Narrative accounts of parenthood following the death of a child to muscular dystrophy

James Randall-James

June 2017

Submitted to the University of Hertfordshire in partial fulfilment of the requirements of

the degree of Doctor of Clinical Psychology

Abstract

Rationale and Aims: Research into the lived experience of parenting children with muscular dystrophy has typically addressed key transitions along the disease trajectory, such as diagnosis or end-of-life care. Families reportedly face continuous challenges as their child's health deteriorates. No research has considered accounts of parenting across the lifespan that look at adaptation following their child's death. This research was conducted in the context of a wish-fulfilment charity that offer experiences for children to be supported in activities that are usually deemed inaccessible. In this context, the study asked how do parents who have lost a child to muscular dystrophy story their experiences of parenting.

Methods: This research used a qualitative approach that explored the accounts of eight parents interviewed in couples, all of who had experienced the death of their child to muscular dystrophy. The study used a semi-structured interview, lasting from 100-150 minutes each. Interviews were video-recorded, transcribed, and analysed using narrative analysis to explore what and how the parents narrated their experiences. Consideration was given to the social and cultural contexts that shaped these.

Analysis: Multiple readings of the transcripts allowed me to develop individual summaries and then construct an analysis across all of the accounts. Three main stories of change, survival and creating change emerged through my analysis. These three stories represented six sub-stories in total: waking up to different futures; being so close, you don't see the deterioration; humour through the struggle; storytelling together; creating a legacy; and living the dream.

Findings: Couples narrated the loss of parental dreams, leading to the need for identity (re)formation. Humour and storytelling together were often used to regulate emotions during the storying telling, and a means of surviving their loss. Parents shared narratives of building legacies and the memories created through 'living the dream', which alluded to an impact that surpassed death itself.

Implications: These findings suggest the need for greater consideration of sense-making, changing identities, and benefit-finding in clinical consultations, at key transitions during the parenting journey and particularly following the death of a child to muscular dystrophy. Accounts suggest that wish-fulfilment events can sustain hope for parents, a proposition that will need further investigation in the future.

Acknowledgements

I would like to extend my sincere thanks to the individuals who participated in this study. Very special thanks are also due to Michael and Sue, who have remained curious, open and supportive throughout the entirety of this journey. I have learned a lot from you both, personally and professionally.

I wish to acknowledge and thank both Dr David Wellsted and Dr Lizette Nolte for their continued encouragement, energy and support throughout this process. Thank you both very much.

There remain some very influential and important individuals who have got me here, and it would be amiss of me to not thank them. Naomi, who has witnessed and tolerated me at my best and worst – so thank you. My parents, Peter and Polly, and brother, Richard…I am still alive, I promise I will come and see you soon! And thank you for all of the support along this arduous journey from dustman to clinical psychologist!

Honestly, and wholeheartedly, I would not have survived this process without my PBL comrades: Amy, Hannah, Jess, Jacqui, and Sarah… I carry our group wherever I may roam, from clinic to campaign; from lecture to pint: We can and will forever 'Help the person, change the world!'

And finally, but by no means least, I thank you Lee.

"Who is Lee?" you may ask. And that dear sir/madam, is the very point.

Narrative accounts of parenthood following the death of a child to muscular dystrophy

Table of Contents

Chapter one: Introduction

This research is about parenting young people diagnosed with muscular dystrophy (MD) who have participated in a third-sector wish-fulfilment event known as a Muscle Dream. This research used interviews to explore couples' accounts of parenting in the context of loss and bereavement. It considered the retelling of lived experiences as vital to the sense-making and identity formation of the interviewed parents. Narrative inquiry was used to explore the content, structure, performance, and discursive elements, of the accounts individually and collectively.

1.1 Overview

Within this chapter I start by briefly introducing the range of conditions considered to be forms of MD. I then present my perspectives on epistemology and the importance of language. These perspectives are shared to help the reader understand how I have developed my position in relation to this research project, which includes my account of why the topic area resonated for me personally and influenced me to undertake the project. In doing so, I consider the epistemological assumptions I make in approaching research in clinical psychology and the implications this has on the language that I use in order to convey meaning and my own particular 'research narrative'. Thereafter, I describe the available research on families living with MD, that includes a systematic literature review and evaluation of those studies included. To conclude this chapter, I provide a rationale for the study that lead to a description of the research aims.

1.1.2 Introducing muscular dystrophy

The term MD indicates a range of neuromuscular diagnoses, with nine main categories that cover over thirty specific subtypes of the condition. There are notable differences in the causes, symptoms and prognosis of each of these conditions.

Myotonic and Facioscapulohumeral MD are both inherited, autosomal-dominant forms of the disease that lead to weakening and the eventual loss of muscle tissue in young adults. Facioscapulohumeral MD is unique in the sense that one inherits particular genes but that these do not alone cause the condition, instead a second process is needed, whereby a particular set of dead cells are 'awoken' – activating the disease (Lemmers et al., 2010).

Duchene and Becker muscular dystrophy are two of the most common forms of the disease to manifest much earlier in childhood, typically by the age of four. Both types are terminal and recessive diseases linked to inherited mutations on X-chromosomes (Webb, 2005). Most children affected by MD are boys, though the condition can occur within young females – often when *both* X-chromosomes inherited carry the mutated gene (Pearson & Van Ommen, 1984).

This study specifically addressed the experience of parenting children diagnosed with MD, who had passed away from the condition before reaching their thirtieth birthday. As such, this focus excludes some of the variations of the condition, such as myotonic MD. The parents of young people who had lived with Facioscapulohumeral or Becker MD could have been included in this study, but those eventually recruited retold their accounts of parenting a child diagnosed with Duchene MD. As such, the focus of this

thesis is purposely narrow, on a specific population within the range of neuromuscular conditions. Before exploring the lived experience of MD for individuals and families, I provide a brief account of how I came to decide on this research area in particular. In doing so, I first present a range of perspectives that inform how I understand the literature and 'data', which I hope will guide the reader as to how I interpret and document the accounts of the parents I interviewed.

1.1 My position

Where the researcher sits in relation to their research has long been a moot point. The debate in part, reflects the underpinning epistemological assumptions made within the aims of any given project (Scotland, 2012). This can be crudely surmised through acknowledgement that some researchers pursue the 'discovery' of scientific knowledges, whereas others 'create' such knowledges through elaboration, documentation and immersion (Speed, 1991). These positions hold important implications for the role of the researcher and the possible impact of their own views and values. For those pursuing the discovery of truth, the self was to be suspended, deemed not relevant to the research or 'end product'. When understanding knowledges as co-constructions, the role of the researcher needs to be openly addressed and potentially considered part of that end product. As such, I detail my own epistemological stance below, outline the implications for my use of language and detail my own personal journey that led me to this research.

1.2.1 Epistemological stance

When I was younger than I find myself today, I 'discovered' constructivism and social constructionism (Gergen, 1985). At the time, I struggled – these theories left me questioning all I had taken for granted. I became overwhelmed by what I feared was the eradication of all certainties and struggled to sleep. Despite this, I was intrigued. This dissonance was much like the moth to the flame. But herein is where my distress unravelled something most useful; if the moth is to burn, then there *must* be some certainty and reality that we can comment on.

The moth's demise cannot simply be 'one of many' ways of seeing the world. I use an analogy here, somewhat ironically, as we are said to co-construct an *approximation to reality* through analogy (Speed, 1991; White & Epston, 1990). At the time, I did not have the language to convey my epistemological position in a way that I do now, but I just knew that I could not condone a hard-line constructivist position in the context of disease, poverty and torture (as influenced by Hagan & Smail, 1997; Smail, 2005). Yes, we construct our own realities to some extent – but the key 'discovery' for me was that we were construing by-proxy; making sense of lived experience through the limits of our own bodies and minds. For this is part of the reality we exist in, our bodies and minds as the tools that limit how far we can reach out towards reality – if only we could reach a little further!

In short, "*Reality Exists, okay!*" (Speed, 1991, p. 395). But the tools we have to pursue it are limited, loaded and often, knowingly or unknowingly, intoxicated with values, principles and socio-cultural histories. My position is that our reality functions on the reciprocity of agentic constructivism, and the systems and structures of social constructionism. We shape, and are shaped by the systems around us. But just like the

chicken and the egg dilemma, 'realities' pre-date us; born into social worlds full of expectation, inter-generational scripts and ways of being, socio-cultural rituals and practices, and politico-economic powers. Perhaps I can now call this approximation to reality a 'critical realism' (Bhaskar, 2013), or a social-materialist approach (Smail, 2005), or even Speed's invention of co-constructivism (Speed, 1991) – either way, the prospect remains the same; *the truth is out there.* We just cannot quite reach 'there'.

1.2.2 Language

Writing in third-person could be argued to establish a sense of distance and suspend the self of the researcher from their subject area, deriving from a positivist hypothetico-deductive approach to research. As made clear in my epistemological position, I uphold the idea that a reality is 'out there' but that we can only study and present this 'reality' through our co-construing of communally acceptable knowledges that are approximations of 'truth'. As such, it is important for our language to not allude to a finality in our understanding or 'concreteness' in our conclusions – but to make it transparent that these are temporally and contextually situated knowledges, construed and presented by myself; the researcher. It is hence proposed, that through carefully choosing which type of pronoun to use in writing, the reader is reminded of my presence and influence on the stories retold through the research process, and on the overall research narrative presented (Webb, 1992).

I have chosen to integrate academic and personal writing styles in order to convey meanings within their particular and appropriate contexts. This represents the notion that context *cannot* be removed from the narrative plot (Hinchman & Hinchman, 1997).

Indeed, this means that I, the researcher, cannot necessarily be disentangled from the findings presented.

For the purposes of this thesis, I have embarked on the strategic use of first- and third-person pronouns. I have tried to embrace my own participation in these evolving knowledges of the parental stories that continue to unfold and take shape. This approach is used to make transparent my attempts at conducting this research from the position of a reflexive researcher (Hosking & Pluut, 2010), which I elaborate on further in the methods section.

1.2.2.2 Defining key concepts

Throughout this thesis I will use terms that could be understood in a range of different ways, particularly cross-culturally. As such, I briefly describe my own use of 'identity' and 'parenthood' in order to offer the framework from which I use and understand these concepts within the presented text.

I understand the term identity to represent a concept that functions to signify something about a person or collective (Tajfel, 1982). Identities can be self-assigned or indeed, prescribed by others (ibid.). In this sense, identity is a concept that allows a defining of people, a distinguishing of the self or selves (Trepte & Loy, 2017). Identity then, can be deconstructed, constructed and/or contested through the retelling of particular stories in ways that demonstrate cultural-relevance, or to the contrary, omit particular stories, depending on the particular context within which these are told (e.g. to convey desirable attributes). The contextual retelling or omission of stories that construct particular identities can also be understood in terms of a purposeful and

chosen identity presentation and/or a more implicit construction of identity through participating in associated actions and practices (Kogan & Gale, 1997). Indeed, this fits with a dramaturgical understanding of self in that identities are considered as roles an individual participates in and as if upon a stage, require particular actions, rituals and investments in order to 'play the part' (Goffman, 1956).

Heidegger (1996) suggests that such a distinction of selfhood is irrevocably linked to the relational; a conceptualisation of 'the other' held in mind. In this light, identity may have a comparative element to it – though this is not necessarily an explicit or effortful drive to define and distinguish oneself from others. As such, identity formation can be seen to function to offer meaning and purpose, sustaining action and the practice of such identities in social arenas. This definition of identity is consistent with much of Heidegger's (1996) theorising about selfhood and identity. In short, identities can be understood as categories or concepts that function to provide meaning to the self and to others, they are linked with how individuals talk and distinguish themselves in relation to their context and others, and enable or provide meaning and implications for practice. Additionally, the concept of identity communicates and distinguishes who one may have been in the past, who they consider themselves to be at present, and how they envision themselves in the future.

Parenthood is one particular example of an identity that someone may be assigned or assign themselves, that links an individual to others, and has particular practices associated with the enactment of such an identity. Indeed, parenthood infers a relational link to a third individual; that is, a child. Cross-culturally, it is accepted that becoming a parent is inexplicitly linked to biology and the reproductive cycle (Steinbock, 2006). However, as noted previously, this identity could be contested by the

individual, the other parent, or the child, based on their fulfilment of the related practices of parenthood. Beyond the biological link, parenthood is also defined by a social element that emphasizes the rearing, nurturing, caring and loving of a child. This social component of parenthood does not necessarily relate to the biological component of parenthood (Steinbock, 2006). As such, children could have multiple parents at once; from biological mother and father, to surrogate mothers, and those that rear the child but do not necessarily have any biological link (e.g. step-parents, foster parents, etc.). Indeed, identities such as parenthood can be viewed as a series of ongoing accomplishments as opposed to a tangible and static 'thing' (Kogan & Gale, 1997). For the purposes of this study, the social component of defining parenthood was used, whereby biological linkages were not assumed to define parenthood.

In recruiting participants, this study was advertised on the basis of exploring *parental* accounts. As such, I had already set a particular discursive framework of which I define here but did not do so for participants. However, this permitted a degree of freedom for participants to consider their identity and only choose to participate if they identified with this concept. This approach seemed consistent with the research aims, as the final sample included a range of families in which not all participants identified as birth parents.

1.2.3 The personal significance of why I chose this research

I came to this project from a position of relative naivety, having only heard about MD in passing and having never really fully considered what it would mean for someone to be born with not only a life-limiting, but life-threatening condition. This being said, the

potential of this project resonated for me for several reasons, which I briefly describe below.

At the time of deciding upon my project, I was at a key transition in my own life. I had recently married, was due to turn thirty and wanted to start a family of my own. I remember seeing snapshots of how joyous the beneficiaries[1] looked, as a representative of the Muscle Help Foundation visited our University to present on a series of 'wish-fulfilment' events entitled Muscle Dreams, delivered to young people diagnosed with MD. Vicariously, and perhaps paternalistically, I felt so relieved that there were pockets of society *enabling* these young people to fulfil some of their dreams without having to necessarily consider the barriers or obstacles that would usually get in their way.

I found it interesting how I could feel inspired and hopeful at the images presented during that presentation; snapshots of lived experience that were narrated before us, albeit visually. At the same time, I too seemed to co-construct narratives about enablement, 'living the dream' and escaping those societal obstacles, which were not overtly described on that day. As an audience member, I seemed to be participating in creating a narrative about living with MD and about the Muscle Dream experience; which interested me. There was a sense of a parallel process occurring whilst I was in my audience role, which led me to wonder what it is like for the parents to experience their children enabled *and* disabled by society in many shapes and forms over time. As such, I became interested in how they would narrate their parenting journey.

Much like my realisation that social constructionism was in danger of neglecting the social-materialist conditions of lived experience (as described above), I became

---

[1] The term beneficiaries is used within the Muscle Help Foundation for those young people who have been in receipt of, and experienced, a Muscle Dream event.

increasingly interested in thinking about the embodied experience. In part, this began when I read a quote from White and Epston's text *Narrative Means to Therapeutic Ends* (1990), which described how in the context of narratives, "there are always feelings and lived experience not fully encompassed by the dominant story" (Bruner, 1986, as cited in White and Epston, 1990, p. 12). Additionally, I wonder whether a lifelong coping strategy of intellectualising could at times make it difficult for me to embrace what my Jungian therapist at the time called the 'felt sense'; namely, and probably described inaccurately, the embodied *and* emotional content of lived experiences. This growing interest in the embodied experience, and the stories that our bodies hold for society, coincidentally tied-in well with another personal transition during training for me. Test results returned ambiguous and 'borderline diagnostic' for me carrying a genetic condition rife in my family and the cause of a premature death of my cousin at the age of thirty, and the cause for her daughter to have a heart transplant in her teens. With this threat of a physical, embodied, life-limiting and life-threatening condition possibly being held within my own body too, the project bore relevance to my own familial circumstances, and conjured up a professional intrigue within me.

Having briefly outlined the project, introduced the condition MD in general, and detailed the relevance of my own position in relation to the research, I now present an overview of the relevant literature with the narrow focus of Duchene MD.

1.3 An overview of the literature

Firstly, I briefly outline some of the literature around the onset, diagnosis and prognosis of MD. This is followed by a closer look at some of the signs and symptoms of MD, living with the condition, and the relevance of community interventions. I then present a systematic literature review addressing the experience of parenting a child with MD over time, based on a specific literature search strategy. The literature is then used to provide a rationale for the need of the current study, concluding the chapter with the specific aims of this research project.

1.3.2 Diagnosing muscular dystrophy

Over 30,000 people are said to be diagnosed with MD or related conditions within the UK (Muscular Dystrophy Campaign, as cited in Cunniff, Chisholm, & Chouliara, 2015). Parents often become concerned at key developmental milestones, reporting problems from the ages of six months to their child's third birthday (Bendixen & Houtrow, 2016). Although MD is often not diagnosed until the child's fifth birthday (Witte, 1985), those with a family history of the condition are increasingly being diagnosed much earlier than was historically the case (i.e. from two years of age; Daack-Hirsch, Holtzer, & Cunniff, 2013). The typical signs and symptoms that indicate the need for testing include delayed walking, a difficulty in rising from the floor or sitting up, a waddling gait, odd posturing, frequent falls, becoming easily tired, and over-developed calf muscles (Bendixen & Houtrow, 2016; Daack-Hirsch et al., 2013). The diagnostic process should involve an initial creatine kinase measurement and if found to be elevated, a referral to a specialist neuromuscular service for genetic testing is required.

The gradual, yet progressive and degenerative impact of MD is caused by a defect in dystrophin, a protein that is needed in the growth of healthy muscle fibres (Cunniff et al., 2015). As a result, the condition causes wastage, damage, and eventual death of skeletal muscle cells, which leads to increasing physical impairment over time as a result (Gagliardi, 1991a, 1991b). Wheelchair dependency often occurs between the ages of 10 and 12 years (Erby, Rushton, & Geller, 2006). To date, there is no curative treatment known for the condition, with individuals often dying in their late teens due to respiratory complications or heart failure (Samson et al., 2009). Although technological advances (e.g. ventilation machines) have improved the provision of care, with the potential to extend these young people's lives, opportunities to plan for such significant transitions in health are often missed (Erby et al., 2006). Even now, only a few individuals with MD will live beyond the age of thirty (Tomiak et al., 2007).

1.3.3 Living with MD

Much like other related diseases, MD is "life-threatening, life-limiting and chronically debilitating" (Waldboth, Patch, Mahrer-Imhof, & Metcalfe, 2016, p. 45). As such, the condition is likely to impact the quality of life of both the young person who is diagnosed and their family. The impact of MD is also relative to the stage of disease progression and is complicated further by the extent of knowledge, awareness and open dialogue about their diagnostic status within the family (Buchanan, LaBarbera, Roelofs, & Olson, 1979; Dawson & Kristjanson, 2003; Gagliardi, 1991a, 1991b).

As the disease progresses, children and young people living with MD are said to experience significant physical pain (Gagliardi, 1991a, 1991b). The disease phases, particularly in relation to ambulation and mobility, are associated with significant

increases in pain that are likely to impede the individual's quality of life unless preventative strategies and interventions are put in place in a timely manner (Bushby et al., 2010a, 2010b). The negative impact of pain on quality of life in general, is well documented (Ferrell, 1995).

When children are not informed of their diagnosis, they often perceive of themselves as 'different' from their peers with little understanding as to why (Buchanan et al., 1979). Buchanan and colleagues (1979) found that individuals often thought of themselves as incompetent and personally responsible for their 'weakness'. There may be cultural differences in how families approach disclosure and communication around diagnoses, with a correlation between difficulties in communication and psychiatric conditions, in particular depression (Fitzpatrick & Barry, 1990). Some of those who become aware of the terminal nature of their condition have been reported to adopt identities as 'futureless persons', in part reflecting an out-dated understanding that suggests death before one's twentieth birthday is inevitable (Gibson, Zitzelsberger, & McKeever, 2009). Even with increasing medical and technological innovations, the condition continues to take the life of young people before or during their early thirties. 'Futureless' identities have been described as leading individuals to a 'shortened lifespan' discourse, resulting in what Gibson and colleagues called 'damaging effects' (Gibson et al., 2009). These damaging effects included the material, social and symbolic isolation of those individuals (ibid.).

Upon leaving the education system, individuals living with MD are said to often lack meaningful activity and vocation thereafter (Abbott & Carpenter, 2014). Abbott and Carpenter hypothesised that the dominance of a 'shortened lifespan' narrative makes planning ahead extremely challenging for the individual and their family. They

proposed that this discourse often creates barriers to quality of life for these individuals, as those in society make assumptions about what the individual can and cannot do in relation to a relatively out-dated understanding of the condition (i.e. death before adulthood). Not only this, but the individuals internalise these narratives, in a way not too dissimilar to internalised stigma (e.g. Bedini, 2000). In Gagliardi's ethnographic research, "the boys withdrew into their own worlds, apparently so as not to confront the limitations imposed by their illness [and society]" (Gagliardi, 1991a, p. 163).

1.3.4 Understanding disability in the context of society

Although some adversities experienced by families living with MD may relate specifically to the degenerative nature of the disease, society may also perpetuate problems when adequate adaptations are not made. Whereas medical discourses may describe the child with MD as disabled, social models of disability posit that the 'disability' should be reallocated and used as a descriptive of society, as opposed to the individual (Shakespeare, 2010). "Disability is viewed as a by-product of a society which is organised around only certain bodies which are defined as normative, in legislation, education, institutions and popular culture" (Oliver, 2004 cited in Spandler, Anderson, & Sapey, 2015, p. 140). Historically and arguably presently, such discourses of disability have dominated practices – leading to inequitable access and support (Spandler et al., 2015).

1.3.5 Muscle Dreams: Intervention in the community

In recognition of the social model of disability, some charities look to create a context within which a child with a life-limiting/threatening condition can participate in activities otherwise inaccessible due to societal constraints. One particular charity is the Muscle Help Foundation, founded by an individual diagnosed with MD himself. The Muscle Help Foundation deliver events that are personalised and tailored for young people diagnosed with MD called Muscle Dreams. Individuals are nominated, often by their parents or carers, and a process begins in which the Muscle Dreams team assess the individual's application based on their interests, history and ambitions. For example, experiences have included beneficiaries who have been enabled to use their journalism degree to interview famous authors and film stars, or to spend a day in the wilderness with a famous outdoor survivals expert.

The intervention begins with a process of the team getting to know the child and their family, familiarising themselves with their likes, dislikes and personality over time. Through this process, an event is designed that would enable the child to 'live their dream' without the barriers or obstacles they would usually face in the outside world. This involves things such as hotel beds being raised in anticipation of the family's arrival in order to ease the use of hoists, adequate transport arrangements, and so on.

Other experiences are delivered in small groups, where the young people can meet peers in similar situations from across the country, who share similar interests (e.g. motorcar racing). Throughout the day, the young people are provided with numerous gifts and surprise activities related to their activity. Each family is provided an assigned volunteer for the day, who gets to know the family, but also responds to any changing needs throughout the day. The Muscle Dream is then closed with a celebratory dinner,

whereby beneficiaries join one another to witness video footage of the day's events, followed by speeches that highlight the individual strengths and interests of each young person there on the day.

It could be said, that the delivery and fulfilment of that Muscle Dream promises an experience in which not only recipients, but their families, may reconstruct their identity in a potentially transformational context that stimulates and empowers. These novel and experiential events offer individuals the opportunity to bypass barriers of health, to access what the families had previously perceived as the unimaginable, and to stimulate their creativity and vision. Indeed, in one of the few papers published relating to wish-fulfilment experiences, Ewing (2009) found that such experiences offer an opportunity for an unified emotional, physical and spiritual experience, even if only temporarily. She suggests that it is through the appropriate psychological support for families that an opportunity to scaffold a sense of empowerment unfolds (Ewing, 2009).

Very little research exists in relation to such wish-fulfilment experiences. Shoshani and colleagues (Shoshani, Mifano, & Czamanski-Cohen, 2016) however, conducted a randomised control trial in the context of such interventions for children diagnosed with terminal cancer. Children were assigned to either a waiting list or to an individualised wish-fulfilment intervention, which was delivered 6 months later to 'fuel anticipation'. They found that five weeks after the intervention, children who had had their wishes fulfilled, had significant reductions in general distress, levels of depression and anxiety, and improved scores in health-related quality of life, hope and positive affect measures. Families often attend such experiences alongside their children, as both observers and participants in the day's events – but Shoshani and colleagues (2016) did not consider the familial experience alongside the children in this study.

1.3.5.2 Linking wish-fulfilment, enabling contexts and bereavement

Families are likely to shape meaning to such wish-fulfilment activities within the context of their own histories, memories and experiences of living with MD within the family context. The Muscle Dream experience for both young people and their families, who may witness and live their own 'dreams' alongside these experiences, remain significantly under-researched. Additionally, wish-fulfilment events could be considered to meet the needs of young people at two levels; enablement in disabling contexts and opportunities for families to come together in the face of the terminal nature of the disease. As such, wish-fulfilment events can facilitate engagement in activities for those whose context or disability makes such involvement difficult or impossible. Secondly, such events could be argued to offer hope, community, connection, and enjoyment when facing death. This is consistent with Ewing's (2009) proposal that such events operationalise emotional, physical and spiritual connection for individuals and the networks around them, then it not only seems relevant to consider such events in the anticipation of death – but also to consider how people make sense of such events beyond death. This research project looks to begin building an understanding of this, albeit in a preliminary fashion due to the Muscle Dream being one particular experience within a much broader story of parenting across the lifespan. However, this research presented the opportunity to explore whether the memories and meanings of such events are still retold or considered important beyond the child's death and if so, in what way or for what purposes are these stories retold.

1.4 Systematic literature review

Below, I elaborate on the search strategy employed in order to systemically review the relevant literature. Thereafter I describe each study in order to provide the reader with an understanding of the available literature on experiences of parenting a child with MD.

