Aims

The specific aim of Strand 2 was to identify and document met and unmet needs of families so that these can be communicated to local and national agencies as evidence for the need for improved services.

4. Methods & Methodology

Appreciative Inquiry (AI) was chosen as the most appropriate approach to underpin Strand 2. Fundamental to this approach is the desire to discover 'what works well' and 'why it works well' (see for example, Cooperrider and Whitney 1999). Appreciative Inquiry has been used effectively within a variety of complex, organisational structures including health and social care settings (Carter, Coad et al 2009). Appreciative Inquiry lends itself well to a pragmatic approach which was felt to have good fit within the current study. In Strand 2, we used one to one or focus group semi-structured interviews in a setting chosen by participants and asked participants using arts-based tools what was good about services or met needs; what could be better about services or unmet needs and what the ideal future for services would look like

4.1. Recruitment of children and families

The study took place from July 2011 to March 2012. In line with the original brief and Research Ethics Committee agreement potential participants (affected children and young people and families including parents, legal guardians, family carers and siblings) were approached through Strand 1 invitation letters. Potential participants returned slips opting into Strands as preferred including Strand 2. Strand 2 also independently approached all the schools across the West Midlands and 29 agreed to take part and send out invitation letters. The inclusion and exclusion criteria were set for Strand 2:

Inclusion criteria:

50 to 80 families using the services of West Midlands Children's Services
Families living in the West Midlands NHS clusters and Gloucestershire NHS Cluster
Children and young people with a diagnosis of a life-limiting/life-threatening illness and in receipt of children's services and their families

Exclusion criteria

Families not using the services of West Midlands Children's Services
Families not living in the West Midlands NHS clusters and Gloucestershire NHS Cluster
Young adults not in receipt of children's services

Recruitment was generally very good *(Table 1, page 11)* with 66 families initially agreeing to take part. Of those 51 families were then interviewed with in-depth interviews in the home setting or focus group workshop as preferred. In total 65 individuals were interviewed and subsequently analysed.

4.2 Data Analysis

The principles of the framework analysis approach were used to examine and explore the data. Briefly, the framework analysis approach consists of three interlinked stages: data management (identifying codes from the data and grouping codes into broader categories); descriptive accounts (mapping the range and diversity of data to initial categories and constantly refining categories until the 'whole picture' emerges); and explaining the findings and in the context of this project brief included making recommendations in relation to the course structure and delivery (Smith and Firth, 2011).

The analytical procedures involved:

1. Initially, five interviews (4 adults and 1 child) were subjected to verbatim transcription and two members of Strand 2 read and re-read the narratives in order to become familiar with the data. Codes and categories were created by considering each line, phrase or paragraph of the transcript in order to summarise what participants were saying with key phrases highlighted and comments written in the margins to record preliminary thoughts.

Key phrases were summarised using participants' own words ('in-vivo' codes) and 'initial categories' were identified. A coding index was developed from the initial categories, which were used as a means of sorting and organising the whole data set. Once all data sets were coded, initial categories were refined and sorted into three main sub-categories within the themes of met needs, unmet needs and implications for future needs (*Figure 1 overleaf*).

2. Descriptive data were extracted from the remaining transcripts and subjected to descriptive data analysis in NVivo 9, using the qualitative data analysis package.

3. Key quotes were extracted using the broad themes and the overall story of participants' met, unmet and implications for future needs could be told using the framework set.

4. Biases were reduced by ongoing Strand meetings between the research team members and solid verification strategies that were set in place to ensure trustworthiness. These processes were invaluable and enabled critical review associated with the analysis.

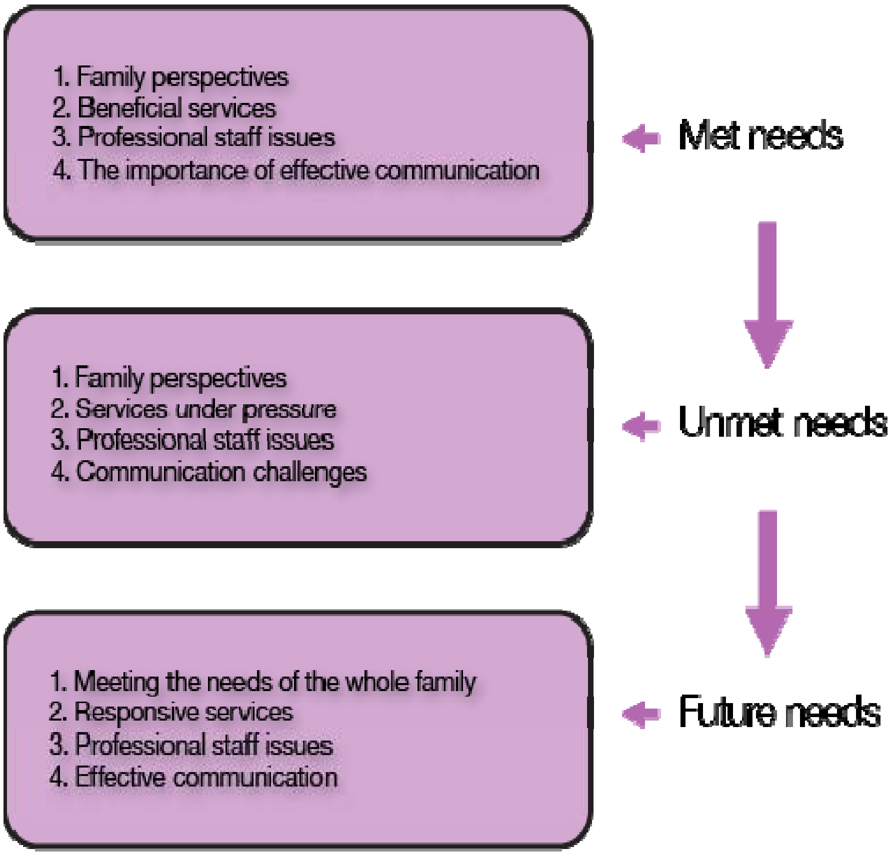


Figure 1: Main sub-categories for Strand 2 data

5. Findings

Direct extracts [as they were said or written] from the data are used in order to illustrate themes and bring the data to life. Individual identities have been protected by not referring to participants by name or specific town, rather using the five West Midlands NHS Clusters and adding a sixth, Gloucestershire NHS Cluster. There are five NHS clusters in the West Midlands but Strand 2 needed to add Gloucestershire as there were a number of family participants who access West Midlands Children's Services but whose home address was within the Gloucestershire cluster (*Figure 2*).

5.1 Demographics

Participant breakdown is included in *Table 1* below. Sixty six families were approached with potentially 180 individuals in total. Of this potential number, fifty one families participated in the study which included 59 individuals (41 adult parent/carers and 18 children and young people aged 5 to 18 years old).

	Families recruited	Potential total Family members	Families interviewed	Individuals interviewed
Adult Data Set (parent/s and family carers)	66	147	39	41 Parent (s)/ Family carers
Children &Young People Data Set (Age 5 years to 18 years)	No child recruited without family consent	33 No child invited without family consent	12	18 Children and Young People (6 parents present with children but who were not interviewed)
Total	66 potential families	180 potential participants	51 families	59 participants

Table 1: Number of Families and Participants Interviewed

The aim of Strand 2 was to understand the met and unmet needs across the West Midlands region. In order to ensure protection of participants Strand 2 used the recent NHS clusters to handle data sets and discuss findings. *Figure 2* overleaf displays the location of where the fifty one families live in terms of the West Midlands NHS Clusters and Gloucestershire NHS Cluster. This represents the total spread of Strand 2 participants who used West Midlands NHS Children's Services.

Figure 2: The Percentage of Participants Living within West Midlands and Gloucestershire NHS Clusters

Given the breadth of family composition within the Strand 2 sample it was important to understand adult's participant's relationship to the child or young person with a life-threatening or life-limiting condition. *Figure 3* shows 74% of adult participants were mothers and 13% are fathers. It also includes a spread of birth grandparents, foster grandparents and adoptive parents who play a primary care role for the child or young person.

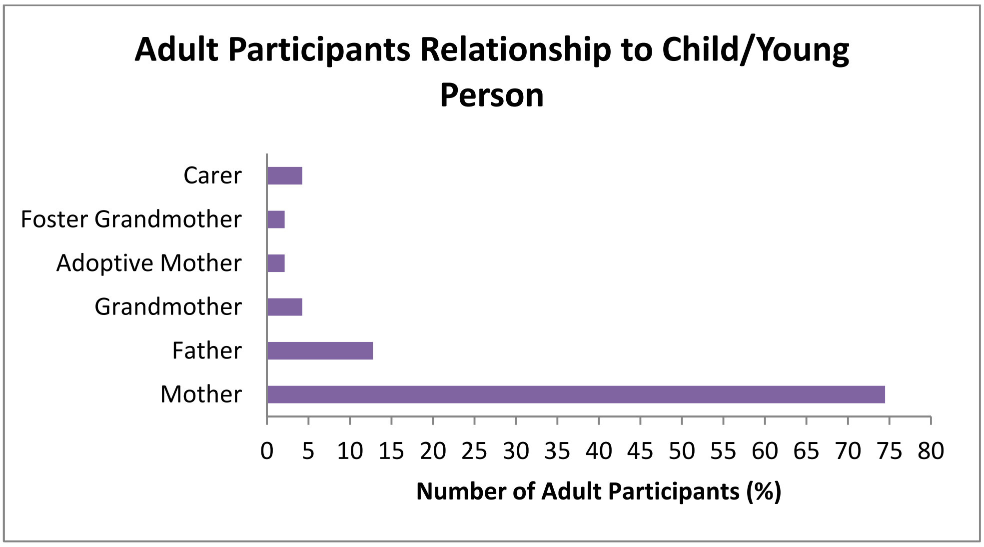


Figure 3: Parent or Carers Relationship to Child/Young Person

Although only 18 children and young people were interviewed, within the fifty one families there were 53 children and young people with life-threatening or life-limiting conditions, of which 55% were male. A variety of age bands of children and young people with a life-threatening or life-limiting condition were accessed as shown in *Figure 4*. Of the total sample, 36% of participants were in the 5-10 age banding and 15% were in the 11-15 age banding.

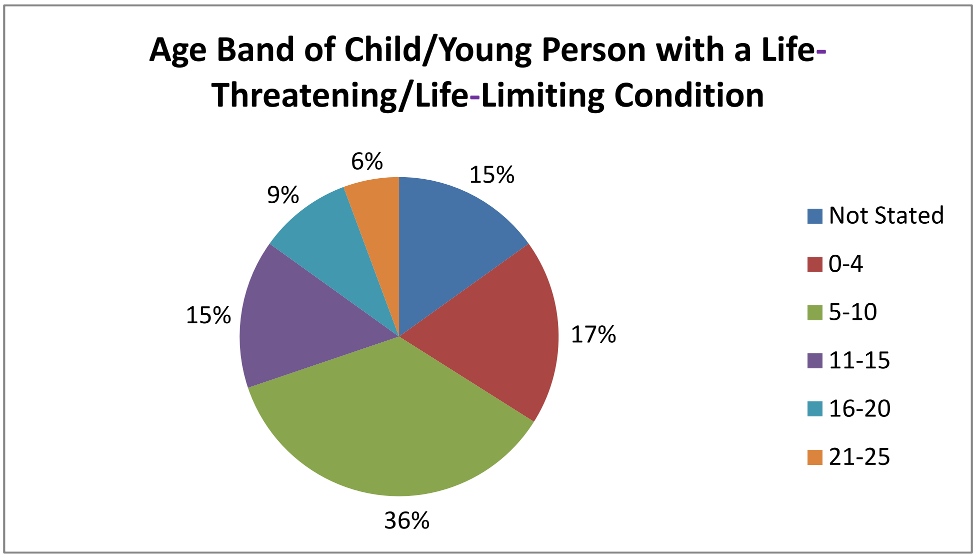


Figure 4: Age Band of Children and Young People of Strand 2 sample

The West Midlands is an ethnically diverse region. *Figure 5* displays the ethnic groups for the participants in Stand 2. The majority of the participants of Strand 2 were White British (67%) which matches the estimated numbers from the Office for National Statistics (2011). British Asian were 11% of participants and 7% were from other Asian backgrounds. This compares with the Office for National Statistics data which states 8.5% of the West Midlands population are Asian or British Asian (Office for National Statistics, 2011).

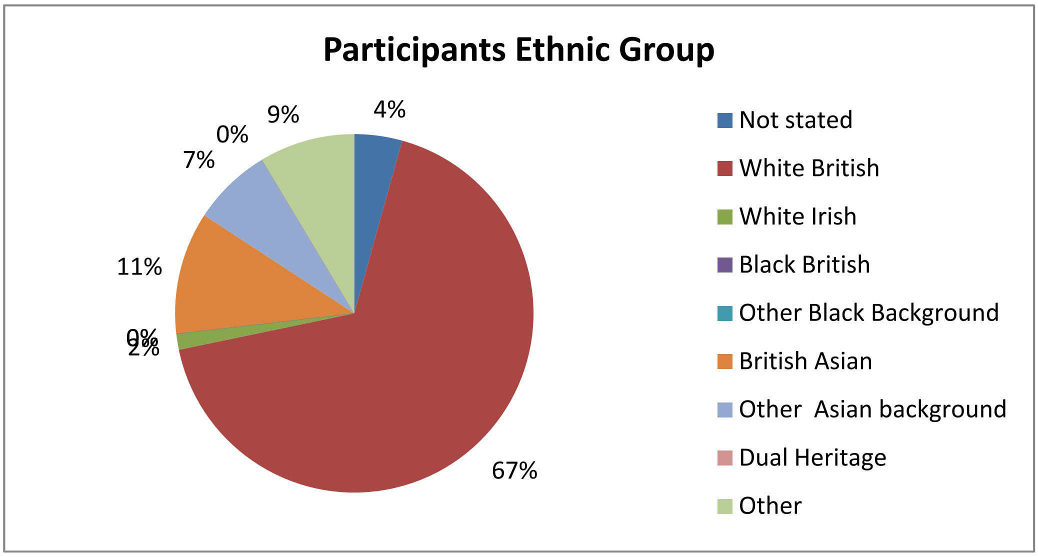


Figure 5: Participants Ethnic Groups from Strand 2

Strand 2 conditions were dealt with using the same criteria for Strands 1 and 4. *Figure 6* shows the variety of the children and young people's conditions. 21% of children and young people's conditions are within the static encephalopathy and congenital and chromosomal group whilst 19% have conditions within the neuromuscular group.

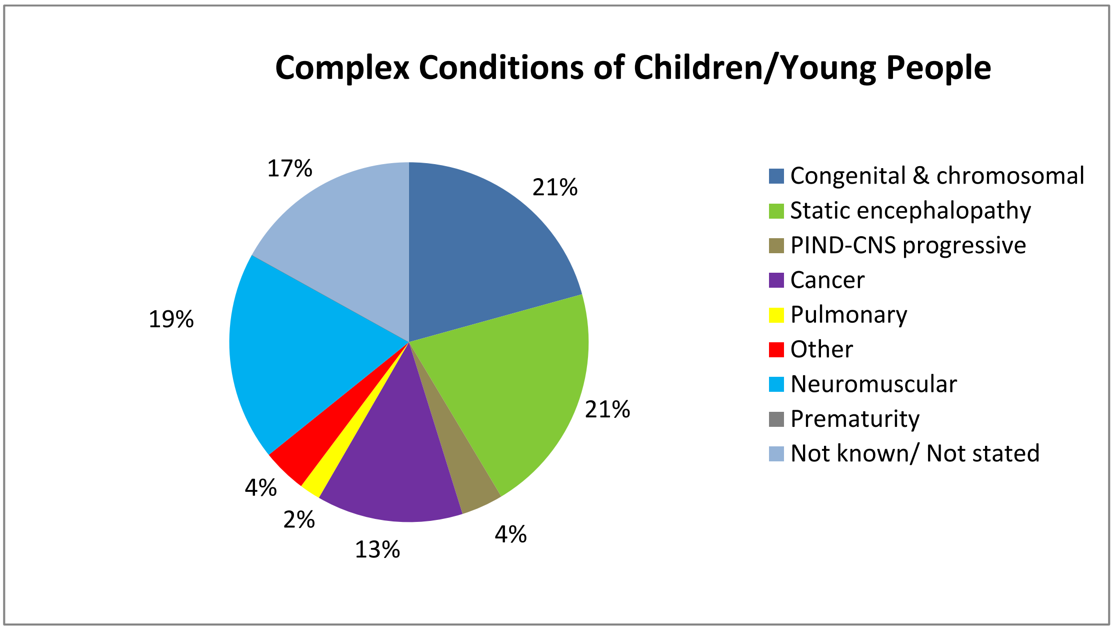


Figure 6: Children and Young Peoples Complex Conditions

Educational provision of the children and young people with life-threatening or life-limiting conditions in the West Midlands has not been mapped previously and findings from Strand 2 data are shown in *Figure 7* overleaf. Within the initial sampling of Strand 2, 29 schools were contacted to ascertain their population of children or young people with life-threatened and life-limited illness. This initial survey revealed very low numbers of children and young people with life-threatened or life-limited conditions in mainstream education. Final number of participants revealed that 51% attended special school and 8% attend mainstream school therefore highlighting that a large number of Strand 2 sample of children and young people with life-threatening and life-limiting conditions were attending special schools and nurseries.

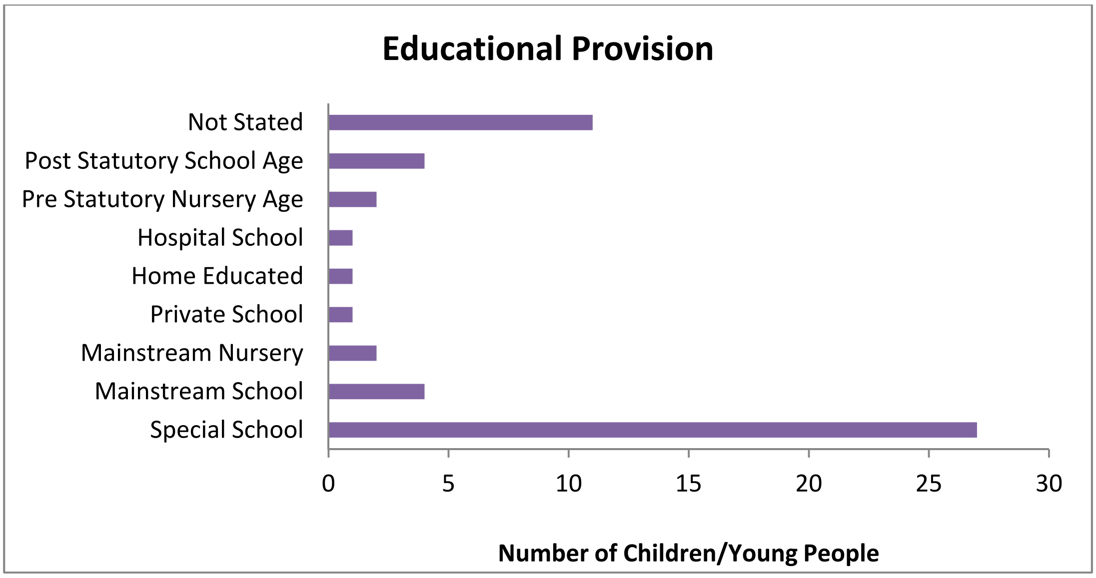


Figure 7: Educational provision attended by children or young people with life-threatening or life-limiting conditions

5.2. Where service needs were met

5.2.1. Family Perspectives

Family coping strategies in order to deal with the situation included parents and carers, such as grandparents or adult siblings, educating themselves and knowing all they need to know to help them to deal with what's happening. Some parents and carers recognised the limitations of what they are able to do, and this was important in helping them discuss their needs and to develop coping strategies. There were also occasions when parents acknowledged the importance to them of talking to their child about the illness and involving the child or young person in decisions that affect them as illustrated;

yes I make sure of it, I make sure they do, I'm an articulate and thoughtful individual and mother of a sick child and I'm not prepared to accept information that I don't necessarily agree with and I will question them about it.
(Participant 9, Mother, West Mercia)

I never hide anything from…(child). I always find a way of telling him. I might make a story out of it or something – I never hide anything – he can't talk but he can hear everything.
(Participant 37, Mother, Arden)

One parent highlighted that their religion (Islam) helps them to cope;

...I think religion... it's really helped because I mean we're Muslims and obviously we believe in god and we believe there's a better place for them ok it's not meant to be here…there's a reason for this and we'll be going to a good place
(Participant 11.1, Mother, Birmingham)

Positive mentality of the parent and the child was important and commonly discussed as a strategy for coping;

There is a really positive side. … is a very happy child. Curiosity is her major thing!
(Family day interview 1, Father, Arden)

She's actually exceeded all my expectations and done far better than I thought she would (Participant 15, Adoptive Mother, Birmingham)

Feeling their role as a parent is to look after the child no matter what and seeing the situation as part of their life helps some parents to cope as illustrated;

I am the sole carer for my son. I gave up my job nine years ago to look after (...) and my husband stopped work a year ago to help me
(Participant 37, Mother, Arden)

5.2.2. Beneficial services

Children and young people with life-threatened and life-limited conditions are living longer. Where needs were met, in terms of a good service, this was one which allowed a child to reach their full potential. Having trust and confidence in a service delivering holistic care to their child was very important to family carers and their day to day normality. All of the parents felt that every child and young person had the right to expect care to be provided at home and that hospital admission should be prevented or reduced to the minimum. Children's hospice services were also perceived as very beneficial to the family members who had used them.

Through the opportunity to access children's hospices facilities, parents and carers are given time and space to separate themselves from their normal caring duties. They felt that the staff were knowledgeable and compassionate to their specific individualised needs but also provided the specialist skills for life-limited children and their families. The choice of leisure activities such as play, information technology or hydrotherapy facilities and opportunities such as social trips for children or young people were highly valued. In terms of regional services, it was identified that hospices were more likely to be accessed by families in the Birmingham NHS Cluster compared to other areas in the West Midlands as highlighted;

When he goes to [Children's Hospice] he'll go around four o clock on one day and then he'll spend all night there and all that day then all night again then I'll go pick him up again in the morning...gives me a good rest so I can sleep, do what I have to do then I'm ready again refreshed for when he comes back (Participant 12, Mother, Birmingham)

Care packages

Care packages were frequently discussed. Planning and delivery of such care packages was a common role that the Community Children's Nurses (CCN) performed and generally this was felt to be a high standard of service given to the child or young person and families. Notable regional areas in alphabetical order were Arden, Birmingham, Staffordshire and West Mercia NHS Clusters. Parents and carers reported care packages were largely complex or continuing care, but as part of the packages of care End of Life (EoL) and palliative care were often frequently cited as most intense. This high quality palliative/end of life care was needed 24/7, based around the families needs. Despite limited resources in some cases, parents felt that teams, especially medical and nursing, accommodated choice and implemented a number of local initiatives to support the whole family. For example, a family who owned a home-based business was able to continue caring for their child at home because carers made a first call to the home in the morning which enabled the mother to prepare breakfast for guests who were staying. On another occasion, one of the community nurses travelled to a hospital appointment with a mother when the child was prone to fits and could not have been left in the back of the car alone during the journey.

A further service that was repeatedly reported by children and families as beneficial was the respite and short break care services offered. Respite offered the families assistance with day to day care to take pressure off parents/carers and makes a positive change to the daily lives of parents. Respite is arranged through different services across West Midlands Children's Services for example through community respite services, hospices and voluntary organisations.

Overall, parents/carers in all NHS Clusters had confidence that the service was there when needed and the continuous support provided is valued. Where service providers are able to respond quickly when a family needs support, this is highly praised as exemplified:

[short break care]...it's just like a big house, its lovely its home away from home... absolutely wonderful place...we usually stay over with her they've got parents accommodation upstairs (Participant 24, Mother, Staffordshire)

(In relation to Respite service for Children with Disabilities*)*

Any time you need them they come out especially if she's ill they come straight out erm and if they can't then they advise you what to do to bring her to the hospital and stuff
(Participant 31, Mother, Birmingham)

We accessed support from … (voluntary agency) right from the start. For the last 3 years we have used … (children's hospice) for respite at weekends. We get about 14 nights a year. To begin with he didn't want to stay on his own so we stayed with him.
(Participant 4, Mother, Gloucestershire)

*I went on trip once and it was great so that was nice (*42 Child, Girl, Arden)

Support within schools for children with life-threatening and life-limiting conditions was very varied but overall was *reported to be* important in meeting needs. Children and young people attended both special schools and mainstream education.

In the latter, inclusion of the child or young person into the setting was generally good. Support for transition between phases of education is varied, but generally parents feel that they are included in decisions about their child. Services that are provided through schools (e.g., physiotherapy) which are accessible under one space/area were very useful for families. These needed to be tailored to families in their required environment and the locality to home was important. Many families reported school staff had gone beyond the call of duty and they appreciated this. Special schools in West Mercia NHS Cluster were highlighted as providing particularly good holistic educational care as shown in the following quotes;

Most of those things are 'actioned' through the education system because she's in a special school...there's on site physio, on site occupational therapy erm visual impairment stuff done through there as well...all her aids are done through school as well
(Participant 21, Mother, Arden)

And ….my nurse told my teachers (about my illness) and they all know and look out for me
(Participant 6, Child, Boy, Arden)

Then my teacher came to visit me in hospital. I was surprised. It was alright
(Participant 4,1 Child, Girl, Arden)

He goes to a special school – they are good there. He has 1-1 support.
(Participant 57, Mother, West Mercia)

Three adult participants from across the West Midlands NHS Clusters discussed that they received a Direct Payment to tailor care plans to their needs, which worked well for them. For example, one carer said this allowed a family member to be employed to provide care which allows the parents to have some time out. Other illustrations are included;

Meeting our needs I would say is the direct payment that does work well for us because we've got something to look forward too at the end of the week...we've got some time to ourselves erm so that works well for us
(Participant 24, Mother, Staffordshire)

.. we have direct payment for a community carer 28 hours a month
(Participant 38, Father, Birmingham)

yes one carer is paid through direct payment and the others social services pay for
(Participant 18, Grandmother carer, Arden)

5.2.3. Professional staff issues

Aspects that children, families and non-nursing participants valued were having Community Children's Nurses and that they needed to be flexible, accessible, knowledgeable, friendly and approachable. Repeatedly, families reported that essential skills included good communication and listening. Being the single port of call, in many cases, they were ideally placed to listen to and care for children with life-threatening and life-limiting illness and their families at home, whilst allowing parents to share both their joys and concerns on a daily basis. A number of families spoke about having a Children's Community Nurse available enabled them to be discharged 'earlier' and that they were able to offer a wide range of services and support such as medical treatment, care and opportunities for social interaction. Community Children's Nursing teams in Staffordshire NHS Cluster were cited as providing essential support that enables young people to participate in leisure and sport activities with their peers. Families are supported by a wide range of services such as physiotherapy, speech and language therapy, play therapy, family key workers, sibling workers, teachers, teaching assistants, social workers and health visitors.

There is evidence that services across the region are responding to the individual needs of families and in some instances they are able to adapt protocols to incorporate holistic care as illustrated in two quotes;

Children Community Nursing really astonishing care, offer a collection of tools, like two hourly slots each week, general activities to help with (...) swimming, guides and dancing sessions and hospital team to check on wounds and dressings
(Participant 33, Mother, Staffordshire)

...the [service delivering milk/medicine]...their wonderful to us...the good thing about it, not a year ago I managed to push for ourselves to go holiday...and they sent me a whole box out...a whole medical box...[service delivering milk/medicine] sorted it all out...
(Participant 25, Father, Black Country)

All the children, young people and families felt that both the personal and professional attributes possessed by staff were important. This impacted on their views of the service and the two were often reliant on the other.

Health and education teams in many cases had also provided personal support to parents who were experiencing enormous stress such as difficulty in the relationship with their partner or at times when parents were feeling particularly exhausted. Children and young people felt that when professional and family carers are of a similar age to life-limited young people they were able to relate to them particularly well and they are able to share some interests in common. Other named characteristics included, being highly competent, autonomous, and skilled.

Having somebody to speak to in confidence, who was well trained with up to date skills, knowledge and understanding was repeatedly identified as an important characteristic of staff. They also felt it was important to have a caring disposition and deliver high quality care to individuals and collective groups of children and families. As well as staff being supportive to parents, parents also highlighted the support provided to their children such as being responsive to their needs and making the experience more enjoyable such as;

It important having someone who knows us (Participant 62, Child, Boy, Arden)

They know what they are doing and I feel safe and they talk about TV shows and what we did at the weekend (Participant 41, Child, Girl Arden)

Children, young people and families in the majority of those interviewed lived as normal lives as possible within what is an abnormal set of circumstances. Maintaining routines and enjoying family celebrations are important to the children and young people. They reported high levels of stress, but also coping mechanisms they build as individuals and families. Therefore, professionals who are flexible within the care pathway is important as each child and young person's context is different and protocols may not always be attuned to that particular child and family's needs at a given time. Feelings of consistency and confidence, continuity of care, and trust are important to an overall feeling of security.

Two carers mentioned that staff needed to be appropriately qualified with CRB clearance. Accessibility of support was highlighted as important as was being able to make contact and to receive support out of normal working hours. Community staff from Arden and West Mercia NHS Clusters were praised for their expertise and ability to match support to individual family needs as illustrated;

...it's having continuous contact with someone that's there, that understands what you're going through... (Participant 20, Mother, Arden)

...to know that I can just phone up and have somebody to talk too who knows all about us and [...], today, anytime, twenty four hours a day is wonderful...now that she knows everyone and she's built up a good relationship with the team in [a hospital]...I just can't fault it...
(Participant 53, Mother, West Mercia)

..Our key worker at Special School is like a friend. She gives me one to one emotional support. She is the first person I would contact if there was a problem and she would get the right help for me. My son gets lots of support from there. (Participant 37, Mother, Arden)

Continuity and consistency of staff was important and firmly linked with familiarity, support and trust. Many of the children referred to how well they knew their carers and that this had developed over an extended time period, in some cases all the child's life. Getting to know the health carer is crucial for trust and confidence and levels of familiarity to develop. Many children and young people considered their professional carers to be like members of their extended family. Comments included;

It's a good service (Participant 66, Child, Girl, Arden)

It's important to me that I get the same person for my personal care (Participant 33.2, Child, Girl, Staffordshire)

Consistent support allows relationships to be developed with staff which allows parents/carers to depend on them for advice and emotional support whenever needed. This was noted in a number of areas but two are drawn from The Black Country and West Mercia NHS Clusters;

She's got a good team of Consultants looking after her and she's got the same carers and nurse coming in week after week so their building a nice relationship...
(Participant 2, Mother, Black Country)

...to know that I can just phone up and have somebody to talk to who knows all about us and [...], today, anytime, twenty four hours a day is wonderful...now that she knows everyone and she's built up a good relationship with the team in [a hospital]...I just can't fault it...
(Participant 53, Mother, West Mercia)

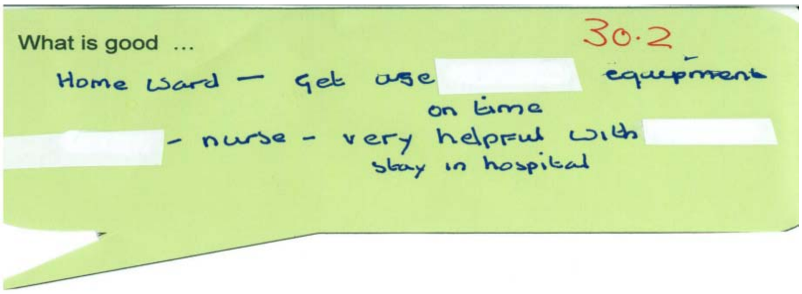
Going the extra mile

In addition to regular service provision, parents and carers across the West Midlands NHS Cluster and Gloucestershire NHS Cluster identified a number of additional services which they found to be beneficial such as transport, specialised beds, leisure and play time activities, These services included additional health services such as Physiotherapy, complimentary therapy and Speech and Language Therapy as well as complimentary therapies for both the child and parent. Parents/carers also directly named members of professional staff who they felt had *'gone the extra mile'* in providing personalised care for their child and helping to make them comfortable and in supporting the role of the parent and carer. Examples included;
He gets physio once a week. A play therapist comes during term time. Then we get speech and language therapy and a dietician as well. (Family day interviews 2, Father, Arden)
(meeting other parents) Yeah through [A Childrens Hospice] they do coffee mornings and parents days out to meet up with other parents and spa days (Participant 31, Mother, Birmingham)

5.2.4. The importance of good communication

A co-ordinated and joined up seamless health and social care service was viewed as very important to children and young people with life-threatening and life-limiting conditions and families. If information is relayed in a positive, clear way this is highly valued by parents/ carers. This included verbal and written communication as well as discussions during organised meetings. Some parents and carers highlighted in interviews specific health and social care professionals or services by name who had been particularly good in communicating with them such as a paediatrician, hospice staff, social worker or a General Practitioner (GP). Being listened to, consulted and included in the 'care team' was also important for parents/carers. Children, young people and families felt that the health professionals provided a much needed and trustworthy line of two-way communication, between themselves and other healthcare professionals. Front line health professionals such as qualified Community Children's Nurses often acted as advocates to the parents, for example offering to be present when other professionals such as doctors were informing them of changes in the care packages. Family members felt that this improved communication and established better levels of consistency and continuity of care across and within services of the West Midlands. The quote below from a young girl highlights this;

….Everybody knows if there's a change in [S] (Participant 66, Child, Girl, Arden)



What is good … 30.2

Home ward — Get use [redacted] equipment on time

[redacted] - nurse - very helpful with [redacted] stay in hospital

Children and young people with life-threatening and life-limiting conditions endorsed a need to voice their views and the importance that professionals place on children and young people's contributions to their holistic care. Some brothers and sisters told of their fears for their sibling. Where services are able to provide opportunities for siblings to meet other children who have a brother or sister with a life threatened or life limited illness, this is viewed as beneficial, providing that families are able to access the support without making further demands on them. Thus, having someone outside their family to whom they could talk provided a much needed 'safety valve' when children and young people were confused or frightened and they did not want to talk to their parents. Parents also spoke of not feeling judged or criticised by service staff and being able to approach them with the seemingly small things that cause them anxiety. They provide counselling and someone for the parents to talk to when they are upset such as;

yeh yeh i get a say in what happens [gets a say in terms of care received] (Participant 8.2, Child, Boy, West Mercia)

She talks to me on my own sometimes … yeah I like that (Participant 41 Child, Girl, Arden)

Yes they talk to me (Participant 44a, Child, Boy, Arden)

…we get involved not just our parents (Participant 11.2 Child, Boy, Birmingham)

Collaborative working was cited in what works well in terms of school nursing; paediatric and community consultants; ward staff; special schools and health visitors. Participants were generally well informed and spoke about positively about how health, education and social care delivery should be jointly planned and delivered in partnership with the users. Thus, fostering multi-disciplinary and multi-agency working with service users was pivotal, which in turn participants felt would ensure more holistic care to children, young people and families.

