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Doctoral Thesis

**A Qualitative Exploration of Family Members' Experiences of Paediatric Chronic
Illness**

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FAMILY MEMBERS' EXPERIENCES OF PCI

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Literature review	7,623	7,345	14,968
Research paper	8,987	9,143	18,130
Critical appraisal	3,929	938	4,867
Ethics section	4,197	4,486	8,683
Total word count	25,033	21,912	46,945

Thesis Abstract

This thesis comprises of two parts aiming to explore family members' experiences of living with a child who has a paediatric chronic illness (PCI) and one part that also reflects on the research process.

The systematic review utilised a thematic meta-synthesis approach to analyse 12 studies, consulting 373 healthy siblings regarding their experiences of living with a PCI. The synthesis resulted in two themes. The first theme was 'Changing relationships' which had two sub-themes: 'Changing family relationships' and 'Changing relationship to self'. The second theme was 'Managing changes' which had three sub-themes: 'Coping and acceptance', 'Support from friends peers and support groups' and 'Negative reactions from others'. Siblings were found to adopt new roles, new skills and eventually develop a new 'prosocial' identity in order to find more socially acceptable ways to meet their needs and the needs of the family.

The research paper explored parents' stories of adjustment to having a son with Duchenne Muscular Dystrophy. Narrative analysis was applied to seven semi-structured interviews of five women and two men. The analysis identified key narrative elements from their stories that were then collated into demographics and contextual data, individual summary stories and three common chapters: 'Investigating and fighting: "Something's not right"'; 'Making meaning of the diagnosis'; and 'Living a normal life'. Parents explored narratives relating to acute and chronic illness, altering their identity from parent to carer and providing a normal and happy life for their son.

Finally, a critical appraisal of the research reflected on the use of qualitative research in health settings and how the associated issues applied to this thesis. Epistemology was used as the lens to view how decisions in the research were made, in particular highlighting the importance of quality, the debate regarding validity and identifying relevant future research.

Declaration

This thesis presents research undertaken for the Doctorate in Clinical Psychology at the Division for Health Research, Lancaster, from (October 2012 until April 2017). I confirm that the work presented in this thesis is my own except where otherwise referenced. The work has not been submitted for any other academic work.

Name: Antoinette Deavin

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There are many people who have made the writing of this thesis possible. First, a special thanks to all the parents who kindly shared their personal experiences. Their honesty brought truth and integrity to the research study. I would also like to thank the paediatric team who volunteered their time and effort in supporting this project, whilst endeavouring to provide the best service for families with paediatric neurological conditions.

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Finally, I will try to put into words the gratitude I have for my family and friends. Thank you, Mum, Dad and Bill for your unconditional love and gentle guidance and support throughout my life. Pete, you are my home that I return to through good times and bad. Despite the challenges we have faced, we have come through the other side stronger. To Arlia and Blake, thank you for being my sunshine and a reminder of why I do the work I do. Also thank you to Lesley, Rosie and Josh for looking after my rays of sunshine whilst I worked on this thesis. To my Grandma, I miss you and I hope I make you proud. To my friends, kindred spirits and soulmates, you know who you are; I thank you for helping me find my place in the world; being understanding when I've had to cancel plans and helping me night and day to finish this behemoth task.

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Section One: Literature Review

**A Synthesis of Children's Perspectives of Living with a Sibling with a Paediatric
Chronic Illness**

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Abstract

Context: Current clinical guidance emphasises the importance of considering the whole family when caring for a child with a paediatric chronic illness (PCI).¹⁻³ However there is a lack of professional guidance and research specific to sibling experience. Both quantitative and qualitative research frequently utilise parental accounts to address family, and particularly sibling experiences, meaning the direct voices of siblings tend to be neglected.

Objective: The meta-synthesis aimed to directly explore the experiences of siblings of children with PCIs to understand their perspectives; in particular what they or others may have done that was found to be helpful.

Results: The thematic synthesis⁴ identified two overarching themes providing new insights. The first theme was 'Changing relationships' which had two sub-themes: 'Changing family relationships' and 'Changing relationship to self'. The second theme was 'Managing changes' which had three sub-themes: 'Coping and acceptance', 'Support from friends peers and support groups' and 'Negative reactions from others'.

Conclusions: The findings outline how changes in family relationships often result in reduced communication and a suppression of healthy siblings' needs. Siblings develop strategies to help them cope with and accept their circumstances, including finding new prosocial ways of meeting their needs in the form of skills and roles they develop.

Keywords: chronic health condition, sibling, meta-synthesis, experience

Introduction

The definition of a chronic illness has frequently been debated,⁵ and therefore requires description to explain how this relates to the literature base and the research question posed by this paper. Here it will relate to a diagnosis in childhood or a paediatric chronic illness (PCI) and is defined as: a) a disease that after diagnosis will be present throughout the child's life i.e. is incurable. This is in order to consider the compounded effects of the disease over time; b) is requiring ongoing care; and c) as having an impact on the child – i.e. adjustments to day-to-day living caused by reduced/altered physical functioning, cognitive impairment or psychosocial consequences stemming from having the condition. It does not have to be genetic, congenital or life-limiting.

The ramifications of these diseases require families to quickly make adaptations, including practical and financial considerations, e.g. attending hospital visits or one parent giving up work to look after the child.⁶ In addition, there is also the emotional toll that families often experience from living with a child who may frequently be unwell or in pain, or struggling to adjust to their condition.^{7,8} These experiences impact on the quality of life of the individuals who have the illness and family members⁹. This is recognised in current professional guidance in the recommendation of family based care and interventions.^{2,3} However, within these guidelines the discussion of siblings is marginal. This is also true of the representation of siblings in research, which generally focuses on overall family experiences or prioritises parental accounts of sibling experiences.¹⁰⁻¹⁶ The paucity of qualitative research also tends to focus on the family experience as a whole.¹⁷⁻²⁰ Consequently the voices and subjective experiences of siblings are overlooked. It has been commented that the effects on siblings has received little attention⁷ and it is difficult to make clinical recommendations based on the available research.²¹

Previous research frequently suggests increased levels of sibling distress compared to peers,¹⁶ however results have often been perceived as mixed with contradictory findings.¹⁵ In their review Sharpe and Rossiter¹⁵ reiterate Cuskelly's statement of how previous research into psychological adjustment of healthy siblings leaves the reader with "...the overwhelming impression of contradiction and confusion."^{22(pp111)} Their meta-analysis revealed that whilst siblings were found to have overall small significant negative effects on their psychological functioning, further analysis demonstrated significant heterogeneity of their effect sizes, which may have contributed to the confusion. On further investigation this appeared to be as a result of the differences in variables explored; with siblings demonstrating higher levels of internalising (i.e. anxiety) than externalising (i.e. aggressive) behaviours. They also noted how historically studies had also shown long-term benefits for siblings' externalising behaviour, for example increased levels of compassion.

It could be argued that the effects of PCIs on healthy siblings may be considered less clinically urgent or necessary to investigate than those immediately experiencing or managing the condition i.e. patients and parents, because the impacts appear to be small, potentially positive in the long-term and unclear. However the field of paediatric clinical psychology, theory and guidance acknowledges the importance of considering the needs of healthy siblings,^{8,23,24} as some do experience distress and should not be overlooked. In addition, recent quantitative research has identified a range of factors potentially contributing to negative outcomes or 'maladjustment'²⁵ for siblings of children with PCIs. A meta-analysis by Vermaes, et al¹⁶ found siblings of children with PCIs presented with lower self-esteem and increased internalising and externalising difficulties i.e. higher levels of depression or aggressive behaviour, than their peers. The authors hypothesised that these findings stem from a systemic and relational cause; for example, siblings do not ask for the

attention they need as they do not want to burden their parents further,^{26,27} or they have learnt that their parents do not have the time or emotional resources to effectively support them, which over time causes them to devalue their own needs. Vermaes et al¹⁶ also found that for older siblings, when the disease was life threatening and/or more intrusive they had an increased risk of psychological difficulties.

The range of systemic variables may also account for some of the variation of findings as healthy siblings' experience is partially dependent on the actions of others, in particular their family members.²⁸ The other issue that may have affected previous results is siblings' ability or openness to communicate their experience, either due to a lack of direct consultation, through the use of parental measures of wellbeing¹¹ or through their own desire to provide socially acceptable answers rather than their own feelings.²⁹ Previous findings have shown that siblings do not want to burden their families further,^{26,27} so this may cause some to answer questions in a manner that is unrepresentative, especially if parents are present for their responses.

Arguably methodology could also have influenced these results. The majority of research into healthy sibling experience is quantitative which had an a priori framework. While this has provided useful information, each study will have focused on different variables and/or measures for wellbeing or distress, which may make some studies difficult to compare and may also decontextualize the results. This argument would be consistent with the finding of Vermaes et al¹⁶ which highlighted the relational nature of their findings. A first-hand subjective account from siblings allows them to explore these variables whilst giving them context and connections. In addition they would be able to discuss anything else they may view as important to their experience, meaning that their viewpoint is privileged.

The study of subjective experience is the realm of qualitative research,³⁰ which has been utilised to understand siblings' experiences; however this has mostly been from a whole family perspective, e.g. all family members are interviewed and analysed together. This approach has been used to explore treatment demands,²⁸ family dynamics,²⁹ perceptions towards genetic testing³¹ and general psychosocial impacts.²⁰ While this may help to provide an understanding of family perspectives, sibling's views may be influenced by other members of the family, therefore failing to identify the distinct voices of the siblings. Many studies include siblings of younger ages e.g. six years old²⁰ and whilst having an adult present and/or their perspective included in the research ensured initial insights into a difficult to engage sample this may mean that siblings provide socially desirable responses.²⁹ The benefit of this approach is that siblings at least have a partial voice which has provided insight into reactions on diagnosis, education on the chronic illness, sibling's involvement in care, sibling relationships, fears and the overall impact of the disease on the sibling.²⁰

To further advocate siblings' experiences this synthesis focuses on siblings' direct reports. For some specific diseases, for example paediatric cancer, the sibling experience is investigated with siblings directly and is generally better documented and understood as demonstrated by three reviews.³²⁻³⁴ However it has been argued that a non-diagnosis based review would focus on the psychosocial commonalities that would help to improve care for families.⁷ A more generic review with the intention of unifying siblings' voices across diseases has not yet been undertaken. Thus, the aim of this meta-synthesis of qualitative research was to fill this gap and address the research question: What are the experiences of siblings of children with a chronic and non-communicable physical health condition?

Method

The Database Searches

Four databases were used to identify articles relating to the experiences of siblings of children with chronic physical health conditions: PsychInfo, CINAHL, PubMed, and Academic Search Complete. The speciality and breadth of subjects, potential audience and preferred methodologies addressed by each database was considered when deciding which combination would provide the best opportunity for capturing all relevant articles. For example, CINAHL was chosen as a global database reporting on research related to nursing and other allied health professionals, which is targeted to a more medical perspective than PsychInfo, which indexes articles relating to psychology.

As all four databases publish articles for different audiences and consequently use different terms, it was necessary to generate a thorough list of key words to maximise the retrieval of relevant articles. Initially key words were generated using four strategies: firstly, by identifying APA recommended vocabulary; secondly, by using each database's specified term generator (e.g. the Thesaurus in PsychINFO); thirdly, by identifying possible key words from relevant articles; and finally reviewing the words with the research team. This led to a final set of search terms that could be used adaptively according to the database (see Table 1).

[Insert Table 1. here]

'Chronic disease/illness' has been shown to have varied meanings.⁵ For the purposes of this review 'chronic' has been defined as a condition lasting longer than 3 months and is currently incurable, which is in accordance with the frequently cited characteristics of length and severity present in the research literature.⁵ The research question also specifies that the disease must be a physical health issue and non-contagious. Experiences relating to physical injury (i.e. brain injury), mental health or addiction were not included.

All the databases used filters for language (English only) and type of article (scholarly/peer-review/article). The methodology filter was also used in PsychInfo, which is a feature that is not available in the other databases. None of the searches were restricted by the publication date. Inclusion and exclusion criteria were carefully selected in order to address the reviews aims. The final systematic search was carried out in April 2016, which resulted in 12 articles being found relevant for the review. A summary of the initial October 2014 and final April 2016 review process is outlined in Appendix 1A.

Inclusion and Exclusion Criteria

Whilst the focus of the synthesis was on healthy sibling experiences, the PCI that affected the unwell sibling must have been physical, chronic, non-communicable, incurable, require ongoing care and have an impact on the affected child. The disease did not need to be genetic, congenital or life-limiting. In order to examine healthy siblings' perspectives only studies that reported using qualitative approaches were included and must have elicited first-hand accounts, in the form of interviews or written responses e.g. open ended questionnaires. Studies that included interviews with other family members were included when the sibling's experiences could be identified independently in the findings i.e. as a separate theme. This was also evidenced by participant quotations.³⁵ Those reporting using a mixed methodology were also included to maintain the breadth of the review and represent the range of studies available in this topic area. The content of the interview had to discuss the siblings' experiences as children (under the age of 20 years old to account for all childhood experiences and also provide a sufficient number of articles for the review), although the age of the participant at interview was unrestricted to allow an overview of experiences and reflections over time. Articles that targeted disease-specific experiences or procedure (e.g.

being a donor for a hematopoietic stem cell transplant)³⁶ as part of the research question were excluded.

The Critical Appraisal Skills Programme³⁷ qualitative checklist was used in conjunction with Kearney's³⁸ typology and application criteria (see Appendix 1B and Table 2). CASP scores were assessed by the researcher and an affiliate external to the research team and then averaged in order to present an unbiased score. These measures are presented to help the reader evaluate the studies, in particular each study's quality and potential relevance to the topic area and the review, whilst highlighting methodological issues.³⁹

[Insert Table 2. here]

Quality was not used to exclude articles. This is an accepted practice, especially when there is a small quantity of papers being reviewed or the quality of the studies in the topic area needs to be highlighted.⁴⁰

Overview of the Selection Process

The selection process had five steps (see Figure 1). Firstly the key terms and filters for each database were applied and then the titles and abstracts were scan read for general fit to the research question. Articles thought to be relevant were flagged for a more detailed review, which involved a more thorough assessment of the inclusion criteria. Particular emphasis was placed on identifying whether articles utilised qualitative methods and related to direct sibling experience. Simultaneously, a review of the inclusion and exclusion criteria were reflected upon within the research team, which lead to the exclusion of papers relating to cancer. Some forms of cancer are recognised as being acute e.g. acute leukaemia, and treatment in other forms can lead to long-term remission of the symptoms, which was felt to make this disease sufficiently different from the sample to exclude childhood cancer from this review. Lastly, a

more thorough reading of the articles and the application of the new inclusion criteria produced the final sample of 12 articles. Duplicates were removed.

[Insert Figure 1. here]

Summaries of the aims, key features and themes of the final sample are provided (see Table 3.).

[Insert Table 3. here]

Meta-synthesis

The systematic review aimed to explore the reported experiences of siblings of children with chronic physical health conditions, to gain their subjective perspective and consider what they may have found helpful. The chosen thematic synthesis⁴ approach addressed these aims by utilising concepts from a range of qualitative methods, predominantly thematic analysis,⁴¹ meta-ethnography⁴² and grounded theory⁴³ to inductively compare findings reported in the primary qualitative studies.

The initial stage of analysis generates 'descriptive themes' from the data, which takes the codes and finds similarities between them. These descriptive themes are then compared and contrasted to each other, and the initial codes, to provide 'analytical themes'. These collate similar concepts together whilst providing links to each other and any subthemes. Through this process a hierarchy is achieved, whereby concepts evolve from data. This is akin to 'third order interpretations' in meta-ethnography, whereby an overarching model is achieved.⁴⁴ The use of coding and the development of descriptive and analytical themes are synonymous to reciprocal translation⁴² whereby comparison between primary sources identifies similarities in the findings.^{44,45} The benefits of this approach are that the analysis remains faithful to the

original articles, whilst allowing a transparent deconstruction (coding), synthesis of the data (descriptive themes) and translation of the concepts (analytical themes).⁴

The analysis is similar to thematic analysis⁴¹ in that the findings from each article are coded line by line. The discussion sections were also reviewed to check for new information, however codes were only included when it was grounded in the data rather than conceptual interpretations. The codes from each paper were colour coded and iteratively arranged into descriptive themes representing similar concepts. Similarities were then identified between themes, which produced the two final analytical themes. Rigour was maintained by the comparison of the analytical themes to the primary articles, ensuring the findings were grounded in the original data. Throughout, notes were taken regarding nuances of the codes, in particular the differences and similarities. Each stage has been documented and examples can be seen in appendices 1C-1F. Epistemological reflections and applications can be found in the critical appraisal paper of this thesis. The synthesis is outlined in written format below.

Findings

The systematic review identified 12 papers which yielded two themes providing insight into the variety of experiences of healthy children who have siblings with a PCI: '*Changing relationships*' and '*Managing changes*'. '*Changing family relationships*' occurred between all members of the family as a result of their changing circumstances and their own emotional responses. This led to a 'changing relationship to self' for siblings as a result of meeting the changing needs of the family and consequently acquiring new roles, skills and responsibilities which frequently became part of their long-term identity. Managing these practical and psychological changes requires siblings to develop ways of coping, accepting and adjusting, which is often influenced by external support or their fear of negative reactions from others.

A visual representation is provided (see Figure 2). The findings are discussed alongside theory, clinical practice and current guidance.

[Insert Figure 2. here]

Theme One: Changing Relationships

This theme outlines siblings' experiences of changing relationships and includes two subthemes: '*Changing family relationships*' and '*Changing relationship to self*'.

Subtheme One: Changing Family Relationships

This subtheme outlines healthy siblings' perceptions of changes to family cohesion, changes to their relationships with their parents and with their unwell sibling.

An altered sense of family cohesion since the onset and diagnosis of their sibling's disease was reported in five of the 12 articles. Healthy siblings expressed views representing both a sense of increased and decreased family cohesion, demonstrating the subjectivity and complexity of their experience. When positive experiences were reported, siblings described a united family environment,^{46,47} with the whole family contributing to their sibling's care,^{47,48} which they felt brought them closer.⁴⁷ This was described as giving the family something in common,⁴⁹ which they shared and talked about together.⁴⁸ The practice of caring was not only focused on the unwell sibling, but led family members to be "nicer to each other",^{49(pp434)} meaning family and individual relationships felt stronger.⁵⁰ Participants felt this level of closeness and bonding was different to other families that did not have to manage a PCI.^{46,49} In contrast, Read et al⁴⁸ emphasised how the division of family activities, due to their sibling's disability, could reduce family cohesion. One example was having to divide the family when their sibling had an hospital admission. Bellin et al⁴⁶ noted how

emotions and reactions within the family were related to the uneven distribution of family resources, which then can impact upon relationships between individuals.

Findings from 10 papers addressed the changing relationship between parents and their healthy children. These highlight the changes in the amount and quality of parental attention given to each of the children,^{47,49-55} the perceived change in parental expectations of the healthy sibling (e.g. helping to care for unwell sibling),^{46,53} the recognition by siblings that parents are worried and their behaviour changes as a result^{47,49,50} and, lastly, healthy siblings then notice a reduced level of communication with their parents.^{47,53,56} These findings are detailed further below.

The majority of reports from healthy children describe a shift of parental attention to their sibling. Healthy children often felt jealous and resentful of the amount of time, protection and 'special treatment' parents bestowed upon their siblings,^{49,50,55} especially when their sibling appeared to be 'doing well'.⁵³ Although, siblings' acknowledged the increased attention was necessary.⁵⁰ In contrast, there were two examples of siblings stating there was no preferential treatment towards their brother or sister.^{46,50}

The sentiments of resentment and jealousy have been discussed within the paediatric psychology literature when the needs of the unwell sibling preoccupy the thoughts and time of parents.⁸ Parents feel that this behaviour is practically necessary but also find it challenging to balance the needs of the illness and the family²³ and are reported in many cases to not have the resources to meet both⁸.

Healthy siblings from two studies described what has been termed as 'parental silence', whereby parents do not talk to the healthy sibling about the illness in order to protect them from the difficult aspects of the disease.^{53,56} This meant parents sometimes did not know how

much siblings knew about the illness.⁵⁶ Participants in the study by Wennick and Huus⁴⁷ discussed how siblings would return the silence as their parents had 'enough to worry about'. This resulted in a reciprocal silence between parents and the healthy sibling.⁵³ This is potentially problematic for family relationships as some participants reflected that openness might provide the antidote to family breakdown.⁵⁵

The relationship between siblings appeared to be equally complex, eliciting a range of emotional responses. For example, the increased time spent with siblings meant for some they felt they had a closer relationship,⁴⁸ describing their sibling as a friend,⁴⁶ whilst others described continuing to have a 'normal' relationship.^{46,51} In contrast six studies found that siblings recognised negative changes in their sibling relationship, describing it as a loss⁴⁹ or distancing.⁴⁸ These were associated with changes in their sibling,^{50,55} for example becoming more prone to mood swings.⁴⁷

The changes in family relationships described by siblings in these studies could be due to changes in the implicit rules of the family resulting from the diagnosis of the PCI.²³ These rules encompass ways of relating to other members of the family in order to meet the needs of the group. In particular they highlight the roles within the system (e.g. becoming the carer of the family) and the acceptable behaviours. Families hold varying beliefs about how to view and approach illness as a unit and as individuals.¹⁸ For some the illness is all but ignored in attempts to create a sense of self that is unaffected. For others they incorporate the illness in the new family identity to the point where it may become the sole focus, causing the family to define themselves based on providing care.²³ The sense of increased family cohesion some healthy children mentioned may therefore result from healthy siblings identifying with the family system's roles and agendas, causing them to feel included. However this has repercussions for siblings who do not fit with the 'acceptable' assumptions of the family

system. For example, one particularly common and potentially damaging issue is the reciprocal silence that occurs when familial beliefs promote a lack of discussion about the illness, in order to protect others or because it is too painful (i.e. 'our family does not talk about how we feel about the illness'). This causes siblings who want to discuss their emotions to feel as if they are threatening the family, perhaps becoming a burden, forcing them to become isolated and unable to address their needs. These internal system boundaries,²³ set up to protect each other, consequently reduce communication and cause members to become distant at times when they need each other the most. Given that the structure and hierarchy of the system is also governed by parental beliefs, it is important to remember not all members have equal power and voice. With healthy siblings being both children and perceived of as being in less urgent need, their voices may often be ignored and suppressed by the adults around them.

Subtheme Two: Changing Relationship to Self

This subtheme synthesises the voices of siblings from all 12 articles. As a result of the changes in the family, healthy siblings became more aware of their role within the family and felt forced to change, which affects how they view themselves in the long-term.

All the articles acknowledged the changes occurring within the family had an impact on the emotional experiences of healthy siblings. The findings from two studies^{46,50} noted there were positive impacts experienced by siblings including a positive sense of self and their life experiences. However, the majority of the findings discussed the overwhelming sense of negativity siblings experienced. This variation of experience is represented by one adolescent sibling's description of an "emotional rollercoaster".^{46(pp204)} In Derouin and Jessee's article⁵⁰ some of siblings went so far as to cite themselves as the "most unhappy member of the family"^(pp142). In some instances siblings discussed the link between their emotions and their

experiences. For example identifying that their emotions resulted from the empathy they felt towards their sibling. They were often sad that they had the disease or became angry when others bullied their sibling because of the disabilities arising from the PCI.^{46,55}

Sadness was explicitly noted as being the most frequently experienced emotion by siblings in the paper by Hutson et al.⁵³ However 'worry' was the most commonly cited emotion across the articles. Worrying about their sibling's health and wellbeing was pervasive.^{49,50,53,55,56}

Some recognised that worrying had begun to interfere with other areas of their life, particularly causing problems with concentration at school.⁵⁶ Siblings also worried about the uncertainty of their future.⁵⁵ Dependent on the type of illness their sibling was diagnosed with, participants reported anxiety about the development and transmission of the illness, specifically how it might affect their health^{49,52} and whether they would be a carrier for their children.^{48,53}

Other emotions reported in the literature were feelings of "survivor guilt",^{46,52,55} loneliness resulting from familial and peer isolation^{46,53,55,56} and jealousy arising from the reduced level of parental attention.^{48,52}

Emotions like jealousy, unhappiness, loneliness and anxiety stem from the changes to the family dynamics and link to siblings' needs to feel supported, cared for and safe. Williams et al.,⁵⁷ used structural equation modelling to examine the interrelationships amongst variables affecting siblings and mothers of children with chronic illness and found that mood and experience of support affects siblings' self-esteem. This finding highlights the importance of meeting siblings' emotional needs in order to maintain their self-esteem.

Findings from Read et al⁴⁸ described the difficulties participants experienced in balancing home demands, including parental and sibling expectations, whilst their own needs were

often overlooked. Given siblings' perceptions that their needs were not as important as their siblings, it is not surprising participants in five articles stated they did not discuss their feelings with their family for fear of their parents becoming angry, increasing their parents' worry⁵² and adding to their burden.⁵⁶ In addition, siblings discussed avoiding conflict^{53,58} and pleasing family members to gain approval.⁵³ The desire to balance the needs of the family, leading to avoidance of communicating their emotions, appears to provide the foundation of siblings' experiences of not recognising,⁵³ deprioritising^{52,58} and suppressing their own needs. This process is identified as the sibling's desire to be emotionally self-sufficient.⁵⁸ Bradford's⁸ hypothesis of "sibling self-sustainability" due to reduced parental attention is synonymous with the synthesis findings of emotional self-sufficiency. "Self-sufficiency" is thought to be promoted by reduced communication from parents in order to protect the sibling. Research also suggests that siblings regularly do not have direct contact with a health professional, compounding the lack of communication and need to be self-sustaining.⁵⁹

In all of the articles healthy children described the new roles and skills they had developed since the onset of their sibling's condition. In contrast to their emotional experiences, these were often described more positively. It could be that the transition to a new, more positively understood role within the family allowed the children to meet their needs without expressing them. Participants described how this felt like a role transfer and children found themselves embodying roles associated with older adolescents or adults.^{48,55} Their new experiences provided them with specialist knowledge of the condition and the responsibilities placed upon them caused them to develop a caring role, which became a part of their identity.^{46,47,55} These skills and tasks ranged from general caring for the family e.g. housekeeping, supervision of their sibling,^{47,49} helping with leisure activities,⁴⁸ to identifying, assessing and managing their

sibling's symptoms.^{48,51,56} The findings of Brennan et al highlight the extent to which the caring persona was internalised and supported by others, demonstrated by friends calling the healthy sibling "Mum".^{58(pp819)} One sibling noted how their experience had made them more compassionate towards other people generally.⁵⁴ Siblings described their caring role with pride, noting the acceptance that came with the role, but also recognising the limitations.^{55,56} The caring role was discussed as increasing over time,⁴⁸ with few breaks,⁵³ and some discussed the shame they experienced when they did not meet their own expectations of the role.⁵²

The acquisition of specialist knowledge, skills and internalised positive roles appears to have provided siblings with an identity that makes them feel unique and special,⁴⁹ which seems to fulfil some of their basic social needs. These skills can be transferred to other areas of their life, for example acting as the "glue" in their relationships⁵⁸ and some acknowledged their experiences shaped their career choices e.g. becoming a family support worker or nurse.⁴⁸

Theme Overview

In summary, the synthesis has found that the majority of siblings alter their behaviour in order to meet the needs of the family and also their own. They take on tasks and skills that include them in the current caring goals of the family. With time and positive feedback, the behaviours that are reinforced become roles within the family and an internalised positive characteristic of their identity. These characteristics are synonymous with prosocial characteristics listed in the clinical and research literature.^{8,23,24,58,60}

The combination of emotional self-sufficiency and increase in prosocial behaviours may cause parents and professionals to perceive the sibling as functioning and thriving. Research has shown parents underestimate the emotional responses and needs of healthy siblings,⁸

which may explain why they are frequently overlooked clinically, in research, and why there is a lack of professional guidance regarding the treatment of siblings through the care process.^{59,61} This is exemplified by the policy statement released by the American Association of Pediatrics, which highlighted the importance of patient and family centred care, but did not mention any specific risks or recommendations for siblings.² Whilst siblings adapt their behaviour to become more prosocial to meet their basic social needs, they are still experiencing high levels of distress. A meta-analysis by Vermaes et al¹⁶ found that siblings had significantly higher levels of internalising problems (e.g. depression and anxiety) than comparisons, which support the current findings and suggest the rates of these issues are higher than the general population.

Theme Two: Managing Changes

This theme outlines how healthy siblings cope with and accept the changes that result from their sibling's disease. This includes navigating the impact to their broader social networks. It consists of three sub-themes: *'Coping, acceptance and adjustment'*, *'Support from friends, peers and support groups'* and *'Negative reactions from others'*.

Subtheme One: Coping, Acceptance and Adjustment

Adjustment over time was a factor described in five articles as having an influence on coping and acceptance. Siblings described the impact of the illness as getting easier over time.⁴⁹ They commented that it was harder when their sibling was first diagnosed⁴⁶ but, where there was a slow progress of the disease this allowed coping and acceptance.⁴⁸ Time was felt to create space to make sense,⁴⁶ allowing the process of accommodating to their sibling's decline.⁵⁶ The importance of adjustment over time and space to process the changes was not identified in any of the papers but became apparent through this synthesis. This perhaps links

with child development and their ability to understand all aspects of the condition. However this was affected by the amount of information they had received dependent on their age and the views of the adults around them. This is important to consider as the reports from siblings in this review suggest there is a balance to be struck between timing and transparency of information to provide optimum conditions for adjustment.

Participants described the evolving process of accruing of information.⁴⁸ The findings highlight the importance of providing siblings with information about their sibling's disease.^{48,53} Some commented that a lack of information, particularly relating to symptom management, led to fear.⁴⁷ Parents were noted as the main source of information⁴⁸; however when they were unwilling to discuss the disease siblings would find alternatives, for example support groups or the internet.^{48,53} For some they felt knowing earlier would have been helpful and changed their sibling interactions, whilst others acknowledged they did not want to know,⁵⁵ often because they were afraid or choosing to not know provided them with a sense of control.⁴⁸

When siblings had access to information they started changing their attitudes towards the disease and its impacts.⁵⁵ Siblings described how they developed insight and understanding of the broader situation⁵⁰ and empathy for their family member's experience.^{48,49,51,56} Understanding sometimes helped them to cope with the imbalance of attention between themselves and their sibling.⁴⁷ For others, they still felt jealousy or neglect but could tolerate and accept the necessity of the situation.⁴⁸ These findings suggest that siblings would prefer to have increased levels of information earlier, relating to the disease and its impacts, which may help build their understanding and empathy, helping them to tolerate and accept the situation and their feelings. This is supported by findings showing strong associations between increased sibling illness knowledge, positive sibling attitude toward the illness and

sibling behaviour.⁵⁷ However it is important to note that knowledge may be impacted by the age of the sibling, with older children having more illness knowledge.⁵⁷

Half of the articles had findings relating to behavioural and cognitive strategies that allowed healthy children to cope with and/or accept having a sibling with a PCI. These appear to be either distancing or integrating techniques. Distancing techniques included avoidance,⁴⁸ distraction,⁵⁰ physical and temporal compartmentalisation.^{53,58} These were often described as “getting on with it”, “focussing on the day-to-day”, “living in the present” and removing themselves from difficult situations, which would include pursuing external interests.⁵⁵

Children described themselves as being different dependent on the environment in order to keep their home and ‘other’ life separate and maintain some level of “normality”. These strategies were used simultaneously alongside humour, having a positive outlook⁴⁸ and talking about their feelings⁵⁰ to help them accept their experience as part of life. Bellin et al⁴⁶ noted how siblings deemed “normalising” as a step toward acceptance and integration of the disease into their lives.

Little is documented on the specific coping strategies used by siblings; however the techniques identified in this meta-synthesis are comparable to those used by children with PCIs. A narrative review by Compas, Jaser, Dunn and Rodriguez⁶² categorised strategies into three types: active, accommodative or passive coping. Active coping is defined as an individual’s attempts to directly influence or change the source of stress. Accommodative coping is defined as an individual’s attempts to adapt to the source of stress through “...reappraisal, positive thinking, acceptance or distraction”.^{62(pp6)} Passive coping includes cognitive and physical avoidance of the stressor. The review found accommodative coping to be the most effective form of coping, with mixed findings for active coping and poorer outcomes for those who utilise passive coping techniques. If the findings from Compas et al

⁶² are applied to healthy siblings in this review then they are using a combination of efficacious and in-efficacious strategies, with the latter being more frequently reported.

Subtheme Two: Support from Friends, Peers and Support Groups

Support was felt to come from family, friends, teachers and support groups.^{48,56} The findings from Gallo et al⁵¹ detail the internal conflict healthy siblings have when telling other people about their sibling's illness and seeking support. Participants reported all options, from feeling comfortable revealing the illness to others (though some would be selective) to others who did not want people to know.

Friendships were discussed in five of the articles,^{46,48,51,53,56} with siblings stating they felt able to have good friendships.⁵¹ The articles reported that friendships were often supportive, mature and inclusive,⁴⁶ providing both practical and emotional support, with space for the healthy sibling to relax and be themselves.⁴⁸ Some of the findings from Malcolm et al⁵⁶ described friends as being the most supportive, understanding and accepting relationship. In contrast, participants from the study by Hutson et al,⁵³ commented that their peers were not supportive and feared that their friends would reject or ignore them due to their siblings.

Support groups were mentioned as a form of support that siblings felt helped them to overcome their isolation and aid their adaptation,⁴⁸ allowing them to share their experiences and knowledge with each other.⁵⁰ They enabled connections to others with shared experiences, expanding their social network and offering them more opportunities to make sympathetic friends.⁴⁹ Groups were valued and enjoyable; however they were too infrequent and/or far apart.⁵⁶

Subtheme Three: Negative Reactions from Others

Findings from seven studies identified the reactions of other people as a worry for siblings. The majority of the concern regarded others reactions to their sibling.⁵⁶ Children were worried their brother or sister might be teased, rejected by others and become upset,^{46,48,51} even so far as for them to be concerned about prejudice from the community.⁵⁶

Children described being scrutinised because of their sibling⁴⁶ and becoming embarrassed by the treatment of their sibling in public.⁴⁹ At times they struggled with the feeling of being judged by others⁴⁶ and felt different compared to their peers.⁵⁸ This was compounded by other differences that siblings found embarrassing, for example, the family experiencing financial difficulties due to the cost of treatments.⁵³

Some children reported their thoughts about the beliefs and actions of others, stating “it’s none of their business”,^{51(pp23),55} or feeling frustrated because others do not understand^{46,55} but also identifying ignorance as a rationale for their negative behaviour.⁵⁶ These experiences led siblings to desire that others would be more accepting and tolerant.⁵⁶ Consequently others’ reactions may contribute to siblings’ uncertainty about asking for help; meaning they have limited avenues for seeking support where they can feel safe and alike.

Theme Overview

In summary the thesis found that siblings have many strategies and resources that they use in order to manage the changes that result from their sibling’s disease. Paramount was the access to relevant, timely and accurate information. Siblings described how they adjusted over time as they assimilated new knowledge and began to adapt to their circumstances and accept their feelings. The timing and appropriateness of the information needed to be considered in relation to the age of the sibling so that they had their questions answered but were also not overloaded or given information that they might find frightening. Siblings

described how timely information led to greater understanding and empathy towards family members, which helped them to manage and tolerate their own emotions; in particular their sense of jealousy regarding the imbalance of parental attention.

Siblings used a variety of cognitive and behavioural techniques, which have been identified in the paediatric patient literature. This review's findings suggest that whilst siblings use active and accommodative coping strategies, they rely more on passive coping e.g. distancing, which has been linked to poorer outcomes.

Siblings described contrasting thoughts about sharing their experiences with others. Whilst they acknowledged that it could be helpful to have someone to talk to, they also feared being rejected due to their sibling's illness. Support groups with others in a similar situation were therefore seen as helpful, but were infrequent. It may be that this kind of external support became a means by which siblings could utilise more accommodative coping strategies to address both their practical and emotional needs i.e. a need for information and a space to discuss how they feel. These supportive relationships have been found to be helpful for siblings of children with cancer by allowing them to access information to help them make sense of the situation, provide opportunities to communicate how they feel, seek reassurance, receive attention to feel valued and maintain self-esteem and a sense of self independent of the illness.⁶³ By utilising a systemic structure it is possible to explain the necessity of these relationships as providing another external system to the family, the roles and agendas of which are similar to the needs of the sibling. However this is complicated by the fear of the negative reactions of others, restricting their possibilities for support and potentially leading them to moderate help seeking from others outside the family.

Conclusion

These findings embody the subjective range and complexity of healthy children's experiences since the onset of their sibling's symptoms. In particular, this review gives voice to healthy siblings' concerns regarding their inability to disclose their emotions and the necessity to develop skills and roles that become internalised in order to meet their own needs, by constructing a positive identity that is concordant with the family's needs. This sets up an ultimatum reflected in both the family and friendship systems i.e. suppress your feelings or risk burdening others or them not understanding.

The alternative of expressing their needs is further hampered by the widening gaps in communication with their support systems, perhaps leading to the clinical symptoms often reported as maladjustment i.e. mood changes, rebellion, attention seeking and somatic complaints.²⁵ Some of these have been well documented previously,^{18,20,28,64} although not necessarily from the sibling's viewpoint. The absence of the sibling narrative has meant these connections described have not been identified by previous quantitative meta-analyses.

Whilst there is some evidence of considering the clinical importance of siblings' experiences,^{8,23,24} it has been recognised that they have been overlooked within the research literature.^{7,21} This may be as a result of siblings appearing to be functioning well and in a desired manner to parents and professionals, combined with their fear of expressing how they feel.

Clinical Implications and Future Research

Current good practice guidelines identify the need to consider the entire family when working with families with a PCI.² However there is little guidance globally as to how this should be implemented with siblings.^{2,3} This synthesis points to several areas for clinical intervention, including early intervention strategies and recommendations for therapists.

The findings emphasise siblings' desire and ability to conceal their needs and emotions. Consequently, sharing the findings from this review with siblings, parents and professionals would help them to be mindful of their needs and monitor the levels of communication or avenues for support available to the sibling. It may be that broader systems need to be involved, for example social services, in order for parents to have the necessary level of resources to support both children.

Siblings should have access to a health professional to have their questions regarding the disease answered, which may reduce anxiety regarding treatments and their uncertainties about the future. Age appropriate leaflets could also normalise the experience for the family. Where possible, families should be encouraged to speak together about the illness with siblings and allow them a forum to express their concerns and help them understand and to feel heard and included. Keeping channels of communication open would also allow parents to problem solve when necessary. Support groups and other positive events, for example charity work or sibling days²⁴ were also found to be helpful but were often too far away for siblings to access regularly. It may be that more online resources and ways of communicating could be developed and encouraged, e.g. sibling forums, buddy systems or websites such as www.sibs.org.uk.

Whilst siblings are proficient at creating a positive prosocial identity, their ability to identify other positive self-attributes are lower than siblings of healthy children.¹⁶ Consequently families and professionals should liaise to ensure that siblings have space and support to develop a positive identity outside of the caring role. At home this could be implemented by ensuring that children are encouraged to have interests and their independent achievements are recognised. More broadly this could be supported by actions to reduce negative reactions

within the general population, possibly due to a lack of understanding. This could involve providing education days at school about chronic illnesses.

Last, it may be necessary for therapists to be involved in the care of the family or the sibling. The limited amount of current guidance recommends a systemic approach to care, which would fit with family therapy or systemically informed therapy.²³ Initial steps to share systemic formulations with families and professionals would identify family dynamics, elucidate siblings' position in the system and highlight potential problems and solutions. The benefits of further therapy in this approach would be providing a space for the family to communicate and promote cohesion rather than isolation, whilst identifying the specific needs of the sibling. Narrative therapy could be used with the sibling to broaden alternative stories about their identity and help them construct a fuller positive narrative about themselves independent of illness.^{23,65} Equally cognitive behavioural therapy⁶⁶ could be utilised to incorporate more accommodating coping strategies e.g. positive thinking,⁶² or help siblings to challenge the core beliefs about themselves (e.g. I am only worthwhile if I help others) that could continue into adulthood.

Potential Limitations of the Study

As a qualitative article the findings do not aim to be valid, reliable and generalisable, as in quantitative research, but provide insight into the experiences of a specific group, which may stimulate new ways of thinking. Consequently it is important to acknowledge the characteristics of the articles, and who they represent, in order for the reader to consider the influence of context on the findings. Some of these have been documented in Table 3. Whilst the synthesis represents the voices of siblings the sample is small and therefore only includes a small range of cultures, disease types, potential age ranges of siblings and research quality. For example, the sample is predominantly of Western cultures and health systems i.e.UK,

America, Canada and Sweden. The small sample also means that a narrow range of diseases are represented. The only diseases to be present in multiple papers are paediatric diabetes and cystic fibrosis. Ten of the papers also focussed on one particular disease rather than multiple similar diseases which further restrained the range of diseases represented. The inclusion and exclusion criteria have also influenced the range of diseases represented. In particular three types of studies were excluded. These were studies that 1) represent very specific experiences e.g. illness specific procedures; 2) could be potentially treatable e.g. paediatric cancer and 3) related to physical injury e.g. head trauma. In addition, the studies often fail to identify the age of the unwell sibling or the current stage of their prognosis.

The small final sample may also be impacted by the researchers in the field. For example it may be that researchers lack the resources to investigate or that they prefer utilising a quantitative methodology to explore the phenomena that families experience. The latter is potentially supported by the frequent presence of mixed methodologies and the relatively low quality of the sample (i.e. a sample CASP mean of 18.9 out of 30 and range of 13- 23.5). Furthermore, when using Kearney's typology and application criteria studies were often found to be descriptive, whereby data is placed into categories with little explicit analysis. Consequently this synthesis has highlighted the need for more qualitative research of healthy siblings' experience and of a higher quality. Future research might focus on how culture may influence family dynamics including elements of communication, narratives regarding illness and gender roles.

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Tables and figures

Table.1

A Table of Search Terms Used for the Meta-synthesis Database Searches

Search terms	Boolean terms: 'OR' used within row and 'AND' used across rows
Sibling	brother, sister, kin
Experience	experien*, cop*, resilien*, manage*, deal*, respon*, adapt*, adjust, understand*, impact, well-being, wellbeing, effect*, thought, think, feel, perspective
Chronic non-communicable physical health condition	chronic health condition, chronic physical health condition, chronic disease, Muscular Dystrophy, Cancer, neoplasm, Asthma, Cystic Fibrosis, Diabetes, Arthritis, life-limiting, fatal, terminal, congenital, genetic, non-communicable, organic disease, non-infectious disease, autoimmune disease, kidney disease, heritable, patient, palliative
Qualitative	qualitative, interview, IPA, grounded theory, subjective, quot*, narrative*, them*, audio, discourse, phenomenon*
Not to be included	autism, ASC, psychosis, psychotic, schizophrenia, bipolar, Alzheimer, attention deficit disorder, ADHD, anorexia, bulimia, HIV, drug

Table 2.