1.4.1 The scope of the systematic literature review

The scope of this literature was limited in part, due to the available timescales of conducting research within the context of clinical training. Additionally, narrowing the focus of the systematic literature search to only papers published using qualitative methods was strategic at a conceptual and epistemological level, with a view to establishing a clear understanding of what current literature there is that reflects the accounts, experiences and sense-making of couples parenting children living with MD. Therefore, a decision was made to conduct the systematic literature review on qualitative research only. This decision enabled a clear, yet narrow focus on the literature. However, this could be considered at the cost of gathering a broader view on the literature that is inclusive of the quantitative literature. In response to this risk, I felt that it was important to still consider the quantitative literature in order to contextualise the findings, and so additional searches were conducted alongside the systematic literature review. These additional searches were used to inform the presented research narrative and ensure that other important findings outside of the qualitative studies were also considered within the text.

1.4.2 Review strategy

A literature search was completed using the databases SCOPUS and PubMed on the 30th November 2016 and repeated on the 5th June 2017. These databases were supplemented by hand-searches, additional electronic sources (e.g. Google Scholar), snowballing through references and citations of obtained texts, and perusal of the 'grey literature' that in this case included unpublished manuscripts, blogs, articles and MD Associations[2]. The search terms included variations on *muscular dystrophy*; *parent, carer, mother/father;* and *qualitative*. Limits were applied only to exclude articles not published in English and biomedical literature.

Following a review of the titles of papers for their relevance to the current research project based on a set of inclusion and exclusion criteria, 111 papers were identified overall. The exclusion criteria included papers that were not available in English, based on different populations (e.g. older adults), and specific evaluations of particular interventions (e.g. peer-support groups). After excluding the remaining duplicated papers and reflective articles from the review, a total of 13 papers were reviewed in depth (see Appendix A for further details). The full breakdown of this process of identifying papers is described in Figure 1.

---

[2] E.g. Muscular Dystrophy UK (http://www.musculardystrophyuk.org); MDA (https://www.mda.org)

Figure 1. *The systematic literature review results.*

SEARCH:

Records Identified and retrieved through database searching

(Scopus n = 50)
(PubMed n = 54)
Overall = 104

Further records identified through citation search on google scholar & reference screening

(Google scholar n = 11)
(Reference checking n = 14)
Overall = 25

Duplicates n = 18

87 titles screened following removal of duplicates

N = 111

Reasons for exclusion (N= 63)
Not published in English.
Research not focusing on lived experience of living with a child diagnosed with muscular dystrophy.
Research/methodology reviews.
Quantitative methodology.
Unrelated condition or population.

Articles selected for review of abstracts

N= 48

35 papers further excluded

Reasons for exclusion:
Studies evaluating interventions.
Participants are those diagnosed, or health care professionals.
Same study published twice.
Review paper.

Articles selected for full text review and quality evaluation

REVIEW: N= 13

1.4.3 Literature review

Thirteen qualitative papers were reviewed in detail and provided me with a further understanding of what it can be like for families when parenting a child living with MD, particularly in terms of some of the challenges faced. In short, the parental experience of

living with MD was often described in relation to the disease-specific challenges faced, how these challenges were approached and overcome, and the emotional and psychosocial impact that these had on individual parents and their identity as a couple. Indeed, parenting a child living with MD was said to equate to "a long, isolating, uphill struggle" (Gravelle, 1997, p. 739). Below, these thirteen papers are reviewed in detail relating to a range of particularly pertinent moments on a parent's journey: from diagnosis; to disease progression and daily life; to deteriorating health and death.

For many parents, the first specific challenge in terms of MD is in identifying that there may be some physical and developmental difficulties for their child (Daack-Hirsch et al., 2013), followed by challenges in seeking and obtaining an appropriate diagnosis (Bendixen & Houtrow, 2016).

1.4.3.2 Diagnosis

Daack-Hirsch and colleagues (2013) explored parental perspectives on the diagnostic process for MD in depth and identified several practical factors that either enabled or hindered access to diagnosis and support. They interviewed 25 families overall using telephone and group interviews, from MD clinics and the MD Association in the USA. Firstly, they analysed the data in order to identify themes, and then used these themes to populate the Stages of Help-Seeking Model (Eiraldi, Mazzuca, Clarke, & Power, 2006), a pre-existing model they deemed applicable to families seeking diagnosis for their sons.

Throughout their findings, Daack-Hirsch and colleagues (2013) propose that early-on in their child's life, parents oscillate between identifying problems, and then seeking help, as they attempt to understand what development is to be expected or not, and at what age. As a result, parents often experienced long delays or repeated similar stages and consultations– but would constantly remain focused on problem-resolution and 'doing more' for their child. From their findings, they suggest that the psychosocial impact of seeking diagnosis and support cannot be so readily disentangled from these pragmatics of 'doing'. For example, they conceptualise 'denial' and stigma as factors that inhibit parents moving onto subsequent stages of help.

Like Daack-Hirsch and colleagues (2013), Bendixen and Houtrow (2016) found that the parental experience of securing a diagnosis is linked to the practicalities of help-seeking and service utilisation. In their study, Bendixen and Houtrow (2016) interviewed 15 parents who were already involved in ongoing studies at academic institutions in the USA. These semi-structured interviews over the telephone were subsequently analysed using thematic analyses, with their findings highlighting some of the interpersonal and relational experiences involved in the diagnostic process.

Bendixen and Houtrow (2016) present an overarching theme of a 'communication breakdown' that emerged from their data. They described how services paid inadequate attention to concerns raised by the parents in their sample, which lead to further delays in appropriate diagnosis. The parents interviewed described feeling their concerns were dismissed by professionals; professionals who often lacked the adequate knowledge-base to identify, perceive and understand MD symptomology. Parents reported feeling that staff were careless in their delivery of the diagnosis, and that post-

diagnostic support and advice was often not communicated adequately. As such, the parents in Bendixen and Houtrow's (2016) study, reported becoming increasingly distressed in relation to these challenges in seeking help.

Both papers discussed above led the authors to suggest the need to create further opportunities for parents to be heard within services. This appeared to become a common thread throughout all of the literature in terms of clinical implications, at whatever stage of the disease progression. Now I will turn to the papers that address subsequent challenges following diagnoses, as the parents and their child face adversities over time relating not only to the disease, but to general developmental stages in life.

1.4.3.3 Disease progression and daily life

Adversities faced by families in relation to the degenerative trajectory of MD not only include how affected the child becomes by the condition and how quickly it progresses, but also factors relating to their developmental stage, and transitions faced by any child growing up (Gravelle, 1997). Gravelle (1997) interviewed 8 mothers and three couples conjointly, meaning that eleven parents were interviewed in total. She initially used an unstructured interview format, which was followed by a semi-structured interview to follow-up on themes from the preceding interview. She recruited parents through two health centres and an MD Association in Canada. Gravelle (1997) used a phenomological analysis to develop a conceptual model of defining and managing adversities throughout the parental journey.

In Gravelle's (1997) sample, parents described an acceptance of the inevitably of deterioration and further challenges, in a way that reflected a pragmatic need to respond without too much worrying. Despite this, with each new situation or challenge for their child, parents reported experiencing a degree of loss themselves. This on-going and adaptive process led to many parents describing how they gained personal strength over time and became more resilient. Some losses were reportedly more significant for parents, such as when their sons could no longer walk and as a result, required a wheelchair – requiring significant renovation or a complete move out of the family home.

Notably, the parents described a position of dissonance, as they felt great sorrow in no longer being able to 'deny' their child's disease prognosis, but also a time of optimism and success – as some would reframe the loss of embodied mobility with an improved mobility, through physical aids such as mobility scooters. Reportedly, this dissonance of loss and freedom appeared to reflect the nature of many of the challenging transitions faced by families living with MD – through managing such adversities, many families appeared to create opportunities together.

Making sense of loss through facing and adapting to adversity in the context of hope appeared to be an important concept for the parents interviewed in Samson and colleagues' work (2009). Their sample included 12 families in total, interviewing nine mothers and then three couples conjointly. Recruiting from a neuromuscular clinic in Canada, Samson and colleagues (2009) used a phenomenological analysis to interpret their data and presented stages of hope and coping.

Reportedly, the parents in Samson and colleagues' (2009) study described initially facing MD as a threat to their child and their way of life, which they adapted to in a passive way, coping through the hope for an eventual medical cure. Through learning to adapt however, the parents talked of noticing an increasing sense of feeling different from others in society, feeling isolated over time. Despite their reported loss of social networks, the parents described how they redefined 'normality' as a response to this loss. Indeed, these parents reported how the illness became incorporated into their daily life and that their hopes became focused on the 'here and now' pursuit of positive and tangible effects on their child's wellbeing.

Beyond the disease, Samson and colleagues (2009) reported that the interviewed parents described how they facilitated and nurtured their child's growth as a person, rather than being overly distracted by caring tasks. These parents described how they would give new meaning to the disease experience, and that the child's developing sense of individuality over time became a great source of hope for those parents interviewed.

As muscle-wastage occurs over time, ominous physical reminders of the disease's terminal nature reportedly become more frequent, leading some parents to look ahead and make preparations for the future, requiring continuous revisions and adjustments to their plans (Dawson & Kristjanson, 2003; Gravelle, 1997). Dawson and Kristjanson (2003) interviewed a total of 16 carers of individuals living with MD and Motor Neuron Disease. Eleven carers from this sample were parents of children with MD, with three retrospective interviews in relation to the loss of a child. They recruited their sample from a MD clinic in Australia. They used a content analysis and the constant-comparison

approach, identifying three core experiences relating to reactions and responses to challenges; health system 'crossing points'; and 'reaching forwards'.

Dawson and Kristjanson (2003) found that families faced losses on an on-going basis, daily. Reportedly, this resulted in a great deal of anxiety for the interviewed parents. With great uncertainty about the reversibility of a crisis, some parents reportedly managed adversity through avoiding contact with health care professionals and hospitals altogether. They described how the interviewed parents experienced points of contact with health care professionals as struggles and 'fights' for what they needed; characterised by varying degrees of trust, tension and inconsistencies in information-giving. It is in this context, they say, that parents often appealed for assistance in planning for the future.

For the families in Gagliardi's ethnographic studies (Gagliardi, 1991a, 1991b), it emerged that families reported to go through a process of recognition to working things out, and identifying resolutions over time. Gagliardi (1991a, 1991b) joined three young families over a ten-week period. She visited weekly, joining the family for a range of activities and also interviewed parents two times during the period of being immersed in the family, and one final interview a year later.

Gagliardi (1991a, 1991b) described how the parents in her study were faced with their child experiencing increasing levels of pain over time due to physical deteriorations, leading them to a state of disillusionment. By this, she described a sense of normality was challenged, and the parents became increasing angry, felt cheated, and saddened by their child's situation. She also noted that families reported how they experienced

society to confirm 'the impossibility of normalcy' through reminders of difference, and inaccessible activities and environments.

Through 'trying to work things out', the parents described how they developed and imposed rules and boundaries in order to regain 'control' over their world, which reportedly became smaller and smaller as the disease progressed. Such rules for example, included to never speak of the condition within the household. Reportedly, families were then faced with the option to 'let go' or 'hang on' – as their sons became more self-sufficient and no longer needed the same degree of protection required before. The dilemma to let go but lose one's caring identity, or to hang-on but prohibit their child's independence, was reportedly resolved through parents becoming increasingly involved in the charitable sector and establishing a community around those with lived experiences of parenting children with MD.

Some of these on-going changes, losses and challenges were addressed by Webb (2005), whose work explored the ways in which parents coped with caring for a child living with MD over the lifespan. She interviewed 15 families in total, of who seven were conjoint couple interviews. She recruited her sample from the internet and a conference, in the USA. She used grounded theory to analyse the content of these interviews. Similar to the findings of Daack-Hirsch and colleagues (2013) and Bendixen and Houtrow (2016), parents reportedly struggled to access diagnoses and support, and when they did, would often react with fear, anger, guilt or 'denial' of the situation (Webb, 2005).

The parents in Webb's (2005) sample reported that they became discontented with the treatment they received from hospitals and schools during their journey, and described a significant struggle to acquire the necessary equipment to assist their child. Indeed, Webb (2005) reflects on how parents should be supported by health care professionals in trusting their instincts early-on, by anticipating a grief-like response to diagnosis, and improving collaborative efforts to obtain assistive-technologies and in learning how to use them together.

The familial experience when children transitioned into adulthood was addressed by Yamaguchi and Suzuki (2015), who interviewed 18 individuals in total, including four fathers. They recruited from a specialist hospital and three MD Associations in Japan, and analysed their findings using grounded theory. They found that parents reported how they provided all care at first, but that over time they had to 'hand-over' the management of physical and medical support to their children. The parents in this sample did not wish to intrude upon their son's care, but maintained a working knowledge of the condition in anticipation of having to 'cover' care at any given point (i.e. to back-up).

Yamaguchi ad Suzuki (2015) detailed how parents described trying to sympathise with their son's situation, recognising that they were susceptible to embarrassment, disappointment, irritation and anxieties due to the challenges and limitations that were faced. In their analysis however, they identified three problems in parents 'becoming backup carers' in that psychological support was rarely offered to parents, that the parents themselves were likely to experience age-related and changing health needs

over time, and that parents would often wish to intervene in their child's decisions, particularly around end-of-life care.

Earlier studies addressing the lived experience of parenting children living with MD, appeared to look specifically at the difficulties associated with facing and adapting to the adversities linked to the degenerative condition. Witte (1985) interviewed what appears to be four families of adolescents diagnosed with MD, with two families each having two siblings both diagnosed with MD – although the sample is not explicitly described in detail. Witte (1985) recruited these families through a special educational needs school that the young boys attended in the UK. He does not describe the method of analysis, but appears to use a phenomenological approach informed by psychoanalytic interpretation. Reportedly, parents in this sample described how their sons withdrew from unfamiliar environments and social groups, fearing that they would not be accepted. Witte (1985) hypothesizes that this could relate to feelings of shame about one's condition, and detailed how the parents reported to not enjoy interacting with the 'outside world' as a result.

Witte's (1985) analysis reported that parents would avoid conversations around the terminal nature of MD and 'detour' discussions around what could happen in the future. As such, he goes on to describe how "not to address their grief with each other [was] to ensure 'normal' family functioning. [This then] condemns each family member to live a guilt-related, yet alienated affect-deadened life in and outside of the family" (Witte, 1985, p. 185). Witte's (1985) lack of transparency in his methods however, warrants that his findings are viewed with caution – particularly as psychoanalytic concepts

appear interwoven with emerging themes from the data, with no acknowledgement within the text.

An even earlier study by Buchanan and colleagues (1979) interviewed 23 mothers and two fathers, recruited from and conducted within a MD clinic in the USA. They do not detail their analytical approach to the data, but appear to use a descriptive approach. Buchanan and colleagues (1979) reported how the parents within their sample described using particular coping strategies to manage the adversities faced. They detailed that parents would use 'denial', 'magical thinking' and 'over-protection' in order to cope but that this would have a negative impact on their own mental health – with 76% of the parents interviewed identifying a psychological problem as their primary concern in having had to adjust to a life of living with MD. They described how parents would 'deny' their emotional reaction to the initial diagnosis, would seek alternative diagnoses and could only discuss factual matters. Buchanan and colleagues (1979) described how parents would see their sons as different to other children diagnosed with MD, becoming convinced the disease would not progress as quickly as others, or that they would eventually recover from it ('magical thinking'). In describing how some parents would 'over-protect' their child, Buchanan and colleagues (1979) noted how parents would isolate their children from the world, often in ways that seemed maladaptive and controlling. They concluded that the most 'well-adjusted' families involved an open and frank communication style between the parents, who were orientated towards accomplishments and available activities, had support networks outside of the family, and had regular and routine forms of recreation for the parents themselves.

1.4.3.4 Gendered stories of parenting a child living with MD

Significant differences in the care-giving role of mothers and fathers have been found across multiple domains, such as adaptation, coping, involvement of care, and psychosocial factors and wellbeing (Katz, 2002). Much of the MD literature indicates that the primary care-giver role disproportionately sits with mothers (Dawson & Kristjanson, 2003), which suggests that gender and gender roles may play an important part in the experience of parenting children living with MD. Tomiak and colleagues' (2007) sample included 11 parents, interviewing five couples separately, and an additional interview with one mother (following the father's decision to withdraw from the study). One interviewee reported on their experiences of parenting a daughter with MD. They recruited the sample from a neuromuscular clinic in Canada.

Tomiak and colleagues (2007) used a phenomenological analysis to explore gender differences in greater detail, and identified five areas in which fathers and mothers reportedly differed in their care-giving. These areas of difference included in the care of the child, their coping styles, relationships with their partners, their career, and their social support networks. Reportedly, most differences tended to relate to the respective roles of primary or secondary caregiver status, which with the exception of one relationship, involved mothers being the primary caregiver. Fathers would reportedly oversee family functioning, provide psychological support and 'moral direction' to their families, whereas mothers would provide direct physical care to their children.

At first, parents were said to perceive differences in their approaches to caring as threatening to the parental relationship, serving to isolate each of them from one another. Mothers were reportedly more likely to experience emotional distress than

fathers, whereas fathers were more likely to experience couple-related distress. Fathers in particular, were reported as being more likely to become distressed in response to the stigmatisation of their child's condition. Mothers were reported to have little or no other choice than to become the primary-caregiver, and it was suggested that many perceived the role to be a moral obligation of being a mother. However, frustrations and resentment were also reported to build up over time, as "the sacrifices women make often go unrecognised" (Gravelle, 1997, p. 744). However, over time, parents reported that they would adapt to one another's differences – often enriching the relationship due to both offering different skills and qualities that aided their situation (Tomiak et al., 2007).

Cunniff and colleagues' (2015) work on *Listening to Fathers*, specifically explored roles and experiences of fathers in parenting a child with MD. They interviewed 15 fathers in total, and a further 55 fathers responded to a 'written comments sheet'. They recruited their sample from four national charity organisations, and used a grounded theory approach to analyse the emerging data. They reported on father's accounts being unique relating to experiences of loss and acceptance, support (versus isolation), fighting for resources, and the importance of 'a race against time'. The fathers in their sample described how transitions were particularly challenging, with previously held hopes threatened by impending changes, particularly early expectations around what becoming a father would look like and what the role would involve over time.

One participant in the study coined the term 'issue fatigue', a phenomenon that Cunniff and colleagues (2015) suggest describes a dilemma of perpetual demand faced by fathers to deal with things, yet having very little time to manage anything else (2015, p.

11). Reportedly, these Fathers felt isolated from the routines that mothers had established over time with their children, and felt unneeded and unwanted in consultations hosted by professionals. At the same time, the fathers described a desire for speed, with appointments not seemingly being offered quickly enough.

The fathers in the study also reported aspirations and plans to 'cram in' as many activities for their children 'before it was too late'. Cunniff and colleagues (2015) suggest that this may be because the fathers were found to live in constant anticipation of the next loss, leading some to 'self-protect' through not becoming too close to their sons. For example, these fathers were reportedly forced to re-evaluate their previous expectations for 'continuing the family name' when faced with the terminal nature of MD.

1.4.4 Quality evaluation of the literature

In assessing the quality of the 13 papers included for systemic review, Elliot, Fisher and Rennie's (1999) criteria for evaluating the quality and credibility of qualitative research were used. Examples of the details of the evaluation are provided in Appendix B. In assessing the quality of published research, Elliot and colleagues provide seven aspects common to high quality quantitative and qualitative research and then a further seven aspects that specifically relate to the quality of qualitative research. I briefly summarise an overview of the quality of the papers included in this review, with a particular focus upon those specific to qualitative research.

1.4.4.2 Guidelines for publication of qualitative and quantitative approaches

All papers reviewed were explicit in addressing the scientific context and purpose of their studies. Appropriate methods were utilised by most researchers to address their aims and most explicitly stated their approach towards respecting their participants through ethical considerations. However, it is noteworthy that the two studies that did not address ethical standards nor detail their methods or analyses, were the most dated papers included for review (Buchanan et al., 1979; Witte, 1985). This may reflect changes in publication procedures and improvements in the reporting of ethical standards over time. All papers included in the review produced appropriate discussions that reflected the research context and findings, with one exception. Throughout his discussion, Witte (1985) appears to employ a psychoanalytic approach to interpreting the data, with no reference to this and no apparent tentativeness in his conclusions. All papers were presented using variations of conventional formats for scientific publication.

1.5 Rationale and aims for the study

1.5.1 Rationale for the study

The literature on parenting a child living with MD is broad in its scope, considering the familial experience of facing and overcoming adversities relating to the degenerative nature of the disease. As observed in the literature, experiences of family members adjusting to diagnosis, changes in mobility, and the dying process, are reported often with a notable psychosocial impact on the individuals involved. What is evident from the literature, and described more fully in the following chapter, are key omissions in the understanding of more situated and contextualised experiences of parenthood.

Explicitly, much of the literature purports to describe parental experiences, and yet often only interview mothers or fathers separately. The qualitative studies explored used a range of methods, but none used narrative inquiry, which has the potential to elicit situated and contextualised understandings of how parent's account for their experiences. All studies addressed specific periods of the disease trajectory, such as diagnosis or end-of-life care, and so enable an enriched understanding of a particular aspect of the parenting journey – but have inherent limitations to understanding the entire parenting story, its role and its relationship to identity within broader narratives. Some of the studies addressed death, and few included retrospective interviews within their sample, but none addressed this or life after the death of the young person. There also remains a lack of exploration and understanding around the delivery of wish-fulfilment events such as Muscle Dreams, particularly the familial experience. Indeed, although some of the studies employed ethnographic methods to research, there was no consideration to the 'live' co-constructing and recounting of experiences, and what this would mean for the parents, the nature of the research findings and what the clinical implications of this would be.

In short, the literature on the experience of parents within this lifespan and relational context of being a couple, as opposed to individuals, has been rarely considered and remains largely unexplored. To meet this research need, I considered narrative inquiry to be an appropriate route to contribute towards these noted omissions within the literature.

1.5.2 Aims of the study

As described in the preceding rationale and as highlighted throughout the literature, it seemed important to consider how partners account for and make sense of living with MD across the lifespan in the context of their relationship (living, experiencing, understanding and retelling together). As such, the core aim of this study was to explore the narrative of parents who have lost a child to MD. In this context, the research addresses multiple stories retrospectively, as parents develop their narrative accounts through the retelling of their experiences from before diagnosis to beyond death.

Therefore, the research question asked:

> *In the context of a delivered Muscle Dream, how do parents who have lost a child to*
> *Muscular Dystrophy story their experiences of parenting and the loss of their child?*

Within this broad question, there are several aspects of further inquiry that the research aims to elaborate on:

- When invited to narrate the parenting story within the context of children who had died, what seem to be the important aspects of the parenting experience that are used to create such accounts?

- In what way is the impact of events such as the Muscle Dream experience told by parents who have witnessed their children participate? For example, are these retold in a way that suggests short or longstanding individual and family 'impact'?

- In what ways do parents narrate their collective experience as a couple?

In answering these questions, I have paid particular attention to the research design and analytical procedures in order to provide an overall research narrative that provides insight into the how couples who have lost a child to MD narrate their parenting journey. The following chapter details the methods in which this has been made possible.

Chapter two: Methods

2.1 Overview

This is an exploratory study using qualitative methods. Specifically, I used narrative inquiry to explore how parents who have lost a child to muscular dystrophy story their experiences of parenting and the loss of their child, in the context of a delivered Muscle Dream. In this chapter, I will first present an account and justification for my choice of methodology, leading to the decision of narrative analysis. Thereafter, I then describe the design of the study with particular consideration to service-user consultation, ethical conduct, and the quality controls employed in the current study. I conclude this chapter with reference to the role of researcher and my participation in a reflexive-researcher approach.

2.2 Qualitative research

The way in which clinical psychologists conduct research has changed over the course of history. Llewelyn, Hardy and Aafjes-van Doorn (2015) analysed papers published between 1952 and 2012. They found that earlier studies tended to be more theoretically orientated, focused on conceptualising diagnostic concepts or discrete categories of personality, and focused on the psychometrics of cognition and personality.

Historically, positivist approaches have sought to identify causal factors to observable phenomena and have dominated much of the research within the field (Kirkman, 2002). In their review, Llewelyn and colleagues (2015) note that there is an increasing use of qualitative methods in clinical research in Britain. Most qualitative research abides by the notion that there is no objective or universal truth that we can identify and is

underpinned by quite different epistemological assumptions (Lyons & Coyle, 2016). As such, qualitative research explores and elaborates on the lived experiences, perspectives and meaning of phenomena in particular contexts (Andrews, Squire, & Tamboukou, 2013).

Qualitative research is particularly useful in under-researched areas (Barker & Pistrang, 2015). Within the context of MD, a range of qualitative methods have been applied – particularly the grounded theory method (e.g. Cunniff et al., 2015; Yamaguchi & Suzuki, 2013). However, in the context of the 'wish-fulfilment' context (I.e. Muscle Dream), to my knowledge, no qualitative research has been conducted previously, with the exception of another project recently completed by the same research team (Nolte, Ragavan, Randall-James, Wellsted, 2017). Additionally, qualitative accounts of parenting have traditionally meant the accounts of individual interviews with one parent, usually the mother (Tomiak et al., 2007). Rarely have qualitative studies interviewed parents conjointly, and to my knowledge, never in the context of a narrative inquiry and analysis.

2.2.2 The case for narrative inquiry

Narrative inquiry presupposes the centrality of language in how we understand social phenomena and create meaning (Riessman, 1993). In social constructionism, the process of narrative formation could be argued to represent the phenomenon of interest in and of itself. Some quantitative and positivist approaches could also recognise the centrality of language in their approaches, but look to control as many

variables as needed to limit the relativity introduced by relying on our communicative tools. For my critical-realist stance, I accept that language plays an essential and *central* part to how we understand phenomena, but also to how we position language as *a tool* in which we can progress towards an understanding of a tangible world around us, and not necessarily an end in itself. That world is layered and nuanced by a complex web of relational, social, discursive and embodied processes and interactions that we may only ever approximate such a reality (as explored previously). More aptly put, through language we co-construct narratives, and through narratives we can reflect, and reflect on the world and the experiences lived within it.