What worked particularly well was the inter-team working and sharing of resources through the Common Assessment Framework (CAF). On the CAF, participants felt that families should have shared records with the multi professionals and that there needed to be a more efficient and responsive service. This is highlighted:

They normally have a CAF...eight people sit round a table and say what he needs and where he's got to go and things like that... I've got a person who comes out to my house [female from Children's Hospice] and she explains what happened in the meeting (Participant 12, Mother, Birmingham)

The best thing is when all the doctors and nurses communicate with one another (named hospital). In all departments they work towards the same goal. And they feed back to me about…(son) (Participant 37, Mother, Arden)

Some parents and carers also appreciated that health professionals treat the child or young person as an individual and communicate information to them in a sensitive, thought out way with language tailored to them as highlighted:

...we've now got erm a Paediatrician that actually listens to us; we've found a GP that listens to us... (Participant 24, Father, Staffordshire)

The ways in which information regarding diagnosis, prognosis and treatment is communicated is praised by some parents/carers. In particular the interpersonal skills of the messenger is important in explaining the situation in a sensitive manner and taking the time to answer any questions as well as the inclusive approach of sharing information with other family members.

I was told the news on the Friday and my whole world fell apart. It was awful as you can imagine. It was all done in a lovely way and the consultant told ... (child) but he didn't use the word cancer. (Participant 57, Mother, West Mercia)

Other forms of communication that parents and carers found useful included online social networking sites and support from the extended family, both of which provided wider emotional support with those on a similar journey as highlighted here;

I've actually found an awful lot of emotional support from the internet [social network website]...managed to meet up virtually as you say with a lot of people who have similar difficulties... brilliant source of support (Participant 32, Mother, Birmingham)

5.3. Where service needs were unmet

5.3.1 Family perceptions

In many of the interviews parents and carers, especially mothers (who were frequently the main carers), expressed their anger and frustrations towards the circumstances which they were in. Children, young people and families felt that being told about their condition was primarily the parent's role, but many thought professional emotional support would also be helpful. There were occasions during interviews when participant's emotions inhibited the discussion of unmet services for their affected child/young person and the family as a whole. The complex psychological support needs of family members were thought to often be left unmet, because of the focus on treating the affected child/young person.

Many fathers and grandparents felt that having a child with a life-threatening and life-limiting illness was not fair and as a result this impacted on how they navigated around services whilst trying to maintain a 'normal family life'. Fathers often concentrated on technology needs and equipment during the interviews whilst mothers discussed wider issues. There was frustration expressed from all family members that they were not being able to have a break from their caring responsibilities.

Mixed responses emerged about mothers feeling that their own mother (child's grandmother) or sister (child's aunt) either helped or hindered their child's care. Many family carers described the exhausting level of care they undertook for the child and often without real thought to their own mental or physical health such as;

I don't think nobody out there understood how much medical needs my daughter needed they must of thought I was just a trouble maker mum I'm labelled as this but it doesn't bother me anymore because I'm stronger than them... (Participant 2, Mother, Black Country)
It's a strain making all the arrangements for (...) care. I do it all. (Participant 13, Mother, Staffordshire)

Me and my husband we can't just go out of the house and leave …(son) and de-stress. (Participant 37, Mother, Arden)

Many families referred to the pervasion of caring for a child at home and although no participant said that they would want this to be different, some commented that their family home was more like a hospital or clinical setting than a real home. This was not only because of the amount of equipment, but the actual presence of carers, especially health services, coming in and out of the family home all the time. Family privacy was therefore compromised.

Being able to take part in social activities was difficult to arrange and therefore families would benefit from services which enabled them to take time out in leisure pursuits. Some participants referred to changes in family and social dynamics and circumstances due to situation of having a child with a complex life-threatening/life limiting condition. This frequently added to the strain of couple relationships;

Our home …it's just a public building I said that to my husband - we both feel like that (Participant 9, Mother, West Mercia)

Changing circumstances

When circumstances change such as holiday time, moving house or changing jobs this appears to put the parental carers and all family members under enormous strain. This demonstrates how stressful having a child with a life threatened or life limited condition can be and in some cases, the guilt felt, as parents cannot spend equal amounts of time with their other children. This is illustrated;

...I've got next week off now it's half term...because what happens is when [...] off we can't spend time together one has to go to work and one has to look after [...]...like when our Christmas I couldn't take any time off...we haven't had a holiday together for a long time... (Participant 25, Father, Black Country)

I feel that I have let my other children down because all I ever did was for... I didn't neglect the other two but I didn't give them any of my time. (Participant 59, Mother, Birmingham)

...I didn't feel like a mum for the first year I felt just like erm ….a secretary because I was constantly chasing up people for appointments. (Participant 24, Mother, Staffordshire)
I am so isolated – the situation is very isolating. (Participant 57, Mother, West Mercia)

Frustration and marginalised

Parent and carer feelings impacted on relationships with professionals especially if they felt frustrated and protective because people were inconsiderate, incompetent or demonstrated a lack of empathy with the child, parent or family members;

it makes me feel humiliated and I'm at the bottom of this little heap asking you for help, and I got a bit personally really I said and you wouldn't like it if people were marching in your house everyday telling you what to do you wouldn't like it would you? And you're standing there so confidently with your paediatrician hat on and your GP hat on you would feel very differently about your identity your children and your families. (Participant 9, Mother, West Mercia)

There were reported situations where parents had lost faith in the quality of service provision which they feel has jeopardised their child's survival and recovery after an acute episode of bad health. There were particular anxieties about their child's eventual transfer from paediatric to adult services in parental reports from The Black Country and in West Mercia NHS Clusters. There is a feeling that adult services are even less likely to meet their needs as highlighted;

…I had a choice whether to keep [...] under children's services or move over to adults and I was so peed off with children's service I decided to go over to adults
(Participant 2, Mother, Black Country)

…I always thought children's services were bad depending on what social worker you got at the time, but adult services are a hundred times worse so at the moment we are kind of enjoying children's services at the moment because we no what's coming when he makes the transition
(Participant 22, Mother, Staffordshire)

Lack of staff expertise concerning a child or young person's condition was cited by parents as a cause for concern and in the case of physiotherapy, one child was unable to have treatment because a school did not administer the medication the child needed before exercise. Lack of communication between Occupational Therapists and neurologists and families was cited in Gloucestershire and Arden NHS Clusters with occasions when phone calls and emails from parents were not responded to such as;

The real problem with OTs was the first one that came was fine but later ones more difficult to access. I've tried the manager and the manager's manager. It has been a complete lack of communication – phone calls are not returned – no response to emails. For example at the moment ... hasn't got a bath sling
(Participant 14, Mother Gloucestershire)

I find my drawback is …..that they [specialist regional service] don't seem to understand, I know that they have got a big work load but when you phone someone up over two weeks ago on their advice helpline you do expect to have some response...that's something that I don't get off my local team we are involved with [...]..I shouldn't be chasing specialist services
(Participant 20, Mother, Arden)

Families discussed on a number of occasions that the quality of the hospital care environment in which children and young people are looked after, could have an adverse effect on the quality of their experience. Parents perceive that unsuitable accommodation in hospital could have a detrimental effect on their child's complex needs. Lack of organisational aspects of care and poorly defined staff responsibilities also impacted adversely, as did aspects like poor diet being provided. Extracts include;

Being in hospital is a horrid, horrid, horrid experience as much as um they do try and make it nice for the parents so many politics about who does what…
(Participant 57, Mother, West Mercia)

The (nutrition) food in (named hospital) is awful. When nutrition is so important because they do not feel like eating, what is served is not going to encourage any child to eat – but that is a small, tiny, tiny thing.
(Participant 58, Mother, West Mercia)

At home, provision of equipment is problematic in some areas due to financial constraints, and parents perceive that their child's quality of life may be compromised by for example, not having the correct wheelchair. In their time of greatest need, parents may approach services for emotional support. If this has not been forthcoming, they have felt abandoned and sold short by the system;

I was fighting to get help when she was first diagnosed and really ill, a nightmare. None of the health visitors phoned social services – no-one would listen. Looking back it, it was really neglectful because that was their job. During the time she was worse than she has ever been – the roughest patch – I rang up to say I really needed some help, but there wasn't any.
(Participant 35, Mother, Arden)

5.3.2. Services under pressure

Equipment needs or issues

Equipment needs at home such as feeding equipment, disposables such as syringes or dressings, nappies and ventilation equipment were a repeated source of concern and contention for the family carers, especially mothers. Some reported substantial delays in the delivery of equipment such as walking frames and wheelchairs.

There was a strong sense of there being inequity in provision of equipment which is subject to a postcode lottery and for some this included having to fight for their rights (across all the West Midlands NHS and Gloucestershire Clusters). Some parents and carers felt that they were particularly challenged by limited availability of supplies. There were occasions when parents commented that the expiry dates on prescribed feeds were too short. Other parent and carers reported that equipment was not age appropriate. However, this disparity was a West Midlands Children's Services regional issue. Some families reported inadequate provision of resources and delivery of supplies in their area (such as Birmingham NHS Cluster) whilst others felt their equipment needs were met. Indeed, two families reported that they had moved, believing resources would be better in another area (Arden and Black Country NHS Cluster). Many family carers, including parents, felt that all families, wherever they lived, should be entitled to "the same" service. The current context of care was unacceptable to many of the parents and some of the older children and young people. Some parents also talked about the adaptations they had to have in their homes and the constant issue of limited space such as;

We have to fight for everything – from wheelchairs to adaptations around the home. It puts a huge strain on your marriage because one or other of you nearly always has to fight for something. If we go away he nearly always ends up in hospital – you end up between a rock and a hard place – you need a break but can you take the risks.
(Participant 3, Mother, Birmingham)

I was very lucky we had the house sorted when [...] was five or six years old but she's now seventeen... and the facilities aren't really, they don't meet her needs completely, there not awful but they don't meet her needs as well as they could do...
(Participant 15, Adoptive Mother, Birmingham)

Service delivery difficulties or constraints

It was reported by many parents whose children had been using health services for many years that the 'recession' and 'NHS cut backs' were responsible for impacting on services. Some suggested that unmet needs were 'getting worse'. Further discussion around the issue of regional or local disparity in care packages, most notably health services, was highlighted in many transcripts and that families were aware of these from one geographical area to another. For example, in the areas of Birmingham, Gloucestershire and Staffordshire NHS Clusters the limited issue of specialist physiotherapy and occupational therapy was raised as shown below;

...I'm asking for a Physio, Physio, Physio but nobody knows...every time I try and phone them and ask I never get a straight answer...the only thing I want really need is just the Physio, I would scrap all the treatments at the hospital because we only go there to talk I mean I travel

forty five minutes to the hospital...they talk to me for twenty minutes and send me back home...all I want them to do is get a Physio
(Participant 12, Mother, Birmingham)

More OTs and physiotherapists are needed. What they say is going to work which is what they can afford, realistically isn't going to work for you as a family
(Participant 14, Mother Gloucestershire)

I would like more Physio support definitely erm we are expected to do an awful lot of Physio...were knackered...we both have full time jobs
(Participant 22, Mother, Staffordshire)

Limited joined up services

Many parents also reported that even locally health, social and education services were not joined up and felt that responsive systems and resources needed to be in place to deliver a modern, health, social care and education service. This included rapid mobile and bleep systems and improved use of information technology across services so they did not have to keep telling their story. Hence a number asked why services could not talk to one another. This is further illustrated in the following extract;

There are a lot of charities and services out there that we are unaware of. What would be really good for me as a parent would be one point of call that would bring it all together. If there was one person you could call and ask 'what is available for this, this and this that would be very good for me. A lot of the time I am having to chase up different people for different things – to find information I get one lead and then another lead and you have to do a lot of leg work yourself. If there was one person then they would have all that sort of information like if I need to know about his medicines or events that are happening in the local area
(Participant 45, Father, Birmingham)

The frustration of service-led working patterns

A further issue of frustration related to working patterns, not only health and social care staff, but also that of education providers. A number of parents talked about health care only being delivered five days a week and between 'standard' working hours (09.00-17.00/ 08.30-16.30) whilst other parents had 24 hour care. The former model of working did not meet the needs of life-threatened/life-limited children/young people and their families. Some family carers who needed support after 5pm and could not access local and expert services, reported how they either had to manage their child on their own, or turn to services such as their GP who they felt provided less expert and informed advice. A few family carers had even resorted to using their local Accident and Emergency Services with the aim of seeing a specialist consultant. Standard health service hours usually coincided with school hours for children, meaning that many reports included children and young people missing school or college time to see a health professional such as a consultant or nurse. If this was compounded by having to travel across the West Midlands region, this added to family stress.

Comments about working patterns also related to respite care/short break care. It was reported by parents in the areas of Birmingham, Black Country and Arden NHS Clusters that there were financial constraints. However, they had received overnight help, sometimes by volunteers, and therefore felt that in spite of cutbacks, that they had a good complex nursing package. In other areas, parents felt that this was more difficult to obtain. Some areas such as Gloucestershire and West Mercia NHS Clusters were reported as having little respite on offer. This is illustrated in the following quotes;

The overnight carers would be nice...even if it was just one night and ….a bit closer together, because for [...] she struggles to recognise people from four months ago...a shorter break on a more regular basis would be a lot better...(Participant 24, Mother, Staffordshire)

We have a lot of issues with things sort of being put in place but somebody would be ill somebody would be away somebody would be on holiday a lot of times and nothing ever gets completed till months down the line and we've got our social worker trying to chase it but it just always seems like silly things like medicines wrong you know syringes are wrong, wrong things sent out, there is always something to be doing every week (Parent comes in) we could train to be chemists, doctors, nurses and we could do it all ourselves (Participant 26, Carer, Staffordshire)

Respite and End of Life Care (EOL)

The two areas of service delivery that created the greatest challenges were respite care or short break care and end of life care. Across all the regions of the West Midlands Children's Services, families felt that both end of life care and respite care created an enormous challenge for many of the health services to respond to. These services are highly valued by families and the demand for them is high as illustrated by the following extracts:

There isn't any opportunity for me to have respite where I still feel like I'm in control with the kids there's not many activities that happen in the short breaks as well for younger children more for five and over very difficult to access things for three year olds (Participant 52, Mother, Arden)

Interestingly, when we lived in Coventry, the respite home that she uses she wasn't suitable ...but as soon as we moved to Warwickshire it was suitable (Participant 21, Mother, Arden)

Clearly, staffing and funding limited the capacity of health and social care teams to respond to a family's choice for their child's end of life care to be provided within their own home. Equally, there were problems in having appropriately trained and experienced nurses to provide the actual care. It was also reported in many accounts of parental sadness and frustration in that a number of the hospices were not now able to care for their child after 18 years of age. It was suggested that as the group of children and young people were living longer the age of cut off should be re-evaluated as highlighted;

[In relation to one of children's hospices] The only thing that was a great shame...they were taking children up until they were twenty five...unfortunately...earlier this year...the trust made the decision that they'd actually stop children going there over eighteen
(Participant 15, Adoptive Mother, Birmingham)
...the things that have been offered so far don't fit in with our needs for example when [...] may have a couple of nights respite at [a children's hospice] or any hospice that can provide for his age group which obviously is a difficult age in a way and with his needs that we would have to

take him......it's too far... because of the erm needs that he has...our needs as well... (Participant 19, Mother, West Mercia)

Residential location and access to services

There is evidence that there are gaps in service provision across the region and in some cases this means that a family has to travel a considerable distance to a specialist children's hospital or unit, some within the region and some to national specialist hospitals. In some cases parents are frustrated because they perceive that a postcode lottery determines whether they are able to access support or not.

We have to go to (city hospital) for first course of each IV treatment. It needs to be done in hospital in case there is a reaction. Why can't it be done at.... (local hospital). It is just six miles down the road, instead of 20 miles. People can choose where they have their operations so I don't see why I can't choose where ... has her treatment. When I spoke to ...(consultant) it was as if I was the only parent with needs outside the postcode lottery support. (Participant 35, Mother, Arden)

Facilities and appointments

However, the biggest frustration by far was lack of facilities at hospitals and health care settings such as GP clinics. This contrasted with good provision in the regional hospices. In terms of hospitals, frustration usually related to having their own space, including storage on the ward and affording them some privacy;

The things that drove me completely mental after having months and months of no sleep was the alarms going off all night long and the pumps going off all night long and the nurses didn't respond to those really...that could be improved I think (Participant 22, Mother, Staffordshire)

Travelling with a child with complex care needs is an enormous undertaking for many parents and often necessitates careful planning. When there are delays in appointment times parents are angry that they are wasting precious time such as;

but it's also the physios and OTs they will make appointments and turn up an hour late and they don't understand that time is precious (Participant 31, Mother, Birmingham)

School or education provision

Schools or education provision needs to have an appreciation of children and young people with life-threatening/life-limiting illness and with complex care needs. Special schools were reported in the previous section as the preferred option for children with complex needs because staff were able to match the teaching and learning to a pupil's individual cognitive ability. In mainstream schools, children often encountered difficulties in respect of learning and listening, reading and concentration, memory and organisation. It was also harder to maintain continuity of learning in the mainstream if the child had frequent appointments which took them away from the school environment.

Some children and young people had medical needs that had to be attended to during the nursery or school day. There were occasions when a child or young person's absence from school meant that they missed assessments such as SATs and this often reinforced the idea for parents and for the child that they were in some way 'different' from their peers such as;

.. the nurseries just wouldn't take her on...the only one we found which was five miles away from home erm we had to travel and it's costing us a fortune to pay for it because we're both in full time work we couldn't get any discount or nothing...

(Participant 25, Father, Black Country)

We wanna keep him at school he only does half days now…but he's like the only one in a wheelchair there like all the time…(Carer comes in) it does affect [the male child] a lot because he does want to be the same as the other children (Parent comes in) this is what we've been fighting for with the school…to them it seems like okay he'll be fine if he stays in his wheelchair all day but to [the male child] that destroys him (Participant 26, Mother and Carer, Staffordshire)

I don't think they really grasps that at all and education has been a nightmare because she is a statement they think they you are willing to home educate if you take them out of school and I'm not because I have so many other things to do just nursing her
(Participant 9, Mother, West Mercia)

I have lots of problems at the moment with [...] eating...They don't have the facilities to have one to one with him at lunch time to get him to eat...limited diet, orally averse eating is such a chore for him (Participant 52, Mother, Arden)

Speech and language service

Some children and young people had limited audible speech and language and communication difficulties. Children, young people and families talked about health and social care providers who would initially talk to the child, but then address the conversation to the parents, (largely the mother), if the child or young person had not responded in a way the professional was able to understand. Thus, the child or young person was excluded from the dynamic nature of the conversation. There were also many occasions when parents and carers made reference to the child or young person communicating non-verbally and, although not stated as an unmet need, there are implications never-the-less for carers to develop some basic skills in sign language and symbol vocabulary. Educational transition was also stressful for parents as highlighted;

..it starts at fourteen... I think we found out about fourteen weeks before he was due to leave his education environment... We did ask the question then he has been in transition since he was fourteen why does it take until almost the very last minute for our security and peace of mind...but we have got that now...but it was a very hard and erm stressful time getting there...
(Participant 19, Mother, West Mercia)

Financial constraints

Financial constraints were identified by parents and carers as an issue in their lives which they were concerned about. These concerns were mainly in relation to three areas. Firstly, the impact on personal circumstances due to a lack of money as summed up by one participant;

I need more financial support ......things are going up. When she was in hospital it was really expensive. I get about £40 a week disability allowance but my expenses are about £200 a week. (Participant 35, Mother, Arden)

Secondly, parents and carers also recognised how high costs are limiting the services they already receive both in terms of equipment and support received as well as changing the way services are being run as highlighted below;

they said they don't provide care assisted chairs because of the cost of one. If we wanted one we would have to buy it ourselves (Participant 18, Grandmother, Arden)
and the hospital said they couldn't cover [....] needs at night anyway, you know i understand that entirely because of the financial issue and nursing constraints that makes total sense to me. Participant 9, Mother, West Mercia)

Sadly because of cuts because of economic problems it used to be a cut off point of 25/ 26 but now it's 18 so [....] won't be able to go again after march next year because that's when the new ruling comes in (Participant 8.1, Mother, West Mercia)

We lost the complex nursing package in the home literally overnight because of cut backs – the powers that be decided that … had shifted from one category of need to another. One day we had support – the next day we didn't. (Participant 3, Mother, Birmingham)

Finally, parents and carers expressed concern regarding financial constraints, was the impact on service provision due to 'cut backs' and their lack of confidence that they would receive services in the future, as reflected by one participant;

I think it's going to be harder because services are going to be cut, certainly in the next decade. Services are going to be cut charities are going to feel the pinch and I really think it looks quite bleak, that's why I can't think about it and its a week at a time you know. (Participant 8.1, Mother, West Mercia)

Children and young people with life-threatening and life-limiting conditions were aware of the current financial constraints affecting their service provision. Young people suggested expenditure should be focused on increasing healthcare services and greater availability of mobility equipment such as wheel chairs. Some of the older young people had responsibility for deciding how they would spend the Direct Payments allocated to them and this gave them a vital sense of responsibility. If financial support was withdrawn, this often compromised the quality of the young person's life and that of their family. Extracts highlight this;

Not good really now that they're stopping the money and everything so less services are going to be given out (Participant 11.2 Child, Boy, Birmingham)

erm the most important thing to spend it on would be healthcare and wheelchairs and stuff like that that's all (Participant 8.2 Child, Boy, West Mercia)

...it was really difficult actually with financially caring... (Participant 25, Father, Black Country)

5.3.3. Professional staff issues - Staff skills and attitudes

There were examples cited where experienced staff inspired parental confidence. Conversely, junior or less experienced staff were often doubted in terms of having the skills and knowledge in the care of children and young people with life-threatening and life-limiting conditions. Parents and family carers also felt that they were experts in their child's care and there was evidence cited of some individual professionals being dismissive of parental skills;

...with some of the more senior staff who had the confidence and experience to say actually I wouldn't recommend that you do that because I've seen this and this before you know and that was a real help because we didn't know what we were dealing with erm but some of the more junior ones you'd say something to them and they didn't really know what drugs they were administering really... I was really amazed that Chemotherapy Nurses could be like that... (Participant 22, Mother, Staffordshire)

Over-familiarity of professional carers with families was also identified as being problematic especially when an individual has been working with a family over a long period. This can be perceived by parents as the professional carer taking advantage of the situation.
Sometimes I think with the carers .. I do find you know .....kind of start take advantage if they think someone's nice... (Participant 11.1, Mother, Birmingham)

Continuity of care

Continuity of care is one of the hallmarks of parental perceptions of what constitutes good care. Continuity inspires confidence and it is also important in maintaining good parent and professional partnership. There are occasions when parents and carers expressed concern that a person named as their point of contact was not a person with whom they felt able to share their concerns;

When our care was transferred to… (local hospital) the doctors and nurses did not communicate with each other. Every time we have to go to A&E or he has to be admitted there is a new doctor each time and I have to give a complete story from when he was born Why isn't the information already there on computer? Why haven't they got his notes? (Participant 37, Mother, Arden)

Parents and carers also mentioned occasions when different professionals gave them conflicting advice and this was particularly disconcerting when parents were learning new complex medical procedures or when parents had to hand over the administering of medicines to their child as highlighted;

He's recently had a gastrostonomy fitted erm we obviously didn't know nothing about it we had a specialist nurse that came out to show us how to use it, what we needed to do. We had the community nurses then come out which did a different practice to the specialist nurse which completely confused us and …I was like come on someone tell me what I'm doing, am I doing it right. …it was very confusing for us… (Participant 26, Carer, Staffordshire)

Parents and carers were also frustrated when they have to make repeated requests about their child's care and they may expend a great deal of time and energy chasing up professionals with their unmet needs or endeavouring to sort out mistakes which could potentially have compromised the care their child received, as illustrated;

They got the chemists kept getting it all wrong all mixed up… (Carer comes in) we've had lots of issues where the wrong medicine, the wrong dosage…I've had to keep going back and going you've not put the right label on…(Participant 26, Carer, Staffordshire)

...I had fights with them GP receptionists on the phone to get the GP to come to the house because they refused and told me to call an Ambulance instead but eventually I got that sorted they came to the house... (Participant 32, Mother, Birmingham)

... you know you have such a turnover of staff in erm any care organisation... you never have that continuity and that relationship build up and when your that far away...understand his needs and identify a change in him that could alert us to something more significant going on... (Participant 59, Mother, Birmingham)

Training of staff

A high proportion of family carers referred to difficulties in terms of availability of community health staff and whether staff had the right training. Some family carers described situations that simply keeled over if trained staff were sick and not able to cover a full shift. When a replacement was unavailable, parents and carers (frequently mothers), reported that they had to take over responsibility for care which added to their stress. Similarly, some families also reported that when their child was admitted to hospital and a ward was understaffed, they were asked (or expected) to care for their child/young person. Some parents talked about being more highly skilled and trained than carers, both in the home and hospital settings, and this did not inspire their confidence. One participant sums this up:
I think a lot of the agencies, when we ask them for carers, they don't send people that are trained having the kind of experience I think that causes a lot of problems….I don't mind training and they listen but they just don't... (Participant 11.1, Mother, Birmingham)

5.3.4. Communication difficulties

Parents and carers rely on good communication between service providers and themselves and between professionals. However, there were many instances cited when communication had broken down and when professionals' interpersonal skills were felt to be lacking. Communication appeared from the data of Strand 2 to be worse in hospital settings, with only one cited breakdown in a children's hospice. Parent and carer experiences were also varied in respect of communication shortfalls across the region. Although Strand 2 data sets can not be generalised, it would appear that Staffordshire had the highest proportion of unmet needs in terms of communication but also included West Mercia, The Black Country and Gloucestershire NHS Clusters. Although Gloucestershire NHS Cluster had the least parental satisfaction, data on this issue was only collected from two families. Parents and carers expressed low satisfaction due to a lack of communication between services and relaying important information to families. Parents and carers were justifiably confused and frustrated by a situation which they perceive as unfair. Staffordshire and Birmingham NHS Clusters scored better, with Arden NHS Cluster scoring highest for communication. However, in all these areas, parents and carers felt they had to tell their stories many times which was exhausting for them. They also complained of struggling to access information about the range of services available via information technology systems. They often expended a great deal of time and energy to get the support they needed, as identified below:

...you have to be in the know to ask for what you want. If you're not aware that something is available you don't know to ask for it...a lot of things over the years we've picked up more and more support but we didn't know to ask for it and then somebody says well aren't you getting that and it's like no I didn't even know that was possible (Participant 21, Mother, Arden)

Communication at the time of the child's diagnosis

Understandably parental anxiety is enormous particularly at the time around their child's diagnosis. The way in which parents received the news varied between face-to-face meetings and through telephone conversations. There was one instance when a mother heard the news through a third person. On several occasions a consultant had broken the news in a matter of fact way and then walked out of the room leaving the parent to cope alone as demonstrated;

Then the paediatrician phoned one evening when my husband was out and said (...) has got spinal muscular atrophy, if you want to look it up on the internet you can find all about it. I remember thinking it was quite callous. It was shocking – I had 6 weeks off work. …..you either go up or go down and I am not going to let … have a miserable life. (Participant 4, Mother, Gloucestershire)

The way we were given the diagnosis wasn't the best – it was in a normal clinic appointment. The doctor was looking at his watch at one point. I asked what sort of research was going on [to help] and the doctor said don't worry about that. – just love him. (Participant 14, Mother, Gloucestershire)

Parental understanding about their child's illness

Parents were unanimous in their need for information about their child's illness from the time of the diagnosis onwards. A large number of families said that they would like more opportunities which enabled them to communicate with other families who were in similar circumstances. This included electronic communication such as the Internet.

In spite of the fact that many parents and carers become experts in the practical care they give their child (including complex medical procedures), there are numerous occasions when it is reported that hospital nurses and consultants address parents either in patronising ways or conversely they speak in medical jargon which is difficult for parents to understand. On a number of occasions there were reports of consultants only talking to the mother when both parents and the child/young person were present. On two occasions it was reported that a hospital doctor failed to give the parent any explanation about the child's illness and just handed her a leaflet;

They (doctors) will be standing there talking about your son and I think please explain it to me. And they get quite offended when I challenge them. I think tell me what is going on. Because I have never been to school – my English is good but my reading and writing isn't good. If you need to fill a form or ask questions it is difficult then. My sister deals with all my paperwork and I feel really bad because I am asking someone else to do it for me. Even the forms – it is horrendous keeping up to date with what you are doing. (Participant 59, Mother, Birmingham)

Failing to take child or parent's views into account

Children, young people and families talked about difficulties in communication between themselves and care professionals within the schools, hospitals and in the community. However, none spoke about this in relation to a children's hospice. The children and young people highlighted the importance of improving communication between health professionals and themselves. There also seems to be a distinct lack of acknowledgement of the parents' and in some cases the children's and young people's level of expertise concerning their condition. This maybe because professionals are unaware of the expertise that families have developed and the complexity of the care they give their child.

In many of the NHS Cluster areas, there were accounts of parents and carers having to repeat their ‘stories’. Some reported there were too many health professionals involved whereas in others, lack of communication was due to the limited number of health professionals and infrequency of appointments. There were no reports of having to repeat stories to social care or education professionals. However, there were occasions when children, young people and families mentioned a lack of congruence between a professional’s verbal language and body language as highlighted;

Whenever we get asked it’s always my mum gets asked ….they won’t ask us (Participant 11.2 Child, Boy and 11.3 Child, Boy, Birmingham)

They don’t tell me everything that’s going on they just briefly say how are you and that’s it…I mean like when the social worker comes … and sometimes then carers with the language barrier you can’t really I can’t really understand what they say (Participant 11.2 Child, Boy, Birmingham)

It was reported that many of the life-threatened and life-limited children and young people frequently experienced an acute medical crisis and had to be admitted to hospital as an emergency. There would appear to be few systems in place which enable a child’s/young person’s case history to be retrieved immediately on their admission to hospital. Parents were frustrated by the length of time taken giving information about their child when it is obvious to them that their child is in a critical condition and they need medication;

Some doctors and nurses won’t listen to you. I told them that (...) was dehydrated and pyrexial and needed IV antibiotics, but the doctor said they had to know all his details and feeding regime first. The antibiotics are important because he gets recurrent chest infections. All his details should be on a computer. Nobody was listening to me.
(Participant 37, Mother, Arden)

Lack of information about support available

The data highlighted that having a child /young person diagnosed with a life-threatening/life-limiting illness plunges parents into a confusing and unknown world. Once they have recovered from the initial shock, most families endeavour to find out as much as they can about the services and support available to them. Across the NHS Cluster areas, the availability of information is very varied and easily accessible information is the exception rather than the rule. Some parents would welcome an opportunity to attend voluntary support groups where they could meet other parents and carers with a child or young person diagnosed with a life-threatening/life-limiting condition. There would appear to be a lack of centralised information about voluntary services/support groups which is easily accessible to families such as;

It is a minefield finding out what you are entitled to. Most of the things we have found out by accident. There are all those services out there but they should make it more transparent – a directory or something. (Family day 1, Father, Arden)

… I mean the care is there out there for you, you just have to find out if somebody sat me down and told me all this someday, nobody told me I had to find out ourselves...
(Participant 25, Father, Black Country)

Collaboration between service providers caring for the child

Most families access a number of services. Collaboration and communication between services is often fragmented and unsatisfactory, which necessitates families having to be proactive in contacting services and updating them with information, often repeating their story over and over again. This is illustrated;

It's hard to check in [...] medication because when they change at the Hospital they don't send the information to the Pharmacy or the Doctors here so I have to go chasing everyone to find the actual proper dosage of medication because they don't want to sign it out or they've put the dosage wrong (Participant 12, Mother, Birmingham)

It is like banging your head against a brick wall. None of the doctors [across region] ...speak to the other – they all do their own thing and that's it (Participant 37, Mother, Arden)

There also appears to be a problem in relation to the information or advice given to parents and carers regarding how to use equipment that a child or young person requires. For example, the following participant's experience captures the lack of communication between a specialist nurse and community nurses;

He's recently had a gastrostonomy fitted erm we obviously didn't know nothing about it we had a specialist nurse that came out to show us how to use it, what we needed to do. We had the community nurses then come out which did a different practice to the specialist nurse which completely confused us and I was speaking to one speaking to the other and I was like come on someone tell me what I'm doing, am I doing it right. In the end I was told by the community nurses not to speak to [a specialist female nurse] to go straight through them which was a bit of a nightmare but that we did, that needs to be improved, they all need to be signing from the same book and saying the same thing…it was very confusing for us…
(Participant 26, Carer, Staffordshire)

Communication between schools and parents

Parent and carers' experience of how well school staff communicated with them is varied. In mainstream settings, they felt that they have to fight for their child to receive the individual support they need if they are going to make progress in their learning as shown in these extracts;

…going back to this (regarding the school)…tried to get [the male child] one to one support in school but the head teacher said no only other child has ever got that and that child has recently passed away so was a lot worse that [the male child] so he'll never get that (Carer comes in) quite dismissive not even looking into it and from our point of view she should look into it and then if it is refused at least we tried.I mean we've been to lots of meetings and we've always come away and been told by the deputy head that actually there is more than one child

in the school which I'm aware of but obviously we're fighting for the child (Participant 26, Mother and Carer, Staffordshire)
When we first went to see my middle school, they didn't have any accessible stuff at all. I was the first person to use a wheelchair. They didn't know what to expect really (Participant 33.2 Child, Girl, Staffordshire)

5.4 Service visions for the future

5.4.1. Meeting the needs of the whole family

Improved emotional support for children, young people and families are strongly indicated from Strand 2 findings. Opportunities provided or offered to enable children, young people and families to take time out such as short breaks and holidays.