Summary of the Primary Studies' Aims, Themes, Quality Scores and Typology

Reference	Study aims	Themes	(CASP) Quality assessment (Score out of 30)	Typology and application (Kearney, 2001)
Bellin, Melissa H. Kovacs, Pamela J. Sawin, Kathleen J.	"To raise awareness about the risk and protective influences present in the lives of siblings of youths with SB so that resources may be mobilised to decrease vulnerability and bolster resilience."	<ul style="list-style-type: none"> • The rewards and consequences • The journey toward acceptance of SB • The emotional climate of siblings • Qualities of the social environment of siblings 	21	A priori and descriptive
Brennan, C., Hugh-Jones, S., & Aldridge, J	"To investigate the experiences of siblings of children with life-limiting conditions and how they cope with those experiences."	<ul style="list-style-type: none"> • Protection of sense of self • Compartmentalising life • Self as glue in relationships • Positioning in the adult world 	22.5	Shared pathway or meaning
Derouin, D., & Jessee, P.	"To investigate siblings' perceptions of family disruption when a brother or sister had cystic fibrosis."	<ul style="list-style-type: none"> • Knowledge of the illness and its impact on the child • Effect of illness on the family • Effect of illness on the responding sibling • Advice to other siblings 	18	Descriptive
Gallo AM, Breitmayer BJ, Knafel KA, Zoeller LH.	"To describe how well siblings view potential stigma in childhood chronic illness."	<ul style="list-style-type: none"> • Revealing the chronic illness to others • Sibling responses to the ill child • Impact on daily living 	13.5	Descriptive
Herrman, Judith W.	"To fill the gap in the literature and attempt to provide health care professionals with siblings' perceptions which may serve to guide teaching and inform counselling."	<ul style="list-style-type: none"> • Costs of diabetes • Pain, interruptions and change • Family and sibling stress • Too much information: illness and consequences • Rewards of diabetes • We have a healthier and closer family • Knowledge and responsibilities. 	21.5	Descriptive
Hollidge C.	"To investigate the psychological experience of well children living in the same household as child siblings with diabetes."	<ul style="list-style-type: none"> • The well child's feelings about living with a diabetic sibling • Expression of worry about health 	14	A priori descriptive

Hutson, S., & Alter, B	"To explore the experiences of healthy siblings of patients with Fanconi Anemia."	<ul style="list-style-type: none"> • Patterns of communication • Containment • Invisibility • Staying under the radar • Worry • Despair 	23.5	Descriptive
Malcolm C; Gibson F; Adams S; Anderson G; Forbat L,	"To report sibling experiences related to these rare degenerative and progressive conditions, in order to inform the future development of supportive interventions."	<ul style="list-style-type: none"> • Perceptions of the condition and its symptoms • Impact of the condition on the family • Emotional responses from the siblings • Coping and support 	21.5	Shared pathway or meaning
Okashah, R., Schoch, K., Hooper, S., Shashi, V., & Callanan.	"To assess (a) the frequency, method, and content of information about 22q11DS that parents share with their unaffected children; (b) the knowledge of 22q11DS among unaffected siblings; and (c) the unaffected siblings' perceptions of the impact of this condition on their affected sibling and themselves....to help genetic counselors develop anticipatory guidance for families with children with 22q11DS."	<ul style="list-style-type: none"> • Less negative emotions (includes worry, stress, guilt). • Less insight or knowledge about condition (includes increased compassion, ability overcome obstacles) • Increased parental time and attention • Better or different relationship with siblings • Be similar to other kids without 22q11.2DS 	13	Descriptive
Read, J., Kinali, M., Muntoni, F., Weaver, T., & Garralda, E	"To obtain descriptive accounts from siblings about impact and coping with Duchenne Muscular Dystrophy and consider implications for psychological function."	<ul style="list-style-type: none"> • Knowledge • Caring responsibilities • Activities • Impact • Coping mechanisms • Supports 	20.5	Descriptive
Velleman, S., Collin, S., Beasant, L., & Crawley, E.	"To understand the impact of paediatric CFS/ME on siblings, to investigate whether any factors are psychologically protective and to discover how services should support siblings of young people with CFS/ME."	<ul style="list-style-type: none"> • Impact on family • Impact on sibling 	17.5	Descriptive
Wennick, A., & Huus, K.	"To illuminate what it is like to be a brother or sister of a child newly diagnosed with type 1 diabetes."	<ul style="list-style-type: none"> • Living differently • Being concerned • Participating in caring for the affected child 	19.5	Descriptive

Figure 1.

An adapted PRISMA (2009) representation of the electronic database literature search run in April 2016

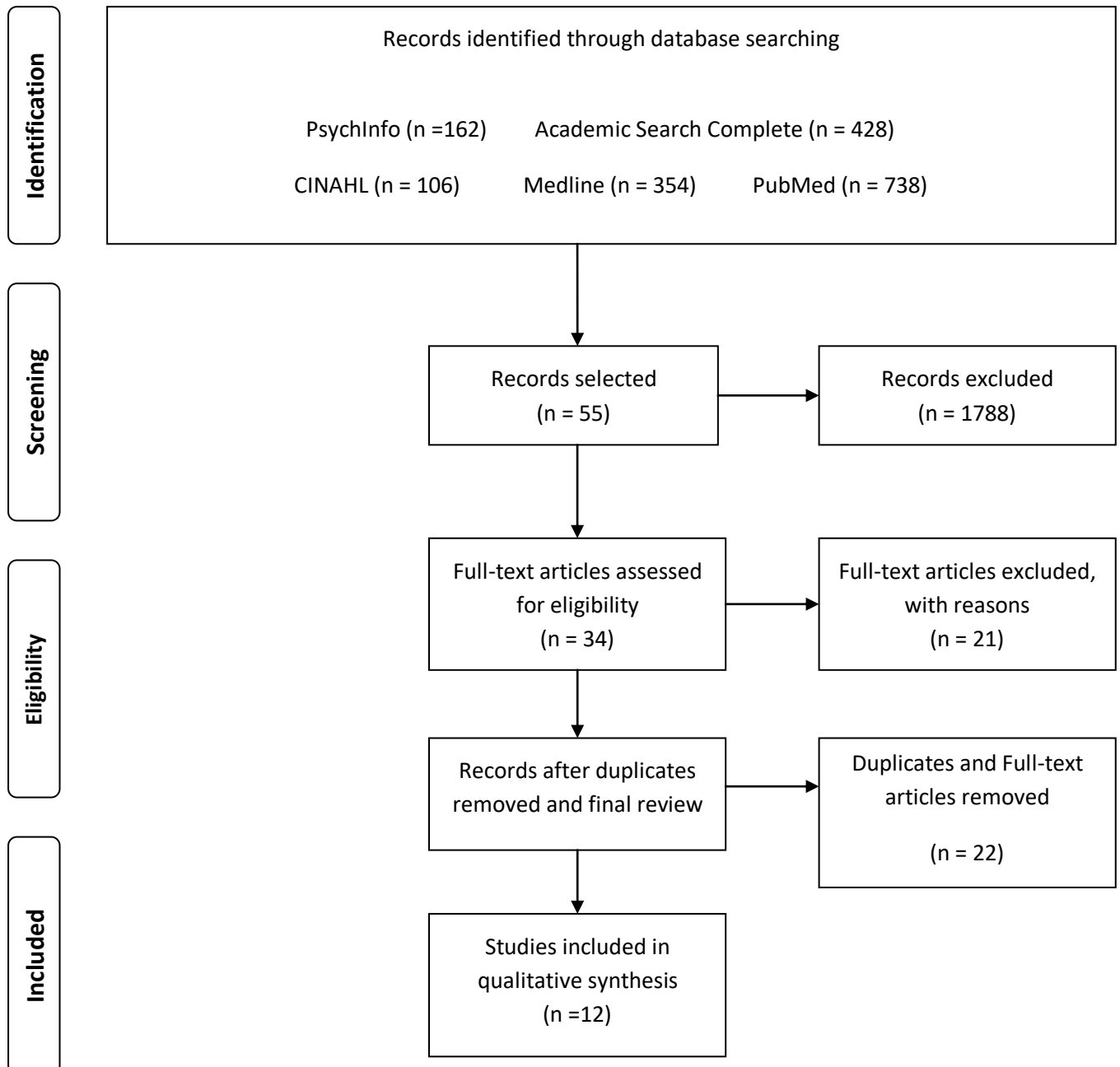


Table 3.

A Summary of the Key Features of the Final Sample of Selected Articles.

Authors and Date	Title	Methodology	Number of participants and type of family member	Average or range of age of healthy (and unwell siblings where given)	Type of illness	Setting and country
Bellin, M., Kovacs, P., & Sawin, K. (2008)	Risk and protective influences in the lives of siblings of youths with spina bifida	Data reported are from a larger mixed method study. One open ended question was posed in the posted study packet. Written responses were returned via post. Reported using a form of content analysis and referenced Morse and Field (1995). Developed themes with no statistics.	155 siblings	13.83	Spina Bifida	Social work, USA
Brennan, C., Hugh-Jones, S., & Aldridge, J. (2012)	Paediatric life-limiting conditions: coping and adjustment in siblings	Mixed method, semi-structured interviews. Reported using grounded theory and referenced Starks and Brown Trinidad (2007).	31 siblings	9.9	Life-limiting conditions	Health Psychology, UK
Derouin D, Jessee P. (1996)	Impact of a chronic illness in childhood: siblings' perceptions	Mixed method, semi-structured telephone interviews based on an interview schedule developed by Tritt (1983), and questionnaires to siblings and parents. The analysis is reported as non-parametric descriptive analyses based on the categories outlined in the interview schedule.	15 Siblings and 14 parents	8-17	Chronic illnesses	USA

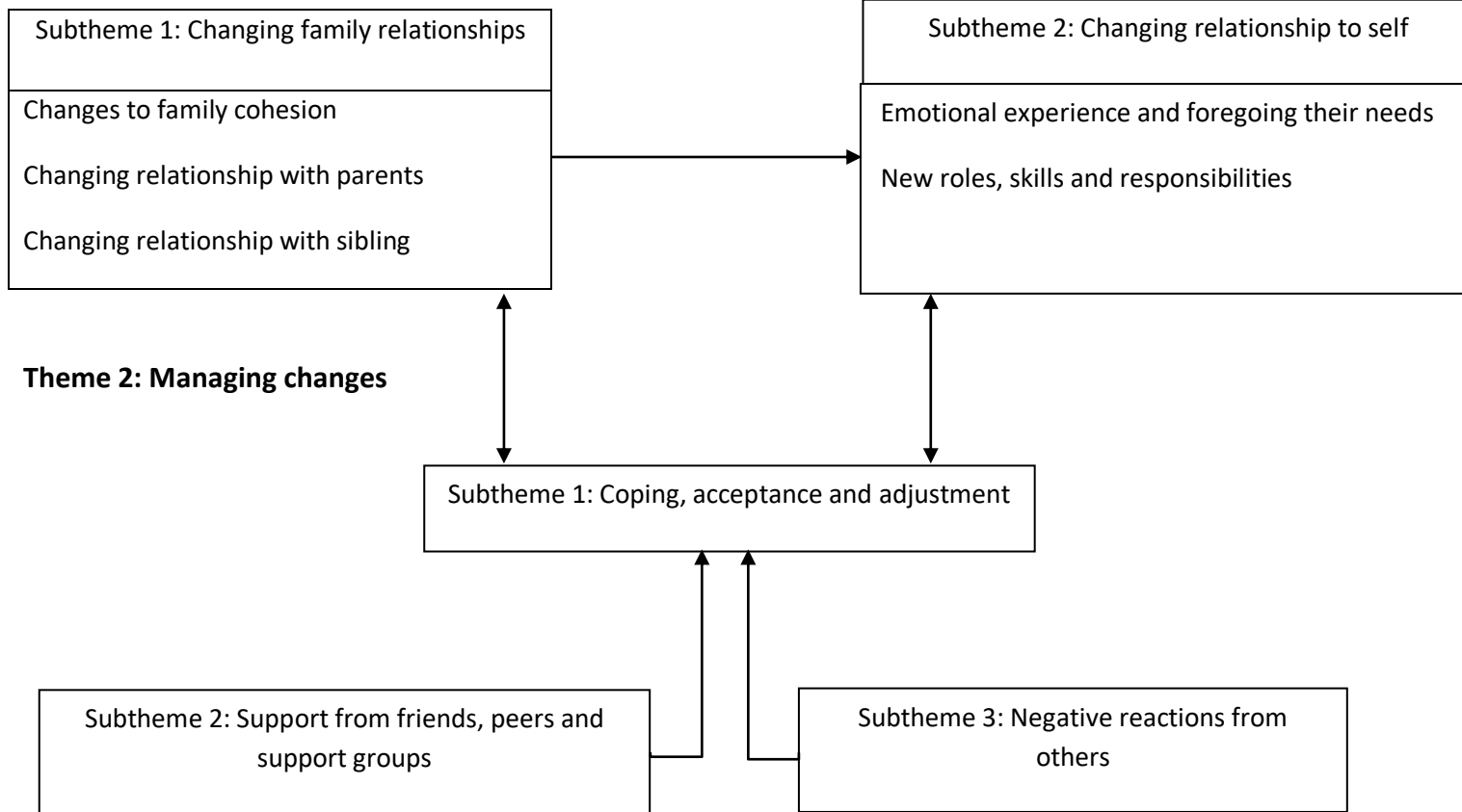
Gallo A, Breitmayer B, Knafl K, Zoeller L. (1991)	Stigma in childhood chronic illness: a well sibling perspective	Part of a larger study. Structured interview of three questions and the results were categorised according to these questions. Referenced using the analysis promoted by Miles and Huberman, (1984).	27 Siblings	6-14	Chronic illnesses	Nursing, USA
Herrman, J. (2010)	Siblings' perceptions of the costs and rewards of diabetes and its treatment.	Semi structured interviews were based on a previous schedule focusing on rewards and costs of having a sibling with diabetes, developed according to social exchange theory (no reference given). Reported using template analysis (Polit and Beck, 2003).	20 Siblings	4-16	Diabetes	Nursing, USA
Hollidge C. (2001)	Psychological adjustment of siblings to a child with diabetes.	Mixed methods, semi-structured interviews. The analysis placed data into three pre-organised themes, relating to siblings feelings, expression of worry about their own health and communication patterns. No references were given for the qualitative part of the analysis.	28 Siblings	8-12	Diabetes	Social Work, Canada
Hutson, S., & Alter, B. (2007)	Experiences of siblings of patients with fanconi anemia	Semi-structured interviews. Reported using qualitative description and qualitative content analysis referenced Sandelowski (2000) and Kearney (2001).	9 siblings	11-21	Fanconi Anemia	Nursing, USA
Malcolm, C., Gibson, F., Adams. S., Anderson, G., & Forbat, L. (2013)	A relational understanding of sibling experiences of children with rare life-limiting conditions: Findings from a qualitative study	A mixed method study. The qualitative section used interviews facilitated by the card sort technique and a schedule designed to support the card sorting. The analysis was informed by grounded theory and referenced Charmaz (2006).	8 siblings	7-12	Mucopolysacch aridoses & Batten disease	Nursing, UK
Okashah, R., Schoch, K.,	Parental communication and experiences and knowledge of	Mixed method, online survey, 2 open ended questions. Reported	29 siblings	12-16	22q11.2 deletion syndrome	Genetic counselling,

Hooper, S., Shashi, V., & Callanan, N. (2015)	adolescent siblings of children with 22q11.2 deletion syndrome	using content analysis and referenced Elo and Kyenga, (2008)				USA
Read, J., Kinali, M., Muntoni, F., Weaver, T., & Garralda, E. (2011)	Siblings of young people with DMD - a qualitative study of impact and coping	Semi-structured interviews. Discusses using thematic analysis (Flick, 1998) and the thematic framework approach (Ritchie and Lewis (2003) for analysis.	35 siblings	11-18 (5-22)	Duchenne Muscular Dystrophy	Neurology, UK
Velleman, S., Collin, S., Beasant, L., & Crawley, E. (2015)	Psychological wellbeing and quality of life among siblings of paediatric CFS/ME patients: A mixed-methods study	Mixed method, semi-structured interviews and analysed using thematic analysis (Braun and Clarke, 2006).	9 siblings	12-17 (8-18, 13.8 mean)	CFS/ME	Clinical Psychology/ Psychiatry, UK
Wennick, A., & Huus, K. (2012)	What it is like being a sibling of a child newly diagnosed with type 1 diabetes: an interview study	Semi-structured interviews and content analysis referencing Burnard (1991).	7 siblings	10-17 (8-13, 9.5 median)	Diabetes	Nursing, Sweden

Figure 2.

A graphical depiction of the themes and connections elicited

Theme 1: Changing relationships



Appendix

- 1A A summary of the October 2014 and April 2016 article review
- 1B A table of the full CASP quality analysis as ascertained by the average from the researcher and an external liaison
- 1C An example from the line-by-line coding table
- 1D A table of articles represented in the themes
- 1E An overview of developing the analytical themes
- 1F An example of a developing analytical theme: Managing change
- 1G Pediatrics manuscript guidelines for publication
- 1H Pediatrics title page
- 1I Pediatrics contributor's statement page

Appendix 1A:

A summary of the October 2014 and April 2016 article review

The final searches in each database had been saved in March 2014. The searches were re-run using the publication date as a filter. All relevant articles found in 2013-2014 and 2014-2016 were subjected to the inclusion criteria. Two new articles were found in the April 2016 search and were considered appropriate to be synthesised into the review.

Below is a table depicting the number of articles found in each new search and the reasons for exclusion. Some of the articles may have been excluded for multiple reasons however detailed below are the ones identified first.

Database (total number of articles found in new search) and reason for exclusion after initial application of inclusion and exclusion criteria	Number of articles excluded in October 2014	Number of articles excluded April 2016
CINAHL (12)(31)		
• Already in saved folder (duplicate)	3	1
• Not sibling focused	5	0
• Focuses on childhood cancer	2	1
• Not related to chronic physical illness	2	0
Academic Search Complete (66)(96)		
• Already in saved folder (duplicate)	5	1
• Not sibling focused	21	0
• Focuses on childhood cancer	4	3
• Not related to chronic physical illness	15	2
• Does not use qualitative methodology	15	2
• Not focused on humans	6	0
• Disease specific experience	0	1
PsychInfo (27)(65)		
• Already in saved folder (duplicate)	3	2
• Not sibling focused	17	0
• Focuses on childhood cancer	2	3
• Not related to chronic physical illness	3	0
• Does not use qualitative methodology	2	0
Medline (50)(61)		
• Already in saved folder (duplicate)	3	2
• Not sibling focused	18	1
• Not age appropriate (e.g. geriatric illness)	1	0
• Focuses on childhood cancer	5	5
• Not related to chronic physical illness	7	1
• Does not use qualitative methodology	12	0
• Not focused on humans	4	0
Pubmed (47)(141)		
• Already in saved folder (duplicate)	0	0
• Focuses on childhood cancer	3	1
• Not sibling focused	1	0

Appendix 1B:

A table of the CASP quality analysis as ascertained by the average from the researcher and an external liaison

Articles	Presence of clear research aims?	Qualitative approach appropriate?	Design appropriate for aims?	Recruitment strategy appropriate?	Does data collection address the research issue?	Presence of reflection on researcher and participant relationship?	Ethical issues considered?	Data analysis rigorous?	Clear statement of findings?	Does research add value?	Total score out of 30
Bellin, M. Kovacs, P. Sawin, K.	3	2.5	1.5	1.5	2	1	1.5	2.5	2.5	3	21
Brennan, C., Hugh-Jones, S., & Aldridge, J	3	2.5	3	2	3	0	2	2.5	2.5	2	22.5
Derouin D, Jessee P.	3	2.5	1.5	2	1.5	1	1.5	1	2	2	18
Gallo A, Breitmayer B, Knafel K, Zoeller L.	3	2.5	1	1	2	0	0	1	1.5	1.5	13.5
Herrman, J	3	2.5	3	2	3	0.5	1.5	2	2	2	21.5
Hollidge C.	2.5	2.5	1.5	2	1	0	0	1	1.5	2	14
Hutson, S., Alter, B	2.5	2.5	3	2.5	3	1	2.5	2	2	2.5	23.5
Malcolm C; Gibson F; Adams S; Anderson G; Forbat L,	3	2.5	3	2.5	2.5	0.5	2	1.5	2.5	1.5	21.5

Okashah, R., Schoch, K., Hooper, S., Shashi, V., & Callanan.	2.5	2	1	1.5	1.5	1	1	1	1	1	13
Read, J., Kinali, M., Muntoni, F., Weaver, T., & Garralda, E.	3	2.5	2	2	2.5	0.5	2	2	2.5	1.5	20.5
Velleman, S., Collin, S., Beasant, L., & Crawley, E.	3	2	1.5	2	1.5	1	1.5	1.5	2	2	17.5
Wennick, A., & Huus, K.	2.5	2.5	2.5	2	2	0.5	2.5	1.5	2	1.5	19.5

Appendix 1C:

An example from the line-by-line coding table

Reference	Themes from original article	Line-by-line codes
Wennick et al.	Living differently Being concerned Participating in caring for the affected child	<p>Living differently</p> <ul style="list-style-type: none"> • A transformed everyday life – physiological, structured and psychological aspects. • They find it odd to see their sibling affected by the illness – injections etc. • It's not like it was before. • Eventually it all became normal. • Life became more structured – scheduling meals. • Meeting the siblings needs resulting from illness. • Affected child was seen as more easily irritated, with mood swings. • Siblings have to be more patient. • Parents also change – e.g. friendlier. • Parents give affected child more attention than before. • Siblings see imbalance of attention as understandable. • Feel sorry for sibling. <p>Being concerned</p> <ul style="list-style-type: none"> • Siblings feel they know little about the illness. • Fear that it will ruin their sibling's life. • Treatment is a reminder of their concern – worry the needle hurts. • Frightening information – what will happen if they don't take insulin. • Feel that parents have enough worries. • Recurrent need to ask sibling if they feel well, to subdue anxiety. • Fearing a negative response to checking their sibling is ok. • Unfamiliarity with management and a need for more information led to fear. • Worry about future health of sibling. • Desire to comfort sibling about a cure. • Often want to be involved in hospital educational sessions, but due to scheduling could not attend. <p>Participating in caring for the affected child</p> <ul style="list-style-type: none"> • Hospitalisation means living as a separated family. • Parents split care when the sibling is in hospital. • Sibling has to assume more responsibility at home.

		<ul style="list-style-type: none">• A family reunion is positive.• Discharge does not imply less responsibility but it is shared.• Helping out consists of housekeeping, glucose testing, motivating sibling to follow treatment.• The whole family contributing to care improved their unity and felt like they were all closer.
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Appendix 1D:

A table of articles represented in the themes

Synthesis theme	Contributing articles (number of codes from the article in ascending order)	Contributing themes from original article
Changing relationships <ul style="list-style-type: none"> • Changing family relationships 	Brennan (0) Malcolm (1) Hollidge (2) Hutson (5) Gallo (5) Velleman (5) Okasha (5) Read (6) Wennick (9) Derouin (10) Herrman (13) Bellin (20)	<ul style="list-style-type: none"> • Perceptions of the condition and its symptoms • The well child's feelings about living with a diabetic sibling • Despair • Staying under the radar • Containment • Impact of the illness on daily living • Sibling responses to the ill child • Communication • Change of focus/role • Less negative emotions • Increased parental time and attention • Be similar to other kids without 22q11.2DS • Impact • Activities • Participating in caring for the affected child • Living differently • Being concerned • Effect of illness on the responding sibling • Knowledge • Effect of illness on the family • Rewards of diabetes to the family • Costs of diabetes to the sibling • Costs to the family • Rewards of treatment of diabetes • Cohesive family relations • Warmth and closeness in the sibling relationship

<ul style="list-style-type: none"> • Changing relationship to self <ul style="list-style-type: none"> ○ Emotional experience and needs 	<p>Wennick (0) Gallo (1) Okasha (2)</p> <p>Malcolm (3)</p> <p>Brennan (3) Derouin (3) Herrman (5)</p> <p>Velleman (8)</p> <p>Read (10)</p> <p>Bellin (11)</p> <p>Hollidge (11)</p>	<ul style="list-style-type: none"> • Qualities of the social environment of siblings • Influence of SB on the sibling relationship • Conflicted, ambivalent feelings about having a sibling with SB • Experiences of differential treatment and expectations • The development of unique knowledge and responsibilities • Perception of sibling with SB as “normal” • Impact of SB on daily life <ul style="list-style-type: none"> • Sibling responses to the ill child • Less negative emotions • Better or different relationship with sibling • Emotional responses from siblings • Perceptions of the condition and its symptoms • Self as glue • Effect of illness on the responding sibling • Cost of diabetes • Costs of treatment of diabetes • Costs of diabetes to the sibling • Communication • Emotional reactions • Impact • Coping mechanisms • Knowledge • The emotional climate of siblings • Conflicted, ambivalent feelings about having a sibling with SB • Protective and empathic concerns for sibling with SB • Questioning the fairness of SB • The omnipresence of SB • Feeling undervalued and overlooked • The well child’s feelings about living with a diabetic sibling • Expression of worry about health • Patterns of communication • Despair • Worry
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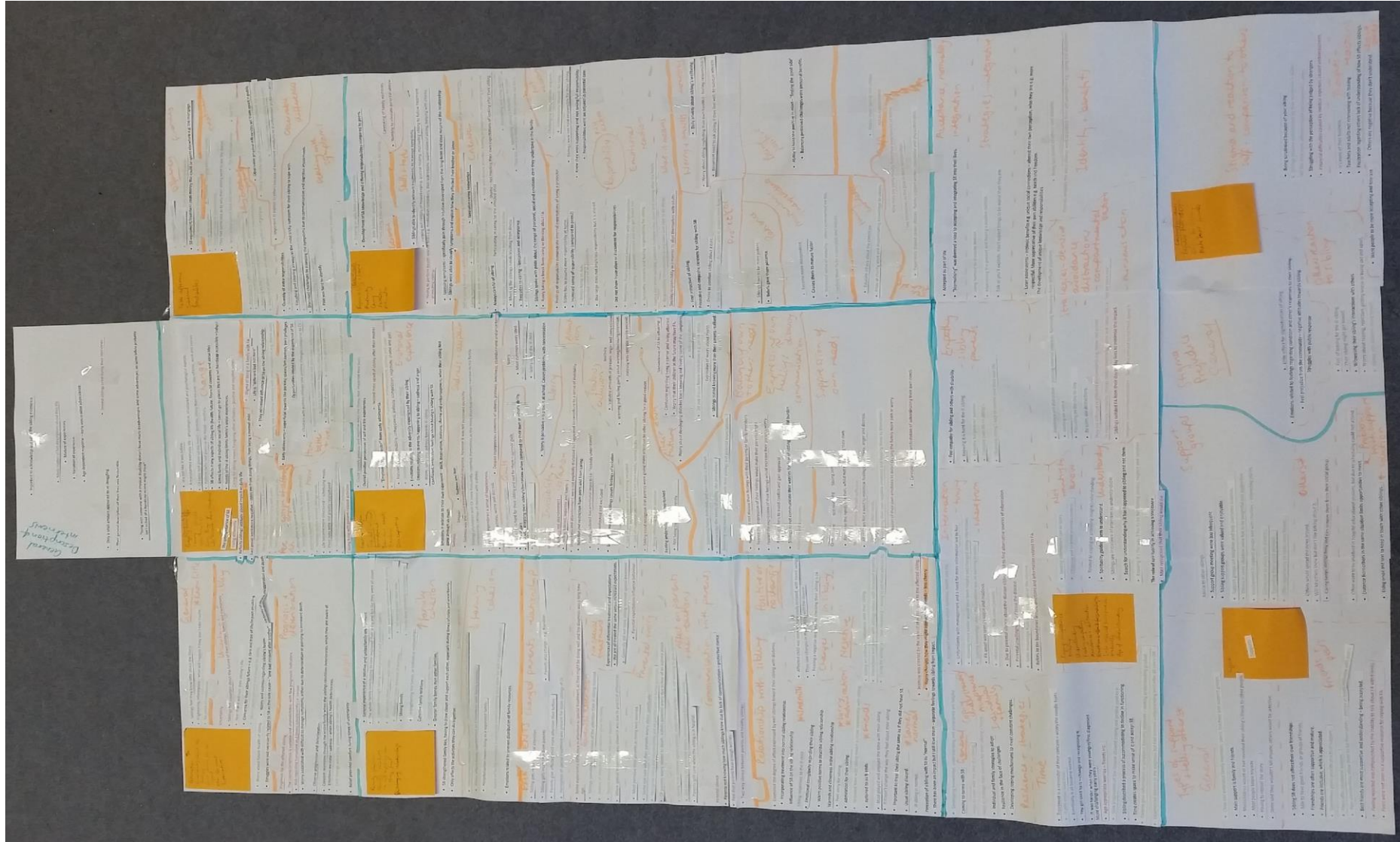
<p>o Roles and skills</p>	<p>Hutson (25)</p> <p>Okasha (1) Derouin (2)</p> <p>Gallo (2) Hollidge (3)</p> <p>Hutson (3) Velleman (4) Brennan (7)</p> <p>Bellin (8)</p> <p>Wennick (8)</p> <p>Herrman (10)</p> <p>Malcolm (11)</p>	<ul style="list-style-type: none"> • Invisibility • Staying under the radar • Containment • Less insight or knowledge about the condition • Effect of the illness on the responding sibling • Advice to other siblings • Impact of the illness on daily living • The well child's feelings about living with a diabetic sibling • Expression of worry about health • Staying under the radar • Positioning in the adult world • Change of role/focus • Self as glue • The development of unique knowledge and responsibilities • The omnipresence of SB • Protective and empathic concerns for sibling with SB • Coming to terms with SB • Opportunities created by the experience of SB • Participating in caring for the affected child • Living differently • Being concerned • Costs to the family • Rewards of diabetes to the sibling • Costs of diabetes to the sibling • Rewards of diabetes • Perceptions of the condition and its symptoms • Impact of the condition on family life • Emotional responses from siblings • Coping mechanisms • Impact • Caring responsibilities • Supports
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<p>Managing changes</p> <ul style="list-style-type: none"> Coping and resilience 	<p>Read (16)</p> <p>Hollidge (0) Okasha (0) Gallo (1) Wennick (3)</p> <p>Derouin (3) Malcolm (3)</p> <p>Herrman (5)</p> <p>Brennan (6) Hutson (7)</p> <p>Velleman (8)</p> <p>Bellin (13)</p> <p>Read (17)</p>	<ul style="list-style-type: none"> Impact of the illness on daily living Living differently Being concerned Advice to other siblings Perception of the condition and its symptoms Emotional responses from siblings Rewards of diabetes to the family Rewards of diabetes to the sibling Costs of diabetes to the sibling Costs of diabetes Compartmentalising life Worry Uncertainty Parental silence Containment Life outside the family Advice to CFS/ME specialist services or to families Not knowing The journey toward acceptance of SB Coming to terms with SB Cohesive family relations More challenging early years The role of spirituality in achieving acceptance Questioning the fairness of SB Experiences of differential treatment and expectations Perception of sibling with SB as "normal" Coping mechanisms Knowledge Impact
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<ul style="list-style-type: none"> • Support 	<p>Hollidge (0) Brennan (0) Velleman (1) Okasha (1)</p> <p>Wennick (1) Derouin (1) Bellin (3) Herrman (3)</p> <p>Hutson (3)</p> <p>Malcolm (6) Read (7)</p> <p>Gallo (7)</p>	<ul style="list-style-type: none"> • Communication • Less insight or knowledge about condition • Being concerned • Advice to other siblings • The nature of peer relations • Rewards of diabetes to the sibling • Rewards of diabetes to the family • Rewards of diabetes • Stigma of being in a family with FA • Being busy with patient-related activities • Coping and support • Supports • Impact • Revealing • Not revealing • Impact of the illness on daily living
<ul style="list-style-type: none"> • Reactions from others 	<p>Derouin (0) Hollidge (0) Wennick (0) Read (0) Okasha (0) Brennan (1) Hutson (1) Herrman (2)</p> <p>Bellin (5)</p> <p>Gallo (5)</p> <p>Malcolm (5) Velleman (9)</p>	<ul style="list-style-type: none"> • Compartmentalising life • Stigma of being in a family with FA • Costs of diabetes • Costs of treatment of diabetes • Protective and empathic concerns for sibling with SB • Struggles with public response • Feeling undervalued and overlooked • Not revealing • Revealing • Sibling responses to the ill child • Emotional responses from siblings • Communication • Social emotional support/impact

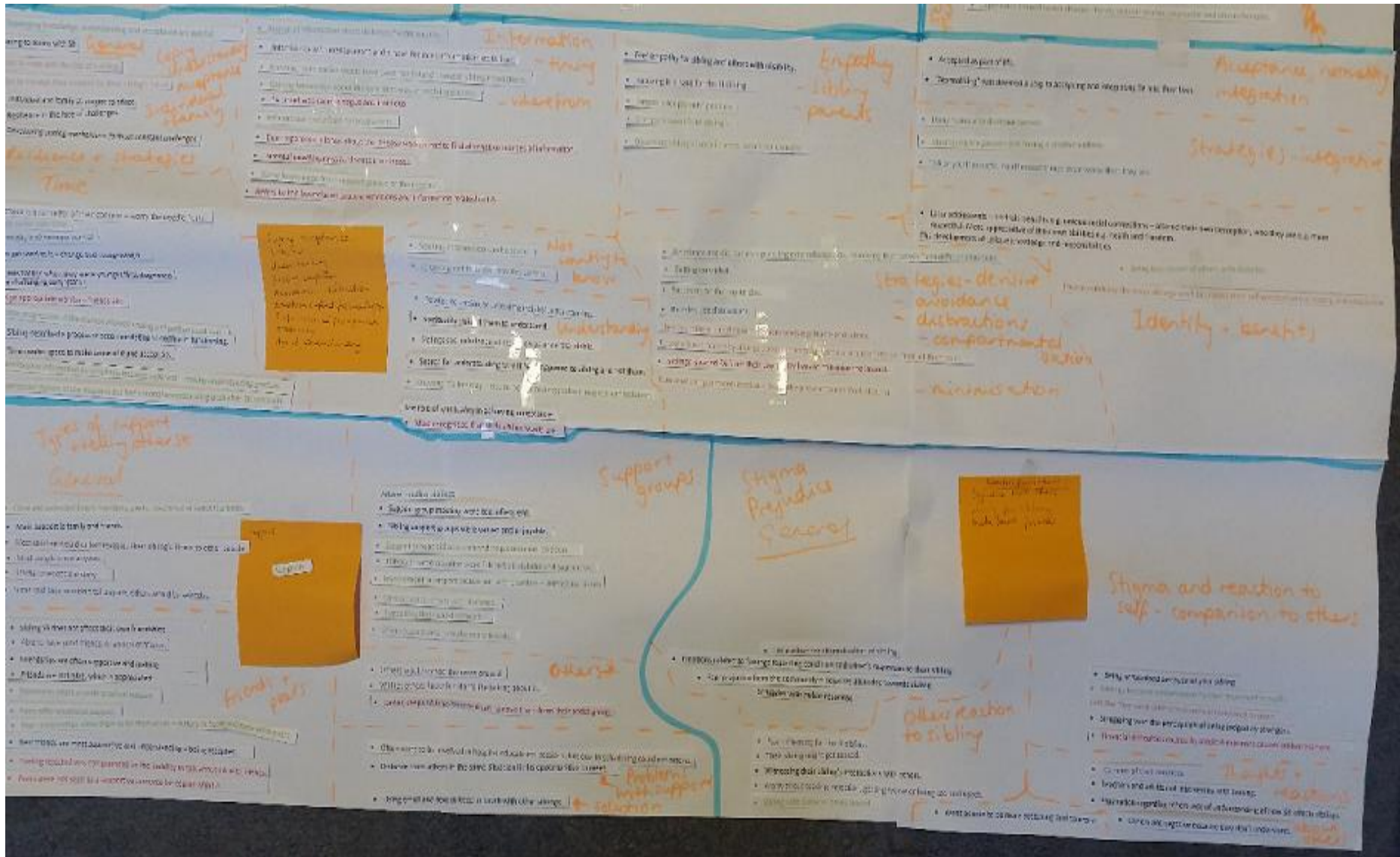
Appendix 1E:

An overview of developing the analytical themes



Appendix 1F:

An example of a developing analytical theme: Managing change



Appendix 1G:

Pediatrics manuscript guidelines for publication

Journal Style

All aspects of the manuscript, including the formatting of tables, illustrations, and references and grammar, punctuation, usage, and scientific writing style, should be prepared according to the most current *AMA Manual of Style* (<http://www.amamanualofstyle.com>).¹

Author Listing. All authors' names should be listed in their entirety, and should include institutional/professional affiliations and degrees held.

Authoring Groups. If you choose to include an organization, committee, team, or any other group as part of your author list, you must include the names of the individuals as part of the Acknowledgments section of your manuscript. This section should appear after the main text prior to your References section. The terms "for" or "on behalf of" must also be used when referencing the authoring group in the by-line.

Titles. *Pediatrics* generally follows the guidelines of the *AMA Manual of Style* for titles. Titles should be concise and informative, containing the key topics of the work. Declarative sentences are discouraged as they tend to overemphasize a conclusion, as are questions, which are more appropriate for editorials and commentaries. Subtitles, if used, should expand on the title; however, the title should be able to stand on its own. It is appropriate to include the study design ("Randomized Controlled Trial"; "Prospective Cohort Study", etc.) in subtitles. The location of a study should be included only when the results are unique to that location and not generalizable. Abbreviations and acronyms should be avoided. The full title will appear on the article, the inside table of contents, and in MEDLINE. Full titles are limited to 97 characters, including spaces. Short titles must be provided as well and are limited to 55 characters, including spaces. Short titles may appear on the cover of the journal as space permits in any given issue.

Abbreviations. List and define abbreviations on the Title Page. Unusual abbreviations should be avoided. All terms to be abbreviated in the text should also be spelled out at first mention, followed by the abbreviation in parentheses. The abbreviation may appear in the text thereafter. Abbreviations may be used in the abstract if they occur 3 or more times in the abstract. Abbreviations should be avoided in tables and figures; if used they should be redefined in footnotes.

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Proprietary Products. Authors should use nonproprietary names of drugs or devices unless mention of a trade name is pertinent to the discussion. If a proprietary product is cited, the name and location of the manufacturer must also be included.

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References

1. Iverson C, Christiansen S, Flanagan A, et al. *AMA Manual of Style*. 10th ed. New York, NY: Oxford University Press; 2007.

2. Lundberg GD. SI unit implementation: the next step. *JAMA*. 1988;260:73-76.
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Formatting Requirements

All submissions must adhere to the following format:

- Times New Roman font, size 12, black
- Title Page, Contributors' Statement Page, Abstract, Acknowledgments, and References should be **single-spaced**
- Only the Main Body Text should be **double-spaced**
- Main Submission Document as Microsoft Word or RTF file (no PDFs)
- Do **not** include page headers, footers, or line numbers in new submissions.
- Do **not** include footnotes within the manuscript body. Footnotes are allowed only in tables/figures.

Refer to the “Article Types” section for specific guidelines on preparing a manuscript in each category. Note in particular the requirements regarding abstracts for different categories of article.

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The “title page” should appear first in your manuscript document, and depending on the individual needs of a paper may encompass more than one page.

Title pages for all submissions **must** include the following items (as shown in the [sample Title Page](#)):

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5. **Financial Disclosure Statement** for all authors. Disclose any financial relationships that could be broadly relevant to the work. If none, say “Financial Disclosure: The authors have no financial relationships relevant to this article to disclose.”
6. **Funding source.** Research or project support, including internal funding, should be listed here; if the project was done with no specific support, please note that here. Technical and other assistance should be identified in Acknowledgments. If your funding body has open access requirements, please contact the Editorial Office prior to submission. Pediatrics has a 12 month embargo on articles (followed by a 4 year open access period) and does not allow articles to be opened for Creative Commons or similar licenses.
7. **Conflict of Interest Statement** for all authors. If none, say “Potential Conflicts of Interest: The authors have no conflicts of interest relevant to this article to disclose.”

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All persons designated as authors should qualify for authorship ([see "Publication Ethics" above](#)), and all those who qualify should be listed. Each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content. The Contributors' Statement Page should list the authors and specify the contribution(s) made by each individual. If multiple individuals have identical contributions they may be listed together; do not list an author more than once.

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Contributors' Statement:

Dr Smith conceptualized and designed the study, drafted the initial manuscript, and approved the final manuscript as submitted.

Drs Jones, Lee, and Weber carried out the initial analyses, reviewed and revised the manuscript, and approved the final manuscript as submitted.

Ms Green designed the data collection instruments, and coordinated and supervised data collection at two of the four sites, critically reviewed the manuscript, and approved the final manuscript as submitted.

Note: Contributors who do not meet the criteria for authorship (such as persons who helped recruit patients for the study, or professional editors) should be listed in an Acknowledgments section placed after the manuscript's conclusion and before the References section. Because readers may infer their endorsement of the data and conclusions, these persons must give

written permission to be acknowledged. These permissions do not need to be submitted with the manuscript unless requested by the editors.

Word Count

To determine article length, count the body of the manuscript (from the start of the Introduction to the end of the Conclusion). The title page, contributors' statement page, abstract, acknowledgments, references, figures, tables, and multimedia are not included.

Figures, Tables, and Supplementary Material

Figures

Authors should number figures in the order in which they appear in the text. Figures include graphs, charts, photographs, and illustrations. Each figure must include a legend (placed on the figure itself or as a list appearing after the References) that does not exceed 50 words. Abbreviations previously expanded in the text are acceptable. If a figure is reproduced from another source, authors are required to obtain permission from the copyright holder, and proof of permission must be uploaded at the time of submission.

Figure arrays should be clearly labeled, preassembled, and submitted to scale. Figure parts of an array (A, B, C, etc.) should be clearly marked in capital letters in the upper left-hand corner of each figure part.

Technical requirements for figures: The following file types are acceptable: TIFF, PDF, EPS, and PNG. Color files must be in CMYK (cyan, magenta, yellow, black) mode.

Style for figures: Readers should be able to understand figures without referring to the text. Avoid pie charts, 3-dimensional graphs, and excess ink in general. Make sure that the axes on graphs are labeled, including units of measurement, and that the font is large enough to read. Generally delete legends or other material from the graph if it makes the picture smaller. Color graphs should be interpretable if photocopied in black and white.

Pediatrics cannot accept Excel or PowerPoint files for any part of your submission.

Tables

Tables should be numbered in the order in which they are cited in the text and include appropriate headers. Tables should not reiterate information presented in the Results section, but rather should provide clear and concise data that further illustrate the main point. Tabular data should directly relate to the hypothesis. Table formatting should follow the current edition of the *AMA Manual of Style*.

Style for tables: Tables should be self-explanatory. Avoid abbreviations; define any abbreviations in footnotes to the table. Avoid excess digits and excess ink in general. Where possible, rows should be in a meaningful order (e.g., descending order of frequency). Provide units of measurement for all numbers. In general, only one type of data should be in each column of the table.

Supplemental Information

Authors may wish to include additional information as part of their article for inclusion in the online edition of *Pediatrics*. References to any online supplemental information must appear in the main article. Such supplemental information can include but are not limited to additional tables, figures, videos, audio files, slide shows, data sets (including qualitative

data), and online appendices. If your study is based on a survey, consider submitting your survey instrument or the key questions as a data supplement. Authors are responsible for clearly labelling supplemental information and are accountable for its accuracy. Supplemental information will be peer reviewed, but not professionally copyedited.

Review Article, Systematic Reviews and Meta-Analyses

Abstract length: 250 words or fewer (structured or unstructured, depending on review type)

Article length: 4,000 words or fewer

Review Articles combine and/or summarize data from the knowledge base of a topic. Preference is given to systematic reviews and meta-analyses of clearly stated questions over traditional narrative reviews of a topic. Both types of review require an abstract; the abstract of a narrative review may be unstructured (no headings, run in a single paragraph). **See below for abstracts of systematic reviews and meta-analyses.**

The general instructions regarding submission (including cover letter, title page requirements, contributors' statement page, journal style guidance, and conflict of interest statements) also apply to Review Articles.

Systematic Reviews and Meta-Analyses

Reports of systematic reviews and meta-analyses should use the PRISMA statement (<http://www.prisma-statement.org/>) as a guide, and include a completed PRISMA checklist and flow diagram to accompany the main text. Blank templates of the checklist and flow diagram can be downloaded from the PRISMA Web site (<http://www.prisma-statement.org/statement.htm>).

Structured abstracts for systematic reviews are recommended. Headings should include: Context, Objective, Data Sources, Study Selection, Data Extraction, Results, Limitations, and Conclusions (see Iverson et al¹[pp22-23]).

Appendix 1H:

Pediatrics title page

A Synthesis of Children's Perspectives of Living with a Sibling with a Paediatric Chronic Illness

Antoinette Deavin (BSc, MRes), Pete Greasley (PhD), Clare Dixon (BSc, DClInPsy, PhD)

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Short Title: Siblings' Perspectives of Living with a PCI

Funding Source: Lancaster University

Financial Disclosure: The authors have no financial relationships relevant to this article to disclose.

Conflict of Interest: The authors have no conflicts of interest relevant to this article to disclose.

Abbreviations: Paediatric Chronic Illness (PCI)

This synthesis voices healthy children's experiences of living with a sibling who has a paediatric chronic illness.

The research literature on healthy siblings' well-being is mixed. A recent meta-analysis suggests that siblings have poorer outcomes than their peers, with higher levels of internalising (e.g. depression) and externalising (e.g. aggression) behaviours, and highlights the importance of systemic factors.

The synthesis of qualitative articles allows siblings' subjective experiences to be heard and understood in context. By gathering these experiences one can take an overview of the factors that they find important and the effect of their experiences.