The medium of communication is varied, with narrative taking the form of spoken word, text and images, sometimes sourced historically or created in current contexts (Andrews et al., 2013). It is largely agreed upon that narratives constitute an ordering of events into a coherent sequence, typically following a chronological order (linear time) that provides explanation or meaning (Gergen, 2009; Wells, 2011). Squire (2008) defines what she calls an 'experience-centred' narrative approach as one that focuses upon human experience, assuming that narratives 're-present' experience in sequential and meaningful ways – often reconstituting it and expressing it in a multitude of ways. As such, narrative inquiry can be multifaceted to reflect the different means of re-presenting human experience, which is reflected in different approaches to data collection and analyses (Wells, 2011).

Ultimately, Squire (2008) suggests that narratives display change and transformation. Such transformational narratives then, do not solely relate to an ordering of events, but have intricate implications for how we see ourselves and the world (i.e. implications for individual and collective identities) (Wells, 2011). For the process of constructing a

coherent sense of identity, individuals story their experiences and piece this together into a storyline – however, this may vary depending on their perceived audience (Murray & Sargeant, 2011). Indeed, "parents are not giving either 'true' or 'false' accounts, but how, faced with talking to a stranger about potentially quite personal things, they produce 'morally adequate' accounts and stories" (Baruch, 1981, as cited in Abbott, 2012, p. 244).

Based upon this understanding of how narratives can be studied to understand human experience, identity and transformation in meaning, narrative inquiry is consistent with my epistemological position presented in Chapter one. Namely, that narrative represents a co-construction (Speed, 1991) and data as a 're-presentation' (Squire, 2008), in which narratives are irrevocably contingent on time, place, occasion, narrator and audience (context) – emphasizing interview talk as 'purposeful social action' (Chamberlain & Thompson, 1998). Narrative inquiry is "trafficking in human possibilities rather than in settled certainties" (Bruner, 1986, p26 as cited in Hydén, 2013, p. 138) in a way that enables the creation of social and clinical opportunities.

The current study used purposefully produced speech as its medium for narrative inquiry and so in this context, will allowed for exploration of the experience of parenting a child who had lived with MD. Whereas other qualitative methods tend to address the content of speech, narrative inquiry enables an exploration of not only the *what* (content), but also enables the researcher to address *how* (structure, performative) and *why* (sense-making, meaning) narratives are presented in such a way, and to also consider the purpose of such talk (Reissman, 2008).

At the same time, narrative analysis allows for wider discourses to be considered throughout – an important concern if we consider that many of the ways we understand

our experiences are contingent on the dominant discourses at any given time (White & Epston, 1990). Considering stories of parenting a child who lived with MD then, narrative analysis can be used to understand the societal and discursive context in which parents make sense of events and imagined futures, what this means for them (e.g. challenges, changes, transformation), and what it is like for them as a couple, and as individuals.

2.3 Design

2.3.2 Service-user consultation

Service-user involvement in research takes many shapes and forms. One cited reason to involve service-users throughout research procedures relates to a commendable desire to move away from disempowering and expert-dominated contexts, towards more civically responsible endeavours (Shippee et al., 2015). The word commendable here is purposeful, as such a drive within research resonates with me personally. I really learned the value of this when I conducted a research project involving service-users diagnosed with dementia in participating in systematic literature reviews (Fisher, Kelly, Lawrie-Skea, Randall, & Wade, 2014). In addition to a more egalitarian research context, service-user involvement also helps to improve, or at least provides checks on the quality of design, the applicability and acceptability of proposals, and advice on appropriate means and methods for dissemination (Morrow, Ross, Grocott, & Bennett, 2010).

The overall project described in this thesis was completed over a period of two years. I conceived the idea for the project following a presentation by a representative of the

Muscle Help Foundation and so wrote a project pitch not too long after this. Having received a two-page summary of my ideas for the project, my supervisors and I met with a team from the charity. It was at this meeting that we discussed the challenges and opportunities within the project, invited the group's feedback about, and advice around working with, and researching in the area of MD.

This meeting was an important moment of service-user consultation, based not only on the content of the proposed project. One representative had asked if I had ever met a young person diagnosed with MD before, which I had not. In asking this, they opened up a learning context in which I could understand and experience the importance of my physical presence, mannerisms and habits, even before an interview had taken place. For example, this addressed the importance of considering the implications of an offered handshake, which not all young people would be able to engage in. Likewise, another representative offered their reflections on the family and carer experience – both from their personal perspective and those families that they had befriended during Muscle Dream events. This meeting formed the basis for three further meetings over the first year of the project, developing a shared and collaborative learning environment, whereby I as the researcher in particular, could consider both content and process in the ongoing project. This included consultation on the research procedures and design of the interview schedule.

In order to familiarise myself with the Muscle Dream experience and the lived experience of young people and their families living with MD, I also attended a Muscle Dream event as a volunteer. This experience enabled me to witness what such opportunities could create for families, to converse about the challenges faced outside of this context, and hear the hopes that these families held for the event. Alongside this,

I observed the community spirit fostered throughout the environment as many young people living with similar challenges were facilitated to engage in activities otherwise inaccessible, and share that experience with others in similar circumstances. I was able to observe the intricacies of such events, such as Muscle Warrior salutes; physical gestures with one's body that symbolises comradeship and being stronger together – an embodied action that appeared to become a communal identity among beneficiaries and abled-bodied allies. There were particular narratives conveyed, such as the precise number of 657 Muscle Dreams to be delivered to match the number of muscles within the body. This ethnographic component to the study enabled me to become immersed in the Muscle Dream experience, and to consider my own 'outsider witness' position. I describe this as an outside witness position because it is equivalent to being an audience to this therapeutic event; "to listen to and acknowledge the preferred stories and identity" (Carey & Russell, 2003, p. 1) of these young people. For me, I considered this an experiential consultation of sorts – becoming immersed and familiar with the relational and potentially transformational reality of not what only takes place, but what this could mean for individuals and families.

A pilot interview was also conducted with a bereaved mother and father in order to trial out procedures, and to seek feedback on the acceptability of interview questions. The pilot interview offered a similar experience, as the interviewees shared story books and poems they had written in relation to their experiences of parenting, that enabled me to try and develop a sensitive awareness to the emotional content my interviews were likely to activate. Although the ethnographic elements described here were not explicitly used to develop the interview procedures or schedule, these experiences

allowed me to contextualise my reading and vision for the project, and consider the challenges and opportunities that could be described to me in the interview context.

2.3.3 Sampling strategy

Purposive sampling was used in order to recruit parents who had experienced the death of a child to MD. In this context, purposive sampling was appropriate as it focuses on only a particular selection of a population and so can be strategic, in order to explore specific experiences in greater depth.

As this research aims to explore parental accounts of living with MD, following the death of a child, the following inclusion criteria were applied:

- To be parents[3] of a child who was diagnosed with MD;

- For the child and family to have been beneficiaries of a Muscle Dream event;

- For the death of the child to have been at least 8 months from first point of contact in research (see Section 2.4.5);

- To be fluent in English[4].

---

[3] For the purposes of this research, parental status was considered a self-assigned identity, independent of legal, biological or social conceptions of parenthood (Meyer, 2006).

[4] Narrative inquiry focuses upon the use of language in co-constructing accounts of events, identity, and meaning. With the centrality on the subtleties and nuances of how language is used and presented, and my own limitations in only speaking English, on this occasion it was not possible or ethical to commit to interviews in other languages.

2.4 Ethical considerations

In this section, I will briefly describe some of the ethical considerations made during the process of developing and conducting this study. In making such provisions, ethical approval was subsequently granted by the Health and Human Sciences Ethics Committee with Delegated Authority, the University of Hertfordshire (Appendix C).

2.4.2 The ethical starting point: Ethnography

This research project touches upon an emotive, sensitive, and potentially distressing topic area. Due to my relative inexperience within the field of MD, I felt that I needed to familiarise myself with learning about and witnessing aspects of the lived experience of parenting someone with MD before conducting any interviews. As previously mentioned, I participated in some ethnographic fieldwork (i.e. Muscle Dream event), which provided me with the opportunity for in-depth, yet experiential consultation. I considered this experience and the pilot interview to equate to a 'warming of the context' (Burnham, 2005, pp. 4-5), enabling a 'readiness' to witness the accounts that families hold to be of great importance. I consider this a genuine and ethical starting point for the project, to understand at some level the lived experience in which you are interested in researching – not necessarily in an intellectual sense, but in a felt and experiential sense. Indeed, this position fits with 'ethical mindfulness' within research practices (Guillemin, McDougall, & Gillam, 2009).

2.4.3 Explaining the research

Individuals who expressed an interest in participating were provided with an information leaflet containing the aims of the research, what participation would involve, and their rights to withdraw at any time (see Appendix D). The process of explaining the research project was gradual and graded over time, to enable families to make a decision that was right for them at that time. This is fully described in Section 2.5 on the research procedures. In short, details about the research were disseminated through press releases prior to recruitment; families were written to in order to provide space and time to consider the project and to note any questions or concerns. Then I contacted potential participants over the telephone in order to respond to any queries, and on the day of any interview, I revisited the research aims, checked-in about and answered any questions, and ensured that interviewees were equipped with all of the information they would need in order to make an informed decision about whether to participate or not. All potential participants had contact details for both myself and the charity, should they have wanted to seek further information or ask any questions at any point throughout the process.

2.4.4 Confidentiality and consent

Potential participants were provided with detailed information relating to the research project, in order to help them make an informed decision as to whether they wished to be interviewed. Participants signed a form to indicate their consent (Appendix E). Crucial to this information was how their personally identifiable information would be stored and used. Confidentiality was maintained through anonymity from the point of

transcription. A transcription company was used for some of the interviews and so a confidentiality agreement was signed (see Appendix F). Raw data was stored on an encrypted device and participants were offered for this data to be destroyed immediately after transcription. All personally identifiable data was anonymised and individuals were allocated pseudonyms for ease of reference.

The consent process included details about how any information gathered could be used thereafter, with safeguards described in order to aid individuals to make an informed decision about their interviews use. Beyond the current remit of this thesis for example, there is a potential for confidentiality to *not* be maintained through the release of some video footage. This was fully approved by the Ethics Board and further details are included in the Appendix G*.* Participants will continue to be consulted beyond the research and be able to opt for their data to be destroyed at any point.

2.4.5 Potential distress

Interviewees in this study were parents who have gone through the Muscle Dream experience, but experienced the loss of their child to MD since. As an ethical safeguard, only those who had experienced a death 8 months ago or longer were invited to participate (as was found advisable in Hynson, Aroni, Bauld, & Sawyer, 2006). The justification for this was that any earlier would be intrusive and will not have allowed the families time to start to process their loss. As such, this measure was upheld as a preventative strategy to not intrude upon grieving families and so as to not unnecessarily distress potential participants by invite to the project.

Bereaved parents will often experience unexpected changes in emotion during interviews and so it is important to anticipate and prepare for these moments (Dyregrov, 2004). The context of a research interview may also enable parents to search for and rediscover a closeness with their child in a safe context (Hooghe, Neimeyer, & Rober, 2012), which without participating in research is an opportunity that many parents do not have (Dyregrov, 2004). As such, during the ethics application I produced a protocol for how I would manage heightened distress within the context of the interview (see Appendix H). Below, I detail some of the ways in which this risk was mitigated:

- I was well placed to managed such incidents due to clinical practice and experience in conducting research projects across a range of complex situations (e.g. carers of those diagnosed with behaviours that challenge in dementia). As such, in designing the protocol I was able to include a range of responses when an individual or couple would become distressed. One example was that I used my body language and silence to acknowledge and not shut-down distress too readily (e.g. by stopping the interview at the first signs of distress).

- As detailed above, the recruitment of participants was also designed in a way that sought to empower families to make the decision that was best suited for their circumstances and wishes. As such, we used a sensitively written letter of invitation, providing time for families to consider the project together before any further contact, and ultimately, provided choice of interview time and location (Hynson et al., 2006).

- Throughout information leaflets and conversations with potential participants, they were reassured that a 'no questions asked opt-out' was available at any point in the research (Hynson et al., 2006).

- The chosen method of narrative inquiry is also consistent with the ethical proposition that bereaved parents wish to engage in 'unhurried' interviews, delivered in a less clinical manner, that allows the parent to lead the pace and direction of the interview (Hynson et al., 2006). As will be described below, the interviews were designed to meet these ethical standards without impeding the credibility of the research.

- Couples in grief are said to regulate not only how close or distant they feel to their child, but also to aid one another in this process too (Hooghe et al., 2012). By regulating one another's emotions, they are said to 'contain' their partner when they appeared to be getting too close to an 'emotional core or sadness' – but also to provoke their partner when they were perceived to be too distant (ibid.). Indeed, in their case study, Hooghe and colleagues noted that the couple "kept an eye on each other and guarded each other's boundaries…When one partner noticed that the other was too close to the emotional core and was having a hard time with it, he or she tried to create some extra space, for instance by taking over the conversation, so that the partner could recover and move back again" (Hooghe et al., 2012, p. 8). Through the composition of joint couple interviews, it was my hypothesis that couples were able to regulate one another's emotions in the interview context, in addition to the strategies I employed to maintain a safe and containing interview space.

- It was anticipated that individuals and/or couples interviewed may wish to seek further support or counsel after their interview. As such, a participant debrief

leaflet was compiled that included helpline addresses, links to information/advice leaflets, access to online communities and support forums (see Appendix I). Individuals were also advised to seek advice from their GP should they need any additional supports. The various resources listed were catered for different situations that participants could find themselves in, for example, there were non-specific bereavement resources listed, alongside specific MD networks, and services that support the siblings of those deceased.

- In planning this research, I also thought it ethical and best practice to consider the impact of the research on my own wellbeing. Indeed, increasingly researchers are addressing the emotional safety of researchers conducting interviews in the context of distressing and traumatic stories (Beale, Cole, Hillege, McMaster, & Nagy, 2004; Bowtell, Sawyer, Aroni, Green, & Duncan, 2013). I initially sought my own personal psychotherapy, but could not continue this to the end of the project. However, I continued to develop my own self-(re)formulations over time (McCormick, 2012) and keep a reflective journal for my own personal wellbeing, in addition to my research specific journal. These endeavours alongside a growing interest and practice in Mindfulness enabled me to 'cycle' around my own and other's emotional contexts.

2.5 Procedure

2.5.2 Recruitment of participants

As described above, recruitment consisted of multiple phases of offering a complete picture of what the research entailed in terms of its aims, the required investment by participants and its potential outcomes. This process is described in depth below:

1. I compiled a press release relating to the collaboration of the University of Hertfordshire and the Muscle Help Foundation in order to inform the public and key stakeholders of the project and its aims (Appendix J). At the same time, I also included a picture and biography in order to offer further information for potential audiences, who may at a later time wish to contact us to seek involvement in the project. This press release and biography was published online and through multiple accounts on social media, including the University of Hertfordshire.

2. Using Hynson and colleagues' (2006) paper that explored what bereaved parents considered to be ethical and appropriate ways of engaging this group in research, participants were at first contacting by post or email from the charity. This initial contact briefly summarised the intentions of the project and advised that a representative from the charity would make contact within two weeks of the letter to discuss further if they wished. This is recommended by the aforementioned service-user led guidance published by Hynson and colleagues (2006), enabling parents to consider the opportunity in their own time, to have conversations about the possibility with their families, and at the same time, to not feel encouraged or pressured to take part.

3. This representative then telephoned potential participants to discuss the research ideas and project as a whole. They did not seek informed-consent per say, but gauged initial levels of interest in the project. Following these conversations, they made a contact list for me. I then distributed the research materials to those interested in learning more about the project and advised that I would contact them again in the near future.

4. I went on to contact interested families over the telephone. This telephone call took place at least two weeks after the research materials had been received and read, and the telephone call was used to check the families understanding of the project. In making contact, I summarised the research to build upon the participants understanding, clarified the particulars of the study, and answered any remaining questions about the project. I took care to discuss the consent process in depth, particularly in relation to video-recording interviews and how these videos could be used in the future. In discussing this, I sought verbal consent to participating in the study and/or arranged a follow-up telephone call a week later to provide time for the individuals to consider the project fully. The same procedure will be followed on this subsequent telephone call as the first.

5. Formal consent was taken prior to commencing the interview in person, with the opportunity to ask further questions and a reminder that they were free to withdraw their consent and participation at any time without having to provide a reason for this.

2.5.3 Participants

The study recruited five parent couples to participate in interviews overall. I made contact with these families, following their details being passed onto me by the charity. Eleven families were identified by the charity as meeting the inclusion criteria, of who six were initially agreeable to learning more about the research project. One family who were initially interested in the project, upon learning more about the content of the interviews decided that this could be too distressing for them and decided to not take part. Another family had not engaged in a Muscle Dream experience, but offered to participate as a pilot interview. All other families contacted were agreeable to participating and offered their informed consent to take part.

Demographic information was gathered through interview, as opposed to a demographic form and some of this information is detailed in Table 1 below. Due to the small size of the charity, as a research team we were concerned the participants could be potentially identified if this information was shared in its entirety. Although this is potentially at the cost of situating the sample as is ideal in narrative inquiry, we felt that the ethics of confidentiality should take precedence in order to respect those who participated in this study, in accordance with our professional code of ethics (BPS, 2014, p. 22).

All interviews lasted between 100 and 150 minutes each, across one-two interview dates per family. The ages of the individual parents who participated ranged from 29-68. Two of the fathers within the sample were step-fathers. The parents had a range of occupations, including professional and computer consultancy, education, labouring, and care working; both paid and caring for family members. The ages in which the parents' children were diagnosed ranged from 2.5-5 years of age.

Table 1. *Participant details.*

Pseudonyms	Jim & Sarah	Beth & Lenny	Ellie & Ricky	Derek & Kim
Young person's pseudonym	Kieran	Owen	Bond	Samuel
Age of death	19-21	16-18	19-21	16-18
Time passed since death	1-2yr	2-3yr	1-2yr	2-3yr
Interview length	150 minutes	100 minutes	145 minutes	120 minutes
Muscle Dream experience	International Rugby game	Dance show	Car television show	Boy band show

2.5.4 The interview process

Riessman (2008) suggests that researchers should always consider the context in which the story is to be told, and as such, I offered to interview couples in their preferred location. All of the parents included in this study opted to be interviewed in their homes, most in their living or dining rooms. Most of these environments also had photos of their sons around the room, which were referred to at times – or introduced to me, during their accounts. I conducted the interviews independently and followed the University of Hertfordshire's lone-working policy.

With consideration to the aims of the research and the need for in-depth, reflective accounts for narrative inquiry, the sample size was expected to fall between four-eight participants, reviewed intermittently as the data was gathering in order to consider whether I needed to recruit further participants. This process resulted in a total of four parent couples being included in this final dataset, with an additional couple being interviewed for the pilot study. Based on the service-user consultation, it was anticipated that interviews could last between 2-4 hours with both parents overall, so it was anticipated that two interview dates would be necessary, each one lasting approximately 1-2 hours. The actual interview lengths overall, ranged from 100-150 minutes each.

At the start of the interviews, the research information sheets were revisited, questions answered and consent forms discussed, reviewed and signed. I set up the video-camera and checked with the couples as to whether they were happy with the selected angle, and reminded them that the recording and/or the interview could be stopped at any time, without them needing to give a reason for this. Overall however, all participants consented to the video-recording and no one asked for recording to be stopped at any point.

On completion of the interviews, the video-camera was turned off and interviewees given the opportunity to reflect on the interview experience. With two participant couples, only one prolonged interview took place due to pragmatic and organisational reasons (e.g. work schedules). However, for the remaining couples, the second interview followed the same procedures as their first interview with an additional acknowledgement that they could revisit or reflect on any conversations from the prior interview if they wished.

With the use of video-recording in this project and potential for accounts to be made publicly-available, it is also important to consider the ways in which the presence of a video camera will have influenced the interview process and results. One could suggest that particular individuals are more or less likely to consent to participating in a video-recorded interview. For example, in the context of a delivered Muscle Dream, were only those who found the experience to be positive, likely to contribute to the study? If so, this would naturally influence *what* and *how* they were likely to talk about their experiences. As such, this could make it more likely that they talk about helpful and pleasant experiences because of the presence of the camera – emphasizing the importance of the interview schedule to promote some boundaries for the topics of conversation. Though the cameras presence and influence on the produced accounts and analysis should be considered throughout. Beyond this, it is also important to consider the cultural context and 'relationship' to video-recording. For example, is the presence of a camera considered to infer safety for some, yet scrutiny and distrust for others? Is the presence of a camera an acknowledgement of the seriousness or desirability of one's account through video-recording – such as the increasing trend of vlogging[5] among young people (Aran, Biel, & Gatica-Perez, 2014; Biel & Gatica-Perez)? Or is the presence of a camera an opportunity for less privileged or subjugated stories to provide a platform to be heard? Do some cultural contexts make it more or less likely for some groups to relate to the presence of a video-camera as an opportunity to raise awareness, establish allies and seek social or political change? As such, acting as a springboard for social change; the camera in Latin America for example has been likened to a gun -leading to widescale investigations relating to forced sterilisation in

---

[5] Vlogging is a term relating to the practice of video-recording one's experiences/accounts and posting these online.

the country (Burton, 1978). It is possible that the participants in this study came with differing relationships to being video-recorded and with having the presence of cameras in general. The presence of cameras can influence talk then not only on the level of instrumentality and how the talker wishes to influence their audience, but they too are also influenced by the cultural meanings of what it means to be recorded, and their own underlying relationship to that cultural milieu (e.g. an unease due to a context of surveillance, or an excitement in the context of vlogging).

2.5.5 The interview structure

The interview schedule was designed in response to the research aims, literature and through service-user consultation. The research context then was no 'blank slate' –my initial hypotheses around what stories *may* be told contributed towards what stories *could* be told. In this context, the general guiding themes of the interview related to anticipated stories of pre-diagnostic and diagnostic times, the Muscle Dream experience, death and dying, and life after the death of their child. As such, the interview schedule was designed around this framework and primes the interview context for particular stories to be told (Hollway & Jefferson, 2008). The interview schedule is included in Appendix K.

2.5.6 Interviewing couples rather than individuals

I felt it was important for me to interview couples together for several reasons. Firstly, I felt the literature pertaining to represent the views of parents could be misleading at times – with most studies often only interviewing one parent, and that would often be mothers. Secondly, I felt that this approach was more naturalistic in a way, as if akin to a semi-structured ethnographic study. I considered this to be an approach that could move closer to some of the stories couples could and would co-construct in their everyday lives. Indeed, this is a position also noted by Bjørnholt and Farstad (2014), who suggest that more elaborate, contested and multifaceted stories that approximate a real life context are elicited when you interview couples together. Thirdly, I was reassured by Hooghe and colleague's (2012) paper on 'cycling around the emotional core', in which interviewees could support one another through distressing moments – something that I thought was extremely important to consider in this context.

Heidegger (1996) uses the concept of 'Dasein' (to exist or to be there) to understand how individuals are continuously engaging in and interpreting things with other relational beings and the contexts within which they find themselves, or indeed, 'imagine' themselves to be in. When they have been advocated, joint interviews have been seen as beneficial in research when couples share a socially defined identity, such as being parents (Taylor & de Vocht, 2011). Indeed, parenthood is an all encapsulating identity, defined and redefined through continuous interactions with one another, others and the contexts they find themselves in. As such, the concept of Dasein not only helps to justify conjoint interviews in the context of parenthood, but could easily be applied to criticise the use of individual interviews. Explicitly, "Dasein alone is incomplete; even when there are no other people around…Virtually everything Dasein

does or is, is implicitly or explicitly linked with others. People therefore form an integral part of a communal world, and do not exist as separate entities; the world and the individual co-constitute meanings and understandings. Our meanings are not constructed as individuals in isolation; we are always in relation with others" (Taylor & de Vocht, 2011, p. 1579). As such, Dasein offers a strong case for conjoint interviewing, particularly when the couple are jointly invested in the topic considered. Despite this, the practice remains extremely rare (Morris, 2001; Torgé, 2013), and so there is a need to employ and evaluate these practices further – as is the case in this study.

2.6 Analysing stories

2.6.2 Interview transcription

The analysis of these accounts could be said to start with the process of transcription (Mishler, 1991). Indeed, transcription itself is a form of interpretation (Riessman, 2008) and not simply about "getting it right" (Poland, 2003). I transcribed three of the eight interviews overall, in order to familiarise myself with the data, the process of transcription and reflect on what was lost or gained through transcription from video recordings. An independent company transcribed the remaining interviews as previously described. All interviews were transcribed verbatim, including pauses, expressive utterances, non-audible speech, overlapping conversation, interruptions, and the use of 'voice' / parodies of others (Wells, 2011). After interviews, I noted changes in body language or gestures that aided the storytelling in my reflective notes – and also made notes following a revisit to the video data post-analysis (e.g. pointing to household objects; describing family photographs). An example of a transcript can be found in Appendix L.

2.6.3 Framework for guiding analysis

Narrative inquiry does not include a formal guide on how best to conduct one's analysis, but instead has a range of possible ways to understand the data (Wells, 2011). Riessman (2008) describes three approaches often used within narrative studies:

- Thematic narrative analysis relates to an exploration of content and what is actually told.

- Structural narrative analysis explores the use of language, and how this is presented and organised.

- Performative narrative analysis looks at how narratives are co-constructed and interactively created within the interview context. This approach combines aspects of the thematic and structural approaches described above.