I would like sibling support and age appropriate things for him to do. He misses out when (...) is having her IV treatment (Participant 35, Mother, Arden)

We would like to just book a holiday and go, without having to lug everything. (Participant 4, Mother, Gloucestershire)

5.4.2. Responsive Services

- Children, young people and families hoped that there would be age appropriate services in the future, whether that is in relation to education requirements, social care or health care and transition from children services into adult services.

- A fully integrated, holistic, equitable, comprehensive, multi-disciplinary and multi-agency seamless service which has clear geographical boundaries across the region of the West Midlands and has a clear responsibility for meeting the needs of all children and young people with a life-threatening/life-limiting condition and their families and one that is *not perceived to be post code lottery*.
- Far more co-ordination between social services, health authority and the education system when it's children so at the moment everything's with separate budgets and [daughter] ticks all three boxes and I'm not sure how well they integrate between the three of them (Participant 2, Mother, Arden)
- An age-appropriate service needs to be centred on children, young people and parental/legal guardian choice and should take account of what is important to each individual child/young person and their families. As the group of life-threatened and life-limited children and young people is growing, in respect of numbers and complexity of needs, any regional service should take account of this in commissioning.

When I go to clinics and stuff I would like more fun things to do and more sweets out and stuff like that. I would like a short movie at the hospital like the pictures – we went on a boat once and they had pictures which was good
(Participant 41, Child, Girl, Arden)

- ❖ A service that ensures there are skilled, trained carers which includes comprehensive symptom management to meet the needs of children, young people and families. This needs to be a regional service that is accessible, fair, comprehensive and flexible as the child or young person's needs change. There is also a need for a wider range of services to meet growing needs that enables children, young people and families to gain knowledge and opportunities to access new services.

It is really, really crucial that all doctors and nurses are trained so that they know how to approach a child with a disability. Other parents have told me this. …If they have some background knowledge they will know how the approach that child. (Participant 35, Mother, Arden)

- ❖ Improvements include more support for care over the 24/7 period, quicker access to services and available information especially on diagnosis and improving communication in order to provide co-ordinated and joined up services to reduce having to repeat stories.

- ❖ I want the [care] package to carry on... if I could change anything I should be more long term orientated and patient driven not money driven... (Participant 50, Mother, Arden) Regional delivery needs improving in respect of specialist physiotherapy as highlighted in Staffordshire, Birmingham and Arden NHS Clusters. More occupational therapy availability was highlighted in Staffordshire and Arden NHS Clusters.

Definitely the OT services need to be you know improved getting the equipment you know sometimes ...it can take ages to get a vital piece of equipment (Participant 6, Mother, Birmingham)

Increase OT and physiotherapy services (Participant 13, Mother, Staffordshire)

- ❖ A regional service that meets not only physical needs but emotional needs of children, young people and families, starting with diagnosis.

I am looking for a brighter future...we've suffered too much now (Participant 25, Father, Black Country)

- ❖ Genuine choice and partnerships about respite/short break care and end of life care that encompasses the home as the preferred place for care but is flexible enough to meet the needs of families changing circumstances.

I'd like to see a palliative nurse in position in every area of the county who is accessible at least by telephone 24 hours a day for families, especially when you haven't got a nurse on duty …..(Participant 9, Mother, West Mercia)

In a dream world I would want her to be normal. In the real world it would be that we could have more respite – if we could have nothing else it would have to be the respite. (Family day interview 1, Father, Arden)

- Care packages that are life-long and which meet the needs of the child, young person and families which are delivered by competent, trained carers.

- Equipment which is provided in a timely and efficient manner which meets the health, social and educational care needs of the child, young person and family and which also acknowledges that children/young people and family considerations are as important as a professional perspective.

there's no reason why she can't have a good quality of life with the right people around her (Participant 2, Mother, Black Country)

- Improved Information technology to strengthen communication channels. Technology that supports the service including databases which can 'talk' across agencies and settings to facilitate sharing of data as well as developments such as telemedicine to enhance care and support and help overcome (as appropriate) some of the geographical distances between services and families.

- Information of available services to be shared more explicitly with children, young people, parents and carers.

Use e-mail for communication between hospital and community services with parents (Participant 13, Mother, Staffordshire)

More use of computerised information for doctors and nurses (Participant 37, Mother, Arden)

- In all areas, access to the right school and access to the right service to meet the needs of the individual child or young person.

Access to …the right schooling and stuff …..that process needs to be a bit easier or a bit more simplified (Participant 52, Mother, Arden)

- Financial help and support, which is equitable and fairly assessed to relieve financial burdens and help maintain the child's care and family needs.

Thinking of new ways of delivering care: I am going to explore the possibility of Direct Payments so that it gives me the flexibility I need. (Participant 14, Mother, Gloucestershire)

- Improved funding for services. Stable funding is important as funds are currently being reduced, e.g. voluntary organisation funding cutbacks. Commissioning which is robustly calculated and resourced so services can meet the needs of children and young people with life-threatening/life-limiting conditions and their families and which can be intelligently managed at a local level.

[the hospice] is a charity and all its money comes from donations and perhaps they should be government supported given what they offer (Participant 21, Mother, Arden)

5.4.3. Professional Staff

- Professional staff (health, social care and education) to have improved skills and competency training relevant to children and young people with life-threatening/life/limiting conditions and their families, which advances their practice. This would seem urgent in relation to ensuring care needs at home are met.

The Community team are great- really great. When people come to the house it is so much better because it cuts the hassle of having to go to the hospital. (Participant 1, Mother, Arden)

- Improved communication training for staff including how to communicate sensitive information and about the importance of listening to the parent's voice.

You need a lot of understanding people to make this possible, to help, support and arrange. You need training in talking to us about this (Participant 4, Mother, Gloucestershire)

5.4.4. Effective Communication

Improved organisation and joined up communication within and across health, education and social care teams and children, young people and families to ensure all teams are fully informed, up to date and are working from the same information.

For the health teams to actually talk to each other...nobody talks to each other and they don't share information...(Participant 24, Mother, Staffordshire)
What would be great is to have one point of contact...a lot of what we do is coordinating across the two different Trusts...so communication...one email address...or a website...everyone can see what the care plans are and when are the next appointments (Participant 22, Mother, Staffordshire)

Better communication to enable better transitions between hospital and home and from hospital to hospital and children's and adult services.

Better communication between all the allied services who links in well with the paediatrician the consultant and if you have an appointment there that's linked in as well so much more joined up, I'm not sure if that's the right term but (Participant 9, Mother, West Mercia)

6. Discussion

The Independent Review of Children's Palliative Care Services (Craft and Killen, 2007) highlighted the inequity of service provision in children's and young peoples' palliative care across England. Furthermore, a review of the literature for Strand 2 revealed a paucity of research in the United Kingdom regarding holistic care. It is also evident that existing literature generally refers to children's and young people's palliative care in a way that fails to take into account regional differences. Strand 2 of The BIG study has highlighted that there are met and unmet needs in service provision and much inequity exists locally as well as regionally as illustrated in *Figure 8* below.

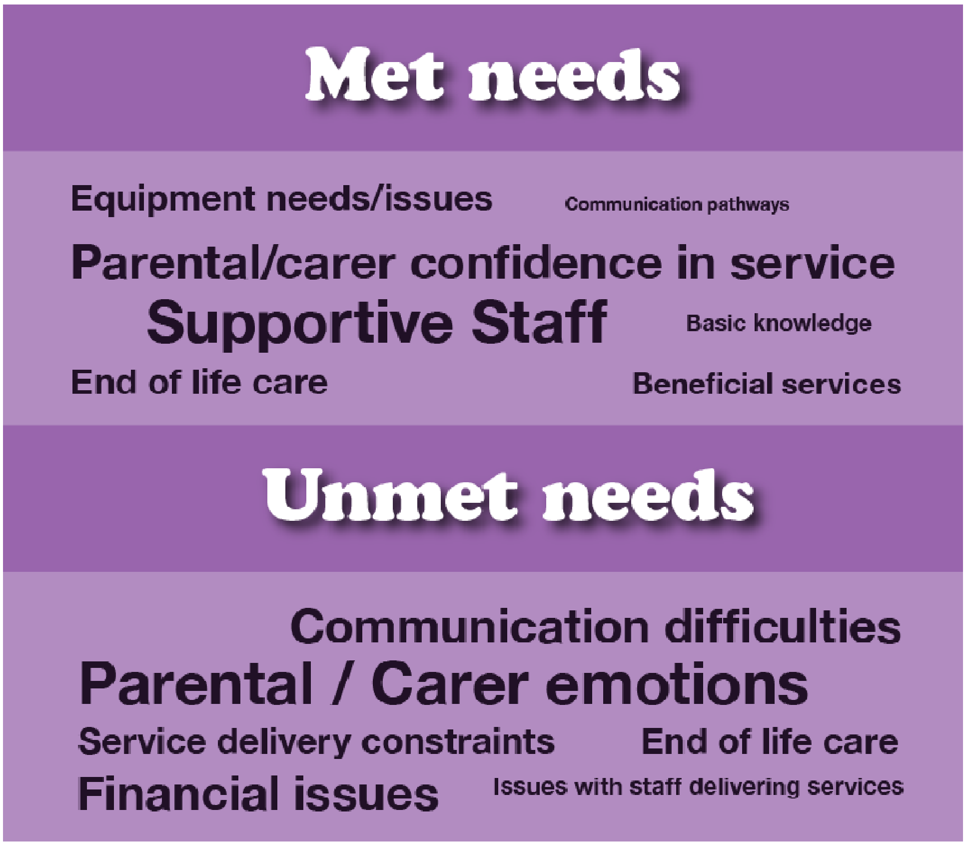


Figure 8: Met and unmet needs drawn from Strand 2 of The Big Study (text size denotes frequency)

6.1. Impact on the family

Findings of Strand 2 highlighted overall that when a child or young person is diagnosed with a life-threatening or life-limiting condition, a family is changed forever. This finding reflects much of the existing literature (Craig, 2006, Brown 2007). In many ways families are bereft as they lose their hopes and dreams for the future and they are plunged into anticipatory grief. This grief is particularly evident at some of the times when normal childhood milestones would have been reached. Many of the family interviews returned to the time when the child/young person was diagnosed. Having to repeat their story many times can impact on the trust families place in a service or on the relationship with the professionals concerned. Data from Strand 2 does however challenge the assumption that families will inevitably struggle with the knowledge that their child's life is at risk (Radriguez and King, 2009). There are numerous examples when parents demonstrate resilience and determination. Against all the odds, families often maintain a sense of optimism, they hope that the situation could be different and this sense of hope may help their determination to continue living their life as fully as possible. Parents and carers frequently said that other parents with a child with a life-threatened/life-limiting condition were worse off than they are. There is a sense of empathetic awareness and a need to be able to access opportunities where parents are able to meet with other parents in similar situations so that they are able to engage in altruistic support.

According to research by Simons et al (2003), parents and carers frequently glean information about services that are available to them in a piecemeal way. Strand 2 findings concur with Simon's et al's findings, revealing that the parents interviewed often gleaned information from other parents or, haphazardly through voluntary organisations. Parents in Strand 2 also made a heartrending plea for easily accessible information, using information technology systems that were responsive and preferably obtainable from a single point of call.

Improved emotional support for children, young people and families are strongly indicated from Strand 2 findings. Findings also highlighted that the families with a life-threatened/life-limited child or young person have a chaotic lifestyle, and whilst some 'coped' well, others just felt they 'coped'. They may 'hold on' to the normal routines of family life while constantly being aware that their child might have a medical crisis at any time. Care needs to directly reflect the expressed needs of the family while also meeting quality indicators. Services should be planned around family lifestyles and routines. Although routines give families a sense of 'normality' and security, but never-the-less these routines are frequently shattered and life is turned 'upside down'. Very often families are unable to plan ahead and they may be reluctant to make even very short-term plans because of the disappointments they have come to expect. For 'well' siblings, this is particularly hard and they may feel apprehensive about taking friends home or planning social events.

Family life is often disrupted and siblings may feel that they are on the periphery of what is happening even though they are living in the same household. They frequently take on extra responsibilities. One of the Key findings of Strand 2 is that some brothers and sisters experience a change of role in the family and a change in status. It was not unusual for siblings to feel intense anticipatory grief in the knowledge that their brother or sister would die. In some families, siblings are the sick child's closest friend and, for young children who have not formed friendships with their peer group, the anticipated loss of their brother or sister means that they will lose the only friend they have. Where services are able to provide opportunities for siblings to meet other children and young people who have a life-threatened or life-limited brother or sister, this is viewed as beneficial, providing that families are able to access the support without making further demands on them.

Similarly, Strand 2 findings shed new light on the experience by grandparents, often as the carers supporting parental care. The geographical location of the grandparent's home in relation to the life-threatened/life-limited child's/young person's home plays a role in the relationship between grandparents and the life-threatened/life-limited child's/young person's family. When grandparents are more readily available, it is not unusual for them to be in almost daily face-to-face contact with at least some members of the life-limited and life threatened child's family. In some cases where a parent is unable to look after their life-limited/threatened child grandparents become primary carers and occasionally they have adopted their grandchild and the study has provided a unique insight into this role.

6.2. Implications for services

Improvements in technology and medicine mean that children and young people with life-threatening or life-limiting conditions are surviving longer and many very complex, dependent children and young people are living at home. Strand 2 of The Big Study has revealed a gap in service provision regionally for this group of children and young people and this is likely to be the case nationally. It is also evident that there may be a real danger that a young person's autonomy will be overlooked under the umbrella of family-centred care, because the focus may be on adults making decisions on their behalf. Some of the young people who participated in Strand 2 were very knowledgeable about the prognosis for their conditions and they needed services matched to their holistic needs. The outcomes of the interviews indicated that planning transition from paediatric to adult services has become a vital component in holistic care and demands that service provision is expanded and improved. For example, the findings from parental reports from The Black Country and West Mercia NHS Clusters indicated that there were particular anxieties about the eventual transfer from paediatric to adult services with the feeling that adult services are even less likely to meet their needs.

An increasing number of children and young people with life-threatening and life-limiting conditions are able to enjoy their entitlement to education. Many mainstream schools see themselves as inclusive, though not all have the experience, skills and resources to make effective provision. The Disability Discrimination Act 2001 calls for schools to make reasonable adjustments to prevent pupils with special needs (including a child with complex health needs) from being at a disadvantage and to plan to improve their access to the curriculum. Denying a child a place in a mainstream school may be discriminatory, but in the West Midland Region, mainstream schools may not have the capacity to provide the differentiated curriculum which is matched to the needs of an individual child. Strand 2 findings endorse the findings of Brown's recent research (2012) which indicates that although the government may endorse the right of all children and young people to attend a mainstream school, parents and carers often opt to send their child to a special school because they consider that the education that their child receives will be better matched to their needs. Special schools in West Mercia NHS Cluster were highlighted as providing particularly good holistic educational care. Furthermore, although in general it was reported that schools provided a balanced, differentiated and broad curriculum, some staff were not confident about supporting pupils with special needs, especially those with complex health needs.

In 2010 the West Midlands region had the lowest rate of employment for the 12 regions of the countries in the UK, and in 2009, median earnings in the West Midlands were lower than the UK as a whole. Furthermore the region has areas of high deprivation (Office for National Statistics, ONS, 2011). Having a sick child inevitably leads to extra costs and it is far more difficult for parents with a life-threatened or life-limited child to manage on the same income as families where people enjoy good health. Many families with a sick child have fewer opportunities to earn, and despite the range of benefits and other help available, they may struggle to find their way through the maze and claim their full financial entitlements. Although the benefit system recognises the potential implications of financial hardship for families, there is evidence from Strand 2 that the benefits currently available do not always match the additional costs incurred. Furthermore, life expectancy for medical conditions has improved dramatically (Department of Health, 2008). Therefore, some families are caring for many years, and the cumulative effect of exhaustion can seriously damage parents' health. Hewitt-Taylor's (2009) small-scale study of parents with a child with complex medical needs concluded that there remained significant inequity of provision and access to recreational activities, social opportunities and transport. Strand 2 findings would seem to reiterate Hewitt-Taylors's (2009) research outcomes.

Respite care or short break care has been recognised as helping to reduce parental stress and fatigue (Craig, 2006). Eaton's 2008 survey provided evidence that families required more overt referral systems and regular assessment of need. Strand 2 work endorsed these findings and additionally highlighted that parents need to know that respite care service provision is sufficiently flexible to be able to respond to a family's needs in a crisis situation. Indeed, access to emergency care and routine short-break care is given equal priority by parents. In relation to respite care, parents reported in the areas of Birmingham, Black Country and Arden NHS Clusters that they were aware of the constraints but had received overnight help, a number of volunteers and generally good complex nursing package whereas parents in other areas felt that this was more difficult to obtain. Some regions such as West Mercia NHS Clusters were reported as having little respite on offer.

Since 2003, national and local governments in England have embarked on reforms aimed at improving and integrating health, education and social services for children and their families. Effective partnership has been a core principle underpinning a number of key policies (Department for Education and Skills and Department of Health, 2007, Her Majesty's Treasury and Department of Education and Skills, 2007, Department of Health, 2008). Strand 2 findings highlighted that poor co-ordination of services had severe consequences for children, young people and families. Joined up working and identifying who is responsible overall for *'getting things done'* would thus seem a priority with this group given their growing needs.

6.3. Professional staff issues

Trust and confidence in a service delivering holistic care is one of the hallmarks of good service delivery and Strand 2 findings highlighted the importance of effective joined up working and collaboration between service providers. Despite financial constraints, many families are enjoying good medical and nursing care. Care in children's hospices is deemed to be excellent and in some cases, to exceed all expectations. However, where there is cause for complaint, about the care provided, some families feel that if they voice their concern, the care they receive may be compromised, and in some cases, that their child's survival may be jeopardised.   For example, a lack of communication between Occupational Therapists and specialist consultants and families were cited in Gloucestershire NHS Cluster with occasions when phone calls and emails from parents were reported as not being responded to. Taylor (2000) identified the adverse impact that different ideologies and attitudes might have on families. In Arden NHS Cluster, it was reported that information technology systems of professional staff needed modernisation to ensure communication was effective. It has also revealed the pivotal role which professionals play when they work both within teams and across teams. The success of the Community Nursing Teams in Arden and Staffordshire NHS Clusters appears to be largely dependent on co-operative partnership between all the organisations concerned and the lead individuals in posts, so that families are able to receive a seamless service.

6.4. The need for effective communication

The need for effective communication that prepares parents and carers for the inevitability that their child will die prematurely has been identified as a core issue (ACT, 2004, Avis and Reardon, 2008, Midson and Carter, 2010). *Figure 9* draws together the overall issues from Strand 2 data.

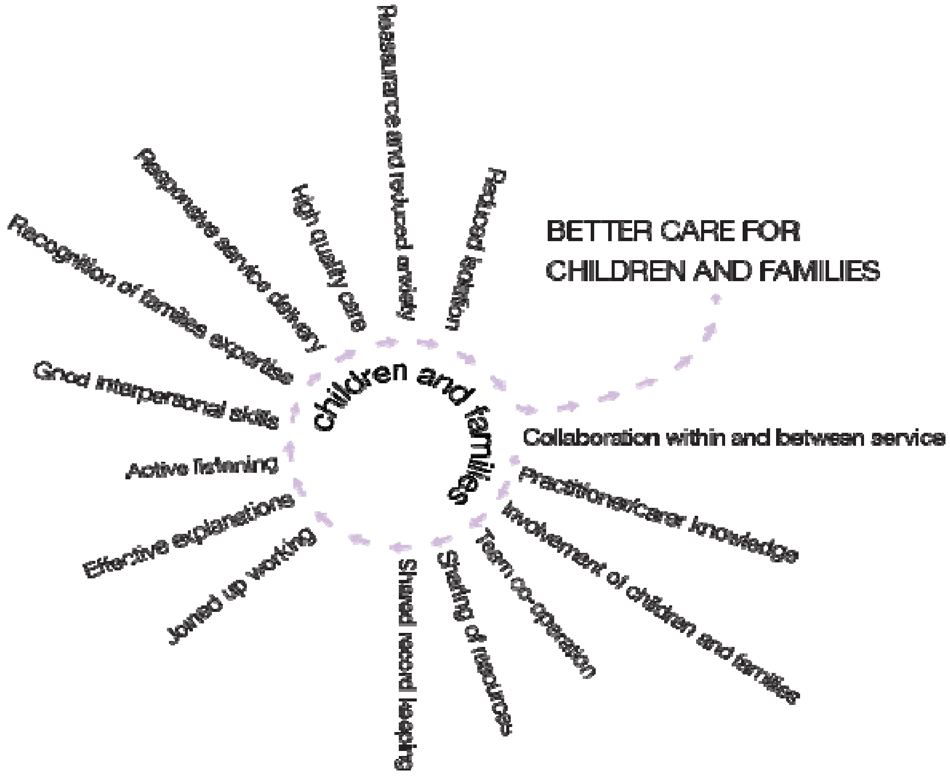


Figure 9. Better care for children and families through effective communication (Strand 2 findings)

The importance of good communication within and between services has also been highlighted as a significant factor in how well families cope with a life-threatened or life-limited child (Down and Simons, 2006). Therefore, it is important to assess the service needs of children and their families both individually and as a unit. One of the significant findings of Strand 2 has been that families have named some of the components that they believe to be present when good communication takes place. Furthermore the interviews have also provided a rich picture of a number of the positive outcomes of good communication for families.

Knowing their child's illness has a label is helpful and empowers parents when they are given opportunities to tell their stories, but families need professionals to recognise the uniqueness of their child (DoH, 2008). There were occasions in Strand 2 when parents, especially mothers, expressed frustration at having to tell and re-tell their stories over and over again to professionals. Furthermore, young people were described by parents and carers as being excluded from these discussions and this was perceived to have a detrimental impact on young peoples' emotional well-being. Evidence from children and young people who participated in the research has challenged the assumption by some professionals that children and young people are unable to express their opinions. If communication is not limited to verbal dialogue, even pre-verbal young children and those who use augmented and assisted communication systems can express what they need and how they feel.

When people are in potentially vulnerable situations it is important that care is focused on their needs and what is important to them. Parents with a life-threatened or life-limited child often have to face questions and painful decisions. Professionals need skills, confidence and information to find out what each family wants and organisational structures which are sufficiently flexible to enable them to provide it. The importance of assessing children within the context of the family has been well documented (DoH, 2004, Kerr, 2001). The ACT Charter (2005) suggests that each child and young person should receive an individual Care Plan which is reviewed at regular intervals and that where possible the child and the family should be active participants in deciding on the Care Plan. Strand 2 findings wholeheartedly endorsed this. Families seldom mind if carers ask about what is important to them, as long as the subject is approached in a sensitive and respectful way. Indeed, interest may well be welcomed and enjoyed.

It is important that information is provided in accurate and accessible ways, particularly when English may be an additional language. The West Midlands has the highest non-White regional population outside London and Asian or Asian British makes up the biggest non-White proportion of the West Midlands population at 8.5% (ONS 2011). This compares with the national average of 6.1%. Randhawa et al (2003) and Katbamna's et al (2000) studies highlighted that there was limited research available that focused on the cultural needs of families. Strand 2 findings indicate that the needs of Asian and British Asian parents and children are unique to each family and that care should be tailor-made to each family member rather than making assumptions that all people from a given ethnic group conform to a stereotype.

A summary of key messages from the findings are outlined below in *Table 10;*

Family Perspectives

- Having trust and confidence in services being able to deliver high quality holistic care for the child and the whole family, is very important to families. For example, when families are able to access respite care it gives them opportunities to take time away from caring duties. Financial pressure and burden of care is high but balancing of a 'normal' family life was important.

Services

- Services need to be responsive to needs. Services are most under pressure regionally in terms of respite care and end of life care. Up to date information technology systems were needed in several regional areas. Paucity of age appropriate service provision and a lack of joined up services are a cause for concern.
- There is inequity in service provision and specialist equipment matched to individual children's and young people's needs, both locally and regionally. This was mapped in the full report. Some families have to travel considerable distances to access specialist services.

Professional Staff

- Parents/carers value staff that are professional, positive in attitude, competent, knowledgeable and dedicated. In particular Community Children's Nurses were highly regarded for their planning and delivering care packages and providing children and young people opportunities to take part in normal activities with their families and peers. The number of staff who are appropriately trained and experienced to provide care is a concern.

Communication

- Good communication and being listened to is valued by parents/carers but in all the regional areas studied, participants had to retell their 'stories' many times because collaboration within and between service providers is often fragmented.

Figure 10: A summary of the key findings from Stand 2 of The BIG Study.

6.5. Limitations

Understanding family needs is a pre-requisite to shaping family-centred and child and young people-centred care. (Brown, 2007, Coad and Houston, 2007). Strand 2 of The BIG Study sought to identify and to document met and unmet needs of families with a life-threatened/life-limited child/young person living in the West Midlands so that these could be disseminated to local and national agencies as evidence for service improvement and development. Whilst methods and analysis are sound, limitations need consideration.

Firstly, the complexities of interviewing children, young people and families who are in potentially vulnerable situations are well known (Down and Simons, 2006, Graham et al, 2006). We have argued that the research methods were appropriate. Appreciative inquiry was used to collect data by Strand 2 in order to discover 'what worked well' for families and 'why it worked well'. Appreciative Inquiry has previously been used within a variety of organisational structures including health and social care settings (Coad and Carter, 2009). Direct extracts from participant interviews were extracted verbatim with great sensitivity and have been used to illustrate themes.

Recruitment in Strand 2 was very good although there were a number of challenges in making initial contact with participants owing to the chaotic lifestyles they experience. Appointments were often cancelled at very short notice. Fifty one families participated in the study. More than two thirds of the participants (67%) were White British which matches estimated numbers for the West Midlands Region from The Office for National Statistics (2011). A further 11% of participants were British Asian and 7% other Asian. Notwithstanding, Strand 2 acknowledges a number of limitations.

- The paucity of published research makes it difficult to compare the findings of the BIG study with a well-established evidence-base for children's and young peoples' palliative care.
- Data from some of the five NHS West Midlands clusters and Gloucestershire is limited because of low recruitment in some areas and therefore data sets cannot be generalised.
- Some children and young people were unable to take part in the study because they were too young or they were only able to communicate using alternative and augmentative communication methods.
- Data validity is limited in respect of intergenerational and cross-cultural perspectives.

6.6. Recommendations

Strand 2 recommends the following:

In terms of family issues:

- Improved provision of professional, emotional support for children, young people with life threatened and life limiting conditions and families is strongly recommended, most especially at diagnosis.

- Families need to be supported as a whole, including support for siblings, bereavement support, and respite available to the whole family, to ensure that families are able to stay together and enjoy activities together.

- Families need to be fully informed of their rights to receive financial support with systems in place to assist with relevant paperwork, so not adding to the parent/carer workload.

- Appropriate support, both for their emotional and physical needs, is also needed for life threatened and life limited young people making the transition from paediatric to adult services in the community and in palliative care settings. This includes bereavement support for all family members from the time of the child's diagnosis to the end of life and beyond.

In terms of responsive services:

- Access for all children and young people to community children's nursing services and a continuing shift in services from hospital to home.
- Regional service provision needs improving for Physiotherapy in Staffordshire, Birmingham and Arden NHS Clusters. The availability of occupational therapy availability needs to be improved in Staffordshire and Arden NHS Clusters.
- Health, social care and education services need to be responsive to, and tailored to the child's or young person's needs throughout their illness trajectory. Care plans and packages need to be updated continuously in partnership and in line with assessing the child's, young person's and family needs.
- There needs to be greater equity and timely provision of specialist equipment in order to support the health, social and education care needs of the child/young person and family.
- The accessibility of 24 hour daily support for families needs reviewing.
- Information about available services needs to be communicated more explicitly and in a variety of formats in order to ensure parents can make fully informed choices.
- The development of forums, using up to date, responsive information technology systems that encourage the voice of the child, young person and families to be heard which informs service delivery. Services need to be increasingly user-led as well as user-centred.
- There needs to be a national commitment to providing short break/respite care to families caring for a child with complex medical needs.
- Families need more opportunities in the community which will support care throughout the trajectory of the child's or young person's illness. For example, young children requiring technological support and those needing end of life care.
- At a time of economic hardship, commissioners need to set realistic targets in order to maintain and develop the provision of service delivery. It is paramount that that any quality indicators developed or used for paediatric palliative care take into account the views of children and families. Measuring and demonstrating quality, within and between services, without their views can not fully take into account the experience from the user viewpoint.

In terms of professional staff:

- Specialised training for carers to meet the complex needs of children; training for doctors in communicating sensitive information and professional development for healthcare staff to meet the emotional and physical needs of parents and children. Given the findings relating to experience of diagnosis and family trust thereafter this needs to be a priority.

- Improve staff skills which enable them to be responsive to the augmented and alternative communication needs of children and young people with complex needs, who may be technology dependent.

In terms of effective communication:

- Improve partnership and collaboration across services to take into account the holistic needs of families from diagnosis onwards.

- Strengthening systems which will improve communication across health, social and education in order to achieve a fully documented, integrated, comprehensive, multi-disciplinary and multi-agency service.

- Information Technology systems to support communication across services. Databases which can 'talk' across agencies and settings, facilitating the sharing of information and developments such as telemedicine to enhance family care and support to overcome existing geographical barriers and delay in access to support.

- There is a need for a national palliative care research project to determine resonance of findings here in the region of West Midlands and that any project needs user views embedded in to it focusing on meeting family needs in the community from an intergenerational, cross cultural perspective.

6.7. Conclusion

Using Appreciative Inquiry and Framework Analysis, Strand 2 has reported on in-depth interviews of met and unmet needs from the perspective of 59 participants, including children, young people with life threatening and life limiting conditions and their families. The expected outcome is to influence and inform health, education and social care services and policymakers which is at a critical stage in its evolution in this field. Seeking the views of children and young people with life threatening and life limiting conditions and their families was very important in the BIG study, in order that their needs are reflected in service provision. The challenge as services move forward is not only to consider care from a one dimensional angle but instead joined up and seamless across health, social care and education services. Moreover, this Strand has also shown how important user voices are. Consequently, any service planning and delivery must be in partnership with the children and young people with life threatening and life limiting conditions and their families. Only with listening and responding to their voices can provision of services continue to grow in order that the unique needs of everyone concerned are met.

References

Association for Children with Life-threatening or Terminal Conditions and their Families (ACT) and Royal College of Paediatrics and Child Health) (RCPCH) (2003) A Guide to the Development of Children's Palliative Care Services. 2nd Edition. Bristol, ACT and RCPCH.

Association for Children's Palliative Care (ACT) (2004) The Transition Care Pathway. A Framework for the Development of Integrated Multi-Agency Care Pathways for Young People with Life-threatening and Life-limiting Conditions. Bristol, ACT.

Association for Children's Palliative Care (ACT) (2005) The ACT Charter. Bristol, ACT.
Association for Children's Palliative Care (ACT) (2007) The Transition Care Pathway. A Framework for the Development of Integrated Care Pathways for Young People with Life-threatening and Life-limiting Conditions. Bristol, ACT.

Avis M., Reardon, R. (2008) Understanding the views of parents of children with special needs about the nursing care their child receives when in hospital: a qualitative study. *Journal of Child Health Care,* 12 (1) 7-17.

Bachmann, M., O'Brien, M., Husbands, C., Shreeve, A., Jones, N. and Watson, J. (2009). Integrating children's services in England: a national evaluation of children's trusts. *Child Care Health Development,* (35):2, 257-65.

Brown, E. and Arens, G. (2005) Siblings Project – Listening to Brothers and Sisters. Birmingham, Acorns Children's Hospice Trust.

Brown, E. (2007) Supporting the Child and the Family in Paediatric Palliative Care. London, Jessica Kingsley.

Brown, E. and White, K. (2008) The transition from paediatric palliative care to adult services. In: Brown, E., Gatrad, R. and Sheikh, A. (2008) Palliative Care for South Asians: Muslims, Hindus and Sikhs. London, Quay Books 131- 45.