Appendix 1I:

Pediatrics contributor's statement page

Contributors' Statement Page

Contributors' Statement:

Ms Antoinette Deavin conceptualised and designed the study, carried out the synthesis and drafted the initial and final manuscript as submitted.

Drs Greasley and Dixon reviewed the initial concept and critically reviewed the manuscript, and approved the final manuscript as submitted.

Section Two: Research Paper

**Parents' Stories of Adjustment to their Child's Chronic Illness: a Narrative Approach
to Duchenne Muscular Dystrophy**

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Word Count (excluding tables, references and appendices): 8,987

Word Count (including tables, references and appendices): 9,143

Prepared for submission to Pediatrics (see appendices 2M-2O for author guidelines, title page and contributors' statement)

Abstract

Background and Objectives: It has been recommended that further qualitative and disease specific research is needed to understand parental adjustment to Duchenne Muscular Dystrophy (DMD) - to elucidate the ‘what’ and the ‘how’.

This study explored the personal and societal narratives that influence parents’ experiences of adjustment to DMD. By understanding these influences it is possible to identify strategies to help support parents and minimise their distress.

Methods: Narrative analysis was performed on seven interviews of British parents of boys with DMD. Parents were purposively sampled utilising local neurological services, posters, and online advertisements. Interviews were recorded, coded and analysed to identify narrative elements.

Results: First, brief summary stories were developed, highlighting the ‘self’ that each parent promoted most vehemently in relation to their adjustment. Second, the narrative analysis yielded three chapters providing common narratives: ‘Investigating and fighting: “Something’s not right”’; ‘Making meaning of the diagnosis’; and ‘Living a normal life’.

Conclusions: The findings outline how parents, a) feel the need to fight for information and support, b) held narratives of health as restitution, and disability as loss, c) noticed how their identity and roles changed from parent or worker to carer, and d) lived day to day, embodying roles that upheld their narratives of normalcy and providing happiness, whilst allowing them to suppress their concerns about the future. Future research should focus on how multiple life events, and varying social and cultural narratives, impact upon adjustment.

Keywords: Duchenne Muscular Dystrophy, parent, narrative analysis, adjustment

Introduction

Recent research, policy and professional guidance on chronic illness, and specifically neuromuscular and rare conditions, reflects the financial, practical and emotional requirements of families and the lack of support they receive. These aspects are discussed in relation to the specific needs of parents of children with Duchenne Muscular Dystrophy (DMD) to provide context for this study and its rationale. This study aims to give voice to parents' experiences of adjustment to having a child with DMD and provide a theoretical understanding of their experiences.

Duchenne Muscular Dystrophy

Duchenne Muscular Dystrophy is a genetic, chronic and progressive disorder that affects approximately 2500 boys in the UK¹ with approximately 13,500 affected from 31 countries on the TREAT-NMD Global DMD Registry.²

DMD results in muscle degeneration in early infancy, which is associated with abnormal bone development and an increase of fat and fibrotic tissue. This causes a range of difficulties, examples of which can be seen in figure 1.

[Insert Figure 1.]

DMD is a life-limiting condition, with individuals having a life expectancy in their 20s. However new treatments have made it possible for some affected males to live into their 30s.^{3,4} DMD is associated with a range of co-morbidities including: respiratory difficulties, cardiac problems, scoliosis, a learning disability and behaviour that parents might find difficult to manage.⁴ A diagnosis of DMD therefore forewarns of potentially distressing physical changes for the child and family.

Rare Diseases: International Collaboration, Funding and Service Provision

A recent bulletin by the World Health Organisation (WHO) highlighted the urgent need for international investment and collaboration in research and treatment for rare diseases, noting that these tasks are too large for any one country to manage alone.⁵ The Walton Report (2009)⁶ highlighted the similar lack of funding and under resourced care for neurological diseases, particularly DMD, in the UK. The report stated that services relied upon charitable funding and had no form of succession planning. It concluded that often the levels of care were found to be “unacceptable” and this was thought to be due to the paucity of specialist adult neuromuscular services across the country.

It was recommended that to address a complex multi-system disorder like DMD a specialist multi-disciplinary approach would be appropriate, including increasing the number of named care coordinators and coordinators for young people when transferring from paediatric to adult services.

In attempts to standardise and improve care the National Health Service (NHS) in the UK had committed to develop clinical guidelines for ‘uncommon neuromuscular conditions’.⁷ However instead, they delegated this task to the Duchenne Muscular Dystrophy Care Considerations Working Group, comprising of international clinicians, researchers and patients,⁸ which subsequently developed international guidance specifically for the treatment of DMD.⁴

Parents’ Wellbeing, Current Psychosocial Recommendations and Adjustment

With large amounts of change to daily life, and potentially little support, it is not surprising that parents of children with DMD have been found to suffer psychological distress that meets diagnostic criteria,⁹ particularly depression.¹⁰

The DMD Care Considerations Working Group has specifically identified psychosocial management as being important when caring for families with children with DMD.⁴ In particular they recognised the experiences and needs of parents, one example being the stress associated with both the implementation of practical changes as a result of the disease and psychosocial problems within the family.¹¹

Similarly, the Walton report⁶ outlined the necessity for UK services to meet social care and quality of life needs, in addition to the health requirements of children with neuromuscular conditions. This included practical support and the promotion of the family's wellbeing; for example faster assessment and equipment provision, which would reduce the financial burden on families, supporting them to lead fuller lives. Regional findings in the north east of England in 2009 also suggested a promotion of wellbeing, with half the families reporting not being satisfied with the emotional support available to them.¹² Consequently specialist psychological input was recommended as part of the multidisciplinary team.

These reports highlight the distress experienced by parents of children with neuromuscular conditions, particularly DMD. This study therefore provided a timely opportunity to explore parents' subjective experiences with the aim of identifying how services can improve support and aid adjustment.

Adjustment

A recent editorial by Moss-Morris¹³ recently highlighted the inconsistent definition and measurement of 'adjustment' in the fields of mental health and chronic illness. The term is often used to describe the presence or absence of psychopathologies and their symptoms, providing a simple form of measurement. However it has been argued that this fails to recognise the multifaceted nature of adjustment and its many potential outcomes, including

factors not relating to the chronic illness e.g. impact on life roles. Consequently the presence of psychopathology may not be an accurate marker for adjustment.¹³

An alternative definition relates to an individual's ability to preserve their daily functioning.¹⁴ This definition was thought to be potentially unrealistic and dependent on the 'functioning tasks' of the specific illness, for example managing chronic pain versus coping with imminent death. Instead it was suggested that adjustment be considered in terms of the individual's ability to manage their emotions.^{15,16}

Moss-Morris considered these arguments and subsequently produced a model defining and operationalising adjustment as returning to a state of equilibrium psychologically, socially and/or physically (see appendix 2A).¹³

Other models presented in the adjustment literature often focus on undefined stressors or threatening events, and relate to the directly affected party i.e. the patient. These models have then been adapted to consider the chronic illness and/or the perspective of a parent. For example the models by Taylor¹⁷ and Lazarus¹⁸ both relate to generic external stressors but have become popular in the field of health psychology,¹³ in particular when considering an individual's adjustment to a chronic illness. These models are similar in that they consider the individual's ability to appraise or understand the problem, find solutions or coping strategies, gain a sense of control or efficacy and therefore restore self-esteem.^{18,19}

These elements are also key to the models of Wallander and Varni²⁰ and Walker,²¹ however they also include factors relevant to chronic illness in general. For example the model by Wallander and Varni⁹ consists of risk and resistance factors for parental adjustment considering the specifics of the disease or disability, which impacts on the functional independence of the child and the subsequent experience of psychosocial stress. These factors

and the parents' ability to process stress, their intrapersonal factors and social-ecological factors all affect adjustment (see appendix 2B).²⁰

Walker identified an interconnectivity between personality traits, stressors, appraisals, psychosocial functioning, coping strategies, affect, disease activity and physical adjustment (see appendix 2C).²²

A Narrative Approach to Parental Adjustment to DMD

Qualitative research aims to understand the subjective conceptualisations of a phenomenon, i.e. the "what" and the "how".²³ The current lack of consensus for what constitutes adjustment, the "what" of 'adjustment', impacts on the measurement of the "how". A qualitative approach would provide the content and links between ideas relevant to an individual's experience. By directly asking parents about their experiences of adjustment, both can be addressed.

Previous reviews^{13,20} have emphasised the need for further disease specific qualitative research in the areas of chronic illness and adjustment due to the disease's specific trajectories and associated life events. This is especially necessary for DMD given its rarity, the unique illness trajectory and the lack of understanding of families in this situation. There is a scarcity of research papers that focus on families', and particularly parental, experiences of DMD. A narrative approach would offer a contextualised definition of adjustment for parents, based on the subjective factors that influence their experience and identify their needs. Once identified, support can be targeted and research can build upon current theory and professional practice.

Narratives have been useful when trying to understand the influences, needs and coping strategies of vulnerable individuals who require chronic care.²⁴ The analysis of people's

stories has been successfully used to understand individual's experiences of health and healthcare,^{24,25} e.g. patient experience and health promotion.²⁵ Consequently a narrative approach could raise the profile of DMD and aid others' understanding of the effects on the family. Lastly, narrative analysis is useful for analysing the development and trajectory of experiences,²⁶ including transitions,²⁷ and would therefore address the recommendation that future studies should investigate adjustment over time.¹³

Aims

This study aims to provide a subjective understanding of parental adjustment to DMD by giving voice to the needs of vulnerable and overlooked parents.

Method

Design

A qualitative approach, entailing narrative interviews and analysis was used to address the research question. Narrative analysis stems from the concept that the stories and narratives we hold about our experiences represent the meaning we make of them,^{28,29} including how we attribute agency to the characters involved and the inferred causal links we make between the events in our lives.²⁷ Narrative analysis identifies these elements and provides insight into the participants' internal world by analysing the content and structure or "the told" and "the telling"²⁹ of their stories.^{27,30} Cross case analyses of stories are used to discover patterns or explore differences of the meaning that individuals make of their experience.²⁹ By accessing participants' stories this study contributes to our understanding of parental experiences of a child's health condition, aspects specific to DMD and parents' subjective understanding of adjustment.

Participants

Five female and two male parents of children with DMD participated in this study, all of whom were the biological parents. All participants had received the diagnosis over two years ago (ranging from 4 to 12 years prior to interviews) and were still caring for the child; this encouraged them to explore their longer-term narratives of adjusting to having a child with DMD. All participants spoke English as their primary language.

As recommended by Murray, a table of demographic and contextual details are presented below.³¹

[Insert Table 1.]

Participants with children who had other co-morbidities unassociated with DMD, e.g. other chronic health illnesses, were not included in an attempt to keep the interview focused on the child having DMD.

Parents with their own chronic health needs that may distinctly affect their perspective were not included. Parents for whom there were ongoing safeguarding concerns i.e. currently under investigation, were also not included. This became relevant when a parent with a previous history of low mood was included after assessment and subsequent consultation with the research team.

Procedure

Recruitment

Participants were purposively sampled and recruitment was based on practical and theoretical factors, i.e. the size and scope of the study and the ability to represent participants' stories effectively and with sufficient detail.

Multiple recruitment strategies were designed to increase access to the study and allow the researcher to manage the potential number of people being recruited. These were a postal mail out, information given by the neuromuscular nurses during clinics, posters, and an online advert on the Contact a Parent website.

Interviews

Participants took part in face-to-face semi-structured interviews. Locations were collaboratively agreed between the researcher and the interviewee, to consider travel distance, suitability for the task and confidentiality. Six chose to be interviewed at home and one at his place of work.

The interviews were based on a schedule developed by the lead researcher (see appendix 2D) and ranged from 52 minutes to 1 hour 40 minutes, with an average of 76 minutes. The schedule was influenced by Flick's³² episodic interview which focuses on specific experiences of change or periods of disruption. Initially participants were asked a narrative inducing question and, as necessary, followed by open ended prompts relating specifically to the research question: e.g. "What has influenced your adjustment?" More generic prompts were developed to encourage the interviewee to continue e.g. "What happened next?" The interview process was designed to ensure that parents' stories were listened to actively but also minimised the researcher's influence on the participant's responses.³³

When necessary the interview schedule was designed to prompt responses relating to influences,^{33,34} identity,³³ chronology/temporality²⁶ and key characters.^{33,34} To ensure validity the transcripts were reviewed between interviews by the research team to consider the formulation of questions. This promoted changes to the questions, which included a final summarising question of "Are there any other events that you feel have been important to you?"

Analysis

All interviews were transcribed by the researcher and initial notes on pertinent features were made. The analysis was influenced by Lieblich, Tival-Mashiach and Zilber's²⁷ holistic versus categorical, and content versus form dimensions. The axes they promote provide a flexible structure for analysis that allow varying amounts of emphasis on the content and structure of participant stories.

The main analysis was informed by Crossley's³⁵ approach to engaging with the data, i.e. familiarisation and using constant iterative comparisons; identifying important concepts, narrative tone, themes and images. The work of McAdams³⁴ was also considered regarding more specific narrative elements to be explored, i.e. key events, significant people, future scripts, stresses and problems and personal ideology. To achieve this, codes were recorded on the transcripts and coloured according to the potential narrative components mentioned previously (see appendix 2E). Key events, role, identities and characters were tabulated (see appendices 2F and 2G), as were similarities in language and potential themes (see appendices 2H-2K). The lead researcher also made reflective notes post interview.

This led to a "cross case analysis",²⁹ identifying common concepts between themes which became the chapters, providing a "categorical-form" approach, to discover "...both the themes that unify the story and the disparate voices that carry, comment on and disrupt the main themes."^{29(pp226)} In order to move the analysis from purely categorical, i.e. similar to a content analysis,²⁷ the lead researcher explored how the parts of the themes became integrated akin to Schleiermacher's "hermeneutic circle", whereby an understanding of the whole reveals the parts which in turn creates a whole.²⁹ The chapters were then discussed with the research team to assess the integration of the themes into chapters.

Finally, reflection on the post interview notes and the cross case analysis highlighted the importance of specific individual narratives. Consequently brief accounts were developed (see appendix 2L) providing a more “holistic-form” approach, to provide further insight into each parent’s personal journey and to highlight any features that were unique or salient in their interview. Murray acknowledges that individuals have many stories about themselves that endeavour to structure our ‘selfhood’.^{30(pp115)} The summary stories were developed to explore the selfhood that the participants promote through their story telling and how these identities influence their adjustment.

Validity and Quality

Unlike quantitative research, narrative analysis is not concerned with the realm of absolute truths and therefore does not seek the same forms of assessment.^{23,27} Alternatively, the trustworthiness, rigour^{37,38} and quality³⁷ of the study needs to be demonstrated. These elements were addressed through frequent supervision with the research team. Transparency has been maintained through the use of audit trails e.g. keeping a reflective journal (other exemplars can be found in appendices 2D-2L), provision of evidence (e.g. participant quotations) and making the audience aware of the researcher’s position to the research.²⁷

Reflective Positioning

Reflecting on the researcher’s position is important in narrative research as the approach acknowledges that the researcher is more than an observer and is part of co-constructing the story.³⁹ Consequently the analysis and write-up were designed to recognise these influences to make them transparent to the reader.

The lead researcher took a critical realist stance towards the research question⁴⁰ in order to provide an analysis grounded in parents’ experiences and understandings, whilst

acknowledging the importance of the researcher's interpretations informed by theory and clinical experience.

Although the lead researcher had no personal experience with paediatric chronic illness (PCI), experiences of working with two young men with DMD and their families, and more recently embarking on the journey of becoming a parent, have developed the researcher's interest during clinical psychology training. Training has also highlighted the need to help empower individuals within healthcare. The researcher's position is explored further in the critical appraisal of this thesis.

Ethics

The British Psychological Society's Codes of Ethics and Conduct ⁴¹ and the Lancaster University and Lancashire Care NHS Trust guidance were considered to identify ethical issues and develop procedures that would enable the safety and comfort of all parties involved in the research. This study was approved by the Lancaster University Research Office, the NHS Research Ethics Committee and the appropriate Trust Research and Development team. Due to the potential for participants to experience distress during the interview the study was submitted to and approved by a NHS Proportionate Review Subcommittee. All the ethical issues and procedures are outlined thoroughly with supporting documents in section four of this thesis. However the main issues related to the management and/or communication of confidentiality, consent, risk management and assessing the level of potential distress to be possibly experienced by the interviewee.

Findings

The Common Narratives

The findings have been organised into an integrated story comprised of three chapters, detailing the common narratives between the parents, representing shared events and experiences: *‘Investigating and Fighting: “Something’s Not Right”*; *‘Making Meaning of the Diagnosis’*; and *‘Living a Normal Life’*. Links to relevant theory and guidance have been made in parallel to help maintain the narrative and provide context for the findings.

Chapter 1: Investigating and Fighting: “Something’s Not Right”

This chapter includes two themes, which outline the parents’ experiences of acquiring practical support and information: *Investigating* and *Fighting*.

Investigating

The first theme describes two times when it has been necessary for parents to investigate: initially, when concerns arose regarding their son’s health, and secondly when trying to understand and find solutions to the implications of the diagnosis.

Prior to diagnosis all the parents had identified that their sons’ were not hitting the developmental milestones at the same times as their peers or siblings “...[they were] not developing as fast as other children were” (April). Lucy described how she compared her son’s ability to his younger sister: “...he would not charge up the stairs...” like she did. The process of comparison demonstrates that parents have a story of “normal” childhood development and that when development is delayed this implies there is “something’s wrong” (Sarah, Dawn, Bob, Lucy, Raphael).

Parents’ desire to compare is in line with Festinger’s social comparison theory,⁴² which states that individuals often “...make upwards comparisons in order to learn how to cope more efficiently”.^{17(pp1165)} An instinctive coping strategy for parents of ill children is to problem solve, usually through seeking advice from a medical professional.⁴³ The majority of parents

in this study (Sarah, April, Raphael and Danielle) relied on health care professionals to provide them with information. If applied to the stress and coping model,¹⁸ information seeking could be a means of appraising the stressor to determine a suitable coping strategy.

The assessment of symptoms was often found to be long and disempowering. Both Sarah and Danielle commented that their initial concerns were disregarded. For example, Sarah was initially told by a health professional that "...if I looked at every kid that's broke one leg and they complain they've broke the other I'd never be away from this place", whereas, April's process of investigation was initiated by a health professional.

Contrastingly, Lucy, Dawn and Bob all expressed how they were empowered to take control of the investigative process due to the skills and knowledge gained in their work or from their partners (Bob coincidentally works with boys with DMD, Lucy's husband was a medic and Dawn worked in an allied field). This meant they had the confidence to seek information when they felt it was needed: "Oh I had to push to get to see the right [person], the first paediatrician basically just ignored everything I said... I just said, how do you know it's not Duchenne? And I think they knew, but they're not going to say outright. They were going to do more assessments..." (Dawn)

Dawn's remark suggests hesitancy from professionals in investigating and confirming a diagnosis. This may feel like a necessary action professionally given the expected impacts of the diagnosis. However for the participants in this study the delay was often viewed as obstructive to the parents' role as information seekers, who were trying to solve what was "wrong" with their son.

Parents found themselves in the information seeker role again after diagnosis. A lack of signposting and information by services left them questioning how to practically manage the condition and its financial implications, e.g. how to get new equipment: "...we don't

generally get that signposting anywhere...” (Danielle). Lucy tried to find other parents to talk to but did not know where to look: “It’d be a lot easier if there was something out there....a group of mums and dads and I could say “So what chair do you use?...you know but things like that, I don't feel like there's anything.” Eventually she found information and contacts through the annual international conference organised by the UK based charity Action Duchenne.

All the parents sought information until their questions were answered. It is important to note in the case of DMD this process may be particularly difficult, because DMD is a rare disease with few points of comparison for parents. Consequently, stressor appraisals¹⁸ may be made in relation to parents of children with other chronic illnesses or no illness, which may set unrealistic expectations. Health professionals are often the means by which parents attempt to fulfil their role as an information seeker and support their role as a healthcare provider. When these roles are hindered this may be when parents construe professionals as being in opposition and parents do not feel heard.

Fighting

The words ‘fight’ and ‘battle’ appeared frequently across many interviews (see appendix 2H) and indicates that these parents see themselves as being in opposition to the professional from which they were seeking support. This is reflected in the parents’ narratives of professionals as obstructions when seeking information about their son’s delayed development, but more crucially when seeking practical support after diagnosis. April, Sarah, Danielle, Lucy and Raphael all stated how they had to fight to have their concerns heard relating to the planning of the adaptations to their homes. They felt they were denied the opportunity to have constructive input into the design, or personal financial input, if the grant would not cover adaptations that would best suit the family’s functioning: “... the things they had planned to

change [to] the house... I didn't like, I didn't think were appropriate.....It was just, it felt unacceptable so I decided to move house and do the adaptations ourselves” (April)

Sarah and Lucy felt that services were not empathetic and thought they needed to “...walk in our shoes for a week and then go back to do the job they’re doing and then they’ll see it differently” (Sarah). Lucy expanded upon this narrative by reflecting upon the professionals’ own roles and identified that they had their own agendas, which was perhaps at odds with her own: “I just felt there was a tick box exercise. She had to come to the house, she had to say she’d been and that was it.”

Unfortunately these apparent conflicts of agendas meant that often the adaptations were not appropriate for the family, even when the family offered to pay the additional costs: “...they didn’t liaise with you throughout it all, it’s like I’ve got a garden that the kids can’t even access, and when I put it to them, look can you not come and do something with the garden I don’t want you to pay for it...I have to walk on a wall to peg my washing out, because I can’t reach the washing line if I’m not on the wall.” (Sarah). After initial consultations some parents (Bob, Dawn, April, Raphael) who had the finances felt that it would be better for them to avoid continuing to fight and take control themselves by making the adaptations themselves and/or moving house. For those the fight was avoided.

These findings represent a common narrative of parents fighting to get support and often feeling that services are in opposition rather than working with them. Consequently, the interactions between parents and professionals provide a decision point in their narrative that forces parents to either accept the adaptations as the professionals present them, or do it themselves.

Overall the parents reported disconnected and sparse care, demonstrated by the lack of information and parents’ need to fight for support. This narrative of a lack of specialist

support is supported by the recommendations within the recent professional guidance literature outlining the need for standardised care pathways and funding for neuromuscular diseases.^{5,6} This is important to consider clinically, as perceived high levels of support are thought to contribute to successful adjustment.¹³ Chapter 1 therefore represents the obstacles to adjustment in the parents' stories and how they fought to overcome them.

Chapter 2: Making Meaning of the Diagnosis

All the participants told the story of the diagnosis and its implications, describing the events, experiences and their viewpoints. This chapter includes two themes: '*Diagnosis and Prognosis*', and '*Impact on the Participant's Identity*'.

Diagnosis and Prognosis

Parental interpretations of the diagnosis were represented in their language. Parents described the diagnosis as either a 'confirmation of their fears' (Lucy) or as 'coming out of the blue' (Raphael). Many used the world as a source of comparison - "that's when our world collapsed a little bit" (Danielle), - to describe the magnitude of the emotions they felt due to their interpretation of the diagnosis as "...a death sentence" (April).

All the parents described the process of receiving the diagnosis and prognosis as a negative, uncontrollable and hopeless experience, e.g. "In reality there is very little that you can do." (Bob). This is important as parental appraisal, particularly perceived control,^{13,22} optimism and hope,^{22,44} are thought to play a key role in parental adjustment to a chronic health diagnosis.

Parental understanding of chronic illness has been found to impact upon their expectations and intuitive ways of coping.⁴³ Parents were found to understand most illnesses to be acute, temporary and controllable or treatable, for example having a broken arm. The parental role

is therefore seen as to reassure, seek an 'expert' opinion to restore health,⁴⁵ and minimise discomfort,⁴³ all of which were seen in Chapter 1 and are incongruous with a diagnosis of DMD. Contrastingly the chronic illness model focuses on managing the condition sufficiently that families can 'live well' and accept an altered life.⁴³

It appears that the parents in this study have adhered to the model of acute illness and restitution (e.g. hoping for a cure "I know there's not a cure, but obviously we're fighting, we're fighting to try and find something" [April]), which promotes values of personal responsibility and control to fight disease and return to a state of normalcy⁴⁶ If this narrative is held by the parent of a child with DMD it may cause them to question their ability to fulfil their parenting role. The disparity between the hoped for 'normal' future and their interpretations of the prognosis may also lead them to experience their son's future as a loss of a normal life.

This sense of loss may be influenced by societal narratives of disability. For example, within the UK people with a disability are often presented as "less productive, needing to be cared for and getting in the way..."^{47(pp7)} and less able to live a full life.⁴⁸ This is apparent in the parents' descriptions of loss and grief regarding their son's physical ability and eventual decline compared to their healthy peers: "...I'd see all the kids on the front playing football, (son) used to go out and sit in his chair watching. Aww god it used to tear me apart." (Sarah). However this was successfully disputed for Danielle when given a positive role model, suggesting parents' expectations of possible futures are potentially inaccurate: "...seeing now that it'd be quite possible to have a conversation with him...Before then it was just like that (son)'ll never go to university...get a job. And it made me think that you know he can have a life rather than just disabled..."

This narrative has also been challenged by superior outcomes for individuals living with DMD in Denmark and the Netherlands, demonstrated by higher rates of supported and independent living.⁶ This is significant clinically as negative illness representations and helplessness are thought to be linked to adjustment difficulties,¹³ which would be perpetuated by negative societal disability narratives.

When this information is applied to the stress and coping model of adjustment¹⁸ it is plausible to see how parents, immediately after the diagnosis, can appraise the stressor as insurmountable (e.g. “a death sentence”) and perceive themselves as unable to cope (e.g. I am either a professional or I am a carer) as they have limited experience. They then subsequently negatively appraise the efficacy of these strategies (e.g. I cannot return his health so he will always have a disability), meaning by their own standards they are constantly failing to adjust to their son’s condition. From a narrative perspective, given the combination of the narratives of restitution, parental responsibility and disability as a loss and the genetic nature of DMD, it is understandable how parents would feel that “it’s their fault”.

Adjustment could therefore be identified as the process by which parents move from an acute to a chronic illness model, moving from the dichotomies of healthy or unhealthy, life and death, disabled or able, and professional or carer to a more integrated understanding of living well.

Impact on the Participant’s Identity

This sub-theme outlines parents’ experiences of how their identity has changed or how they expect it to change as their son’s health declines. Parents described several roles, including being the protector, the sacrificer and the carer.

Once diagnosed the parents felt strongly about protecting their sons against the emotional and psychological implications of the diagnosis. This was performed in two ways: through minimising the impact of their emotions on their son and by sensitively explaining the diagnosis.

For example, through counselling, Lucy became more aware of her emotions and how these may affect her son: "I've got to get myself sorted, got to be right in myself because that influences him....". Lucy endeavoured to protect her son by carefully explaining the diagnosis and trying to "... put a positive slant on anything...". Dawn also carefully shared the diagnosis with her son, balancing the truth and appropriateness of the explanation she gave: "We've always said we'd be open and honest with them. Obviously within the scope of where they're at".

Due to both her sons being unable to walk Sarah described herself as a mother, to the exclusion of other parts of her identity, which manifested in her feeling obligated to stop working: "...I don't feel like a person in my own right if you like...I just felt that something had to give and it had to be me..." Sarah described her family's values of working: "When you come from a family that's always worked, that's what you do, you go out to work..."; however when she sacrificed this aspect of herself in order to care for her sons she felt like a "second class citizen". This narrative is sadly supported by the Department for Work and Pension's report documenting the public perception of those on benefits as 'idle'. ^{49(pp13)}

Dawn shared Sarah's feelings of sacrifice despite working part-time. She noted the limitations placed upon her work life, e.g. time out of work to attend hospital appointments. This then led to a conflict of roles, specifically the desire to develop a career versus caring for her son: "I'd like to do more... But can you take it on, because you've got so much going on with him."

Dawn's narrative was situated in the present and related to current experiences, whereas Bob expressed comparable concerns but relating to the role he feared he would have to lose in the future: "I still like to feel that I'm the top of my game. And I might have to give that up I think, but that wouldn't be very comfortable."

Parents may feel particularly helpless and possibly resentful towards these changes given that they expressed that they felt forced due to a lack of alternatives. Dawn and Sarah both discussed feeling forced into a caring role due to their desire to help and a lack of choice. This was represented by a dichotomy between the parent and carer roles: "We try and keep it very much a parent.... You do get pushed to do caring role because you haven't got a choice." (Dawn); "...because I'm the mum I've got to do it. Aren't I doing above and beyond of that of a mother, how many mums do you know who wipe their 18 year old's backside?" (Sarah)

In summary, this chapter has explored parents' narratives relating to the meaning they make from the diagnosis. These include: diagnosis as "a death sentence", disability and illness as a loss, and being forced to take on an alternative role.

Chapter 3: *Living a 'Normal Life'*

This chapter has two themes: '*Living Day to Day, Normalcy and Happiness*' and '*The Present versus the Future*'.

Living Day to Day, Normalcy and Happiness

Parents stated that they eventually entered a period of relative stability in which their son's physical deterioration slowed "...between age 4 and 5 where with Duchenne they plateau..." (Dawn). They felt this allowed them to live more day to day and return to the task of being a 'normal' family once again before having to consider the impacts of future events: "...other

things get in front like making sandwiches for school and day to day run ins” (April), which then reminded parents that their children’s identity was more than the DMD and consequently served as: “Sort of a reminder that everyday issues you're dealing with your children are because they're children...not necessarily because he has DMD.” (Lucy)

In addition, parents discussed how they strived for normalcy: “...what we try to do is to live as normal a life as we possibly can” (Bob). This was present when they compared parenting to their healthy children: “And when he's naughty he gets told off, like his brother.” (April). Raphael also spoke about treating his children similarly or rather “fairly” and “equally”.

In some cases ‘being normal’ and ‘living day to day’ became a way of coping by ignoring their son’s disability. This was temporarily possible as their sons’ condition was stable and therefore they had fewer reminders: “...we, just generally tried to ignore it I think. And that was quite successful in some ways until the last couple of years... but we buried our heads in the sand....And we by and large, we tried to live life as normally as possible.” (Bob).

The following quote from Raphael suggests parents’ ideas of normal is influenced by their own upbringing and that happiness was integral to that: “...doing just the family things that I remember from my childhood, doesn't have to be anything fancy but just spending time doing something happy really.”

Being happy and providing happiness (“...as long as he's happy” (Dawn)), was an intertwined narrative with striving for a normal life, which was often linked to living day-by-day: “...you know be positive and happy and enjoy every day that's what we do” (April).

Some families adjusted and felt happiness by reflecting on the aspects of their life that they considered normal and stable: “And I think that's sort of like a coping mechanism I suppose, knowing that there are people far worse off than me. At least my kids are happy, they're as healthy as they can be, I've got my health, I've got a home.” (Sarah)

For this sample of parents providing ‘normalcy’ and ‘happiness’ were important values that they endeavoured to uphold, which may stem from their own childhoods. This would support the proposition that approaches to parenting are influenced by the individual’s experiences of familial, cultural and societal narratives,⁵⁰ for example legislation regarding children’s welfare (e.g. the promotion of happiness in the Convention on the Rights of the Child).⁵¹ In this instance their subsequent beliefs identify them as being responsible for their children’s happiness, which given the additional circumstantial difficulties makes these expectations more difficult to achieve and may not match their experience. It is apparent that parents come to embody the role of happiness and normalcy providers; however this role may feel futile if the parents believe their child is incapable of a normal life, as suggested in Chapter 2.

If a stress-coping approach¹⁸ is applied then parental experiences of vigilance,⁵² perception of reduced parenting self-efficacy⁵³, having to maintain many roles (e.g. health care provider, problem solver),⁵⁴ fear and insecurity about the future,⁵⁵ become understandable as a response to perceiving PCI as a threat to their parenting identity.

The Present versus the Future

Distraction and ignoring the prognosis were actions used to manage thoughts of their son’s future and minimise their own distress:

“...my concern in all of this in the way that I have adapted to his illness is that I can say I've always worked hard and always enjoyed it but it has become, it has at times been a shield, in a sense that I've kept myself very busy because I haven't really wanted to think about (son)’s prognosis so I have used it (keeping busy) as a bit of a crutch I think.”

(Bob)

Parents most frequently used language like “not dwelling” and “getting on with it” as the means by which they adjusted at this time: “I think sometimes if you sit and dwell you’ll never get out of the bed in the morning because you feel so sorry for yourself and your kids” (Sarah); “...the future we don't know what that holds it's just something we try not to dwell on” (Danielle)

These quotes state that parents feel that if they think about the prognosis they will reside in those thoughts. The counter to this position of stagnation is to keep moving and “get on with it”. This is confirmed by parents describing characteristics such as being “organised”, and “pragmatic”, which continue to describe parents actions of moving forward by being “proactive” and living day to day. However these terms have a more positive connotation. This aspect of their identity allows them to manage the daily difficulties of the condition without having to think too far ahead. Dawn encapsulated this sentiment in two quotations: “I think a lot of that is doing research planning ahead, being pragmatic and not dwelling on the crap side of it basically”; “Don’t think too far ahead, because it's not a pretty picture.” (Dawn)

These ways of thinking and acting allowed parents to feel adjusted to their current situation. It moved them from a position of extreme and seemingly uncontrollable negative emotions to a position of power and control i.e. they can choose to get on with life or let life and its miseries keep them still. Adjustment was described as an ongoing process dependent on the family’s circumstances. This became relevant when parents talked about the future and the uncertainty that it held for them: “What I'm uncertain about is how we will adjust and in particular and how will we fit our working lives around the need to care for (son)” (Bob). This suggests that the parents recognise that they will need to adjust again as their son deteriorates and eventually dies: “...the only thing that sort of concerns me is when I've no

longer got the boys what do I do? But I try and not think about it ...because at the moment there's nothing I can do until that point comes.” (Sarah)

The only certainty is that their son’s prognosis is fatal, whatever the circumstances around this, i.e. when, how he will cope until then, and how the parents will adjust, are uncertain. By “not dwelling” on an uncertain future parents can live in the now and provide a normal life. However they are aware that the next major event in their story will come and they will need to make meaning of this as they had with the diagnosis. In thinking about the future, they would be revisiting the events and narratives that were present at the diagnosis.

Conclusion

This study gives voice to the experiences and meanings that parents make when trying to adjust to having a child with DMD. Models of adjustment have previously focused on specific categories that relate to discreet factors without providing the context.^{13,20,22} These findings provide the context for these factors and consequently are able to explore the subjective ‘what’ and ‘how’ of adjustment. A strength of the narrative methodology used is that it elucidated why parents make certain appraisals of their situations. In addition, by focusing on DMD directly rather than PCIs, the findings also identified how the pause in their son’s deterioration allowed time for reflection.

By using an inductive approach the study begins to challenge the idea of adjustment as a final destination that has reliable factors that can be measured. Adjustment could represent parents’ movement from an acute to a chronic model of understanding based on the illness specific information that they gather, integrated with their cognitions based on previous experiences. These cognitions reflect personal and cultural narratives relating to their identity. This definition would encompass the narratives parents described in this study (including being an information seeker and fighter, diagnosis as “a death sentence”, disability

and illness as a loss, being forced to take on an alternative role and not feeling valued, being ultimately responsible for their children's health and being a good parent), which informed their appraisals of stressors and subsequent affect response and whether they felt 'adjusted' or not. This understanding of parental adjustment to DMD also accounts for Moss-Morris's¹³ comments regarding changes over time, which can be explained by parents being introduced to new information, which helps them to make sense of their experience and provides a new subjectively 'acceptable' identity with new roles and strategies.⁵⁶ This definition of adjustment provides new therapeutic avenues and modes of intervention.

Clinical Implications and Future Research

The adjustment models of Wallandar and Varni²⁰ and Walker²² do not mention the important role that health professionals play in parental stories of adjustment, however Moss-Morris¹³ alludes to the importance of these relationships in the review but does not provide clinically useful detail. The narrative analysis highlights two main areas of intervention for health professionals that would provide a multi-disciplinary model of working to support parents throughout the care of their son.

The first aim would be to minimise parents' need to seek information and fight for support. This would mean considering the diagnostic process and ongoing care to provide accurate information regarding the disease in a timely manner. This could be addressed by a standardised pathway for diagnosis that particularly considers how the diagnosis is sought and shared, paying attention to validating parents' concerns and providing a message of hope. To meet these needs clinicians should consider the diagnostic process in terms of: who is invited to appointments⁵⁷; considering how the diagnosis is explained, in particular what information is given when; ensuring it is a person centred meeting with relevant signposting.

Parents will also require practical support. Signposting to peer support might be helpful when parenting concerns arise related to the disease e.g. how to discuss the condition with their son. However more practical information might be more efficiently given by a healthcare professional after diagnosis. This could be face to face or in the form of a series of leaflets addressing FAQs at key stages. For example, what needs to be considered when getting a wheelchair and/or signposting to other sources of information like the charity Action Duchenne. Parents may also need help with completing forms for extra support (e.g. disability benefit forms) and advocacy to ensure they receive the support they need (e.g. when discussing adaptations for their house).⁵⁸

The second aim would be to help parents make meaning of their circumstances and understand their reactions. First, it might be helpful to share the findings of this and other relevant studies that provide insight into parental experiences with parents to help 'normalise' their reactions and provide hope. Peer support could also address this need but it would need to be organised so that help came from parents at a similar stage. This might also help them to remember that they are still a parent with normal parenting concerns.

Sharing these findings with health professionals will allow them to keep these in mind when talking with families, to help identify potential issues related to adjustment. If identified, a stepped approach to care dependent on the level of need might be beneficial, with health professionals initially assessing and supporting the parents. The aim would be to encourage thinking about DMD as a manageable chronic disease, whilst normalising their experience. Dependent on the level of experience of the health professional and the distress of the parent, this role may need to be facilitated by a mental health specialist i.e. a clinical psychologist or require referral for direct therapeutic involvement.

Dependent on the presenting issue, clinical psychologists might consider:

First, helping them discern acute versus chronic illness models, breaking down their understanding of restitution and where this knowledge comes from. More specifically cognitive behavioural therapy⁵⁹ could be used to recognise and challenge their thoughts regarding their son's ability to be happy and live a normal life.

Second, acceptance and commitment therapy⁶⁰ to acknowledge their parenting ideals, whilst helping parents accept what is out of their control and encouraging them to recognise how they can continue to follow their values.

Last, narrative work^{61,62} could also be used to identify their changing roles and understand how they can coexist. It could also externalise the diagnosis to minimise parents' experience of blame and burden.

Potential Limitations of the Study

A potential limitation of the study was the amount of service user and participant involvement in the design and analysis, an increase in which would arguably make the study more valid. For example participant feedback on the interview questions may have shaped the schedule, perhaps tailoring it to chronic illness experience. However, it was felt that due to the exploratory nature of the research, broad open questions and prompts would be sufficient to allow the participants to discuss their relevant experiences. More service user involvement may be beneficial in the future when the research question is more refined.

Initially member checking was considered as a device to ascertain validity, however during development of the chapters it was felt that participants' involvement in the analysis, would not necessarily be appropriate. The chapters represent the entire dataset, not just individual narratives. Consequently the participants should recognise themselves in the narratives but the analysis will not be a representation of their story alone, thereby making it difficult for

them to assess how valid the chapters are to the sample. This argument is supported by the researcher's critical realist stance whereby the researcher identifies 'tendencies' across the dataset which are dependent on context. This combined with the narrow timescale of the study led to a re-evaluation of its necessity. However member checking may be more useful should individual narratives be focused upon in more detail.

A limitation is that member checking may have been useful for the summary stories, which focused on a salient characteristic that each participant embodied during their narrative. These stories did not aim to provide an overall summation of each participant's experiences; they instead aimed to highlight one thread that directly affected their adjustment. This formed an additional extra to the main analysis that might help practitioners to identify individual needs but due to time constraints participants' feedback on these characteristics was not ascertained.

The sample of parents in this study was small and culturally homogeneous, which was beneficial as these similarities provide a detailed understanding of the experiences of the group leading to clear applications and recommendations. However the small variation in available social support, socio-economic and particularly financial statuses represented may mean that they are more able than other parents whom are less fortunate to deal with multiple life events and other social stressors that might affect adjustment. Consequently future research could focus on how multiple life events and varying social and cultural narratives impact upon adjustment.

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Tables and Figures

Figure 1.

Timeline of the major physiological changes associated with DMD

One to three years

- Children with Duchenne MD usually start to have noticeable symptoms between the ages of one and three years.
- The muscles around their pelvis and thighs tend to be affected first and often appear bulkier than normal.

Three to eight years

A child with Duchenne MD may:

- have difficulty walking, running or jumping
- have difficulty standing up
- learn to speak later than usual
- be unable to climb the stairs without support
- have behavioural or learning difficulties

Eight to fourteen years

- As their muscles weaken and they lose the ability to walk.
- They can also develop scoliosis, which is where the spine begins to curve sideways. This can lead to one shoulder or hip being higher than the other.

Mid-teens

- Some people with DMD will develop dilated cardiomyopathy. This is where the condition affects the heart muscles, causing the chambers of the heart to enlarge and the heart walls to become thinner.

Late teens and adulthood

- People with DMD can begin to have breathing problems. The condition can affect the intercostal muscles (muscle tissue between the ribs) and the diaphragm (main muscle between the chest and abdomen used during breathing).
- Once the heart and respiratory muscles are damaged, DMD becomes life threatening. With medical care, most people with DMD die from heart or respiratory failure before or during their 30s.

Table 1.