In trying to explore the narratives of parents living with a child diagnosed with MD, this study used all three layers of analysis in order to depict *what* the parents retold, *how* the retelling was presented, and with what *context* in mind. These multiple layers of analysis seem pertinent to understanding how couples narrate shared experiences, particularly as these are often narrated by individuals independently. "Narrative sense-making is a communicative process, one in which spouses organize and understand their individual and relational lives by putting their experiences into narrative form and testing these stories out on an audience" (Kellas, Trees, Schrodt, LeClair-Underberg, & Willer, 2010, p. 177). With notions of 'testing' accounts out on audiences, performative analysis becomes particularly useful in order to understand the *what* and *how* of co-constructed narratives within specific contexts. For example, it can be useful to consider how 'present' particular audiences are during the storytelling, even if they are not

physically present ('ghostly audiences'; Minister, 1991). This notion is particularly relevant in regards to whether particular stories are told with the charity, other parents, or society in mind at the time (among many other possible audiences).

Performative analyses can inform the clinical practice of psychology, as the ways in which individuals tell their stories differ according to the perceived context. For example, an assessment that could determine access to treatment may result in someone's story being retold in a different way as opposed to the retelling to a supportive friend. Performative analysis can enable the clinical psychologist then, to consider the contextual aspects of talk in order to make sense of why particular stories may be chosen above others (e.g. to mourn the loss vs. to celebrate the life lived before the loss), the ways in which the telling of stories can be supported or hindered by physical environments or objects (e.g. using puppets and play), and/or supported by co-storytellers (e.g. 'informants', carers).

2.6.4 Analytic process

With Riessman's (2008) analytic framework in mind, the analysis of the presented accounts are considered to be "situated retellings" (Mishler, 1991, p. 51). I began the analysis process through firstly listening to, and then watching the complete video-recordings of the interviews. I then went on to read through the transcripts, in order to familiarise myself with the accounts. Following this, transcripts were then read in terms of content, using the margins of the page to describe the data. This was followed by a reading relating to a structural analysis; noting broader themes within the accounts and recurring motifs throughout the text. Transcripts were then read in order for a performative analysis that paid particular attention to the joint-telling and interactions

between couples and the context in which these are told (e.g. audiences, discourses). Finally, the videos were watched over with the performative analysis kept in mind and any additional observations noted within the transcript (e.g. the use of gestures within the storytelling).

Although described below as independent forms of analyses, it is important to consider thematic, structural and performative analyses as synergistic components that inform and develop upon an overall narrative analyses. Similarly, the couple accounts presented demonstrate each aspect of narrative analysis, and are also part of the wider collective narrative analyses across the accounts. As such, any particular aspect of analysis should be considered alongside other elements of storytelling (e.g. performative aspects of talk being informed and informing the content/themes of talk).

2.6.4.2 Thematic narrative analysis

When considering the content of the accounts presented, I noted explorative comments and emergent themes within the margins of the transcript. In doing so, these highlighted changes, challenges, opportunities, and reflections (including sense-making, hopes, principles/values) retold within the narrative accounts of parenting. Due to the literature indicating a psychosocial impact on parents, I also thematically analysed the emotional experiences retold.

2.6.4.3 Structural narrative analysis

The structural analysis allowed me to look at *how* parents accounted for their experiences. This included consideration of the ordering of events, whether based upon

a thematic progression or temporal and chronological development. In the organisation of the accounts, where parents began and ended their stories was noted overall, but also how changes were presented through the juxtaposition of stories to one another. I noted recurring themes and motifs throughout the transcripts in order to consider whether these were presented in a way that structured the narrative to resonate with particular perceived audiences; for example, the use of analogy to create imagery or emphasize the emotional experience of a situation in the retelling.

In considering structure within this context, I also considered how the accounts were co-constructed between the participants as a couple (Kellas et al., 2010). This included consideration of both the verbal and non-verbal interactions between them. Kellas and Trees (2006) consider there to be two types of joint-storytelling available for analysis – namely, conversational and shared joint-storytelling. Through conversational analysis, I explored how the couple used one another as co-authors to the account, influencing the story in the moment through interruptions, questions, clarifications, and expressions of understanding and/or disagreement. Through shared joint-storytelling, I noted when stories were extended and elaborated upon in an accumulative and supplementary manner – building upon the established narrative.

2.6.4.4 Performative narrative analysis

In reading the transcripts for performative features, I considered the context of the storytelling and the interactive aspects of the interview. I also considered how my questions or comments could influence the process (Mishler, 1991), potentially as another co-author within a conversational analytic sense. Independent of shaping the accounts witnessed by the questions I asked, I also kept in mind the role of 'audiences'

throughout the performative analysis. My physical presence during the interview was considered, particularly in terms of visible or voiced signs of difference (Burnham, 2012). Explicitly, were interviewees narrating to me as a young white man; a researcher; a psychologist; a student? Along similar lines, I considered the wider context of perceived audiences, which seemed particularly relevant due to the video-recording of interviews. As such, the analysis considered whether parents were narrating in relation to wider audiences (Andrews, 2007), such as the charity or other parents in the community (particularly due to the explicit invitation to consent to producing accessible video accounts).

2.6.5 Gaining alternative perspectives

In qualitative research, it is not uncommon to return to participants post-analysis in order to explore the ways in which the researcher had presented their accounts (Lyons & Coyle, 2016). Indeed, member-checking can be considered an ethical approach that aids the credibility of the research (Elliott et al., 1999). However, in this context, the act of member-checking would create a further layer of analysis beyond the remit of this thesis. This is because, as earlier stated, my epistemological position considers narratives to be re-presentations of lived experience that are co-constructed and situated within multiple present or ghostly contexts. As such, member-checking in this context would equate re-presentations of re-presentations, and possibly require further analysis itself. Within the context of bereavement, it is also possible that such a process could be unnecessarily distressing for participants.

Without member-checks from the participants themselves, it was important for me to employ some other means of quality control through the analysis process. Three peers

and I established a narrative workshop group and met formally on three occasions over the research period, with facilitation from an expert in narrative inquiry. These groups were used to discuss narrative approaches and collaborate on analysing transcript extracts. Three peers independently coded a sample of an interview, discussing and reflecting on the similarities and differences in our analyses. Following this, my second supervisor independently analysed another interview in its entirety, and again, met to discuss and reflect on the similarities and differences in our analyses.

2.6.6 Representing the narratives

There are different approaches to how one can present narrative analyses of different accounts of lived experience. In the context of my clinical training and requirements for this thesis, it is important to consider the extent to which I can credibly adhere to my ethical stance in presenting a transparent account of the conversations held, at the same time as being transparent that this is context-specific, situated within epistemological assumptions that shape how I 're-present' the narratives presented to me (Etherington & Bridges, 2011). Indeed, reflexivity is not only limited to procedures of research, but also to how aware I am in influencing the presented narratives – contributing through my emphasis on particular accounts and my omission or under-recognition of other important stories (Lather & Smithies, 1997). One inevitable loss in my own retelling of these accounts, lies in the fact that "the body and its parts are used as communicative instruments and as resources for structuring and interpreting stories" (Hydén, 2013, p. 139). I make attempts at the retelling of such embodied narratives through a performative analysis in this thesis, but due to the reliance on textual presentation,

aspects of any narrative are inevitably reduced to something much less 'alive' than the specific context in which the story was retold.

In 're-presenting' the accounts of these parents, I have decided to present a short summary of each of narrative account, and present my analyses of these accounts by grounding the assertions in quotes that I have interwoven throughout the text. Beyond these presented couple accounts, I present collective narratives in consideration to the commonalities of experience retold across the sample group.

2.7 Credibility and pragmatic use

As per the literature review described in chapter one, I have attempted to adhere to the quality guidelines for qualitative research as posited by Elliott and colleagues (1999). Through its design and application, qualitative research aims to make claims of credibility in its presentation of findings (Elliott et al., 1999). Credibility in qualitative research is said to be achieved in part through degrees of reliability, replicability, and consistency (Elliott et al., 1999; Tracy, 2010; Yardley, 2007). Such measures look to situate both the researcher (owning one's perspective) and the sample within their context (Elliott et al., 1999). It also aims to ground findings in interview data to aid transparency, to develop a coherence that resonates with its audience, to be clear on its general or specific research task, and to elaborate upon the methods employed to ensure interpretations are credible (ibid.). To ensure the analyses and findings generated in this research are credible, reasonable and grounded in the data, I have sought to meet each of these criteria (see Table 2 on page 75-76).

Research findings can and should be used to form useful contributions to clinical practice and guide future research (Riessman, 2008). The sharing of narratives also creates a context for social action, empowerment and change (Andrews et al., 2013). I address these implications for services, research and the public in the final discussion chapter. The potential for disseminating this research holds many opportunities, particularly in terms of contributing towards the understanding of the way in which parents narrate their lived experience, but also to look at how parents take account of the impact of 'wish-fulfilment' experiences for their children. On completion of this thesis, I will prepare an article for publication in an academic journal and present the findings at a conference.

Table 2. *Self-evaluation of quality standards employed.*

Criteria	Approach employed to maintain standard
Explicit scientific context & purpose	Systematic literature search completed and presented in context of other relevant research, UK context, and research aims.
Appropriate method	Narrative methods used to elaborate on how parents account for lived experience, including content, performative aspects and structural aspects.
Respect for participants	Due to sensitive research area, extensive efforts to adhere to ethical approach of recruitment, based in literature and guidelines. Extensive service-user consultation. Confidentiality maintained throughout. Additional checks regarding consent. Participants provided with as much choice as possible throughout. Ethical mindfulness in research.
Specification of method	Method described in full, including development of interview schedule, procedure in which this was conducted and levels of analysis.
Appropriate discussion	Discussion based on findings from interviews, using quotes to illustrate this. Used to corroborate, supplement and critique current literature. No claims made beyond the scope of the literature. Implications for clinical practice detailed.
Presentation	Presented using conventional framework for presentation.

Criteria	Approach employed to maintain standard
Contribution of knowledge	This study corroborates much of the literature available in terms of services needing to attend to the family perspectives across the disease trajectory. It goes beyond the current literature to rethink how parents adapt to loss, and how services could contribute to this process.
Coherence	There is a coherent narrative to the research, whereby similarities and differences in experience are linked across accounts, grounded in the data.
General vs specific	This research was specific in its aims to explore parenting accounts that demonstrate particular stories of adaptation to living with MD. As such, no claims to generalise the findings are made – but aspects may be representative of other accounts and those in similar situations may benefit from related implications.
Grounding in examples	All findings supported by extracts from the data.
Resonating with readers	Narrative inquiry documents the retelling of lived experience in rich detail that illustrate life events, identity and meaning-making. This method elicits stories that are likely to resonate with a range of audiences. This is particularly true due to the range of similarities and differences throughout the documented accounts.

2.7.2 Reflexivity

Reflexivity can be used to consider one's own participation in the particular contexts, such as research interviews – but also in terms of the entire process, from conception to dissemination. Self-reflexivity is required to ensure data remains credible, in terms of reviewing findings (your own or other papers), and can be used to ensure interviews are delivered in accordance with the appropriate standards. As narratives are arguably produced collaboratively – with "the interviewer playing an active part in the production of talk and meaning" (Silverman, 2006 as cited in Abbott, 2012, p. 244). This not only means the researcher should reflect retrospectively, but should also anticipate and reflect in the moment in order to maintain a high quality of research. Such reflections could relate to how my own personal experiences or principles influence how I engaged with the different aspects of the research.

Following the interviews, I recorded my reflections on a Dictaphone. In my reflections I considered a range of topics, for example I considered the ways in which the interview context was created – who played the most influential part; what audiences were held in mind at different times, what surprised me most; what emotions came up for me; what identities did I participate in, at different times (e.g. researcher, clinician, youthful self, male self, etc.) – and how these aspects may have contributed to the co-construction of the narrative. For example, the emotional aspects of conducting research in the area of child bereavement had an impact upon how I conducted the interviews; finding myself more aligned to sitting with silence during the interviews, as opposed to interjecting with further inquiry. It is possible that through empathising with the parent's loss, I focused upon stories of survival and togetherness in talk and analysis, as a means of comprehending and making sense of such loss. Becoming

mindful of this response to the emotional content of asking, telling and analysing, in part enabled a richer analysis that named such survival or adaptation in the context of its origins; pain, loss and bereavement. The process of reflecting on one's role within and relationship to their research is however an ongoing and dynamic process, whereby one can never become fully 'reflective' – and so I could only aspire to dedicate myself to this process and work towards as reflective and reflexive a relationship with all aspects of this research over time. As such, my research narrative itself should be read within its own context and considered a situated account in itself.

Following my reflections, I would collate my thoughts in a more thorough, critically reflective and ethically mindful manner through use of a reflective journal. I have used these as a basis for my personal reflections in concluding this thesis. In anticipation of my role as a reflexive-researcher, I also sought quotes that would challenge my thinking in terms of my role and support me to become more attuned to the influence of self in research (an example is included in Appendix M).

In the next chapter, I shall present the findings of this study.

Chapter three: Findings

3.1 Overview

This chapter provides an overview of the analysis and interpretation of the accounts by parents whose child has passed away from muscular dystrophy. This reflects the stated research question of how parents who have lost a child to MD story those experiences of parenting. Firstly, I present the individual accounts. The purposes of this is to introduce the reader to the main storyline and performance of their narrative. This in turn answers the questions of *what* is talked about, and *how* the parents story their experiences (aspects of the research aims). Thereafter, I present my interpretation of collective storylines across all of the accounts.

In this chapter then, my research questions underpin the re-presentation of accounts that illustrate what seem to be important aspects of the parenting experience for the participants, and the ways in which events and the impact of events such as the Muscle Dream are retold. Throughout, I also explore the ways in which parents narrate their collective experience as a couple through a performative lens, as per my research questions.

All names used are pseudonyms and all personally identifiable information has been omitted or anonymised. Due to size of the charity, the anonymization necessary for confidentiality unfortunately restricts the degree to which I can situate the sample below.

3.2 Introducing the individual accounts

3.2.2 Jim and Sarah's story

Jim and Sarah worked in consultancy and care work. They were birth parents to Kieran and his younger sister, Bea (by two years). The first time that I met with Jim and Sarah in their home was on the day of a coffee morning to raise money for a MD charity that their daughter had organised. Despite this, they offered as much time as was needed and so established a scene in which MD was still very much a part of their lives.

Jim and Sarah were engaging narrators, for the most part presenting a coherent and chronological account of their experiences. Jim was particularly expressive and compelling in conveying their account through words, expressions and reference to objects. Both Jim and Sarah often used active voicing (Wooffitt, 1992) for themselves and others, including their late son. I found these approaches to storytelling particularly engaging, promoting perspective-taking for me, positioning and imagery of person, event or place.

Jim and Sarah described a "difficult" route to becoming parents, with a history of pregnancies struggling to take hold and Sarah having had several miscarriages before Kieran was eventually born. Jim declared that "all our dreams [were] answered" with the birth of their first child, Kieran. Two years later, their daughter Bea was born. Fearing "dodgy hips", the couple sought professional consultation when Kieran was young, resulting in a series of tests. The couple spoke of the panic of not knowing, when their GP called just three days later asking for a consultation. Jim vividly recollected the "absolute bombshell" when the GP alluded to Kieran's condition; *"it can be serious"*. Renovating their home alone at the time, Jim recalled pacing around like "a caged lion"

"in floods of tears", trying to desperately track Sarah down (laughing that this was before the times of mobile phones). The couple recalled hearing of muscular dystrophy for the first time, and seeking subsequent muscle biopsies to identify the type – hoping for Becker's, instead of the more life-limiting Duchene.

Kieran was diagnosed with Duchene before the age of four. Told not to look on the internet as "anxiety levels were through the roof", the couple recall being given the hope of "time"; time to find a means to delivering the cure. The couple talked of time also diminishing their hope; "too much damage done" as Kieran's "body deteriorated over the years".

The couple recalled the different challenges at key transitions through Kieran's life: remembering barriers to getting to school trips and limitations in activities during those trips, to inescapable buildings during the event of a fire and becoming panicked by regular lift breakdowns.

Jim in particular, talked with great sadness, of how Kieran was "denied so much" throughout his life. The couple talked of the struggles of having to observe as their son become effectively invisible to his peers, including his own cousin, during the time that "teenagers…become dehumanised". The couple talked of frequent instances of Kieran being superficially included in activities: "it was inclusion and exclusion at the same time…When you sit in a room and nobody even knows".

Jim recollected being invited to write the nomination form for a Muscle Dream; determined to get his son this opportunity. The couple recalled Kieran being invited directly (rather than through his parents), the environment being tailored to him (e.g.

hotel beds adjusted for hoists) and the build-up of surprise after surprise; "the perfect experience, no stone left unturned".

The couple frequently talked of their "fight" and "battling on" as a family; not allowing the condition to direct their parenting style. Eventually, the couple described "stepping back" and allowing Kieran to become more independent in his later teens. The couple eventually described themselves as being "so *close*", that they "could not *see*" Kieran's deterioration over time: introducing "the cough" that was the beginning of the end. They recalled his subsequent hospitalisation and realising that things "weren't right" when he slept so much and wouldn't eat his usual foods during a home visit. Through tears, Jim retold his son's determination to "fight this" as his best friend visited, and Kieran's worries about not having brought his dad a Father's Day present (with a week to go). Kieran passed away on the Tuesday before Father's Day.

The couple recalled how "heaved" his funeral was, as unfamiliar faces approached to reminisce about their love for their friend endearingly referred to as "wobbly legs". Jim struggled to talk about how popular his son appeared to be, as he contrasted this with memories of how angry and tearful his lonely son could become at times. Since Kieran's death, the couple described a struggle "to pick your life back up again…[and] to soldier on". They talked of missing the "madness" and the "noise" in their house in particular; "just plodding along" now, having to find something else to focus on, such as work.

3.2.3 Beth and Lenny's story

Beth and Lenny worked within care and manual labour. Beth was Owen's birth mother and Lenny was described as his step-father, joining the family in Owen's early teens. The

first time that I met with Beth and Lenny was in their home, where several other family members were present and curious about the research. In the background, Owen's two siblings and friends played and watched television. The setting seemed to establish a scene in which family functioning revolved around being together and supporting one another.

Beth and Lenny were engaging narrators, for the most part presenting a coherent, although not always chronological account of their experiences. Indeed, for the second interview, Beth was left on her own and used this opportunity to tell her account of parenting before Lenny came into her life. Beth and Lenny used active voicing too (Wooffitt, 1992); again for themselves, others, and their late son. Again, I found these approaches to storytelling particularly engaging, promoting imagery of person, event or place, and encouraging perspective-taking from my position as their immediate audience.

Beth was a single-parent for the first decade of Owen's life, having a turbulent relationship with Owen's father and managing to move out of "a difficult neighbourhood". Beth talked of the journey of parenthood as a rollercoaster of the "emotional, physical, mental and everything" – in which she *had* to be stronger and took much of her inspiration from Owen himself. She recalled Owen "tripping over fresh air" and having "clicky hips" as he walked "like the honey monster". This led to her seeking several hospital appointments, which seemed to "last forever", until Owen was finally diagnosed. Having never heard of MD, she turned to searching for answers and looking for the fix – which kept her "in a bubble" for some time – but then she "broke" and "changed that day". "It changes you, inside and out": diagnosed with depression, and experiencing anxiety and panic attacks.

Beth spoke of adapting and staying strong for her children, taking each day a day at a time – which eventually ended with the couple taking each hour, an hour at a time. Beth recalled putting up as many barriers as she could when she first met Lenny, detailing that "nobody's gonna want to take on baggage with me". However, "it didn't faze him" and their relationship blossomed.

Beth and Lenny spoke of making decisions in confidence, with Owen always being included – putting their "own fears and questions aside". The couple talked of being "very straight", with no "pussyfooting around" issues. Due to Beth needing to look after the other children, Lenny spent almost three months with Owen in hospital and this "opened [his] eyes" – particularly when Owen had a cardiac arrest one morning. This was particularly difficult for Lenny, who had his "head in his hands" much of the time, with no official paperwork to name him as Owen's stepfather – hospital staff could not permit him to make final decisions on behalf of Owen's care. With other children in the home, Beth and Lenny talked of how it was "difficult to balance everything" and at times as a couple, they "were just ships that passed in the night", didn't feel they could sleep and were "obviously on the go all the time".

From the moment of being selected, Beth and Lenny recalled how excited Owen was to go and see a dance act for his Muscle Dream experience. They recalled how the facilitator was relatable to the young people and inspired them; being in a wheelchair and living with MD himself. They spoke of meeting other families in similar situations, in a beautiful setting – something they could never have afforded themselves. They spoke of witnessing the energy of the day as Owen continued to "whiz" around as they collapsed onto their bed to rest. "Another fantastic memory that [they] will always have…priceless…just nice to have that quality time". Beth said: "I felt special and I'm

only mum, so God knows how Owen felt". Indeed, they recalled conversations between Owen and his friends, as he described the day as "sick"[6] and boasted of the events for months thereafter.

In talking about death, Owen would use humour to protect his parents – for example, declaring "oh, great, morbid one today then". The couple talked of not feeling in control throughout the latter stages of the illness, but Beth suggested "to gain a bit of control back is to give Owen his voice, to sit and listen, and to do what he wants". As such, Owen returned home for his final days – which involved laughter and tears. Beth and Lenny were "a bit of scaffolding" to one another, where behind closed doors they could "be a wreck" – which was important for them: "we're only human". At his funeral, the couple spoke of their pride in their son, but Beth also said "I can't remember, I remember seeing maybe two faces that day".

3.2.4 Ellie and Ricky's story

Ellie and Ricky worked in care and consultancy work. Ellie was Bond's birth mother and Ricky, his step-father from a very early age. Bond had several siblings. Ellie and Ricky were engaging narrators, for the most part presenting a coherent, although not always chronological account of their experiences.

The narrative content and performative expression in presenting humour and laughter through struggles, juxtaposed a stark contrast in the content and emotional resonance of the narrative; with empathy and position-taking becoming particularly compelling

---

[6] This is popular slang in the UK that monopolises on inverting meanings in order to emphasize one's point. In this case, by saying 'sick' the suggestion is that the event, experience or state went 'well', was good, exciting, stimulating, and so on.

for the audience. Ellie and Ricky also used active voicing (Wooffitt, 1992); again for themselves, others, and their late son. They used humour throughout the storytelling and often laughed when retelling stories of struggle and about the challenges they faced as a family.

Ellie and Ricky began by negotiating the starting point of their narrative as distinguishing between the amount of years living with Bond as different to those years living with MD. This provided the sense that the couple clearly distinguished person from disease and indicated the person-first discourse that the couple would go on to present. Following a series of hospital appointments and an initial 'diagnosis' of "youngest child syndrome", Bond was eventually diagnosed with Duchene MD before the age of four. Ellie talked of her shock at how insensitive the initial diagnostic consultation was, as she was advised of his terminal illness but "reassured" that he would never be incontinent. The couple described the diagnosis as like a "death sentence", but not long after they "started to find [their] feet again and sort of dip [their] toes in…a different world".

Bond needed a wheelchair before the age of ten, but the couple talked of fostering the drive to do anything and make the most of life. Ricky described how there was "always a fight" in trying to make the best of their situation. In this sense, Bond would take every opportunity to get involved in school trips, campaigning and television interviews, and if this seemed challenging at first, the family would do their best to persevere and change this. Indeed, they "conquered a few mountains, chiselled a few ways forward".

The couple spoke of how all the family thrived on surprising one another, which also led to Bond developing initiation rituals for new carers due to his "evil sense of humour". Ricky and Ellie used their love of surprising one another to include Bond's own Muscle

Dream. Bond would go on to retell all of his friends about this for months thereafter, creating memories for the family that they could never have pursued on their own without the charity's persistence in liaising with a television company. Ellie and Ricky talked of their difficulty in entrusting others to support Bond on his journey towards independence, but he eventually moved away and lived independently for three years. He returned home after college, which surprised Ricky and Ellie at first – but they described how he may have wanted to be at home with his family nearer the end of his life.

After Bond passed away at home, the family held a "celebration", as opposed to a funeral, in which hundreds of people attended. Ellie and Ricky felt that it was important that Bond attend his own celebration dressed as a superhero and have since continued some of Bond's unfinished campaigns.

3.2.5 Derek and Kim's story

Derek and Kim worked in care and consultancy. They were parents to Samuel and his younger brother, Peter. I met with Derek and Kim in their home, with the conservatory doors open and the sound of children playing in the park. This scene setting seemed like a reflection on their openness to sharing their story and almost a juxtaposition to the otherwise quietness within the home, having had Samuel pass away and Peter move out.

Derek and Kim were reflective and direct narrators, and although not in chronological order, presented an account of their experiences that seemed coherent through significant themes and issues. Kim positioned herself as a care worker early on in the

interview and appeared to align herself with the perceived "expert" audience within me, utilising medical discourses and terms at particular points in the account. Both Derek and Kim used active voicing (Wooffitt, 1992) from time to time, but not as frequently as other interviewed couples did. I found these approaches to storytelling particularly engaging, and provided a clear and coherent understanding of their positions within the family, their experiences over times and series of "fights" they faced.

From the beginning of their account, Derek and Kim held other parents in mind – detailing that their story was "probably slightly different to others". They positioned themselves in this way as they storied a struggle in conceiving. This was a long and extensive process, in which they prepared for parenthood, thinking through how they would manage a range of different parenting scenarios – hoping for a child. They talked of their expectations of parenthood being important in shaping how different their story was, and explicitly talked of their hopes to have a child who was healthy not to have a disability; hopes relating to Kim's experiences working with families professionally. By juxtaposing their expectations to the unfolding account, they seemed to emphasize the struggles they faced in trying to meet their son's needs over time; "because [disability] would make life a lot harder".