Brown, E. (2012) The Education of the Life-limited Child. In Goldman A., Hain, R. and Liben S. (eds.) The Oxford Textbook of Palliative Care for Children. Oxford, Oxford University Press.

Brykczynska,G. and Simons, J. (2011) Ethical and Philosophical Aspects of Nursing Children and Young People. London, Wiley Blackwell.

Carter, B., Coad, J. and Goodenough, T. (2009) *Community Children's Nursing in England: An appreciative review of CCNs*. Department of Health Report. NHS at Home: Community Children's Nursing Services. http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_1248-98.

Coad, J. and Houston R. (2007) Voices of Children and Young People. Involving children and young people in the decision-making processes of healthcare services. A review of the literature. London, Action for Sick Children.

Coad, J., Houston, R., Widdas, D. and Brown, E (2012) Giving children a voice on making decisions on care and services: how involved are children and young people receiving palliative or end of life care? (In press). *International Journal of Palliative Nursing*.

Coad J. and Shaw, K. (2008) Is children's choice in health care rhetoric or reality? A scoping review. *Journal of Advanced Nursing,* 64(4):318-327.

Cooperrider, D. and Whitney, D. (1999) Appreciative Inquiry: a positive revolution in change. In *The Change Handbook: Group Methods for Shaping the Future*. Holman, P. & Devane, T. (eds.), San Francisco, Berrett-Koehler Publishers, pp. 245-261.

Craft, A. and Killen, S. (2007) Palliative care services for children and young people in England: an independent review for the secretary of state for health. London, Department of Health.

Craig, F. (2006) Adolescents and Young Adults. In: Goldman, A., Hain, R. and Liben, S. Oxford Textbook of Palliative Care for Children. London, Oxford University Press, 108-18.

Danvers, L., Freshwater, D., Cheater, F. and Wilson, A. (2003) Providing a seamless service for children with life-limiting illness – experiences and recommendations of professional staff at The Diana Princess of Wales Children's Community Service, *Journal of Clinical Nursing 12* (3): 351-9.

Department for Children, Schools and Families (2007) The Children Plan. London, Department for Children, Schools and Families.

Department for Education and Skills and Department of Health (2004) Every Child Matters: Change for Children. London, Department for Education and Skills and Department of Health.

Department of Health (2004) National Service Framework for Children, Young people and Maternity Services. London, Department of Health.

Department for Education and Skills (DfES) (2005) Professional Guidance for Children with Additional Needs. London, Department for Education and Skills.

Department of Health (2007) Making it Better for Children and Young People. London, Department of Health.

Department of Health (2008) Better Care: Better Lives – improving outcomes and experiences for children, young people and their families living with life-limiting and life-threatening conditions. London, Department of Health.

Doig, J., McLennan, J., and Urichk, L. (2009) 'Jumping Through Hoops': parents experiences with seeking respite care for children with special needs. *Child Health and Development* , 35(2): 234-42.

Doug, M., Adi, Y., Williams, J., Paul, M., Kelly, D., Petchey, R. and Carter, Y. (2012) Transition to adult services for children and young people with palliative care needs: a systematic review. *BMJ Supportive & Palliative Care,* 1 (2) 167-173.

Down, G. and Simons, J. (2006) 'Communication'. In Goldman, A., Hain, R. and Liben, S. (eds.) Oxford Textbook of Palliative Care for Children. London, Oxford University Press, 28-41.

Eaton, N. (2008) 'I don't know how we coped before': a study of respite care for children in the home and hospice. *Journal of Clinical Nursing,* 17 *(23): 3196-204.*
Elston, S. (2003) Assessment of Children with life-limiting conditions or terminal conditions and their families. Bristol, Association for Children with Life-limiting conditions or Terminal Conditions and their Families (ACT).

Enskar, K. (2000) Important aspects of care and assistance for children with cancer. *Journal Pediatric Oncology Nursing ,*2000; 17(4):239-249.

Glendinning, G., Kirk, S., Guiffrida, A. and Lawton, D. (2001) Technology-dependent Children in the Community: definitions, numbers and costs. *Child Care, Health and Development,* 27 (4) 321-34.

Graham, R., Dussel, V. and Wolfe, J. (2006) 'Research'. In: Goldman, A., Hain, R. and Liben, S. (eds.) Oxford Textbook of Palliative Care for Children. Oxford: Oxford University Press p. 615.

Grinyer, A., Payne, S. and Barbarachild, Z. (2010) Issues of power control and choice in children's hospice respite care services: a qualitative study. *International Journal of Palliative Nursing* ,*16*(10) 505-10.

Heaton, J., Noyes, J., Sloper, P. and Shah, R. (2005) Families' experiences of caring for technology dependent children: a temporal perspective. *Health and Social Care*, 13(5) 441-50.

Hewitt-Taylor, J. (2009) Children with complex continuing health needs and access to facilities. *Nursing Standard,* 23 (31) 35-41.

Hewitt-Taylor, J. (2008) Children who have complex health needs: parents experiences of their child's education. *Paediatric Nursing,* 20(8) 20-3.

Hsiao, J., Evan, E. and Zeltzer, L. (2007) Parent and child perspectives on physician communication in pediatric palliative care. *Palliative and Supportive Care,* 5(4):355-365.
HM Treasury and Department for Education and Skills (2007) Aiming High for Disabled Children. London, HM Treasury and Department for Education and Skills.

Jassal, S. and Sims, J. (2006) Working as a Team. In: Goldman, A., Hain, R. and Liben, S. (eds.) Oxford, Oxford Textbook of Palliative Care for Children. Oxford University Press. p. 518.

Katbamna, S., Padma, B. and Parker, G. (2000) Perceptions of disability and care-giving relationships in South Asian communities. In: Ahmad, W. (ed.) Ethnicity, Disability and Chronic Illness. Buckingham, Open University Press.

Kelly, A., Ktatz, B. and Bielski, M. (2002) Implementing transitions for youth with complex chronic conditions using the medical home model. *Pediatrics, 110: Supplement* 3 1304-6.

Kerr, G. (2001) Assessing the needs of learning disabled young people with additional disabilities: implications for planning adult services. *Journal of Learning Disabilities.,* 5(2) 157-174.

Kirk, S. (2001) Negotiating lay and professional roles in the care of children with complex care needs. *Journal of Advanced Nursing,* 34 (5) 593-602.
Knapp, C. and Contro, N. (2009) Family support services in pediatric palliative care. (2009) *American Journal of Hospital Palliative Care,* 26(6): 476-82.

Knapp, M., Perkins, M., Beecham, J., Dhanasiri, S. and Rustin, C. (2008) Transition pathways for young people with complex disabilities: exploring the economic consequences. *Childcare Health and Development,* 34(4) 512-20.

Lenton, S., Stallard, P., Lewis, M. and Mastroyannopoulou, K. (2001) The Effects of Childhood Non Malignant Life Threatening Illness on Parent Health and Family Functioning. *Journal of Child Psychology and Psychiatry,* (38) 7, 823-9.

Lester, P., Chesney M. and Cooke, M. (2002) Diagnostic disclosure to HIV-Infected children: How parents decide when and what to tell. *Clinical Child Psychology and Psychiatry*, 7(1):85–99.

Lyon, M., Garvie. P., McCarter, R., Briggs, L., He, J. and D'Angelo L. (2009) Who will speak for me? Improving end-of-life decision-making for adolescents with HIV and their families. *Pediatrics,* 123(2):199-206.

Midson, R. and Carter, B. (2010) Addressing end of life care issues in a tertiary treatment centre: lessons learned from surveying parents' experiences. *Journal of Child Health Care,* 14 (1) 52-66.

Ministry of Health (1959) The Welfare of Children in Hospital (The Platt Report). London, HMSO.

Noyes, J. (2006) Resources use and service costs for ventilator-dependent children and young people in the UK. *Health and Social Care in the Community,* 14(5) 508-22.

Office for National Statistics (2011) Census 2011. London, Office for National Statistics.

Radriguez, A., and King, N. (2009) The lived experience of parenting a child with a life-limiting condition: a focus on the mental health realm. *Palliative Support Care,* 7 (1) 7-12.

Randhawa, G., Owens, A., Fitches, R. and Khan, Z. (2003) Communication in the Development of Culturally Competent Palliative Care Services in the UK: a case study. *International Journal of Palliative Nursin, 9*(1): 24-31.

Ranmal, R., Prictor,M. and Scott, J. (2009) Interventions for improving communication with children and adolescents about their cancer (Review). *Cochrane Database of Systematic Review* , Issue 4.

Simons, J. (2002) Practical issues in the care of the dying child. In: Goldman, A. (ed.) Care of the Dying Child. (2nd edition). Oxford, Oxford University Press.

Sloper, T. and Beresford, B. (2006) Families and Disabled Children: social and economic needs are high but remain largely unmet. *British Medical Journal,* 333(4) 9289-9.

Smith, J. and Firth, J. (2011). Qualitative data analysis: the framework approach. *Nurse Researcher*, 18 (2), 52-62.

Swallow, V. and Jacoby, A. (2001) Mothers' evolving relationships with doctors and nurses during the chronic childhood illness trajectory*. Journal of Advanced Nursing,* 36(6) 755-64.

Taylor, C. (2000) The Partnership Myth? Examining Current Options. *Journal of Child Health Care*, 2(2) 72-75.
The Department of Health, Social Services and Public Safety (DHSSPS) (2003) Breaking Bad News: Regional Guidelines. Belfast, DHSSPS.

Tuchman, L., Slap, G. and Britto, M. (2008) Transition to adult care: experiences and expectations of adolescents with chronic illness. *Child Health Care and Development,* 34(5): 557-63.

UNESCO (1991) United Nations Convention on the Rights of the Child. Geneva, UNESCO.

While A., Cockett, A., and Lewis, S. (2004) Children and young people requiring home assisted ventilation in the South of England: incidence, receipt of care support and components of the care package. *Children and Society,* 18(3) 207-17.

Zwaanswijk, M., Tates, K., Van, D., Hoogerbrugge, P., Kamps, W.. and Bensing, J.. (2007) Young patients', parents', and survivors' communication preferences in paediatric oncology: Results of online focus groups. BMC pediatrics 2007; 7 , 2007. Article Number: 35. Date of Publication: 09 Nov.:35

Zwaanswijk, M., Tates, K., Van, D., Hoogerbrugge, P., Kamps, W., and Beishuizen, A. (2011) Communicating with child patients in pediatric oncology consultations: A vignette study on child patients', parents', and survivors' communication preferences. *Psycho-Oncology*, 20(3):269-277.

Strand 3 Report:
An evaluation of professional networks, co-ordination, co-operation and collaboration in the West Midlands Paediatric Palliative Care Network

By the Centre for Nursing and Healthcare Research at the University of Greenwich

Professor Elizabeth West
Val Chandler
Anna Wik
Helen Magee
Dr David Baron

Contents

Strand 3	page
1. Introduction	137
1.1 Background	137
1.2 Scope	137
1.3 Report	137
2. Context	139
2.1 Children’s Palliative Care and health policy	139
2.2 Clinical Networks	140
2.3 Concepts of social network analysis	141
3. The Big Study strand 3	142
3.1 Aims of strand 3	142
3.2 Research design and methods	142
3.3 Results of the electronic questionnaire	143
3.4 Results of the Social Network	153
3.5 Summary and Conclusions	157
References	159

Table of Figures

Figure: 1 Timeline
Figure: 2 Duration of membership
Figure: 3 Meeting attendance
Figure: 4 Stay in touch with the network
Figure: 5 Improving practice
Figure: 6 Change in practice
Figure: 7 Access to resources
Figure: 8 Career related opportunities
Figure: 9 Professional relationships
Figure: 10 Social relationships
Figure: 11 Increased influence
Figure: 12 Influence over groups
Figure: 13 Reasons for influence
Figure: 14 Membership benefits
Figure: 15 Representation of service users
Figure: 16 Benefits for service users
Figure: 17 Membership constraints
Figure: 18 Negative effects
Figure: 19 Sense of contribution
Figure: 20 Support from employer
Figure: 21 Membership in other networks
Figure: 22 People not in the network
Figure: 23 Organisations not in the network
Figure: 24 Professional background
Figure: 25 Organisations represented
Figure: 26 "Is this person known to you"
Figure: 27 Distribution of degree in figure 26
Figure: 28 " Would you go to this person for advice"
Figure: 29 Distribution of degree in figure 28
Figure: 30 " Does this person occupy a leadership role"
Figure: 31 Distribution of degree in figure 30
Figure: 32 "Do you see this person as influential outside the network?"
Figure: 33 Distribution of degree in figure 32

1 Introduction

This is a report on Strand 3 of the Big Study, which studied the West Midlands Paediatric Palliative Care Network. The Big Study was funded by The Big Lottery Fund and Strand 3 of the Big Study was researched by the Centre for Nursing and Healthcare Research in the School of Health and Social Care at the University of Greenwich.

1.1 Background

The West Midlands Paediatric Palliative Care Network began as an interest group which started in the year 2000, with 6 to 10 members and grew. At one stage it was allied to the Birmingham Cancer Network and funded by the NHS Strategic Health Authority and at this stage it became more representative of services and West Midlands geography. It has existed in its current format, as a voluntary clinical network to promote paediatric palliative care and share best practice since 2009.

The membership is wide and inclusive which means 30 to 40 people may attend the meetings which are held on a bimonthly basis and are hosted and supported charitably. Subgroups are now used to manage work in specific areas e.g. transition or clinical standards. There are links to other related networks with reciprocal membership and informal links to NHS commissioners who may seek advice.

1.2 Scope

This strand of the Big Study focused on the West Midlands Paediatric Palliative Care Network. The geographical area of the West Midlands Paediatric Palliative Care Network includes Birmingham, Coventry, The Black Country, Herefordshire, Shropshire, Solihull, Staffordshire, Stoke-on-Trent, Telford and Wrekin, Warwickshire and Worcestershire.

All members of the WMPCCN and the organisations they represent were included in the study. Both NHS and non-NHS organisations offering clinical services to any children requiring palliative care were represented. Excluded from this study was the detailed examination of any of the other networks, e.g. children's speciality networks or networks covering smaller geographical areas, to which members belonged.

1.3 Report

This report will present the results of an analysis of the responses to an online questionnaire and Social Network data from semi structured telephone interviews. This data was collected during the period February to June 2012.

The approach included analysing the online survey data in order to understand the benefits and constraints of the network for individual members and Social Network Analysis of data derived from telephone interviews to explore the flow of knowledge, communication and information within the network.

This report will consist of 3 different sections, with Section 1 focusing on children's palliative care policy, the development of clinical networks and social network analysis concepts. Section 2 will focus on the research design and methods. Section 3 presents the results of the study and the final section provides a summary and conclusions of the analysis.

2 Context

This section will give the reader a contextual background to children's palliative care, clinical networks and social network analysis. This will inform the reader's understanding of the results and conclusions of Strand 3.

2.1 Children's palliative care and health policy

Adult palliative care developed in the 1960s, but it was not until the 1980s that Great Ormond Street Hospital appointed the first consultant in paediatric palliative care, the world's first children's hospice was opened in Oxford, and the Association for Children's Palliative Care (ACT) was established.

Children with terminal conditions survive for longer now than in the past and the focus of care has increasingly centred on enabling a child to live as normal a life as possible. The Royal College of Paediatricians and Child Health (RCPCH) and the Association for Children's Palliative Care (ACT) define the palliative care of children and young people as:

"an active and total approach to care, embracing physical, emotional, social and spiritual elements. It focuses on enhancement of quality of life for the child and support for the family and includes the management of distressing symptoms, provision of respite and care through death and bereavement." (ACT/RCPCH Guide 2003).

In 2003 the government presented its Green Paper, *Every Child Matters,* in response to the inquiry into the death of Victoria Climbié. It proposed measures to improve children's care and enhance their life chances. In tune with the government's commitment to more integrated working, the subsequent Children's Act of 2004 based the changes around agencies and professionals working together. In the same year, the Department of Health published *The National Service Framework for Children, Young People and Maternity Services* setting out the standards necessary to achieve the government's policy aims. It states an expectation that children with complex health conditions receive co-ordinated, needs-led child- and family-centred services; that palliative care is available for those who need it and a range of flexible, sensitive services supports families in the event of the death of a child.

Children's palliative care has traditionally relied heavily on voluntary donations. In 2006, the Department of Health announced a three year funding package of £27 million, but concern over future sustainability led to an independent review in 2006/07. Kraft and Killen found a mixed picture in the provision of services. They reported a failure to understand that palliative care means more than just end of life care and a continuing lack of recognition of children's palliative care as a speciality in its own right. Despite policy commitments to partnership working, a recurrent theme was the lack of inter-agency co-ordination. The development of regional Paediatric Palliative Care Networks launched by the DH in 2006 was criticised as "patchy". The review made a number of recommendations, including the development of a national strategy, more sharing of best practice and the strengthening of regional networks.

The first ever national strategy for children's palliative care - *Better Care, Better Lives* - was published in 2008. It recognised that different agencies were involved and that the commissioning and delivery of health services should be planned in close partnership with education, children's services and the voluntary sector. It recommended the further development of children's palliative care networks as a focal point for regional strategic planning.

The new coalition government explained how their policies for the NHS could improve services for children and young people in *Achieving Equity and Excellence for Children* (2010). The emphasis was on shared decision-making, personalised care and choice. The Secretary of State for Health also commissioned an independent review of the funding of palliative care for adults and children in England. The review was asked to develop a per-patient funding mechanism for palliative care - the first time in the world that such a funding arrangement has been introduced for child palliative care services. The Government is currently supporting children's hospices with an annual revenue grant of £10m. In 2010/11, the Department of Health made a further non-recurrent grant of £30m available in support of local projects to improve paediatric palliative care services.

Thus recent years have seen a growing recognition of paediatric palliative care as a speciality in its own right and an increase in funding. In line with other aspects of children's services, there has been a shift in emphasis from care based on the needs of the service to more personalised and community-based child and family-centred care. A common theme of all the key policy developments has been the principle of partnership working. Children's palliative care networks are seen as an important way of facilitating this.

2.2 Clinical networks

In contrast to the hierarchical structure of the past and the market model of the 1980s, the New Labour government's reforms of the NHS were based upon collaboration and partnership working. This model was first developed within cancer services. The Calman-Hine Report (1995) had recommended that care for cancer patients should be based around managed networks of expertise and include the voluntary sector. These networks that moved beyond the informal professional groupings of the past gradually spread across a wide range of specialities.

The most widely accepted definition of a clinical network was first set out by the Scottish Office Department of Health in 1999: "Linked groups of professionals and organisations from primary, secondary and tertiary care, working in a co-ordinated manner, unconstrained by existing professional [and organisational] boundaries, to ensure equitable provision of high quality, clinically effective services." Networks increasingly came to be seen as a solution to the co-ordination of services for patients who require care from multi-disciplinary and multi-site teams. Partnership and network based working at all levels of care was a major policy theme in the NHS Plan (2000).

Within paediatric care, *Getting it right for children and young people* (2010) proposed that the development of successful networks was the best means to manage the complexity of the NHS and enable children to move seamlessly from one service to another. The most effective were considered to be the more formalised and managed networks as already found in newborn, palliative and surgical care. Recommendation 35 states: "There should be local networks through which services can share information electronically and protocols should be agreed by the networks to provide for this.”

As networks have developed and their impact upon the delivery of health care become more important, researchers have looked to social science theories to examine their strengths and weaknesses and possible implications for the NHS.

2.3 Concepts of social network analysis

What is Social Network Analysis? NHS Evidence provides us with this definition written by Shaunagh Robertson:

"Social network analysis is the mapping and measuring of relationships and flows between people, groups, organisations, computers or other information/ knowledge processing entities." (Valdis Krebs, 2002). “

Who goes on to say:

“In the context of knowledge management, social network analysis (SNA) enables relationships between people to be mapped in order to identity knowledge flows: who do people seek information and knowledge from? Who do they share their information and knowledge with?

In contrast to an organisation chart which shows formal relationships - who works where and who reports to whom, a social network analysis chart shows informal relationships - who knows who and who shares information and knowledge with who.

It therefore allows managers to visualise and understand the many relationships that can either facilitate or impede knowledge creation and sharing. Because these relationships are normally invisible, SNA is sometimes referred to as an 'organisational x-ray' - showing the real networks that operate underneath the surface organisational structure.”

So how does social network analysis help us to understand the West Midlands Paediatric Palliative Care Network? There follows a list from NHS Evidence that describes some of the uses of SNA.

- Identification of teams and individuals playing central roles - thought leaders, key knowledge brokers, experts, etc.
- identification of isolated teams or individuals
- detection of information bottlenecks
- spotting opportunities for knowledge flow improvements
- accelerating the flow of knowledge and information across functional and organisational boundaries
- improving the effectiveness of formal communication channels
- targeting opportunities where increased knowledge flow will have the most impact
- raising awareness of the importance of informal networks.

In examining the Social Network Analysis of West Midlands Paediatric Palliative Care Network we consider network characteristics and concepts that require some explanation. These key terms accompany the results in Section 3.4.

3 The BIG Study – Strand 3

This section explains the purpose and aims of Strand 3 of the Big Study and details how the study was conducted.

3.1 Aims of Strand 3

(a) To describe the pattern of formal relationships in the network focusing on the aspects of co-ordination, collaboration and co-operation.

(b) To identify the distinctive characteristics of the network that may explain areas of strength and weakness in operation.

(c) To collect information from the network members about their perceptions of the functioning of the network and any concerns that they might have about the quality of service that is being provided.

3.2 Research design and methods

Using systematic review methods we searched the academic and policy literature for recent work on children's palliative care services in the UK and the information was used to review clinical networks, policy on children's palliative care and social network theory.

Strand 3 of the Big Study was initially planned as a qualitative research project. The data would be collected through telephone interviews with the members of the West Midlands Paediatric Palliative Care Network. However, the project was subsequently modified into a two stage process, with an electronic questionnaire being the first stage and a telephone interview the second part of the data collection. Ethical approval was sought for the electronic survey and its accompanying documents (a participant information sheet for professionals explained the outline of the study and how it was to be conducted).

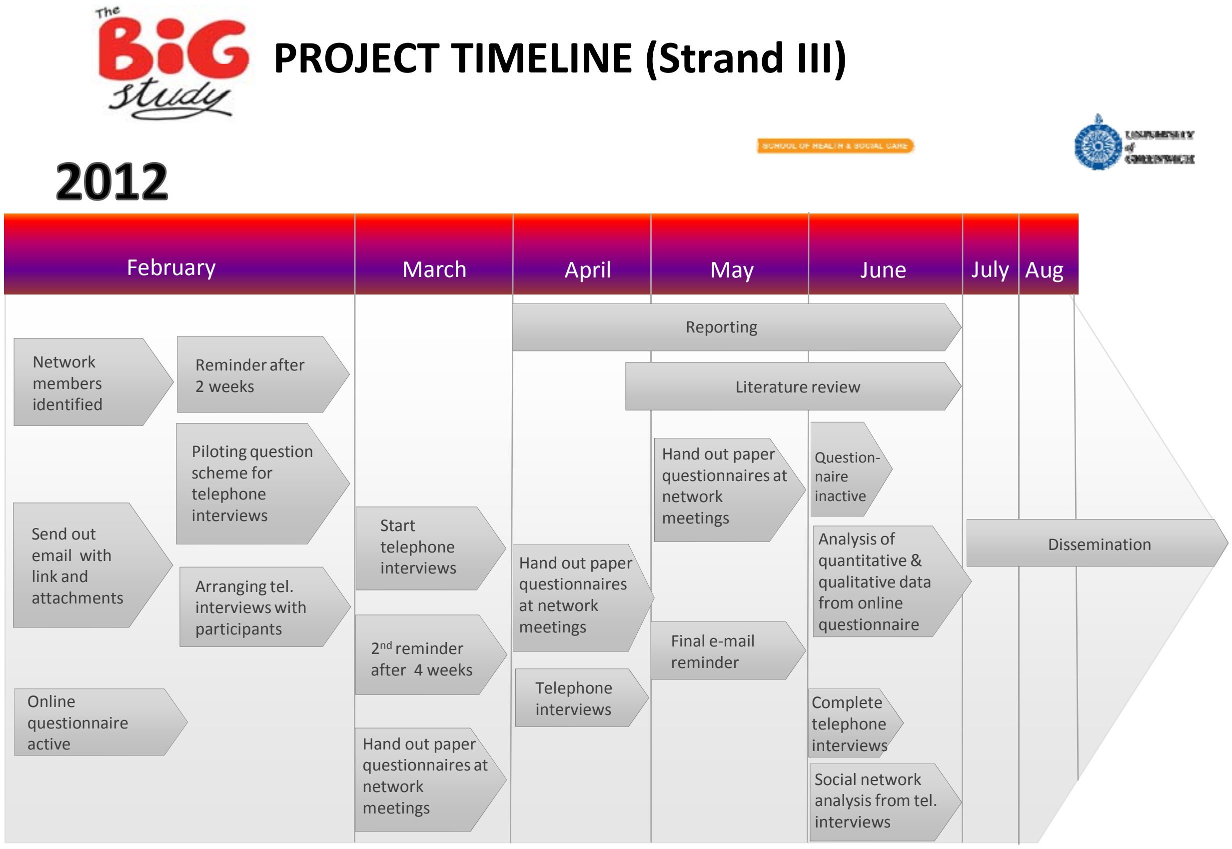
The questions for the electronic questionnaire was developed and designed by the research team of Strand 3. The platform for the electronic questionnaire was agreed to be Qualtrics. Qualtrics software is a web based service that enables users to design and implement their own online survey. The results from the online questionnaire were analysed using the statistical software package SPSS. Basic descriptive statistical analysis was conducted and a thematic framework was developed for the open ended questions in the survey.

The members of the West Midlands Paediatric Palliative Care Network were identified through the chair Claire Thomas and a list of member names and e-mail addresses was made available to the research team at the University of Greenwich. An invitation to participate in the survey was distributed by e-mail to network members, who clicked on the attached link to access the survey. The survey took approximately 15 minutes to complete. At the end of the questionnaire respondents were asked to take part in a semi-structured telephone interview of about 45-60 minutes. Any recipient who did not click the link in the e-mail was sent an electronic reminder after two weeks inviting them to take part in the survey. A second reminder was sent out after four weeks to those who had not yet taken the survey. A paper version of the survey was also handed out at network meetings.

For those respondents who indicated their interest in the telephone interview, a time convenient to both the interviewer and the interviewee was immediately set up. The telephone interviews varied in length, from 50 minutes to 90 minutes. Before the interview participants were sent a list of all the network members as well as the organisations they represented. The interviewer asked the participant questions about whom she/he knew in the network and of what nature that relationship was. Questions about leadership and influence of the members were also asked. The data collected was analysed by David Barron using the Social Network Analysis software tool *R*.

The results from both stages were then collated and reported in this document including a discussion of the findings. The project timeline of Strand 3 is illustrated below.

Figure 1. Timeline



3.3 Results of the electronic questionnaire

28 members from the West Midlands Paediatric Palliative Care Network decided to take part in the study by responding to the electronic survey on Qualtrics. There were also 5 responses to

the paper survey handed out at network meetings. This gave a response rate of 42 %. Graphs displaying the survey results are depicted below.

Membership and attendance

The majority of respondents (4 out of 5) had been members of the Network for 1 to 6 years. The longest serving members had belonged to the Network for 12 years. Only 6 % of respondents had joined the network in the last year (see figure 2).

Almost 7 out of 10 members said they attended most or every meeting and where they couldn't attend they stayed in touch by email or face to face contact (see figures 3 and 4).

Figure 2. Duration of membership

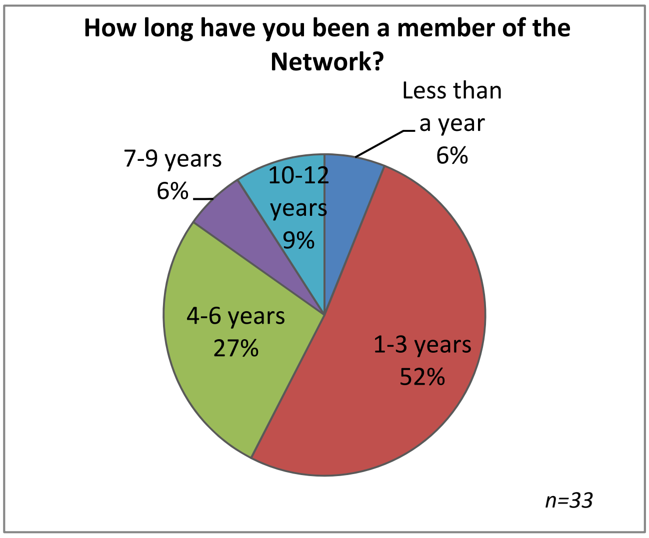
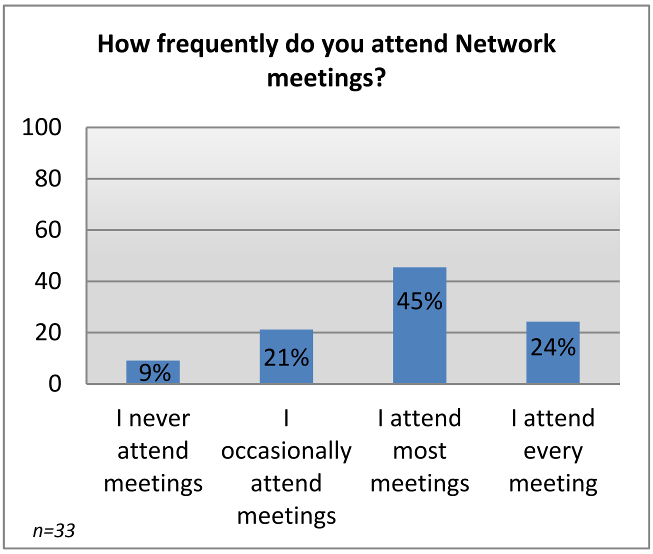


Figure 3. Meeting attendance



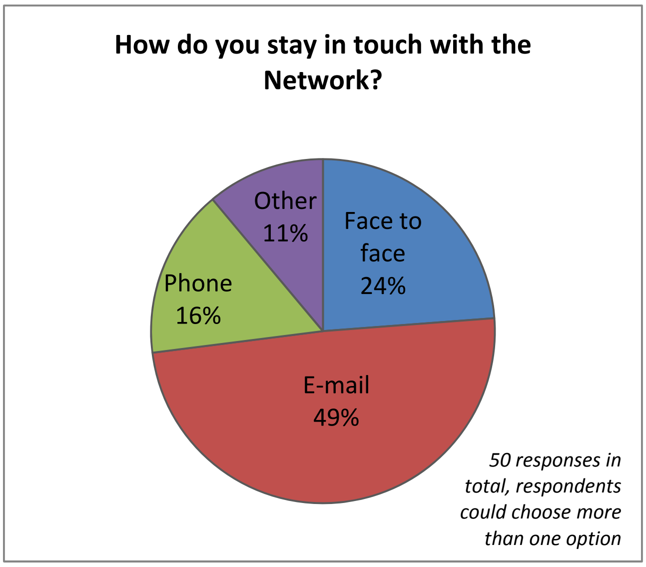


Figure 4. Staying in touch with the network

Clinical Practice

91% of respondents said they had acquired new ideas, information or evidence about paediatric palliative care from the Network, whilst 74% said that their practice had changed as a result of belonging to the Network.

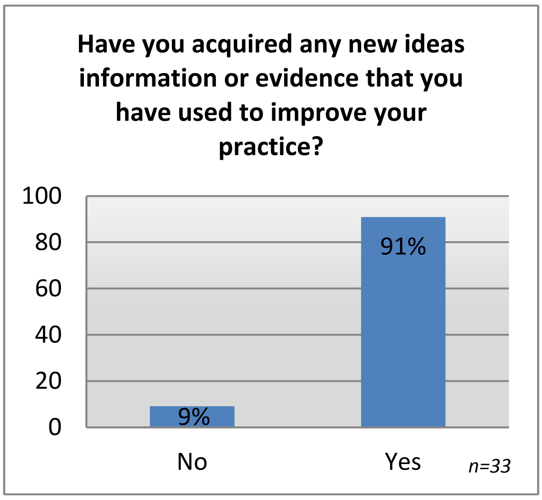


Figure 5. Improving practice

25 respondents commented on the acquisition of new ideas, 9 mentioned the toolkit and 2 mentioned the advanced care plan. Improved knowledge was cited by a number of respondents:

"I am a GP so have gained knowledge of the network, resources and management of children with life limiting illness" and

"bringing information directly from practising clinicians into commissioning" although one respondent said "more reinforcement of practice as opposed to brand new ideas"

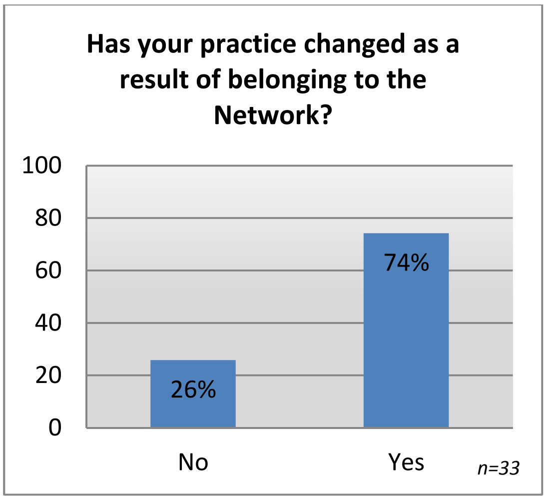


Figure 6. Change in practice

19 respondents commented on changes in practice, 3 mentioned the Toolkit and 6 mentioned advance care plans as having an impact on practice. Examples provided by respondents also included:

"We have a practice register of children with life limiting illness and I have had discussions with parents about the role of the GP and access to our services."