A table of participant's demographic and contextual information

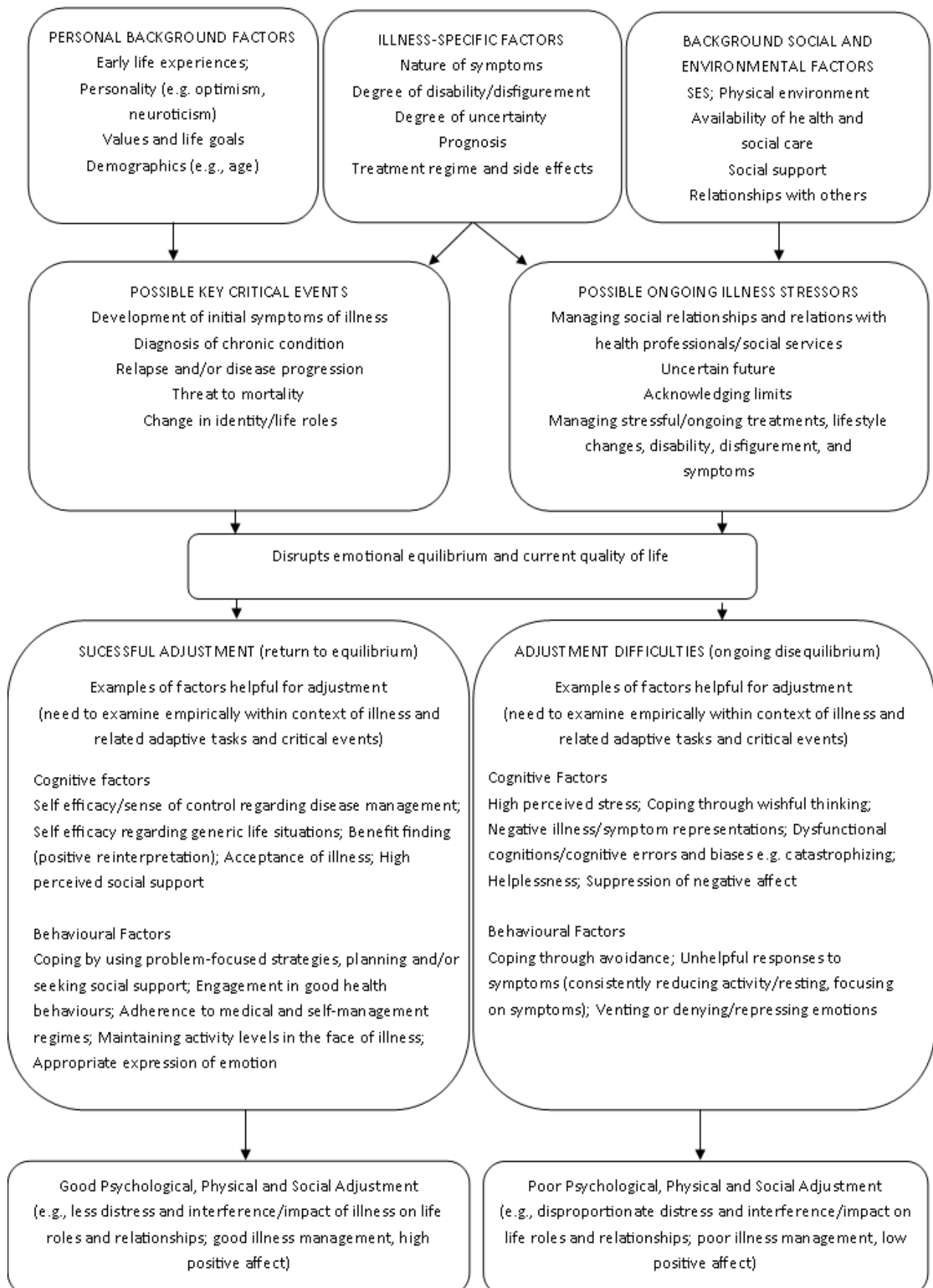
Pseudonym	Sarah	Lucy	Dawn	April	Raphael	Bob	Danielle
Parent related information							
Age	44	41	50	41	42	54	34
Gender	Female	Female	Female	Female	Male	Male	Female
Partner	Yes, second marriage	Yes	Yes	Yes	Yes	Yes	Yes
Work	Not currently	Yes, but type not mentioned	Yes. In research, part-time	Yes, in health, part- time	Yes, in health, full-time	Yes. In health, full-time	Not currently
Ethnicity	White British	White British	White English	White British	White British	White British	White British
Child related information							
Age of children with DMD (years)	18 and 12	10	12	8	8	14	8
Time since diagnosis	12 years	5 years	7 years	4 years	4 years	12 years	3 years
Number of children (ages in years)	2 sons (18, 12) 2 step sons (15, 12)	Son (10) Daughter (8)	Daughter (16) Son (15) Son (12)	2 sons (8, 6)	2 sons (8, 6)	Son (17) Son (14) Daughter (7)	Daughter (16) Son (8) Daughter (1)
Other relevant information	Both boys have DMD	Had psychological input				Works with boys with DMD	Living with mother in law

Appendix

- 2A Moss-Morris's model of adjustment¹³
- 2B Wallander and Varni's model of adjustment²⁰
- 2C Walker's model of adjustment²²
- 2D An example of the semi-structured interview schedule
- 2E An example from a completed interview and coding
- 2F An example of the key narrative elements
- 2G An example of a participant timeline which contributed to the brief summaries and chapter development
- 2H An example of similarities in the imagery and language used across participants
- 2I A diagrammatic representation of chapter 1 and its themes part way through the analysis
- 2J A diagrammatic representation of chapter 2 and its themes part way through the analysis
- 2K A diagrammatic representation of chapter 3 and its themes part way through the analysis
- 2L Participant summary stories
- 2M Pediatrics manuscript guidelines for publication
- 2N Pediatrics title page
- 2O Pediatrics contributor's statement page

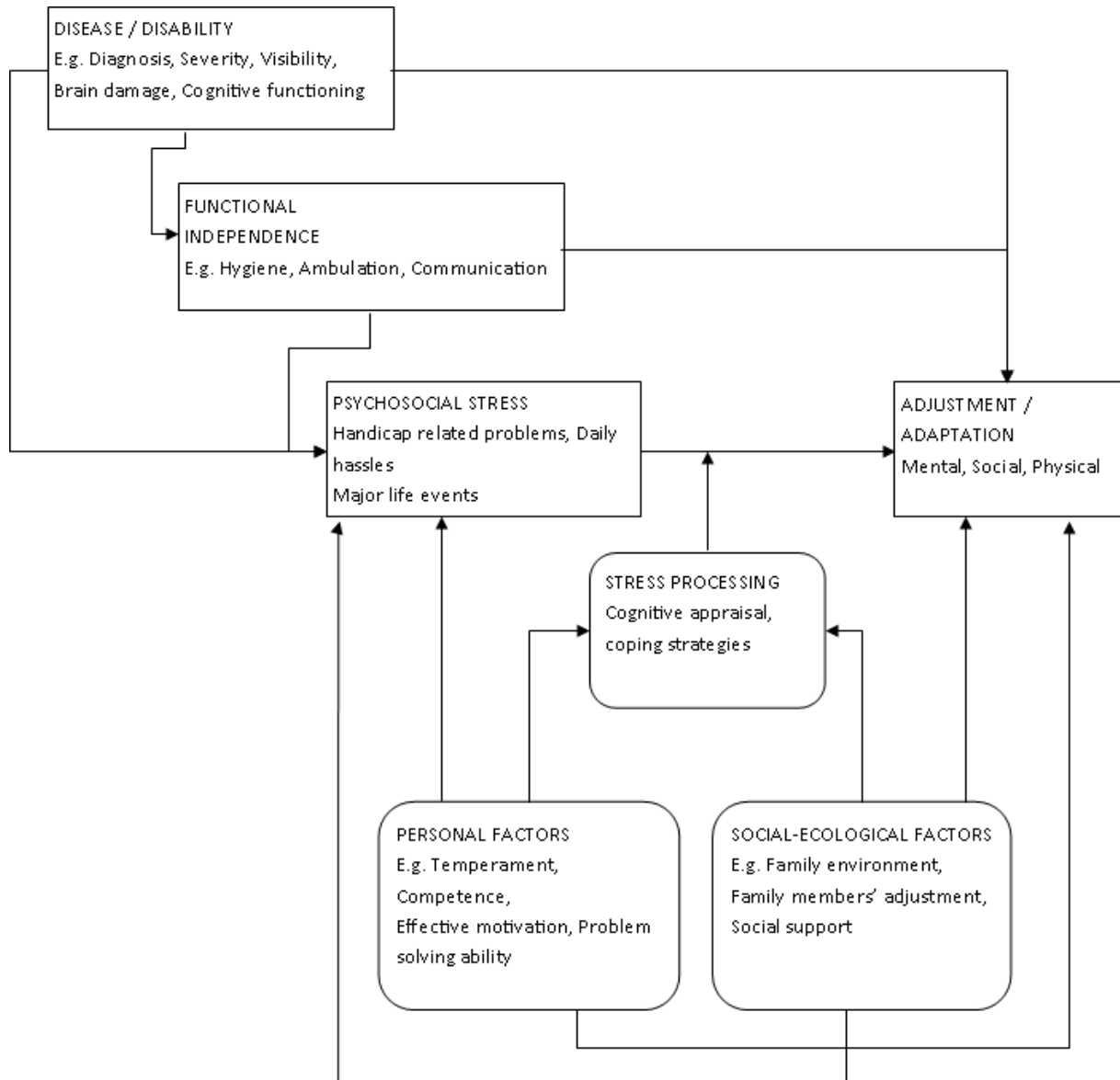
Appendix 2A.

Moss Morris' model of adjustment



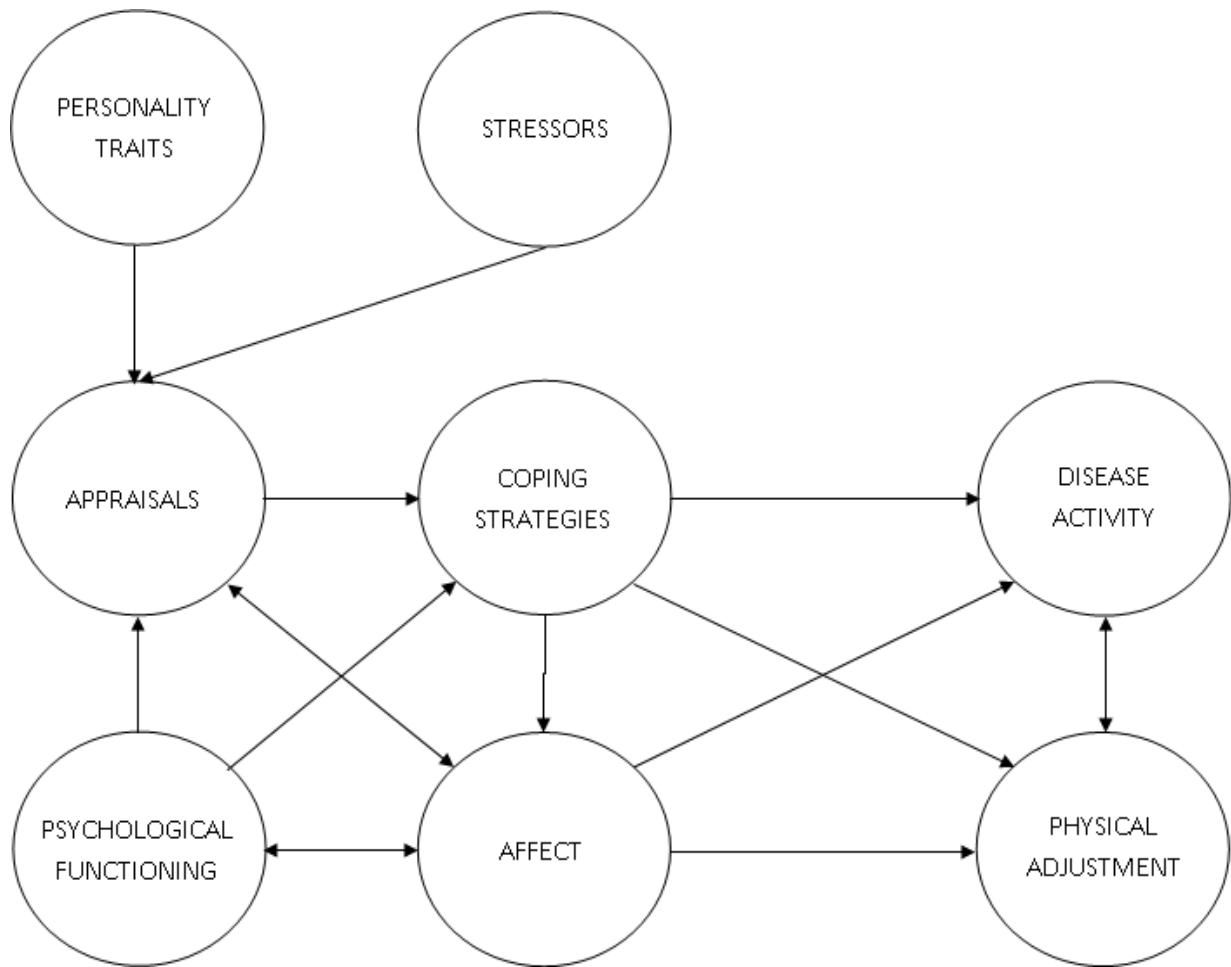
Appendix 2B:

Wallander and Varni's model of parental adjustment²⁰



Appendix 2C:

Walkers' model of adjustment²²



Appendix 2D:

An example of the semi-structured interview schedule

Opening question	Examples of Prompts
<ol style="list-style-type: none"> 1. Hello... 2. Make comfortable...Before we get started would you like a drink? (whatever is available: tea/coffee/water/squash). Biscuits? 3. Introductions 4. Go through the participant information sheet 5. Check criteria/demographic information 6. Consent form 7. So we've talked about risk and confidentiality and when I would need to break confidentiality. And I really want to make sure that all the information you give me is kept personal and private. I also want to make sure that you and the others around you are safe. 8. Right, I've already done a lot of talking and asked you a lot of questions. Is there anything you would like to ask me before we start? 	<ul style="list-style-type: none"> • My name and title • What a trainee psychologist is? • Why I am doing the research. • Age • The diagnosis must have been given at least 2 years ago. • Currently caring for the child? • The child must not have any other serious illnesses not associated with DMD. • The family must not be under investigation. • So to keep what you say private, I am using false names to replace the names of the parents I talk to. What name you would like to have on the interview transcript and any quotes I might use from our discussion? <ul style="list-style-type: none"> ➤ this is to make sure you cannot be identified.

<p><u>SWITCH ON TAPE RECORDER!</u></p> <p><u>Introduction to the process and topic area</u></p>	<p>I am interested in your adjustment to having a child with Duchenne. I want to know what you think adjustment is and what has been important to you along this journey.</p> <p>Today we will have up to an hour and a half for you to tell me your story. I may occasionally say something but I will try to keep this to a minimum. If you brought a meaningful item with you, you can use this to help remind you of things to discuss.</p>
<p><u>Specific prompts</u></p>	<ul style="list-style-type: none"> • So where is a good place to start? • What have the key events been in this process? • What has influenced your adjustment? • How has this affected how you see yourself? • Can you tell me what things were like before....(the diagnosis)? • So, tell me about after...(the diagnosis)? • And later.... • Did you bring an object that represents your adjustment? If so, tell me about it. • Who has been important on your journey? • Tell me about how you see your future. • Where would you say you are on your journey of adjustment? • <i>Any other events that you feel have been important to you?</i>
<p><u>Generic prompts</u></p>	<ul style="list-style-type: none"> • Tell me more about that... • Describe that to me.... • What happened next? • What influenced that? • <i>Why was that important?</i>
<p>Ask any questions? When and how can I contact you about the results?</p>	

Appendix 2E:

An example from a completed interview and coding

INTERVIEWER	PARTICIPANT ID	INTERVIEW DATE	INTERVIEW TIME	VENUE
Principal Researcher	April	24.03.14	9:30	April's home
DURATION	WORD COUNT	PART. GENDER	PART. AGE	
67 minutes 25 seconds	10,107	Female	41	

Colour key for concepts to be aware of (Informed by Crossley, 2007 & McAdams, 1993):

Roles	Potential chapters	Key events	Significant people	Personal thoughts/ideology
Narrative tone	Imagery	<u>Societal influences</u>		

TRANSCRIPTION NOTATION

- I:** Interviewer speaking
- R:** Respondent speaking
- ... Long pause
- (words) Transcribers interpretation/suggestion of what was said (if difficult to hear/understand)
- (-----) Unclear or imperceptible speech
- [0:00] Digital audio file time marker (appears in line number column at 5 minute intervals)

LINE No.	TEXT	NOTES/CODES
[0:00] 1	I: So erm, having explained a bit of what I'm looking for and	
2	hopefully that's clear, where do you think is a good place to	
3	start.	
4	R: From the beginning I think, when I, when we thought there	Beginning: thinking
5	was something wrong. we didn't, we thought there was	something was wrong
6	something wrong with (son) and um, he wasn't developing as	
7	fast as other children were, he was a very late walker he didn't	Comparing
8	walk until he was nearly two and he shouldn't develop,	development to other
9	developmental on with other children of his age and we noticed	children, Late walker
10	he was having problems getting up from sitting, he'd be sat	
11	down and then he'd really struggle to get up. He'd probably	
12	crawl over to something to lift himself up to get up, and he was	Not hitting milestones
13	just like an old man going up the stairs he'd be like ooh. where	- getting up from
14	as a child would be running up and the school noticed this, he	sitting
15	was in a preschool and he was, I don't know, he was at	Like an old man
16	preschool and he was under. I'm trying to think what age he	School noticed
17	was, he must have been about 3 or 3.5 and we had the physio	
18	involved and she was giving him, he's got very weak trunk	
19	muscles so she was giving him exercises and she said we'll just	
20	take a blood sample just so, just in case there's any underlying:	Diagnosis - through
21	we're missing anything, anyway they found out got em back	physio appointments
22	he'd got Duchenne muscular dystrophy, which I knew nothing	
23	about at all. But they didn't tell me, they made an appointment,	
24	it was our younger specialist paediatrician doctor and we went	

Appendix 2F:

An example of the key narrative elements

Participant	Key event/storyline	Roles/identity	Key characters	Language and imagery
Lucy	Suspicions Blood tests Diagnosis Telling son Visit from specialist nurse Son doing well Help didn't feel relevant No advice for parents Action Duchenne conference Finally getting advice Speaking to other parents Not telling anyone Time makes it easier Counselling Not wanting the wheelchair Genetic testing Loving my son again Remembering he is a child Why us? Grieving the loss of a son without DMD Psychological and practical adjustments Not knowing where to find support Receiving empathy Telling school Hospital appointments Life events— death in the family Physical activities Meeting other families at a similar stage Helping others go through it Clinical trials Parenting issues Dealing with his sadness Peaks and troughs Things getting worse	Diagnosis sharer with son Unsupported Unheard Knowing what to do/say Remover of blame Blaming myself Griever Hiding my sadness from son Not taken care of/or interest in Initially feeling weaker/less able to cope with life Needing normalisation of the process Need to make headway in some area/do something helpful Having some normality – being able to provide some normality Being positive Burden carrier Seizer of opportunities Having faith Now a stronger person More accepting of others feelings I've got myself sorted	Specialist nurse Her husband Her other child Friends Her son	"Our world fell apart."

Appendix 2G:

An example of a participant timeline which contributed to the brief summaries and chapter development

Events and possible chapters	Codes relating to the events and chapters
<p>Life before the diagnosis—Information seeking and fighting for support</p> <p>Suspicious Blood tests</p>	<p>Suspicious something was wrong Husband's role provides some insight Getting confirmation of their fears Referred to hospital for blood tests The tests to prove the worst Others wouldn't be able to tell Comparisons with sister</p>
<p>Diagnosis and making meaning/grieving</p> <p>Telling son Visit from specialist nurse Son doing well Help didn't feel relevant No advice for parents Action Duchenne conference Finally getting advice Speaking to other parents</p> <p>Not telling anyone Time makes it easier Counselling Not wanting the wheelchair Genetic testing Loving my son again Remembering he is a child Why us? Grieving the loss of a son without DMD Psychological and practical adjustments</p>	<p>Didn't think she'd react like this – it's better now? Finding out was hard. Our world fell apart It was hard to accept...it still is Hard to tell him – so not say anything at first No advice on how to be Visit from a specialist which was a waste of time Nurses role and perspective The help that was offered wasn't relevant Yes he has DMD BUT he's doing well Proof of the difference Nurse proposing a picture that didn't fit Events have proven the nurses timing to be wrong Advice was totally irrelevant No real support as parents No advice on what to say to him Support as a tick box exercise The box – visited the family, So felt nothing was provided Continuing as best as you can on your own, Conference –gaining information Information is beneficial e.g. how to talk to son - knowing what to say (detail in quote) Answer his questions in a way that's appropriate for his age Despite having advice – planned or blurted? Telling him what he had was hard Telling him so he didn't blame himself Not sure he understood At least providing an answer Being able to provide reasons No support for parents Conferences are reasonably good – information gathering Conflict - Speaking to other parents i.e. needs to be at the same age At the time he was doing well , that is not the case anymore Feeling up and down Diagnosis was the worst thing ever Coping by not telling anybody Knowing what would happen if it's said out loud. Hiding ourselves away for a bit Time makes things easier Counselling to deal with own influence on situation Not being a carrier but still blaming self Counselling was beneficial Not wanting to have the wheelchair Counselling had an impact</p>

<p>Not knowing where to find support</p> <p>Diagnosis and making meaning/grieving</p> <p>Receiving empathy</p>	<p>Loving my son again Feeling blame for loving him less Feeling better about self allowed reflection on relationship with him Talking reminds you children are children Remembering the issues are because he is ten not because he has DMD Talking is better than bottling up Why questions "Why us?" (why information might be helpful) Knowing that there's no way we could have known – and it started with him Initially how and why Meeting with geneticists Google (good and bad) What could I have done differently What could I have done differently? Having to go through a process Time is the biggest healer – getting used to the idea Questioning ability to accept it ever Getting used to losing somebody Having it explained like grieving The loss of a child without DMD Conflict: Acceptance increases whilst his ability decreases Two adjustments getting used to the idea dealing with the practical consequences It's emotional support that I wanted Not knowing where to find support More emotional support in the early stages More support from the specialist nurse Signposting for emotional support Nobody says 'how are you? How are you coping?' It's such a big thing Somebody of that ilk – professional? What to expect – normalising how awful Gaining information without going on the internet Timing of information – not seeing pictures If you google DMD you get wheelchairs and ventilators You don't want to know it You don't want to know it then especially Other parents – setting expectations and letting you know it will be ok Having someone with specific understanding Expressing how I feel Receiving empathy affects the parent and has a knock on effect for the child You will feel like..... It's ok to feel all of the emotions It's like a grieving process It's ok</p>
<p>Live day-to-day</p> <p>Telling school</p> <p>Hospital appointments</p> <p>Life events— death in the family</p>	<p>Telling the school. School have been supportive/amazing. Anything we want/need we can have. Hospital appointments -hard at first, hard for him You get used to it – appointments Rarity means there aren't many parents to get support from. Support from friends and family ebbs and flows Interest in how you are declines with time It's just the way it is Making headway – accepted onto trial</p>

<p>Live day-to-day</p> <p>Physical activities Meeting other families at a similar stage Live day-to-day Helping others go through it Clinical trials</p> <p>Parenting issues Dealing with his sadness Peaks and troughs</p>	<p>Something's happening. Life is up and down It depends on what else is going on in your life Coping threshold reduces with events Feeling less able to deal with the life events others also experience Life events stop progression? Cope less well than used to Conflict: Talking about it makes it easier Coping mechanisms are weaker Coping together with partner Biggest negative influence – lack of external support Sibling relationship impacts</p> <p>She's amazing with him Siblings allow you to do the more physical family activities – feels like avoiding the word 'normal' The trials feel like you're doing something (to help?) Faith helped Physical activities to cope Coping by contributing Exercise makes you feel better in yourself Friends doing extra to include him in normal things Friends wanting to include him Being a normal 10 year old boy Friends make life more normal for the family Meeting other families with children at a similar stage Helping others go through the process Providing the support you wish you'd had He's very aware of what he can't do I can't reason with him or explain why Trying and failing to explain Trying to be positive Coping with it for each member of the family is cumulative but makes it easier for them Repetition of 'burdened' Burdened with more than just what's in it for you Life has to go on, live day to day Make the most of what you've got It's created opportunities Opportunity - trials meant spending 1-1 quality time together Go with the flow and accept it Moving on is better than dwelling Talking to God Try to enjoy life Shut out the not so pleasant stuff Having someone positive (husband) Finding the good side is therapeutic Time to get used to the idea It's not as bad as it was initially Conflict: I am a stronger person More accepting of others feelings Others not being tolerant of her sadness Feeling rubbish but saying I'm fine Using experience to understand others I'd rather have their life à everyone has hard things Conflict: I am a stronger person</p>
<p>The immediate future</p>	<p>Not speaking to parents of older children They're so much further down the line</p>

	<p>I don't want to know that bit yet More support from professionals opposed to friends and family Conflict: meeting other parents, but hasn't tried Stage appropriate conversations (transitioning) You find your way Mums and Dads for practical peer support – which wheelchair? Helpful conversations are currently relevant A stumbling block is our house Once the practical stuff is sorted the heartache of seeing his struggle is reduced</p>
<p>The blank chapter Things getting worse</p>	<p>Dealing with going downhill Dealing with using the wheelchair Initially having hope that it'll never happen Facing the reality These things will happen Everything will get harder It won't get any easier because he will get worse Hoping for a cure</p>

Appendix 2H:

An example of similarities in the imagery and language used across participants

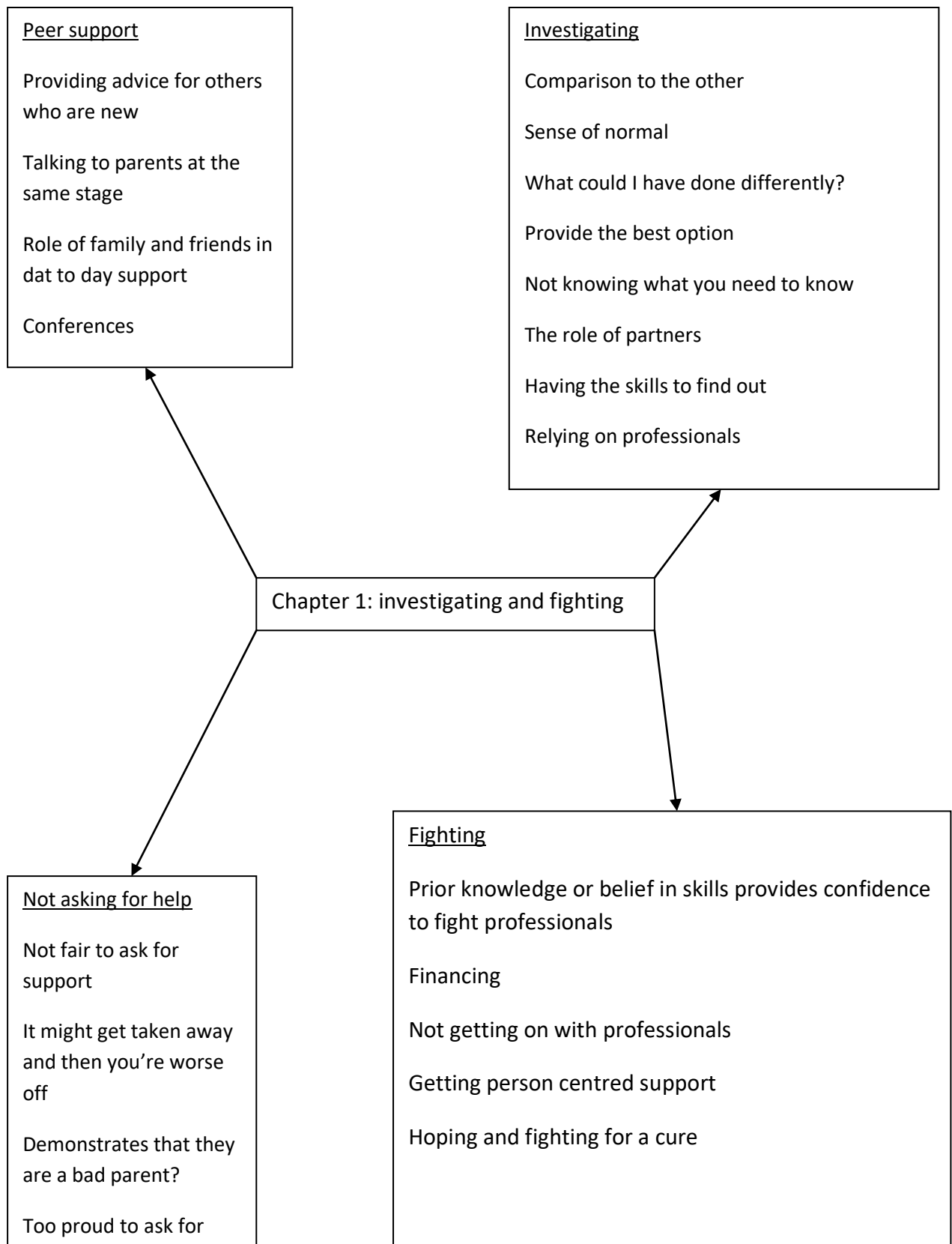
Similarities	Sarah	Lucy	Dawn	April	Raphael	Bob	Danielle
Fighting language - battles	<p>"...you do have to fight..."</p> <p>"For me the main issue's the battles you have to fight"</p> <p>"...you fight each battle as it comes..."</p>		<p>"but it's whether you fight for more support, to get help"</p> <p>"But yeah my role is his mum, and I am fight doing as much of the caring role as I can within reason because I don't think it's fair"</p> <p>"I'm kind of fighting my corner a bit more"</p> <p>"it's like you've got enough crap on your plate without thinking oh jeez here we go another battle"</p> <p>"I mean it's always been a battle with him doing homework"</p>	<p>"I know there's not a cure, but obviously we're fighting, we're fighting to try and find something"</p>			<p>"so we kind of got that extra help but I know that people are still fighting whose boys are 10 or 11 just to try and get a lift in that accommodates the wheel chair"</p> <p>"AND she's been fighting and fighting and fighting and to be honest she's on her own. Unless you can afford, or you know, you kind of know somebody, you are on your own quite a lot."</p>

Other similarities in language and imagery included references to:

1. Normality
2. Living day to day/each day as it comes,
3. Not right/something's wrong,
4. Obstacles/stuckness/struggles/barriers/problems,
5. Powerlessness/lack of choice/control/not much you can do,
6. Sacrifice/giving up,
7. Dwelling,
8. Sadness/shock of diagnosis e.g. heart-breaking,
9. Getting on with it,
10. Ups and Downs/downhill,
11. Guilt and blame,
12. Burdens,
13. Death and/or grieving,
14. Strength,
15. Being realistic/pragmatic,
16. Restrictions/limited/stop/constraints,
17. Mollycoddling/cotton wool/overly worry,
18. Uncertainty/not knowing/not wanting to know/not sure,
19. Being unsupported/overlooked
20. Knowledge.

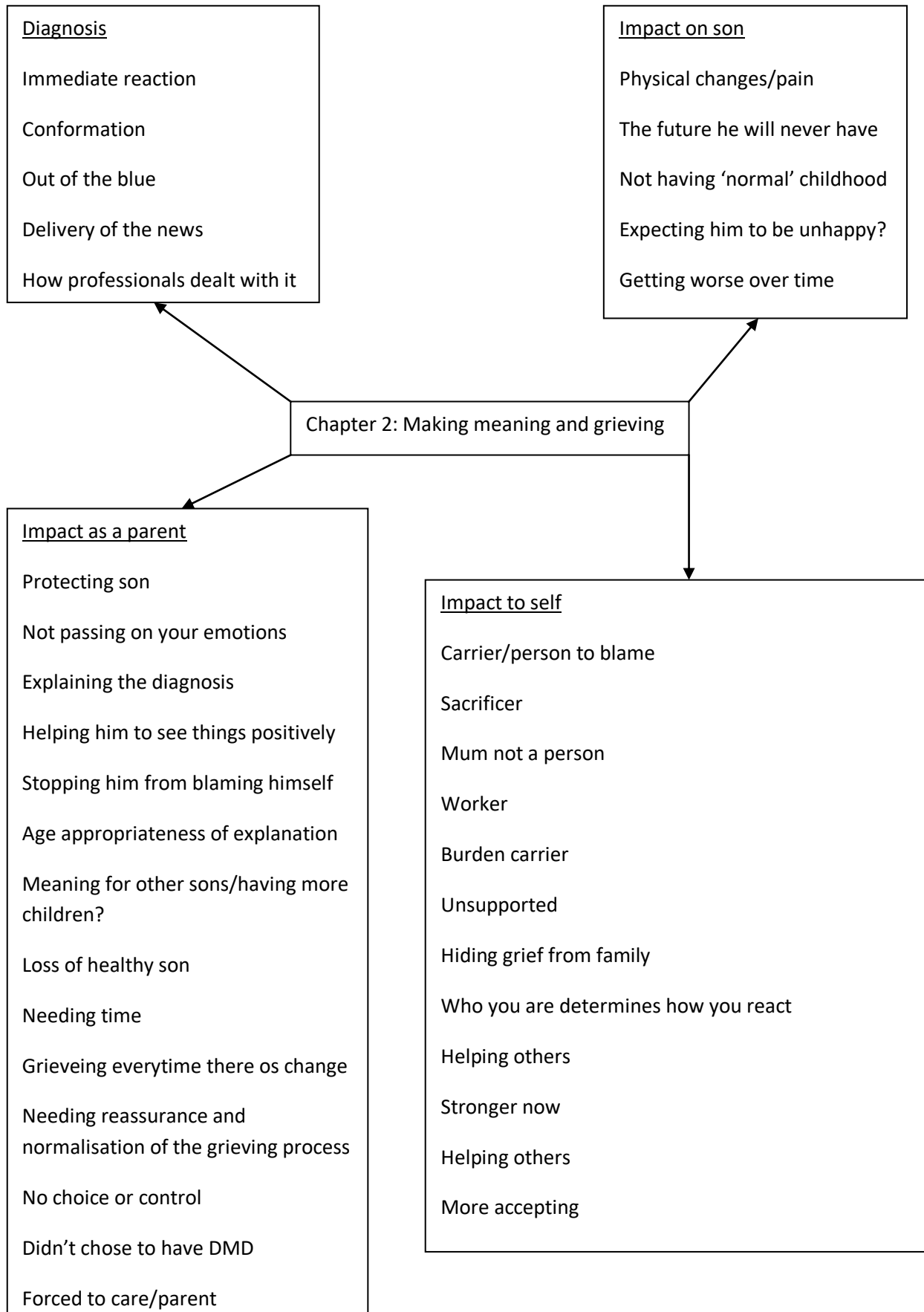
Appendix 2I:

A working diagram representing chapter 1 and its themes part way through the analysis



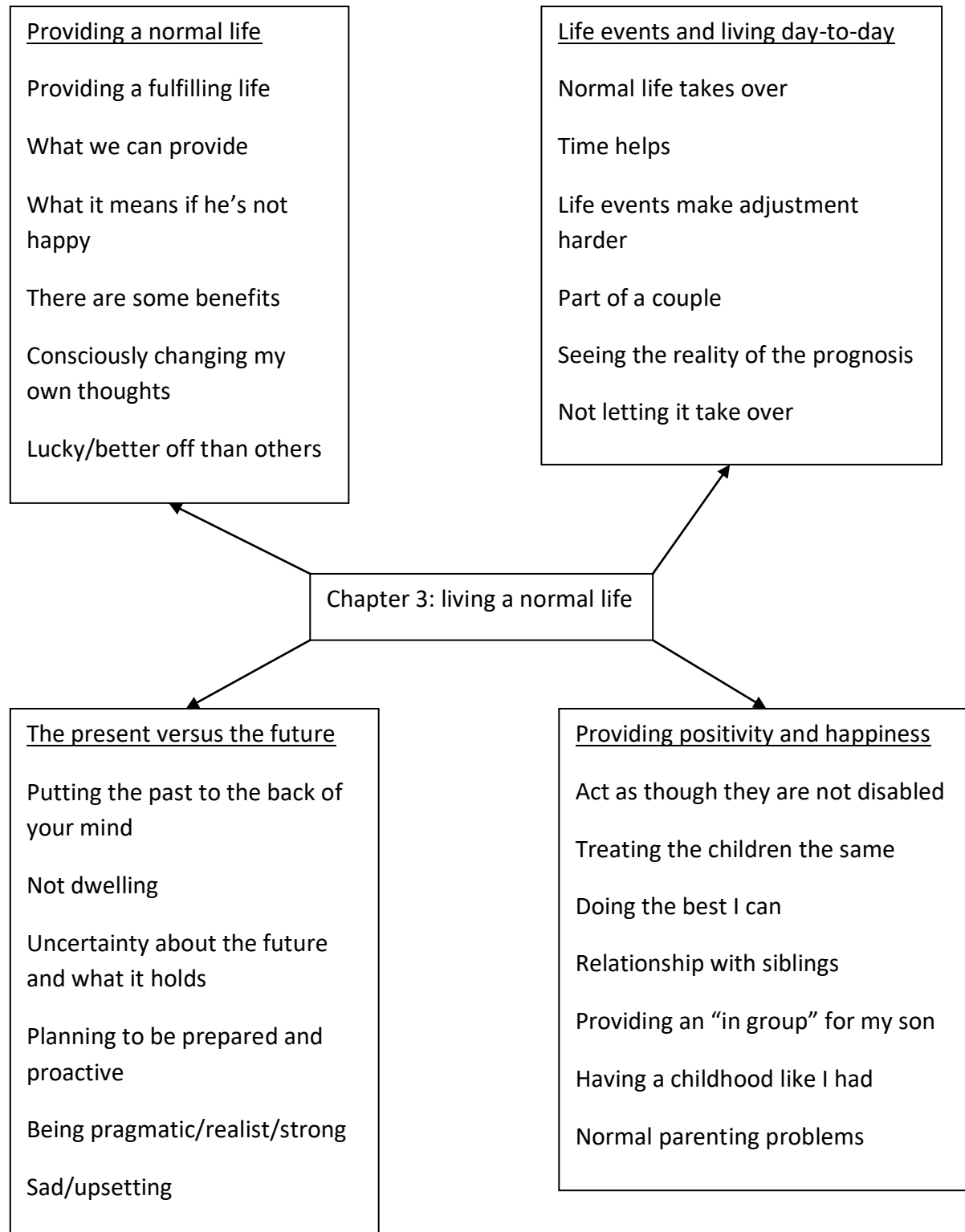
Appendix 2J:

A working diagram representing chapter 2 and its themes part way through the analysis



Appendix 2K:

A working diagram representing chapter 3 and its themes part way through the analysis



Appendix 2L:

Participant summary stories

The individual narratives discussed below provide an insight into the parents' personal journeys and highlight the events and salient roles they embody that might influence the meaning that they make of their experiences of having a child with DMD.

Sarah: I'm a Mum not a person

Sarah was the only parent interviewed who had two sons with DMD. This required her to give up her job to become their full-time carer. She described how being a Mum has replaced being "a person in her own right". Whilst she took on the responsibilities gladly, she missed the variety of experiences that came with meeting new people and doing new things every day. She described the loneliness that comes from sitting at home with 'the four walls' and not having interesting topics of conversation when someone does ask about her day. She would like to go back to college, as she viewed herself as unemployable, however this would put financial strain on the family, which she felt would ultimately take away resources from her sons, at a time when they will need increased levels of support. She also expressed concerns about the future, particularly "what will I do without the boys?" This pointed to ideas about who will she be once her role as a Mum has gone.

Lucy: The one to blame and burden carrier

Lucy described the impact that the diagnosis had on her as a parent. When she learnt about the genetic nature of the condition and the impact that it would have on her son she described experiencing guilt and blamed herself. She wondered if she could have done anything differently. This led Lucy to feel sad and distressed, which she did not want her family to see, particularly her son. Consequently she became the carrier of her own "burden", which she described as the grief and frustration associated with believing her son's illness "was still her fault" and the realisation that she had "lost a son without DMD". Lucy described attending counselling which helped her to feel better about herself and allowed her to love her son again. The guilt had started to cause a distance between her and her son, which was resolved when she was able to see that it was not her fault. This meant that both the way she viewed parenting issues and how she dealt with these issues changed. For example

she was able to see problems with bedtime routines as a part of normal childhood and consequently became calmer.

Dawn: The pragmatist and realist

Dawn described how being a researcher had impacted on all aspects of her experience, starting from diagnosing her son's condition. Her role provided her with the skills to seek appropriate information, interpret it and the confidence to follow her beliefs; to the point where she felt able to challenge professionals who may be seen as the 'experts' by others. Dawn described herself as a "pragmatist" and a "realist", which were parts of her identity that were associated with her role as a researcher. Later in her story these characteristics were helpful in her roles as a planner – which helped her to face the changes that her son was going to face and view them in a matter of fact way; for example, buying a wheelchair before he needed it so that he could get used to it. She mentioned how knowing these plans had been put in place and having a "realistic" attitude had also helped her to adjust emotionally.

April: Finding her old self to cope

April described the "emotional rollercoaster" that she had been on since they first suspected "something was not right". Her journey started with being without her partner on the day of the diagnosis. Despite feeling as though her "heart had been ripped out" she was left with the responsibility of processing the diagnosis on her own and feeding it back to her husband later that day. Throughout her story April discussed how she worried about the others in her family; how her husband and her parents would react and how she could explain the prognosis to her son. These anxieties added to her initial sense of vulnerability, self-blame and grief, which required her to take some time off work immediately after the diagnosis. At this point she did not feel like her usual capable self. Over time she became aware that she felt gradually better, having more good days than bad. She was able to put her worries and sadness to the back of her mind and get on with her life. She attributed this to her noticing that her son's health was not declining as she had expected and consequently her family were able to lead a relatively "normal" life. Although she expressed that she does still experience distress when she is reminded of what the future holds.

She commented that deciding to run for the Muscular Dystrophy Campaign was a "turning point", which allowed her to take control of an aspect of her life related to

DMD. Through running she recognised that she had become stronger, physically and psychologically, much like she had previously been before the diagnosis.

Raphael: The Optimist

Raphael was the only parent who commented that everything was new and challenging as they were new parents. He described his role as being the best father he could be, which meant using his son's happiness as the 'barometer' by which to measure his success. He endeavoured to make sure that his son has a fulfilling and relatively "normal" life, which required them to plan a little but generally they "live day by day". He described himself as an "optimist", which allows him to turn events into "positives" and stops him from "dwelling" on the negatives, which he acknowledged could easily happen. He also described himself as being quite "realistic", which he partly attributes to working in a healthcare profession and allowed him to find information and understand the prognosis and treatment. This helped to set his expectations, whilst providing hope that there could be a development in treatment.

Bob: The "Type A personality"

Early on Bob mentioned that both he and his wife were doctors. Two years prior to his son's diagnosis Bob started working with teenage boys with DMD, as a specialist in his field. This meant that Bob had a detailed knowledge of the disease and its prognosis, which he felt was beneficial for him as he did not have to research the disease.

Bob mentioned that he strongly identified with the stereotype of his profession, which he referred to as being a "type A personality" i.e. those who could be described as ambitious, rigidly organised, high achieving, outgoing and focused.⁶³ He described taking great pride in his work and being at the "front edge" of his field. This required him to work long hours, but immersed him in work that he enjoyed. Up until now he had been able to keep his work and personal life separate. His son had been sufficiently mobile that family life had stayed fairly "normal" and they had worked hard to ensure that his son's disability did not restrict their activities as a family. However he felt that most of his current worries related to the increased need for him to forego his work life and take on more caring duties, which would start to blur the boundaries and alter his job role. He mentioned that on reflection the division between work and home might have been a coping mechanism to help him deal with his son's prognosis and which led him to describe his need to work as "selfish".

Danielle: The adapter

Danielle described how a series of life events had shaped her past and present thinking about life and herself. Prior to having her son, Danielle had been through some experiences that she described as “troublesome” and “tough to deal with”. These included leaving an abusive relationship after having her daughter at the age of 17, and the death of her husband’s father. Danielle attributed these events to the experiences of “depression” she has occasionally felt. Her attitude towards these events now is “if you deal with things that are a bit rubbish, it’s just something else and you just get on with it.” She felt that she used her sarcastic sense of humour to make life feel less serious, which helped her to “push through”. Other people have also been valuable in forcing her to do things or making her think about how she parents her son. For example when her son was diagnosed with DMD she started to “mollycoddle” him and having two other children to look after meant she saw him differently and has forced him to be more independent. She is now at a point where she has seen positive examples of boys with DMD that have given her hope for her son having a fulfilling future, which have led her to feel like she does not want DMD to take over their lives.

Appendix 2M:

Pediatrics manuscript guidelines for publication

Journal style

All aspects of the manuscript, including the formatting of tables, illustrations, and references and grammar, punctuation, usage, and scientific writing style, should be prepared according to the most current *AMA Manual of Style* (<http://www.amamanualofstyle.com>).¹

Author listing

All authors' names should be listed in their entirety, and should include institutional/professional affiliations and degrees held.

Authoring groups

If you choose to include an organization, committee, team, or any other group as part of your author list, you must include the names of the individuals as part of the Acknowledgments section of your manuscript. This section should appear after the main text prior to your References section. The terms "for" or "on behalf of" must also be used when referencing the authoring group in the by-line.

Titles

Pediatrics generally follows the guidelines of the *AMA Manual of Style* for titles. Titles should be concise and informative, containing the key topics of the work. Declarative sentences are discouraged as they tend to overemphasize a conclusion, as are questions, which are more appropriate for editorials and commentaries. Subtitles, if used, should expand on the title; however, the title should be able to stand on its own. It is appropriate to include the study design ("Randomized Controlled Trial"; "Prospective Cohort Study", etc.) in subtitles. The location of a study should be included only when the results are unique to that location and not generalizable. Abbreviations and acronyms should be avoided. The full title will appear on the article, the inside table of contents, and in MEDLINE. Full titles are limited to 97 characters, including spaces. Short titles must be provided as well and are limited to 55 characters, including spaces. Short titles may appear on the cover of the journal as space permits in any given issue.

Abbreviations

List and define abbreviations on the Title Page. Unusual abbreviations should be avoided. All terms to be abbreviated in the text should also be spelled out at first mention, followed by the abbreviation in parentheses. The abbreviation may appear in the text thereafter.

Abbreviations may be used in the abstract if they occur 3 or more times in the abstract. Abbreviations should be avoided in tables and figures; if used they should be redefined in footnotes.

Units of Measure

Like many US-based journals, *Pediatrics* uses a combination of Système International (SI)^{2,3} and conventional units. Please see the *AMA Manual of Style* for details.

Proprietary Products

Authors should use nonproprietary names of drugs or devices unless mention of a trade name is pertinent to the discussion. If a proprietary product is cited, the name and location of the manufacturer must also be included.