Derek and Kim described being "knocked sideways" by the diagnosis process, experiencing "quite the shock". They went to their GP just before Samuel was three years old to suggest that he had "joint problems" to be told that it was a muscular problem – but nothing else. From this point, the family suspected MD as they had seen a documentary about this on television at the time. Kim and Derek described a reluctance of consultants and charities to provide any information about MD, before Samuel was officially diagnosed – which required his blood tests and biopsy results. Indeed, they

described being "incredibly fraught, incredibly stressed, because in one breath [they] were…fighting like mad to get information". Samuel's diagnosis of Duchene MD was disclosed by accident by a secretary when Kim phoned up to chase the results, and found this to be a particularly stressful period.

Derek and Kim talked of finding support in unexpected places, particularly through friends of friends. They soon made a decision to invest in developing a quality of life for Samuel, independent of his life-limiting condition: a decision influenced by Kim's belief in a social model of disability (utilising a medical discourse to position the family's socio-political motivations) (e.g. Shakespeare, 2010). Again, Derek and Kim framed their journey in the context of a "fight" – detailing "I was determined, and, as Samuel got older, he was also determined that we would fight for everything we needed". As Samuel grew older, he became integrated into the local community and others would "look out for him". Through continual "battles", the family managed to secure funding for 24hour care provision, so that Samuel was able to leave home to live independently. Over time, Derek and Kim described how their son learnt "the fight" from them and went on to get involved in local decisions around adaptation, advocate on behalf of others, and campaign. This also reflected Samuel's "extra angst" in his teenage years in which he held some "extra anger" and had "wonderful rows" with his parents. Overall, Kim and Derek described seeking their "wants" as a family, as opposed to just what they needed – which they presented as a costly endeavour, with renovations and costly adaptations to vehicles, in order to achieve a quality of life equitable to those without disability.

The Muscle Dream experience highlighted the strong connections Samuel had developed outside of the family for Derek and Kim, as they witnessed the confidence

and commitment of his carer who nominated Samuel. The event was said to have created lasting memories, something that the couple described was important for the parenting journey as a whole. Derek and Kim's account described how Samuel's death came when "everything was just coming together" – for a period of time, Samuel was said to be "very miserable" but that he had acquired a job with his favourite sports team. Kim and Derek did not explicitly detail Samuel's funeral, but talked of their discussions as a family about how to commemorate his life. As such, they described a yearly visit to Samuel's favourite park bench to share a drink and eat some food in his memory.

3.3 Emerging storylines

Following individual analyses, I considered the parental accounts collectively. The focus of this collective analysis was on similarities and differences in the key stories told, and familial identities conveyed in consideration to the research aims. Through this analysis, I observed three main stories being told in the accounts: stories of 1) change, 2) surviving times of change, and 3) creating times of change. These narratives included particular stories within each (see Table 3).

Table 3. *Storylines and sub-stories*

Story	Sub-stories
Stories of change	Waking up to different futures
	Being so close, you don't see the deterioration
Stories of surviving	Humour through the struggle
	Storytelling together
Stories of creating change	Creating a legacy, making a difference
	Living the dream

Below, I will address these main stories in turn.

3.3.2 Stories of change

All parents described the process of seeking consultation around the early signs that something was wrong with their child's development. These accounts described numerous hospital appointments, tests and a lack of communication from health professionals about the process and the condition itself. These experiences appeared to reflect the beginnings of broader stories of busyness that all parents described, forcing adaptation to changing circumstances without necessarily having the time to process these transitions and what that means for their identity as a parent.

3.3.2.2 Waking up to different futures

All of the accounts detailed how difficult they found it when their son was diagnosed with MD. They all described changes in their sense of self, realising that envisioned

futures of who their son could become and the parent they could have been for them, shattered through the delivery of a diagnosis. This is demonstrated by Ricky and Ellie as they retold their experience of the diagnostic consultation, demonstrating how content, structural and performative aspects of storytelling synergistically convey meaning on a multitude of levels. For example, juxtaposing the 'death sentence' of diagnosis to the 'mourning' and loss of what their son could have become; constructing a narrative of loss in terms of selfhood, child and future.

> Ricky: *"It was almost like, it was a death sentence. It, it's described erm, life limiting… in the first sentence, Duchenne muscular dystrophy, life limiting, muscle …*
>
> Ellie: *… wasting neurological condition, isn't it? [R: yeah] But I think erm, we mourned the loss of Bond then, that's how I feel. [R: yeah] Because … we couldn't see the wood for the trees at that point, could we? [R: No] We just saw the fact that he's gonna get weaker and weaker and weaker and that'll be it" [723-736]*

Similarly, Jim and Sarah talked of shock and pain of diagnosis, construing a sense of one's identity and selfhood being shattered in response. Indeed, their words convey a literal shattering of self: "I was in so much bits". This suggests that their experience of loss from that lived experience is very much carried with them, perhaps as an embodied narrative that continues a connection to their son's embodied story that remains unheard directly (Frank, 2006). Despite this, Jim tells of the immediate claim to a new father identity, not only utilising discourses of 'fight' common to all of the accounts, but also influenced by gendered discourses around masculinity and taking control of one's situation (MacDougall, 1997). This approach is likely to be compounded by the

'restitution narratives' (Frank, 1995) that are readily available throughout society's approach to healthcare, based on medicalised discourses of curing disease. At the same time however, although this discourse motivates him – within his broader narrative it inhibits him, due to the lack of cure for his son's condition. Through drawing on these discourses, Jim makes sense of his loss of self and future in a way that enables him to adapt and claim new identities thereafter. In doing so, Jim addresses the pain and struggle he had to overcome in order to tell himself a different anticipated story at the time.

> *Jim: "We just did not stop, I just did not stop crying, didn't stop crying, it was almost, it was, it was like losing him and after all the hard work we'd had to get him … you know, and the joy we had when he was born, aah, it, and- and I remember sitting on the sofa in here 'cause I, you know, I couldn't, I couldn't even finish off the decorating, I was in so much bits. And I thought to myself, "Jim, you're either gonna fight this and do the best by that boy or you're just going to lie down like this" and, and so the decision was made, "Come on, you've got to get strong now" and er, but God it was awful, wasn't it?" [95-104]*

For Derek and Kim, the point of diagnosis was storied as a disruption to planned for identities as parents, and a challenge to hoped-for identity of their child. In their story of hoping for 'a healthy child', Derek and Kim appear to use discourses of 'normality' in the context of societal discrimination of visible difference in order to comfortably hold their position. There was almost a sense that they had been 'cheated', in a socio-cultural context whereby healthy, thriving nuclear families are posited as the norm, to be expected of couples. By contrast, this narrative of loss appears to pre-empt a broader

and dominant motif in their account, discourses of enablement through means of conflict (e.g. fighting for services). Indeed, this reflects a broader story that Frank (1995) calls a 'quest narrative' – in which individuals (in this case parents) orientate their accounts towards a meaningful pursuit of particular goals.

> Kim: "At the point of diagnosis, it was quite - well, it would always be a shock, but because it wasn't in our family, and because we'd been told this was a healthy child, it was, erm, quite a shock, really...it comes, you know, completely out of the blue, really, and knocked sideways immediately" [176-188]

The following extract from Beth and Lenny's account again details how diagnosis was narrated as a shattering of self and parental dreams. Beth retells her attempts to make sense of things, but stories a struggle to comprehend the change in her circumstances, struggling to gain a sense of coherence within her new, yet chaotic narrative. Through the performative use of active voice, Beth emphasizes not only the narrative content of distress and grief, but also the broader motif of facing loss and adversity.

> Beth: "It was, it, for a good two, three weeks I think I was in this bubble and erm, I didn't really get upset, didn't really get mad, I didn't really feel much, I was quite numb and then I broke, just oh God, cried like a baby ... I bet that was for nearly a fortnight, me mum come down and stayed with us, erm, and I just couldn't, couldn't comprehend any of it, I couldn't take it on board or anything, erm, "What am I gonna do? What am I gonna do? What's he gonna do?"

As she continues to narrate her experience, Beth presents a sense of urgency, a sense of being controlled by her change in circumstances, and feeling pressured to know, adapt and cope. It is possible that cultural discourses around motherhood influenced this narrative, presenting subtle assumptions about how Beth as a woman and mother, *should* have felt at the time and what she *ought* to have done, and so on. As such, she narrates a forced restructuring of identity in response to her new, changed reality but also possibly by discourses of motherhood. As such, utilising discursive aspects of narrative, Beth informs *how* she goes on to tell her story in this particular context and within the broader context of her story as a whole. In doing so, Beth continues to use imagery that conveys the shattering of her identity as she struggles to 'pull herself together', almost as if different aspects of her are left at different points within the story. Indeed, she draws this 'small story' of diagnosis, to a point in which her older self is no longer thought of as being present.

> Beth: *"Oh my God", it was just, but ev-everything was like, I was jumping into everything, erm and then I had to sort of pull myself together and be like, "Right, hang on, one question at a time, one answer at a time" and from then on in, I took one day at a time and when Owen got poorly poorly, I took it hourly, erm, because it's just, your mind … it, it, I don't know, it can run away with you, you know, it really did mess me up but at about six months Owen was diagnosed, erm, oh I couldn't sleep, I couldn't function and erm, I was diagnosed with depression, so they put me on tablets because I couldn't sleep, I couldn't switch off at night erm, and throughout the day I had panic attacks, anxiety, just totally did not feel me at all. I changed that day, that diagnosis come and I, I've never been right since to be honest, erm, only now*

Owen's no longer with us, I'm different again, do you know what I mean? All these things in life, the impact it has, it- it changes you, inside and out, erm and I don't, I can't remember the last time I truly felt like Beth" [948-962]

Throughout all of the accounts co-constructed by the parents were stories involving hopes about what parenthood would involve and the opportunities their children would have. These 'dreams' appeared to be based upon wider discourses of 'normal' or 'conventional' life trajectories, which were often then juxtaposed to the lived reality of parenting a child diagnosed with a life-limiting condition.

In Beth and Lenny's account, Beth construed this parental dream as something that Lenny could choose to participate in – a choice that she initially puzzled over. This may relate to the broader story of how diagnosis of one's child can shatter a parent's identity and force them into claiming a new identity situated in different and unanticipated futures. Below, Beth's puzzlement emphasizes the loss of selfhood that one may experience when becoming a parent to a child with a life-limiting disease, as opposed to Lenny not having to join what Beth has effectively storied as a less conventional trajectory of parenthood.

Beth: "I think it's just, I don't know, if somebody had a choice, I was Mum, I didn't have a choice, we got dealt them cards, we had to deal with it, erm, for, for Lenny, for me it was more, mmmm, why would somebody choose to take this road? To take our journey with us? Erm, I- I found that really difficult to digest and then I also had the fear of erm, "Oh God, what if they get close?" and it got, you know, Lenny got put off in any way and ended up go- it was so hard because there was so much going on but me being Mum, just wanted to protect everyone I think? Erm, but he persevered, erm and when

Lenny come on the scene, erm, about two and a half years down the line, Owen was deteriorating quite quick, erm, he was in hospital for months on end" [52-61]

For Derek and Kim, despite their planning for parenthood, they still ended up parenting their children quite separately at times, which they narrated as never being part of their plan. This appears to relate on the suddenness of change, limiting opportunity to reflect and process, and instead forcing reaction and consideration of the next challenge ahead. As such, Derek and Kim present a narrative in which they were close to adapting to the 'here and now' of caring, that they fell into unexpected patterns and roles as parents, which they had hoped to avoid.

Kim: "It was different. That was - that wasn't the plan, the plan was that we would be joint parents and that we would share everything, and make sure that we were on the same track, and that, you know, we were doing the same thing. But, in fact, it didn't quite work out like that, did it?

Derek: No, it didn't. And, I mean, yes, you, you really don't know whether it would have worked out differently if Samuel hadn't been disabled" [1659-1666]

The delivery of a diagnosis appeared to lead to a loss of hope. Those identities informed by socio-cultural discourses around parenthood and ideas about what life would have in store for them, no longer seemed possible. Through diagnosis, there was a shattering of these hopes, dreams and identity. In answering how parents story their experiences of parenting a child with MD following their death, narrating the loss of hopes and dreams for parenthood appears very important. In doing so, parents juxtapose one's envisioned

and actual life trajectory, that emphasize the struggles and challenges that follow, but also allows the audience to empathise with the significance of this loss. As such, the structural and performative delivery of one's narrative, emphasizes the importance of the content talked about; in this case, the loss of perceived identities and futures. Indeed, Bowman describes such moments as "the loss of an emotionally important image of oneself, one's family, or one's situation; the loss of what might have been; abandonment of plans for a particular future; [and] the dying of dreams" (Bowman, 1999, p. 181). However, due to the continued challenges of parenting a child with MD, the parents talked of having to adapt swiftly, and would claim new identities, adapted to the presence of MD in their lives. These made use of gendered discourses, as Jim chose to 'fight' for his son, and Beth 'pulled herself together' in order to protect her son. Through adapting however, the parents accounted for having little time to process the loss of their previous identities or dreams before having to move onto the practical management of a subsequent challenge.

3.3.2.3 Being so close, you don't see the deterioration

The accounts all depicted a closeness and distancing to the condition over time, making some transitions appear to happen suddenly and without prior warning, independent of knowing that such changes *would* happen at some point. The following quote from Lenny demonstrates how his 'distance' from the condition initially allowed him to see past Owen's future deterioration, but that through daily caring tasks he occasionally found himself confronted by a painful future. As can be seen here, the performative aspect of active voice is also used to gain a distance from the emotional content of the talk, reflecting the broader narrative and discourses around the life-limiting disease.

> Lenny: "I was unaware of the condition and everything at the time...so I was quite oblivious to everything at the time and I, I just, liked seeing past it every single day. Obviously, the caring side still makes you think about it every...It used to like hit you there and then sort of thing, like "wow, there's gonna be a day that's gonna come and you're not gonna be able to do this anymore" [42-47]

Like Lenny's sudden insights into future losses of ability, others described a similar suddenness to their son's actual deterioration. Presented in the context of relentless 'battles' and 'fights' for resources, Jim and Sarah narrate a sudden loss of mobility whilst feeling so rushed at the time. In their retelling, it would seem that a discourse of remaining steadfast and persevering through the toughest of times, with the declaration of 'hey ho' being reminiscent of British discourses of maintaining 'stiff upper lip'; that is to remain in complete control when confronted with distressing problems (Capstick & Clegg, 2013). Active voice on this occasion, again permits a distancing from the emotional content of the story being retold, almost as if allowing another person to tell the story on their behalf. This distancing within the narrative is achieved through a performative element, possibly in acknowledgement of that painful content, and the broader story of deterioration.

> Jim: "By the time you got that thing put in the house, you were onto the next thing, you know, so now you're battling for that and oh, and, and , so that was just a constant sort of grief, you know, but hey ho, it was as it was but yeah, I'll never forget, we were laying in bed one morning 'cause Kieran could still, you know, walk a little bit but he spent most of his time in a chair and he just, he just called from his bedroom and he said, "I can't walk anymore" and

it was just as simple as that weren't it, "I can't walk anymore", just like

that... So the day before he could walk, the day after he couldn't" [275-287]

It would seem that the parents narrated a suddenness at times of change, perpetuated by a closeness that limited their perspective at the time and limited their ability to anticipate the changes ahead, that they otherwise would have been able to 'see' with some distance. Through their stories, they spoke of a sense of constant change, with little time to stop and process; limiting opportunities to grieve, protest or lament such change. Changes and deterioration then, were narrated as small moments of shock, disbelief and hurt, but were quickly taken over by further challenges, new 'battles' and future changes. Ellie and Ricky's account demonstrates their own story of sudden deterioration too, as they retell their confusion over Bond's transition from walking to wheelchair, through a consultation with a physiotherapist:

Ellie: "[The Physiotherapist] came round and sort of very patiently said to me, "Do you remember I said he'd go off his feet?" and I went, "Yeah", she went, "Well that's what he's done", I went "But that was quick" and she went, "It's been going on for years" [laughs], because you know you just get so used to things as they are and then donk, isn't it? It's another change

Ricky: It was a change in some, some respects, erm ...

Ellie: But it was, you know like every major change, isn't it?

Ricky: It was almost, it was almost a relief as well that ...

Ellie: For him ...

> *Ricky:* *Yeah, and physically for us and for him, it became … erm, his lack of mobility was, in terms of getting around, was becoming quite hard work and for him to be able to do that on wheels… made his life a lot easier and to be honest it kinda made ours. That transition was, was always going to happen and when it did happen, it kind of, it opened a new chapter in, erm, his life but it also sort of turned a page for us as well.*
>
> *Ellie:* *And you just think, "Okay move on" …" [1888-1908]*

In Jim and Sarah's account they narrated the impact of Kieran transitioning into a chair in the context of an ever-increasing visibility of deterioration and/or disability. In their account, they voiced similar frustrations to the other parents in this sample – the visible signs created barriers to relationships outside of the home, limited opportunities as a result, and often had a detrimental effect on one's wellbeing due to the responses of others. In presenting this, Jim and Sarah utilise discourses around exclusion, stigma and discrimination within society, enabling the audience to empathise with their struggles. Indeed, the extract below depicts aspects of this and opens with a contestation of dominant discourses around disability and wheelchair users:

> *Sarah:* *"It was just his body that didn't work, he was a, you know, cognitively he was brilliant, it was just that wheelchair and the fact that his body didn't work and he hated the way people would perceive him just because he was in a wheelchair…He was just, he said "I'm just like everyone else, Mum, my body just doesn't work and why don't people see that?" …Because all they see is the wheelchair to start with.*

Jim: And, and we, that was a, a, you know, walls that we fought so hard to break

down; stop looking at the wheelchair, look at the person in that wheelchair,

it, it's just a wheelchair, you know, there's a person there" [2678-2689]

The motif of closeness and distance throughout the accounts detailed transitions as 'out of the blue' and unanticipated, as parents were 'too close' to see the deterioration over time. Only through retelling these experiences following their son's death, could they gain the distance to make sense of the broader story of deterioration. Through this closeness, the parents also found themselves in some respect, blinded to changes in their son's health until crisis point or significant changes in their needs. These changes were storied with a suddenness, and individuals spoke of their shock to such changes – again, having little time to process, with the requirement to swiftly adapt to these new circumstances becoming imperative.

In 'being so close, that you don't see the deterioration', all of the parents directed their account towards the dying process and eventual death of their sons. The accounts depicted a long period of time in which the parents lived with the knowledge that their child was dying, gradually over time. This journey cannot be dislocated from the wider cultural stories of parenthood, that presuppose that parents can/should 'fix' things for their child and prevent the 'bad things' from happening to them (e.g. deterioration). Indeed, such journeys from ill health to resolution are reflected in much of the way that society expects individuals to story their journey through living with disease, treating accounts of health as 'signs' that in principle connects people with one another – in what

Frank (1995) describes as restitution narratives (from sickness to health). Needless to say, for these parents however, they had to live with the knowledge that they could not 'fix' the situation nor 'prevent' deterioration or death, and so rather than 'connect' with dominant accounts of health, these families have to make sense of their experiences and story them along different trajectories.

The couples narrated particular ways of facing these challenges in their accounts. In preparing for death, Beth and Lenny talked of developing an openness within the family that allowed them to discuss distressing subjects openly. Beth acknowledges the challenges in being unable to 'fix' the situation, and talked of her own avoidance of the issues at times, but ultimately utilises alternative discourses to 'protecting' her child from knowing about death, employing instead a discourse around the resilience of children. This is perhaps in stark contrast to her own storied loss, of there being nothing left for her. In the extract below, Beth recognises the challenges that she has faced as a parent on the journey, but emphasizes the importance of open dialogue between parents and children about death and dying. This demonstrates the performative progression of active voice, elaborating on the content and characters within the narrative. This re-presentation of events also utilises the broader and structural aspects of the narrative, conveying a journey that informs and is informed by discourses of death and dying.

> Beth: "You can't sugar coat terminal illness, erm, it is what it is, I understand people can bury their heads, I did that myself, but I do believe that kids are more resilient than adults without a doubt and you've got to g-, you know, you've got to give them some credit because as much as it, I think it's harder for the parents, erm, to approach kids, erm, you know, on anything like this

but the hardest thing is them being diagnosed and you living with it, the hardest thing then obviously is erm, when you lose your child, you know, there's nothing more but on that journey, I think you get to a, to a point where … you have to get hard, you have to, you know, it's a case of there is that much going on in your head, it's not about "Right, well, we'll sit down and, and I'll just approach the idea" and you know, softly, softly, it just, it can't be because there's that much going on, it has to be you know, like "Right, listen, this is the situation, right, what are we going to do with it? What are your ideas?", everything needs to be out in the open, everything. I find a lot of it is, erm, you know, behind closed doors, adults will have a quick chin wag" [174-189]

All of the accounts detailed the significant loss faced when their son passed away. The narrative co-constructed by Jim and Sarah below demonstrates the impact of this on them as parents and also their perceived futures:

Jim: *"It's like losing a … wheel off your wagon, you know, just, just, it just isn't the same… He was the centre of our world, we were the centre of his weren't it, you know?*

Sarah: *Yeah, 'cause you, you were hands on with him all the time …*

Jim: *And you could still, you know, we- we've talked about that and, and I can still feel … every area of his body, you know, when you used to shower him or dress him or turn him over, you could still sense those, those, you know those connections, that touch … and his face felt, you know, his little beard and, yeah, never lose those sensations.…I just don't feel the passage of time now,*

it, it's, because it, life is a struggle now isn't it, you know, but we have to stay strong for Bea…I know that Kieran is out there, he is out there, you know and 'cause of our beliefs and erm, and I know that he wouldn't like to see us even sitting here like this…he'd want to see us happy because we will meet again, but it's, it's just, it's just a matter of time. But it is difficult to pick your life back up and, and to soldier on, you know, it really is…

Sarah: When your life has been solely about looking after one person…when your life has been so intently … erm, structured to look after the per- this person, Kieran, that needs this, this, this and this and it's just … your way of life and that is changed, it's just taken away dramatically…It's really hard" [1413-1460]

The proceeding extract from Beth takes the audience back to the moment in which she and her son were advised that there were no longer any possible interventions, and that Owen was dying. She begins with using 'active voice' to present her son declaring that she had given up on him, perhaps allowing her to re-present this content to her audience in the knowledge that the broader plot is known. She retells of her reaction, almost as if in defence of herself and her commitment to fight every possible cause for him – perhaps linked to how she narrated carrying this voice with her ever since; an 'unresolved fight' on behalf of him, perhaps. This could represent the lack of opportunity to process things at the time and perhaps, the continued attempts to process and reclaim her identity as a 'fighter' for her son. In this latter respect, her involvement in this research could be considered part of that continued reclamation – a process that Beth acknowledged as therapeutic when ending her interview. Through this persistence to 'fight' on behalf of one's son, it could be considered that Beth utilises

a broader 'quest narrative' (Frank, 1995) whereby one's story is presented with moral objectives, a clear manifesto for pursuing change and for action – in this case, even beyond death.

> Beth: *"I still hear that voice today, you know, "basically you're giving up" and I'll always carry it with me but I got upset and mad and I shouted back, "It's not about that!", because I don't, I didn't want that to be in the front of his head, it's so not about giving up, everything, when you've got a poorly child, there's a fight, whether it's equipment, whether it's you know, today, today, everything is a fight, erm and he, he was like and I'm saying to him, I'm speaking to him saying, erm, "Look, you know, there comes a time when your body can't take no more" [373-381]*

The couples in this study had to continue parenting over time with the knowledge that their child had a terminal condition; an awareness that initially changed their perceived course of parenting, and challenged societal discourses about what it means to be a parent. Through storying the end of life, the parents were faced with dilemmas as to whether to avoid or confront the issue of death with their sons. To approach death, Beth for example would have to relinquish her 'fighter' role for her son, which seemed at odds with the relentless busyness of family life and parenting a child with MD. It was almost as if, to not be fighting, was to be giving up. Whereas for Ricky, he storied a broader motif – a fight that was informed by death, one that would encourage the family to achieve as much as possible during their son's reduced lifespan. Overall, end-of-life care and death were key moments for the parents where their identities were once

again shattered, no longer able to fight, and faced with a degree of powerlessness to protect – confronting discourses of parenting.

3.3.2 Stories of surviving

3.3.2.2 Humour through the struggle

Throughout all of the narratives, humour appeared to be a motif and was key to the performative delivery of the parental accounts. Humour was used to present the character and strength of the individuals within the narrative, alongside a familial thread of perseverance through change, social challenges and deterioration:

> *Kim: "You know, it's all - it's, it's a forever process of struggle and laughter" [1462-1463]*

Humour was conveyed as something that could bring the family members together during difficult times. It was often used to position oneself against a stance or experience found to be unpleasant or abusive. This is observed in the below extract from Jim and Sarah's account where they narrate their son's journey from child to teenager and young man, in the context of testing peer relationships:

> *Sarah: Because it, teenagers, they, their empathy goes, when they hit a certain age*
>
> *...*
>
> *Jim: I, I say they become dehumanised and they, I mean that in the nicest way and a joking way because as Sarah just said they, a year before, before all those hormones kicked in, you know, they were, they were, Kieran was in the*

room, a year after when they're then becoming pretty boys and pretty girls

...

Sarah: It wasn't cool to be seen with a disabled ... person! [laughs]

Jim: I think when they get to 18 or 19, they become human again ...