Another respondent said they were "more aware of how our particular hospice fits into the wider West Midlands area which will definitely enhance collaborative working with the statutory sector and voluntary sector colleagues".

Resources

70% of respondents said that since joining the Network they had had access to information about funds that they might not otherwise have heard about, and 67% had heard about jobs or courses through the Network.

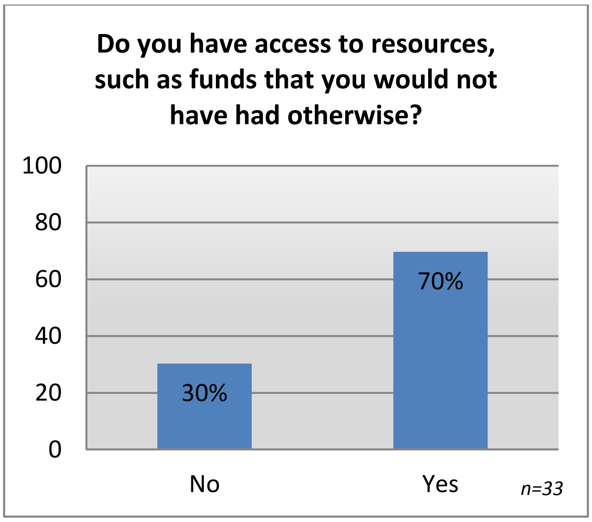


Figure 7. Access to resources

20 respondents commented positively on access to resources and 8 mentioned the £30 million programme. The strength of the network was described: "the joint bid by the network for palliative care monies for regional training would not have been available to a single PCT"

One respondent pointed out that there were "no funds within the network itself", just "access to information and contacts" and support for bids to the Department of

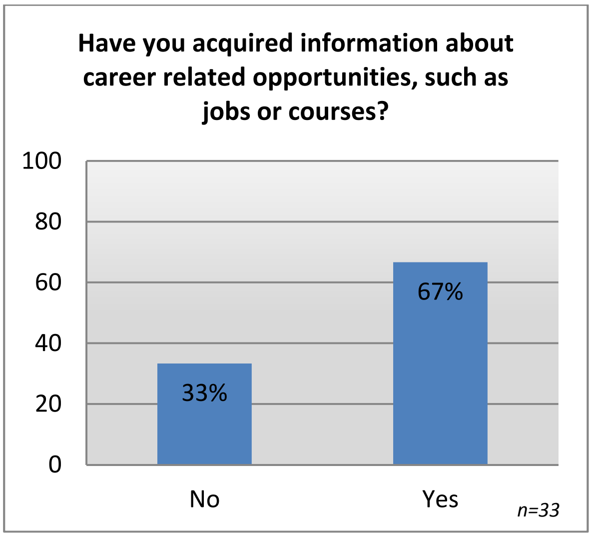


Figure 8. Career related opportunities

17 respondents made comments about career related opportunities, 11 about courses, many very positively about communications courses. E-learning is also mentioned, specifically a web based palliative care course.

Professional and social relationships

94% of respondents said that they had developed new professional relationships through the West Midlands Paediatric Palliative Care Network whilst less than 20% had made new social relationships.

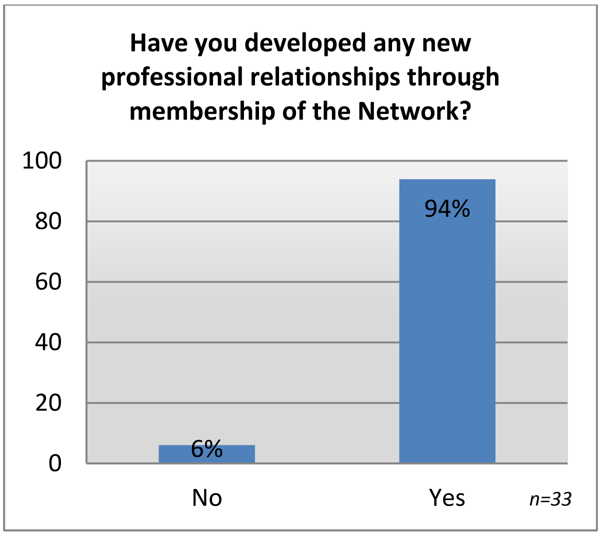


Figure 9. Professional relationships

23 respondents commented on professional relationships, 6 of the examples given mentioned team working and one respondent said that "meeting people face to face does foster better professional relationships". Closer working with community teams and the voluntary sector were mentioned, as well as "Lead nurses…… are coming together to look at provision of 24/7 care across the West Midlands. This group will continue to meet to share best practice, information and explore ways of developing paediatric palliative care nursing services"

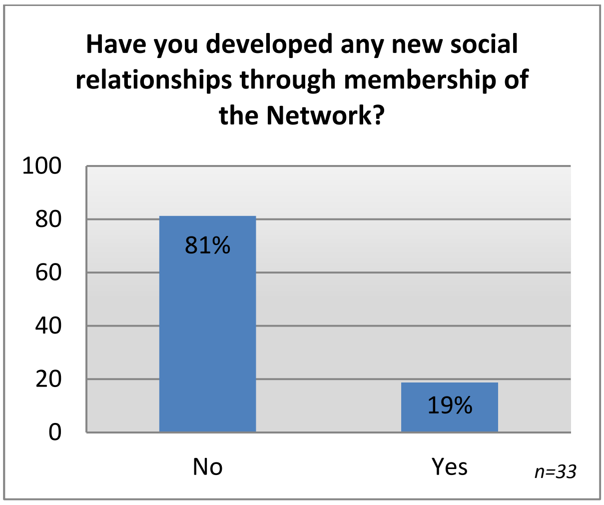


Figure 10. Social relationships

When asked about new social relationships, 4 respondents commented about renewing old relationships, network members who are friends and shared interests with old and new contacts, but 81% of members had not developed any new social relationships

The Network would appear to be predominately business orientated.

Influence

88% of respondents felt they had more influence as members of the Network, mainly on practitioners, commissioners and policy makers (83% of all parties named). 8 of 10 members felt that membership of the network added to their job role, contacts and reputation to enable them to exert influence.

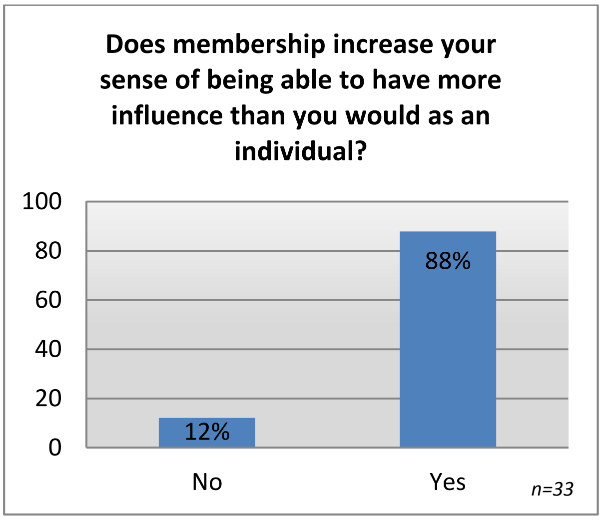


Figure 11. Increased influence

18 respondents gave examples of increased influence, 4 of these commented on securing funding, “joined up working” and effective lobbying of commissioners is also mentioned. Another respondent said “My Trust gives greater credence to quality standards that have the backing of the Network

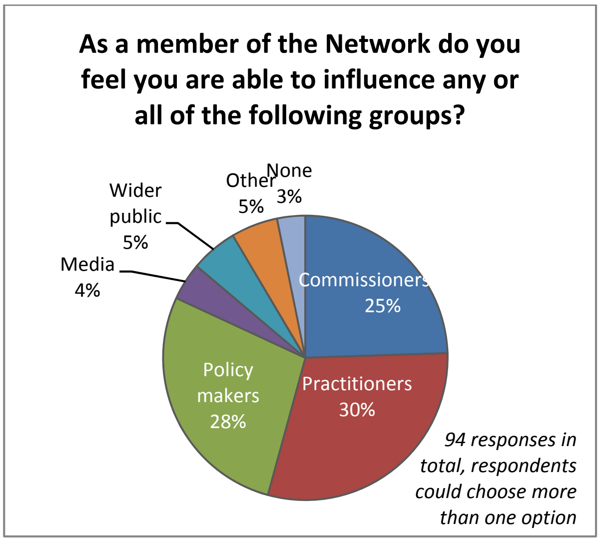


Figure 12. Influence over groups

Respondents could choose as many options as they wanted for this question our groups mentioned were strategic health authorities, Gp's, consortiums and mid sector groups.

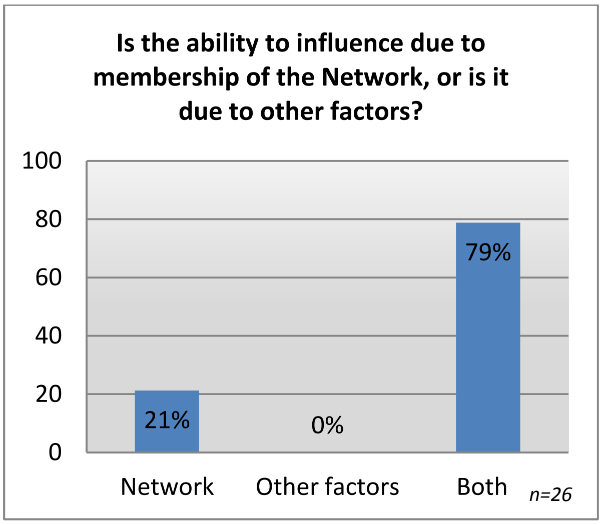


Figure 13. Reasons for influence

Where influence was attributed to both membership of the Network and job role, 11 examples were given, some naming specific forums which were influenced, another commenting that "it often carries more weight if one can demonstrate appropriate stakeholder collaboration; the network provides a good mechanism for demonstrating this". The value of a "collective voice" is mentioned, as is "credibility within my own area".

Benefits of Network Membership

59% of respondents said that there were other benefits to network membership with 97% saying that service users benefited from the network and 79% indicating that they felt more able to represent service users because of their membership.

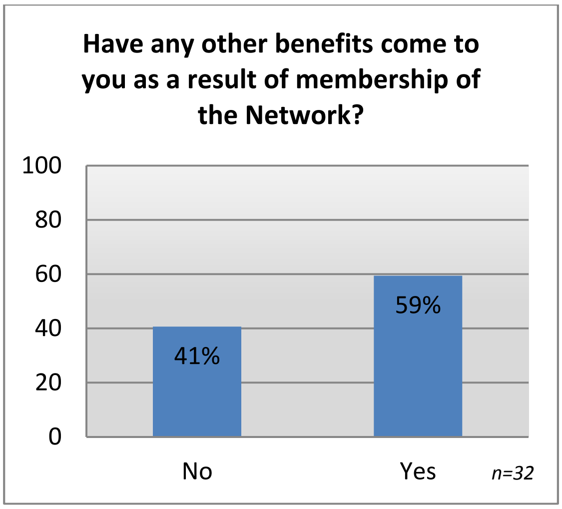


Figure 14. Membership benefits

15 respondents gave examples of other benefits, which included speed of response to calls for pilots, involvement in the Big Study, membership of subgroups, lessening of isolation and improved knowledge and access to information. One respondent said they were "confident to offer home as a place of death knowing there was a wealth of knowledge at the press of a button".

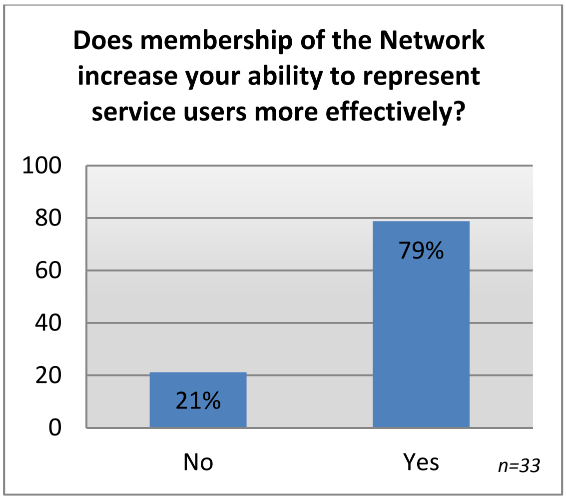


Figure 15. Representation of service users

79% of respondents felt that membership of the network increased their ability to represent service users. There were 17 comments, some mentioned specific examples of service user requests (e.g. for complementary therapies) and areas of work e.g. transition. One respondent felt local groups, which included parent representatives and made them "an integral part of decision making", were valuable and another comment was that "hopefully service users will be representing themselves at future (WMPPC) Network meetings

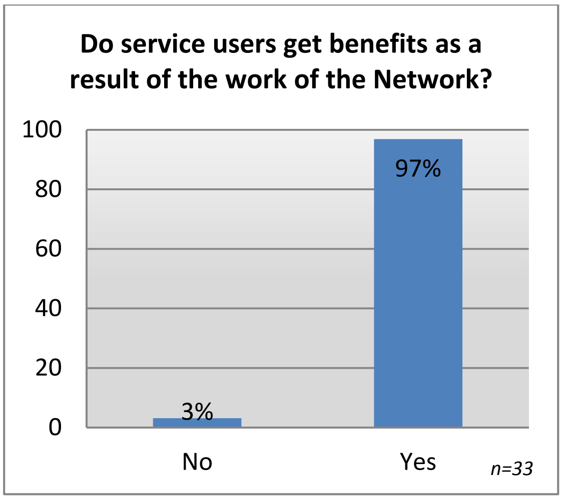


Figure 16. Benefit of service users

22 comments were made on benefits to service users. Indirect benefits were identified such as shared values and collaborative working which were perceived as enhancing consistency and standards through better informed, uniform care. The toolkit was identified as a direct benefit, as was more integration of services and partnership working.

Constraints and Contributions

The vast majority of respondents (97%) did not feel that the Network constrained them in any way and less than 20% identified any negative aspects to membership, with 8 out of 10 respondents feeling that they had been able to make a contribution to the Network. More than 80% of respondents said that their organisation supported their membership.

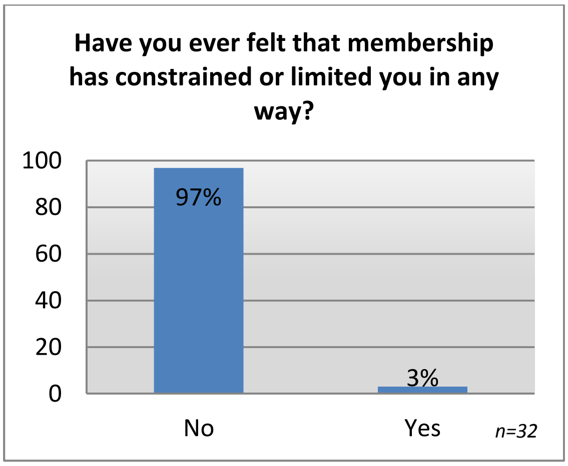


Figure 17. Membership constraints

When asked about whether membership of the network has provided any constraints, there was only one respondent who commented that “it can feel intensely political at times”.

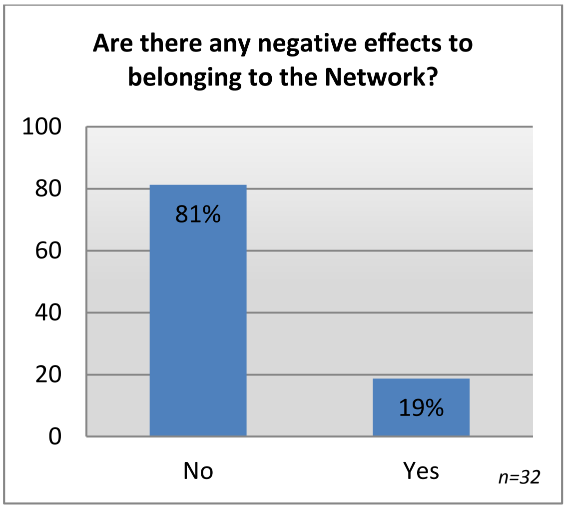


Figure 18. Negative effects

6 respondents commented on negative effects to belonging to the network, 4 of 6 respondents commented on negative effects to belonging to the network, 4 of these were on the time consuming nature of membership especially where membership was “additional to existing commitments”.

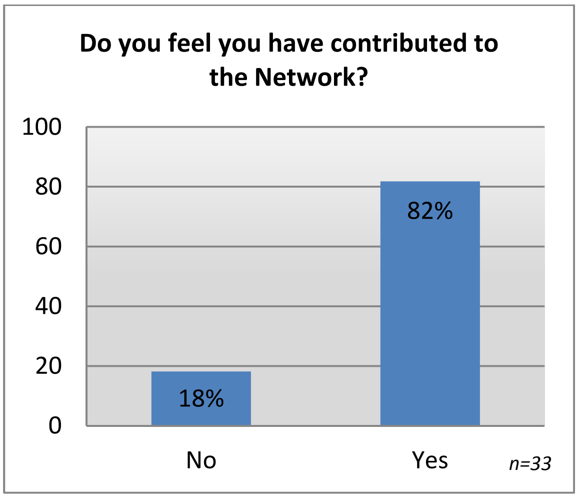


Figure 19. Sense of contribution

21 comments were made on contributing to the network. Membership of subgroups that drive change and contributing to bids for funds were most commonly mentioned. Discussions at meetings “where you feel valued” were also important

Figure 20. Support from employer

17 respondents were given time to attend meetings by their organisations and time and support to disseminate information and tools provided by the Network

Membership

Doctors and Nurses made up 88% of respondents with 6 out of 10 respondents employed by the NHS and 3 out of 10 by charities. Over 60% of these were also members of other related Networks. 45% of respondents identified “missing” people and organisations.

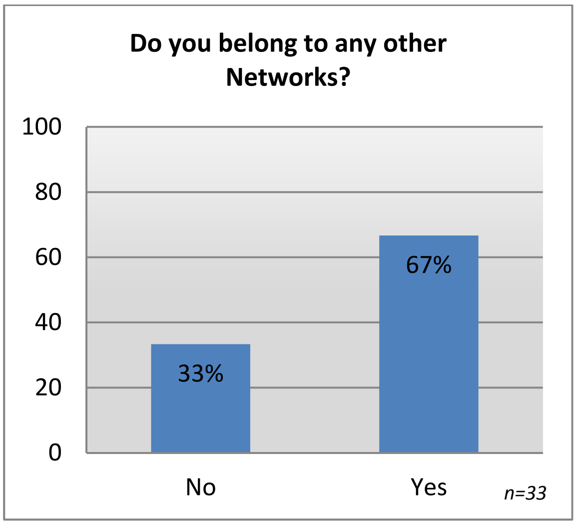


Figure 21. Membership in other networks

17 respondents commented on belonging to other networks. These included other children’s networks (e.g. safeguarding, children’s oncology, and children’s community nurse network), other West Midlands Networks(e.g. West Midlands paediatric network, WM long term ventilation network, WM children’s cancer network) and other locality networks both outside and inside the West Midlands (e.g. North Wales Paediatric palliative care network and Coventry and Warwickshire multiagency Strategic network for paediatric palliative care

The following question asked about the level of activity in the other networks respondents belonged to. There were 18 responses, 8 said they were most active in the WMPPCN. 2 were most active in other specialty networks e.g. neurology, oncology, one was most active in another locality – Wales, and the remainder were equally active in all the networks they belonged to.

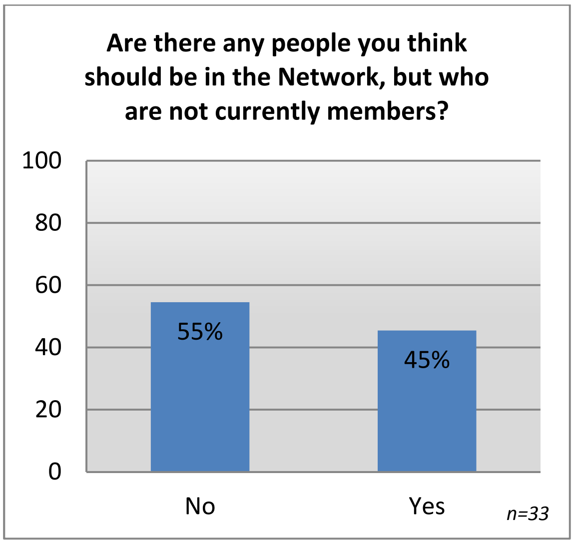


Figure 22. People not in the network

There were 13 responses, job roles identified that should be included were: Acute sector consultants and Allied Health Professionals including ambulance personnel, GP Commissioners and Service Users.

Services not represented included adult palliative care services, social care and disabled children's services. One respondent felt that more children's hospices should be represented.

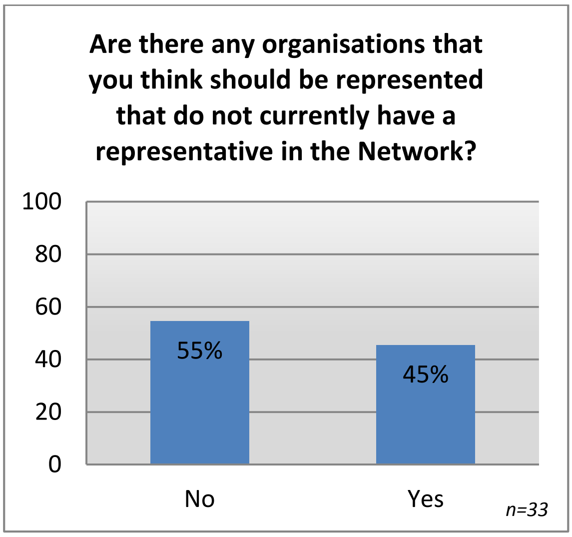
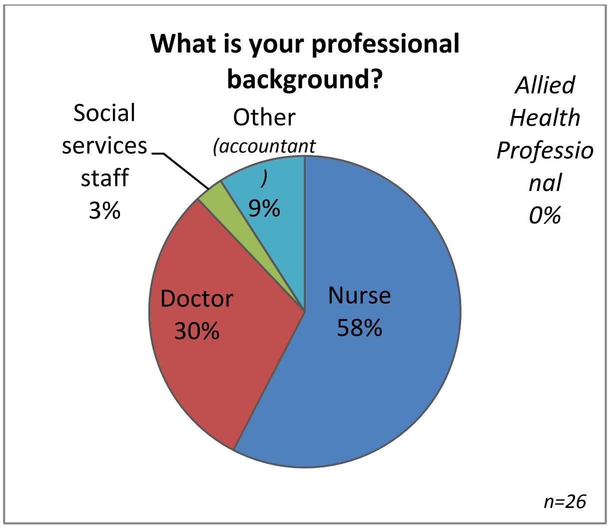


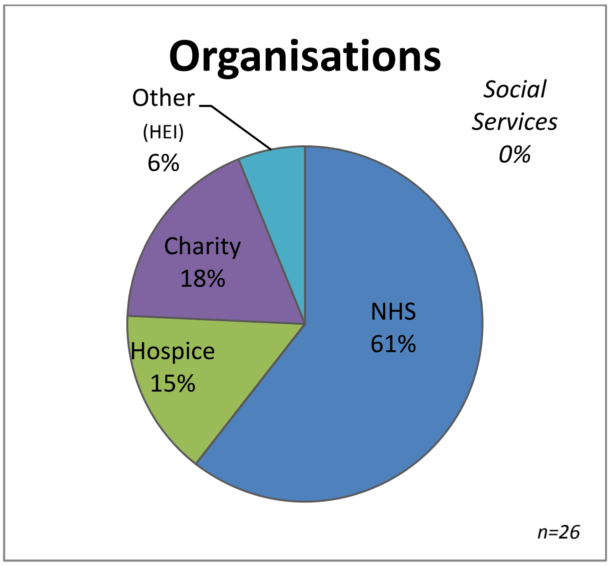
Figure 23. Organisations not in the network

There were 12 responses about organisations, including commissioners, education, ambulance, social care, local authority, special school education and Marie Curie.



In addition to stating their professional background, respondents were also asked to specify their job title. The most common job titles amongst respondents were nurses and consultant paediatricians. There were also quite a few Directors/Heads of Care, Clinical Team Leaders and Medical Officers. A few respondents were Finance and Managerial Staff.

Figure 24. Professional background



In addition to stating their professional background, respondents were also asked to specify their job title. The most common job titles amongst respondents were nurses and consultant paediatricians. There were also quite a few Directors/Heads of Care, Clinical Team Leaders and Medical Officers. A few respondents were Finance and Managerial Staff.

Figure 25. Organisations represented

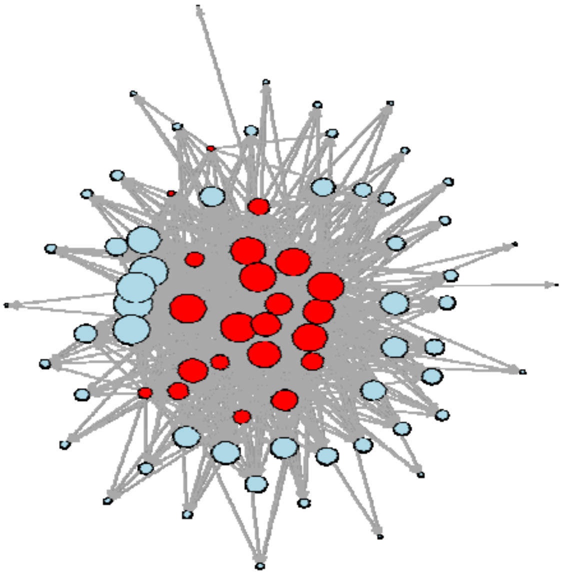
3.4 Results of the Social Network Analysis

We set out to obtain telephone interviews with all 78 members of the network but recruitment was slow. Liz West attended 2 network meetings and presented strand 3 at the National Networks Conference. Data collection opportunities remained open as late as possible and we slowly gathered network data from 22 people in total. Although it would have been better to have had more respondents, we have sufficient data to give a picture of the functioning of the network. In this report we will describe some of the main features of the network by examining the results derived from four questions: who do you know? Who would you go to for advice? Who provides leadership in the network and who is influential outside of the network.

Key Terms

- Node or actor: the unit of analysis, often a person or an organisation.
- Edge: a tie indicating a relationship usually represented as a line, sometimes with an arrow to indicate direction.
- Reciprocal ties: when the line has arrows at both ends.
- Multiplicity: the idea that nodes can be linked across different kinds of relationships, for example, someone can be a relative and a friend, or a friend and a colleague
- Strong and weak ties: measuring the strength of the relationship in some way, such as how well do you know this person or how much time do you spend with them.
- Density: a measure of the number of ties that exist, relative to the number that could exist. In a very dense network some of the ties might be described as "redundant" as they could be removed without great damage being done to the flow of information throughout the network
- Centrality: an actor is defined as central if they are on the pathway between many other actors and there are few other actors in the network also working as intermediaries. Highly central actors can be described as "brokers" or "bridges".
- Centralisation: highly centralised networks are like traditional hierarchies and in the extreme all ties would go to one actor. In a decentralised structure there are few if any actors who can be seen as focal points.

“Is this person known to you?”



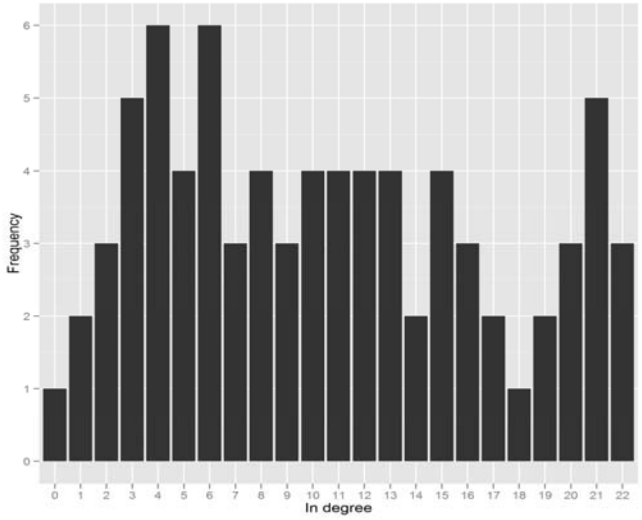
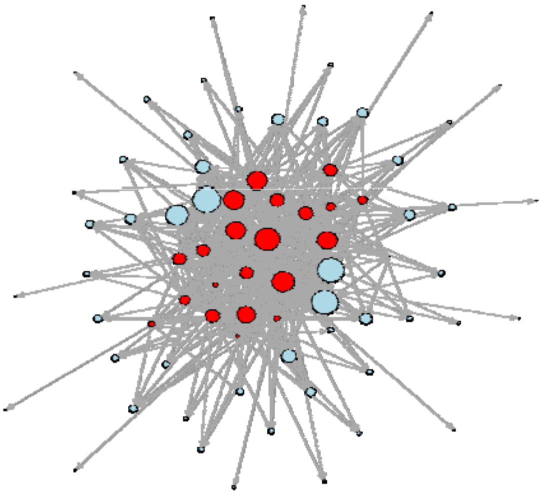


Figure 26. "Is this person known to you?"

Figure 27. Distribution of degree in figure26

Red indicates an interviewee. Size of the node is proportional to its in-degree. There was one “isolate” i.e. someone who was not known to any of the interviewees—we have removed this person from the diagram. There are quite a number of people in this network who are known by many of the interviewees and a significant proportion who are only known by a few people. This suggests that a core and periphery model might fit the data but there may be more complex models that will fit the data more perfectly. This will entail looking for “community structure” where we might find some densely connected sub-groups with relatively sparse connections between them.

“Would you go to this person for advice?”



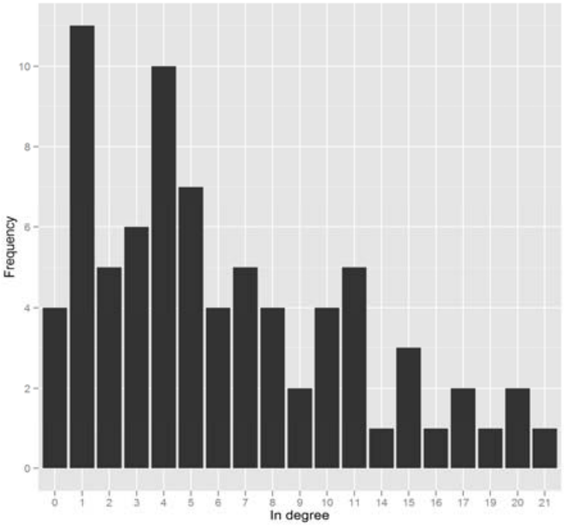


Figure 28. “Would you go to this person for advice?”

Figure 29. Distribution of degree in figure28

There were only four people whom the interviewees would not go to for advice. Most people in the network give advice to a few people and some might be asked for advice at some time by all the interviewees. This suggests that the network is drawing on expertise quite widely across the membership and may mean that individuals are seen to have specialist expertise or personal qualities that are valued in the course of decision making. In future analyses we will examine the data to see if "knowledge brokers" can be identified, that is people who if they were removed from the network would make the connections between other individuals in the network longer or more complex or disconnect them entirely from the network. However, at this stage the high density of ties in this network suggests that there might be many alternative pathways that information and knowledge could take around this network. It will be interesting as well to superimpose the formal organisational structures onto this network to see if some people are depending mainly on colleagues or seeking information from outside organisations.

"Does the person occupy a leadership role?"

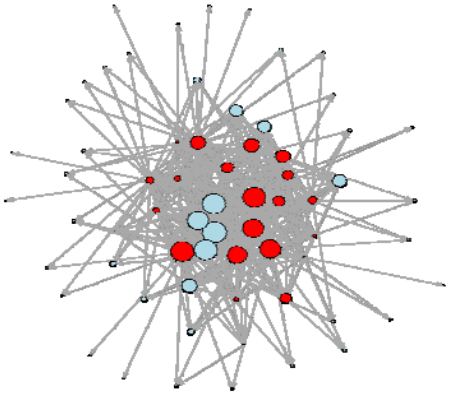


Figure 30. "Does the person occupy a leadership role?"

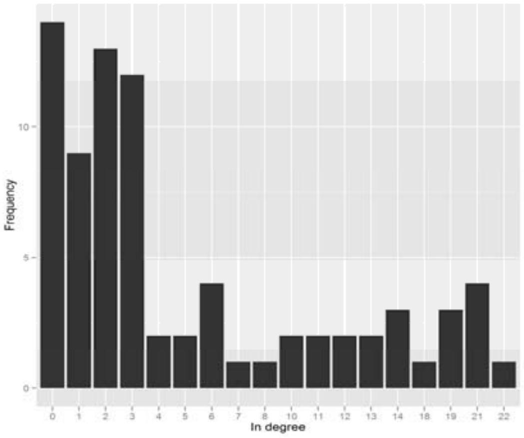


Figure 31. Distribution of degree in figure 30

Many people in the network are seen as occupying a leadership role. Some of the people who are identified as leaders did not volunteer to be interviewed at this time. There may be a difference between the people who offer strategic leadership and those who are most influential through helping the sub-groups to achieve their goals, which were described by one interviewee as the "movers and shakers".

“Do you see this person as influential outside the network?”