References

Authors are responsible for the accuracy of references. Citations should be numbered in the order in which they appear in the text. Reference style should follow that of the *AMA Manual of Style*, current edition. Abbreviated journal names should reflect the style of Index Medicus. Visit: <http://www.nlm.nih.gov/tsd/serials/lji.html>

References

4. Iverson C, Christiansen S, Flanagan A, et al. *AMA Manual of Style*. 10th ed. New York, NY: Oxford University Press; 2007.
5. Lundberg GD. SI unit implementation: the next step. *JAMA*. 1988;260:73-76.
6. Système International conversion factors for frequently used laboratory components. *JAMA*. 1991;266:45-47.

Formatting requirements

All submissions must adhere to the following format:

- Times New Roman font, size 12, black
- Title Page, Contributors' Statement Page, Abstract, Acknowledgments, and References should be **single-spaced**
- Only the Main Body Text should be **double-spaced**
- Main Submission Document as Microsoft Word or RTF file (no PDFs)
- Do **not** include page headers, footers, or line numbers in new submissions.
- Do **not** include footnotes within the manuscript body. Footnotes are allowed only in tables/figures.

Refer to the “Article Types” section for specific guidelines on preparing a manuscript in each category. Note in particular the requirements regarding abstracts for different categories of article.

Title page

The “title page” should appear first in your manuscript document, and depending on the individual needs of a paper may encompass more than one page.

Title pages for all submissions **must** include the following items (as shown in the [sample Title Page](#)):

12. **Title** (97 characters [including spaces] or fewer)
13. **Author listing.** Full names for all authors, including degrees, and institutional/professional affiliations.
14. **Corresponding Author.** Contact information for the Corresponding Author (including: name, address, telephone, and e-mail).
15. **Short title** (55 characters [including spaces] or fewer). Please note: the short title may be used on the cover of the print edition.
16. **Financial Disclosure Statement** for all authors. Disclose any financial relationships that could be broadly relevant to the work. If none, say “Financial Disclosure: The authors have no financial relationships relevant to this article to disclose.”
17. **Funding source.** Research or project support, including internal funding, should be listed here; if the project was done with no specific support, please note that here. Technical and other assistance should be identified in Acknowledgments. If your funding body has open access requirements, please contact the Editorial Office prior to submission. Pediatrics has a 12 month embargo on articles (followed by a 4 year open access period) and does not allow articles to be opened for Creative Commons or similar licenses.
18. **Conflict of Interest Statement** for all authors. If none, say “Potential Conflicts of Interest: The authors have no conflicts of interest relevant to this article to disclose.”
19. **If applicable, Clinical Trial registry name and registration number.** We adhere to ICMJE guidelines, which require that all trials must be registered with ClinicalTrials.gov or any other WHO Primary registry.
20. **Abbreviations.** List and define abbreviations used in the text. If none, say "Abbreviations: none".
21. **Table of Contents Summary.** This is required for all articles with abstracts. This brief summary is limited to 25 words. For accepted manuscripts, this will appear under the author names in the table of contents to give the reader a brief insight into what the article is about. It should entice the reader to read the full article. For example: *"Through linkage of state Medicaid and Child Protective Services*

databases, this study captures similarities and differences in health care expenditures based on a history of child maltreatment."

22. For Regular Article submissions, include the “**What’s Known on This Subject; What This Study Adds**” (see below under article type for description). This is not needed for any other article type.

If a title page does not include all of the above items, the submission may be returned to the authors for completion.

Contributors' statement page

All submissions must contain a Contributors’ Statement Page, directly following the Title Page. Manuscripts lacking this page will be returned to the authors for correction.

All persons designated as authors should qualify for authorship ([see "Publication Ethics" above](#)), and all those who qualify should be listed. Each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content. The Contributors' Statement Page should list the authors and specify the contribution(s) made by each individual. If multiple individuals have identical contributions they may be listed together; do not list an author more than once.

You must follow the required format shown in this example when creating your Contributors’ Statement Page or your manuscript will be returned for correction. Each author should only appear once. Use names, not initials. If multiple authors have identical contributions, you can list them on the same line; otherwise, list each author separately.

Contributors' Statement:

Dr Smith conceptualized and designed the study, drafted the initial manuscript, and approved the final manuscript as submitted.

Drs Jones, Lee, and Weber carried out the initial analyses, reviewed and revised the manuscript, and approved the final manuscript as submitted.

Ms Green designed the data collection instruments, and coordinated and supervised data collection at two of the four sites, critically reviewed the manuscript, and approved the final manuscript as submitted.

Note: Contributors who do not meet the criteria for authorship (such as persons who helped recruit patients for the study, or professional editors) should be listed in an Acknowledgments section placed after the manuscript’s conclusion and before the References section. Because readers may infer their endorsement of the data and conclusions, these persons must give

written permission to be acknowledged. These permissions do not need to be submitted with the manuscript unless requested by the editors.

Figures, tables, and supplementary material

Figures

Authors should number figures in the order in which they appear in the text. Figures include graphs, charts, photographs, and illustrations. Each figure must include a legend (placed on the figure itself or as a list appearing after the References) that does not exceed 50 words.

Abbreviations previously expanded in the text are acceptable. If a figure is reproduced from another source, authors are required to obtain permission from the copyright holder, and proof of permission must be uploaded at the time of submission.

Figure arrays should be clearly labeled, preassembled, and submitted to scale. Figure parts of an array (A, B, C, etc.) should be clearly marked in capital letters in the upper left-hand corner of each figure part.

Technical requirements for figures: The following file types are acceptable: TIFF, PDF, EPS, and PNG. Color files must be in CMYK (cyan, magenta, yellow, black) mode.

Style for figures: Readers should be able to understand figures without referring to the text.

Avoid pie charts, 3-dimensional graphs, and excess ink in general. Make sure that the axes on graphs are labeled, including units of measurement, and that the font is large enough to read.

Generally delete legends or other material from the graph if it makes the picture smaller.

Color graphs should be interpretable if photocopied in black and white.

Pediatrics cannot accept Excel or PowerPoint files for any part of your submission.

Tables

Tables should be numbered in the order in which they are cited in the text and include appropriate headers. Tables should not reiterate information presented in the Results section, but rather should provide clear and concise data that further illustrate the main point. Tabular data should directly relate to the hypothesis. Table formatting should follow the current edition of the *AMA Manual of Style*.

Style for tables: Tables should be self-explanatory. Avoid abbreviations; define any abbreviations in footnotes to the table. Avoid excess digits and excess ink in general. Where possible, rows should be in a meaningful order (e.g., descending order of frequency). Provide units of measurement for all numbers. In general, only one type of data should be in each column of the table.

Supplemental information

Authors may wish to include additional information as part of their article for inclusion in the online edition of *Pediatrics*. References to any online supplemental information must appear in the main article. Such supplemental information can include but are not limited to additional tables, figures, videos, audio files, slide shows, data sets (including qualitative data), and online appendices. If your study is based on a survey, consider submitting your survey instrument or the key questions as a data supplement. Authors are responsible for clearly labeling supplemental information and are accountable for its accuracy. Supplemental information will be peer reviewed, but not professionally copyedited.

Regular Article

Abstract length: 250 words or fewer (structured, as noted below)

Article length: 3,000 words or fewer

Regular Articles are original research contributions that aim to inform clinical practice or the understanding of a disease process. Regular Articles include but are not limited to clinical trials, interventional studies, cohort studies, case-control studies, epidemiologic assessments, and surveys. Components of a Regular Article include:

What's known on this subject

What this study adds

These two brief summaries are each limited to 40 words. Please use precise and accurate language in paragraph form (i.e., not bullet points). For manuscripts accepted as Regular Articles, these summaries will become a highly visible part of your published paper, with prominence on the first page. Moreover, these summaries may be highlighted and presented in other areas of the journal. It is therefore paramount that you use language of the same caliber as the rest of your paper.

Structured abstract (four paragraphs with headings in boldface type; single-spaced)

The abstract should consist of: Background (or Objectives, or Background and Objectives), Methods, Results, and Conclusions. The Objective should clearly state the hypothesis; Methods, inclusion criteria and study design; Results, the outcome of the study; and Conclusions, the outcome in relation to the hypothesis and possible directions of future study.

Body of article

For the body of your article, follow this general outline:

Introduction

A 1- to 2-paragraph introduction outlining the wider context that generated the study and the hypothesis.

Patients and methods

This section should detail inclusion criteria and study design to ensure reproducibility of the research. All studies that involve human subjects must be approved or deemed exempt by an official institutional review board; this should be noted here.

Results

This section should give specific answers to the aims or questions stated in the introduction. The order of presentation of results should parallel the order of the methods section.

Discussion

The section should highlight antecedent literature on the topic and how the current study changes the understanding of a disease process or clinical situation, and should include a section on the limitations of the present study.

Conclusion

A brief concluding paragraph presenting the implications of the study results and possible new research directions on the subject.

General submission instructions (including cover letter, title page requirements, contributors' statement page, journal style guidance, and conflict of interest statements) apply to Regular Articles.

Appendix 2N:

Pediatrics title page

Parents' Stories of Adjustment to their Child's Chronic Illness: a Narrative Approach to Duchenne Muscular Dystrophy

Antoinette Deavin (BSc, MRes), Pete Greasley (PhD), Clare Dixon (BSc, DClInPsy, PhD)

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Short Title: Parental Stories of Adjustment to DMD

Funding Source: Lancaster University

Financial Disclosure: The authors have no financial relationships relevant to this article to disclose.

Conflict of Interest: The authors have no conflicts of interest relevant to this article to disclose.

Abbreviations: Duchenne Muscular Dystrophy (DMD), Paediatric Chronic Illness (PCI)

This qualitative study analyses parents' personal narratives to explore their experiences of adjustment to Duchenne Muscular Dystrophy.

Parents of children with DMD often experience clinical levels of distress. It has been recommended that qualitative and disease specific research is undertaken to understand their distress and adjustment, which would help raise awareness and identify avenues for support.

This study provides an understanding of the societal and personal influences on parental adjustment to DMD. Parents found themselves stuck in dichotomies of understanding: e.g. acute versus chronic illness models, parent versus carer roles and found ways to cope accordingly.

Appendix 20:

Pediatrics contributors' statement page

Contributors' Statement:

Ms Antoinette Deavin conceptualised and designed the study, carried out the research and drafted the initial and final manuscript as submitted.

Drs Greasley and Dixon reviewed the initial concept and critically reviewed the manuscript, and approved the final manuscript as submitted.

Section Three: Critical Appraisal

“Truth Seekers or Truth Makers” (Noblit): Examining Illness and Health Care Experiences in Health Research

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Word Count (excluding tables, references and appendices): 3,929

Word Count (including tables, references and appendices): 938

“Truth Seekers or Truth Makers” (Noblit): Examining Illness and Health Care Experiences in Health Research

Findings Overview

This thesis comprised a systematic review and an empirical paper exploring the subjective experiences of immediate family members when a child has a paediatric chronic illness (PCI). The meta-synthesis utilised a qualitative approach to take a broad view of children’s experiences of having a sibling with a PCI. The findings identified a lack of communication within the family regarding the illness, which led siblings to turn to other sources for information and support. More important are the novel findings regarding siblings’ construction of a more socially acceptable identity in order to meet their needs. This included the development of new roles, skills and responsibilities, often relating to supporting the family (e.g. housekeeping) and caring for their unwell sibling (e.g. managing their sibling’s symptoms). Consequently ‘caring’ became not only an activity but a part of their identity. This was usually at the expense of expressing their own needs directly, which may explain the reported increase of internalising problems compared to their peers.¹ This links directly to their use of short-term strategies, referred to as passive coping (e.g. avoidance), in order to maintain their new identity. These strategies have been found to be potentially unhelpful in the long-term and associated with poorer outcomes.²

The empirical paper analysed parental interpretations of privately held thoughts and broadly held social narratives relevant to their experiences of adjusting to having a child with Duchenne Muscular Dystrophy (DMD). The brief stories highlight the importance of person centred care to support individual needs; whilst the joint narratives can be applied to broader clinical practice and service delivery to aid parental adjustment. The joint narratives formed chapters of a story of adjustment. Initially parents discussed how they identified “something was not right” with their child, which related to their understanding of illness as something to

be resolved and healthcare as restitution. At times, their experiences of professionals conflicted with this narrative, causing them to feel like they had to “fight” for information. Parents based their understanding of the diagnosis on an acute rather than chronic model of health. Consequently they believed they would be unable to provide their son with a ‘normal’ and happy life, and disability represented a loss to be grieved. The restitution narrative was eventually contradicted as time passed. Their narratives also showed distressing changes to their identity. For example they described the movement from valued parent/worker to unvalued carer. Lastly, parents discussed “not dwelling” or “getting on with it”, which reflected coping through distraction and ignoring their son’s prognosis. They acknowledged this was possible due to the period of physical stability that occurs for boys with DMD after diagnosis. Parents “lived day to day” planning far enough ahead to be “proactive” and achieve their immediate parenting goals, but suppressed thoughts and feelings regarding the future. Consequently adjustment is described as a personal and fluid process, occurring at multiple time points, influenced by individuals’ experiences and narratives.

On reflection I feel strongly that these novel findings were as a result of using a qualitative and inductive approach. The remainder of this appraisal focuses on four areas of reflection: 1) the current epistemological position of healthcare 2) how qualitative approaches can be used to address health related research questions 3) the issues when considering epistemology and quality in research, and 4) future research.

Epistemology and Research in Healthcare

Epistemology is known as the “theory of knowledge”,³ and relates to how we come to “know”.⁴ This includes methods of enquiry and consequently relies on a philosophical understanding of what is to count as knowledge.³ This raises questions regarding the definitions of truth, knowledge and belief. Viewpoints vary from subjectivism and

constructionism and the notion that all knowledge is constructed within the thoughts and social actions of the knower(s) to objectivism and positivism where there are constant and knowable truths that can be discovered and generalised.^{5,6} The debate surrounding these questions has impacts for the field of research.^{5,7}

Quantitative methodology embodies positivistic and deductive reasoning. This is thought to be operationalised through the “universal scientific method”, which purports to minimise the effect of biases and reduce the likelihood of the result being due to chance. This leads to a rejection of the null hypothesis and an acceptance of the proposed hypothesis, that is deemed valid and generalisable.⁸⁻¹⁰ By accepting a hypothesis it can appear we are accepting a ‘truth’, at least temporarily, until it is refined by further research.⁹

This methodology has become the ‘gold standard’ within health research in the Western world.¹¹ This seems true for institutions like the NHS in the UK where guidance evaluating research places non-statistical data analysis at the bottom of the hierarchy of evidence, with meta-analyses of randomised controlled trials at the top.^{12,13} This quantitative skew has also been demonstrated in published health research in the UK¹⁴ and globally.¹⁵ For example, a study analysing the qualitative content of six widely read British health journals between 1990-2000 found that whilst qualitative research had increased by 2.1% in general it continued to represent a small percentage of the total, with 96.9% of articles being quantitative in 2000. Daniels et al¹⁵ and 170 co-signatories recently wrote an open letter to reopen the debate surrounding the paucity of published qualitative research in health settings,¹⁶ which consequently limits the kinds of research clinicians and the public can access.¹⁴

The prevalence of quantitative research stems from the beliefs of the individuals involved in the processes of research, teaching and dissemination. For example, it was found that a belief

in the universality of the scientific method^{8,10,17} and the ‘proof’ it produces,^{8,18} spanned teachers, researchers and clinicians. This is reflected in students’ potentially “naïve” preconceptions of science,^{8,19} despite pedagogical research recommending students be taught the breadth of epistemic understanding underlying scientific methodology and practice.^{4,20} This creates a cycle promoting quantitative approaches.

Equally, global political pressures may have played a role in promoting ‘science’, or rather the positivistic and frequently biological model of health science.²¹ The World Health Organisation identifies the role of neoliberalism and free markets in changes to health care since the 1980s.²¹ They report that whilst health reforms were meant to be “context sensitive” they eventually led to “...a limited menu of measures assumed to be valid everywhere.”^(pp.19) The felt necessity for prioritising quantitative research may also arise from the historical need for establishing disease aetiology, the focus of health as being the absence of disease^{22,23} and the emphasis on evidence-based medicine³; all of which aim to provide a standardised model, easily fitting a positivist framework. In the absence of understanding the principles of the nature of science (NOS) and epistemology, the ‘certainty and generalisability’ that this form of investigation claims to offer also suggests minimising risk with maximum impact. Within the context of reducing illness this is a powerful sentiment. Consequently the perceptions of professionals have maintained the ongoing prevalence of quantitative health research, either through ignorance of NOS, or the minimising of alternative research methods.¹¹

Despite the pedagogical and historical political movement towards positivism, increasingly health care endeavours to address broader social and psychological factors to promote health and prevent disease.^{21,23} This identifies a subtle change in emphasis from illness to health and health care, which are more complex issues. Consequently the ways in which these variables are researched need to represent the variation and subjectivity of the change of focus.²³ This

includes the role of systems in health care, unlike biological research which focuses on the mechanics of physiology.

Decisions Regarding the Method and how this is Reflected in the Findings

Utilising Qualitative Methods to Address Health Issues

It is argued that qualitative approaches provide relativism, subjectivity and uncertainty.^{24,25}

However it is recognised that even within a positivistic model of knowledge and research, scientists use creativity and do not adhere to the supposed standardised scientific method⁸ and undertake many subjective decisions.²⁶ In this way researchers cannot remove themselves from the scientific process. It is often argued that a strength of qualitative research is the acknowledgement of the role of the researcher in the research process to provide transparency.

The prevalence of positivism means qualitative research can feel irrelevant and inaccessible to health care professionals. Qualitative research is often seen as ancillary²⁴ or as too specialist to be practical,¹⁶ which was important to consider when designing and writing up the thesis. I needed the final reports to be useful for its audience, which would ultimately be other psychologists and health care professionals who might adhere to a positivistic perspective. This issue meant that sometimes there was a struggle between the methodology, methods and write-up, which needed careful consideration in order to maintain the quality of the project.³ In utilising qualitative approaches I was able to own these reflections and make them transparent for the reader to allow them to come to their own conclusions. Some of my personal considerations are outlined further below.

The Researcher's Position

My research narrative has developed over time, ranging from a positivist scientist in school undertaking my GCSEs to social constructionist whilst studying for my Masters in Research Methods. My current thinking reflects a balance of these stances dependent on the purpose and context of the research. Generally this places me as a critical realist who recognises that rather than having available ‘truths’ there are ‘tendencies’ dependent on context.^{27,28} Houston exemplifies this as “...people’s actions will be influenced by innate psychological mechanisms as well as wider social mechanisms.”^{27(pp. 850)} However in the world of healthcare where the dominant structure is still positivism, I find myself leaning towards a more relativist position.

With patients not often being consulted regarding the policies that will affect them²⁹ I feel the current societal narrative continues to promote a ‘done to’ model of care, despite professional guidance to the contrary and the promotion of person-centred care. It has been interesting then to consider what narratives and care parents receive regarding the illness of their child and how parents can manage these conditions within the systems provided. As a parent myself I am aware of my desire to assess my children’s wellbeing in accordance with an acute model of health, despite being aware of alternative understandings. I feel this demonstrates the strength of these narratives and the power professionals hold in being the storytellers. I believe to counteract the power differential between patients and professionals we must gain the testimony of those who have experienced illness, consequent care and the impacts of both to elucidate their stories of success and failure. Privileging those experiences produces grassroots accounts that should be considered when delivering healthcare; hence my decision to use a qualitative approach. I thought a narrative approach would address the research aims, meet my own criteria and allow for understanding changes over time, which had been missing from previous research.³⁰

Narrative analysis stems from the concept that the meaning individuals make of their lives is storied.³¹ They attempt to provide structure where there had been chaos³² and therefore analysis of an individual's account enables us to sympathise with their understanding of events and experiences. This process necessarily involves the story teller to reflect on their position in the story and therefore represent their "sense of selfhood" or identity in its telling.³³ The changing identity of participants was found to be an important factor in both the systematic review and the empirical paper, which enabled the identification of novel clinical applications that previous deductive research had not identified. The subjectivity and complexity of identity construction means that a quantitative investigation would struggle to capture this issue, whilst qualitative approaches, and in particular narrative analysis, are ideal for this type of research question.

In telling their stories participants explored personal experiences e.g. daily events and interactions, and the social, political and cultural themes that have influenced their understanding.³⁴ The findings from the systematic review identified many social and cultural influences that affected the siblings' experiences e.g. narratives regarding families not discussing their ill child's condition. The empirical paper documents identities influenced by society e.g. being a mum not a person or being the one to blame. Recognising these contextual factors that may influence an individual's experience and their subsequent interpretation are key tenets of narrative analysis.³⁵

McAdams³¹ suggests individuals will have many stories regarding their lives but each integrates subjectively important 'scenes/chapters' based on their experience, which may also reflect others' experiences. Whilst these scenes/chapters are subjective, the chapters in the research paper represent unifying experiences that are present by virtue of having similar cultural backgrounds e.g. health as restitution. I feel this homogeneity in the findings represents both a strength and an area for further research. The strength lies in its utility for

health professionals, as it addresses the question of generalisability, however it requires the reader to be mindful of the participants' demographics as they all described themselves as white, all a similar age and most acknowledged their education or financial situation as supportive factors. Consequently their experiences may not consistently reflect that of others from differing backgrounds.

Quality and Epistemological Coherence

There are differing views as to how quality should be achieved dependent on the approach. Below I outline my considerations.

As per good practice I undertook a quality assessment of the papers to be synthesised; two important quality issues were identified. First, the number of qualitative studies in this area is small and the quality available is low, as explained below. Second, in identifying the low level of quality it became necessary to make this transparent to the reader for them to make their own conclusions and highlight the methodological issues in this area,³⁶ hence no articles were excluded. It is crucial to notice that generally the low quality was due to the research being undertaken with a quantitative understanding. For example, when the research methods were mixed the qualitative element was often ancillary and poorly operationalised therefore not meeting the qualitative guidelines. However, given that the subsequent data was inductively produced and evidenced it was still credible and of value when placed within the context of the synthesis.

I engaged with varying guidelines endeavouring to establish quality in qualitative research^{3,37-39} to consider the quality of my work. This provided a bottom-up understanding, from methods to epistemology, which complimented the top-down academic understanding that had taken place when starting to broadly read about qualitative research design for my empirical paper. Consequently, I identified the need for me to consider the balance of

epistemological and functional issues in my research. Some of the guidelines directly outlined their links to epistemological stances, whereas others assumed a stance and focused on providing functionality. The Critical Appraisal Skills Programme (CASP)³⁹ is an exemplar of the latter. I decided to utilise the CASP to assess the quality of the papers reviewed in the synthesis after discussion with the research team and feedback from course teaching. In addition, one of my personal aims for the research was to introduce psychological ideas to a broader audience. The CASP is a widely known appraisal tool and therefore would be easily accessible to the potential audience. I also chose it for its functionality. As a novice researcher it provides a simple set of guidelines that are easy to follow, which would also be beneficial for the reader, should they be unacquainted with qualitative research methodology. The undertaking of the synthesis and empirical paper posed different quality issues.

For the synthesis I generally referred to standardised assessment tools as guidance for quality of the synthesis (e.g. CASP, PRISMA). These tools provided sufficient guidance to ensure the quality of the synthesis met the expectations of the potential audience but did not consider the coherence between epistemology, methodology and methods. However I consulted a paper by Creswell and Miller⁴⁰ which describes the process of coherence between the research's epistemological approach, its methodology and its methods it outlines transparent methods that demonstrate a study's validity dependent on its philosophy of knowledge. For the synthesis I took a critical realist position, which allowed me to present findings as truths whilst being immersed in context.^{27,28,41} As a post positivist stance they suggested triangulation, liaising with participants to confirm the credibility of the results and an audit trail to exhibit grounding in the data. Triangulation took the form of liaison with the research team, which helped with the development of themes to expand my thinking and to consider the findings from research and clinical perspectives. As a synthesis relying on the research of others I was unable to perform member checks for credibility however I only included papers

with quotations to evidence their participation and perspective, and I endeavoured to incorporate contrasting participant voices to explore the depth of the themes. The result of which was then thoroughly compared with the relevant bodies of literature. An audit trail has been demonstrated in appendices, which document part of the constant iterative process that developed the themes.

Decisions Relating to Epistemological Coherence

Epistemological coherence relates to the idea that the researcher's understanding of truth and knowledge compliments the methodology and methods that are utilised and represented in the research write-up. Coherence has been noted as way of determining the quality of a study.³ As mentioned previously the question of relativism meant I needed to reflect on the thesis' utility as the research would hopefully be read by health professionals working in a variety of contexts with varying research perspectives. Whilst nursing journals appear to represent the range of research methods, with The Journal of Advanced Nursing publishing nine articles including qualitative methods out of 14 in October 2016, publication of qualitative articles in major medical journals remains low,¹⁶ meaning that medics are generally exposed to and most likely adhere to a positivist model of knowledge. Consequently it was necessary for me balance coherence and audience accessibility and utility. For the systematic review the tension between advocating subjective and unheard voices whilst producing helpful accounts was minimised by the fact the primary research had already been undertaken and there was enough to perform a meta-synthesis. The remit of the synthesis was to bring together those voices to collaboratively state what they felt and identify any new information that would promote further understanding of sibling's experiences of chronic illness akin to Noblit's line of argument synthesis.⁴²

Consequently the impact of epistemological coherence on the design of the review was relatively small as my personal aims were in line with the research aims. This has clearly been shown in the findings where a fuller account of siblings' experiences is provided from a varied range of primary sources. For example, the identification of the 'changing relationship to self' whereby siblings' identities changed over time to include prosocial characteristics that had previously met the needs of the family, whilst minimising their own, was shown consistently across studies but not previously identified. The presence of this 'one truth' is given power by the unity of the voices that came from a foundation of inductive based research. Without a synthesised qualitative approach the psychological process of sibling behavioural adaptation that can lead to a change in identity in order to meet their needs would not have been identified.

Narrative analysis is a less standardised form of research, which allows for greater flexibility but can make it difficult to ensure the quality of the research, especially for a novice.³⁵

Narrative methodology openly acknowledges the role of epistemology, and often aligns with a 'realist' position,³³ however this is not a prescriptive relationship. It is the role of the research team to determine the epistemological stance the research will take and reflect on how this translates into its method. In contrast to the synthesis my personal and research aims for the empirical paper reflected a more critical paradigm. Utilising the Creswell and Miller⁴⁰ matrix would suggest research reflexivity, collaboration and peer debriefing as its methods for verifying validity (see Figure 1.).

[Insert Figure 1. here]

Aspects of these concepts are evidenced, however coherence was difficult to balance given the conflicting needs within the research i.e. my needs as a student, my personal and research aims and the needs of the audience. Reflexivity was a constant process and is demonstrated

by my use of a research journal, liaison with the research team and a thorough engagement with surrounding literature. Liaison with the research team was particularly important when considering when to stop recruiting. Narrative analysis does not determine a sample size or a process by which to make this decision, unlike other qualitative analyses like some forms of grounded theory which might require thematic saturation. Consequently, an initial discussion balancing the aforementioned needs was undertaken. These aspects all related to providing a sample that was large enough to provide commonalities, whilst not being unwieldy. Repeat evaluation conversations throughout the analysis led to a final total of seven participants. Reflexivity and credibility were difficult to demonstrate at times due to the limited word counts afforded by the assignments. However the critical appraisal has provided space to evidence some reflection. In regards to collaboration, a large portion of time was dedicated to developing the interview schedule to ensure the interview process was collaborative and led by the participant. The introduction was designed to be transparent regarding consent and the following analysis of the interviews. Questions and prompts were open and intended to put participants at ease, whilst adhering to features that would be relevant for the narrative analysis. As the schedule was flexible the first and second interview transcripts were reviewed by a member of the research team to assess the construction and relevance of the questions. Questions were added and adjusted as relevant. The research team formed the basis for peer debriefing, whom endeavoured to provide a sounding board and challenge the research process and findings.^{40,43,44} This was fulfilled through the research supervisors who specialise in paediatric clinical psychology and research methods, enabling a thorough interrogation of the research content and process.

Future Research

The suggestions for future research in both the systematic review and empirical papers relate to the complexity of human experience and its influences. The synthesis pointed towards how culture influences family dynamics, including communication, narratives of illness and gender roles and the empirical paper suggested investigation into how multiple life events and varying social and cultural narratives impact upon adjustment. These are not easily measurable concepts and therefore qualitative approaches would be most useful, however that is not to say positivist methods should be excluded and would depend on the stance of the research team and the development of the research question.

The issues raised by this thesis often point to interventions by health care professionals. Should professionals implement these suggestions it would be worthwhile investigating the influence of these changes on family's experiences of services, and in coping with and managing their distress, in order to provide continued development and quality of care. Ultimately this would address family's concerns regarding having to fight against services, as they would feel involved. Ideally family members would be involved in the design of the research, which was a weakness of this thesis. Whilst the research addressed the needs of families and their perspectives the design process did not include service user liaison. This premise was debated within the research team and it was felt that due to the scale of the project it was not an urgent requirement of the research. Equally, I felt that given the epistemological stance taken it was not necessary, although it would have been preferable to ensure the involvement and advocacy of families in their care and in the field of research.

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Tables and Figures

Figure 1.

A copy of the Creswell and Miller method matrix (2000)

Paradigm/Lens	Postpositivist	Constructivist	Critical
Lens of researcher	Triangulation	Disconfirming evidence	Research reflexivity
Lens of participants	Member checking	Prolonged engagement	Collaboration
Lens of people external	The audit trail	Thick, rich description	Peer debriefing

Section Four: Ethics Section

Parents' Stories of Adjustment to their Child's Chronic Illness: a Narrative Approach to Duchenne Muscular Dystrophy

Antoinette Deavin

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Word Count (excluding tables, references and appendices): 4,197

Word Count (including tables, references and appendices): 4,486

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please enter a short title for this project (maximum 70 characters)

Parents' stories of adjustment to their child's chronic illness

1. Is your project research?

Yes No

2. Select one category from the list below:

- Clinical trial of an investigational medicinal product
- Clinical investigation or other study of a medical device
- Combined trial of an investigational medicinal product and an investigational medical device
- Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- Basic science study involving procedures with human participants
- Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- Study involving qualitative methods only
- Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- Study limited to working with data (specific project only)
- Research tissue bank
- Research database

If your work does not fit any of these categories, select the option below:

Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? Yes No
- b) Will you be taking new human tissue samples (or other human biological samples)? Yes No
- c) Will you be using existing human tissue samples (or other human biological samples)? Yes No

3. In which countries of the UK will the research sites be located? *(Tick all that apply)*

- England
- Scotland
- Wales
- Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

NHS REC Form

Reference:
13/SC/0649

IRAS Version 3.5

- England
 Scotland
 Wales
 Northern Ireland
 This study does not involve the NHS

4. Which review bodies are you applying to?

- NHS/HSC Research and Development offices
 Social Care Research Ethics Committee
 Research Ethics Committee
 National Information Governance Board for Health and Social Care (NIGB)
 National Offender Management Service (NOMS) (Prisons & Probation)

For NHS/HSC R&D offices, the CI must create Site-Specific Information Forms for each site, in addition to the study-wide forms, and transfer them to the PIs or local collaborators.

5. Will any research sites in this study be NHS organisations?

- Yes No

5a. Are all the research costs and infrastructure costs for this study provided by an NIHR Biomedical Research Centre, NIHR Biomedical Research Unit, NIHR Collaboration for Leadership in Health Research and Care (CLAHRC) or NIHR Research Centre for Patient Safety & Service Quality in all study sites?

- Yes No

If yes, NHS permission for your study will be processed through the NIHR Coordinated System for gaining NHS Permission (NIHR CSP).

5b. Do you wish to make an application for the study to be considered for NIHR Clinical Research Network (CRN) support and inclusion in the NIHR Clinical Research Network (CRN) Portfolio? Please see information button for further details.

- Yes No

If yes, NHS permission for your study will be processed through the NIHR Coordinated System for gaining NHS Permission (NIHR CSP) and you must complete a NIHR Clinical Research Network (CRN) Portfolio Application Form immediately after completing this project filter and before completing and submitting other applications.

6. Do you plan to include any participants who are children?

- Yes No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

- Yes No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the NIGB Ethics and Confidentiality Committee to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

NHS REC Form

Reference:
13/SC/0649

IRAS Version 3.5

 Yes No**9. Is the study or any part of it being undertaken as an educational project?** Yes No

Please describe briefly the involvement of the student(s):
The student will be the chief investigator of the study.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate? Yes No**10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?** Yes No**11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?** Yes No

Integrated Research Application System
Application Form for Research involving qualitative methods only


Application to NHS/HSC Research Ethics Committee

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting [Help](#).

Please define any terms or acronyms that might not be familiar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
Parents' stories of adjustment to their child's chronic illness

Please complete these details after you have booked the REC application for review.

REC Name:
Hampshire-B REC

REC Reference Number:
13/SC/0649

Submission date:
02/12/2013

PART A: Core study information
1. ADMINISTRATIVE DETAILS
A1. Full title of the research:

Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy

A2-1. Educational projects

Name and contact details of student(s):

Student 1

	Title Forename/Initials Surname
	Ms Antoinette Deavin
Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire
Post Code	LA1 4YG
E-mail	a.deavin@lancaster.ac.uk
Telephone	01524592754
Fax	

Give details of the educational course or degree for which this research is being undertaken:

NHS REC Form

Reference:
13/SC/0649

IRAS Version 3.5

Name and level of course/ degree:
Doctorate in Clinical Psychology (DClinPsy)

Name of educational establishment:
Lancaster University

Name and contact details of academic supervisor(s):

Academic supervisor 1

	Title	Forename/Initials	Surname
	Dr	Pete	Greasley
Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire		
Post Code	LA1 4YG		
E-mail	p.greasley@lancaster.ac.uk		
Telephone	01524593535		
Fax			

Please state which academic supervisor(s) has responsibility for which student(s):

Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

Student(s)	Academic supervisor(s)
------------	------------------------

Student 1 Ms Antoinette Deavin

Dr Pete Greasley

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

A2-2. Who will act as Chief Investigator for this study?

- Student
 Academic supervisor
 Other

A3-1. Chief Investigator:

	Title	Forename/Initials	Surname
	Ms	Antoinette	Deavin
Post	Trainee Clinical Psychologist		
Qualifications	Psychology BSc, Psychology MRes		
Employer	Lancashire Care		
Work Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire		
Post Code	LA1 4YG		
Work E-mail	a.deavin@lancaster.ac.uk		
* Personal E-mail			

Work Telephone 01524592754
 * Personal Telephone/Mobile
 Fax

** This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.*

A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?

This contact will receive copies of all correspondence from REC and R&D reviewers that is sent to the CI.

	Title Forename/Initials Surname
	Ms Debbie Knight
Address	Research Support Office, B Floor, University House, Lancaster University, Lancaster
Post Code	LA1 4YW
E-mail	ethics@lancaster.ac.uk
Telephone	01524592605
Fax	

A5-1. Research reference numbers. *Please give any relevant references for your study:*

Applicant's/organisation's own reference number, e.g. R & D (if available):

Sponsor's/protocol number: 1

Protocol Version: 1

Protocol Date:

Funder's reference number:

Project website:

Additional reference number(s):

Ref.Number	Description	Reference Number
------------	-------------	------------------

Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

Yes No

Please give brief details and reference numbers.

2. OVERVIEW OF THE RESEARCH

To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.

A6-1. Summary of the study. *Please provide a brief summary of the research (maximum 300 words) using language*

easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, this summary will be published on the website of the National Research Ethics Service following the ethical review.

Duchenne Muscular Dystrophy (DMD) is a genetic, chronic and life limiting disorder that affects approximately 1 in 3,600 boys in the UK. It results in muscle degeneration, abnormal bone development and an array of other physiological changes, the onset of which can be seen in early infancy. These physiological problems present in everyday life as low endurance, difficulties standing and walking, loss of movement, paralysis, and possibly intellectual impairment. The average life expectancy of children diagnosed with DMD is approximately their late 20s.

Understandably a diagnosis of DMD and the subsequent changes that occur will have an impact not only on the child but their family. Parents in particular are expected to manage their child's health, emotional and psychological needs in addition to their own, requiring a level of adjustment.

Current literature has begun to highlight generic factors relevant to parental adjustment to paediatric chronic illness; however it does not take into account the meaning of these experiences for parents and consequently what they find important for adjustment, which would account for the interactions between factors. Additionally, there is a lack of qualitative research relating to DMD generally and specifically parental adjustment.

Consequently narrative analysis will be used to hear parents' stories of adjustment, allowing them to express what has been important to them in their adjustment and how they understand that process.

Up to 12 parents/ long-term guardians will be recruited for face to face interviews that will last up to an hour and a half and will be located in their homes or local family based services.

Recruitment will take place in two stages, dependent on need:

1. Through an on online advert; posters in local services and an information mail out to those who have been referred to local paediatric services.
2. Through a brief presentation to support groups and contacting local children's hospices.

A6-2. Summary of main issues. *Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.*

Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, R&D office or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.

Ethical issues

Recruitment will primarily rely on participants opting in by contacting the researcher, meaning that the researcher will only receive the participant's contact details when the participant gives them to the researcher directly.

During the introduction to the study confidentiality, anonymity, data protection, consent and withdrawal will be discussed.

Confidentiality is documented in the Participant Information Sheet and will be checked on the Consent Form. Parents will be reassured that they can share as much information as they feel comfortable, however if risk to themselves or others has been disclosed then confidentiality will need to be broken. If this occurs the limits of confidentiality will be discussed again and where possible the next steps of the process will be collaborative, demonstrated by the researcher maintaining a friendly and supportive manner. The researcher will attempt to assess the level of risk and urgency present. Initially, an appropriate member of the research team will be consulted to determine the course of action, which might include sign-posting to another service or getting other services, like social services or the [REDACTED] contact cannot be made with a member of the research team and the risk to the participant is high then other external services may need to be contacted. This process is designed to identify, assess and manage risk and at no point will the researcher be able to offer therapy.

Participants are not expected to find the interview emotionally or psychologically difficult. It will be noted before the start of the interview that if participants become distressed, interviewees will be consulted as to how they would like to proceed i.e. continue, take a break or terminate the interview. The voice recorder will be switched off during breaks and interviewees will be notified when this happens.

Any hard copies of personal information that is collected at the interview (e.g. consent forms) will be stored in designated lockable containers at Lancaster University. Any electronic data, including audio recordings and transcripts will be stored on a password protected computer or laptop. The researcher's laptop will also have an encrypted drive that all files will be stored in. A copy of these will also be kept on the University's network drive which is on Lancaster University's virtual private network and is password protected.

Participants will nominate a pseudonym or 'alternative' name, that can be used on the transcripts and on any quotations in the write-up, to keep their information private. Once a transcript is anonymised the research and field supervisors will be able to view the transcripts to aid analysis.

Participants will have received information on the study prior to the interview but the interviewer will repeat the information documented on the Participant Information Sheet and Consent Form to gain informed written consent. Capacity to consent will need to be considered before the interview and will be generally assessed by the interviewer by going through the Participant Information Sheet and Consent Form.

The researcher will endeavour to keep all processes and decisions transparent to the participant and explain all aspects of the study until the interviewee is satisfied. Their transcript can also be removed from the analysis for up to two weeks after the interview, without giving a reason. This time is provided to allow the participant time to reflect on the interview, whilst providing a timeline that will support the researcher in continuing the analysis.

Practical issues

Recruitment of participants will require information packs to be sent out and other forms of promotion, like posters, will need to be produced. Costs for essential expenses include, but are not limited to: paper, printing, photocopying, postage and phone calls which will be covered by Lancaster University under their research resources for trainees' policy.

Interviews will be sited at participants' homes or in local family based services. This will help to ensure anonymity, as the participants could be attending another appointment. This will also minimise risk as there will be other individuals on site that the researcher or interviewee can contact in an emergency e.g. if a participant collapses. The researcher has considered the Lancashire Care Foundation Trust Lone Worker Procedure and has developed a lone working system in accordance with this documentation. The researcher will use the buddy system to keep track of the interviewer and their expected movements. If support should be required then the nominated buddy can be contacted. An email with relevant details will be sent prior to meeting the interviewee, so that the buddy can access all the information they need, should there be a problem. If the interviewer does not check in with the buddy at the given time then the buddy will initially attempt to contact the researcher and if there is no response or returned call within an hour and a half they will call the police. A set phrase will be agreed upon between the buddy and the interviewer to denote an inability to communicate risk, threat or harm due to fear.

Participants will be given the opportunity to receive notification of any dissemination of the study e.g. organise individual meetings.

A6-3. Proportionate review of REC application *The initial project filter has identified that your study may be suitable for proportionate review by a REC sub-committee. Please consult the current guidance notes from NRES and indicate whether you wish to apply through the proportionate review service or, taking into account your answer to A6-2, you consider there are ethical issues that require consideration at a full REC meeting.*

Yes - proportionate review No - review by full REC meeting

Further comments (optional):

Note: This question only applies to the REC application.

3. PURPOSE AND DESIGN OF THE RESEARCH

A7. Select the appropriate methodology description for this research. Please tick all that apply.

- Case series/ case note review
- Case control
- Cohort observation
- Controlled trial without randomisation
- Cross-sectional study
- Database analysis
- Epidemiology
- Feasibility/ pilot study
- Laboratory study
- Metanalysis

- Qualitative research
- Questionnaire, interview or observation study
- Randomised controlled trial
- Other (please specify)

A10. What is the principal research question/objective? *Please put this in language comprehensible to a lay person.*

What are the adjustment narratives of parents with a child who has Duchenne Muscular Dystrophy?

A11. What are the secondary research questions/objectives if applicable? *Please put this in language comprehensible to a lay person.*

- 1: To use parental narratives to understand parents' interpretations of their experience and consider the issues that they face.
- 2: The prevailing models of parental adjustment have been generic to chronic illness and were developed over 10 years ago (Wallender, 1998). This study would aim to consider parental narratives, their content and structure in comparison to current literature and ascertain the similarities and differences to this model, in order to add to the adjustment literature.
- 3: Narrative research is becoming more influential in developing clinical practice (Overcash, 2003). This study will use parental experience to consider how services could be improved whilst highlighting what is important to parents.
- 4: Duchenne Muscular Dystrophy is a severe and fatal disease, which compared to other chronic illnesses, like Leukaemia or Cystic Fibrosis, is relatively unknown. Therefore this study aims to help raise awareness/strengthen the voices of the families that this disease affects.

A12. What is the scientific justification for the research? *Please put this in language comprehensible to a lay person.*

Research has shown that parents often suffer from severe psychological distress related to their experience of having with a child with a chronic paediatric illness. This highlights the importance of investigating experiences of adjustment, both good and bad, as a way of ascertaining the subjective needs and coping strategies of parents. This would provide information that professionals might find helpful when developing good practice within services when working with parents with children who have chronic illness.

There are generic models of adjustment but these are at least ten years old and therefore do not account for the changes in treatment and society which may influence parental experience and their interpretation of these experiences. The models are also based on quantitative research which has highlighted isolated factors involved in the process of adjustment. Reviews have suggested that qualitative research needs to be undertaken to understand the broader picture and the role that the factors have in parental experience. This has occurred with many chronic illnesses e.g. paediatric cancer, however there is very little focussing on Duchenne Muscular Dystrophy (DMD). Unlike other high profile diseases, DMD is relatively unknown by the general public and is comparatively less well supported by services. This may have an impact on parental experiences and the meaning they have for parents.

A qualitative approach like narrative analysis allows parents to express their stories of adjustment and give them the freedom to discuss what has been important and meaningful to them. The stories may include issues raised by previous research but will provide context and meaning that has been lacking. It will also provide an opportunity for parents to discuss how they have coped. Identifying these narratives will add to the knowledge base of parental adjustment to paediatric chronic illness generally but also consider how this applies to DMD specifically. This research will therefore help to raise awareness for this disease, whilst utilising parents' expertise to consider their needs and how best to support them.

A13. Please summarise your design and methodology. *It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.*

The study's research question required a qualitative approach to be taken. A narrative analysis was deemed the most appropriate analysis to answer the research question, whilst being appropriate for the size and scope of the study. A narrative analysis will allow the detail and subjectivity of the interviewees' stories to be maintained whilst analysing the content and structure of the interview. This will help to analyse parents' understanding of their experiences, which will be useful for professionals.

Up to 12 long-term parents/guardians will be recruited and must fit the inclusion/exclusion criteria. Other factors, including proximity, will be considered when arranging interviews.