Sarah: [laughs]" [426-475]

At the performative level of analysis, this was observed when interviewees prompted and encouraged laughter from one another – sometimes, drawing the other away from more distressing memories, or as juxtaposed in a way that conveyed shock, disbelief or upset. As such, the performative aspect of storytelling was irrevocably part of the narrative content and structural presentation of accounts. Ellie and Ricky's narrative demonstrates that humour was also construed of as an active choice the family made – perhaps representing an underlying family value and principle of how one face's adversity:

Ellie: "The one thing we always did with Bond was if it was a negative, well with all
the kids, we always tried to turn a negative into a positive because that's the
only way you can survive, I think if you see everything as a negative when
you're going through lots of different systems to make things right for your
family, erm, you have to find the funny side don't you and the positive side?
It's hard sometimes though, isn't it?" [1979-1986]

In Derek and Kim's account, they narrated this ability to laugh through adversity as something that they taught and nurtured in their son – almost as if a needed survival strategy to navigate his way through future struggles:

Kim: *"From an early age I fostered in him the feisty, erm, difficult side of him, you know, because to get on as a disabled person you've got to be a bit of a pain the arse. And, of course, as a teenager that meant he was a pain in the arse [Laugh]…and I persuaded him to be different as a child, because I knew he needed to be feisty and difficult. But I could have done without him being that with me! [Laugh]"* [1508-1523]

This 'feistiness' and 'taught fight' appeared to provide a continuity of strength; a humour that overcomes the struggle. This seemed to provide a sense of pride for the parents, even in the painful retelling of death – as illustrated below as Beth and Lenny describe their reaction to being told that Owen was dying. At this point, the performative aspects of the narrative become increasingly collaborative, as the content is produced by each speaker in turn, using the motif of humour within a much broader story of loss and death.

Beth: *"They'd took him to a room, erm…And had the talk about erm, "we don't think he's gonna pull through" etc., this that and the other.*

Lenny: *Where he's come to in his life now… [shaking head] … "He is dying".*

Beth: *Yeah and its always the same talk…they tell you these things but each and every time, Owen…*

Lenny: *He proved them wrong… [laughs]*

Beth: *Showed them the middle finger basically! [laughs] And…always pulled through and so it was a case of, "oh you know, yeah, get on with it, you know what you're on about, he'll be all right", only this time he weren't"* [317-329]

Within the interviews themselves, parents used humour when narrating experiences of discrimination, unfairness and injustice. This was in contrast to expected emotional responses such as frustration, anger, and outrage. In doing so, the parents appeared to provide permission to one another to address difficult content within safer contexts, to regulate the emotions of distressing stories, but to also create a safer context for their audience to witness these accounts. Having said this, there was also an element of reliving their child's sense of humour through the performative mode of humour and laughter in the face of adversity, in a way that pays tribute to the loss of their physical presence, but invites their character and zeitgeist not only into the narrative, but into the room for the immediate retelling. One could question whether omitted feelings of anger and irritation remain untold due to the perceived expectations of one's audience; the delivery of a coherent story whereas the overcoming of adversity is emphasized more so than the challenges themselves. On the other hand, I wonder whether the operation of hope itself regulates these expressions of anger and irritation. I have found this myself, during times where I can be hopeful of change and felt empowered to seek that change, I could mobilise my pain or anger with a direction and action to overcome it. In this context, parents spoke from a retrospective position, distanced from the moment itself – whereby their hopes for change could have roused such potent emotions at the time. Through the distance to that experience, and the lack of utility in hope for change within that story, it becomes understandable as to why such strong emotions would play smaller parts and become directed through humour. Whereas anger and protest has the potential to alienate others, humour often invites collaboration. As such, through a retelling that uses humour, the audience can be invited into the dissonance of shock and disbelief expressed through laughter. In one sense, the

use of humour could enable parents to survive difficult times, but also through their retelling, to invite others to action (i.e. their audience).

The motif of humour was present across all of the accounts, often presenting a resilience to adversity and a way of uniting when faced with injustice. At the performative level during the actual retelling, humour could regulate one another's closeness to painful content. Humour appeared to function at two levels then, a natural and nurtured way of surviving adversities, and secondly, as a choice to unite and look after one another during difficulties (including their retelling). This reflects the research question of how couples narrate their experiences together, considering humour's place where other emotions such as anger would usually be expected. Humour can engage and unite audiences in shock at injustices faced when parenting a child with MD, so that it can foster motivation to act on or change things for others.

3.3.2.3 Storytelling together

In order to tell their stories, the parents interviewed would often lead each other to the next part of the story with pauses, direct invitations (e.g. "remember that time…"), subtle gesturing or glancing for the other to join or 'chip-in' at that point. This collaborative storytelling approach was particularly noticeable during emotionally potent stories, where partners appeared to offer the other a chance to distance themselves from becoming overly distressed. For example, in the story of Kieran's passing, Sarah confidently offers a delay and break to Jim when he appears to be becoming distressed:

Jim: I'll never forget that.

Sarah: And just, it was just amazing but I mean …

Jim: Oh, it was. Very emotional, yeah.

Sarah: … and very emotional.

Jim: It's still very raw. That, that last couple of days [of Kieran's life], all the conversations that we had and … it's all still so emotional, I can feel myself going now…

Sarah: I know … We'll talk, obviously talk about that maybe a bit later 'cause it, it is very upsetting …" [1028-1034]

This performative element of narrating the parental account appeared to regulate the retelling of distressing experiences and so facilitated a range of content within the broader narrative. At times, this appeared to be self-employed, at other times, invited or directed by the other. On occasion, this could involve apparently inconsistent expressions of emotions, such as painful tears alongside laughter. Swift changes in emotion from distress to laughter were not uncommon in the accounts. In the following extract from Beth and Lenny's account, shortly after reflecting on the loss of Kieran's life, Lenny becomes tearful but swiftly invites a humorous story from Beth, who in fact, redirects this narrator role back to Lenny:

Lenny: And that's it and it's been two years on now, hasn't it? …

Beth: Erm, but it's, yeah, everything is still … referred to Owen, definitely.

Lenny: Definitely.

Beth: Definitely.

Lenny: Still makes me cry [tearful]. What about the Man City shirt? [looking to Beth] I don't know if we should say that! [laughs]

Beth: You tell him" [828-838]

There were times where couples spoke on behalf of the other, representing a shared narrative; "we thought". Indeed, couples frequently relived dialogues through 'active voice' (Wooffitt, 1992) and brought in the voice of the other throughout the overall narrative. On the one hand, they would present the voice of their own internal monologue at the time, on the other, they would present a dialogue between characters. These were often interwoven in the narrative, particularly during stories of consultation, as demonstrated by the quote from Derek and Kim's account below:

Derek: "At the time, the campaign were not at all helpful in that sort of situation, and they just said, "no", yeah, "we're not going to talk to you about what even, what the implications might be, because he might not have muscular dystrophy"…

Kim: And the then medical advisor felt that they shouldn't, we shouldn't talk to people until the diagnosis was in place.

Derek: Which we felt wasn't particularly helpful, because, you know, we, we wanted to know what the possibilities, probabilities were? Erm, and, yeah, we were - we were quite prepared to say, "yes, okay, this is what it might be"…and then for them to say, "no, it's not muscular dystrophy" and we'd feel "thank goodness it's not that!" You know?" [234-245]

As observed above, the internal monologue or collective perspective would often be offered as commentary between voices. The frequency of such an exchange between these 'active voices' seemed to increase in relation to the emotional valence of the story (e.g. the revelation that Kieran was dying in Jim and Sarah's account). This again suggests that emotional cost of narrating specific content or acknowledging the broader story of death, could be mediated through the performative means of retelling.

Couples also used their bodies as tools to present their accounts that seemed to add to the emotional meaning and/or sense of time, urgency and proximity; for example, pointing (direction or object orientation), rolling fingers in a circular motion (time, kinetics, motion, representing hopes of 'getting on with things'), and the grimacing of one's face, shaking of head or the clenching of hands (representation of pain/frustration). For example, in Ellie and Ricky's account, Ellie snapped her fingers to symbolise the suddenness of a friend's death, conveying a sense of shock and lack of preparation – but also used her body to convey humour, such as observed in the following extract:

Ellie: "Bond was there going [pulls 'feel sorry' face] with this look...[laughs]" [492-493]

Through the retelling of the parenting experiences, couples often aided one another in co-constructing a narrative account. Through 'chipping-in', clarifying, emphasizing, asking questions, and so on, the narrative appeared to be supplemented by one another. Through interruptions and 'corrections', the narrative appears to be re-directed. These appeared to work on the basis of collaboratively prioritising particular stories over

others, perhaps in relation to the audience and questions at hand. Indeed, through these interactional components of the narration, it appeared that couples regulated one another's emotional distress reactively and in a preventative manner – steering conversation away from particular stories at particular times. These appear to be the ways in which couples collectively narrate their experiences of parenting a child with MD, as per the research aims.

3.3.3 Stories of creating change

3.3.3.2 Creating a legacy, making a difference

The co-constructed accounts detailed how many of the parents' sons had pursued change in society and made a difference to other people's lives. The quote below from Beth and Lenny's account details how Owen's approach to life in the context of his prognosis, offered inspiration for those around him:

> Beth: *"It was just a case of Owen sort of plodded on and just, he never, ever whinged, he never moaned...it's just the person he was, and he will always be, you know what I mean? I still draw strength from Owen, I always will do. Without a doubt.*

> Lenny: *I think anybody would to be honest with you, I don't think we're ever gonna meet anyone as, I don't know, inspirational as what he were. It was just unbelievable, honestly, what a guy... [Interviewer: Can you tell me more?] ...Where do you start? [Lenny laughs; Beth smiles] ... It's, every, even though he was like the younger of everybody else, he was... everybody looked up to him, didn't they? He was like ... the leader, so to speak.*

Beth: He was the core, weren't he? Yeah, he was the cog … that you know, he, he just constantly, you know, got on and did things, but the inspiration from Owen I think is the sheer love for life, knowing … in his little head, there's not much ahead of him but it didn't stop his, erm, his, you know …

Lenny: Goals in life" [520-546]

In retelling of this lasting legacy and inspiration to others, the parents in this study narrated an impact beyond death. The following extract from Ellie and Ricky's account demonstrates how they conceived of their son's impact to others around him, even beyond his death:

Ellie: "He did more in his life than a lot of people are doing who live a lot longer – so…. You know [laughs]

Ricky: The good thing, you know… [hesitates] the legacy that Bond has left, is the fact that his friends who were all, who have similar or erm, disabilities have found motivation in their own lives to go and do something. They saw Bond as an inspiration and there were quite a few friends that didn't feel sorry for themselves because of their disability, but they felt limited because of their disability…

Ellie: They had been a bit closeted because of it as well, didn't they?

Ricky: He told them…" Get out there and do it…If you wanna do it, do it. Don't ask, just do it." And now, some of them are living independently, some are going on cruises,…

Ellie: Yeah… one went to Uni, someone who wouldn't have been able to go before…Well – he's now working! [laughs].

Ricky: I think that's probably… Bond's best legacy was that he proved to people that, uhm, you can do stuff….

Ellie: Don't let your wheels hold you up…" [388-404]

In all of the accounts, the legacy left by their sons frequently involved changes to environments, alongside impacting on relationships and inspiring others. The content of such talk likely reflects the broader narrative of death, but also reflect a performative element in which the parents may have been talking to 'ghostly audiences' such as other parents, making it more important to select some content over other stories. The proceeding extract from Jim and Sarah's account demonstrates this:

Jim: "[Kieran] actually blazed a trail everywhere and, and he seemed to blaze a trail all through his education was, you know, the adaptations that all the schools made and I think Kieran was probably one of the first pupils, you know, to be in a wheelchair, you know…So they had to put ramps in and all that so, but of course that legacy Kieran left behind for the other kids coming through, disabled kids, you know, the, the things were in place for them and so that, that's always been nice for us" [179-187]

Stories of creating a legacy and making a difference for others, were often retold in the context of 'fight' discourses – analogies that in themselves, convey challenge, struggle and commitment to pursuing change. These discourses appear to be used to foster empathy within the audience and highlight injustices within the community. By doing so, these stories juxtapose 'normal' family trajectory to the teller's different path,

inviting the audience to consider their own participation in discursive actions that may help or hinder other families currently living with MD or related conditions. Indeed, in Derek and Kim's account, it was described how this fight was exchanged and learned from one another, for each other and others in similar situations to themselves in the future:

> Kim: "I was determined, and, as Samuel got older, he was also determined that we would fight for everything we needed. Erm, and from Samuel's point of view, it was because if we couldn't get it, then how could any other child get it, and he wanted to do things better for those coming after him" [465-470]

Again, Derek and Kim accounted for how Samuel thrived through the fight to leave a legacy behind and change things for the better. In their retrospective account, they mention the efforts required and arguments that ensued only in passing – focusing more on the satisfaction of the 'fight' and ultimately, on the satisfaction of the 'win'. This could relate on their perceived audiences in this particular retelling, with other parents in mind, it is possible that Derek and Kim storied their experience on this occasion to focus upon the 'ending' of their journey, as opposed to the alluded struggles that came before it. On the other hand, it is possible that through the loss of their son, such stories of overcoming adversity are thickened through retelling and function to sustain the narrative of legacy, strength and continuation. This narrative approach functions to sustain their identity as parents, and as successful parents, and seems to convey a moral message to their audience – that the struggles are overcome in time, are worthwhile, and shape one another's identities within the family.

> Kim: "So, yeah, so I think we did achieve his quality of life…but with a lot of effort. Lots and lots of arguments.

Derek: He got a lot of satisfaction out of that as well.

Kim: Oh yeah, yeah, he enjoyed doing it, yeah.

Derek: He enjoyed winning anyway.

Kim: ...enjoyed challenging people...particularly when he felt they weren't being helpful, but, to some extent, that was, I think, I think that was learnt behaviour, to some extent, in that he...we always challenged them, so he'd grown up in that sort of environment. Erm, so, you know, he felt - what he enjoyed was that he felt he would make a difference for people coming after him. That's what mattered to him" [721-743]

Parents narrated a long-lasting impact of their son's lives on themselves, others and the environments around them. Typically, such narratives used discourses of 'battle' in which the ultimate 'victory' story was prioritised over the challenges and struggles that preceded those achievements. Emphasized stories that omit or lessen the stories of struggle that came before them may well aid parents to sustain hope in the context of bereavement, and to avoid returning to the grief and loss of earlier envisioned futures and parental identity. A legacy can justify the losses that parents have had to make in the past, whereas a story without legacy would potentially become personally threatening. At the same time, through the retelling of legacy, the parents potentially foster hope in other families and so almost provide a moral message to others. In short, narratives of legacy sustain and foster hope for both tellers and listeners, alleviating some of the pain in bereavement and the retelling of loss.

3.3.3.3 Living the dream

This research created an opportunity to explore how parents storied the impact of the wish-fulfilment events such as the Muscle Dream experience, including how parents made sense of and narrated this in the context of their child's death. All of the parents constructed narratives around the Muscle Dream experience in similar ways. These events were deemed to make possible otherwise inaccessible or seemingly impossible activities, in a personable way that could inspire the young people, particularly by seeing what the chief executive could achieve whilst having MD himself.

> Ricky: "It was interesting from [the charity's] point of view as well – as Bond was one of the first dreams that he'd had done. So, there was nothing written down really about what they should be doing, so it was all kind of off the cuff – but it worked out fantastic. The coordination. [Ellie: EVERYTHING]. I think because [someone] was obviously in wheelchair himself…
>
> Ellie: He's got the empathy to understand what people need and how to go about getting it. You know" [215-222]

A common way of narrating the impact of the Muscle Dream experience was in terms of 'creating memories' for all involved. As the extract from Jim and Sarah's account demonstrates below, this was felt to have an impact at the time, but also to continue forth with the families involved. In this light, content may reflect the performative aspect of storytelling for one's perceived audiences; whether for one's self, their partner, other parents or the charity themselves. At the same time, the stories told at this point are likely informed by the discursive context, recurring motifs and broader narrative. In narrating the importance of such experiences, Jim also considers the wider

context and dominance of curative research invested in by many charities, which emphasizes an importance in having both curative and experience orientated charities.

> Jim: "What they do for erm, you know, the disabled lads … is, is just outstanding, it, it is a perfect experience, no stone is left unturned and you know, whilst as I said, without repeating myself, the science bit behind it and the cures and treatments is important, this is also equally important and…just a great, you know, just, and they felt so special, Kieran felt, I could just sense it coming off him, he, he, he was buzzing with it and I was too, given us some lovely photos and lovely memories, you know?" [1854-1871]

Beyond the memories, this process was described as perspective-changing for some of the parents, realising the important aspects of their lives when readily distracted by the demands of life beforehand. The below extract from Derek and Kim conveys this:

> Kim: "Making memories is what it's all about, really. I don't think you know that, and certainly I don't think we knew that as parents.

> Derek: No.

> Kim: I don't think we consciously thought we were making memories, because we were busy being parents and in the middle of a family, and living our lives.

> Derek: Mm-mm. But when you look back now, those were important making memory points, and I think when you're older and, you know, for older adults and things, who, you know, then they're just a couple, so then they can go through a bucket list, or start making memories like that. In the middle of a busy life with a family, I don't think you have that opportunity,

and you don't think that through, but they have been important points…in

our lives" [1249-1266]

In Beth and Lenny's account, they story the importance in the opportunity to focus on intimate familial relationships during the Muscle Dream experience, construing these as possible through the supportive, empathic and relatable organisers:

Lenny: "You felt like you'd known them for a long time, like they was really, really

close friends.

Beth: Oh God, yeah…And Owen were whizzing around and whatnot, and we sat on

the bed just watching him for a while, didn't we? Just go mad and erm, it

was just, it was nice, it was erm, it was another fantastic memory that we'll

always have, which again, priceless. It was just nice to have that quality

time where it was me and Lenny and Owen, whereas obviously at home

you're juggling aren't you with the other siblings as well, it was just nice to,

you know, to solely have, well for Owen to solely have our attention…We got

so much out of it, but I can't really put into words, erm, other than what I

have, what it was, we all got out of it, erm, I felt special and I'm only Mum,

so God knows how Owen felt" [739-767]

The Muscle Dream experience was considered as something that could 'lift' up the young people (a term also used by Jim and Sarah) during times in which they were often struggling with the challenges of becoming young adults, finding work, and establishing relationships, when faced with deteriorating health. The extract from Derek and Kim's account below expresses this point, and appears informed by the broader narrative and

the way in which they re-present their story through a less chronological, and more thematic ordering of challenges faced.

> Kim: *"I think that the, the Muscle Dream experience and, and those sorts of experiences are - they give a lift, they make you feel special at a time where life doesn't necessarily make you feel very special. Certainly, for him, he wasn't feeling very special at all, and this came along and made him feel special. And I think that's very important, erm, because the challenges are enormous and that little bit of support on the way is great"* [1767-1774]

Narratives of the Muscle Dream experience detailed an intervention that offered the timely chance for young people living with MD to not be disabled by their environments and to enjoy an experience that would be otherwise inaccessible. Narratives were similar in their use of a motif about 'creating memories', both in the moment and beyond the experience itself. For parents, they storied an escape from busy lives and caring for their sons, whereby they could simply join them with their enjoyment. Some of the families used similar imagery, narrating a 'lift' in their child that created continued conversations with friends thereafter. Much like the creation of legacies, the creation of memories appeared to sustain a degree of hope for the parents and to potentially 'lift' them to a distance away from the core emotional distress of their loss.

3.4 Summary

Throughout the interviews, there were narratives of loss in terms of the parent identity and envisioned futures, particularly at the point in which their sons were diagnosed

with MD. This reflects an interesting aspect of how parents who have lost a child to MD story their experiences, the key research question in this study. Namely, that aspects of identity are often 'shattered' through the immediacy of challenges faced with parenting a child with MD, with little time to process these challenges, parents are faced with the task of claiming new identities negotiated between one another, their child, the condition and their changing futures.

The next chapter will discuss the findings with particular reference to the research aims and wider literature, followed by my thoughts on the clinical relevance of this study and suggestions for further research.

Chapter four: Discussion

4.1 Overview

The parental accounts witnessed throughout this project were analysed using content, structural and performative narrative analyses. In doing so, as a researcher, I too have become 'part' of these accounts through the ways in which I have chosen to share such rich and complex stories. Even from the conception of my research questions, I have invited particular stories for the parents to consider in their narration of the parenting journey. I think that this is an important point to illustrate before I summarise the findings from this study; in that these findings are situated within my own time and context in which I peruse, make sense of, and convey such interpretations. For the findings presented in the previous section represent the aspect of the parenting journey deemed important to the broader and collective stories, and presented the different ways in which conjoint storytelling can occur – which in part, offer examples that go some way to answer the research questions. I hope that this approach gives due respect not only to the participants who gave their time for this project, but also provides a mark of respect for the souls who were no longer here to voice their own accounts. Indeed, I write now, only from the position that I strive towards an understanding of, and create an approximation to, the lived experience of those parents involved. For "life experience is richer than discourse. Narrative structures organise and give meaning to experience, but there are always feelings and experience not fully encompassed by the dominant story" (Bruner, 1986, p. 143).

4.2 Discussion

From the parental accounts witnessed, three broad stories emerged relating to change, surviving times of change, and creating change. Within these stories emerged important narratives with implications for changes in identity, and how discourses around parenthood and disease are negotiated when facing a child's mortality. Narratives of a closeness that blinds one to the deterioration occurring before their eyes. And a humour that prevails and surpasses death itself, in which legacies are created during a co-constructed retelling of lives lived and continued. In this discussion, I will pay homage to these narratives in light of the existing literature and consider how these narratives reveal many answers to my key research questions of what become important aspects in parental accounts, how parents narrate the impact of events within and through their stories, and how their collective retelling organises narratives in a way that frames evolved and changed identities.

4.2.2 Shattered dreams and identity formation

When facing times of change such as one's child being diagnosed with a life-limiting condition, a parent's identity and envisioned future becomes uncertain. When we consider something to have been a loss, this can mean that our perceived trajectory in life has been interrupted and disrupted, resulting in the loss of a future story (Lester, 1995). Indeed, couples are faced with the shattered dreams of parenthood they once held and may have held for many years. Families are then faced with a struggle to create new identities for themselves (Parkes & Prigerson, 2013). Bowman describes these losses as the 'shattering of dreams'; a shattering of one's perceived or envisioned future

(Bowman, 1999). He described moments whereby one abandons hopes and plans as 'the dying of dreams', which he argued were due to the losses of emotionally important views of the self, family and situation. Through parenting a child with MD, it would seem that the parents had to engage in continuous sense-making of shattering dreams and future selves that appeared to be silenced by the disease trajectory. This reflects a similar process to the continual redefinition of adversity in order to manage, as posited by Gravelle's (1997) model, albeit with a much greater focus on identity formation.

For the process of constructing a coherent sense of identity, individuals story their experiences and piece these together into a coherent narrative and storyline that may vary depending on the perceived audience (Murray & Sargeant, 2011). Arguably, narrative presents the possibility for the continuation of relational identities; i.e. shared and co-constructed identities such as being parents. What was important from this study was that these were narrated as *different* parental identities – as if forced by circumstances to adapt. In doing so, parents adopted discourses of battle, to fight for 'normalcy' and for society to enable rather than disable their sons. At the same time, it was observed that parents were indeed fighting for their own sense of self, developing a new identity over time, that arguably would need to prevail over their lost sense of self to withstand the grief of alternative futures not lived and no longer possible. Sense-making is proposed as an integral process in the Integrated Model of Meaning Reconstruction (Gillies & Neimeyer, 2006), in which benefits must be found in order for identities to be reconstructed in an adaptive and helpful route to new meaning and direction. Interestingly, this model is conceptualising specifically in the context of actual and physical loss, which the parents in this sample experienced, but I would also argue that this process is as equally important for their own loss of envisioned selves. It would

seem that the overcoming of lost selves and the formation of parental identities seem to be important features when narrating parental accounts of bereavement. Within this sample, this way of narrating the self seemed adaptive, but there may also be other contexts in which rediscovering those shattered dreams in order to 're-identify' with what one has lost along the way, may enable a parent to move forwards beyond their grief.

4.2.3 Building identities, storying together and regulating emotions

In answering the research questions I put forward earlier in this thesis, it would seem that identity formation is an important aspect in storying one's account of parenting. If identity was not in flux at the time of storying these accounts, they could be considered in line with Frank's (1995) restitution narratives; as parents storied the replacement of or fixing of a shattered self. However, 'older selves' did not appear to be replaced or fixed, but simply that different selves formed that recognised the importance of former dreams.

Another point to consider here is that it is through the actual telling and listening to accounts that questions of identity are brought to the forefront of the parenting experience. If communication and dialogue are necessary means for sense-making and identity formation, does this pose an identity threat for parents currently living with the ominous and cyclical losses of parenting a child with MD? As if their sense of self or as a parent will continue in a state of flux because of the relentless schedule of caring? Beth's account of never feeling like her older self again could be deemed testament to these thoughts, and yet at the same time – this remains her continued position, even in

bereavement. However, "it is through telling stories to ourselves and to others that we define ourselves. Narrative provides [that] continuity in our sense of self" (Murray & Sargeant, 2011, p. 165). Based on this, it perhaps becomes important to take a both/and position in terms of identity formation, and in fact, consider that *identities* are formed through narration – to witness both the significant loss and enthused conjuring of selves.

Another interesting finding from this study involved a closer look at how parents performed their narratives together; that is, how they co-constructed their accounts through their interaction and negotiating of storytelling. Importantly, joint storytelling appeared pertinent to how the parents tolerated distress through retelling and enabled a relational-regulation of emotions throughout. Indeed, just as Hooghe and colleagues (2012) described a 'cycling around the emotional core', the narratives in this project appeared to steer closer or more distant to the emotional experience of losses faced at different points.

Two ways in which the parents from Hooghe's (2012) study felt able to regulate one another's closeness to pain in particular, was through not talking directly about the loss and through talking in the context of research. Through not talking directly, parents embraced analogy in order to not feel overwhelmed. This was observed in the current study, and could also be applied to understand some of the moments in which parents re-directed the storytelling or used humour to reminisce over situations or character, rather than grief or pain (I discuss humour further below). In light of Hooghe and colleagues' (2012) recognition of the research context as a safe place for distressing discussions, otherwise not shared, this also seems to relate to the current study – with many participants reflecting their gratefulness for the opportunity to discuss their

relationship with their sons in such depth, some detailing how therapeutic the process was, which is reflected in the wider literature (Birch & Miller, 2000; Murray, 2003). This suggests that some contexts and audiences present differing opportunities to establishing narrative coherence and that through relational techniques and interactions, couples will make attempts to manage this through togetherness.