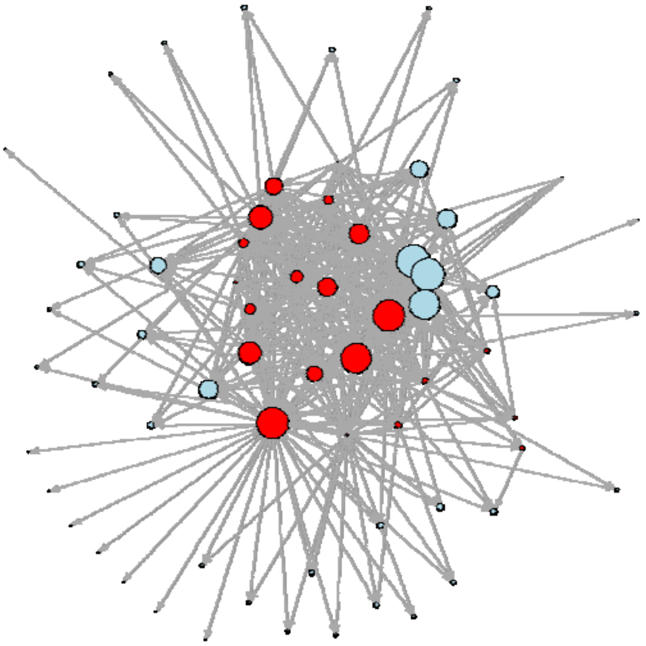


Figure 32. "Do you see this person as influential outside the network?"

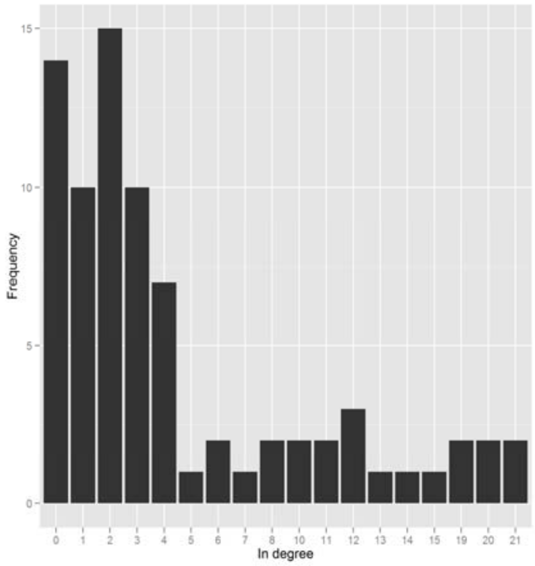


Figure 33. Distribution of degree in figure 32

The majority of people that were known to the interviewees were seen to be influential outside the network with only 14 people seen as not being influential by all the interviewees and a few were seen as influential by almost all. Again this suggests that many of the members of the network are seen to be high profile in paediatric palliative care outside of the local area and may have a national or an international profile.

3.5 Summary and conclusions

This report sets out the rationale for the study, summarises the policy context within which palliative care is being delivered, describes the methods used to gather the empirical data and sets out preliminary findings.

The email survey had a response rate of 42 % by the members of the West Midlands Paediatric Palliative Care Network. The results showed that the network had grown by over 50% in the last few years although 15% had been members for between 7 and 12 years.

Key benefits that members derived from the network included obtaining new ideas to improve practice, access to resources, new professional relationships, an increased sense of efficacy and ability to represent service users. Only a tiny minority felt that service users did not benefit from the network or that the network limited the members in some way, but it was seen as an additional time commitment by some. The majority of members felt that they had contributed to the network. They perceive that the network and its subgroups have delivered improvements e.g. resources, such as tools for improving care as well as successfully bidding for research funding.

Many members belong to other networks, which suggest that there might be an interesting pattern of overlapping memberships evolving over time. Some people and organisations that might benefit from membership were seen to be missing from the network, e.g. social services, education and allied health professionals. The network membership is largely composed of nurses (58%) and doctors (30%) working in the NHS (60%) but 15% and 18% respectively work for hospices and charities.

Initial social network analysis suggests that this network is densely connected, with a core and periphery structure. It appears relatively decentralised with many people being seen as sources of advice, leadership and influence. This is a valuable dataset which will be amenable to further scientific study.

Areas of development for the network might be, to extend representation to all health, social care and third sector organisations providing services to children needing palliative care, as well as service users who the network already plans to include in meetings and who might also be involved in the work of sub-groups.

Sub-groups might focus on transition and service integration, to capitalise on the extended, more complete membership of the network and facilitate communication, cooperation and collaboration at an operational level, as well as responding to feedback from service users described in Strand 5.

In order to improve visibility and communication of their important work, a network website could be developed. This could also be a pathway for the service users to get in touch with the network with questions and suggestions.
The challenge now is to find a way to communicate the findings to the network members in an ethical way to ensure that the study can be of some practical value in the future development of the network.

References:

Department of Health (2004): *National service framework for children, young people and maternity service.* Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4089100

Every Child Matters (2003). Online: http://www.education.gov.uk/consultations/downloadableDocs/EveryChildMatters.pdf
Department of Health (2007): Palliative care services for children and young people in England: an independent review for the Secretary of State for Health. Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_074459

Department of Health (2008): *Better Care: Better Lives.* Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_083106

Department of Health (2010): *Achieving Equity and Excellence for Children.* Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_119449

Department of Health (1995): A policy framework for commissioning cancer services: A report by the Expert Advisory Group on Cancer to the Chief Medical Officers of England and Wales (The Calman-Hine Report). Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4071083

Scottish Office Department of Health (1998). *Acute services review report.* Edinburgh: Stationery Office.
Department of Health (2000): *The NHS Plan: a plan for investment, a plan for reform.* Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4002960

Department of Health (2010): *Getting it right for children and young people.* Online: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_119445

The BiG study

YORK
Health Economics
CONSORTIUM

Strand 4 Report:
An economic evaluation of service delivery

Contents

Strand 4	page
1. Introduction	163
1.1 Background	163
1.2 Analysis methodology	163
1.3 Results	164
1.4 Conditions	164
2. Methodology	166
2.1 Approaches	166
2.2 Literature Review	166
2.3 Costing methodology	166
2.3.1 Cost burden to families	166
2.3.2 Costing Methodology	167
2.3.3 Cost burden to the public services	168
2.3.4 Overall cost estimates for the West Midlands	168
3. Methods	169
3.1 Purpose of literature review	169
3.2 Methodology	169
3.3 Methods of care	169
3.3.1 Short breaks and respite care	169
3.3.2 Hospices	170
3.3.3 Community models	170
3.4 Burden on families	170
3.5 Summary & Conclusions	171
4. The financial burden for families	172
4.1 Introduction	172
4.2 Additional Costs to families	172
4.2.1 Recurring costs	173
4.2.2 Non Recurring costs	175
4.3 Reduced Employment	178
5. The cost to public services	180

5.1 Introduction	180
5.2 Hospital Care	180
5.2.1 Inpatient Care	180
5.2.2 Outpatient Care	183
5.2.3 Diagnostic Tests	184
5.2.4 Overall hospital based care	185
5.3 The costs of other care	186
5.4 The costs of short breaks	187
6. The Broader perspective	188
6.1 The wider cost impact of life limited conditions	188
6.1.1 Costs to families	188
6.1.2 Cost to care services	189
6.2 Unmet needs	191

1 Introduction

1.1 Background

The exact number of children and young people with life limiting conditions in the UK is unknown. Joy (2005) gives a prevalence figure of 25,000 for those under the age of 19, of which between 9,000 – 12,000 have ‘substantial care needs’. Around 3,400 children and young people (up to the age of 24 and excluding neonatal deaths) die each year, mostly of life limiting conditions. Of these, around 20% are affected by cancer, whilst the remaining 80% have a wide range of conditions (Joy 2005). This wide age range indicates that the children, and their families, can be affected over many years. The child may face a slow decline in health over several years: 21% live to beyond 15 years. The management of such children is complex involving a wide range of services and service providers, and crucially, underpinned by care provided by the family, normally the mother.

This study examined the West Midlands region of England. 21 service centres, both NHS and non-NHS participated and they provided contact for the study teams with over 1,000 families and carers of children and young people with life-limited conditions in the region. Strand 4 of the study provided an economic analysis of the provision of services for these children and the impact on their families. The approach consisted of two elements:

- A literature review to identify relevant literature around the costs of different models of care for children and the economic impact on families. This provides a ‘top-down’ contextual analysis for the study.
- Analysis of data gathered by the Strand 1 study team.

1.2 Analysis methodology

The approach to the literature review consisted of an extensive search for literature and studies relevant to children and young people with life-limiting conditions. Snowballing techniques were used to access additional references contained within literature that was reviewed.
For the costing work the methodology was based on the data provided by respondents to the survey of families of children and young people with life-limiting conditions. A range of data was provided including:

- The additional costs to families of caring for children and young people with life-limiting conditions, including any income lost as a result of having to reduce or give up employment;
- The costs of healthcare, including primary, community and acute care, over a period of six months.

The data were used to estimate costs, and median costs were calculated to help quantify costs where data was incomplete. Nationally available cost data for healthcare resources were used to estimate the costs of healthcare.

1.3 Results

Caring for children and young people with life-limiting conditions imposes a considerable cost on families and carers and this is not often recognised in economic studies in this area. There are many hidden costs or resources that families employ which if they were not provided would significantly increase the healthcare costs to public services. One member of the Parents Advisory Group cited an example whereby she has been trained to give her child injections which would otherwise have to be provided by medically trained staff. This kind of care, administered at home, is not often taken into consideration when the costs of caring for life-limited children are estimated.

The findings from the literature review demonstrated that there are very few economic studies of models of care for children and young people with life-limiting conditions. Those that do exist do not necessarily reflect the full costs falling on families. So while models of care such as hospice and community-based care may appear cost effective, they do not always consider the costs that families have to bear to make these models work.

The costing work found that families have to bear costs for a range of everyday and recurring items, including food, clothing, travel and heating. They also have to pay for less regular but more expensive costs such as housing and vehicle adaptations. For the survey respondents, the costs of recurring items was found to be more than £250,000 per year, which when extrapolated to the wider population in the West Midlands, came to an annual cost of over £1.65 million per year. The survey respondents reported that they had spent nearly £1.4 million on non-recurring items and this did not take into account the costs of wheelchairs. This translated as a cost burden of £8.75 million across the West Midlands, in addition to the recurring daily costs of care.

The total costs of hospital-based care for survey respondents were estimated at over £1.3 million per year. This equates to an overall costs for the West Midlands of around £8.4 million per year. Non hospital-based care was estimated at over £200,000 per year or around £1.3 million across the West Midlands.

The cost of short breaks for families in the survey was estimated at nearly £600,000 per year or £3.7 million across the West Midlands. It was not possible from the responses to ascertain the extent to which families had to bear these costs themselves, but some respondents indicated that they had received financial assistance from either the public or voluntary sectors.

1.4 Conclusions

The families and carers of children and young people with life-limiting conditions bear a considerable cost in addition to the healthcare that their children receive. This review examined some of the aspects of those additional costs, as well as the healthcare costs, and they are clearly a significant burden to most families.

Further work should be done to build on these findings so that the true nature of these costs can be fully understood and relevant support can be provided. There may be significant health inequalities in the support and care that is provided to families. The survey responses indicated that in a number of cases, financial support was received from either the public or voluntary sector to pay for certain resources, such as short breaks. Without knowing more about individual family circumstances it is not possible to assess the impact of that support.

The costing work undertaken was limited in that it relied on surveys completed by families. No qualitative work was carried out to support the quantitative analysis and so the analysis was completely reliant on the responses to the survey. Other limitations are cited in the report, including the fact that the cohort of children for whom respondents replied to the survey were of a range of ages and diagnostic conditions and were also at different stages in their conditions.

The findings therefore need to be used with caution. However, they provide a snapshot of the costs required to care for children and young people with life-limited conditions in the West Midlands. Further, more robust analysis, could build on this work.

2 Methodology

2.1 Approaches

Two approaches were adopted to undertake the economic analysis: ‘top-down’ providing context, and ‘bottom-up’ to assess the detail.

The ‘top-down’ approach consisted of a review of the literature around the costs of caring for children with disabilities and life-limiting conditions and of the cost effectiveness of models of care for children with palliative care needs.

The ‘bottom-up’ approach used the data gathered from parents of children and young people with life-limited conditions to estimate the costs of providing healthcare to those children, as well as the additional financial costs to families of caring for those children.

2.2 Literature review methodology

The literature identified for this review came from three sources:

- A focused literature search. Titles and, where available, abstracts of all literature identified were examined, from which were selected studies which addressed costs of services; costs of managing disabled children; burden on families; and economic studies;
- Previous and relevant studies undertaken by the team from YHEC were re-examined and findings within or relevant literature were included;
- Snowballing; identifying relevant titles from bibliographies or the body of articles identified from the above methods.

The review includes studies employing a wide range of methods and no quality assessment of the literature, the studies or their results (and hence robustness) was undertaken. Only literature published in English was included.

The findings from the literature review are summarised below. A more detailed report is available, including detailed analysis of the literature reviewed.

2.3 Costing methodology

2.3.1 Cost burden to families

Data was collected during the course of the study from 188 families of children and young people with life-limiting conditions in the West Midlands. A considerable amount of data was collected on the economic burden of looking after children with life limiting conditions, including:

- One-off costs such as equipment or wheelchairs;
- Ongoing annual costs to parents such as heating or travel.

The interpretation of this data is problematic because it relies on carers being aware of the costs of items they have purchased. In many cases costs given are estimates and in some cases it is not clear what time period the cost covers, i.e. per week or per month. Some respondents reported that they had additional costs but either could not or did not quantify them.

The approach to cleaning the data was therefore important. Where individual costs were clearly stated they were used to build up a framework of costs. Using that data, median costs were extracted. Median costs were chosen over mean costs in the analysis because they helped to mitigate the large range of costs provided. Where respondents had indicated an additional cost but had either not quantified it, or provided a cost that could not be adequately interpreted, the median cost was attributed to that respondent. The result provided an estimated aggregate cost for all respondents who indicated that they had incurred additional costs in a particular category.

Interpretation of this data needs to be carried out carefully. The range of costs incurred and the fact that many respondents did not report additional costs in particular categories meant that it is hard to generalise. Factors affecting these costs are the range of conditions affecting the children in the survey as well as how far advanced the child's condition is at the point at which the survey was taken. Some children may be in an acute stage of their condition while others may have more manageable symptoms. The survey has no way of taking account of this, which probably helps to explain why such a range of costs was presented.

This additional cost burden is made more difficult to bear, in many cases, when parents and carers are unable to continue to work, either completely or partially. Families were asked for details of time they had had to take off work, or where they had had to give up work completely, as a result of the need to provide care for their child.

2.3.2 Costing methodology

The costs used were either the cost provided by the respondent or the median cost where a cost was indicated but could not be quantified. This was the methodology used for all of the cost categories except for wheelchairs. The Parents Advisory Group advised that the data did not appear to be realistic in terms of the potential costs of wheelchairs so these costs were excluded from the analysis.

The data were analysed using the diagnostic categories developed by Strand 1 of the big project. Strand 1 collected ICD10 classification data for all of the 1,180 children and young people to whose families' questionnaires were sent. These data were then used to build six diagnostic categories into which all the children were classified. The economic analysis has used the diagnostic categories to collate the costs for those 188 children whose families returned the survey.

For the cost burden associated with the reduction or loss of employment, a national estimate of average earnings was applied to the amount of working time respondents stated that they and their families had lost. A median cost was calculated and applied to those surveys where a respondent had indicated that there was a loss of earnings but which they were not able to quantify. No account was taken of any additional benefits that families receive to support their care activities or loss of employment. While respondents indicated whether or not they were in receipt of benefits, it was not possible to quantify how much they received.

Details of the analysis of the cost burden to families are provided in Section 4.

2.3.3 Cost burden to the public services

Data was also gathered in the survey, over a period of six months, on:

- The number and type of admissions to hospital;
- Outpatient visits to hospital;
- Diagnostic tests carried out;
- Visits to or by other community, social and voluntary care professionals;
- Any short breaks provided for the child, such as in a hospice, on a family break or to a specialist unit.

From this it was possible to provide estimates of the annual costs to public services of providing care for children and young people with life limiting conditions. The data were analysed and NHS Reference Costs were used to quantify the cost of inpatient and outpatient episodes, as well as the cost of diagnostic tests. The data were analysed using the diagnostic categories and they were also examined from the perspective of demographic differences relating to income, geographical area and the reported ethnic group of the child.

Details of the analysis of the costs to the public services of providing healthcare to children and young people with life-limiting conditions are given in Section 5.

2.3.4 Overall cost estimates for the West Midlands

The results were extrapolated to build up a picture of costs across the West Midlands, using the ICD10 and diagnostic categories for all the families surveyed. The Strand 1 team gathered data on all 1,180 families to whom the survey was sent and they were able to categorise them by diagnosis. These aggregate numbers were then used to extrapolate the wider costs to families in the West Midlands, based on the survey findings.

Details of this analysis are provided in Section 6.

3 Findings from literature review

3.1 Purpose of literature review

Two approaches have been adopted to undertake the economic analysis: ‘top-down’ providing context, and ‘bottom-up’ to assess the detail. The top-down approach includes a review of the literature around costs of caring for children with disabilities and life-limiting conditions and of the cost effectiveness of models of care for children with palliative care needs.

3.2 Methodology

The literature identified for this review came from three sources:

- A focused literature search. Titles and, where available, abstracts of all literature identified were examined, from which were selected studies which addressed costs of services; costs of managing disabled children; burden on families; and economic studies;
- Previous and relevant studies undertaken by the team from YHEC were re-examined and findings within or relevant literature were included;
- Snowballing; identifying relevant titles from bibliographies or the body of articles identified from the above methods.

The review includes studies employing a wide range of methods and no quality assessment of the literature, the studies or their results (and hence robustness) was undertaken. Only literature published in English was included.

The findings from the literature review are summarised below. A more detailed report is available, including detailed analysis of the literature reviewed.

3.3 Models of care

The literature review examined three models of care:

- Short breaks and respite care;
- Hospices;
- Community models.

3.3.1 Short breaks and respite care

Broadly, despite a few reservations expressed in the literature, respite care and short breaks are valued by families. Crucially, delivery of these services should be flexible in order to meet the varying needs of families and be part of a tailored package of support. Evaluation of the Short Break programme funded by recent governments has been positive, with a positive impact noted on families’ quality of life, stress levels, sibling behaviour, and need for long term residential care.

Calculations of their cost effectiveness have produced mixed results. The method of their delivery appears to affect the cost benefit equation. Copps and Heady (2007) produced a negative cost benefit equation, drawing on evidence from a range of studies and sources of costs. On the other hand, NEF Consulting (2009) in their evaluation of the Short Breaks programme produced a positive cost benefit equation.

3.3.2 Hospices

Hospices provide a variety of services, including hospice at home, and evidence of use and experiences of hospices is mostly very positive. Evidence [which evidence?] does suggest that hospice care, given the geographic variation in availability, could be more carefully targeted towards those children with more complex needs. On the other hand, where other service options for families are limited, hospice care is a valuable source of support to families.

It can be argued [it already is!] that children's hospices are mostly cost effective models of care given that the majority of funding is independent of government. However, there are variations in hospice costs and hospices should be encouraged to move to more cost effective models, for example, increased availability of hospice at home.

3.3.3 Community models

There are many models, mostly community based, of care and support which provide alternatives to the more traditional approaches of inpatient hospitals and hospices. Assistive technology to support children and families at home is developing, such that technology dependent children are increasingly being managed at home. The value of key workers, increasingly necessary for families who are in contact with multiple agencies, has been demonstrated although evidence of their cost effectiveness is mixed. Evaluations tend to be model specific. However, it appears that broadly, community based models are well received by families and are cost effective in that visits to hospitals and length of stay whilst there, are both reduced.

3.4 Burden on families

The financial burden falling on families can be analysed under four broad themes: loss of income associated with reduced work opportunities; costs associated with receipt of treatment such as travel and parking; additional costs over and above treatment such as nutritional requirements and equipment; and additional home and living costs such as wear and tear on furniture and clothing and childcare arrangements, as well as one-off large costs of home improvements. (Emerson and Hatton, 2007, Lowson et al, 2007) In addition as a result of these additional financial burdens, which may not be adequately reimbursed by benefits, families may find themselves having to borrow money, access grants and benefits and become in debt. The financial, social and psychological impact of caring for a disabled child is well documented with many studies collecting compelling evidence of the problems faced by these families. Housing is a particular problem faced by these families, who are more likely to live in unsuitable accommodation. Given the potential large number of additional needs of children and young people with life-limiting conditions, this evidence is likely to represent the minimum burden experienced by these families. Of importance for this *Big Study* are the findings by Beresford et al (2007) that parents of disabled children want a better balance between caring and parenting, and that whilst their needs may not be great, they wanted sufficient practical and financial resources to meet their needs and those of their child.

3.5 Summary and conclusions

Our review of the literature indicates a paucity of economic studies on models of care for children and young people with life-limiting conditions. The quality of the economic data in those that we have reviewed is also variable although the evidence broadly points to cost effective community based alternatives to hospital care. These models address supporting families across the totality of the pathway of care from diagnosis to the child’s death, including short breaks, bereavement and the management of acute episodes, as well as the ongoing management of a chronic condition. However, for these models to be cost effective, the significant costs borne by the families such as loss of or reduction in employment and family income, additional costs of caring for the child such as nutrition, clothing and travel costs to receive treatment, and large costs such as housing adaptations, are rarely taken into account.

In these times of increased financial constraints, there is a clear need for evaluations of models of care to include robust economic evaluations. Costing should be more refined and reflect the true costs to society of managing these children, which can be considerable and fall on many agencies and not just health services. The full costs falling on families should also be reflected and acknowledged in these studies, and in government policies.

Services for children and young people with life-limiting conditions are also increasingly provided outside the state sector, with charitable bodies and the independent sector significant providers. Less than 20% of hospice funding is from local health commissioners. These organisations should also look at the cost effectiveness of their models of care, recognising that government funding may decline over time, and that alternative community models are less expensive than traditional inpatient care. Alternative models to enable families to support their children at home, including enabling children to die at home are also likely to be provided by these organisations and will enable the state sector to deliver models of care that are more cost effective and hopefully may therefore increase access and choice to families.

4 The financial burden for families

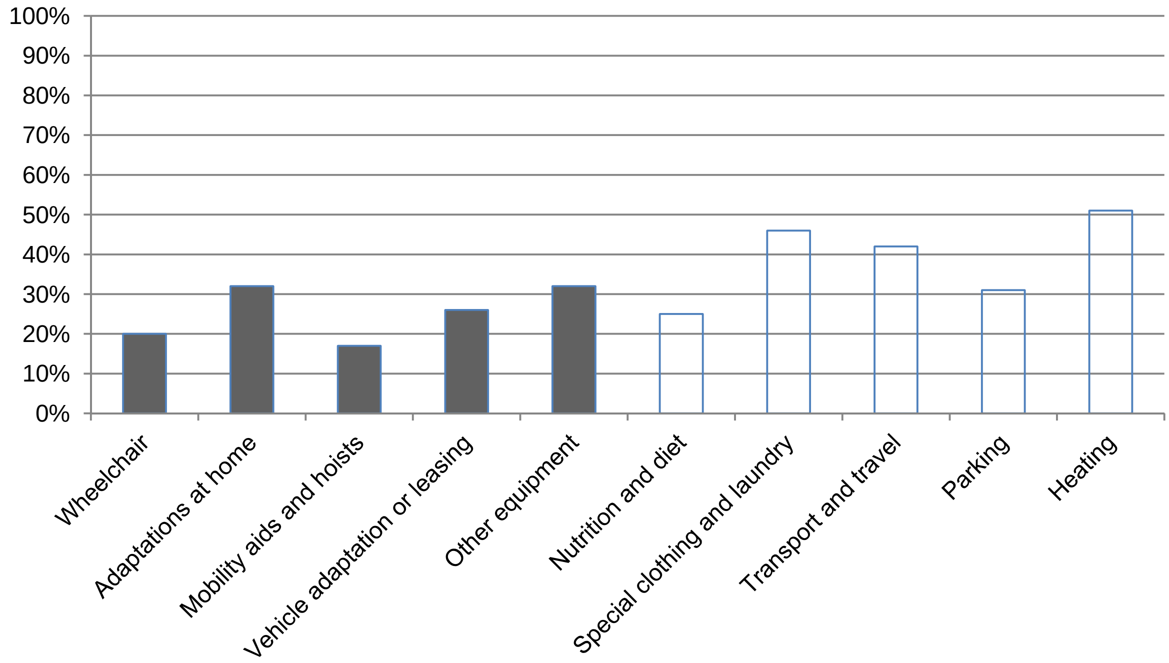
4.1 Introduction

The families of children and young people with life limiting conditions can face considerable financial burdens. In many cases, carers and parents need to spend more money than other families on items such as wheelchairs, travels costs and home adaptations. Additionally, many parents and carers have to reduce their hours or give up work altogether to care for their children and this places an additional burden on families.

4.2 Additional costs to families

The parents' survey asked families about additional costs that they had incurred as a result of caring for a child with a life-limiting condition. We classified these into costs which are likely to be 'one-off' or non-recurring costs, and those which are likely to be ongoing or recurring costs. Non-recurring costs were those for wheelchairs; home adaptations; mobility aids and hoists; vehicle adaptation or leasing; and other equipment. Recurring costs were those for nutrition and diet; special clothing and laundry; transport and travel; parking; and heating.
More than half the respondents stated that they had incurred additional costs for heating and around a third or more reported this for equipment, special clothing and laundry, transport and travel, parking and home adaptations. Figure1 shows the percentage of families reporting additional costs in the survey.

Figure 1: Percentage of families reporting additional costs



Respondents were asked to provide an estimate of the additional costs they incurred for each category. Some caution has to be used in interpreting these responses as respondents were not asked for this information in a consistent format. Nonetheless, the data allowed an estimate to be provided of the ongoing additional cost burden of caring for children and young people with life-limiting conditions.

4.2.1 Recurring costs

For recurring costs, families reported additional costs were annualised. The estimates provided were considered to be robust enough to report, as respondents generally either provided an annual cost or a monthly or weekly estimate that could be annualised.
Table 1 provides details of the numbers of respondents who indicated that they incurred additional recurring costs as a result of caring for a child with a life-limiting condition. The additional costs were estimated at around £264,000 per year.

Table 1: Respondents reporting additional recurring costs

Cost category	Number	Median cost (£)	Cost range (£)	Total cost (£)
Nutrition and diet	30	780	20 – 2,600	28,590
Special clothing and laundry	64	390	50 – 10,000	41,363
Transport and travel	56	675	60 – 10,400	93,580
Parking	37	300	40 – 3,000	15,605
Heating	68	400	20 – 12,000	57,930
Other	35	520	180 – 5,200	26,720
Total				263,788

Respondents described some of the elements of the additional recurring costs that they had recorded:

- For nutrition and diet, 30 respondents reported additional costs. The median additional cost was £780 with a range between £20 and £2,600. Respondents referred to a variety of different supplements that they needed to buy for their children. Both high (ketogenic) and low fat diets were referred to as well as specific items such as boxed milk. Some carers also referred to an increased food intake for their children, particularly when taking steroid-based medicines. Omega 3 supplements were also referred to.

- For special clothing and laundry, 64 respondents reported additional costs. The median additional cost was £390 with a range between £50 and £10,000. Respondents referred to additional costs of cleaning clothes and bedding due to incontinence and also staining caused by chemotherapy. Carers reported having to buy larger sizes of clothing to fit nappies and additional footwear.

- For transport and travel, 56 respondents reported additional costs. The median additional cost was £675 with a range between £60 and £10,400. The main expense referred to was in relation to regular trips to hospital or clinics, either by car, train or taxi. Some respondents also referred to additional costs of travelling outside the region, for example to London to see specialists.

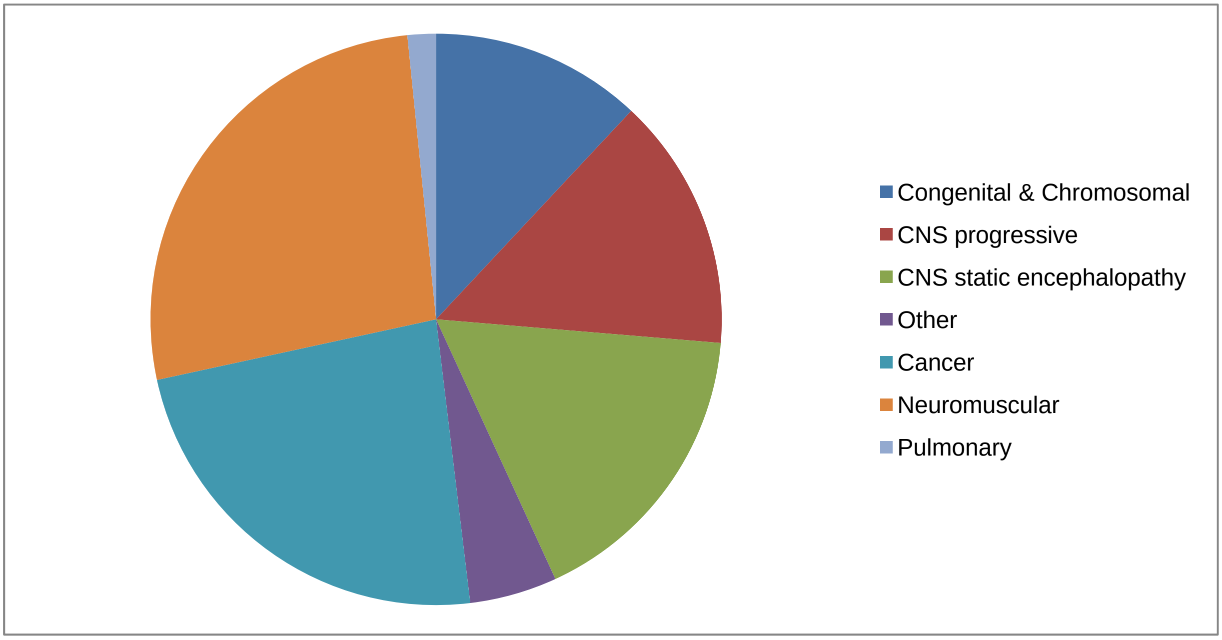
- For parking, 37 respondents reported additional costs. The median additional cost was £300 with a range between £40 and £3,000. Respondents reported additional costs in relation to visits to hospital.

- For heating, 68 respondents reported additional costs. The median additional cost was £400 with a range between £20 and £12,000. Some respondents referred to the need to keep their house warmer than usual due to their child's condition. Some also referred to the need to keep the heating on during the day or overnight for carers.

- Respondents were asked about any other additional costs they incurred, aside from those covered by the headings provided in the survey. 35 respondents reported additional costs. The median additional cost was £520 with a range between £180 and £5,200. Some respondents referred to the need to buy additional play equipment and entertainment for their children, including toys, DVDs and video games and trips to the theatre and cinema. A number of respondents also referred to the need to buy additional nappies.

Figure 2 provides a breakdown of the total additional recurring costs by the type of diagnosis of the child with a life-limiting condition.

Figure 2: Additional recurring costs by type of diagnosis



Respondents with children included in the cancer and neuromuscular disease categories reported the highest proportion of additional recurring costs associated with their child's condition. An average for each disease category was calculated based on the total number of respondents in each category. Again, the cancer and neuromuscular disease categories had the highest average costs (Table 2).

Table 2: Average additional recurring costs by disease category

Cost category	Total reported cost (£)	Number of respondents	Average cost per respondent (£)
Congenital & chromosomal	31,580	47	672
CNS progressive	37,850	26	1,456
CNS static encephalopathy	44,375	37	1,199
Cancer	62,015	31	2,000
Neuromuscular	70,648	31	2,279
Pulmonary	4,260	8	533
Other	13,060	8	1,633
Total	263,788	188	1,403

The average cost per respondent only provides an illustration of the disease categories that had the highest additional costs. It is important to stress that costs for individual families vary depending on the type and severity of the condition that their child has.

4.2.2 Non-recurring costs

Costs that were categorised as non-recurring are generally those costs that are not incurred on a frequent basis. The problem with quantifying these costs is that they are not incurred regularly and so can make estimation difficult. One of the particular problems in estimating non-recurring costs in relation to this survey is that there is no indication of when the costs were incurred. A housing adaptation that took place ten years ago will cost considerably less than if it was being paid for now. Despite these issues, a considerable amount of useful data was gathered on non-recurring costs. The only category that has not been reported is that for wheelchairs, where the costs and data specifications were considered to be unreliable by the Parents Advisory Group.

Table 3 provides details of the numbers of respondents who indicated that they incurred additional recurring costs as a result of caring for a child with a life-limiting condition. The additional costs were estimated at almost £1.4 million.

Table 3: Respondents reporting additional non-recurring costs

Cost category	Number	Median cost (£)	Cost range (£)	Total cost (£)
Adaptations at home	48	7,000	300 – 200,000	870,250
Mobility aids and hoists	26	230	70 – 45,000	57,900
Vehicle adaptation or leasing	42	4,400	350 – 50,000	406,379
Other equipment	47	650	50 – 14,000	60,966
Total				1,395,495

Respondents described some of the elements of the additional non-recurring costs that they had recorded:

- For adaptations at home, 48 respondents reported additional costs. The median additional cost was £7,000 with a range between £300 and £200,000. Some respondents referred to major structural changes to their homes to add or extend bedrooms and bathrooms. Respondents also referred to enhancements made to outdoor areas such as gardens.