Participants will potentially become aware of the study in 1 of 5 ways in two stages: 1) Through posters, an advert on the Contact a Parent forum or as part of an information pack posted to parents who have been referred to local paediatric services. Specialist nurses within the paediatric services will also discuss the study with prospective parents in their daily meetings and give them an information pack.

2) If possible through a presentation to local support groups or leaving information packs at local children's hospices. Parents/guardians will need to opt in to the study by contacting the chief investigator directly on the details given on the Participant Information Sheet, on the poster or the advert. If they respond to a poster then the Participant Information Sheet will need to be posted to them before they participate.

Once an enquiry has been made by a parent they will be asked to notify the chief investigator within 1 week as to whether they would like take part. It is at this stage that the inclusion and exclusion criteria will be checked.

If they are happy to proceed then a convenient time and place will be arranged to meet.

The purpose of the meeting is for the chief investigator to conduct a face-to-face, semi-structured interview to discuss parents'/guardians' experiences and stories of adjustment.

At the beginning of the interview there will be general introductions and the interviewer will go through the participant information sheet again to make sure that the participant understands all aspects of the study. The interviewer will then go through the consent form. The participant will have their right to withdraw and the principle of confidentiality explained to them at this point. The specifics of confidentiality and the process of breaking confidentiality due to risk and danger will be discussed with the participant before they are asked to consent.

The participant will be able to ask questions at any point but they will be specifically given the opportunity to do this after the participant information sheet and consent form has been explained. They will only be asked to give consent when they are happy they understand and want to continue.

After the interview the carer will be asked if they have any questions and whether they want to be contacted to find out about the findings. If they say yes, they will be called within 4 months of their interview to arrange a way to give this information.

Timetable:

November 2013: apply for ethical approval.

November 2013- December: Once ethics has been approved then start recruitment.

November 2013– February 2014: conduct interviews, transcription and coding.

May 2014: Submission of final report

September 2014: Submit paper for publication

A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?

- Design of the research
- Management of the research
- Undertaking the research
- Analysis of results
- Dissemination of findings
- None of the above

Give details of involvement, or if none please justify the absence of involvement.

Participants will have the opportunity to review their re-storied accounts to promote rigor and trustworthiness.

4. RISKS AND ETHICAL ISSUES

RESEARCH PARTICIPANTS

A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

Individuals eligible to participate in this study must be parents or long-term guardians of children with a diagnosis of Duchenne Muscular Dystrophy. If the participant is a long-term guardian then they must have cared for the child prior to the displaying of symptoms and/or a diagnosis.

In order to focus on adjustment and not specifically diagnosis or grief approximately two years must have passed since diagnosis and they must be currently caring for the child. Parents must speak English as a first language.

A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

The child must not have co-morbidities that are unassociated with DMD, e.g. other chronic health illnesses. Expected co-morbidities will not be excluded, examples include: respiratory difficulties/night time ventilation; cardiac problems; scoliosis, ADHD and requirements for spinal surgery.

To keep the sample reasonably homogeneous parents with their own chronic needs that may distinctly affect their perspective cannot be included, for example, but not limited to: those who have had a heart transplant or have a learning disability.

Families where there are current safeguarding concerns, i.e. are under investigation will also not be included.

RESEARCH PROCEDURES, RISKS AND BENEFITS**A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. These include seeking consent, interviews, non-clinical observations and use of questionnaires.**

Please complete the columns for each intervention/procedure as follows:

1. Total number of interventions/procedures to be received by each participant as part of the research protocol.
2. If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
3. Average time taken per intervention/procedure (minutes, hours or days)
4. Details of who will conduct the intervention/procedure, and where it will take place.

Intervention or procedure	1	2	3	4
Recruitment	3	0	10	Participants will be recruited through posters, an information mail out to service users and an online advert on Contact a Parent's forum. The specialist nurses will also discuss the study with parents they meet during their clinics/meetings and give them an information pack so they can contact the chief investigator.
Giving information	1	0	5	The chief investigator will hand out and explain the Participant Information Sheet just before the interview.
Seeking consent	1	0	5	The chief investigator will explain the Consent Form and seek informed written consent from the participant just before the interview starts.
Interview	1	0	90	The chief investigator will conduct the interview.
Post interview questions	1	0	10	The chief investigator will receive post interview questions.
Feedback	1	0	30	The chief investigator will contact the interviewees to ask if they would like to receive a summary of the study.
Review of participant's account	1	0	30	The chief investigator will contact the interviewee to discuss their re-storied account.

A21. How long do you expect each participant to be in the study in total?

Each participant will be involved in an interview (up to 1.5 hours) and a review of their storied account (up to 1 hour) as part of the analysis. These two meetings could be a few months apart.

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

There are no expected risks for taking part in this study. Participants are not expected to find the interview emotionally or psychologically difficult. Participants will be notified that they do not have to share information that makes them uncomfortable or upset, however, it will be noted before the start of the interview that if participants become distressed, interviewees will be consulted as to how they would like to proceed i.e. continue, take a break or terminate

the interview. The voice recorder will be switched off during breaks and interviewees will be notified when this happens. If there is no perceived risk but the interviewee is distressed then they will be recommended to seek advice from their GP or other services that may already be involved for further support.

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

Yes No

If Yes, please give details of procedures in place to deal with these issues:

Confidentiality is documented in the Participant Information Sheet and will be checked on the Consent Form. Parents/guardians will be reassured that they can share as much information as they feel comfortable, however if risk to themselves or others has been disclosed then confidentiality will need to be broken. If this occurs participants will be reminded of the discussion that took place in the introduction and consent process regarding confidentiality. The researcher will continue to discuss the need to break confidentiality and how this applies to their situation. The researcher will aim to make this process collaborative and where possible get the participant to agree with the need to break confidentiality. If they do not agree then the researcher will reiterate their obligation to share this information and what the likely next steps will be. The researcher will initially attempt to contact an appropriate member of the research team to determine the course of action. If appropriate the researcher will aim to do this with the interviewee present so that they are involved in the process. The outcome of this conversation may result in actions like, sign-posting to another service or recommending they seek advice from their GP. If the participant is no longer present then this information will be fed back to them. However, if the level of risk is high and contact cannot be made with a member of the research team the researcher will endeavour to keep the participant calm and in the interview room whilst contacting more immediate services, like other agencies involved or the police. This process is designed to identify, assess and manage risk and at no point will the researcher be able to offer therapy. If participants become distressed but have no or low risk then they will be advised to speak with their GP or other agencies involved as appropriate. The interviewer will also seek supervision from an appropriate member of the research team. This will not require disclosure of identifiable information of the participant but will be a process to support the interviewer should they have any emotional reactions to the interviews. If through this process risk is identified later then the interviewer will contact the interviewee to further discuss the problem and follow the risk process documented previously.

A24. What is the potential for benefit to research participants?

There is no direct benefit for participants. Their participation may provide information that professionals and services find useful when improving their practice to target the needs of parents and guardians of children with Duchennes Muscular Dystrophy.

A26. What are the potential risks for the researchers themselves? (if any)

There should not be any potential risks for the researcher. The nature of the sample i.e. a healthy participant sample, suggest that the participants should not pose a threat to the researcher. The location of the interviews will also provide added support should there be a problem, with individuals contactable in an emergency e.g. if the participant collapses. The researcher will also follow the lone working protocol as documented by the Lancashire Care NHS Foundation Trust and the [REDACTED] by implementing a buddy system for each interview. The researcher will also attend regular supervision to manage any practical and emotional difficulties.

RECRUITMENT AND INFORMED CONSENT

In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? For example, identification may involve a disease register, computerised search of GP records, or review of medical records. Indicate whether this will be done by the direct healthcare team or by researchers acting under arrangements with the responsible care organisation(s).

Participants will be opportunistically sampled and identification of participants will take 6 forms in a staged approach,

based on an opt in recruitment process:

Stage 1

1: Online – via a Contact a Parent discussion board.

2: Posters based at paediatric services involved with the research and other local community services (GP practices, libraries etc).

3: Via an opt-in mail out by local paediatric services based on parents who have been referred to the service. A member of the administration team, who is not involved in the research, will mail out packs of information to parents/guardians who have been referred to the service and have a child with Duchenne Muscular Dystrophy. The parent will need to contact the researcher on the contact details provided to let them know they wish to participate and therefore the administration team will not know who has agreed to participate and the researcher will not have any personal details until the interviewee gives them.

Recruitment will be reviewed after two months of interviewing. If there is a problem then stage 2 of recruitment will be initiated.

Stage 2

4: Via an opt-in mail out by another local paediatric service based on parents who have been referred to the service, following the same mail out procedure as above.

5: Contact support groups (chat/present ideas)

6. Contact children's hospices e.g. [REDACTED] and leave information packs with staff.

In all instances contact details for the researcher will be left and it will be up to the participants to contact the researcher directly.

Should parents/guardians from these recruitment paths contact the researcher and wish to find out more about the study a participant information sheet can be emailed or posted, as they choose. Participants will be asked if further contact to make arrangements can be done by phone to provide the maximum opportunity for participants to attend an interview slot. If they refuse then the researcher will use the preferred method of contact.

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

Yes No

Please give details below:

The paediatric services have minimal databases used to log referrals of parents and guardians using the service. This has a small amount of personal information about the parent/guardian, including addresses. A member of the administration team, who is not involved in the research, will mail out packs of information to parents/guardians who have been referred and appear to fit the inclusion/exclusion criteria. Initially, the mail out will be confined to those who were referred between (April 2011 to April 2012), of which there are approximately (65). This time-scale will be reviewed and extended if there are problems recruiting. The parent will then need to contact the chief investigator on the contact details provided to let them know they wish to participate and therefore the administration team will not know who has agreed to participate.

A27-4. Will researchers or individuals other than the direct care team have access to identifiable personal information of any potential participants?

Yes No

A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?

Yes No

If Yes, please give details of how and where publicity will be conducted, and enclose copy of all advertising material (with version numbers and dates).

Yes. Posters will be put up in local family based services, including but not limited to paediatric services base, local sure start centres and GP practices.

Parents/guardians will also be contacted via an online advert on the Contact a Parent discussion board.

Support groups may also be contacted to arrange a brief presentation summarising the research and leaving information packs and contact details for parents/guardians to contact the researcher.

A29. How and by whom will potential participants first be approached?

A member of the paediatric administration team or the principal researcher will be responsible for approaching participants. This will depend on the method of recruitment.

The paediatric administration team, who will have already had contact with the parent/guardian, will be responsible for the information mail out to parents/guardians selected from the database.

Specialist nurses will also discuss the study during the meetings and provide any families that are interested with an information pack.

The researcher may approach carers in an informal group setting by presenting to local support groups and leaving information for the carers.

All forms of recruitment will function on an opt-in basis, where the foster carer must contact the researcher if they wish to participate.

A30-1. Will you obtain informed consent from or on behalf of research participants?

Yes No

If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

Parents/guardians will be required to give consent to participate at the beginning of the interview during the introduction when they will be talked through the participant information sheet with the researcher. This will be documented on the consent form. If capacity to consent is viewed as a potential problem, then the risk protocol will be followed.

If you are not obtaining consent, please explain why not.

Please enclose a copy of the information sheet(s) and consent form(s).

A30-2. Will you record informed consent (or advice from consultees) in writing?

Yes No

A31. How long will you allow potential participants to decide whether or not to take part?

Once the recruitment period starts after ethical approval participants will be able to contact the researcher and ask questions. However once participants have made an initial contact then the researcher will request that the potential participant notifies the researcher of their decision within a week, to allow for the organisation of the interview and give sufficient time for the participant to make the decision. The researcher will ask if the participant minds being contacted the day before the end of this time period to act as a reminder for the participant.

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs?(e.g. translation, use of interpreters)

Due to financial constraints the study will not be able to make arrangements for specific communication needs.

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? Tick one option only.

The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.

The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.

- The participant would continue to be included in the study.
- Not applicable – informed consent will not be sought from any participants in this research.
- Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

The length of the contact between the participant and the researcher is sufficiently brief (a maximum of an hour and a half) that this is highly unlikely. If however, the researcher has concerns regarding capacity at any point during the interview then this will potentially pose a risk issues and the researcher will follow the identified risk protocol.

If you plan to retain and make further use of identifiable data/tissue following loss of capacity, you should inform participants about this when seeking their consent initially.

CONFIDENTIALITY

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

Storage and use of personal data during the study

A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)? (Tick as appropriate)

- Access to medical records by those outside the direct healthcare team
- Electronic transfer by magnetic or optical media, email or computer networks
- Sharing of personal data with other organisations
- Export of personal data outside the EEA
- Use of personal addresses, postcodes, faxes, emails or telephone numbers
- Publication of direct quotations from respondents
- Publication of data that might allow identification of individuals
- Use of audio/visual recording devices
- Storage of personal data on any of the following:
- Manual files including X-rays
 - NHS computers
 - Home or other personal computers
 - University computers
 - Private company computers
 - Laptop computers

Further details:

University computers or the chief investigator's personal laptop will be used to store data, all of which will be password protected and encrypted. The data stored on the University computers will be saved on the Lancaster University secure network drive, which is located on the virtual private network and is only accessible to the researcher.

The digital recorder cannot be encrypted, consequently following the interview the digital recorder will be stored securely and the audio files will be transferred to a suitably secure computer as quickly soon as possible. After transfer the audio files will be deleted from the digital recorder.

Any paperwork that has personal information on will be stored in a lockable cabinet at Lancaster University.

A38. How will you ensure the confidentiality of personal data? Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.

All electronic data (i.e. audio recordings, not anonymised and anonymised transcripts) will either be stored on the

Lancaster University secure network drive which is located on a virtual private network and is password protected, or it will be stored in an encrypted drive on the chief investigator's personal laptop, which is also password protected. All paperwork containing personal information will be stored in a lockable cabinet at Lancaster University.

Participants will also be asked to give a preferred pseudonym that will be utilised in the transcription and alongside quotes used in the final report and any publications.

Once the transcripts have been anonymised using the pseudonym and removing any potential identifiers the research supervisor will be able to view the transcripts to aid analysis. The field supervisor will only input into the analysis once the transcripts have been coded, thereby making sure that participants' identities are fully protected.

The opt-in recruitment process also means that the researcher will only have participant's personal information once it has been volunteered.

All procedures have been designed in line with the BPS code of ethics and conduct (2009) and the HPC guidance on conduct and ethics for students (2009).

A40. Who will have access to participants' personal data during the study? Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.

The chief investigator will have access to the participant's personal data. The designated 'Buddy' from the lone working protocol will also temporarily have access to this information during the interview to ensure the researcher's safety.

This will be deleted once the safety of the researcher is established. The attachments containing the information will only be opened as per the agreed lone working protocol, and therefore no-one other than the researcher should have access to this information unless there is a perceived danger.

Storage and use of data after the end of the study

A43. How long will personal data be stored or accessed after the study has ended?

- Less than 3 months
 3 – 6 months
 6 – 12 months
 12 months – 3 years
 Over 3 years

INCENTIVES AND PAYMENTS

A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?

- Yes No

A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?

- Yes No

A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?

- Yes No

NOTIFICATION OF OTHER PROFESSIONALS

A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?

Yes No

If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.

PUBLICATION AND DISSEMINATION

A50. Will the research be registered on a public database?

Yes No

Please give details, or justify if not registering the research.

No, however the research will be registered when the researcher pursues a publication.

Registration of research studies is encouraged wherever possible.

You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

A51. How do you intend to report and disseminate the results of the study? Tick as appropriate:

- Peer reviewed scientific journals
- Internal report
- Conference presentation
- Publication on website
- Other publication
- Submission to regulatory authorities
- Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- No plans to report or disseminate the results
- Other (please specify)

Provide a summary of the findings to the participants and paediatric services involved if they are interested. This could be a presentation or a brief summary of the write-up. This research will also be presented at the Lancaster University thesis presentation day open to peers and other allied professionals.

A53. Will you inform participants of the results?

Yes No

Please give details of how you will inform participants or justify if not doing so.

Participants will be asked whether they would like to receive feedback on the results. They will be offered the opportunity to do this one-to-one, over the phone or face-to-face.

5. Scientific and Statistical Review

A54. How has the scientific quality of the research been assessed? Tick as appropriate:

- Independent external review
- Review within a company

- Review within a multi-centre research group
 Review within the Chief Investigator's institution or host organisation
 Review within the research team
 Review by educational supervisor
 Other

Justify and describe the review process and outcome. If the review has been undertaken but not seen by the researcher, give details of the body which has undertaken the review:

A preliminary research proposal was presented by the researcher and this was peer reviewed at a meeting attended by trainee clinical psychologists and two academic tutors. At this meeting the researcher was required to justify the feasibility of their decisions on the design of the project. This feedback has been used to identify potential ethical, practical and theoretical problems and make modifications. The outcome of this meeting was then summarised and fed back to the two tutors at the meeting, with the suggested alterations that could be made.

The research protocol has then been draft read and discussed with the research and field supervisors. A research contract has been agreed with both the field and research supervisors, stating the level of input they are required to help support the chief investigator and also utilise their expertise.

All paperwork has been reviewed by the Research Support Office to check the quality of the study and subsequently granting indemnity.

For all studies except non-doctoral student research, please enclose a copy of any available scientific critique reports, together with any related correspondence.

For non-doctoral student research, please enclose a copy of the assessment from your educational supervisor/ institution.

A59. What is the sample size for the research? *How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.*

Total UK sample size: 12
 Total international sample size (including UK): 12
 Total in European Economic Area: 12

Further details:

A sample size of between 6-12 participants has been estimated for this study, but will need to be monitored and reviewed throughout the analysis.

A60. How was the sample size decided upon? *If a formal sample size calculation was used, indicate how this was done, giving sufficient information to justify and reproduce the calculation.*

Up to 12 interviews/participants is sufficient for an in-depth narrative analysis.

A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.

A qualitative methodology, specifically narrative analysis, has been chosen to focus on the experiences and understanding of parents/guardians of children with Duchenne Muscular Dystrophy, whilst allowing the data to be inductively analysed to generate ecologically valid, subjective and findings (Elliott, Fischer & Rennie, 1999).

Interviews will be transcribed and analysed. Narratives will be identified within each transcript allowing the researcher to explore how parents/guardians understand their experience. Crossley's (2007) narrative analytic model will be used to guide the analysis and Braun and Clark's (2006) 15 point checklist will be considered to help maintain quality.

Currently it is expected that this will be done by hand but the researcher hopes to develop skills to utilise a transcription and coding programme called NVIVO, which should speed up the analysis and make the process of analysis more accessible.

The anonymised findings will be discussed within the research team to strengthen the quality of the analysis.

6. MANAGEMENT OF THE RESEARCH

A63. Other key investigators/collaborators. Please include all grant co-applicants, protocol co-authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.

	Title	Forename/Initials	Surname
	Dr	Pete	Greasley
Post	Teaching Fellow in Research Methods		
Qualifications	BSc (Hons), PhD, PGCHEP		
Employer	Lancashire Care NHS Foundation Trust		
Work Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire		
Post Code	LA1 4YG		
Telephone	01524593535		
Fax			
Mobile			
Work Email	p.greasley@lancaster.ac.uk		

	Title	Forename/Initials	Surname
	Dr	Clare	Dixon
Post	Clinical Psychologist & Clinical Tutor		
Qualifications	BSc (Hons); PhD; DClinPsy		
Employer	Lancashire Care NHS Foundation Trust		
Work Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire		
Post Code	LA1 4YG		
Telephone	01524593535		
Fax			
Mobile			
Work Email	c.dixon@lancaster.ac.uk		

A64. Details of research sponsor(s)

A64-1. Sponsor

Lead Sponsor

- Status: NHS or HSC care organisation
 Academic
 Pharmaceutical industry
 Medical device industry
 Local Authority
 Other social care provider (including voluntary sector or private organisation)
 Other

Commercial status:

If Other, please specify:

Contact person

Name of organisation Lancaster University
Given name Debbie
Family name Knight
Address Research Support Office, B Floor, University House, Lancaster University
Town/city Lancaster
Post code LA1 4YW
Country UNITED KINGDOM
Telephone 01524592605
Fax
E-mail ethics@lancaster.ac.uk

Is the sponsor based outside the UK?

Yes No

Under the Research Governance Framework for Health and Social Care, a sponsor outside the UK must appoint a legal representative established in the UK. Please consult the guidance notes.

A65. Has external funding for the research been secured?

- Funding secured from one or more funders
 External funding application to one or more funders in progress
 No application for external funding will be made

What type of research project is this?

- Standalone project
 Project that is part of a programme grant
 Project that is part of a Centre grant
 Project that is part of a fellowship/ personal award/ research training award
 Other

Other – please state:

A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?

Yes No

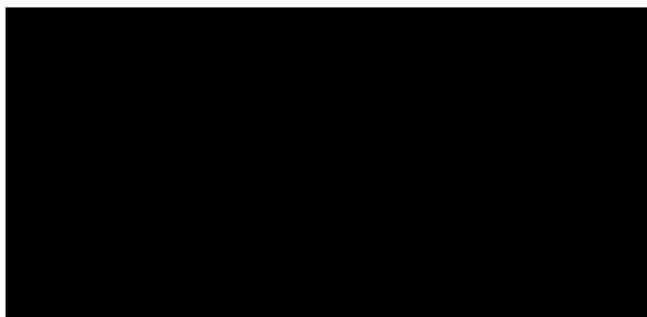
Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

A68-1. Give details of the lead NHS R&D contact for this research:

Title Forename/Initials Surname

Organisation
Address

Post Code
Work Email
Telephone
Fax
Mobile



Details can be obtained from the NHS R&D Forum website: <http://www.rdforum.nhs.uk>

A69-1. How long do you expect the study to last in the UK?

Planned start date: 01/10/2013

Planned end date: 30/06/2014

Total duration:

Years: 0 Months: 8 Days: 30

A71-2. Where will the research take place? (Tick as appropriate)

- England
- Scotland
- Wales
- Northern Ireland
- Other countries in European Economic Area

Total UK sites in study 2

Does this trial involve countries outside the EU?

- Yes No

A72. What host organisations (NHS or other) in the UK will be responsible for the research sites? Please indicate the type of organisation by ticking the box and give approximate numbers of planned research sites:

- NHS organisations in England 2
- NHS organisations in Wales
- NHS organisations in Scotland
- HSC organisations in Northern Ireland
- GP practices in England
- GP practices in Wales
- GP practices in Scotland
- GP practices in Northern Ireland
- Social care organisations
- Phase 1 trial units
- Prison establishments
- Probation areas
- Independent hospitals
- Educational establishments

- Independent research units
 Other (give details)

Total UK sites in study: 2

A76. Insurance/ indemnity to meet potential legal liabilities

Note: in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland

A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? Please tick box(es) as applicable.

Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (NHS sponsors only)
 Other insurance or indemnity arrangements will apply (give details below)

Lancaster University legal liability cover will apply.

Please enclose a copy of relevant documents.

A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the sponsor(s) or employer(s) for harm to participants arising from the design of the research? Please tick box(es) as applicable.

Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (protocol authors with NHS contracts only)
 Other insurance or indemnity arrangements will apply (give details below)

Lancaster University legal liability cover will apply.

Please enclose a copy of relevant documents.

A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?

Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.

- NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)
 Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

This is covered by standard NHS Indemnity.

Please enclose a copy of relevant documents.

PART C: Overview of research sites

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. For NHS sites, the host organisation is the Trust or Health Board. Where the research site is a primary care site, e.g. GP practice, please insert the host organisation (PCT or Health Board) in the Institution row and insert the research site (e.g. GP practice) in the Department row.

Research site	Investigator/ Collaborator/ Contact
Institution name	Title
Department name	First name/ Initials
Street address	Surname
Town/city	
Post Code	
Participant Identification Centre(PIC)-Collaborator/ Contact	

PART D: Declarations

D1. Declaration by Chief Investigator

1. The information in this form is accurate to the best of my knowledge and belief and I take full responsibility for it.
2. I undertake to abide by the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research.
3. If the research is approved I undertake to adhere to the study protocol, the terms of the full application as approved and any conditions set out by review bodies in giving approval.
4. I undertake to notify review bodies of substantial amendments to the protocol or the terms of the approved application, and to seek a favourable opinion from the main REC before implementing the amendment.
5. I undertake to submit annual progress reports setting out the progress of the research, as required by review bodies.
6. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to security and confidentiality of patient or other personal data, including the need to register when necessary with the appropriate Data Protection Officer. I understand that I am not permitted to disclose identifiable data to third parties unless the disclosure has the consent of the data subject or, in the case of patient data in England and Wales, the disclosure is covered by the terms of an approval under Section 251 of the NHS Act 2006.
7. I understand that research records/data may be subject to inspection by review bodies for audit purposes if required.
8. I understand that any personal data in this application will be held by review bodies and their operational managers and that this will be managed according to the principles established in the Data Protection Act 1998.
9. I understand that the information contained in this application, any supporting documentation and all correspondence with review bodies or their operational managers relating to the application:
 - Will be held by the REC (where applicable) until at least 3 years after the end of the study; and by NHS R&D offices (where the research requires NHS management permission) in accordance with the NHS Code of Practice on Records Management.
 - May be disclosed to the operational managers of review bodies, or the appointing authority for the REC (where applicable), in order to check that the application has been processed correctly or to investigate any complaint.
 - May be seen by auditors appointed to undertake accreditation of RECs (where applicable).
 - Will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.
 - May be sent by email to REC members.
10. I understand that information relating to this research, including the contact details on this application, may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 1998.
11. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named below. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Contact point for publication *(Not applicable for R&D Forms)*

NRES would like to include a contact point with the published summary of the study for those wishing to seek further information. We would be grateful if you would indicate one of the contact points below.

- Chief Investigator
 Sponsor

NHS REC Form

Reference:

IRAS Version 3.5

- Study co-ordinator
- Student
- Other – please give details
- None

Access to application for training purposes (Not applicable for R&D Forms)*Optional – please tick as appropriate:*

I would be content for members of other RECs to have access to the information in the application in confidence for training purposes. All personal identifiers and references to sponsors, funders and research units would be removed.

Signature: Toni Deavin

Print Name: ANTOINETTE DEAVIN

Date: 02.12.2013 (dd/mm/yyyy)

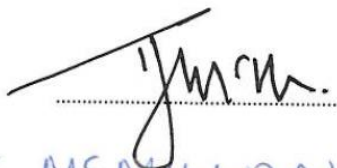
D2. Declaration by the sponsor's representative

If there is more than one sponsor, this declaration should be signed on behalf of the co-sponsors by a representative of the lead sponsor named at A64-1.

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements, as described in question A76, will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.
6. The duties of sponsors set out in the Research Governance Framework for Health and Social Care will be undertaken in relation to this research.
7. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Signature:



Print Name: T. McMILLAN

Post: PRO VC RESEARCH

Organisation: LANCASTER UNIVERSITY

Date: 28-11-13

(dd/mm/yyyy)

D3. Declaration for student projects by academic supervisor(s)

1. I have read and approved both the research proposal and this application. I am satisfied that the scientific content of the research is satisfactory for an educational qualification at this level.
2. I undertake to fulfil the responsibilities of the supervisor for this study as set out in the Research Governance Framework for Health and Social Care.
3. I take responsibility for ensuring that this study is conducted in accordance with the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research, in conjunction with clinical supervisors as appropriate.
4. I take responsibility for ensuring that the applicant is up to date and complies with the requirements of the law and relevant guidelines relating to security and confidentiality of patient and other personal data, in conjunction with clinical supervisors as appropriate.

Academic supervisor 1

Signature:



Print Name:

PETE GRAYLEY

Post:

TEACHING FELLOW

Organisation:

LANCASTER UNIVERSITY.

Date: 02/12/2013 (dd/mm/yyyy)

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please enter a short title for this project (maximum 70 characters)
Parents' stories of adjustment to their child's chronic illness

1. Is your project research?

Yes No

2. Select one category from the list below:

- Clinical trial of an investigational medicinal product
- Clinical investigation or other study of a medical device
- Combined trial of an investigational medicinal product and an investigational medical device
- Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- Basic science study involving procedures with human participants
- Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- Study involving qualitative methods only
- Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- Study limited to working with data (specific project only)
- Research tissue bank
- Research database

If your work does not fit any of these categories, select the option below:

Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? Yes No
- b) Will you be taking new human tissue samples (or other human biological samples)? Yes No
- c) Will you be using existing human tissue samples (or other human biological samples)? Yes No

3. In which countries of the UK will the research sites be located?(Tick all that apply)

- England
- Scotland
- Wales
- Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

- England
- Scotland
- Wales
- Northern Ireland
- This study does not involve the NHS

4. Which review bodies are you applying to?

- NHS/HSC Research and Development offices
- Social Care Research Ethics Committee
- Research Ethics Committee
- National Information Governance Board for Health and Social Care (NIGB)
- National Offender Management Service (NOMS) (Prisons & Probation)

For NHS/HSC R&D offices, the CI must create Site-Specific Information Forms for each site, in addition to the study-wide forms, and transfer them to the PIs or local collaborators.

5. Will any research sites in this study be NHS organisations?

- Yes No

5a. Are all the research costs and infrastructure costs for this study provided by an NIHR Biomedical Research Centre, NIHR Biomedical Research Unit, NIHR Collaboration for Leadership in Health Research and Care (CLAHRC) or NIHR Research Centre for Patient Safety & Service Quality in all study sites?

- Yes No

If yes, NHS permission for your study will be processed through the NIHR Coordinated System for gaining NHS Permission (NIHR CSP).

5b. Do you wish to make an application for the study to be considered for NIHR Clinical Research Network (CRN) support and inclusion in the NIHR Clinical Research Network (CRN) Portfolio? Please see information button for further details.

- Yes No

If yes, NHS permission for your study will be processed through the NIHR Coordinated System for gaining NHS Permission (NIHR CSP) and you must complete a NIHR Clinical Research Network (CRN) Portfolio Application Form immediately after completing this project filter and before completing and submitting other applications.

6. Do you plan to include any participants who are children?

- Yes No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

- Yes No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the NIGB Ethics and Confidentiality Committee to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

Yes No

9. Is the study or any part of it being undertaken as an educational project?

Yes No

Please describe briefly the involvement of the student(s):
The student will be the chief investigator of the study.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

Yes No

10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

Yes No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

Yes No

Site-Specific Information Form (NHS sites)

Is the site hosting this research a NHS site or a non-NHS site? NHS sites include Health and Social Care organisations in Northern Ireland. The sites hosting the research are the sites in which or through which research procedures are conducted. For NHS sites, this includes sites where NHS staff are participants.

- NHS site
 Non-NHS site

This question must be completed before proceeding. The filter will customise the form, disabling questions which are not relevant to this application.

One Site-Specific Information Form should be completed for each research site and submitted to the relevant R&D office with the documents in the checklist. See guidance notes.

The data in this box is populated from Part A:

Title of research:
Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy

Short title: Parents' stories of adjustment to their child's chronic illness

Chief Investigator: Title Forename/Initials Surname
Ms Antoinette Deavin

Name of NHS Research Ethics Committee to which application for ethical review is being made:
Hampshire-B REC

Project reference number from above REC: 13/SC/0649

1-1. Give the name of the NHS organisation responsible for this research site

[REDACTED]

1-3. In which country is the research site located?

- England
 Wales
 Scotland
 Northern Ireland

1-4. Is the research site a GP practice or other Primary Care Organisation?

- Yes No

2. Who is the Principal Investigator or Local Collaborator for this research at this site?

Select the appropriate title: Principal Investigator
 Local Collaborator

Title Forename/Initials Surname

Post
Qualifications
Organisation
Work Address

PostCode
Work E-mail
Work Telephone
Mobile
Fax



a) Approximately how much time will this person allocate to conducting this research? *Please provide your response in terms of Whole Time Equivalent (WTE).*
0.03 WTE

b) Does this person hold a current substantive employment contract, Honorary Clinical Contract or Honorary Research Contract with the NHS organisation or accepted by the NHS organisation? Yes No

A copy of a current CV for the Principal Investigator (maximum 2 pages of A4) must be submitted with this form.

3. Please give details of all locations, departments, groups or units at which or through which research procedures will be conducted at this site and describe the activity that will take place.

Please list all locations/departments etc where research procedures will be conducted within the NHS organisation, describing the involvement in a few words. Where access to specific facilities will be required these should also be listed for each location.

Name the main location/department first. Give details of any research procedures to be carried out off site, for example in participants' homes.

Location	Activity/facilities
1 [Redacted]	The [Redacted] will help with a small amount of administration. The chief investigator will put together information packs that an administrator at the service will add labels of names and addresses to and post them out. This will require up to an hour of the administrator's time. Nurses within the team will also discuss the study with patients during their meetings. Parents will not be directly recruited. This should take approximately 5 minutes per meeting. There will also be rooms available to book for the interviews within the building.
2 Various local family services e.g. Children's centres, GP practices, libraries. At the interviewees house.	Interviews may be conducted at a variety of locations, dependent on the needs of the interviewee. Ideally these will be located in a building which holds local family services, however the interview can be done at the interviewees home if necessary.

4. Please give details of all centres where potential participants for this research site will be identified.

Participant Identification Centre	Name(s) of individuals involved in identifying potential participants
-----------------------------------	---



5. Please give details of all other members of the research team at this site.

1

Title Forename/Initials Surname



Work E-mail

Employing organisation

Post

Qualifications

Role in research team:

other (please specify)

Project advisor

a) Approximately how much time (approximately) will this person allocate to conducting this research? Please provide your response in terms of Whole Time Equivalents (WTE).

0.02 WTE

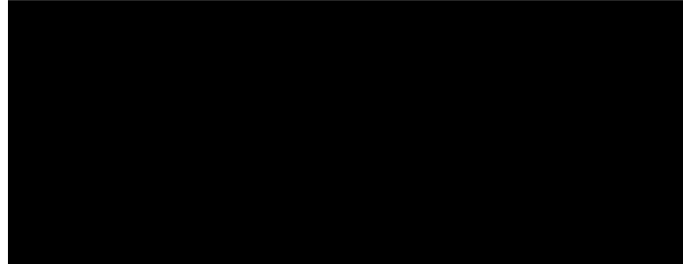
b) Does this person hold a current substantive employment contract, Honorary Clinical Contract or Honorary Research Contract with the NHS organisation or accepted by the NHS organisation?

Yes

No

2

Title Forename/Initials Surname



Work E-mail

Employing organisation

Post

Qualifications

Role in research team:

other (please specify)

Recruitment

a) Approximately how much time (approximately) will this person allocate to conducting this research? Please provide your response in terms of Whole Time Equivalents (WTE).

0.02 WTE

b) Does this person hold a current substantive employment contract, Honorary Clinical Contract or Honorary Research Contract with the NHS organisation or accepted by the NHS organisation?

Yes

No

3

Title Forename/Initials Surname



Work E-mail

Employing organisation	[REDACTED]	
Post	[REDACTED]	
Qualifications	[REDACTED]	
Role in research team:	other (please specify)	Recruitment

a) Approximately how much time (approximately) will this person allocate to conducting this research? Please provide your response in terms of Whole Time Equivalents (WTE).
0.02 WTE

b) Does this person hold a current substantive employment contract, Honorary Clinical Contract or Honorary Research Contract with the NHS organisation or accepted by the NHS organisation? Yes No

6. Does the Principal Investigator or any other member of the site research team have any direct personal involvement (e.g. financial, share-holding, personal relationship etc) in the organisation sponsoring or funding the research that may give rise to a possible conflict of interest?

Yes No

7. What is the proposed local start and end date for the research at this site?

Start date: 01/10/2013
End date: 30/06/2014
Duration (Months): 8

8-1. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. (These include seeking consent, interviews, non-clinical observations and use of questionnaires.)

Columns 1-4 have been completed with information from A18 as below:

- Total number of interventions/procedures to be received by each participant as part of the research protocol.
- If this intervention would have been routinely given to participants as part of their care, how many of the total would have been routine?
- Average time taken per intervention (minutes, hours or days)
- Details of who will conduct the procedure, and where it will take place

Please complete Column 5 with details of the names of individuals or names of staff groups who will conduct the procedure at this site.

Intervention or procedure	1	2	3	4	5
Recruitment	3	0	10	Participants will be recruited through posters, an information mail out to service users and an online advert on Contact a Parent's forum. The specialist nurses will also discuss the study with parents they meet during their clinics/meetings and give them an information pack so they can contact the chief investigator.	Antoinette Deavin (chief investigator for the study). [REDACTED]
Giving information	1	0	5	The chief investigator will hand out and explain the Participant Information Sheet just before the interview.	Antoinette Deavin (chief investigator).
Seeking consent	1	0	5	Th chief investigator will explain the Consent	Antoinette Deavin (chief

			Form and seek informed written consent from the participant just before the interview starts.	investigator).	
Interview	1	0	90	The chief investigator will conduct the interview.	Antoinette Deavin (chief investigator).
Post interview questions	1	0	10	The chief investigator will receive post interview questions.	Antoinette Deavin (chief investigator).
Feedback	1	0	30	The chief investigator will contact the interviewees to ask if they would like to receive a summary of the study.	Antoinette Deavin (chief investigator).
Review of participant's account	1	0	30	The chief investigator will contact the interviewee to discuss their re-storied account.	Antoinette Deavin (chief investigator).

8-2. Will any aspects of the research at this site be conducted in a different way to that described in Part A or the protocol?

Yes No

If Yes, please note any relevant changes to the information in the above table.

Are there any changes other than those noted in the table?

10. How many research participants/samples is it expected will be recruited/obtained from this site?

Between 6-12 participants will be recruited for interview.

11. Give details of how potential participants will be identified locally and who will be making the first approach to them to take part in the study.

In the first instance participants will be recruited through posters, the specialist nurses discussing the study with parents at meetings and an information post out to service users of the [REDACTED]. Parents will need to contact the chief investigator for more information if they have seen a poster, thereby opting-in to being contacted.

The chief investigator will leave information packs with the nurses and parents will again need to contact the chief investigator to opt-in.

One of the [REDACTED] will send out prepared information packs to parents who have been in contact with the service. The parent will then need to contact the chief investigator to opt-in to the study. This will ensure that the chief researcher will not know who has been contacted and the [REDACTED] will not know who is being interviewed.

12. Who will be responsible for obtaining informed consent at this site? What expertise and training do these persons have in obtaining consent for research purposes?

Name	Expertise/training
Antoinette Deavin	I am the chief investigator of the project and therefore I am in a position to answer questions about the project. I have been involved in at least three other research projects where informed consent was required and as such I am fully aware of the process. I am also currently training to be a clinical psychologist where we regularly discuss consent and confidentiality. We have thorough training throughout the course to address this topic and the principles of best practice.

15-1. Is there an independent contact point where potential participants can seek general advice about taking part in research?

There is an independent contact point where potential participants can seek general advice about taking part in

research. Participants can contact Beverley Lowe who is the Lancashire Care Research & Development Senior Officer, contactable at:

Nursing and Governance Directorate
Lancashire Care NHS Foundation Trust
T: 01772 773498 (Anisa Lakhi, R&D Secretary)
M: 07508 601925

The Participant Information Sheet has details of individuals participants can contact to discuss any concerns or complaints about the research.
The Participant Information Sheet also includes details of the participant's right to withdraw and confidentiality. These will also be discussed with the interviewee before they give consent.

15-2. Is there a contact point where potential participants can seek further details about this specific research project?

Participants will be able to contact the chief investigator for more information. The contact details are on the posters and participant information sheets.

16. Are there any changes that should be made to the generic content of the information sheet to reflect site-specific issues in the conduct of the study? A substantial amendment may need to be discussed with the Chief Investigator and submitted to the main REC.

No, there are no changes to the generic content of the information sheet that need to be made to reflect site-specific issues.

Please provide a copy on headed paper of the participant information sheet and consent form that will be used locally. Unless indicated above, this must be the same generic version submitted to/approved by the main REC for the study while including relevant local information about the site, investigator and contact points for participants (see guidance notes).

17. What local arrangements have been made for participants who might not adequately understand verbal explanations or written information given in English, or who have special communication needs? (e.g. translation, use of interpreters etc.)

Due to financial constraints the study will not be able to make arrangements for individuals with specific communication needs.

18. What local arrangements will be made to inform the GP or other health care professionals responsible for the care of the participants?

GPs or other health care practitioners will not be informed of the interviewees participation in the study unless there is a need to break confidentiality due to perceived risk. Whilst the participants (parents) may have been involved with services, like the [REDACTED] or social services, they will not be directly under their care and therefore these services will not need notifying of their participation.

19. What arrangements (e.g. facilities, staffing, psychosocial support, emergency procedures) will be in place at the site, where appropriate, to minimise the risks to participants and staff and deal with the consequences of any harm?

Wherever possible interview locations have been chosen to be local, familiar and safe for the participants. Local family services can be used to conduct the interviews and will ensure that there are other professionals within a brief distance should an emergency arise.

Participants will also be explicitly told prior to the interview that they do not need to discuss any topics which they may find distressing. They will also be notified of their rights relating to the research, e.g. right to withdraw, confidentiality etc. Should a participant become distressed during the interview and there are no risk concerns then they will be asked how they would like proceed for example, take a break or stop the interview. If appropriate they will also be recommended to seek support from their GP or social worker. It may also be appropriate for the researcher to suggest that the interviewee seeks support from the [REDACTED] as they are a service specifically designed to help support families with children with a chronic illness.

If a potential risk has been identified then the interviewer will follow the risk protocol.

The interviewer will ensure that at least two people are aware that she is attending an appointment. This will be so that support can be sought from her local collaborator if a risk issue should arise and also a buddy will be notified so that they can follow the buddy/lone working system that has been put in place should the interviewer not check in at the

allocated time.

The interviewer will also seek supervision when appropriate to help assess their interview skills or process any of their own reactions to the interviews. As the names and any identifiers of the participant will not be used in this discussion, confidentiality will not be broken. Supervision will be used to focus on and support the researcher and not the interviewee.

20. What are the arrangements for the supervision of the conduct of the research at this site? Please give the name and contact details of any supervisor not already listed in the application.

This project will be supervised by a research/academic supervisor from Lancaster University, a field supervisor and a local collaborator from the [REDACTED]. All of these supervisors have been identified previously in the application.

21. What external funding will be provided for the research at this site?

- Funded by commercial sponsor
- Other funding
- No external funding

How will the costs of the research be covered?

Costs of the research will be covered by Lancaster University as part of the contract for the Doctorate in Clinical Psychology, which the chief investigator is undertaking.

23. Authorisations required prior to R&D approval

The local research team are responsible for contacting the local NHS R&D office about the research project. Where the research project is proposed to be coordinated centrally and therefore there is no local research team, it is the responsibility of the central research team to instigate this contact with local R&D.

NHS R&D offices can offer advice and support on the set-up of a research project at their organisation, including information on local arrangements for support services relevant to the project. These support services may include clinical supervisors, line managers, service managers, support department managers, pharmacy, data protection officers or finance managers depending on the nature of the research.

Obtaining the necessary support service authorisations is not a pre-requisite to submission of an application for NHS research permission, but all appropriate authorisations must be in place before NHS research permission will be granted. Processes for obtaining authorisations will be subject to local arrangements, but the minimum expectation is that the local R&D office has been contacted to notify it of the proposed research project and to discuss the project's needs prior to submission of the application for NHS research permission via IRAS.

Failure to engage with local NHS R&D offices prior to submission may lead to unnecessary delays in the process of this application for NHS research permissions.

Declaration:

I confirm that the relevant NHS organisation R&D office has been contacted to discuss the needs of the project and local arrangements for support services. I understand that failure to engage with the local NHS R&D office before submission of this application may result in unnecessary delays in obtaining NHS research permission for this project.

Please give the name and contact details for the NHS R&D office staff member you have discussed this application with:

Please note that for some sites the NHS R&D office contact may not be physically based at the site. For contact details refer to the guidance for this question.