In surviving times of change, couples co-constructed narratives through collaboration and interaction with one another as the stories unfolded. In doing so, they negotiated the important aspects of what would be shared and what would remain untold. As described, this seemed to represent a process of emotional regulation, reacting to the other's talk and/or steering conversation away from stories that could overwhelm themselves or the other. As such, there were both familiar stories for the couples but also safety mechanisms in the way they told their stories which allowed for the exploration of unfamiliar grounds and exploration of the changes in their identity as parents over time. One particular way of surviving was through the use of humour, allowing for a distance from the pain of loss and an invitation to one's audience to empathise without becoming overwhelmed themselves.

4.2.4 Humour will prevail

In addressing how the parents survived times of change, they frequently narrated "a forever process of struggle and laughter" (Kim, 1462-1463). Humour and laughter was both expressed, and woven into the narrative of survival. There were times of significant hardship, pain and relentless losses – but these adversities were often greeted with, or processed by, a playfulness that bonded the parents in unity – often

with their child in mind, envisioned as a collaborator or instigator of the laughter (i.e. a 'ghostly audience'). Societal discourses around disability and injustices are utilised to draw the listener in, to build an understanding of the pain of discrimination but in a way that was buffered by humour so as not to become overwhelming. This functioned in a similar fashion to the way in which the parents regulated one another's emotions, and so in this light, could be argued to also regulate the emotions of their audience through the use of humour in their telling of accounts.

There was not one account in this study that did not foster this 'humour through the struggle' – a way of narrating the parenting journey observed in the pilot interview also. There does appear to be some literature on the role of humour in the context of terminal illness, but this tends to be focused on the utility of health care professionals 'using' humour as an intervention (Bakerman, 1997; Åstedt-Kurki, Isola, Tammentie, & Kervinen, 2001), rather than something that is organic to the familial relationships and a self-initiated means of survival. This study suggests that in the context of family relationships, humour is used in the retelling of some of the most painful of situations, such as discrimination and death.

There were occasions in which humour appeared at odds with the context of talk, as if to confront a harsh reality that would otherwise be too painful – observed at times through shocked laughter or sarcasm. At times, this appeared to be less protective of one's audience and more aligned to a compelling 'call to arms' in reflection of the frequent use of discourses around 'battles' and 'fights' for equality and quality of life for their sons. Indeed, Dean and Major (2008) suggest that humour helps to fosters relationships between parties, eases tensions, manages emotions and helps communication. In a sense, this dissonant painful-laughter had the function then, of

fostering empathy as opposed to sympathy between parties, easing and managing tense emotions, whilst communicating something potentially difficult to hear. Such humour in the context of injustices then, could arguably enable pained-discontent to lead to hopeful-action beyond those immediate contexts.

4.2.5 Closeness that blinds

When parents talked of facing times of change, they retold stories of being so close, that they didn't see deterioration relating to MD in the moment. Narratives emerged of being too close to see the changes that had occurred within recent days, months and years. Parents storied the apparent 'suddenness' of deterioration; unexpected and premature plummets in one's journey, such as sons losing their ability to walk. From the accounts in this study, stories emerged of continuous challenges that demanded close attention and care from the parents – sometimes at the cost of seeing the broader picture. The task of caring was storied in such a way that a narrative emerged, of being 'too close to see' the deterioration and loss of health, that was seemingly obvious to others. It is only through the distance gained over time, that these parents could story that they were 'so close' to caring and the daily struggles, that they lacked the distance to 'see', that for example, their son was dying before them.

One of course can also wonder whether storying parenting using the dynamic of closeness-distance can function to keep distress and regrets at bay, whether consciously or unconsciously. To recognise deterioration in retrospect may become painful when one wonders how one could not have recognised this at the time.

The visceral imagery of being so close to someone you love that you miss a painful truth, capitalises on many socio-cultural understandings of what it means to be a parent – to protect from future dangers and to 'fix' problems. For parents to gain distance and consider the broader situation at the time, could arguably become overwhelming and a challenge to that discursively-informed identity as parents. Stories of what it was like to persevere with knowing the broader story of terminal illness, whilst having to remain focused and hopeful, remained relatively untold. In considering the research question of what is important within the narration, perhaps points to what is important to hold-back on or minimise within the story. Indeed, "there [is] no remittance from…losses, only the awareness that more [are] to come" (Gravelle, 1997, p. 743). Through storying oneself as so close that deterioration was overlooked at times, possibly functions to explain how parents sustain hope and keep momentum going during the process of relentless challenges.

Perhaps within narratives of parenting, knowing that one was *so* present, reflects small acts of resistance to the broader dominance of 'restitution narratives' (Frank, 1995) in the media, and a protestation against the 'futureless' discourse (Gibson et al., 2009) in reclaiming the 'smaller' victories through parenting. To be present and 'too close' is adaptive then, buffering broader anxieties and feelings of parental powerlessness; protecting, fixing, surviving, and becoming victorious within the smaller moments.

4.2.6 Legacies to tell

In the presented accounts of parenting, couples narrated long-lasting legacies and impact that surpassed death. Stories were shared of sons who made a significant

difference to other's lives and the world that they inhabit; leaving a legacy for generations to come. Such changes included inspiring peers to live their lives as fully as possible, to pursuing permanent changes to increase access to buildings, and campaigning to change laws and local policies. It could be argued the narrative construction of legacies provides meaning to otherwise incomprehensible loss. In this light, meaning is found in loss through sense-making, benefit finding, and eventual identity change – as posited in the Model of Meaning Reconstruction (Gillies & Neimeyer, 2006). Indeed, throughout the study these three elements of meaning-making were evident, as parents made sense of their journeys from a position of 'no regrets'. Through this way of retelling the parenting experience, ghostly audiences such as other parents in similar situations were offered an alternative narrative to loss, one that celebrates achievement, highlights 'benefits' and indicates an impact that lasts beyond their son's death.

Legacy making and legacies to be retold seem important aspects of the parenting journey that suggesting it could be useful for the networks around families to nourish the parents' efforts in legacy-making. In clinical contexts for example, the Clinical Psychologist Linda Moxley-Haegert (2015) has developed a particular narrative approach of 'letting the legacy live' for parents of children with terminal illnesses – where all family members co-create ways forward beyond death. Underlying this notion of retelling legacies, is arguably a desire to reconnect with lost loved ones, a desire for remembrance, and for their presence to be felt not just by the self, but also by others. These principles are influenced by the earlier work of Michael White (M. White, 1988), Dennis Klass (Klass, Silverman, & Nickman, 1996), Robert Neimeyer (Neimeyer, 2012; Neimeyer, Baldwin, & Gillies, 2006), and other narrative and social-constructionist

orientated clinicians and researchers. The approach essentially deconstructs grief, so as to suggest that there are potential gains and opportunities for connection, alongside the loss and disconnection. In practice, these approaches seek to foster reconnection and the continuing of bonds (Klass et al., 1996) beyond death; developing and building upon conversations with the deceased. Narratives of connection, much like the retelling of legacies, can involve the use of objects, photographs, and the creative arts. These narratives often foster changes in sense-making, intentions, practice, and a sense of community and belonging.

This research in part, tells the story of a similar therapeutic process to narrative therapy approaches. Although from outside of the clinic room in its retelling, these findings could be drawn upon in clinical settings in order to support families in not only being able to gather and tell their stories, but to potentially change their relationship to life after loss. These findings demonstrate the importance of careful consideration to context, alludes to potential ways to invite stories of togetherness, and suggests a therapeutic power of narrative in and of itself (Tait, Schryer, McDougall, & Lingard, 2011).

The stories told in the interviews of this study reflect a complex and emotive composition of the 'what', 'why' and 'how' of re-presenting lived experience. These stories have been invited, witnessed and subsequently interpreted by myself – meaning that I have played a key part in the production of these particular accounts. In clinical and therapeutic practice then, it seems important to consider the ways in which we can invite more nuanced stories from our service-users that reflect a reality of mixed feelings, relationships, and identities. Therapists can do so, by moving beyond active-listening towards an approach that Michael White (2000) calls 'double-listening'. This

approach enables different types of stories to be 'heard' at the same time. For example, in this study the expression of pride and hope was implicitly accompanied by stories of pain and loss when retelling of legacies. This example perhaps demonstrates my invitation to a 'togetherness' in retelling, but within clinical practice systems and contexts elicit and invite 'problem-saturated narratives' (White & Epston, 1990). Within these contexts, double-listening provides an opportunity for more hopeful, connecting or preferred stories to be witnessed. In doing so, these parallel stories and experiences can be drawn out from the implicit subtexts of accounts and explored through curiosity, questioning and naming. Indeed, Lorraine Hedtke (2014) uses an analogy of light seeping through the cracks – and that it is through this process of providing a platform to all types of stories or experiences, as opposed to potentially silencing or encouraging one over the other, that avenues for movement and desirable, helpful paths forward can be created (ibid.).

Stories of adversity that precede such loss were given less time and focus in the accounts in this study, as parents appeared to privilege stories of legacy. As parents in a state of bereavement, this selectivity of stories to tell or not to tell, bear similarity to the process in which stories are constructed; something Frank (2006) terms 'subjectifiers'. Frank (2006) recognises that we intuitively conceive of our health as a reality that resides within our bodies, and yet our experience of the body could be said to be shaped by our evaluations and ideals over time. So, by telling particular stories over others, we construct our own embodied realities in particular ways. This is interesting in the case of bereaved parents, having storied their parenting experiences of MD and the subsequent losses that often depicted a shattering of oneself, being 'in bits'. In the context of the legacy making that appeared to enable parents to reclaim hope, stories

that described struggles and/or failed attempts of pursuing change, were either untold or minimised in order to emphasize the positive impact their sons had made. Indeed, Frank (2006) suggests that such stories compete for one's attention, leading to one selecting particular stories that alert them to their embodied experiences, but that this can sometimes be to one's detriment. Take for example, the story of never becoming the 'old Beth' again, that she had gone, and been lost along the journey of parenting; it could be argued that this a subjectifier story that dislocates Beth's ideal self from her current embodied narrative – one that she states has been changed inside and out of her. Indeed, reiterated throughout the narrative is this imagery of a shattered self (e.g. 'pull yourself together'). On the other hand, others suggest that "by weaving the threads of illness events into the fabric of our personal lives, physical symptoms are transformed into aspects of our lives, and diagnoses and prognoses attain meaning within the framework of personal life" (Hydén, 1997, p. 53). This suggests the need for particular contexts that support parents to tell stories that would be helpful to their development of a coherent and embodied narrative of identity and health, but with the flexibility to adapt to challenging transitions that require the adoption of new identities over time. The reciprocity of narrating and embodied identities also suggests then, that the clinical application of double-listening (White & Epston, 1990) could impact upon one's embodied self – creating different opportunities in not only storying but also embodying health and identity. This emphasizes the importance in not only designing spaces that invite storytelling and collaboration, but also the use of techniques that provide a platform to content, emotional complexity and process.

4.2.7 Living the dream: What dreams may come?

Through the narrative of 'living the dream' there were stories of surpassing 'disability' in the sense that Muscle Dream experiences were events designed at one level, to counter disabling social experiences and environments. These were events that captured individual's and family's aspirations and enjoyments, that in everyday life had been 'denied' and the young people 'excluded' from (Jim's words). This is consistent with the Social Model of Disability (Shakespeare, 2010). Through their own lived experience of the Muscle Dream, the parents storied a delivery of hope and a 'lifting' of their sons during vulnerable, painful and distressing times. These narrative accounts would seem to corroborate Ewing's (2009) suggestion that such events offer a unified emotional, physical and spiritual experience.

The accounts in this study conveyed a surpassing of the experience in itself, a creation of connection and sense of community beyond one's immediate lived experience. These elements of the story seem to suggest a long-standing impact of such events, which I had sought to explore through the research aims. The creation of memories appeared to foster the continuation of conversations, and the passing on of dreams for others in the future. Elsewhere, with our colleague Romila Ragaven (Nolte, Ragavan, Randall-James, & Wellsted, 2017), we have specifically looked at the Muscle Dream experience in depth using a mixed methods approach. In this, we analysed 82 feedback forms and 153 thank-you cards and letters, which largely corroborated the various aspects of the narratives detailed in this thesis. Namely, that families surpassed disability and 'lived the dream' through building communities (the Muscle Warrior 'tribe') and social networks; felt enabled and understood; developed confidence and learned new skills; were 'lifted' ('a pocket of smiles' through challenging times); were inspired by the facilitator's presence and achievements; discovered and reclaimed hope; and created

long-lasting memories. This study goes beyond these findings in documenting the rollercoaster-like journey that parents take from birth to continued legacies, highlighting the continuation of memories beyond not only the event, but also beyond the young person's life and the ways in which these are retold.

I have addressed how parents narrate a long-standing impact of experiencing environments and events that enable, within the context of a broader story of an inequitable world full of denied or missed opportunities. Indeed, stories are said to call-out to, and connect with people from all walks of life and so understandings evolve from any initial retelling of experience (Frank, 2006). By inviting parents to retell their stories in the context of this research, particular expectations were established around the areas of interest as directed by the interview schedule, and also by the parents' perceived audiences. As such, parents selected and omitted particular stories in developing their accounts and in doing so, highlighted to me the aspects of their account in which they deemed most important and in need of being voiced to others - meeting the aims of this research project in particular.

Narrative inquiry is well suited to further informing practices then and in helping to contribute to the experience of parenting children with MD over time in as helpful a fashion as possible. Below, I draw out some of these implications for services and further research in relation to the findings of this project.

4.3 Clinical relevance and implications

The findings from this study suggest that families would benefit from consultation that is timed and aligned to key milestones on the disease trajectory, as suggested by previous research (e.g. Gravelle, 1997). However, this research extends the need for consultation to provide a forum for couples to consider their changing identities as parents. Considering the ways in which parents narrated a shattering of self, it would be important for clinicians consider available methods of well-being and mental health screening, in order to improve access to psychological interventions if necessary. Indeed, this need to consider the losses involved in caring for a child diagnosed with a terminal illness, and the impact that this can have on the individuals, couples and families as a whole, reflects a key priority from within the King's Fund report entitled *Bringing Together Physical and Mental Health: A new frontier for integrated health* (Naylor et al., 2016).

With consideration to the significant loss of identity, this study also reinforces the need to improve the diagnostic process; not only in how diagnoses are delivered, but in the level of support offered thereafter. For example, pre-diagnostic counselling could prepare families for possible outcomes, help plan how to communicate diagnoses and prognosis to who, when and how. Consultation that covers the period of uncertainty to the point of diagnosis and beyond could empower those families in making the best decisions for their circumstances and planning ahead; perhaps establishing a forum for exploring changes in identity and futures early-on, to allow for adequate time to process the changes diagnosis introduces to one's family life.

To my knowledge, little research has been published on 'wish-fulfilment' activities. At face value, such events appear to have significant impact on the wellbeing of young

people and their families. The preliminary evidence is supportive of this (Ewing, 2009; Nolte et al., 2017; Shoshani et al., 2016). From the described study, there is some suggestion that these events are important to parents in making sense of their loss, letting the legacy live on, and retelling their stories (i.e. sense making, benefit finding, and identity formation). These are aspects that aid processing loss and working through grief (Gillies & Neimeyer, 2006).

In the context of health services, where consultation may be disproportionately orientated to problem-saturated narratives and/or pathology (Barry, Stevenson, Britten, Barber, & Bradley, 2001), it would seem important to consider alternative stories for young people and their families, such as these ideas around legacy. Those consulting families then, should consider signposting to such events like Muscle Dreams that could enable a 'thickening' of strength and ability based stories (White & Epston, 1990, 2005), aiding psychosocial adjustment to life-limiting conditions. If services are to advocate greater use of the third-sector, I wonder whether there is also an ethical need to evaluate those services referred to or at the very least, utilise inter-agency consultation approaches to ensure a high-quality service for individuals and their families.

4.4 Methodological reflections

4.4.2 Strengths

This study built upon previous research that addressed the familial experience of living with MD stretching across decades (e.g. Buchanan et al., 1979; Gagliardi, 1991a, 1991b; Gravelle, 1997; Samson et al., 2009; Witte, 1985). Whereas much of the previous

research has used only one parent to 'represent' the parental account, this study interviewed couples together in order to elaborate on joint storytelling and shared narratives. As discussed, this is arguably closer to a real-life context and a closer approximation to lived experience (Bjørnholt & Farstad, 2014). Indeed, the performative analysis considered how parents could support one another and regulate their closeness and distance to distress – perhaps making a preliminary case for how interviews around significantly distressing topic areas can be explored ethically and safely for those participating when employing conjoint interviews.

To the author's knowledge, no other study has focused specifically on the accounts of bereaved parents in the context of MD (although some have had a mixed sample; e.g. Dawson, S. & Kristjanson, 2003). This aspect of the design may have given room for different aspects of experience to arise, that have not previously been considered (e.g. legacies).

4.4.3 Limitations

There are some notable limitations to this study. Firstly, it is limited in that it has a small sample size. This is not uncommon in narrative inquiry however, which looks to elaborate on experience, meaning and identity in depth rather than make generalisations or claims of 'truth'. Secondly, recruitment took place in partnership with the Muscle Help Foundation and so only granted access to a particular sub-sample of the wider population of parents to children diagnosed with MD. How similar or different parental accounts are to those families that do not access third-sector support in this instance remains unclear.

Families retold their experiences from the context of the charity rather than the NHS, which may or may not have led to particular stories being prioritised over others. Another limitation of recruiting from this context was that in order to ensure confidentiality is upheld, I could not describe the sample as fully as I would have liked to, due to the small size of the charity making it possible that other families or associated stakeholders could identify participants. As such, through respecting our participant's rights to confidentiality, the readers will not be able to situate the sample as fully as would have been desirable.

Inherent in my epistemological approach, I also consider that there are multiple and equally valid ways in which to analyse and understand the interview data presented (Riessman, 2008). This is to say that my interpretations are situated within a particular time and context, and the frame of my research questions. This means that it is both possible that others' interpretations could differ to those presented in this thesis, and so with this in mind, the research was designed and evaluated with quality standards in mind (as discussed in chapter two) - ensuring that the reported findings are credible.

4.5 Suggestions for further research

This study highlighted the importance of identity formation for couples narrating their parenting experiences, following the death of a child to MD. It would be interesting to specifically consider the notion of dreams, envisioned futures and parental identity at the point in which one's child is diagnosed with a terminal illness. This could address whether the shattering of dreams and identities are indeed as poignant at that time, as these retrospective accounts allude to. If so, a study that explored this could unravel factors that help or hinder the processing of these losses and changes. If not, further

exploration and theorising as to what makes these loses so poignant later down the line of parenting, would be helpful in shaping how services respond to bereaved parents. A project that addresses this for example, could use an ethnographic approach to explore changes in parental identities in depth, and over time.

Little research on 'wish-fulfilment' events have been published. Both outcome and experience based research would enable these interventions to be understood in much more depth and this understanding applied in similar and different contexts. Ultimately, further research could improve the delivery of such events. Importantly, this also highlights the need for further inquiry into the individual's experience of such an event – an area overlooked to date.

Methodologically, further research is required into narrative interviews with two or more individuals conjointly. To my knowledge, there are few examples in the qualitative literature and so this could prove a fruitful avenue for further exploration, inquiry, and elaboration – particularly in areas of apparent 'shared' identities such as being parents.

This research also invited individuals to participate within a particular discursive context; namely, by agreeing to participate in the study, individuals were agreeing to my labelling of them as 'parents' without necessarily having a shared understanding of what was meant by this label. As discussed earlier in this thesis, although there is a biological basis for becoming a parent, the parenthood of interest in this study was more aligned to the social component; the rearing, nurturing and continued support of a child within a partnership of individuals (Steinbock, 2006). Naturally this adds variation to the definition of parenthood in this context, whereby birth parents, step-parents, adoptive parents, and other types of parents, are considered within the same discursive umbrella. In this study, it was both the parental identity and accounts of parenting the

child that were of interest. The chosen method allows for this fluidity of identity, and could have allowed for the parental identity to be contested and/or refuted within the accounts – although this was not the case on this occasion. A limitation in this light is to not have fully elaborated upon this sense of identity with participants and to have made explicit, my understanding of parental identity at the point of re-presenting one's account.

In presenting my findings, I have also placed emphasis on 'togetherness' and considered the individual accounts to not only represent individual stories told conjointly, but also to represent collective 'parental accounts'. This notion that narratives can be 'owned' *between* people is contentious in part, because it involves turn-taking and can indeed involve disagreement, re-direction, interruption, and so on. Should someone from a single-parent household have had participated, it is reasonable to assume that this aspect of analysis may have taken a different angle – namely, because at least one interview would have been with one parent only. As such, analyses of 'togetherness' may in part be reflective of the sample context and lack of a more individual account of parenting a child to MD. However, my efforts in presenting 'couple narratives' is consistent with my initial aims of exploring the way in which couples narrate together, and reflective of an ethnographic approach to research. The stories told reflect this particular research context; an invitation to participate as a couple, to take a position in relation to being video-recorded, and to embrace a performative element of talk that reflects a potential wider audience accessing and viewing their accounts. These stories then are equally important for researchers to hear, just as other accounts where context permits or encourages the retelling of separation and differences, that draw primarily on the pain and mental health impact involved in parenting and losing a child are. In

noting the irrevocable influence of context on talk and the purpose of talk, it would seem important for clinicians to consider the psychological impact of telling one's account in differing ways. In doing so, clinicians can provide the most useful and beneficial environments and contexts for storytelling that promotes adaptation if that is at the person's request. For example, the impact of physical environments on mental wellbeing is well evidenced but not fully understood (Daykin, Byrne, Soteriou, & O'Connor, 2008). Understanding how and in what ways a change in environment can invite particular ways of talking and collaborative retelling could help to illuminate the context-wellbeing links.

It would also be interesting to explore whether similar aspects of togetherness are reflected in conjoint interviews for couples retelling their experiences of parenting, but of who have now separated. This could inform future decisions made when recruited parents into research, when aiming to make inferences about their accounts in line with the concept of 'parenthood'.

4.6 Personal reflections

I am proud to have written this thesis through the transparent lens of my epistemological position and an openness to my personal resonance towards the topic area. I believe that these factors significantly influence our direction, whether we are aware of this or not. So, to embark on a research journey in which we are to become more aware of the roles we participate in and how these shape the contexts in which we find ourselves, felt genuine and was an exciting prospect for me. Here, I will present a selection of my reflections from my time working on this project.

4.6.2 Taking the risk to be seen

In 'outing' my epistemological mask earlier in the text, I hope to have made clear that the narrative presented is a co-construction from multiple stakeholders – myself included. What this research process has also taught me however, is that ethically, there needs to be some sort of hierarchy in which we privilege the voices of our participants and in a clinical context, our service-users. This is why I have tried to adhere to a reflexive researcher position throughout: to be seen *and* heard. In becoming reflexive, the process of research can become personally threatening – firstly, because you consider the self not only within the process but also within the content of research. And secondly, it conjures up a vulnerability of mine about 'getting it wrong' – because what if I slip up and miss particular stories, whilst thickening others disproportionately? By being part of the narrative however – by embracing my own Dasein (Heidegger, 1996), I hope to have taken these relational risks with my audiences (Mason, 2005). In doing so, I have tried to build a credible case to influence change in some shape or form, somewhere along the line - along similar lines to 'a difference that makes the difference' (Bateson, 1979, p. 99).

4.6.3 The selves in process

Just as participants will have had a range of audiences which they spoke to, the same could be said about me throughout the process of planning, developing and writing this thesis. One particular 'audience' that it seems important to reflect on is the charity. In

the early stages of this process, I had to pitch my initial proposal to the charity and consider the interests of all involved. In some ways, this reminded me of a network meeting whereby lots of inter-agency bodies meet together in order to make similarities and differences in perspectives visible, and attempts to partially align our aims are made (Fredman, 2014). I had familiarised myself with this way of working on clinical placement, but very often upheld a sense of control in the role of the 'conductor'.

We all had similar interests in establishing a working relationship of some sorts, but also differences in our expertise (e.g. lived experience of the condition; academic expertise). We initially held different visions for what the project could uncover in terms of impact, and different perspectives on the value in quantitative and qualitative approaches. However, the charity seemed open to my suggestion of understanding the experience of parents in greater depth and readily embraced the qualitative angle I chose.

In some ways, much as I likened this situation to a network meeting across agencies, I also wonder whether this bore similarities to developing relationships with commissioners or line-managers who do not have research backgrounds. I would suggest that the qualities that held the process together appeared to be an openness, drive, and vision exhibited by us all. Needless to say, these all sound more fitting to a marketing course than perhaps clinical psychology. However, I wonder whether therein lies the problem. For us to be able to thrive in multi-agency contexts, in an increasingly competitive milieu as the NHS struggles to tread water in these austere times, perhaps clinical psychology really does need to embrace different, unfamiliar and competitive languages. In doing so, we can attempt to direct services towards collaborative, co-produced and therapeutic ends. This needs to be embraced in a reflexive manner, of

course, and with an ethical heart as its lowest common denominator. This project has provided me with a rich exposure to such an approach.

4.6.4 Asking of dreams

One discovery along the way for me, is that dreams seem pretty damn important – and yet how often do we ask one another about these, whether personally or professionally? If dreams can become shattered, and a loss so deep be simply unnoticed or remain unvoiced, it seems almost like we are devaluing those aspects of our identity that hold-on so dearly to those dreams. Giving permission for the dream to be witnessed could create the opportunity for a change to take place, or at least for someone to feel validated and heard. Indeed, if "what we call 'reality' resides and is expressed in one's description of events, people, ideas, feelings, and experiences," (Sluzki, 1992, pp. 218-219) then to ask of one's dream is to conjure up the possibility of it being realised, is it not?