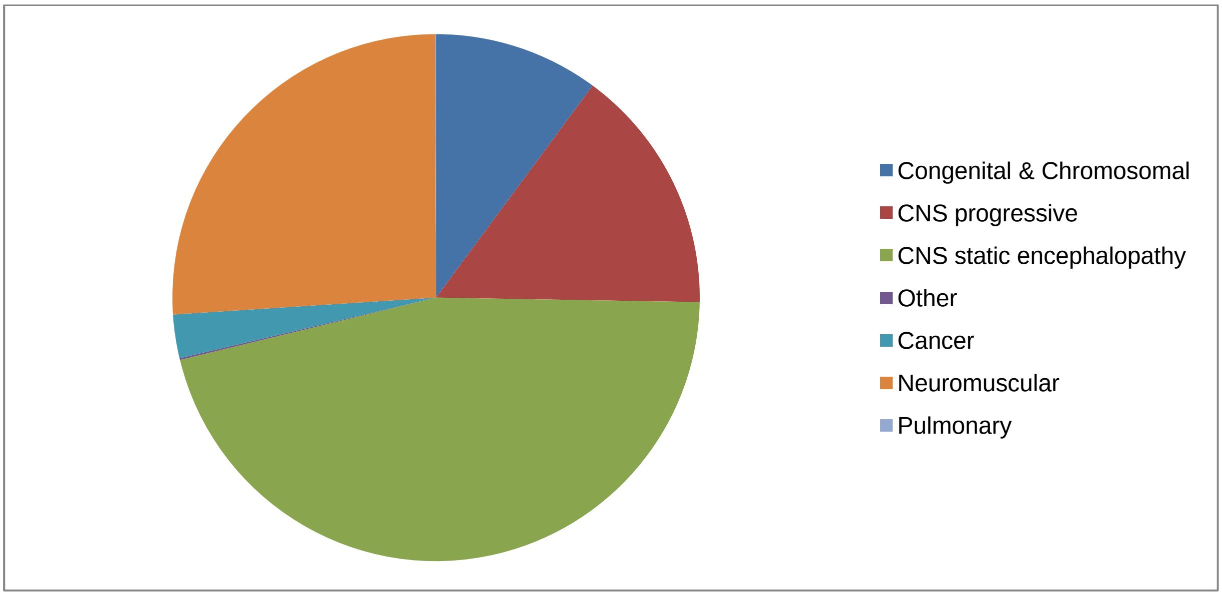
- For mobility aids and hoists, 26 respondents reported additional costs. The median additional cost was £230 with a range between £70 and £45,000. Respondents cited specific expenditure on a stair lift, a mobile hoist and slings.

- For vehicle adaptation or leasing, 42 respondents reported additional costs. The median additional cost was £4,400 with a range between £350 and £50,000. Respondents referred adaptation of their car to accommodate a wheelchair and a number referred to having to buy additional and expensive vehicles as a result of their child's condition.

- For other equipment, 47 respondents reported additional costs. The median additional cost was £650 with a range between £50 and £14,000. Respondents reported a wide range of equipment that they had purchased in relation to their child's condition, including bicycles, a soft play room, a walker, a specialist bed, ramps, a sensory room, special chairs and coagulation and saturation monitors.

Figure 3 provides a breakdown the total additional non-recurring costs by the type of diagnosis of the child with a life-limiting condition.

Figure 3: Additional recurring costs by type of diagnosis



Respondents with children included in the CNS static encephalopathy disease category reported almost half of the additional recurring costs associated, with the neuromuscular disease category accounting for around a quarter of the total costs. An average for each disease category was calculated based on the total number of respondents in each category. Again, the CNS static encephalopathy and neuromuscular disease categories had the highest average costs (Table 4).

Table 4: Average additional recurring costs by disease category

Cost category	Total reported cost (£)	Number of respondents	Average cost per respondent (£)
Congenital & chromosomal	141,240	47	3,005
CNS progressive	211,400	26	8,131
CNS static encephalopathy	640,846	37	17,320
Cancer	37,290	31	1,203
Neuromuscular	362,019	31	11,678
Pulmonary	1,100	8	138
Other	1,600	8	200
Total	1,395,495	188	

As for the recurring costs, the average cost per respondent only provides an illustration of the disease categories that had the highest additional costs. It is important to stress that costs for individual families vary depending on the type and severity of the condition that their child has.

The data provided by respondents in relation to wheelchair costs were not considered to be accurate enough for reporting. The major problems with the data were that the nature of the item purchased was not known. For example, costs reported for the wheelchair category ranged from £50 to £18,000, so without knowing what the expenditure was for, it was impossible to provide an accurate estimate of the cost for each category. Initial reporting of average costs to the Parents Group, confirmed the view that the costs were potentially a significant underestimate. On that basis it was decided that it was best to exclude these costs from the overall estimate of additional costs for families.

It is clear that these costs are a significant additional burden on families. The Muscular Dystrophy Campaign reports that the true average cost of providing an adequate powered wheelchair for a child with a life-limiting condition is in the region of £17,500.[5] Therefore, while the costs obtained from this survey are not considered to be reliable, it is nonetheless very important that the considerable extra costs of wheelchairs is borne in mind in estimating the additional costs of caring for children and young people with life-limiting conditions.

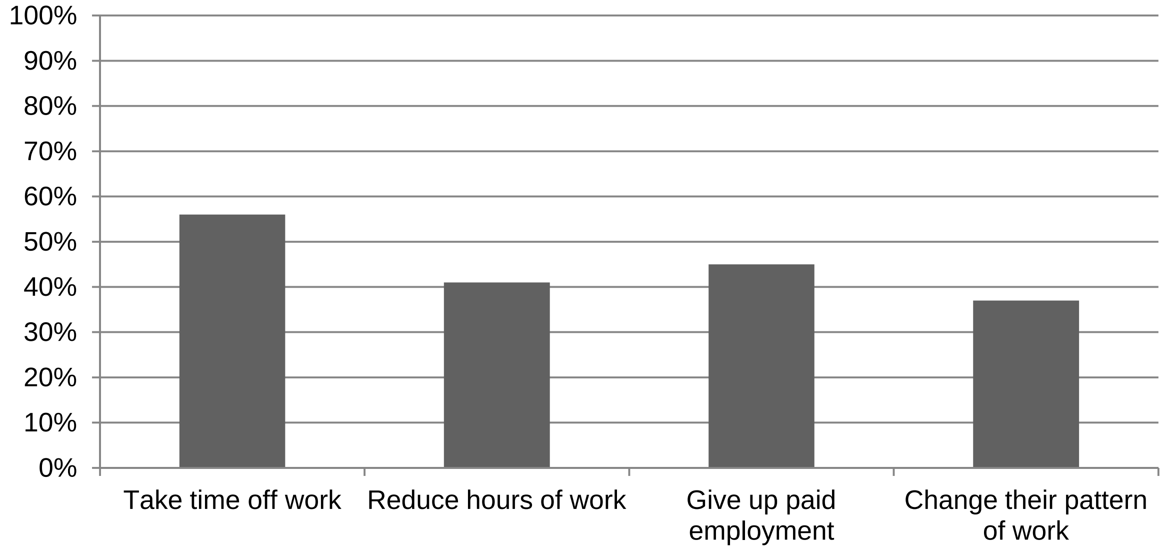
---

[5] Muscular Dystrophy Campaign. *Equipment shortfall: How disabled children are being failed*. www.muscular-dystrophy.org

4.3 Reduced employment

Significant numbers of parents and carers reported that they had had to take time off work or give up work altogether as a result of their child's condition. Figure 4, below, indicates the numbers of respondents who reported that they had had to reduce their employment

Figure 4: Percentage reporting that one or more family members had to reduce work



The annual loss of earned income was quantified by multiplying the reported time off work by the national average annual wage. This was assumed to be £26,100 which was the median gross annual earnings for full-time employees in April 2011.[6]113 respondents reported that either they or their partner, or both, had lost earned income as a result of caring for their child. The median amount of time lost reported by those who provided a figure was three months, which using the average wage, was calculated as a loss of £6,525 per family. That figure was used to provide a cost for those respondents who indicated that they had lost earned income but who were unable to quantify this. On that basis the total annual lost income through reduced employment for the cohort of families who responded was estimated at over£1 million.

This breaks down as follows:

Table 5: Average reported loss of income by disease category

Diagnostic category	Total reported loss of income (£)	Number of respondents	Average loss of income per respondent (£)
Congenital & chromosomal	282,533	47	6,011
CNS progressive	81,563	26	3,137
CNS static encephalopathy	133,001	37	3,595
Cancer	314,831	31	10,156
Neuromuscular	125,824	31	4,059

---

[6] Office for National Statistics. 2011 Annual survey of hours and earnings.

Pulmonary	24,469	8	3,059
Other	64,141	8	8,018
Total	1,026,362	188	

Parents and carers of children and young people with cancer reported the largest loss of earned income as a result of their child's condition.

Many families that responded reported that they were in receipt of benefits as a result of their child's condition. While the loss of work may have other consequences such as the psychological impact on parents and carers, financial benefits may help to offset the economic impact of reducing or losing employment. From an economic perspective, whether or not families receive benefits that make up for their loss of income, there is still a cost to be borne by the state which provides the benefits. Whether the loss of income is borne by families or the state an economic loss is incurred.

The table below indicates the percentage of families who reported that they were in receipt of benefits:

Table 6: Percentage of families in receipt of benefits

Families in receipt of:	Percentage:
Disability benefit	63
Carer allowance	58
Mobility allowance	39
Housing benefit	15
Supplementary benefit	4

Discussion with members of the Parents Group for the study gave the study team an understanding of why every family is not claiming disability benefit or carer allowance. The views of the Group were that the complexity of the benefits system can lead to situations where it becomes economically disadvantageous to claim certain benefits, if this puts at threat other sources of income. The Parents Group also believed that the complexity of the system puts some people off claiming benefits to which they are entitled.

5 The cost to public services

5.1 Introduction

Children and young people with life-limiting conditions tend to have multiple co-morbidities, i.e. they have a primary diagnosis with a particular condition but they may also be susceptible to other conditions in addition to or as a result of their primary condition. Care for these children and young people is an ongoing, complex process and there is no simple care pathway that can be observed. Each child has their own care pathway and alongside scheduled care such as tests and follow-ups, they will have unplanned episodes of illness. This means that existing sources of data on the costs of care, such as Hospital Episode Statistics and NHS reference costs cannot be used to provide general ranges of costs for particular conditions.

In this study, families were asked to record the episodes of care that their children received over a period of six months. This provides an opportunity to build up some information on the costs of care provided for children and young people with life-limiting conditions. The sample of children is relatively small and so care must be taken in drawing conclusions from the costs generated in this study. Nevertheless, the cost data provides some useful insights into the cost of care for children and young people with life-limiting conditions and the extent to which this may vary between conditions.

5.2 The costs of hospital care

The key components of hospital care are:

- Inpatient care, where a child is admitted to hospital;
- Outpatient care, where a child attends hospital for scheduled follow-up meetings with clinicians;
- Diagnostic tests.

5.2.1 Inpatient care

Respondents to the survey were asked to report details about the number of times their child was admitted to hospital in the previous six months. Around half of respondents reported a range of reasons for admission to hospital. These included various infections; viruses; gastrointestinal complaints; sleep studies; insertions of enteral feeding tubes investigations and tests; fractures and surgery. They also reported their lengths of stay and place of admission.

These data were analysed and assumptions made about the specialties to which the child would have been admitted. Reference costs were then used to calculate a cost for each admission, based on the length of stay reported. These were aggregated for each of the diagnostic categories within the group of respondents. The costs of inpatient stays for each diagnostic category were as follows:

Table 7: Estimated costs of admission to hospital by diagnostic category

Diagnostic category	Number	Cost (£)	Average (£)
Congenital & chromosomal	47	283,901	6,040
CNS progressive	26	124,806	4,800
CNS static encephalopathy	37	138,212	3,735
Cancer	31	416,453	13,434
Neuromuscular	31	93,134	3,004
Pulmonary	8	45,521	5,690
Other	8	45,521	5,690
Total	188	1,147,548	6,104

Costs were extrapolated for a year and an average calculated. The results indicate that the cancer diagnostic category incurs a higher inpatient care cost than the other diagnostic categories. It is important that these results are treated with caution as they are likely to be sensitive to changes in incidence and unit costs. For example those diagnostic categories with higher average costs may be the result of one or two children who experienced atypical levels of illness during the six month period. Nevertheless, the data provides an indication of the costs of providing inpatient care for a group of children and young people with life-limiting conditions.

Analysis of the inpatient costs was carried out to examine any potential differences depending upon the demographic profile of the respondents. As the numbers of respondents in some of these categories were small, the median cost was used as an indicator, rather than calculating the full reported cost. The data was analysed by:

- Income group: those stating their family income was below £30,000 per year versus those stating that their income was above £30,000 per year;
- PCT cluster: PCTs were clustered into 5 areas within the West Midlands – Arden; Birmingham and Solihull; Black Country; Staffordshire; and West Mercia;
- Child ethnic group: Black; Mixed; Other; South Asian; White British.

The estimated median cost of inpatient care for those respondents whose families were in the lower income group was lower than that for the higher income group (Table 8). There was also variation in the median cost of inpatient care across the different PCT clusters, with Arden having the highest cost and the Black County the lowest cost (Table 9). The data relating to the ethnic group of the child was too limited to report for three of the categories (Table 10). The estimated median costs for the two largest groups, White English and South Asian, were relatively similar.

Table 8: Estimated median costs of admission to hospital by income group

Income group	Number of people responding	Median cost (£)
Under £30,000 per year	60	3,195
Over £30,000 per year	26	7,490

Table 9: Estimated median costs of admission to hospital by PCT cluster

PCT cluster	Number of people responding	Median cost (£)
Arden	11	6,681
Birmingham and Solihull	23	4,772
Black Country	17	3,050
Staffordshire	18	3,579
West Mercia	15	5,726

Table 10: Estimated median costs of admission to hospital by child's ethnic group

Ethnic group	Number of people responding	Median cost (£)
Black	n/a	n/a
Mixed	n/a	n/a
Other	n/a	n/a
South Asian	14	4,772
White British	67	4,004

Estimated inpatient cost data were also analysed by the Measures of Processes of Care (MPOC) survey that was carried out amongst respondents. The MPOC recorded respondents' attitudes towards the care their child receives, and this was categorised into:

- Enabling and partnership;
- Providing general information;
- Providing specific information about the child;
- Coordinated and comprehensive care for the child and family;
- Respectful and supportive care.

The data were analysed to determine the upper and lower quartiles of respondents for each. Upper quartile respondents were those who were most positive about the particular aspect of care and the opposite was true for the lower quartile respondents. The median costs are summarised in Table 11.

Table 11: Estimated median costs of admission to hospital by MPOC category

	Lower quartile number	Median cost (£)	Upper quartile number	Median cost (£)
Enabling and partnership	22	6,204	24	4,772
Providing general information	21	11,121	27	3,340
Providing specific information	22	5,726	24	2,863
Coordinated and comprehensive care	21	5,726	23	4,772
Respectful and supportive care	22	4,865	20	4,772

While the data has limitations in terms of small numbers, it provides an indication that the costs of inpatient care were lower for those respondents who were more satisfied with the care their child has received. Further costing work to examine this issue further would be helpful.

5.2.2 Outpatient care

Outpatient follow-up visits may be scheduled as part of a child's ongoing care for their condition or as a result of an inpatient episode for a complication. Most of the 137 respondents whose children had attended outpatient clinics in the last six months, reported the nature of those clinics, such as examinations for respiratory conditions or routine clinics for ENT check-ups. These clinics were categorised into specialties and reference costs were applied to calculate the overall costs, detailed in the table below:

Table 12: Estimated costs of outpatient care by diagnostic category

Diagnostic category	Number	Cost (£)	Average (£)
Congenital & chromosomal	47	33,512	713
CNS progressive	26	13,969	537
CNS static encephalopathy	37	15,162	410
Cancer	31	58,673	1,893
Neuromuscular	31	7,382	238
Pulmonary	8	3,885	486
Other	8	7,665	958
Total	188	140,248	746

There was a significantly higher cost burden associated with cancer patients. This appears to relate to the number of clinics and outpatient appointments these children have, rather than increased costs of the clinics themselves.

The estimated median cost of outpatient care for those respondents whose families were in either income group was reasonably similar (Table 13). There was variation in the median cost of outpatient care across the different PCT clusters, with Arden having the highest cost (Table 14). The data relating to the ethnic group of the child was too limited to report for three of the categories (Table 15). The estimated median costs for the two largest groups, White English and South Asian, were relatively similar.

Table 13: Estimated median costs of outpatient care by income group

Income group	Number of people responding:	Median cost (£)
Under £30,000 per year	87	630
Over £30,000 per year	47	532

Table 14: Estimated median costs of outpatient care by PCT cluster

PCT cluster	Number of people responding:	Median cost (£)
Arden	15	1,050
Birmingham and Solihull	36	630
Black Country	27	630
Staffordshire	30	475
West Mercia	23	420

Table 15: Estimated median costs of outpatient care by child's ethnic group

Ethnic group	Number of people responding:	Median cost (£)
Black	n/a	n/a
Mixed	n/a	n/a
Other	n/a	n/a
South Asian	21	630
White British	105	525

The estimated median costs of care were also analysed by the MPOC category (Table 16).

Table 16: Estimated median costs of outpatient care by MPOC category

MPOC category	Lower quartile number	Median cost (£)	Upper quartile number	Median cost (£)
Enabling and partnership	25	525	37	735
Providing general information	29	735	40	630
Providing specific information	27	532	37	630
Coordinated and comprehensive care	27	525	37	735
Respectful and supportive care	30	531	32	739

While the data has limitations in terms of small numbers, it shows that median costs for the upper quartile were mostly higher than for those in the lower quartile. Further costing work to examine this issue further would be helpful.

5.2.3 Diagnostic tests

An important aspect of follow-up care is diagnostic tests. In children with complex conditions these are numerous and frequent, so the costs of these tests have been factored into the analysis as an additional cost. NHS reference costs were used to calculate the total cost for each of the diagnostic categories. The estimated costs of diagnostic tests were:

Table 17: Estimated costs of diagnostic tests by diagnostic category

Diagnostic category	Number	Cost (£)	Average (£)
Congenital & chromosomal	47	8,396	179
CNS progressive	26	3,339	128
CNS static encephalopathy	37	4,657	126
Cancer	31	28,689	925
Neuromuscular	31	2,255	73
Pulmonary	8	2,057	257
Other	8	1,055	132
Total	188	50,448	268

As with outpatient visits, cancer patients had the highest costs in relation to diagnostic tests.

5.2.4 Overall hospital based care

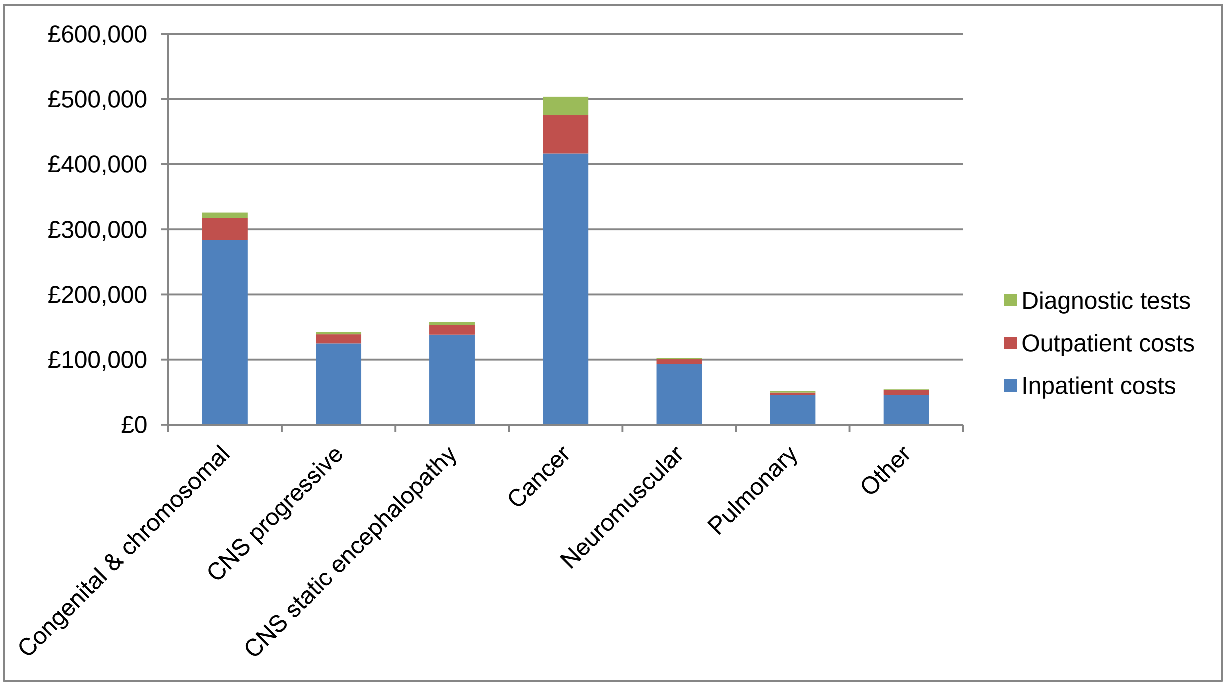
The overall annual cost burden for hospital based care, for the cohort of children and young people whose families responded to the survey was around £1.34 million. This was calculated by aggregating the costs of inpatient and outpatient care, along with the costs of diagnostic tests. This breaks down as follows:

Table 18: Estimated overall costs of hospital care by diagnostic category

Diagnostic category	Number	Cost (£)	Average (£)
Congenital & chromosomal	47	325,809	6,932
CNS progressive	26	142,114	5,466
CNS static encephalopathy	37	158,031	4,271
Cancer	31	503,815	16,252
Neuromuscular	31	102,771	3,315
Pulmonary	8	51,463	6,433
Other	8	54,241	6,780
Total	188	1,338,244	7,118

The cancer diagnostic category had the largest estimated cost and also the highest average cost per child.

Figure 5: The costs of hospital-based care by diagnostic category



Any new data here not reported in other research?

5.3 The costs of other care

Families also reported data about the numbers of times they had contact with non-acute care professionals, including:

- Community children's nursing teams;
- GPs;
- Clinical psychologists;
- Allied health professionals, such as physiotherapists, speech and language therapists and occupational therapists.

The costs of this care were estimated using standard costs derived from the Unit costs of Health and Social Care, published annually by the Personal Social Services Research Unit (PSSRU) of the University of Kent. The costs were as follows:

Table 19: Estimated costs of other care by diagnostic category

Diagnostic category	Number	Cost (£)	Average (£)
Congenital & chromosomal	47	32,908	700
CNS progressive	26	39,836	1,532
CNS static encephalopathy	37	51,131	1,382
Cancer	31	43,398	1,400
Neuromuscular	31	33,048	1,066
Pulmonary	8	3,280	410
Other	8	8,656	1,082
Total	188	212,257	1,129

Respondents who were carers of children and young people in the cancer, CNS progressive and CNS static encephalopathy categories had higher average costs than other diagnostic categories.

5.4 The costs of short breaks

Short breaks can consist of time the child spends away from their family or with their family in a variety of settings. These can include hospices, other specialist units or holidays. The costs of short breaks may be funded by the family themselves or they may receive some financial assistance for these breaks. It is not clear from the data whether or not the costs associated with these short breaks were self-funded or funded through voluntary or statutory means. Nevertheless, from an economic perspective, they are additional costs associated with the child's life-limiting condition. The costs were calculated using PSSRU data and other data derived from the literature review. The costs were as follows:

Table 20: Estimated costs of short breaks by diagnostic category

Diagnostic category	Number	Cost (£)	Average (£)
Congenital & chromosomal	47	211,850	4,507
CNS progressive	26	78,592	3,023
CNS static encephalopathy	37	135,640	3,666
Cancer	32	79,224	2,476
Neuromuscular	30	87,484	2,916
Pulmonary	8	0	0
Other	8	1,184	148
Total	188	593,974	3,159

6 The broader perspective

6.1 The wider cost impact of life-limiting conditions

The costs estimated and reported in Sections 4 and 5 indicate a significant cost burden among the families that completed the survey, alongside the NHS and other support service costs. These results were used to extrapolate costs for the population of families in the West Midlands that was sent a survey and for whom the Strand 1 team was able to gather diagnostic data.

6.1.1 Costs to families

Applying the costs estimated for the survey families to the wider population numbers, the costs for the West Midlands region can be estimated. The following tables summarise the cost burden for the population of identified families with children and young people with a life limiting condition in the West Midlands.

Table 21: Estimate of additional recurring costs for West Midlands families

Diagnostic category	Total population	Respondent population	Respondent costs (£)	Total costs (£)
Congenital & chromosomal	297	47	31,580	199,559
CNS progressive	175	26	37,850	254,760
CNS static encephalopathy	271	37	44,375	325,017
Cancer	148	31	62,015	296,072
Neuromuscular	135	31	70,648	307,661
Pulmonary	62	8	4,260	33,015
Other	92	8	13,060	150,190
Total	1,180	188	263,788	1,655,691

Table 22: Estimate of additional non-recurring costs for West Midlands families

Diagnostic category	Total population	Respondent population	Respondent costs (£)	Total costs (£)
Congenital & chromosomal	297	47	141,240	892,517
CNS progressive	175	26	211,400	1,422,885
CNS static encephalopathy	271	37	640,846	4,693,764
Cancer	148	31	37,290	178,030
Neuromuscular	135	31	362,019	1,576,534
Pulmonary	62	8	1,100	8,525
Other	92	8	1,600	18,400
Total	1,180	188	1,395,495	8,758,958

Table 23: Estimate of loss of income for West Midlands families

Diagnostic category	Total population	Respondent population	Respondent costs (£)	Total costs (£)
Congenital & chromosomal	297	47	282,533	1,785,368
CNS progressive	175	26	81,563	548,982
CNS static encephalopathy	271	37	133,001	974,142
Cancer	148	31	314,831	1,503,064
Neuromuscular	135	31	125,824	547,943
Pulmonary	62	8	24,469	189,635
Other	92	8	64,141	737,622
Total	1,180	188	1,026,362	6,442,059

The overall additional cost burden for families in the West Midlands is estimated at more than £1.65 million per year. It is also estimated that these families incur costs of more than £8.5 million for items such as additional equipment or adaptations. As is highlighted in Section 4, this does not include the additional cost to families of wheelchairs. In addition, these families have collectively lost nearly £6.5 million in income per year through having to reduce or give up their employment, albeit with some families claiming additional benefits to offset this loss.

6.1.2 Costs to care services

The same process was applied to the costs of care to extrapolate total costs for the West Midlands.

Table 24: Estimate of total hospital costs for West Midlands families

Diagnostic category	Total population	Respondent population	Respondent costs (£)	Total costs (£)
Congenital & chromosomal	297	47	325,809	2,058,836
CNS progressive	175	26	142,114	956,537
CNS static encephalopathy	271	37	158,031	1,157,470
Cancer	148	31	503,815	2,405,310
Neuromuscular	135	31	102,771	447,551
Pulmonary	62	8	51,463	398,838
Other	92	8	54,241	623,772
Total	1,180	188	1,338,244	8,399,617

Table 25: Estimate of other care costs for West Midlands families

Diagnostic category	Total population	Respondent population	Respondent costs (£)	Total costs (£)
Congenital & chromosomal	297	47	32,908	207,951
CNS progressive	175	26	39,836	268,127
CNS static encephalopathy	271	37	51,131	374,500
Cancer	148	31	43,398	207,190
Neuromuscular	135	31	33,048	143,919
Pulmonary	62	8	3,280	25,420
Other	92	8	8,656	99,544
Total	1,180	188	212,257	1,332,251

Table 26: Estimate of short break costs for West Midlands families

Diagnostic category	Total population	Respondent population	Respondent costs (£)	Total costs (£)
Congenital & chromosomal	297	47	211,850	1,338,712
CNS progressive	175	26	78,592	528,985
CNS static encephalopathy	271	37	135,640	993,471
Cancer	148	31	79,224	378,231
Neuromuscular	135	31	87,484	380,979
Pulmonary	62	8	0	0
Other	92	8	1,184	13,616
Total	1,180	188	593,974	3,728,135

Using this methodology the annual costs to public services of providing care and treatment for children and young people with life limiting conditions in the West Midlands is estimated almost £10 million. £8.4 million of this relates to hospital based care with around £1.3 million spent in primary and community care and the voluntary sector.

The cost of short breaks is also substantial, at £3.7 million. It is likely that these costs are incurred by the public and voluntary sectors as well as by families themselves. However, it is not clear in what proportion these costs are borne. The survey indicated that some families bear these costs themselves while others are able to access funding from a variety of sources.

6.2 Unmet need

The data gathered from the 21 organisations involved in the study provided contact details for just over 1,100 families of children and young people with life limiting conditions. Epidemiological data suggests that this accounts for around 50% of the likely number of children and young people in the West Midlands with the identified conditions.

If the numbers of children and young people with these conditions is doubled it would not necessarily be correct to double the costs to families and care services. One of the reasons that families may not have been identified by the organisations involved in the study is that they may not be in contact with those organisations because their children require less care beyond their family group. Equally it could be because they are unaware of these services or do not want to access them for particular reasons. It is therefore impossible to speculate whether or not there is an economic cost associated with these families and what that cost would be.

Families who responded to the survey reported about whether or not certain needs were met, and in some cases, whether or not they were paying for those needs themselves. There was not enough data in the surveys to fully quantify the extent of these unmet needs and further research in this area would be beneficial.

Strand Five Report:
Patient and Public Involvement in the Big Study

Sophie Staniszewska and Lee Gunn

Contents

Strand 5

page

1. Introduction 196
1.1 Aims 196
1.2 The importance of context, process and impact 196
1.3 Key messages and priorities for actions 197

2. Methods of involvement 198
2.1 Aims for groups 198
2.2 Recruitment of group members 198
2.3 Group meetings 199
2.4 Participants and researchers 199
2.5 Levels and stages of involvement and specific activities 200
2.6 Impact methods of investigation and analysis 200
2.7 Support and group transition 200

3. Analysis 202
3.1 Overview 202
3.2 Methods of Analysis 202

4. Context and process of involvement 203
4.1 Recruitment of Participants 203
    4.1.1 Payment and recognition 203
    4.1.2 Responsiveness to personal and family circumstances 203
    4.1.3 Ways of working – meeting with parents and carers 204
    4.1.4 Ways of working – remote contact with parents and carers 204
    4.1.5 Ways of working – children and young people 204
    4.1.6 Summary – context and process factors 205
4.2 The nature and impact of group feedback 206
    4.2.1 Feedback on methods 206
    4.2.2 Feedback on results 207
    4.2.3 Identification of Study limitations 207
    4.2.4 Final meetings 208
    4.2.5 Summary – feedback 208
4.3 Diary data on impact of patient and public involvement 209
    4.3.1 Diaries as a tool to collect data 210
    4.3.2 Diaries from members of the parents' advisory group 210
    4.3.3 Diaries from researchers 211
    4.3.4 Summary -findings on diary entries 212

5. Discussion 213
5.1 Context process and impact 213
5.2 The use of feedback from patient and public involvement groups 213
5.3 Assessing the impact of involvement 214

5.4 Challenges and limitations 5.5 The future for Big Study Collaborations 5.6 Challenges & Limit	214 214 214
6. Conclusions	216
References	217 - 218

Tables

Table 1: Context and process factors
Table 2: Types of feedback provided by advisory groups
Table 3: Themes from diary data

Terminology

Terms used for the activities and approaches described in this report vary across disciplines. For consistency, we have usually chosen to refer to ‘patient and public involvement’ when discussing involvement more generally, and to the ‘parents’ advisory group’ and ‘children and young people’s advisory groups’ to refer to specific groups as appropriate.

In this report, ‘service users’ refers to children and young people with life-limiting or life-threatening conditions and their family members of all ages (including young siblings) who access services relevant to the Big Study. The terms ‘parents’ and ‘parents and carers’ both refer to family members who hold a parental or guardianship role in relation to the child or young person receiving services, though they may in fact be (for example) grandparents or foster parents.

1 Introduction

Patient and public involvement (PPI) in HTA and health research has become increasingly common internationally. In the UK Dame Professor Sally Davies (2009), Director General of NHS Research and Chief Medical Officer stated that involvement should be the norm, not the exception, in health research and this is being reflected internationally (1). In health and social care service provision patients and the public have become involved in developing plans and making decisions about local services, with a focus on developing appropriate and relevant services, and on a broader vision of encouraging participative democracy, public accountability and transparency in society (2). A recent white paper in the UK has also emphasised the important role of patient and the public in health care (3). In research, the aims are similar with involvement seen as an attempt to enhance the quality, relevance and appropriateness of health technology assessment and also to contribute to the broader democratisation of research, through participatory forms of involvement that encourage partnership. Within the UK considerable effort has been focused on developing an infrastructure to operationalize the policy commitment to PPI, through the work of organisations such as INVOLVE (4) and the Research Design Services (5) in the UK, which enable researchers to embed patient and public involvement into their work. Together, the policy focus and the infrastructure have created a supportive context in which PPI in research has flourished.

1.1 Aims

The Big Study aimed to involve parents, children and young people in key stages of the study. The purpose of this involvement was to ensure that the study and its focus was relevant, acceptable and appropriate from the parents and children and young people's perspectives, so enhancing the quality and utility of the findings and contributing to the development of key messages from the study.

In addition, strand 5 of the study aimed to assess the impact of patient and public involvement within the study and contribute to methodological development in understanding the difference involvement makes to research. The model of involvement we drew on was a collaborative one, although some elements were consultative (6). In reality the models of involvement can often vary within one study over time, and this was the case with strand 5 of the Big Study.