Title Forename/Initials Surname
[REDACTED]

NHS SSI

IRAS Version 3.5

Work E-mail
Work Telephone


Declaration by Principal Investigator or Local Collaborator

1. The information in this form is accurate to the best of my knowledge and I take full responsibility for it.
2. I undertake to abide by the ethical principles underpinning the World Medical Association's Declaration of Helsinki and relevant good practice guidelines in the conduct of research.
3. If the research is approved by the main REC and NHS organisation, I undertake to adhere to the study protocol, the terms of the application of which the main REC has given a favourable opinion and the conditions requested by the NHS organisation, and to inform the NHS organisation within local timelines of any subsequent amendments to the protocol.
4. If the research is approved, I undertake to abide by the principles of the Research Governance Framework for Health and Social Care.
5. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to the conduct of research.
6. I undertake to disclose any conflicts of interest that may arise during the course of this research, and take responsibility for ensuring that all staff involved in the research are aware of their responsibilities to disclose conflicts of interest.
7. I understand and agree that study files, documents, research records and data may be subject to inspection by the NHS organisation, the sponsor or an independent body for monitoring, audit and inspection purposes.
8. I take responsibility for ensuring that staff involved in the research at this site hold appropriate contracts for the duration of the research, are familiar with the Research Governance Framework, the NHS organisation's Data Protection Policy and all other relevant policies and guidelines, and are appropriately trained and experienced.
9. I undertake to complete any progress and/or final reports as requested by the NHS organisation and understand that continuation of permission to conduct research within the NHS organisation is dependent on satisfactory completion of such reports.
10. I undertake to maintain a project file for this research in accordance with the NHS organisation's policy.
11. I take responsibility for ensuring that all serious adverse events are handled within the NHS organisation's policy for reporting and handling of adverse events.
12. I understand that information relating to this research, including the contact details on this application, will be held by the R&D office and may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 1998.
13. I understand that the information contained in this application, any supporting documentation and all correspondence with the R&D office and/or the REC system relating to the application will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.

Signature of Principal Investigator
or Local Collaborator:

Trui Deavin

Print Name:

ANTOINETTE DEAVIN

Date:

06.12.13

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please enter a short title for this project (maximum 70 characters)
Parents' stories of adjustment to their child's chronic illness

1. Is your project research?

Yes No

2. Select one category from the list below:

- Clinical trial of an investigational medicinal product
- Clinical investigation or other study of a medical device
- Combined trial of an investigational medicinal product and an investigational medical device
- Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- Basic science study involving procedures with human participants
- Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- Study involving qualitative methods only
- Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- Study limited to working with data (specific project only)
- Research tissue bank
- Research database

If your work does not fit any of these categories, select the option below:

Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? Yes No
- b) Will you be taking new human tissue samples (or other human biological samples)? Yes No
- c) Will you be using existing human tissue samples (or other human biological samples)? Yes No

3. In which countries of the UK will the research sites be located? *(Tick all that apply)*

- England
- Scotland
- Wales
- Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

- England
- Scotland
- Wales
- Northern Ireland
- This study does not involve the NHS

4. Which review bodies are you applying to?

- NHS/HSC Research and Development offices
- Social Care Research Ethics Committee
- Research Ethics Committee
- National Information Governance Board for Health and Social Care (NIGB)
- National Offender Management Service (NOMS) (Prisons & Probation)

For NHS/HSC R&D offices, the CI must create Site-Specific Information Forms for each site, in addition to the study-wide forms, and transfer them to the PIs or local collaborators.

5. Will any research sites in this study be NHS organisations?

- Yes
- No

5a. Are all the research costs and infrastructure costs for this study provided by an NIHR Biomedical Research Centre, NIHR Biomedical Research Unit, NIHR Collaboration for Leadership in Health Research and Care (CLAHRC) or NIHR Research Centre for Patient Safety & Service Quality in all study sites?

- Yes
- No

If yes, NHS permission for your study will be processed through the NIHR Coordinated System for gaining NHS Permission (NIHR CSP).

5b. Do you wish to make an application for the study to be considered for NIHR Clinical Research Network (CRN) support and inclusion in the NIHR Clinical Research Network (CRN) Portfolio? Please see information button for further details.

- Yes
- No

If yes, NHS permission for your study will be processed through the NIHR Coordinated System for gaining NHS Permission (NIHR CSP) and you must complete a NIHR Clinical Research Network (CRN) Portfolio Application Form immediately after completing this project filter and before completing and submitting other applications.

6. Do you plan to include any participants who are children?

- Yes
- No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

- Yes
- No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the NIGB Ethics and Confidentiality Committee to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

Yes No

9. Is the study or any part of it being undertaken as an educational project?

Yes No

Please describe briefly the involvement of the student(s):
The student will be the chief investigator of the study.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

Yes No

10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

Yes No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

Yes No

Integrated Research Application System
Application Form for Research involving qualitative methods only

NHS/HSC R&D Form (project information)

Please refer to the *Submission and Checklist* tabs for instructions on submitting R&D applications.

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting [Help](#).

Please define any terms or acronyms that might not be familiar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
Parents' stories of adjustment to their child's chronic illness

PART A: Core study information

1. ADMINISTRATIVE DETAILS

A1. Full title of the research:

Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy

A2-1. Educational projects

Name and contact details of student(s):

Student 1

	Title Forename/Initials Surname
	Ms Antoinette Deavin
Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire
Post Code	LA1 4YG
E-mail	a.deavin@lancaster.ac.uk
Telephone	01524592754
Fax	

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:
Doctorate in Clinical Psychology (DClinPsy)

Name of educational establishment:
Lancaster University

Name and contact details of academic supervisor(s):

Academic supervisor 1

	Title Forename/Initials Surname Dr Pete Greasley
Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire
Post Code	LA1 4YG
E-mail	p.greasley@lancaster.ac.uk
Telephone	01524593535
Fax	

Please state which academic supervisor(s) has responsibility for which student(s):
Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

Student(s)	Academic supervisor(s)
Student 1 Ms Antoinette Deavin	<input checked="" type="checkbox"/> Dr Pete Greasley

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

A2-2. Who will act as Chief Investigator for this study?

- Student
 Academic supervisor
 Other

A3-1. Chief Investigator:

	Title Forename/Initials Surname Ms Antoinette Deavin
Post	Trainee Clinical Psychologist
Qualifications	Psychology BSc, Psychology MRes
Employer	Lancashire Care
Work Address	linical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire
Post Code	LA1 4YG
Work E-mail	a.deavin@lancaster.ac.uk
* Personal E-mail	
Work Telephone	01524592754
* Personal Telephone/Mobile	
Fax	

* This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.

A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?

This contact will receive copies of all correspondence from REC and R&D reviewers that is sent to the CI.

	Title Forename/Initials Surname
	Ms Debbie Knight
Address	Research Support Office, B Floor, University House, Lancaster University, Lancaster
Post Code	LA1 4YW
E-mail	ethics@lancaster.ac.uk
Telephone	01524592605
Fax	

A5-1. Research reference numbers. *Please give any relevant references for your study:*

Applicant's/organisation's own reference number, e.g. R & D (if available):

Sponsor's/protocol number: 1

Protocol Version: 1

Protocol Date:

Funder's reference number:

Project website:

Additional reference number(s):

Ref.Number	Description	Reference Number
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Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

Yes No

Please give brief details and reference numbers.

2. OVERVIEW OF THE RESEARCH

To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.

A6-1. Summary of the study. *Please provide a brief summary of the research (maximum 300 words) using language easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, this summary will be published on the website of the National Research Ethics Service following the ethical review.*

Duchenne Muscular Dystrophy (DMD) is a genetic, chronic and life limiting disorder that affects approximately 1 in 3,600 boys in the UK. It results in muscle degeneration, abnormal bone development and an array of other physiological changes, the onset of which can be seen in early infancy. These physiological problems present in everyday life as low endurance, difficulties standing and walking, loss of movement, paralysis, and possibly intellectual impairment. The average life expectancy of children diagnosed with DMD is approximately their late 20s.

Understandably a diagnosis of DMD and the subsequent changes that occur will have an impact not only on the child but their family. Parents in particular are expected to manage their child's health, emotional and psychological needs in addition to their own, requiring a level of adjustment.

Current literature has begun to highlight generic factors relevant to parental adjustment to paediatric chronic illness; however it does not take into account the meaning of these experiences for parents and consequently what they find

important for adjustment, which would account for the interactions between factors. Additionally, there is a lack of qualitative research relating to DMD generally and specifically parental adjustment. Consequently narrative analysis will be used to hear parents' stories of adjustment, allowing them to express what has been important to them in their adjustment and how they understand that process. Up to 12 parents/ long-term guardians will be recruited for face to face interviews that will last up to an hour and a half and will be located in their homes or local family based services. Recruitment will take place in two stages, dependent on need:

1. Through an on online advert; posters in local services and an information mail out to those who have been referred to local paediatric services.
2. Through a brief presentation to support groups and contacting local children's hospices.

A6-2. Summary of main issues. *Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.*

Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, R&D office or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.

Ethical issues

Recruitment will primarily rely on participants opting in by contacting the researcher, meaning that the researcher will only receive the participant's contact details when the participant gives them to the researcher directly. During the introduction to the study confidentiality, anonymity, data protection, consent and withdrawal will be discussed.

Confidentiality is documented in the Participant Information Sheet and will be checked on the Consent Form. Parents will be reassured that they can share as much information as they feel comfortable, however if risk to themselves or others has been disclosed then confidentiality will need to be broken. If this occurs the limits of confidentiality will be discussed again and where possible the next steps of the process will be collaborative, demonstrated by the researcher maintaining a friendly and supportive manner. The researcher will attempt to assess the level of risk and urgency present. Initially, an appropriate member of the research team will be consulted to determine the course of action, which might include sign-posting to another service or getting other services, like social services or the [REDACTED] involved. If contact cannot be made with a member of the research team and the risk to the participant is high then other external services may need to be contacted. This process is designed to identify, assess and manage risk and at no point will the researcher be able to offer therapy.

Participants are not expected to find the interview emotionally or psychologically difficult. It will be noted before the start of the interview that if participants become distressed, interviewees will be consulted as to how they would like to proceed i.e. continue, take a break or terminate the interview. The voice recorder will be switched off during breaks and interviewees will be notified when this happens.

Any hard copies of personal information that is collected at the interview (e.g. consent forms) will be stored in designated lockable containers at Lancaster University. Any electronic data, including audio recordings and transcripts will be stored on a password protected computer or laptop. The researcher's laptop will also have an encrypted drive that all files will be stored in. A copy of these will also be kept on the University's network drive which is on Lancaster University's virtual private network and is password protected.

Participants will nominate a pseudonym or 'alternative' name, that can be used on the transcripts and on any quotations in the write-up, to keep their information private. Once a transcript is anonymised the research and field supervisors will be able to view the transcripts to aid analysis.

Participants will have received information on the study prior to the interview but the interviewer will repeat the information documented on the Participant Information Sheet and Consent Form to gain informed written consent. Capacity to consent will need to be considered before the interview and will be generally assessed by the interviewer by going through the Participant Information Sheet and Consent Form.

The researcher will endeavour to keep all processes and decisions transparent to the participant and explain all aspects of the study until the interviewee is satisfied. Their transcript can also be removed from the analysis for up to two weeks after the interview, without giving a reason. This time is provided to allow the participant time to reflect on the interview, whilst providing a timeline that will support the researcher in continuing the analysis.

Practical issues

Recruitment of participants will require information packs to be sent out and other forms of promotion, like posters, will

need to be produced. Costs for essential expenses include, but are not limited to: paper, printing, photocopying, postage and phone calls which will be covered by Lancaster University under their research resources for trainees' policy.

Interviews will be sited at participants' homes or in local family based services. This will help to ensure anonymity, as the participants could be attending another appointment. This will also minimise risk as there will be other individuals on site that the researcher or interviewee can contact in an emergency e.g. if a participant collapses. The researcher has considered the Lancashire Care Foundation Trust Lone Worker Procedure and has developed a lone working system in accordance with this documentation. The researcher will use the buddy system to keep track of the interviewer and their expected movements. If support should be required then the nominated buddy can be contacted. An email with relevant details will be sent prior to meeting the interviewee, so that the buddy can access all the information they need, should there be a problem. If the interviewer does not check in with the buddy at the given time then the buddy will initially attempt to contact the researcher and if there is no response or returned call within an hour and a half they will call the police. A set phrase will be agreed upon between the buddy and the interviewer to denote an inability to communicate risk, threat or harm due to fear.

Participants will be given the opportunity to receive notification of any dissemination of the study e.g. organise individual meetings.

3. PURPOSE AND DESIGN OF THE RESEARCH

A7. Select the appropriate methodology description for this research. Please tick all that apply:

- Case series/ case note review
- Case control
- Cohort observation
- Controlled trial without randomisation
- Cross-sectional study
- Database analysis
- Epidemiology
- Feasibility/ pilot study
- Laboratory study
- Metanalysis
- Qualitative research
- Questionnaire, interview or observation study
- Randomised controlled trial
- Other (please specify)

A10. What is the principal research question/objective? Please put this in language comprehensible to a lay person.

What are the adjustment narratives of parents with a child who has Duchenne Muscular Dystrophy?

A11. What are the secondary research questions/objectives if applicable? Please put this in language comprehensible to a lay person.

- 1: To use parental narratives to understand parents' interpretations of their experience and consider the issues that they face.
- 2: The prevailing models of parental adjustment have been generic to chronic illness and were developed over 10 years ago (Wallender, 1998). This study would aim to consider parental narratives, their content and structure in comparison to current literature and ascertain the similarities and differences to this model, in order to add to the adjustment literature.
- 3: Narrative research is becoming more influential in developing clinical practice (Overcash, 2003). This study will use parental experience to consider how services could be improved whilst highlighting what is important to parents.
- 4: Duchenne Muscular Dystrophy is a severe and fatal disease, which compared to other chronic illnesses, like Leukaemia or Cystic Fibrosis, is relatively unknown. Therefore this study aims to help raise awareness/strengthen the

voices of the families that this disease affects.

A12. What is the scientific justification for the research? *Please put this in language comprehensible to a lay person.*

Research has shown that parents often suffer from severe psychological distress related to their experience of having with a child with a chronic paediatric illness. This highlights the importance of investigating experiences of adjustment, both good and bad, as a way of ascertaining the subjective needs and coping strategies of parents. This would provide information that professionals might find helpful when developing good practice within services when working with parents with children who have chronic illness.

There are generic models of adjustment but these are at least ten years old and therefore do not account for the changes in treatment and society which may influence parental experience and their interpretation of these experiences. The models are also based on quantitative research which has highlighted isolated factors involved in the process of adjustment. Reviews have suggested that qualitative research needs to be undertaken to understand the broader picture and the role that the factors have in parental experience. This has occurred with many chronic illnesses e.g. paediatric cancer, however there is very little focussing on Duchenne Muscular Dystrophy (DMD). Unlike other high profile diseases, DMD is relatively unknown by the general public and is comparatively less well supported by services. This may have an impact on parental experiences and the meaning they have for parents.

A qualitative approach like narrative analysis allows parents to express their stories of adjustment and give them the freedom to discuss what has been important and meaningful to them. The stories may include issues raised by previous research but will provide context and meaning that has been lacking. It will also provide an opportunity for parents to discuss how they have coped. Identifying these narratives will add to the knowledge base of parental adjustment to paediatric chronic illness generally but also consider how this applies to DMD specifically. This research will therefore help to raise awareness for this disease, whilst utilising parents' expertise to consider their needs and how best to support them.

A13. Please summarise your design and methodology. *It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.*

The study's research question required a qualitative approach to be taken. A narrative analysis was deemed the most appropriate analysis to answer the research question, whilst being appropriate for the size and scope of the study. A narrative analysis will allow the detail and subjectivity of the interviewees' stories to be maintained whilst analysing the content and structure of the interview. This will help to analyse parents' understanding of their experiences, which will be useful for professionals.

Up to 12 long-term parents/guardians will be recruited and must fit the inclusion/exclusion criteria. Other factors, including proximity, will be considered when arranging interviews.

Participants will potentially become aware of the study in 1 of 5 ways in two stages: 1) Through posters, an advert on the Contact a Parent forum or as part of an information pack posted to parents who have been referred to local paediatric services. Specialist nurses within the paediatric services will also discuss the study with prospective parents in their daily meetings and give them an information pack.

2) If possible through a presentation to local support groups or leaving information packs at local children's hospices. Parents/guardians will need to opt in to the study by contacting the chief investigator directly on the details given on the Participant Information Sheet, on the poster or the advert. If they respond to a poster then the Participant Information Sheet will need to be posted to them before they participate.

Once an enquiry has been made by a parent they will be asked to notify the chief investigator within 1 week as to whether they would like take part. It is at this stage that the inclusion and exclusion criteria will be checked. If they are happy to proceed then a convenient time and place will be arranged to meet.

The purpose of the meeting is for the chief investigator to conduct a face-to-face, semi-structured interview to discuss parents'/guardians' experiences and stories of adjustment.

At the beginning of the interview there will be general introductions and the interviewer will go through the participant information sheet again to make sure that the participant understands all aspects of the study. The interviewer will then go through the consent form. The participant will have their right to withdraw and the principle of confidentiality explained to them at this point. The specifics of confidentiality and the process of breaking confidentiality due to risk and danger will be discussed with the participant before they are asked to consent.

The participant will be able to ask questions at any point but they will be specifically given the opportunity to do this after the participant information sheet and consent form has been explained. They will only be asked to give consent when they are happy they understand and want to continue.

After the interview the carer will be asked if they have any questions and whether they want to be contacted to find out about the findings. If they say yes, they will be called within 4 months of their interview to arrange a way to give this information.

Timetable:

November 2013: apply for ethical approval.

November 2013- December: Once ethics has been approved then start recruitment.

November 2013– February 2014: conduct interviews, transcription and coding.

May 2014: Submission of final report

September 2014: Submit paper for publication

A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?

- Design of the research
- Management of the research
- Undertaking the research
- Analysis of results
- Dissemination of findings
- None of the above

Give details of involvement, or if none please justify the absence of involvement.

Participants will have the opportunity to review their re-storied accounts to promote rigor and trustworthiness.

4. RISKS AND ETHICAL ISSUES

RESEARCH PARTICIPANTS

A15. What is the sample group or cohort to be studied in this research?

Select all that apply:

- Blood
- Cancer
- Cardiovascular
- Congenital Disorders
- Dementias and Neurodegenerative Diseases
- Diabetes
- Ear
- Eye
- Generic Health Relevance
- Infection
- Inflammatory and Immune System
- Injuries and Accidents
- Mental Health
- Metabolic and Endocrine
- Musculoskeletal
- Neurological
- Oral and Gastrointestinal
- Paediatrics
- Renal and Urogenital
- Reproductive Health and Childbirth
- Respiratory
- Skin

<input type="checkbox"/> Stroke	
Gender:	Male and female participants
Lower age limit: 18	Years
Upper age limit:	No upper age limit

A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

Individuals eligible to participate in this study must be parents or long-term guardians of children with a diagnosis of Duchenne Muscular Dystrophy. If the participant is a long-term guardian then they must have cared for the child prior to the displaying of symptoms and/or a diagnosis.

In order to focus on adjustment and not specifically diagnosis or grief approximately two years must have passed since diagnosis and they must be currently caring for the child. Parents must speak English as a first language.

A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

The child must not have co-morbidities that are unassociated with DMD, e.g. other chronic health illnesses. Expected co-morbidities will not be excluded, examples include: respiratory difficulties/night time ventilation; cardiac problems; scoliosis, ADHD and requirements for spinal surgery.

To keep the sample reasonably homogeneous parents with their own chronic needs that may distinctly affect their perspective cannot be included, for example, but not limited to: those who have had a heart transplant or have a learning disability.

Families where there are current safeguarding concerns, i.e. are under investigation will also not be included.

RESEARCH PROCEDURES, RISKS AND BENEFITS

A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. These include seeking consent, interviews, non-clinical observations and use of questionnaires.

Please complete the columns for each intervention/procedure as follows:

1. Total number of interventions/procedures to be received by each participant as part of the research protocol.
2. If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
3. Average time taken per intervention/procedure (minutes, hours or days)
4. Details of who will conduct the intervention/procedure, and where it will take place.

Intervention or procedure	1	2	3	4
Recruitment	3	0	10	Participants will be recruited through posters, an information mail out to service users and an online advert on Contact a Parent's forum. The specialist nurses will also discuss the study with parents they meet during their clinics/meetings and give them an information pack so they can contact the chief investigator.
Giving information	1	0	5	The chief investigator will hand out and explain the Participant Information Sheet just before the interview.
Seeking consent	1	0	5	The chief investigator will explain the Consent Form and seek informed written consent from the participant just before the interview starts.
Interview	1	0	90	The chief investigator will conduct the interview.
Post interview questions	1	0	10	The chief investigator will receive post interview questions.
Feedback	1	0	30	The chief investigator will contact the interviewees to ask if they would like to receive a summary of the study.
Review of participant's	1	0	30	The chief investigator will contact the interviewee to discuss their re-storied account.

account

A21. How long do you expect each participant to be in the study in total?

Each participant will be involved in an interview (up to 1.5 hours) and a review of their storied account (up to 1 hour) as part of the analysis. These two meetings could be a few months apart.

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

There are no expected risks for taking part in this study. Participants are not expected to find the interview emotionally or psychologically difficult. Participants will be notified that they do not have to share information that makes them uncomfortable or upset, however, it will be noted before the start of the interview that if participants become distressed, interviewees will be consulted as to how they would like to proceed i.e. continue, take a break or terminate the interview. The voice recorder will be switched off during breaks and interviewees will be notified when this happens. If there is no perceived risk but the interviewee is distressed then they will be recommended to seek advice from their GP or other services that may already be involved for further support.

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

Yes No

If Yes, please give details of procedures in place to deal with these issues:

Confidentiality is documented in the Participant Information Sheet and will be checked on the Consent Form.

Parents/guardians will be reassured that they can share as much information as they feel comfortable, however if risk to themselves or others has been disclosed then confidentiality will need to be broken.

If this occurs participants will be reminded of the discussion that took place in the introduction and consent process regarding confidentiality. The researcher will continue to discuss the need to break confidentiality and how this applies to their situation. The researcher will aim to make this process collaborative and where possible get the participant to agree with the need to break confidentiality. If they do not agree then the researcher will reiterate their obligation to share this information and what the likely next steps will be. The researcher will initially attempt to contact an appropriate member of the research team to determine the course of action. If appropriate the researcher will aim to do this with the interviewee present so that they are involved in the process. The outcome of this conversation may result in actions like, sign-posting to another service or recommending they seek advice from their GP. If the participant is no longer present then this information will be fed back to them. However, if the level of risk is high and contact cannot be made with a member of the research team the researcher will endeavour to keep the participant calm and in the interview room whilst contacting more immediate services, like other agencies involved or the police. This process is designed to identify, assess and manage risk and at no point will the researcher be able to offer therapy.

If participants become distressed but have no or low risk then they will be advised to speak with their GP or other agencies involved as appropriate.

The interviewer will also seek supervision from an appropriate member of the research team. This will not require disclosure of identifiable information of the participant but will be a process to support the interviewer should they have any emotional reactions to the interviews. If through this process risk is identified later then the interviewer will contact the interviewee to further discuss the problem and follow the risk process documented previously.

A24. What is the potential for benefit to research participants?

There is no direct benefit for participants. Their participation may provide information that professionals and services find useful when improving their practice to target the needs of parents and guardians of children with Duchennes Muscular Dystrophy.

A26. What are the potential risks for the researchers themselves? (if any)

There should not be any potential risks for the researcher. The nature of the sample i.e. a healthy participant sample, suggest that the participants should not pose a threat to the researcher. The location of the interviews will also provide added support should there be a problem, with individuals contactable in an emergency e.g. if the participant

collapses. The researcher will also follow the lone working protocol as documented by the Lancashire Care NHS Foundation Trust and the Central Manchester University Hospitals Foundation Trust, by implementing a buddy system for each interview. The researcher will also attend regular supervision to manage any practical and emotional difficulties.

RECRUITMENT AND INFORMED CONSENT

In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? For example, identification may involve a disease register, computerised search of GP records, or review of medical records. Indicate whether this will be done by the direct healthcare team or by researchers acting under arrangements with the responsible care organisation(s).

Participants will be opportunistically sampled and identification of participants will take 6 forms in a staged approach, based on an opt in recruitment process:

Stage 1

- 1: Online – via a Contact a Parent discussion board.
- 2: Posters based at paediatric services involved with the research and other local community services (GP practices, libraries etc).
- 3: Via an opt-in mail out by local paediatric services based on parents who have been referred to the service. A member of the administration team, who is not involved in the research, will mail out packs of information to parents/guardians who have been referred to the service and have a child with Duchenne Muscular Dystrophy. The parent will need to contact the researcher on the contact details provided to let them know they wish to participate and therefore the administration team will not know who has agreed to participate and the researcher will not have any personal details until the interviewee gives them.

Recruitment will be reviewed after two months of interviewing. If there is a problem then stage 2 of recruitment will be initiated.

Stage 2

- 4: Via an opt-in mail out by another local paediatric service based on parents who have been referred to the service, following the same mail out procedure as above.
- 5: Contact support groups (chat/present ideas).
6. Contact children's hospices e.g. [REDACTED] and leave information packs with staff.

In all instances contact details for the researcher will be left and it will be up to the participants to contact the researcher directly.

Should parents/guardians from these recruitment paths contact the researcher and wish to find out more about the study a participant information sheet can be emailed or posted, as they choose. Participants will be asked if further contact to make arrangements can be done by phone to provide the maximum opportunity for participants to attend an interview slot. If they refuse then the researcher will use the preferred method of contact.

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

Yes No

Please give details below:

The paediatric services have minimal databases used to log referrals of parents and guardians using the service. This has a small amount of personal information about the parent/guardian, including addresses. A member of the administration team, who is not involved in the research, will mail out packs of information to parents/guardians who have been referred and appear to fit the inclusion/exclusion criteria. Initially, the mail out will be confined to those who were referred between (April 2011 to April 2012), of which there are approximately (65). This time-scale will be reviewed and extended if there are problems recruiting. The parent will then need to contact the chief investigator on the contact details provided to let them know they wish to participate and therefore the administration team will not know who has agreed to participate.

A27-3. Describe what measures will be taken to ensure there is no breach of any duty of confidentiality owed to patients, service users or any other person in the process of identifying potential participants. *Indicate what steps have been or will be taken to inform patients and service users of the potential use of their records for this purpose. Describe the arrangements to ensure that the wishes of patients and service users regarding access to their records are respected. Please consult the guidance notes on this topic.*

All recruitment will be based on an opt in process where the carers will have to initially contact the researcher to access further information or arrange an interview.

Only individuals who are part of the paediatric services administrative team will know who has had information sent out to them and participants will have to contact the researcher as above. Therefore the research team will not have access to participant's information until the parent/guardian provides it and the administration team will not know who has agreed to participate.

Carers will be offered a few locations to be seen in the community and, if necessary, at home. This will help them to feel comfortable and maximise confidentiality and anonymity.

A27-4. Will researchers or individuals other than the direct care team have access to identifiable personal information of any potential participants?

Yes No

A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?

Yes No

If Yes, please give details of how and where publicity will be conducted, and enclose copy of all advertising material (with version numbers and dates).

Yes. Posters will be put up in local family based services, including but not limited to paediatric services base, local sure start centres and GP practices.

Parents/guardians will also be contacted via an online advert on the Contact a Parent discussion board.

Support groups may also be contacted to arrange a brief presentation summarising the research and leaving information packs and contact details for parents/guardians to contact the researcher.

A29. How and by whom will potential participants first be approached?

A member of the paediatric administration team or the principal researcher will be responsible for approaching participants. This will depend on the method of recruitment.

The paediatric administration team, who will have already had contact with the parent/guardian, will be responsible for the information mail out to parents/guardians selected from the database.

Specialist nurses will also discuss the study during the meetings and provide any families that are interested with an information pack.

The researcher may approach carers in an informal group setting by presenting to local support groups and leaving information for the carers.

All forms of recruitment will function on an opt-in basis, where the foster carer must contact the researcher if they wish to participate.

A30-1. Will you obtain informed consent from or on behalf of research participants?

Yes No

If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

Parents/guardians will be required to give consent to participate at the beginning of the interview during the introduction when they will be talked through the participant information sheet with the researcher. This will be documented on the consent form. If capacity to consent is viewed as a potential problem, then the risk protocol will be followed.

If you are not obtaining consent, please explain why not.

Please enclose a copy of the information sheet(s) and consent form(s).

A30-2. Will you record informed consent (or advice from consultees) in writing?

Yes No

A31. How long will you allow potential participants to decide whether or not to take part?

Once the recruitment period starts after ethical approval participants will be able to contact the researcher and ask questions. However once participants have made an initial contact then the researcher will request that the potential participant notifies the researcher of their decision within a week, to allow for the organisation of the interview and give sufficient time for the participant to make the decision. The researcher will ask if the participant minds being contacted the day before the end of this time period to act as a reminder for the participant.

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs?(e.g. translation, use of interpreters)

Due to financial constraints the study will not be able to make arrangements for specific communication needs.

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? Tick one option only.

- The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.
- The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.
- The participant would continue to be included in the study.
- Not applicable – informed consent will not be sought from any participants in this research.
- Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

The length of the contact between the participant and the researcher is sufficiently brief (a maximum of an hour and a half) that this is highly unlikely. If however, the researcher has concerns regarding capacity at any point during the interview then this will potentially pose a risk issues and the researcher will follow the identified risk protocol.

If you plan to retain and make further use of identifiable data/tissue following loss of capacity, you should inform participants about this when seeking their consent initially.

CONFIDENTIALITY

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

Storage and use of personal data during the study

A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)?(Tick as appropriate)

- Access to medical records by those outside the direct healthcare team
- Electronic transfer by magnetic or optical media, email or computer networks

- Sharing of personal data with other organisations
- Export of personal data outside the EEA
- Use of personal addresses, postcodes, faxes, emails or telephone numbers
- Publication of direct quotations from respondents
- Publication of data that might allow identification of individuals
- Use of audio/visual recording devices
- Storage of personal data on any of the following:
 - Manual files including X-rays
 - NHS computers
 - Home or other personal computers
 - University computers
 - Private company computers
 - Laptop computers

Further details:

University computers or the chief investigator's personal laptop will be used to store data, all of which will be password protected and encrypted. The data stored on the University computers will be saved on the Lancaster University secure network drive, which is located on the virtual private network and is only accessible to the researcher.

The digital recorder cannot be encrypted, consequently following the interview the digital recorder will be stored securely and the audio files will be transferred to a suitably secure computer as quickly soon as possible. After transfer the audio files will be deleted from the digital recorder.

Any paperwork that has personal information on will be stored in a lockable cabinet at Lancaster University.

A37. Please describe the physical security arrangements for storage of personal data during the study?

All paperwork with confidential information (i.e. the consent forms) will be stored in a lockable cabinet at Lancaster University.

A38. How will you ensure the confidentiality of personal data? Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.

All electronic data (i.e. audio recordings, not anonymised and anonymised transcripts) will either be stored on the Lancaster University secure network drive which is located on a virtual private network and is password protected, or it will be stored in an encrypted drive on the chief investigator's personal laptop, which is also password protected. All paperwork containing personal information will be stored in a lockable cabinet at Lancaster University.

Participants will also be asked to give a preferred pseudonym that will be utilised in the transcription and alongside quotes used in the final report and any publications.

Once the transcripts have been anonymised using the pseudonym and removing any potential identifiers the research supervisor will be able to view the transcripts to aid analysis. The field supervisor will only input into the analysis once the transcripts have been coded, thereby making sure that participants' identities are fully protected.

The opt-in recruitment process also means that the researcher will only have participant's personal information once it has been volunteered.

All procedures have been designed in line with the BPS code of ethics and conduct (2009) and the HPC guidance on conduct and ethics for students (2009).

A40. Who will have access to participants' personal data during the study? Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.

The chief investigator will have access to the participant's personal data. The designated 'Buddy' from the lone working protocol will also temporarily have access to this information during the interview to ensure the researcher's safety. This will be deleted once the safety of the researcher is established. The attachments containing the information will only be opened as per the agreed lone working protocol, and therefore no-one other than the researcher should have access to this information unless there is a perceived danger.

Storage and use of data after the end of the study

A41. Where will the data generated by the study be analysed and by whom?

The data will be transcribed and anonymised by the chief investigator. The anonymised transcripts will then be analysed by the chief investigator and the research supervisor. Analysis will take place at the chief investigator's home and on the Lancaster University campus, where there will be access to safe data storage.

A42. Who will have control of and act as the custodian for the data generated by the study?

	Title	Forename/Initials	Surname
	Ms	Antoinette	Deavin
Post	Trainee clinical psychologist		
Qualifications	Psychology BSc, Psychology MRes		
Work Address	C16 Furness College Lancaster University Lancaster, Lancashire		
Post Code	LA1 4YG		
Work Email	a.deavin@lancaster.ac.uk		
Work Telephone			
Fax			

A43. How long will personal data be stored or accessed after the study has ended?

- Less than 3 months
 3 – 6 months
 6 – 12 months
 12 months – 3 years
 Over 3 years

A44. For how long will you store research data generated by the study?

Years: 5
Months: 0

A45. Please give details of the long term arrangements for storage of research data after the study has ended. Say where data will be stored, who will have access and the arrangements to ensure security.

All personal data will be destroyed when the report is submitted to the University. All other data will be transferred from the chief investigator to Lancaster University when the main report is written. This will then be stored in a secure, locked cabinet at Lancaster University for 5 years.

INCENTIVES AND PAYMENTS

A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?

Yes No

A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?

Yes No

A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?

Yes No

NOTIFICATION OF OTHER PROFESSIONALS

A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?

Yes No

If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.

PUBLICATION AND DISSEMINATION

A50. Will the research be registered on a public database?

Yes No

Please give details, or justify if not registering the research.

No, however the research will be registered when the researcher pursues a publication.

Registration of research studies is encouraged wherever possible.

You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

A51. How do you intend to report and disseminate the results of the study? Tick as appropriate:

- Peer reviewed scientific journals
- Internal report
- Conference presentation
- Publication on website
- Other publication
- Submission to regulatory authorities
- Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- No plans to report or disseminate the results
- Other (please specify)

Provide a summary of the findings to the participants and paediatric services involved if they are interested. This could be a presentation or a brief summary of the write-up. This research will also be presented at the Lancaster University thesis presentation day open to peers and other allied professionals.

A52. If you will be using identifiable personal data, how will you ensure that anonymity will be maintained when publishing the results?

Pseudonyms will be used alongside participant quotes in the write up.

A53. Will you inform participants of the results?

Yes No

Please give details of how you will inform participants or justify if not doing so.

Participants will be asked whether they would like to receive feedback on the results. They will be offered the opportunity to do this one-to-one, over the phone or face-to-face.

5. Scientific and Statistical Review

A54. How has the scientific quality of the research been assessed? Tick as appropriate:

- Independent external review
- Review within a company
- Review within a multi-centre research group
- Review within the Chief Investigator's institution or host organisation
- Review within the research team
- Review by educational supervisor
- Other

Justify and describe the review process and outcome. If the review has been undertaken but not seen by the researcher, give details of the body which has undertaken the review:

A preliminary research proposal was presented by the researcher and this was peer reviewed at a meeting attended by trainee clinical psychologists and two academic tutors. At this meeting the researcher was required to justify the feasibility of their decisions on the design of the project. This feedback has been used to identify potential ethical, practical and theoretical problems and make modifications. The outcome of this meeting was then summarised and fed back to the two tutors at the meeting, with the suggested alterations that could be made.

The research protocol has then been draft read and discussed with the research and field supervisors. A research contract has been agreed with both the field and research supervisors, stating the level of input they are required to help support the chief investigator and also utilise their expertise.

All paperwork has been reviewed by the Research Support Office to check the quality of the study and subsequently granting indemnity.

For all studies except non-doctoral student research, please enclose a copy of any available scientific critique reports, together with any related correspondence.

For non-doctoral student research, please enclose a copy of the assessment from your educational supervisor/ institution.

A59. What is the sample size for the research? How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.

Total UK sample size: 12
Total international sample size (including UK): 12
Total in European Economic Area: 12

Further details:

A sample size of between 6-12 participants has been estimated for this study, but will need to be monitored and

reviewed throughout the analysis.

A60. How was the sample size decided upon? *If a formal sample size calculation was used, indicate how this was done, giving sufficient information to justify and reproduce the calculation.*

Up to 12 interviews/participants is sufficient for an in-depth narrative analysis.

A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.

A qualitative methodology, specifically narrative analysis, has been chosen to focus on the experiences and understanding of parents/guardians of children with Duchenne Muscular Dystrophy, whilst allowing the data to be inductively analysed to generate ecologically valid, subjective and findings (Elliott, Fischer & Rennie, 1999).

Interviews will be transcribed and analysed. Narratives will be identified within each transcript allowing the researcher to explore how parents/guardians understand their experience. Crossley's (2007) narrative analytic model will be used to guide the analysis and Braun and Clark's (2006) 15 point checklist will be considered to help maintain quality.

Currently it is expected that this will be done by hand but the researcher hopes to develop skills to utilise a transcription and coding programme called NVIVO, which should speed up the analysis and make the process of analysis more accessible.

The anonymised findings will be discussed within the research team to strengthen the quality of the analysis.

6. MANAGEMENT OF THE RESEARCH

A63. Other key investigators/collaborators. *Please include all grant co-applicants, protocol co-authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.*

	Title Forename/Initials Surname
	Dr Pete Greasley
Post	Teaching Fellow in Research Methods
Qualifications	BSc (Hons), PhD, PGCHEP
Employer	Lancashire Care NHS Foundation Trust
Work Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire
Post Code	LA1 4YG
Telephone	01524593535
Fax	
Mobile	
Work Email	p.greasley@lancaster.ac.uk

	Title Forename/Initials Surname
	Dr Clare Dixon
Post	Clinical Psychologist & Clinical Tutor
Qualifications	BSc (Hons); PhD; DClinPsy
Employer	Lancashire Care NHS Foundation Trust
Work Address	Clinical Psychology, Faculty of Health and Medicine, C16 Furness College, Lancaster University, Lancaster, Lancashire
Post Code	LA1 4YG
Telephone	01524593535

Fax
Mobile
Work Email c.dixon@lancaster.ac.uk

A64. Details of research sponsor(s)

A64-1. Sponsor

Lead Sponsor

Status: NHS or HSC care organisation

Commercial status:

Academic

Pharmaceutical industry

Medical device industry

Local Authority

Other social care provider (including voluntary sector or private organisation)

Other

If Other, please specify:

Contact person

Name of organisation Lancaster University

Given name Debbie

Family name Knight

Address Research Support Office, B Floor, University House, Lancaster University

Town/city Lancaster

Post code LA1 4YW

Country UNITED KINGDOM

Telephone 01524592605

Fax

E-mail ethics@lancaster.ac.uk

Is the sponsor based outside the UK?

Yes No

Under the Research Governance Framework for Health and Social Care, a sponsor outside the UK must appoint a legal representative established in the UK. Please consult the guidance notes.

A65. Has external funding for the research been secured?

Funding secured from one or more funders

External funding application to one or more funders in progress

No application for external funding will be made

What type of research project is this?

Standalone project

Project that is part of a programme grant

- Project that is part of a Centre grant
- Project that is part of a fellowship/ personal award/ research training award
- Other

Other – please state:

A66. Has responsibility for any specific research activities or procedures been delegated to a subcontractor (other than a co-sponsor listed in A64-1) ? Please give details of subcontractors if applicable.

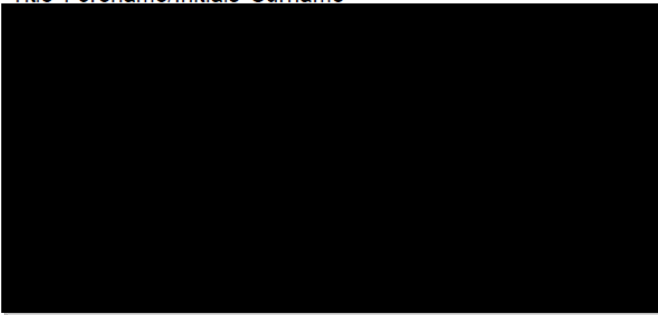
- Yes No

A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?

- Yes No

Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

A68-1. Give details of the lead NHS R&D contact for this research:

Title	Forename/Initials	Surname
Organisation		
Address		
Post Code		
Work Email		
Telephone		
Fax		
Mobile		

Details can be obtained from the NHS R&D Forum website: <http://www.rdforum.nhs.uk>

A69-1. How long do you expect the study to last in the UK?

Planned start date: 01/10/2013
Planned end date: 30/06/2014
Total duration:
Years: 0 Months: 8 Days: 30

A71-1. Is this study?

- Single centre
- Multicentre

A71-2. Where will the research take place? (Tick as appropriate)

- England

- Scotland
- Wales
- Northern Ireland
- Other countries in European Economic Area

Total UK sites in study 2

Does this trial involve countries outside the EU?

- Yes No

A72. What host organisations (NHS or other) in the UK will be responsible for the research sites? Please indicate the type of organisation by ticking the box and give approximate numbers of planned research sites:

- NHS organisations in England 2
- NHS organisations in Wales
- NHS organisations in Scotland
- HSC organisations in Northern Ireland
- GP practices in England
- GP practices in Wales
- GP practices in Scotland
- GP practices in Northern Ireland
- Social care organisations
- Phase 1 trial units
- Prison establishments
- Probation areas
- Independent hospitals
- Educational establishments
- Independent research units
- Other (give details)

Total UK sites in study: 2

A73-1. Will potential participants be identified through any organisations other than the research sites listed above?

- Yes No

A73-2. If yes, will any of these organisations be NHS organisations?

- Yes No

If yes, details should be given in Part C.

A73-3. Approximately how much time will these organisations expect to spend on screening records and/or provision of information to potential participants, and how will the costs of these activities be funded?

The recruitment from the service would require a small amount of time from a member of the team to identify potentially relevant individuals from the service's database and stick labels with their addresses onto pre-made information packs. This would be done in groups of 20 so that we could monitor the progress of recruitment and ensure that we do not over/under-recruit and therefore use time efficiently. For each batch of 20 addresses it will take approximately an hour.

A74. What arrangements are in place for monitoring and auditing the conduct of the research?

The conduct of the research will be the responsibility of the chief investigator, under supervision from the field and research supervisors. A research contract has been agreed to delegate tasks and agree timelines to ensure that all parties are aware of their roles and to keep the research on track. Supervisors will also advise on more technical aspects of the research and analysis to share expertise. The chief investigator will meet with the research supervisor at least once a month, with email and phone contacts as necessary. The field supervisor will see the chief investigator three or four times minimum over the course of the study and will be available by email and phone as necessary.

A76. Insurance/ indemnity to meet potential legal liabilities

Note: in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland

A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? Please tick box(es) as applicable.

Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (NHS sponsors only)
 Other insurance or indemnity arrangements will apply (give details below)

Lancaster University legal liability cover will apply.

Please enclose a copy of relevant documents.

A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the sponsor(s) or employer(s) for harm to participants arising from the design of the research? Please tick box(es) as applicable.

Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (protocol authors with NHS contracts only)
 Other insurance or indemnity arrangements will apply (give details below)

Lancaster University legal liability cover will apply.

Please enclose a copy of relevant documents.

A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?

Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.

- NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)
 Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

This is covered by standard NHS Indemnity.



Please enclose a copy of relevant documents.

A78. Could the research lead to the development of a new product/process or the generation of intellectual property?

Yes No Not sure

PART C: Overview of research sites

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. For NHS sites, the host organisation is the Trust or Health Board. Where the research site is a primary care site, e.g. GP practice, please insert the host organisation (PCT or Health Board) in the Institution row and insert the research site (e.g. GP practice) in the Department row.

Research site		Investigator/ Collaborator/ Contact	
Institution name		Title	
Department name			
Street address			
Town/city			
Post Code			
Participant Identification Centre(PIC)-Collaborator/ Contact			

PART D: Declarations

D1. Declaration by Chief Investigator

1. The information in this form is accurate to the best of my knowledge and belief and I take full responsibility for it.
2. I undertake to abide by the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research.
3. If the research is approved I undertake to adhere to the study protocol, the terms of the full application as approved and any conditions set out by review bodies in giving approval.
4. I undertake to notify review bodies of substantial amendments to the protocol or the terms of the approved application, and to seek a favourable opinion from the main REC before implementing the amendment.
5. I undertake to submit annual progress reports setting out the progress of the research, as required by review bodies.
6. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to security and confidentiality of patient or other personal data, including the need to register when necessary with the appropriate Data Protection Officer. I understand that I am not permitted to disclose identifiable data to third parties unless the disclosure has the consent of the data subject or, in the case of patient data in England and Wales, the disclosure is covered by the terms of an approval under Section 251 of the NHS Act 2006.
7. I understand that research records/data may be subject to inspection by review bodies for audit purposes if required.
8. I understand that any personal data in this application will be held by review bodies and their operational managers and that this will be managed according to the principles established in the Data Protection Act 1998.
9. I understand that the information contained in this application, any supporting documentation and all correspondence with review bodies or their operational managers relating to the application:
 - Will be held by the REC (where applicable) until at least 3 years after the end of the study; and by NHS R&D offices (where the research requires NHS management permission) in accordance with the NHS Code of Practice on Records Management.
 - May be disclosed to the operational managers of review bodies, or the appointing authority for the REC (where applicable), in order to check that the application has been processed correctly or to investigate any complaint.
 - May be seen by auditors appointed to undertake accreditation of RECs (where applicable).
 - Will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.
 - May be sent by email to REC members.
10. I understand that information relating to this research, including the contact details on this application, may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 1998.
11. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named below. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Contact point for publication*(Not applicable for R&D Forms)*

NRES would like to include a contact point with the published summary of the study for those wishing to seek further information. We would be grateful if you would indicate one of the contact points below.

- Chief Investigator
- Sponsor

NHS R&D Form

IRAS Version 3.5

- Study co-ordinator
- Student
- Other – please give details
- None

Access to application for training purposes (Not applicable for R&D Forms)*Optional – please tick as appropriate:*

I would be content for members of other RECs to have access to the information in the application in confidence for training purposes. All personal identifiers and references to sponsors, funders and research units would be removed.

Signature:

Toni Deavin

Print Name:

ANTOINETTE DEAVIN

Date: 02.12.2013

(dd/mm/yyyy)

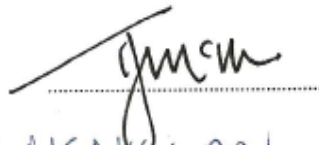
D2. Declaration by the sponsor's representative

If there is more than one sponsor, this declaration should be signed on behalf of the co-sponsors by a representative of the lead sponsor named at A64-1.

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements, as described in question A76, will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.
6. The duties of sponsors set out in the Research Governance Framework for Health and Social Care will be undertaken in relation to this research.
7. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Signature:



Print Name:

T MCNEILAN

Post:

PRO VC RESEARCH

Organisation:

MANCHESTER UNIVERSITY

Date:


28.11.13

(dd/mm/yyyy)

D3. Declaration for student projects by academic supervisor(s)

1. I have read and approved both the research proposal and this application. I am satisfied that the scientific content of the research is satisfactory for an educational qualification at this level.
2. I undertake to fulfil the responsibilities of the supervisor for this study as set out in the Research Governance Framework for Health and Social Care.
3. I take responsibility for ensuring that this study is conducted in accordance with the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research, in conjunction with clinical supervisors as appropriate.
4. I take responsibility for ensuring that the applicant is up to date and complies with the requirements of the law and relevant guidelines relating to security and confidentiality of patient and other personal data, in conjunction with clinical supervisors as appropriate.

Academic supervisor 1

Signature: .....

Print Name: **PETE GRAYLEY**

Post: **TEACHING FELLOW**

Organisation: **LANCASTER UNIVERSITY**

Date: **02/12/2013** (dd/mm/yyyy)

Research Protocol: Version 2

Title: Parents' stories of adjustment to their child's chronic illness: a narrative approach to
Duchenne Muscular Dystrophy.

Chief researcher: Antoinette Deavin

Field supervisor: Dr Clare Dixon

Research supervisor: Dr Pete Greasley,
Lancaster University

Introduction to Duchenne Muscular Dystrophy

Duchenne Muscular Dystrophy (DMD) is a genetic, chronic and progressive disorder that affects approximately 1 in 3,600 boys in the UK. It results in muscle degeneration, the onset of which can be seen in early infancy. The muscle degeneration along with the associated abnormal bone development and increase of fat and fibrotic tissue result in: low endurance, difficulties standing and walking, loss of movement, paralysis, and possibly intellectual impairment. DMD is a life limiting condition which means that individuals have a shortened life expectancy. Currently the average life expectancy of an individual with DMD is their 20s, although in some cases changes in treatment have made it possible for people to live into their 30s (Rinaldi, Mayer, & Dichiario, 2013). Therefore a diagnosis of DMD forewarns distressing physical changes for the child, which may also lead to psychological distress. Other challenging factors associated with DMD may include: approximately one in three children with DMD has a severe learning disability, whilst another one in three has a mild learning disability and behaviour problems are also frequent. Understandably a diagnosis of DMD and the subsequent changes and challenges that occur will have an impact not only on the child but their family. Parents in particular are expected to manage the health, emotional and psychological needs of the child with DMD, whilst looking after any other family members, in addition to their own needs. Qualitative research into parents of children with cancer has demonstrated that meeting these needs often results in parents re-evaluating the meaning and purpose of life and redefining themselves, as their experience contrasts with their expectations, which understandably requires a level of adjustment (Schweitzer, Griffiths, & Yates, 2012).

Research into Parental Adjustment to Paediatric Chronic Illness

Parental adjustment has been looked at in chronic illness generally (Wallander & Varni, 1998) and studies have shown that parents often suffer psychological distress that meets

diagnostic criteria (Wallander, 1993). A selective narrative review of the models of adjustment by Wallander and Varni (1998) recognises that whilst parental adjustment to their child's chronic illness is a serious problem they also emphasise variation in this population and ability for good adjustment. However, they predominantly reviewed quantitative studies, with little reference to parental personal experience, which meant they derived 'categories' that are pertinent to parental adjustment. The categories they felt were most important included parental psychosocial stress, hope, perceived competence in managing chronic illness and the extent to which the disease restricted the parent's ability to pursue their own interests. These all relate to parents appraisals of experience, which influenced how they felt and subsequently how they managed to cope and adjust. This suggests a complex and personal interpretation of experiences shape parental adjustment. Given the subjectivity of the process a quantitative approach, as promoted by Wallander and Varni (1998), does not account for the meaning that parents create from their experiences and how this relates to adjustment.

Qualitative Research into Parental Experiences, Adjustment and DMD

The need for more qualitative research in adjustment to paediatric chronic illness was recognised by Wallander and Varni (1998). Their recommendation has been acknowledged for many childhood chronic diseases and there is now qualitative research available for parental experiences in paediatric cancer (Schweitzer et al., 2012), kidney disease (Tong, Lowe, Sainsbury, & Craig, 2008) and epilepsy (Mu, 2008).

However there is a scarcity of papers that focus on the parental experiences of DMD generally. The most recent identified piece of research based on parental experience of DMD was by Samson, Tomiak, Dimillo, Lavigne, Miles, Choquette, Chakraborty and Jacob's (2009) paper on parental hope of which adjustment was identified as a main theme. The identification of this theme demonstrates its importance and consequently supports further

research into adjustment. However, there is very little research on adjustment to DMD and, as noted above, it tends to be quantitative, which identifies categories without understanding their meaning and significance for parents. Other qualitative publications that focus specifically on family adjustment to DMD include work by Poysky and Kinnett (2009). Whilst their report is not research based, the documentation of their international workshop with parents and professionals begins to take a qualitative approach to supporting people involved with children with DMD. The inclusion of parents in this discussion highlights the need to consider parental perspectives to adjustment to help inform service provision and improve care.

The absence of literature on DMD generally and parental experience more specifically, may be due to its comparatively low prevalence in the general population, which could have minimised its research status. Although, as a severe and life limiting condition it is surprising that there is little emphasis placed on understanding the subjective experiences of the families who are affected by DMD. This is demonstrated by the lack of qualitative research in this field and consequently little is known about parental experiences. There is none that allow parents to tell their story of how their understanding of events and experiences has shaped their adjustment. Consequently this study will enable parents to have a voice and tell their story. To accomplish this narrative analysis will be used to explore parental narratives to ascertain how parents make sense of their experience and how this relates to adjusting to their child having DMD.

Research question and aims

Research question: What are the adjustment narratives of parents with a child who has Duchenne Muscular Dystrophy?

Specific aims:

1. As an exploratory study I aim to get an in-depth understanding of parental experiences and their meaning through the content and construction of their narratives.
2. The results will be compared with the prevailing models of parental adjustment which relate to parental adjustment to paediatric chronic illnesses generally and are over 10 years old (Wallander & Varni, 1998) to see how meaning making may influence the factors that have already been suggested in research i.e. stress, hope, perceived competence in managing chronic illness and the ability for parents to pursue their own interests.
3. Specific differences across illnesses e.g. disease symptoms, level of support, prevalence, perceptions of society and immediate peers, funding for new treatments etc, may affect parents/guardians appraisals of their experience and therefore adjustment. Where disease specific qualitative research has already been completed this study can compare similarities and differences.
4. Narrative research is becoming more influential in developing clinical practice (Overcash, 2003). This study will use parental interpretations of their experience and how this relates to adjustment to consider how services could be improved whilst highlighting what is important to parents.

Method

Participants

The study will recruit between 6-12 parents or long-term carers of children with a diagnosis of Duchenne Muscular Dystrophy to provide an appropriate range of in-depth narrative accounts (Howitt, 2010). As an exploratory study a range of experiences will be accepted to provide a base for future research, although participants will not be recruited on specific aspects of their experience. In this way the study will benefit from emphasis on the different parts of the story that parents feel are appropriate to their experience of adjustment. Participants can be recruited as individuals or as a couple, although they will be recommended to attend individual interviews as this will allow more time to tell their story, without other influences that may alter their perspective and meaning of the story. All participants will need to fit the inclusion and exclusion criteria.

Inclusion criteria

Individuals eligible to participate in this study must be parents or long-term guardians of children with a diagnosis of Duchenne Muscular Dystrophy. If the participant is a long-term guardian then they must have cared for the child prior to the displaying of symptoms and/or a diagnosis.

In order to focus on longer-term adjustment, and not specifically diagnosis or grief, approximately two years must have passed since diagnosis and participants must still be caring for the child. Participants must also have English as their primary language.

Exclusion criteria

The child must not have co-morbidities that are unassociated with DMD, e.g. other chronic health illnesses. Expected co-morbidities will not be excluded, examples include: respiratory difficulties/night time ventilation; cardiac problems; scoliosis, requirements for spinal surgery, learning disability and attention deficit (hyperactivity) disorder. To keep the sample

reasonably homogeneous parents with their own chronic needs that may distinctly affect their perspective cannot be included, for example, but not limited to: those who have had a heart transplant or have a learning disability.

Families where there are current safeguarding concerns i.e. are under investigation will also not be included.

Design

A qualitative approach has been chosen to best address the research question, specifically exploring the experiences of 6-12 parents and/or long-term carers. To achieve this narrative analysis will be used. Narrative analysis stems from the concept that the meaning that individuals make of their lives and themselves is storied (McAdams, 2008). McAdams (2008) suggests that individuals will have many stories regarding their lives but each integrates subjectively important 'scenes' based on their experience. Narratives will also reflect the culture and beliefs of the individual and will be constructed accordingly. These narratives may also then identify changes over time. Accessing and analysing these stories will help to develop an understanding of the subjective influences on parents and how they make sense of them in relation to adjustment.

Materials

See Appendix C for an example interview schedule.

The interviews will be recorded using a digital voice recorder.

Procedure

Recruitment strategy

Parents of children with DMD have many daily commitments, often related to the specific health needs of their child e.g. hospital appointments. The pressure placed on their time may mean that parents feel unable able to attend an interview. Therefore it is necessary to consider how to broaden the sample pool and how to support parents to attend.

Multiple recruitment strategies have been devised to ensure that enough parents attend to complete the analysis. In order to manage the recruitment process a two staged approach has been developed which will allow a review of recruitment. The review will enable the chief investigator to assess the number of participants recruited. If the review demonstrates that the sample is too small then the second stage of recruitment will be instigated.

Stage 1

- 1: A mail out will be sent by a member of the [REDACTED] team to parents who have been referred to the service. The service will be given information packs which will include the Covering Letter (see Appendix D) discussing why the parents have been approached and the Participant Information Sheet (see Appendix A) with the details of the study and how to contact the chief investigator if they are interested, including a stamped addressed envelope. The service will then initially send out 20 packs at random to eligible parents. Three weeks after the initial mail out recruitment will be re-assessed and another 20 packs can be sent out if needed. There are approximately 60-100 potential participants within the service and therefore this procedure can be repeated up to five times, however given the time constraints it will be re-assessed weekly after the second mail out. This process will also ensure that confidentiality has been maintained as the research team will not know who has been contacted and the administration staff will not know who has responded.
- 2: The neuromuscular nurses will be given information packs to hand out to parents during their daily meetings e.g. clinics. The nurses will then describe the study and how to contact the chief investigator but will not recruit directly.
- 3: Posters (see Appendix E) will be put up in local community services (GP practices, libraries etc).

4: An advert will be posted online on the Contact a Parent parental discussion boards/forums (<http://www.makingcontact.org/forums/>). There are six forums that would be appropriate to post an advert i.e. medical issues, behaviour, working, growing up and transition, share your day, and consultations, questionnaires and events. The advert will also be posted on the Contact a Family facebook page. I have spoken with the charity and they have approved this dependent on ethical approval from the NHS REC for the study. The advert will be a modified version of the poster (see Appendix G). Currently the site has a total of approximately 21 parents located in the North of England or with the specific location not given: Cheshire East x 1, 'England' x 7, Lancashire x 1, Location not given x 8, Staffordshire x 2 and Yorkshire x 2.

Stage 2 (dependent on weekly reviews to assess whether further recruitment is needed)

5: If required a mail out from the [REDACTED] can be organised, utilising the same process as documented above for the [REDACTED].

6: Approach local support groups allied to charities (e.g. Muscular Dystrophy Campaign Muscle Groups) about attending a session to present the research. This will give parents an opportunity to discuss the research and take an information pack if they are interested.

7: Approach local children's hospices e.g. [REDACTED] regarding putting up posters and leaving information packs at the sites.

Participants will be opportunistically sampled but the decision to be included in the study will be considered in relation to a number of factors, including: meeting the inclusion and exclusion criteria, location of residence, and number of participants already recruited etc. However recruitment has been designed to work on an opt-in basis, with carers being

required to contact the chief researcher directly on the contact details given to express their interest. Consequently the chief researcher will only gain access to participants' personal information when the potential participant gives it and the services involved will not know who has volunteered, maintaining the participant's anonymity.

When parents have made an enquiry of interest but an interview has not been scheduled, participants will be asked to notify the researcher within 1 week from the first contact to discuss whether they would like to participate and subsequently arrange a meeting. The researcher will ask if they would like to be contacted at the end of the week's period to remind them and all communication thereafter will be using the participant's preferred method i.e. by phone or email.

All participants are asked to answer brief questions relating to the inclusion/exclusion criteria when they express their interest (see Appendix A). These questions will allow the carer to be notified quickly if it becomes apparent that they are not suitable to take part in the study. Once fit with the criteria has been established participants will be invited for interview and will be sampled opportunistically.

If the criteria are met and they wish to continue then a convenient time and place will be arranged to meet. To decrease travelling and time constraints for parents they will be offered the option of having the interview at their home, although this will be offered alongside alternative locations in the community that are easily accessible. Other examples of available locations include rooms in a local counselling service, sure start centres and GP surgeries. In the event of a home visit Lancashire Care Foundation Trust and [REDACTED] [REDACTED] lone working protocols have been considered and a protocol has been devised (see Appendix F)

Interviews

The basic format of the meeting will consist of: introductions, a semi-structured interview, closing questions from the interviewee and arrangements for feeding back the findings. On arrival there will be general introductions about who the researcher is and the purpose of the research. The interviewer will then go through the participant information sheet (see Appendix A) with the participant, answering any questions and making sure that they understand the form. The interviewer will then go through the consent form (see Appendix B), which details participants' right to withdraw and the principle of confidentiality. Participants will only be asked to give consent once they have agreed that they understand and have had their questions answered to their satisfaction. Insufficient capacity to consent should not be a problem within this sample; however the thorough explanation of the forms will give the interviewer an opportunity to quickly consider this issue and if there are any concerns it will be treated as a risk issue. Participants will then be asked to give a pseudonym that will be used in transcription of the interviews and alongside any quotations that are in the report. Participants will also be asked to give some demographic details to be considered in the analysis (see Appendix C).

The interview will be face-to-face and audio recorded, lasting approximately 1-1 ½ hours at one of the locations previously mentioned. The interviews will be semi-structured allowing the interviewer to be able to provide occasional prompts if required but usually letting the interviewee tell their narrative uninterrupted. Examples of the types of prompts that will be used are in Appendix C. The prompts will be reviewed throughout the interviews, to allow the researcher to be responsive to parents' discussion of their experiences (see Appendix C). Participants will also be encouraged to bring objects that relate to their experience of adjustment, e.g. photographs or appointment letters, to act as prompts and support their narratives, although this will be at the participant's discretion.

The interviewee will be notified that if they become distressed at any point they will be consulted as to how they would like to proceed e.g. take a break, terminate the interview or continue. After the interview the parent will be asked if they have any questions and whether they want to be contacted to find out about the findings. If they say yes, they will be called within 4 months of their interview to arrange a way to give this information.

Confidentiality

All paperwork containing personal data (e.g. consent forms) will be stored in designated lockable cabinets at Lancaster University. All electronic data (audio recordings and incomplete transcripts) will be stored on a password protected laptop and then in an encrypted drive at the researcher's residence or transferred to the University's network drive, which is located on a virtual private network and is only accessible to the researcher, requiring a password. Personal data will be destroyed when the report is submitted. Transcripts will be kept by Lancaster University for five years after the completion of the study.

Analysis

This study aims to investigate parents' understanding and experience of their child having Duchenne Muscular Dystrophy through their narratives of adjustment. Semi structured interviews will allow participants to provide detailed subjective and storied accounts prioritising experiences they feel are relevant. Consequently it is difficult to precisely predetermine the number of interviews that should be undertaken, however recent studies suggest that up to 12 participants should be sufficient (Guest, 2006).

The interviews will be transcribed by the chief researcher to gain an initial understanding of the nuances and commonalities in the data. Transcripts will be analysed using narrative analysis. Interviews will be coded to generate subjective narratives relevant to the research

question. Crossley's (2007) analytic method will be used to guide the analysis. This outlines six stages which will help to focus the analysis:

1. Reading and familiarisation with the transcripts, i.e. a process of persistent engagement.
2. Identifying important concepts to look for.
3. Identifying narrative tone.
4. Identifying narrative themes and images.
5. Weaving a coherent story together.
6. Writing up as a research report.

Although Braun and Clarke's (2006) 15 point quality checklist focuses on thematic analysis, the principles apply to qualitative analysis in general and therefore will be used to ensure the quality of the study. This will be demonstrated by use of thorough audit trails, coding manual and direct participant quotations.

Process	No	Criteria
Transcription	1	The data have been transcribed to an appropriate level of detail, and the transcripts have been checked against the tapes for 'accuracy'.
	2	Each data item has been given equal attention in the coding process.
	3	Themes have not been generated from a few vivid examples (an anecdotal approach), but instead the coding process has been thorough, inclusive and comprehensive.
	4	All relevant extracts for all each theme have been collated.
	5	Themes have been checked against each theme have been collated.
	6	Themes are internally coherent, consistent, and distinctive.
	7	Data have been analysis – interpreted, made sense of – rather than just paraphrased or described.
	8	Analysis and data match each other- the extracts illustrate the analytic claims.
	9	Analysis tells a convincing and well-organized story about the data and topic.

Overall	10	A good balance between analytic narrative and illustrative extracts is provided.
	11	Enough time has been allocated to complete all phases of the analysis adequately, without rushing a phase or giving it a once-over-lightly.
	12	The assumptions about, and specific approach to, thematic analysis are clearly explicated.
	13	There is a good fit between what you claim you do and what you show you have done- i.e, described method and reported analysis are consistent.
	14	The language and concepts used in the report are consistent with the epistemological position of the analysis.
	15	The researcher is positioned as active in the research process; themes do not just 'emerge'.

Practical issues

Practical issues relate to expenses, interviewing, recruitment and ensuring confidentiality.

The location of the interview will be negotiated with each of the participants ensuring that confidentiality and safety are considered. A 'buddy' system in line with the Lancaster University, Lancashire Care NHS Foundation Trust and the [REDACTED]

[REDACTED] lone working protocols has been created, which will be used at all locations (Appendix F).

The digital recorder cannot be encrypted but will be stored securely until the audio recordings can be transferred to password protected computers and encrypted drive either at Lancaster University or at the home of the chief researcher. Further security is available on the researcher's laptop, where files will also be stored on an encrypted drive. The interviews will then be transcribed and pseudonyms will be used to anonymise the transcripts. The research tutor will only be able to view the transcripts once they have been anonymised. The field supervisor will give support and only view the data once the themes have been identified. The pseudonyms will be used alongside any quotations in the report and write-up for publication.

Ethical concerns

Participants will be allowed to withdraw from the study for up to 2 weeks after interview, which is documented on both the Participant Information Sheet and the Consent Form (see Appendices A & B). This deadline has been put in place to allow a reasonable amount of time for participants to consider whether they wish to withdraw. After this time data analysis will begin, ending the allocated withdrawal period.

Data will be anonymised using a pseudonym inserted during transcription and individual quotes will be assessed within the write up to determine whether any situational cues could cause their identity to be ascertained. If this were deemed to be the case then details will be modified to preserve their anonymity.

Timescale

November/December 2013: apply for ethical approval.

November-February 2014: once ethics has been approved then start recruitment.

November – February 2014: conduct interviews, transcription and coding.

May 2014: submission of final report.

September 2014: submit paper for publication.

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Appendix A

Participant Information Sheet

Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy.

My name is Toni Deavin and I am doing this research as a trainee clinical psychologist on the Clinical Psychology Doctorate programme at Lancaster University.

What is the study about?

Caring for children with a chronic illness can be a stressful job and this study wants to ask parents i.e. Mums and Dads, about their experiences of adjustment to their child having Duchenne Muscular Dystrophy. This will help professionals to understand what is important to parents and the findings from the study could provide useful information to develop services to support your needs.

Why have I been approached?

You have been approached because you are a parent or long-term carer of a child who has Duchenne Muscular Dystrophy. When I say long-term carers I mean carers who look after children in foster care and the placement is meant to last until the child is 18 years old. If you are a long-term carer then you must have cared for the child since before the symptoms started or a diagnosis was given. The child's diagnosis must have occurred over two years ago and you must currently be caring for the child. Unfortunately we cannot interview you if your child has many serious problems that are not associated with Duchenne. This might include other chronic illnesses like cystic fibrosis. If you are not sure about this you can always call the number below and check.

It is important to note that the research team for the study (myself and two supervisors) will not have access to your contact details or any personal information unless you give them to us.

Do I have to take part?

No. It's completely up to you to decide whether or not you take part. If you decide to take part you are free to stop at any time throughout the research process and you can ask to remove your interview from the dataset for up to 2 weeks after the meeting. You do not need to give a reason.

What will I be asked to do if I take part?

If you decide you would like to take part, you will need to let me know you are interested by contacting me on the details at the bottom of this form. I will then contact you on your preferred method of communication, either phone call or email, to arrange a time and place to meet for a face to face interview with me, lasting approximately one to one and a half

hours to discuss your experiences. You can bring with you an item that you connect with your adjustment, for example, this might be a letter or a photograph that will help you to tell your story.

On arrival I will go through the information on this sheet with you and you will be given the opportunity to ask questions. Once you are satisfied you understand the study and if you still wish to take part, then you will be asked to complete the Consent Form before starting the interview. The interview will be recorded so that I can type out the conversation into a paper version of the interview called a 'transcript'. After the interview you will be free to ask any questions that you may have. You will also be asked if you mind being contacted to discuss the results of the study, which is part of the analysis. If you agree to be contacted then you will hear from me within 2 months of your interview to arrange a meeting or a chat over the phone.

Will my data be confidential?

Your data will be kept confidential in two ways: anonymisation and secure data storage.

These are described below:

1. You will be asked to provide another name that is not your own, known as a pseudonym, which will take the place of your name on the transcripts and appear alongside any direct quotations in the report or publications.
2. The data collected for this study will be stored securely and only the researchers conducting this study will have access to this data:
 - Audio recordings will be stored securely on a password protected computer and will be encrypted. Only I will listen to the interviews to type them out to make the transcripts. The audio recordings will then be deleted after the report has been completed.
 - Hard copies of personal information (i.e. participant consent forms) will be kept in a locked cabinet and be destroyed on completion of the report.
 - At the end of the study, hard copies of transcripts will be kept securely in a locked cabinet for five years at Lancaster University. At the end of this period, they will be destroyed.

However there are some limits to confidentiality: if what is said in the interview makes me think that you, or someone else, is at significant risk of harm, I will have to break confidentiality and speak to my supervisors. If possible, I will tell you if I have to do this. If I then need to do something, like speak to your social worker, I will discuss this with you beforehand, so you know what is going on. This will be explained again at the beginning of the interview, where you can ask questions.

What will happen to the results?

The results will be summarised and reported in a project that is part of the evaluation process for the Doctorate in Clinical Psychology at Lancaster University. This will be completed in June 2014. Findings may then be submitted for publication in an academic or professional journal.

Are there any risks?

There are no risks expected with participating in this study, although some participants may find discussing events and feelings that have occurred in the past upsetting. **However, you**

are not required to share any information that makes you feel uncomfortable and it is your choice what you share with me. Should you feel upset at any point, but able to complete the interview, you can ask to take a break and the digital recorder will be paused until we start again. If you feel unable to complete the interview then you can tell me this at the time and we will stop. There will also be time at the end of the visit to discuss any concerns or queries you may have. However, if you experience any distress following participation you are encouraged to inform me and I can let you know about some helpful services. You may also wish to contact your GP or other agencies involved for further support.

Harm

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against Lancaster University, but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

Are there any benefits to taking part?

There are no direct benefits in taking part. However, you may want to take part as your involvement will help add to professionals' understanding of parental adjustment to their child's diagnosis of DMD and their needs. Services may then be able to take this into account when they work with families, to provide a better service.

Who has reviewed the project?

This study has been checked by Lancaster University through a peer-review process and then the Doctorate of Clinical Psychology exam board and the Research Support Office. The study was then checked and approved by the NRES Committee (TBD) and the [REDACTED] Research and Development department.

Where can I obtain further information about the study if I need it?

If you have any questions about the study, please contact the researcher:

Toni Deavin (trainee clinical psychologist at Lancaster University)

Email: a.deavin@lancs.ac.uk

Tel: (TBD)

Complaints

If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher, you can contact:

Sheila Payne

Head of Division, Health Research,

Tel: (01524) 593308

Email: k.froggatt@lancaster.ac.uk

Lancaster University, Lancaster, LA1 4YD

If you wish to speak to someone outside of the Clinical Psychology Doctorate Programme, you may also contact:

Professor Paul Bates

Associate Dean for Research

Faculty of Health and Medicine

(Division of Biomedical and Life Sciences)

Tel: (01524) 593718

Email: p.bates@lancaster.ac.uk

Lancaster University, Lancaster, LA1 4YD

Thank you for taking the time

If you would like to take part in the study, please contact me on the details below or return the cut off slip to the address provided.

Toni Deavin
Clinical Psychology,
Division of Health Research,
Lancaster University
Lancaster
LA1 4YT

Tel: *(I am in the process of arranging for the university to loan me a mobile phone. The allocated number will be inserted here)*

Email: a.deavin@lancaster.ac.uk

Participant's name: _____

Contact details (preferred form, i.e. email address or phone number):

How long ago was your child diagnosed with Duchenne Muscular Dystrophy?

How many children do you have?

If you are a guardian/long-term foster carer, did you know the child had Duchenne when you took the placement? And did they have any symptoms?

Do you have a partner?

Are you currently caring for the child?

Appendix B

Consent Form

Study Title: Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy.

We are asking if you would like to take part in a research project that will ask parents and long-term carers about their stories of adjustment to their child having Duchenne Muscular Dystrophy.

Before you consent to participating in the study we ask that you read the participant information sheet and mark each box below with your initials if you agree. If you have any questions or queries before signing the consent form please speak to the chief investigator, Toni Deavin.

Please initial box after each statement

1. I confirm that I have read the information sheet and fully understand what is expected of me within this study.
2. I confirm that I have had the opportunity to ask any questions and have them answered.
3. I understand that my interview will be audio recorded and then made into an anonymised written transcript using a pseudonym of my choice.
4. I understand that audio recordings will be kept until the research project has been examined.
5. I understand that I am not obliged to take part in this study and can withdraw my participation before, during, or up to 2 weeks after my interview.
6. I understand that any hard copies of personal information (like the consent form), will be destroyed on submission of the final report.
7. I understand that the information from my interview will be pooled with other participants' responses, anonymised and may be published. I understand that when direct quotes are used in the write-up the quote will be anonymised and the pseudonym will be used.
8. I consent to information and quotations from my interview being used in reports, conferences and training events.
9. I understand that any information I give will remain strictly confidential and anonymous unless it is thought that there is a risk of harm to myself or others, in which case the chief investigator will need to share this information with her supervisors.
10. I consent to Lancaster University keeping written transcriptions of the interview for 5 years after the study has finished. At which time they will be destroyed.
11. I understand that relevant data collected during the study, may be looked at by individuals from Lancaster University, from regulatory authorities or from the NHS Trust, where it is

relevant to my taking part in this research. I give permission for these individuals to have access to this data.

12. I understand that I am not obliged to take part in this study and can withdraw my participation before, during, or up to 2 weeks after my interview.

13. I consent to take part in the above study.

Name of Participant _____

Participant Signature _____

Date _____

Name of Researcher _____

Researcher Signature _____

Date _____

Appendix C

Example of the semi-structured interview schedule:

Opening question	Examples of Prompts
<p>9. Hello...</p> <p>10. Make comfortable...Before we get started would you like a drink? (whatever is available: tea/coffee/water/squash). Biscuits?</p> <p>11. Introductions</p> <p>12. Go through the participant information sheet</p> <p>13. Check criteria/demographic information</p> <p>14. Consent form</p> <p>15. So we've talked about risk and confidentiality and when I would need to break confidentiality. And I really want to make sure that all the information you give me is kept personal and private. I also want to make sure that you and the others around you are safe.</p> <p>16. Right, I've already done a lot of talking and asked you a lot of questions. Is there anything you would like to ask me before we start?</p>	<ul style="list-style-type: none"> • My name and title • What a trainee psychologist is? • Why I am doing the research. • Age • The diagnosis must have been given at least 2 years ago. • Currently caring for the child? • The child must not have any other serious illnesses not associated with DMD. • The family must not be under investigation. • So to keep what you say private, I am using false names to replace the names of the parents I talk to. What name you would like to have on the interview transcript and any quotes I might use from our discussion? <ul style="list-style-type: none"> ➤ this is to make sure you cannot be identified.

<p><u>SWITCH ON TAPE RECORDER!</u></p> <p><u>Introduction to the process and topic area</u></p>	<p>I am interested in your adjustment to having a child with Duchenne. I want to know what you think adjustment is and what has been important to you along this journey.</p> <p>Today we will have up to an hour and a half for you to tell me your story.</p> <p>I may occasionally say something but I will try to keep this to a minimum. If you brought a meaningful item with you, you can use this to help remind you of things to discuss.</p>
<p><u>Specific prompts</u></p>	<ul style="list-style-type: none"> • So where is a good place to start? • What have the key events been in this process? • What has influenced your adjustment? • How has this affected how you see yourself? • Can you tell me what things were like before....(the diagnosis)? • So, tell me about after...(the diagnosis)? • And later.... • Did you bring an object that represents your adjustment? If so, tell me about it. • Who has been important on your journey? • Tell me about how you see your future. • Where would you say you are on your journey of adjustment?
<p><u>Generic prompts</u></p>	<ul style="list-style-type: none"> • Tell me more about that... • Describe that to me.... • What happened next? • What influenced that?
<p>Ask any questions?</p> <p>When and how can I contact you about the results?</p>	



Appendix D
Cover letter for mail out

Hello,

I am a trainee clinical psychologist at Lancaster University, and I am currently looking to find parents or guardians of children who have Duchenne Muscular Dystrophy, to take part in the research project I am doing as part of my studies. The title is: **“Parents’ stories of adjustment to their child’s chronic illness: a narrative approach to Duchenne Muscular Dystrophy.”** I would be very grateful if you would consider taking part.

The study has been checked and approved by the NHS Research Ethics Committee and the project is also being helped by the [REDACTED].

If you have received this letter by post it is because the [REDACTED] has sent it out as part of a general postal recruitment drive for the study. You have not been singled out for any specific reason, other than you are a parent/guardian in the local area and you have a child with Duchenne. The research team and I have not seen your contact details or any personal information and you will not hear from us again unless you decide to take part.

Included with this letter is a Participant Information Sheet, which contains all the specific information about the study and how you can contact me if you decide you would like to ask any questions or take part.

If you are unable or do not want to participate in the research but know someone who would be suitable and might be interested, please pass on this information.

I really appreciate you taking the time to read this letter. If I do not hear from you within 2 weeks of this letter being posted I will assume you do not wish to take part.

Thank you in advance.

Best wishes

Toni Deavin
Trainee Clinical Psychologist
Lancaster University/Lancashire Care NHS Foundation Trust

Are you a parent of a child with Duchenne Muscular Dystrophy?



Would you like to tell your story?

You are invited to take part in research on “Parents’ stories of adjustment to their child’s chronic illness: a narrative approach to Duchenne Muscular Dystrophy.”

What is the study about?

Caring for children with chronic illness can be a demanding job and this study wants to ask parents and long-term carers about their experiences of adjustment to their child having Duchenne Muscular Dystrophy. This study aims to gather these stories to provide personal and relevant perspectives of adjustment, specifically noticing what has influenced their stories. This will help professionals to understand what is important to parents and consequently to consider how support can be provided to fit parents’ needs.

How you can help?

I am looking for up to 12 parents or long-term guardians to attend face-to-face interviews, lasting approximately a 1-1½ hours.

The interview will include:

- Your story of adjustment.
- This might include:
- What you think adjustment is
 - Your choice of events that have been influential to your adjustment.
 - What it is like caring for a child with Duchenne Muscular Dystrophy.
 - How life compared before symptoms became apparent.

Interested?.....What you need to do

Please contact me on the details below, giving your name, address or email address and I will send you a detailed information sheet outlining the study, what is involved, and what happens next.

Toni Deavin (trainee clinical psychologist)
Clinical Psychology, Division of Health Research,
Lancaster University, Lancaster, LA1 4YT

Tel: **being arranged**

Email: a.deavin@lancaster.ac.uk

Toni Deavin
(trainee clinical psychologist)
Clinical Psychology, Division
of Health Research,
Lancaster University,
Lancaster, LA1 4YT
Tel: **being arranged**
Email:
a.deavin@lancaster.ac.uk

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(trainee clinical psychologist)
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(trainee clinical psychologist)
Clinical Psychology, Division
of Health Research,
Lancaster University,
Lancaster, LA1 4YT
Tel: **being arranged**
Email:
a.deavin@lancaster.ac.uk

Appendix F

Interview safety procedure

1. Once a time and place has been organised for the interview I will send two emails, one to Dr [REDACTED] and one to Dr Pete Greasley, who will be my designated Buddy.
2. The email to the allocated supervisor will look like:

Dear [REDACTED],

I have organised an interview in the local area. This email is to notify you that it will take place on **(insert date)** at **(insert time)**. This is to make you aware that I may need to contact you around this time if there are any risk concerns. I will let you know if the interview is cancelled. Please respond to this email so that I know you have received it. If you know that you will not be available at this time, please let me know so that I can make other arrangements.

I will also contact Pete Greasley to let him know so that he can act as my lone working buddy.

Best wishes

Toni

3. The email to Pete will look like this:

Dear Pete,

I have organised an interview in the local area. This email is to notify you that it will take place on **(insert date)** at **(insert time)**. This interview is due to finish at **(insert time)**. Please respond to this email so that I know you have received it and whether you are able to be my buddy at this time, so I can make other arrangements if necessary. I will let you know if the interview is cancelled.

Summary of procedure

- I will call you within half an hour of the interview completion time.

- If you do not hear from me please call my mobile numbers (insert my research and personal mobile numbers).
 - **If I respond** and use the *safe word* (to be agreed between Pete and myself), please open both attached documents and give the enclosed details to the police.
 - **If I do not respond** and you do not hear from me, try again in 30 minutes. If you cannot get through to me after an hour of the interview completion time, please open the attachment document called 'location'. If the location is a community service base, please call them to ascertain whether I am in the building or if I have left and under what circumstances. If the location is the interviewee's home then please call the interviewee's home number to check my safety.
- If you have not been able to reach me for an hour and a half after the interview completion time and you have gone through the above steps, please open the 'personal information' document and give all details to the police.

N.B I will not text you. If you receive a text please react as if I have not called you.

Best wishes

Toni

4. The first attached document will be called 'location' and look like:

Name of interviewer: Toni Deavin

Location of interview (delete as appropriate): Base in the community / interviewee's home

If the interview is being held at a base in the community, please provide the address and telephone number for the facility. If it is at an interviewee's home, please provide the landline telephone number only:

5. The second attached document will be called 'personal information' and look like:

PERSONAL INFORMATION

EMERGENCY CONTACT AND PERSONAL IDENTIFICATION DETAILS

Lone worker's name:	
Interviewee's name:	
Address and telephone number of interviewee	

Lone worker's Home address:		
Date of birth:		
Contact telephone Numbers:		
MOBILE-WORK		
<u>MOBILE-PERSONAL</u>		
Personal description: (height, build, hair and eye colour, any distinguishing marks, features etc)		
Car details:		
Make:		
Model:		
Colour:		
Registration plate details:		
Nominated emergency contact person (friend or relative): Husband		

Name:		
Address:		
Contact telephone numbers:		
MOBILE		
Next of kin details:		
Name:		
Address:		
Contact telephone numbers:		

Appendix G – Online Advert**Are you a parent of a child with Duchenne Muscular Dystrophy?
Would you like to tell your story?**

You are invited to take part in research on “Parents’ stories of adjustment to their child’s chronic illness: a narrative approach to Duchenne Muscular Dystrophy.” (Ethical approval given by Lancaster University and the NHS Research Committee)

What is the study about?

Caring for children with chronic illness can be a demanding job and this study wants to ask parents i.e. Mums and Dads, and long-term carers about their experiences of adjustment to their child having Duchenne Muscular Dystrophy. This study aims to gather these stories to provide personal and relevant perspectives of adjustment, specifically noticing what has influenced their stories. This will help professionals to understand what is important to parents and consequently to consider how support can be provided to fit parents’ needs.

How you can help?

I am looking for up to 12 parents or long-term guardians to attend face-to-face interviews, lasting approximately a 1-1½ hours.

The interview will include:

- Your story of adjustment.

This might include:

- What you think adjustment is
- Your choice of events that have been influential to your adjustment.
- What it is like caring for a child with Duchenne Muscular Dystrophy.
- How life compared before symptoms became apparent.

Interested?.....What you need to do

Please contact me on the details below, giving your name, address or email address and I will send you a detailed information sheet outlining the study, what is involved, and what happens next.

Toni Deavin (trainee clinical psychologist)
Clinical Psychology, Division of Health Research,
Lancaster University, Lancaster, LA1 4YT

Tel: *being arranged*
Email: a.deavin@lancaster.ac.uk



Health Research Authority

NRES Committee South Central - Hampshire B

Bristol REC Centre
Level 3 Block B
Whitefriars
Levens Mead
Bristol
BS1 2NT
Telephone: 01173 421383
Facsimile: 01173 420455

11 December 2013

Ms Antoinette Deavin
Clinical Psychology, Faculty of Health and Medicine,
C16 Furness College, Lancaster University,
Lancaster, Lancashire
LA1 4YG

Dear Ms Deavin

Study title: Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy
REC reference: 13/SC/0649
Protocol number: 1
IRAS project ID: 136834

The Proportionate Review Sub-committee of the NRES Committee South Central - Hampshire B reviewed the above application on 10 December 2013.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the REC Manager, Libby Watson nrescommittee.southcentral-hampshireb@nhs.net

Ethical opinion

The Committee raised concerns that participants would not be able to withdraw their data 2 weeks after the interview.

On behalf of the Committee, the sub-committee gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see



Health Research Authority

“Conditions of the favourable opinion” below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

1. Please correct the typographical error at item 6 of the Consent form, as this does not currently make sense.
2. Please ensure that the Chief Investigator's name is spelt consistently throughout the Participant Information Sheet.
3. Please make it clear in the PIS whether parent's means both mums and dads.
4. In the protocol please provide greater clarity or examples for the online recruitment plans.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission (“R&D approval”) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites (“participant identification centre”), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non clinical trials this is not currently mandatory.



Health Research Authority

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The documents reviewed and approved were:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Advertisement	V.1	02 December 2013
Evidence of insurance or indemnity		11 July 2013
Interview Schedules/Topic Guides	V.1	02 December 2013
Investigator CV	A Deavin	02 December 2013
Letter from Sponsor		26 November 2013
Letter of invitation to participant	V.1	02 December 2013
Other: C.V - C Dixon		
Other: C.V - [REDACTED]		
Other: C.V - P Greasley		
Other: Interview Safety Procedure		
Participant Consent Form	1	02 December 2013
Participant Information Sheet	1	02 December 2013
Protocol	1	02 December 2013
REC application	136834	02 December 2013

Membership of the Proportionate Review Sub-Committee

The members of the Sub-Committee who took part in the review are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review



Health Research Authority

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

information is available at National Research Ethics Service website > After Review

13/SC/0649

Please quote this number on all correspondence

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

With the Committee's best wishes for the success of this project.

Yours sincerely

Dr Chris Markham
Alternate Vice-Chair

Email: nrescommittee.southcentral-hampshireb@nhs.net

Enclosures: List of names and professions of members who took part in the review

"After ethical review – guidance for researchers" [SL-AR2]

*Copy to: Ms Debbie Knight
Ms Alison Robinson, Central Manchester University Hospitals
Foundation Trust*



Health Research Authority

NRES Committee South Central - Hampshire B

Bristol REC Centre

Level 3 Block B

Whitefriars

Lewins Mead

Bristol

BS1 2NT

Telephone: 0117 342 1384

Facsimile: 0117 342 0445

20 December 2013

Ms Antoinette Deavin
Clinical Psychology, Faculty of Health and Medicine,
C16 Furness College, Lancaster University,
Lancaster, Lancashire
LA1 4YG

Dear Ms Deavin

Study title: Parents' stories of adjustment to their child's chronic illness: a narrative approach to Duchenne Muscular Dystrophy
REC reference: 13/SC/0649
Protocol number: 1
IRAS project ID: 136834

Thank you for your letter of 19 December 2013. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 11 December 2013

Documents received

The documents received were as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Advertisement	Online Advert v1	16 December 2013
Participant Consent Form	2	19 December 2013
Participant Information Sheet	2	19 December 2013
Protocol	2	16 December 2013

Approved documents

The final list of approved documentation for the study is therefore as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Advertisement	V.1	02 December 2013
Advertisement	Online Advert v1	16 December 2013
Evidence of insurance or indemnity		11 July 2013
Interview Schedules/Topic Guides	V.1	02 December 2013
Investigator CV	A Deavin	02 December 2013



Health Research Authority

Letter from Sponsor		26 November 2013
Letter of invitation to participant	V.1	02 December 2013
Other: C.V - C Dixon		
Other: C.V - [REDACTED]		
Other: C.V - P Greasley		
Other: Interview Safety Procedure		
Participant Consent Form	2	19 December 2013
Participant Information Sheet	2	19 December 2013
Protocol	2	16 December 2013
REC application	136834	02 December 2013

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

13/SC/0649

Please quote this number on all correspondence

Yours sincerely

Libby Watson
REC Manager

E-mail: nrescommittee.southcentral-hampshireb@nhs.net

Copy to: *Ms Debbie Knight,*

[REDACTED]



Antoinette Deavin
Clinical Psychologist
Child & Adolescent Psychiatry
Lancaster University
Lancaster LA1 4YGT

Our Ref. [Redacted]

Dear Antoinette

Study: Parents' stories of adjustment to their child's chronic illness
Sponsor: Lancaster University
Chief Investigator: Antoinette Deavin
Local Liaison: [Redacted]
[Redacted] R03486

We have received a request for authorisation for our Trust to become involved as a **Participant Identification Centre (PIC)** for the above study, and [Redacted] confirm that as a PIC site all research activity will take place off site.

Following receipt of the documentation listed at the foot of this letter, we have completed the minimum governance checks required (for PICs) and can confirm our Trust's agreement.

I would like to take this opportunity to wish you well with your research.

Yours sincerely

[Redacted signature]

Date: 28/01/2014

cc [Redacted]