1.2 The importance of context, process and impact

Good patient and public involvement in research depends on a number of key context and process factors being in place. These factors provide architecture for involvement and are important for creating the environment for involvement to make a difference or have an impact. As the context and process of involvement and its relationship to impact is generally a poorly understood area of research, primarily to do with poor reporting in papers (7,8), in addition to exploring impact, the Big Study examined the context and process factors that are important in involving parents, children and young people as the precursor to any type of impact or difference. We hope our findings will help ensure future studies consider context, process and impact when planning the involvement of children, young people and parents.

1.3 Key messages and priorities for action

Advisory group members confirmed their support for key messages identified across the study. In addition, the following key messages were synthesized from advisory group discussions:

Families need help, not hurdles: Family-centred support needs to be co-ordinated, from the time of diagnosis to bereavement and beyond, so that families can better carry out their central role in caring for a child or young person with a life-limiting or life-threatening condition.

Involvement matters: Advisory group members believe that research should make a difference. Involving children, young people and parents throughout studies, including making use of findings, can be key to making this happen.

Priorities for action were identified in connection both with research and with service improvements. Firstly, it was recommended that a compelling campaign for better care needs to take place, underpinned by the research findings. In addition:

- More research is needed on family costs, including one-off expenses, and costs for families before, during and after bereavement.
- Involvement of parents and children and young people needs to be co-ordinated throughout research studies, including taking action on findings.
- The centrality of the family's role in caring for a child with a life-limiting or life-threatening condition needs to be recognised, and support needs to be provided for the whole family.
- The effectiveness and expertise of parents should be respected and strengthened, with access for everyone to information and advocacy services.
- Emotional support should be made available to family members, in addition to practical and financial support, from the time of diagnosis to bereavement and beyond.
- There should be much better co-ordination of services to meet the needs of each individual family, including identified and empowered key workers, and co-ordinated service management.
- Improvements need to be made in finance and funding - including after bereavement.
- Information about services and funding should be easily available when families need it and when they can take advantage of it.
- Communication with children and young people is important, including providing opportunities for them to talk with someone when they do not want to worry other family members.
- High quality services should be equally provided, with identified gaps addressed – for example, provision for under-fives.

2 Methods of involvement

Patient and public involvement was an important element of the Big Study from its inception. The charity ACT (now merged with Children's Hospices UK to form Together for Short Lives) held a key role in developing the proposal alongside the academic partners. Several of the researchers involved had experience of patient and public involvement, and consultations took place with service users to identify areas of relevance and concern to inform the preparation of the proposal (9). Research teams held further discussions with service users during detailed planning and preparation of data collection tools, including consultations with members of ACT and with members of the Comensus (Community Engagement and Service User Support) group at the University of Central Lancashire. This work provided an important foundation for the development of the parents' and children and young people's advisory groups. A research fellow was appointed to develop the groups of parents and of children and young people, and to work with service users and researchers on collaborative involvement. This role was particularly important as it cut across the four strands and included significant negotiation of input from parents, children and young people on the different strands.

The parents' advisory group met bi-monthly, with additional communications by telephone and email. The West Midlands Medicines for Children Research Network young people's advisory group provided feedback at three of their regular meetings, to supplement input from young people who volunteered to take part in a Big Study children and young people's advisory group.

2.1 Aims for groups

Our aim for the groups was that they should work as collaboratively as possible with the researchers on the Big Study. Where collaborative involvement was not possible, consultative involvement was utilised, where individuals are asked to comment on aspects of the study, but may not be involved as full partners (6). Many aspects of strand 5 activity focused on facilitating those interactions and trying to make them as collaborative as possible.

2.2 Recruitment of group members

Recruitment for the groups was integrated with recruitment for the study as a whole, so that families were not confused by a number of separate invitations to take part. Some parents volunteered to become members of the parents' advisory group as a result of contacts with ACT or with one of the academic institutions. Others (the majority) volunteered when completing the survey administered by Strand 1 of the study. Volunteers were contacted by telephone or email and invited to become members of the parents' advisory group. They were given leaflets about the advisory group and information sheets on writing diaries about their involvement, together with references to further information on the study as a whole.

It proved difficult to identify very many children and young people with life-limiting or life-threatening conditions who were willing and able to take part in an advisory group, despite considerable efforts for recruitment of children and young people - perhaps reflecting the challenges that exist in this area of research when children or young people are not very well and maybe prefer to focus on other activities. We invited several children and young people who had been volunteered by their parents to join the group, providing leaflets and information sheets. Two young service users responded to our invitation and took part in a helpful discussion. To ensure that young people's perspectives informed the study as fully as possible, we also made contact with the West Midlands young people's group of the Medicines for Children Research Network (MCRN), and held three lively and productive discussions on the research.

2.3 Group meetings

After consulting volunteers about their preferences, we arranged for the parents' advisory group to meet face to face bi-monthly as this seemed a reasonable timescale with their other commitments. Terms of reference for the group were agreed at the first meeting. Email and telephone contacts were made between meetings.

One strand of the Study usually provided the main focus for discussion, in order to facilitate involvement across the study, with the final meeting considering key messages and priorities identified by all strands. In addition, at each meeting the group followed up actions initiated at previous sessions. Parents and carers who could not attend meetings were encouraged to take part by means of telephone and/or email at times of their choosing, and some individual meetings also took place face to face in locations to suit parents' convenience. All discussions were documented and meetings were recorded, with permission from participants, as a record of their feedback. Minutes were taken at bi-monthly meetings of the parents' group and both minutes and detailed notes on feedback given for each strand of the research were reviewed and agreed by attendees. Group members completed diary entries to assist with the exploration of the impact of public and patient involvement.

2.4 Participants and researchers

The parents' advisory group included a 'core' group of five parents and carers who maintained a substantial level of involvement at every stage once the group was established, providing feedback on many aspects of the study. Members of a wider group (12 more parents and carers who responded to our invitation) gave their views and responded to specific questions by telephone or email. The 'core' group included a grandparent who was a particularly active member, and the wider group included a foster parent (both carers for a child or young person with a life-limiting condition). Four bereaved parents were members of the group, two of them in the 'core' group.

Though we did not ask for personal details about the children and young people who took part in discussions, some of them indicated that they had experience of services investigated as part of the Big Study. One advisory group member in particular had extensive experience of services, and a sibling also took part.

Five researchers representing strands 1-4 of the research each came to one or more parents' group meetings. Two researchers from strand 5 and a representative of Together for Short Lives also attended most meetings. The strand 5 research fellow acted as facilitator throughout, as well as co-ordinating communications between meetings.

2.5 Levels and stages of involvement and specific activities

A collaborative style of involvement informed all interactions with members of the groups, which includes respectful listening and valuing all contributions. Collaboration specifically informed the definition of the role of parents' advisory group members, the terms of reference, and the processes of involvement.

Group members assisted with the interpretation of qualitative data from the survey in relation to the identification, validation and consolidation of key themes. They contributed to the discussion of key messages from all strands of the Study, and collaborated in identification of priorities. Additional activities fitted with a consultation model rather than a collaborative or user-led model, with group members commenting on methods, explaining and illustrating aspects of the meaning of data and themes, and answering specific questions to help resolve apparent ambiguities in the data. In addition, group members identified limitations of the research and suggested subjects that need to be addressed in future.

2.6 Impact – methods of investigation and analysis

Members of the advisory groups and members of the research teams were invited to keep diaries of their involvement. It was explained that this would help the team to explore the experience and the impact of user involvement. Information sheets were given to members of the advisory groups, and informed consent was obtained for their participation in this part of the study. Information sheets were also provided for researchers and the team agreed that researchers' submitting a diary for analysis would constitute consent (minutes of Big Study team meeting 1 May 2011).

A suggested format for diary entries was provided, and a sample diary was made available, though it was emphasized that alternative formats were also welcomed. Assistance in completing the diaries was offered. In practice, most members of the advisory groups chose to follow a suggestion made at the first meeting to submit a diary entry after each discrete event, using the blank diary format provided (with suggested questions to be used as prompts). This format was also used by some researchers, while others expressed a preference for a more individual approach.   Diary entries have been received from all members of the 'core' parents group and from researchers, and these are being subjected to thematic content analysis (10), with initial results informing this report.

2.7 Support and advisory group transition

Processes were put in place for support to be accessible for advisory group members if it was required, and contact details of appropriate organisations were provided. It was recognised that taking part in advisory groups might be stressful for group members who already had many emotional and practical demands made on them, and we carefully took this into account in all of our interactions. Several parents and carers said that they enjoyed and appreciated the opportunity to talk about sensitive issues and that their input and feedback would help make a difference to others facing associated difficulties.

Group members have been invited and encouraged to keep in touch with Together for Short Lives in associated work after the Big Study is formally completed, and details of local research groups have been provided. Some group members may also choose to assist with further dissemination where appropriate, such as being involved in writing peer-reviewed papers. This reflects good practice in supporting individuals who wish to continue their association with a particular area of research or activity, as discussed by Morrow et al. (10).

3 Analysis

In addition to facilitating parents' and children and young people's involvement in the Big Study, we carried out detailed analysis specifically for strand 5 of the study.

3.1 Overview

Three separate sets of analysis have been carried out, to reflect different areas of focus for this strand of the study.

Firstly, we analysed context and process elements, vital to our understanding of the potential for impact to take place. These are based on relevant sections of meeting minutes and associated documentation.

Secondly, we analysed feedback provided by the parents' and the children and young people's advisory groups for researchers on each strand of the study, and for the study as a whole, based on detailed notes made from recordings of discussions and from email responses to questions.

Thirdly, we analysed data on the impact of user involvement, based on diaries completed by members of the advisory groups and by researchers.

3.2 Methods of analysis

We are making use of thematic content analysis (11) and qualitative analysis software (12) to explore context and process factors, types of feedback, and diary entries.

This report provides indicative findings, with detailed findings to be reported in peer-reviewed papers.

4 Results

The results are divided into a number of sections. The first considers the context and process of involvement, as these are vital factors in developing an environment in which involvement can have an impact on research or on individuals (researchers or members of advisory groups). We have identified some key context and process factors which were important in developing the involvement of parents' and children's and young people's groups in strand 5 of the Big Study, and which have implications for future studies. The second section considers the feedback that members of groups provided and some potential associated impacts. The third section considers the diary data from members of groups and from researchers.

4.1 Context and process of involvement

As outlined previously, knowledge of the context and process factors in relation to impact is not well developed (6). The Big Study addressed this key issue in order to develop our understanding of key factors future studies should consider when they plan to involve families in research.

4.1.1 Payment and recognition

Payment and recognition have been previously recognised as an important factor in the context underpinning involvement (13).The role and importance of payment as a way of valuing contribution is contested in patient and public involvement, with some people adopting a volunteering approach which suggests that payment can change the nature of interaction, while others view it as a key way in which people feel valued and an indicator of equity, as others attending meetings are often in paid employment. A decision was made to pay actively involved members of Big Study advisory groups an amount of money corresponding to four to five hours of time contributed during the study, according to the payment criteria utilised by UNTRAP – The University of Warwick Public Involvement Network (14). Feedback indicates that the payment is appreciated, though several people said that they became involved in order to help with the research, and did not expect payment. An additional issue is that payment can affect some families' benefits, and so discourages involvement, as financial stringencies are a key issue for many of the families in the Study. In addition to the one-off payment, members of Big Study advisory groups had expenses covered for the meetings they attended.

4.1.2 Responsiveness to personal and family circumstances

Families of children with life-limiting or life-threatening conditions need to balance multiple, complex, often unpredictable and stressful demands on them. Many parents who volunteered for groups found it difficult or impossible to attend meetings at specific times, even though they expressed a keen desire to help with the research in any way they could. To respond to their preferences and needs as productively and sensitively as possible, we developed processes in collaboration with individuals and also with the 'core' group within meetings. The research fellow spent time getting to know individual parents and carers and their particular interests and areas of expertise, and encouraged them to take part as much or as little as they wished in providing their feedback. Most conversations took place initially over the phone and then by email, though the research fellow also visited two families at home and two parents in their workplaces (at their suggestion). In later stages, she was then able to direct questions to family members with specific relevant expertise.

4.1.3 Ways of working – meetings with parents and carers

The importance of ways of working has been identified by a range of organisations including INVOLVE (15, 16). Much less is written about the content of good ways of working in order to inform future practice. Parents' advisory group meetings were considered important, not only for attendees but also for members of the wider group of parents and carers. They provided a structure for involvement activities, a point of communication and a means of addressing issues in depth. There was very careful selection of meeting venues to ensure the location was convenient and to ensure the ambience of the meeting place was appropriate for the study and would enable parents to feel comfortable. The meeting time was limited to two hours (except for the longer final meeting), to fit in with other commitments and in recognition of the intensive nature of the proceedings. The research fellow leading on involvement facilitated each meeting with the aim of enabling contributions from the parents, so that all parents felt able to contribute, that everyone listened in a respectful way to others' contributions, that researchers had a chance to raise and discuss key issues and that all contributions were valued. In this way, we aimed to create an appropriate environment for contribution, with clear leadership from the facilitator to encourage discussion. Members of the wider group received details of discussions in advance of each meeting and after it, and some asked for specific items to be covered.

It was helpful for researchers to meet parents and carers in person, especially for those who did not make face to face contact with families as part of their own strand research. Parents and carers who attended meetings appreciated meeting each other on a regular basis, and built up a substantial amount of expertise about the study which informed their ongoing contributions.

4.1.4 Ways of working -remote contact with parents and carers

While some parents and carers were able to meet on a regular basis, with additional email contacts between meetings, some parents who wanted to be involved could not attend any of the meetings because their circumstances left little flexibility for this kind of commitment. The research fellow worked with these parents to find alternative ways of communicating with them to ensure their input was integrated into the study wherever possible (17).

4.1.5 Ways of working – children and young people

Discussions with children and young people took place in a family home and at meetings of the West Midlands Medicines for Children Research Network (MCRN) young people's group. A parent helped with the discussion in a family home, and facilitators of the MCRN group assisted with the MCRN group discussions.

It was helpful to be able to consult children and young people from a family who had taken part in the Big Study, and also to access an established group who had associated experiences and who were already used to involvement in research. We asked for group members' views on the best ways for us to work with children and young people, which informed our plans and conduct of later contacts. We also sought their views on emergent findings, while taking into account the complexities of children and young people's involvement in data analysis (18). We involved children and young people in interpreting, commenting on, illustrating and prioritising the issues raised by family members on changes in services they would like to see. We also asked for their views on key messages and priorities for the study as a whole. We encouraged group members to talk about their own perspectives on the subjects under discussion, and they responded with insight, sensitivity, humour, and a considerable amount of expertise. As with the parents' group, sessions were recorded (with participants' permission) and detailed notes were made and shared with other researchers so that members' contributions could be fully taken into account.

4.1.6 Summary-context and process factors

The importance of context and process in developing the potential for impact of involvement has been recognised as vital in developing our understanding of the impact of public involvement more broadly. The Big Study is one of the first to explore these issues with this particular group of families. The context and process factors that are being identified in this study should be considered by future studies that are attempting to enable parents to contribute. The table below (Table 1) lists a selection of context and process factors identified.

Subtheme	Context and process factors
Payment and recognition	Value of contributions affirmed
	Complexity of financial issues acknowledged
Responsiveness	Meetings set up to fit in with group members' circumstances
	Flexible communication outside meetings, with support and contact by email and phone
	Understanding the circumstances, expertise and interests of each member and working in accordance with these
Ways of working – parents meetings	Importance of meetings recognised and valued, with documentation of all feedback
	Input from those who could not attend in person
	Careful selection of venue, convenient location, convenient time to fit with other commitments
	Facilitation from convener to enable contributions
Ways of working - remote contact	Understanding individual circumstances, expertise and interests, and agreeing best means and timing of communication

	Encouragement to participate as much or as little as appropriate
	Keeping members aware of progress, without pressure always to contribute
Ways of working – children/young people	Fitting in with individual circumstances to ensure that everyone is comfortable in contributing
	Keeping a focus on group members' perspectives and expertise, and valuing all contributions

4.2 The nature and impact of group feedback

This section is concerned with the feedback that family members provided in their capacity as collaborators and consultants on the study. We aimed to involve parents and carers, and also children and young people where possible, about each strand of the research. Group members provided feedback on methods and assistance with the interpretation of early results. They also contributed to discussion of key messages, identification of priorities, and planning dissemination and future work associated with the study findings.

4.2.1 Feedback on methods

Early involvement of parents, children and young people helped shape the original bid (9). As the study developed, when they met with the parents' advisory group, researchers from each strand explained the methods they were using to collect and analyse data. In most cases, members of the group provided confirmation that methods were appropriate, though some limitations were identified and alternative approaches suggested for future research. For example, it was noted that some questions to families could have been misinterpreted and costs underestimated accordingly.

There were expressions of disappointment that it was not feasible to make some suggested changes to methods at later stages of the study, although this would be expected at this stage when funders have already agreed direction and ethics committees agreed the methods and questionnaires. Some parents and carers had specific expertise related to aspects of the study and were able to engage in discussions on methods at a more informed level than anticipated by members of the research teams.

The children and young people were especially interested in methods of gaining the views and experiences of people in their age group, and they suggested ways in which recruitment for advisory groups might be improved in future research projects, for example, by age-specific materials, online discussions and use of online videos.

4.2.2 Feedback on results

The parents’ advisory group and the West Midlands MCRN young people’s group provided substantial feedback on the key messages and priorities resulting from the study. In addition, at an earlier stage, researchers from strands 1 and 4 presented early emergent findings for the advisory groups to consider and to contribute their expertise in helping to interpret the findings. Feedback on processes and associated concerns was given for all strands.

In most cases, group members provided validation for emergent findings and key messages, confirming that these resonated with their personal experiences and those of other families they knew. They added illustrative detail to less clearly specified responses, for example on the complications of arranging for suitable adaptations to meet changing needs over time. They presented explanations for unexpected results, and corrected some potential misinterpretation of data, for example, on reasons for relatively low take-up of some benefits.

There was substantial involvement with early analysis and interpretation of qualitative responses to the open-ended question in the survey, where families identified changes they thought could most improve services. For example, a key change suggested was better co-ordination of services, including the provision of information to families at appropriate times. Both the parents’ and the young people’s groups discussed and endorsed the researchers’ initial classification of responses, and they suggested key priorities for change. As expected, these priorities were different for parents and for young people, though both also acknowledged the importance of the other’s perspective. For example, members of the young people’s group felt that the ways in which service providers communicated with children was an especially important issue. Bereaved parents identified their specific priorities for change, including (for example) the abrupt cessation of financial help for families after bereavement.

Issues of finance and funding were of particular concern to group members, and it was emphasised that costs to families require further detailed research, as an underestimate of costs can have adverse effects.

In addition to the contributions made during meetings, nine parents responded to a specific email request for clarification of acceptable distances from appropriate services (for example, hospices).

As well as researchers hearing feedback first hand at meetings, detailed notes were provided for researchers on relevant strands so that they could take feedback fully into account in their work.

4.2.3 Identification of Study limitations

Parents in particular felt that it was important to identify the limitations of the study, and this was included at their request in the parents’ advisory group terms of reference. Group members were concerned that future research should be planned to address gaps where appropriate. Accordingly, impact is expected to extend beyond the boundaries of the study towards future research.

4.2.4 Final meetings

The final meeting of the parents' advisory group provided feedback on key messages and priorities within and across the Study. All members of the 'core' group attended, along with researchers from strands 1, 2 and 5 of the Study. The group provided validation of messages and priorities identified from the research. Group members suggested increased emphasis on needs for emotional support for family members, on co-ordination of services (and identifying who is responsible overall for getting things done), and on services for under-fives. The potential role of families sharing their expertise was raised, with the suggestion of a particular role for parents who had the opportunity to develop expertise over a number of years. In addition, issues for bereaved parents were discussed, together with pre-bereavement and post-bereavement experiences and associated needs for support.

In addition to previously identified priorities (finance and funding, information provision, and co-ordination of services), these issues were noted for attention in the definition of key messages from the Study.

The final meeting with the West Midlands MCRN young people's group provided validation of key messages and priorities from results of the survey and interviews with families (Strands 1 and 2). They confirmed their own list of priorities identified at previous meetings (responsive services, better communication with children and young people, and support for the whole family). Members of the group expressed a particular interest in issues of communication within the health service (including the use of new technologies for communication), and also the promotion of wider understanding of the lives and the needs of children and young people with life-limiting or life-threatening conditions.

4.2.5 Summary – feedback

The table below (table 2) lists different kinds of feedback provided by the groups.

Table 2 –Types of feedback provided by advisory groups

Strand of research	**Type of feedback and impact**
All	Emphasis on importance of practical use of research Validation of key messages across all strands Identification of priorities for action Identification of study limitations Identifying audiences and appropriate formats for dissemination Adding detailed perspectives of bereaved parents Insight into expertise of families and its potential for application
Strand 1	Interpretation of qualitative responses to survey questions Identification and illustration of priorities for change Linking and grouping results to create a coherent view Validation of emergent findings Comments and suggestions about methods Providing detailed feedback on acceptable distance from services
Strand 2	Validation of methods and findings Suggesting parents' priorities (for example, caring staff) Illustrating issues of communication with children and young people
Strand 3	Suggesting focus on responses to family concerns Emphasis on need for practical co-ordination of services Request for more information on aims/achievements of network
Strand 4	Interpretation of early results, with illustrative details Presenting explanations for unexpected results Warning of dangers of underestimating costs Comments and suggestions about future research Validating the importance of finance and funding to families
Strand 5	Feedback on context and process factors of involvement Identifying what works well and what needs improvement Provision of insights into experiences and impacts

4.3 Diary data on impact of patient and public involvement

Diary entries were collected from the five 'core' members of the parents' advisory group and from researchers, focusing on the experience and the impact of involvement. We also requested dairies from members of the wider group of parents and from the children and young people who volunteered to help with the research, and it is possible that these may be returned to us and analysed at a later stage. For this report, we have carried out an initial thematic analysis of the parents' and researchers' diary entries that we have received to date.

4.3.1 Diaries as a tool to collect data

We used diaries as the main tool to collect data on the impact of involvement (19). This has the advantages of allowing for reflection on the part of individual participants, and enabling our understanding of a range of different perspectives. We encouraged diarists to include positive and negative items, and emphasized that we valued all of their views and contributions.

4.3.2 Diaries from members of the parents' advisory group

We have received diary entries from five members of the parents' advisory group to date. These comprised entries for individual meetings that each of these participants attended. Preliminary analysis of the diaries indicates that members of the group found meetings interesting and useful:

"Good to meet people who share parts of your experience and realise that you are far from being alone. Good to try to give something back"

"I can … give my opinions and know that I am being taken seriously."

"I felt that … we gave a lot of important feedback and some findings were … understood more clearly as a result of our experiences"

There were suggestions about how the group might function more effectively, which we took into account as the study progressed:

"…more parents present at these events would offer a much wider perspective of experiences…"

"May have more influence if we went to the strand rather than they came here"

Some comments indicated that parents thought their contributions had an influence on the research:

"I believe the researchers took [our views] on board and saw our individual points…"

Some measure of scepticism was also apparent, for example:

"I would only be able to judge [the difference involvement made] at the end of the process ..."

Members of the group were very committed to research having a practical impact on improvement of services. Several members suggested that it would be desirable for the group to continue beyond the end of the study:

"… this group should be an ongoing one to regularly feedback and discuss how care is being delivered and to take actions to address the gaps that exist".

4.3.3 Diaries from researchers

We received diary entries from researchers, commenting on a variety of issues associated with the experience and impact of working with advisory groups. In addition, many researchers attending a team meeting to consider results across the study, also attended by two members of the parents' advisory group, contributed their views.

Entries indicate that the involvement of family members was very much appreciated.

For example:

"It is really helpful to get some personal perspectives on the … data that we are processing."

"I feel that the parents' advisory group provided helpful and insightful comments/feedback and put forward challenging, interesting questions that were thought provoking for the research team"

"Made [the discussion] more grounded and sharper. Made us all think about bringing it together and making it real."

Researchers also raised useful and interesting points about the challenges and the detailed processes of involvement. For example:

"I think it was underestimated how long it would take to establish these groups…"

"Some of the parents in attendance were bereaved parents whereas others were not and this may have been distressing for those parents whose child was alive to hear stories of those who child had died. Does this need more reflection?"

"Perhaps the parents could have been asked to submit some questions to the team before the group took place."

At a final team meeting held to discuss the reporting of findings, researchers confirmed the importance of involving children, young people and parents as fully as possible across the Study.

4.3.4 Summary - findings on diary entries

The table in this section lists a number of themes identified from the diaries received.

Table 3 – Themes from diary data

Source of data	Themes
Advisory group members	Importance of focus on what the research can achieve
	Wish to help – in particular, to improve services
	Enjoyment of meetings, including meeting other parents
	Appreciation of comprehensive feedback
	Differing views on processes and detailed arrangements
	Need for ongoing group to carry the work forward
Researchers	Appreciation of advisory group contributions
	Useful to meet family members/gain insights into their experiences
	“Keeps it real”
	Challenges for researchers (for example, questioning their work)
	How involvement ‘fits’ with research processes
	How involvement could be better integrated with research

5 Discussion

The Big Study included a strand which focused specifically on involvement within the other four research strands, with the intention of contributing to the acceptability, relevance and utility of the research and findings. This reflects the current focus within health and social care research which acknowledges the important contribution that patients and the public make to research (7).

5.1 Context, process and impact

A key issue within such an endeavour is developing people's involvement in ways that reflect the particular situation of participants. The importance of acknowledging context and process in PPI has only been recently recognised (7). Context and process factors outlined in this report need to be in place within a study for parents, children and young people to have the chance of making a difference or an impact on a study. Context and process factors are wide ranging and multi-facetted. A key concept in this context is study responsiveness, which describes the way in which the study responds to an individual's particular situation. An example is organising meetings to fit in with people's personal circumstances, such as the best time of day, the most convenient location and the most appropriate venue. While seemingly minor, these can be important context factors for ensuring involvement makes a difference to the study. In the Big Study all these elements were important, alongside flexibility in communication such as email and phone. These enabled parents to communicate with the researcher when it was convenient for them and so did not have a negative impact on their care responsibilities. Future studies should explore the potential for remote collaboration more extensively, particularly in groups for whom regular meetings are not realistic. If attention is not paid to context and process factors such as these in future studies, then the potential for positive impact will be considerably reduced. Context and process factors will be fully explored and reported in a peer-reviewed paper which will aim to make an important contribution to our understanding in this field.

5.2 The use of feedback from patient and public involvement groups

The Big Study adopted a collaborative approach with the advisory groups involved in the study. This differs from including individuals as research subjects or sources of data. In this study parents, children and young people provided feedback in a range of ways outlined in this report in table 2. This feedback led to a range of changes made to how data was being interpreted or linked. The specific impact of this feedback is considered in the next section. It is also important to observe how much advisory group members enjoyed being involved in the study in this way and the extent to which they valued the potential to contribute to a significant study that might shape future services. In turn, the researchers valued their contribution, their confirmation of many of the study findings, and the explanatory power parent feedback provided, as a form of community validity (20) that enabled the researchers to feel that study results had a resonance with the real world of parents', children's and young peoples' experiences.

5.3 Assessing the impact of involvement

In this strand, impacts were identified using a range of methods, including reflective diaries and analysis of meeting notes. It must be recognised that within strand 5 of this study, advisory group members were collaborators and not subjects of research. As such, their feedback was concerned with fundamental aspects of the research, including methods and results. In this study, the parents, children and young people contributed most to the analysis, synthesis and interpretation of data. This included validation of emergent findings and importantly how they were being interpreted and linked to other findings. Contributions to prioritisation of findings and key messages were also important, with parents keen to be involved with the campaigning activities of Together for Short Lives to ensure the study findings are disseminated and used as widely as possible to develop better services. As with feedback, the impact that parents, children and young people made will be reported in peer-reviewed papers to ensure a strong contribution to knowledge in this field.

5.4 The ‘positioning’ of PPI within a study

As Staley emphasizes (1), the impact of involvement is highly context-specific. Though there is a growing literature on involvement, little work has been published on involvement across multi-stranded mixed methods research such as this. In addition, accounts of work that focuses explicitly on the impact, conceptualisation and theorisation of user involvement are comparatively rare (7).

Originally strand 5 was designed to underpin all the other strands and embed involvement into the study. In reality, having a separate strand for this activity posed some challenges in the extent to which involvement could run alongside all the research activities. In hindsight, we would design the study so that involvement was an integral part of each strand. While many aspects of involvement worked well in the study, a closer integration into each of the themes may have resulted in the development of closer collaborations between the parents, children and young people and the researchers leading each strand.

5.5 The future for the Big Study collaborators

The parents, children and young people made important contributions to the study and many reported that they had enjoyed it. Their interest in research was developed, and they expressed a keen interest in ensuring key study messages are implemented in the development of better services. The research team have facilitated opportunities for advisory group members to become involved in other activities, such as other groups run by Together for Short Lives and the Warwick University Research Network for User Involvement (UNTRAP), so that further opportunities for involvement may be developed through ongoing relationships as discussed by Morrow (10).

5.6 Challenges and limitations

While patient and public involvement was accepted as important by all partners in the Big Study, its separation into a separate strand may have provided some challenges in the fluidity with which researchers and members of the advisory group could interact and the extent to which the parents’ advisory group could contribute to the research. As a result we would recommend that future studies integrate their involvement activities within each activity as this may increase the potential for impact on the research.

Though we found that it was helpful to include a diverse set of parents in the parents' advisory group, the difficulties of enabling all group members to meet together face to face in some ways resulted in less intensive involvement. Family members who were able to meet each other reported that they appreciated this direct interaction. Additional contacts took place on an individual basis between family members and strand 5 researchers, who then co-ordinated responses and presented them for further discussion at meetings. Some group members were reluctant to share their email addresses more widely, or to deal with multiple emails; but it is possible that carefully controlled use of a social networking site may have been a valuable option.

Group members and researchers provided data for the assessment of impact in the form of diaries. In some ways, this required them to be research participants as well as collaborators, and this dual role presented some challenges for the research team in the design and conduct of associated processes. However, we feel that the resulting insights will be both interesting and valuable to others undertaking similar work.

6 Conclusions

Strand 5 of the Big Study has made an important contribution to our understanding of how parents, children and young people can be involved in research. The parents' and children and young people's advisory groups appear to have had a clear impact on later stages of the study, particularly on the interpretation of data and the identification of priorities for action. In addition, the research helped us to identify context and process factors of importance to patient and public involvement, to better understand what matters to parents and to children and young people taking part, and to appreciate the varied perspectives of researchers working on the Study. This strand will disseminate its findings through peer-reviewed publication and provide important insights for those wishing to develop collaborative research studies in the future.

We hope that group members are able to continue their involvement through associated organisations in the future, building on the success that been achieved as a result of their expertise, commitment and determination to make a difference.

References

Staley K (2009). Exploring Impact: Public involvement in NHS, public health and social care research. INVOLVE, Eastleigh

Staniszewska S (2009). Patient and public involvement in health services and health research: a brief overview of evidence, policy and activity. Journal of Research in Nursing*. Jul 2009; vol. 14: pp. 295 – 298*

Equity and excellence: Liberating the NHS (2010). Department of Health, London

INVOLVE http://www.invo.org.uk/ (Accessed 27 July 2012)

Research Design Services: http://www.nihr.ac.uk/infrastructure/Pages/infrastructure_research_design_services.aspx (Accessed 27 July 2012)

Brett, J, Staniszewska, S, Mockford,C, Seers K, Herron-Marx S and Bayliss H, (2010). The PIRICOM Study: A systematic review of the conceptualisation, measurement, impact and outcomes of patients and public involvement in health and social care research, UK Clinical Research Collaboration, London

Brett J, Staniszewska S et al (2012). Mapping the impact of patient and public involvement on health and social care research: a systematic review. Health Expectations doi: 10.1111/j.1369-7625.2012.00795.x

Staniszewska S, Brett J, Mockford C, Barber R (2011). The GRIPP checklist: Strengthening the quality and transparency of reporting for patient and public involvement in research. International Journal of Technology Assessment for Health Care, 27 (4):391-399.

Hunt A. et al, "Why does it happen like this?" Consulting with services users and providers prior to developing bid to evaluate services for children with life-limiting conditions and their families in the West Midlands, paper in development, 2012.

Morrow E et al, (2010). A model and measure for quality service user involvement in health research, International Journal of Consumer Studies, 34, 532-539

Green J and Thorogood N (2009). Qualitative Methods for Health Research, Sage, London

NVivo Software (Copyright QSR International Pty Ltd, Version 8, 1999-2009)

Payment for involvement: A guide for making payments to members of the public actively involved in NHS, public health and social care research (2010) INVOLVE, Eastleigh

UNTRAP, University/User Teaching and Research Action Partnershiphttp://www2.warwick.ac.uk/fac/cross_fac/healthatwarwick/untrap/ accessed 28 July 2012

INVOLVE (2010) INVOLVE Ways of Working, INVOLVE, Eastleigh

Branfield F (2009). Developing user involvement in social work education, SCIE, London

Read S and Maslin-Prothero S (2011). The involvement of users and carers in health and social research: the realities of inclusion and engagement, Qualitative Health Research 21, 704-713

Coad J and Evans R (2007). Reflections on practical approaches to involving children and young people in the data analysis process, Children and Society volume 22, 41-52

Charmaz K (2006). Constructing Grounded Theory, Sage, London

Staniszewska S, Haywood KL, Brett J, Tutton E (2011). Patient and public involvement in PROMs : Evolution not revolution. The Patient: Patient-Centred Outcome Research (in press)