A conclusion of sorts: The ending is the beginning

In designing this study, I considered a narrative approach appropriate to develop upon how the public and healthcare professionals understand lived experiences of parenting children with MD across the lifespan. The research questions considered the perceived impact of caring, loss and how events of enablement were made sense of. I have detailed how couples narrate the experience of parenting children with MD through making sense of loss, building new or different identities as parents, and through regulating a

partner's distress in storytelling. I have explored the dynamic retelling of closeness and distance in a way that circles the condition of MD, but permits resistance and survival in the context of societal discourses of parenthood and death. I have presented narratives of survival through the use of humour within stories and through retelling, in ways that foster empathy and compel audiences to establish their own position in relation to the narrated struggles that arguably surpass individual accounts. Ultimately, through narrated legacies and memories, a communal zeitgeist that surpasses individuals and death seemed to emerge. Perhaps this reflects the essence of narrative inquiry, in that it creates something beyond the immediate retelling; that "this process of narrative sharing…becomes a means of empowerment and potentially leads to forms of social action and change" (Murray & Sargeant, 2011, p.165).

References

Abbott, D. (2012). Other voices, other rooms: reflections on talking to young men with Duchenne muscular dystrophy and their families about transition to adulthood. *Children & Society, 26*(3), 241-250.

Abbott, D., & Carpenter, J. (2014). 'Wasting precious time': young men with Duchenne muscular dystrophy negotiate the transition to adulthood. *Disability and Society, 29*(8), 1192-1205. doi:10.1080/09687599.2014.916607

Andrews, M. (2007). Exploring cross-cultural boundaries. *Handbook of narrative inquiry: Mapping a methodology*, 489-511.

Andrews, M., Squire, C., & Tamboukou, M. (2013). *Doing narrative research*. London: Sage.

Aran, O., Biel, J.-I., & Gatica-Perez, D. (2014). Broadcasting oneself: Visual discovery of vlogging styles. *IEEE Transactions on multimedia, 16*(1), 201-215.

Åstedt-Kurki, P., Isola, A., Tammentie, T., & Kervinen, U. (2001). Importance of humour to client–nurse relationships and clients' well-being. *International journal of nursing practice, 7*(2), 119-125.

Bakerman, H. M. (1997). Humour as a nursing intervention. *Axone (Dartmouth, NS), 18*(3), 56-61.

Barker, C., & Pistrang, N. (2015). *Research methods in clinical psychology: An introduction for students and practitioners*. West Sussex: John Wiley & Sons.

Barry, C. A., Stevenson, F. A., Britten, N., Barber, N., & Bradley, C. P. (2001). Giving voice to the lifeworld. More humane, more effective medical care? A qualitative study of doctor-patient communication in general practice. *Social Science and Medicine, 53*(4), 487-505. doi:10.1016/S0277-9536(00)00351-8

Bateson, G. (1979). *Mind and nature: A necessary unity*. New York: Dutton.

Beale, B., Cole, R., Hillege, S., McMaster, R., & Nagy, S. (2004). Impact of in-depth interviews on the interviewer: Roller coaster ride. *Nursing & Health Sciences, 6*(2), 141-147.

Bedini, L. A. (2000). "Just sit down so we can talk:" perceived stigma and community recreation pursuits of people with disabilities. *Therapeutic Recreation Journal, 34*(1), 55-68.

Bendixen, R. M., & Houtrow, A. (2016). Parental Reflections on the Diagnostic Process for Duchenne Muscular Dystrophy: A Qualitative Study. *Journal of Pediatric Health Care, 31*(3):285-292. doi:10.1016/j.pedhc.2016.09.002

Bhaskar, R. (2013). *A realist theory of science.* Abingdon, Oxfordshire: Routledge.

Biel, J.-I., & Gatica-Perez, D. (2010). *Voices of Vlogging*. Paper presented at the AAAI International Conference on Weblogs and Social Media (ICWSM), Washington DC. Retrieved on the 16th September 2017 from: http://www.idiap.ch/~gatica/publications/BielGatica-icwsm10.pdf

Bjørnholt, M., & Farstad, G. R. (2014). 'Am I rambling?'on the advantages of interviewing couples together. *Qualitative Research, 14*(1), 3-19.

Bowman, T. (1999). Shattered dreams, resiliency, and hope:"Restorying" after loss. *Journal of Personal & Interpersonal Loss, 4*(2), 179-193.

Bowtell, E. C., Sawyer, S. M., Aroni, R. A., Green, J. B., & Duncan, R. E. (2013). "Should I send a condolence card?" Promoting emotional safety in qualitative health research through reflexivity and ethical mindfulness. *Qualitative Inquiry, 19*(9), 652-663.

BPS (2014). *Code of Human Research Ethics*. Leicester: The British Psychological Society.

Bruner, E. M. (1997). Ethnography as narrative. In: L. P. Hinchman & S. Hinchman (Eds.). *Memory, identity, community: The idea of narrative in the human sciences*, (pp. 264-280). USA: Suny Press.

Buchanan, D. C., LaBarbera, C. J., Roelofs, R., & Olson, W. (1979). Reactions of families to children with Duchenne muscular dystrophy. *General Hospital Psychiatry, 1*(3), 262-269. doi:10.1016/0163-8343(79)90028-8

Burnham, J. (2005). Relational reflexivity: a tool for socially constructing therapeutic relationships. In: C. Flaskas, B. Mason & A. Perlesz (eds.)*, The Space Between: Experience, Context and Process in the Therapeutic Relationship.* (pp. 1-17). London: Karnac.

Burnham, J. (2012). Developments in Social GRRRAAACCEEESSS: Visible-invisible and voiced-unvoiced. In: I.B. Krause (Ed.), *Culture and reflexivity in systemic psychotherapy: Mutual perspectives* (pp. 139-160). London: Karnac Books.

Bushby, K., Finkel, R., Birnkrant, D. J., Case, L. E., Clemens, P. R., Cripe, L., . . . Pandya, S. (2010a). Diagnosis and management of Duchenne muscular dystrophy, part 1: diagnosis, and pharmacological and psychosocial management. *The Lancet Neurology, 9*(1), 77-93.

Bushby, K., Finkel, R., Birnkrant, D. J., Case, L. E., Clemens, P. R., Cripe, L., . . . Pandya, S. (2010b). Diagnosis and management of Duchenne muscular dystrophy, part 2: implementation of multidisciplinary care. *The Lancet Neurology, 9*(2), 177-189.

Burton, J. (1978). The Camera as "Gun": Two Decades of Culture and Resistance in Latin America. *Latin American Perspectives, 5*(1), 49-76. doi:10.1177/0094582X7800500105

Capstick, A., & Clegg, D. (2013). Behind the stiff upper lip: War narratives of older men with dementia. *Journal of War & Culture Studies, 6*(3), 239-254.

Carey, M., & Russell, S. (2003). Outsider-witness practices: Some answers to commonly asked questions. *International Journal of Narrative Therapy & Community Work,* 3(1): 1-22.

Chamberlain, M., & Thompson, P. R. (1998). *Narrative and genre* (Vol. 1). London: Psychology Press.

Cunniff, A. L., Chisholm, V., & Chouliara, Z. (2015). Listening to Fathers of Sons with Duchenne Muscular Dystrophy. *New Male Studies: An International Journal, 4*(2), 5-23.

Daack-Hirsch, S., Holtzer, C., & Cunniff, C. (2013). Parental perspectives on the diagnostic process for Duchenne and Becker muscular dystrophy. *American Journal of Medical Genetics Part A, 161a*(4), 687-695. doi:10.1002/ajmg.a.35810

Dawson, S., & Kristjanson, L. J. (2003). Mapping the journey: family carers' perceptions of issues related to end-stage care of individuals with muscular dystrophy or motor neurone disease. *Journal of palliative care, 19*(1), 36-42.

Daykin, N., Byrne, E., Soteriou, T., & O'Connor, S. (2008). The impact of art, design and environment in mental healthcare: a systematic review of the literature. *The Journal of the Royal Society for the Promotion of Health, 128*(2), 85-94. doi:10.1177/1466424007087806

Dean, R. A. K., & Major, J. E. (2008). From critical care to comfort care: the sustaining value of humour. *Journal of clinical nursing, 17*(8), 1088-1095.

Dyregrov, K. (2004). Bereaved parents' experience of research participation. *Social science & medicine, 58*(2), 391-400.

Eiraldi, R. B., Mazzuca, L. B., Clarke, A. T., & Power, T. J. (2006). Service utilization among ethnic minority children with ADHD: A model of help-seeking behavior.

Administration and Policy in Mental Health and Mental Health Services Research, 33(5), 607-622.

Elliott, R., Fischer, C. T., & Rennie, D. L. (1999). Evolving guidelines for publication of qualitative research studies in psychology and related fields. *British journal of clinical psychology, 38*(3), 215-229.

Erby, L. H., Rushton, C., & Geller, G. (2006). "My Son Is Still Walking": Stages of Receptivity to Discussions of Advance Care Planning Among Parents of Sons With Duchenne Muscular Dystrophy. *Seminars in Pediatric Neurology, 13*(2), 132-140. doi:10.1016/j.spen.2006.06.009

Etherington, K., & Bridges, N. (2011). Narrative case study research: On endings and six session reviews. *Counselling and Psychotherapy Research, 11*(1), 11-22.

Ewing, B. (2009). Wish fulfillment: Palliative care and end-of-life intervention. *Pediatric nursing, 35*(2), 81-85.

Ferrell, B. R. (1995). The impact of pain on quality of life. A decade of research. *The Nursing Clinics of North America, 30*(4), 609-624.

Field, P. A., & Morse, J. M. (1985). Nursing research: the application of qualitative methods. Beckenham: Croom Helm.

Fisher, M., Kelly, R., Lawrie-Skea, A., Randall, J., & Wade, N. (2014). Involving people living with dementia in systematic reviews. *Generations Review, 7*, 27-31.

Fitzpatrick, C., & Barry, C. (1990). Cultural differences in family communication about Duchenne muscular dystrophy. *Developmental Medicine & Child Neurology, 32*(11), 967-973.

Frank, A. W. (1995). *The Wounded Storyteller: Body, Illness, and Ethics*. London: The University of Chicago Press.

Frank, A. W. (2006). Health stories as connectors and subjectifiers. *Health, 10*(4), 421-440.

Fredman, G. (2014). Weaving Net-works of Hope with Families, Practitioners and Communities: Inspirations from Systemic and Narrative Approaches. *Australian and New Zealand Journal of Family Therapy, 35*(1), 54-71.

Gagliardi, B. A. (1991a). The family's experience of living with a child with Duchenne muscular dystrophy. *Applied Nursing Research, 4*(4), 159-164.

Gagliardi, B. A. (1991b). The impact of Duchenne muscular dystrophy on families. *Orthopaedic Nursing, 10*(5), 41-49.

Gergen, K. (2009). *An Invitation to Social Construction*. Los Angeles: Sage.

Gergen, K. J. (1985). The social constructionist movement in modern psychology. *American psychologist, 40*(3), 266.

Gibson, B. E., Zitzelsberger, H., & McKeever, P. (2009). 'Futureless persons': Shifting life expectancies and the vicissitudes of progressive illness. *Sociology of Health and Illness, 31*(4), 554-568. doi:10.1111/j.1467-9566.2008.01151.x

Gillies, J., & Neimeyer, R. (2006). Loss, grief, and the search for significance: Toward a model of meaning reconstruction in bereavement. *Journal of Constructivist Psychology, 19*(1), 31-65.

Glaser, B., & Strauss, A. (1967). *The discovery of grounded theory*. London: Weidenfield & Nicolson.

Goffman, E. (1956). The Presenting of Self in Everyday Life. New York: Anchor Books.

Gravelle, A. M. (1997). Caring for a child with a progressive illness during the complex chronic phase: parents' experience of facing adversity. *Journal of Advanced Nursing, 25*(4), 738-745.

Guillemin, M., McDougall, R., & Gillam, L. (2009). Developing "ethical mindfulness" in continuing professional development in healthcare: use of a personal narrative approach. *Cambridge Quarterly of Healthcare Ethics, 18*(02), 197-208.

Hagan, T., & Smail, D. (1997). Power-mapping—I. Background and basic methodology. *Journal of Community & Applied Social Psychology, 7*(4), 257-267. doi:10.1002/(SICI)1099-1298(199709)7:4<257::AID-CASP428>3.0.CO;2-P

Hedtke, L. (2014). Creating stories of hope: A narrative approach to illness, death and grief. *Australian and New Zealand Journal of Family Therapy, 35*(1), 4-19.

Heidegger, M. (1996). *Being and time: A translation of Sein und Zeit*. New York: Suny Press.

Hinchman, L. P., & Hinchman, S. K. (1997). *Memory, identity, community: The idea of narrative in the human sciences*. USA: State University of New York Press.

Hollway, W., & Jefferson, T. (2008). The free association narrative interview method. In: L.M. Given (Ed.) *The Sage encyclopaedia of qualitative research methods.* London: Sage Publications.

Hooghe, A., Neimeyer, R. A., & Rober, P. (2012). "Cycling Around an Emotional Core of Sadness" Emotion Regulation in a Couple After the Loss of a Child. *Qualitative health research, 22*(9), 1220-1231.

Hosking, D. M., & Pluut, B. (2010). (Re)constructing reflexivity: A relational constructionist approach. *Qualitative Report, 15*(1), 59-75.

Hydén, L. C. (2013). Bodies, Embodiment and Stories. In M. Andrews, C. Squire, & M. Tamboukou (Eds.), *Doing Narrative Research* (pp. 126-141). London: Sage.

Hydén, L. C. (1997). Illness and narrative. *Sociology of health & illness, 19*(1), 48-69.

Hynson, J. L., Aroni, R., Bauld, C., & Sawyer, S. M. (2006). Research with bereaved parents: a question of how not why. *Palliative Medicine, 20*(8), 805-811.

Karlsson, G. (1993). *Psychological qualitative research from a phenomenological perspective*. London: Coronet Books.

Katz, S. (2002). Gender differences in adapting to a child's chronic illness: A causal model. *Journal of Pediatric Nursing, 17*(4), 257-269.

Kirkman, M. (2002). What's the plot? Applying narrative theory to research in psychology. *Australian Psychologist, 37*(1), 30-38.

Klass, D., Silverman, P. R., & Nickman, S. L. (1996). Continuing bonds. *London und Philadelphia*.

Koenig Kellas, J., & Trees, A. R. (2006). Finding meaning in difficult family experiences: Sense-making and interaction processes during joint family storytelling. *The Journal of Family Communication, 6*(1), 49-76.

Koenig Kellas, J., Trees, A. R., Schrodt, P., LeClair-Underberg, C., & Willer, E. K. (2010). Exploring links between well-being and interactional sense-making in married couples' jointly told stories of stress. *Journal of Family Communication, 10*(3), 174-193.

Kogan, S. M., & Gale, J. E. (1997). Decentering therapy: Textual analysis of a narrative therapy session. *Family process, 36*(2), 101-126.

Lather, P., & Smithies, C. (1997). *Troubling the angels: Women living with HIV/AIDS*. USA: Westview Press, Inc.

Lemmers, R. J. L. F., van der Vliet, P. J., Klooster, R., Sacconi, S., Camaño, P., Dauwerse, J. G., . . . van der Maarel, S. M. (2010). A Unifying Genetic Model for Facioscapulohumeral Muscular Dystrophy. *Science, 329*(5999), 1650-3. doi: 10.1126/science.1189044

Lester, A. D. (1995). *Hope in pastoral care and counselling*. USA: Westminster John Knox Press.

Llewelyn, S., Hardy, G., & Aafjes-van Doorn, K. (2015). Psychologists as researchers. In: J. Hall, D. Pilgrim, & G. Turpin (Eds.), *Clinical Psychology in Britain: Historical Perspectives* (pp. 154-167). Leicester: The British Psychological Society.

Lyons, E., & Coyle, A. (2016). *Analysing qualitative data in psychology*. London: Sage.

MacDougall, G. (1997). Caring—a masculine perspective. *Journal of Advanced Nursing, 25*(4), 809-813.

Malik, R., & Krause, I.-B. (2004). Before & Beyond Words: Embodiment & Intercultural Therapeutic Relationships in Family Therapy. In: C. Flaskas, B. Mason & A. Perlesz (eds.)*, The Space Between: Experience, Context and Process in the Therapeutic Relationship.* (pp. 95-109). London: Karnac.

Mason, B. (2005). Relational risk-taking and the therapeutic relationship. In C. Flaskas, A. Perlesz, & B. Mason (Eds.), *The space between: Experience, context and process in the therapeutic relationship.* (pp. 157-170). London: Karnac.

McCormick, E. W. (2012). *Change for the better: self-help through practical psychotherapy.* London: Sage.

Meyer, D. D. (2006). Parenthood in a time of transition: Tensions between legal, biological, and social conceptions of parenthood. *The American Journal of Comparative Law, 54*, 125-144.

Minister, K. (1991). A feminist frame for the oral history interview. In S. Gluck and D. Patai (eds.) *Women's words: The feminist practice of oral history*, 27-41. Abingdon, Oxford: Routledge.

Mishler, E. G. (1991). Representing discourse: The rhetoric of transcription. *Journal of narrative and life history, 1*(4), 255-280.

Morris, S. M. (2001). Joint and individual interviewing in the context of cancer. *Qualitative Health Research, 11*(4), 553-567.

Morrow, E., Ross, F., Grocott, P., & Bennett, J. (2010). A model and measure for quality service user involvement in health research. *International Journal of Consumer Studies, 34*(5), 532-539.

Moxley-Haegert, L. (2015). Leaving a legacy'and'letting the legacy live': Using narrative practices while working with children and their families in a child palliative care program. *The International Journal of Narrative Therapy and Community Work*(2), 58-70.

Murray, B. (2003). Qualitative research interviews: therapeutic benefits for the participants. *Journal of Psychiatric and Mental Health Nursing, 10*(2), 233-236.

Murray, M., & Sargeant, S. (2011). Narrative Psychology. In D. H. a. A. Thompson (Ed.), *Qualitative Research Methods in Mental Health and Psychotherapy : A Guide for Students and Practitioners.* London: Wiley.

Naylor, C., Das, P., Ross, S., Honeyman, M., Thompson, J., & Gilburt, H. (2016). *Bringing together physical and mental health*. London: King's Fund.

Neimeyer, R. A. (2012). Correspondence with the deceased. *Techniques of grief therapy*, 259-261.

Neimeyer, R. A., Baldwin, S. A., & Gillies, J. (2006). Continuing bonds and reconstructing meaning: Mitigating complications in bereavement. *Death studies, 30*(8), 715-738.

Nolte, L., Ragavan, R., Randall-James, J., & Wellsted, D. (2017). *Evaluation of Muscle Dream Experiences: A mixed-methods retrospective evaluation of the impact of the Muscle Help Foundation's Muscle Dream experiences for beneficiaries and their families.* University of Hertfordshire. Unpublished manuscript.

Omery, A. (1983). Phenomenology: a method for nursing research. *Advances in nursing science, 5*(2), 49-64.

Parkes, C. M., & Prigerson, H. G. (2013). *Bereavement: Studies of grief in adult life*. London: Routledge.

Pearson, R. & van Ommen, G. (1984) Recent developments in DNA research of Duchenne muscular dystrophy. In: L. Ten Kate, P. Pearson & A. Stadhouders (Eds.), *Research into the origin and treatment of muscular dystrophy* (91–100). Amsterdam, the Netherlands: Excepta Medica.

Poland, B. D. (2003). Transcription quality. In J. Holstein and J. Gubrium (eds.) *Inside interviewing: New lenses, new concerns* (pp. 267-287). London: Sage publications.

Riessman, C. K. (1993). *Narrative analysis: Qualitative research methods series 30.* Boston University: Sage Publications.

Riessman, C. K. (2008). *Narrative methods for the human sciences*. London: Sage Publications.

Samson, A., Tomiak, E., Dimillo, J., Lavigne, R., Miles, S., Choquette, M., . . . Jacob, P. (2009). The lived experience of hope among parents of a child with Duchenne muscular dystrophy: perceiving the human being beyond the illness. *Chronic Illness, 5*(2), 103-114. doi:10.1177/1742395309104343

Scotland, J. (2012). Exploring the philosophical underpinnings of research: Relating ontology and epistemology to the methodology and methods of the scientific, interpretive, and critical research paradigms. *English Language Teaching, 5*(9), 9-16.

Shakespeare, T. (2010). The social model of disability. In: L. Davis (Ed.), *The Disability Studies Reader* (pp. 266-273). New York: Routledge.

Shippee, N. D., Domecq Garces, J. P., Prutsky Lopez, G. J., Wang, Z., Elraiyah, T. A., Nabhan, M., . . . Murad, M. H. (2015). Patient and service user engagement in research: a systematic review and synthesized framework. *Health Expectations, 18*(5), 1151-1166. doi:10.1111/hex.12090

Shoshani, A., Mifano, K., & Czamanski-Cohen, J. (2016). The effects of the make a wish intervention on psychiatric symptoms and health-related quality of life of children with cancer: a randomised controlled trial. *Quality of Life Research, 25*(5), 1209-1218.

Sluzki, C. E. (1992). Transformations: A blueprint for narrative changes in therapy. *Family process, 31*(3), 217-230.

Smail, D. J. (2005). *Power, interest and psychology: Elements of a social materialist understanding of distress*. Ross-On-Eye: PCCS Books.

Spandler, H., Anderson, J., & Sapey, B. (2015). *Madness, distress and the politics of disablement*. Bristol: Policy Press.

Speed, B. (1991). Reality exists OK? An argument against constructivism and social constructionism. *Journal of Family Therapy, 13*(4), 395-409.

Squire, C. (2008). Experience-centred and culturally-oriented approaches to narrative. In: M. Andrews, C. Squire, & M. Tamboukou (Eds.), *Doing narrative research* (pp. 41-63). London, UK: Sage.

Steinbock, B. (2006). Defining parenthood. In M. Freeman (Ed.) *Children's Health and Children's Rights* (pp. 311-334). Leiden: Brill Publishers.

Tait, G. R., Schryer, C., McDougall, A., & Lingard, L. (2011). Exploring the therapeutic power of narrative at the end of life: a qualitative analysis of narratives emerging in dignity therapy. *British Medical Journal Supportive Palliative Care, 1*(3):296-300. doi: 10.1136/bmjspcare-2011-000051

Tajfel, H. (1982). Social psychology of intergroup relations. *Annual Review of Psychology, 33*(1), 1-39.

Taylor, B., & de Vocht, H. (2011). Interviewing separately or as couples? Considerations of authenticity of method. *Qualitative Health Research, 21*(11), 1576-1587.

Tomiak, E. M., Samson, A., Miles, S. A., Choquette, M. C., Chakraborty, P. K., & Jacob, P. J. (2007). Gender-specific differences in the psychosocial adjustment of parents of a child with duchenne muscular dystrophy (DMD) - Two points of view for a shared experience. *Qualitative Research Journal, 7*(2), 2-21. doi:10.3316/QRJ0702002

Torgé, C. J. (2013). Using conjoint interviews with couples that have been living with disabilities and illnesses for a long time–implications and insights. *Qualitative Studies, 4*(2), 100-113.

Tracy, S. J. (2010). Qualitative Quality: Eight "Big-Tent" Criteria for Excellent Qualitative Research. *Qualitative Inquiry, 16*(10), 837-851. doi:10.1177/1077800410383121

Trepte, S., & Loy, L. S. (2017). Social Identity Theory and Self-Categorization Theory. In: P. Rössler, C. Hoffner, & L. van Zoonen (Eds), *The International Encyclopedia of Media Effects.* London: John Wiley & Sons, Inc.

Waldboth, V., Patch, C., Mahrer-Imhof, R., & Metcalfe, A. (2016). Living a normal life in an extraordinary way: A systematic review investigating experiences of families of young people's transition into adulthood when affected by a genetic and chronic childhood condition. *International Journal of Nursing Studies, 62*, 44-59. doi:10.1016/j.ijnurstu.2016.07.007

Webb, C. (1992). The use of the first person in academic writing: objectivity, language and gatekeeping. *Journal of advanced nursing, 17*(6), 747-752.

Webb, C. (2005). Parents' perspectives on coping with Duchenne muscular dystrophy. *Child: Care, Health and Development, 31*(4), 385-396. doi:10.1111/j.1365-2214.2005.00518.x

Wells, K. (2011). *Narrative inquiry*. USA: Oxford University Press.

White, M., & Epston, D. (1990). *Narrative means to therapeutic ends.* New York: WW Norton & Company.

White, M. (1988). Saying hullo again: The incorporation of the lost relationship in the resolution of grief. *Dulwich Centre Newsletter, 3*, 29-36.

White, M. (2000). *Reflections on narrative practice: Interviews and essays*. Adelaide, Australia: Dulwich Centre Publications.

White, M., & Epston, D. (2005). Externalizing the problem. In C. Malone, L. Forbat, M. Robb, & J. Seden (Eds.), *Relating Experience: Stories from Health and Social Care* (pp. 73-78). USA: Routledge.

Witte, R. A. (1985). The psychosocial impact of a progressive physical handicap and terminal illness (Duchenne muscular dystrophy) on adolescents and their families. *British Journal of Medical Psychology, 58,* 179–187. doi:10.1111/j.2044-8341.1985.tb02632.x

Wooffitt, R. (1992). *Telling tales of the unexpected: The organization of factual discourse*. Lanham, UK: Rowman & Littlefield.

Yamaguchi, M. & Suzuki, M. (2015). Becoming a back-up carer: Parenting sons with Duchenne muscular dystrophy transitioning into adulthood. *Neuromuscular Disorders, 25(1)*: 85-93.

Yardley, L. (2007). Demonstrating validity in qualitative psychology. In: J. Smith (Ed.), *Qualitative psychology: A practical guide to research methods (2nd Ed),* (pp. 235-251). London, UK: Sage Publications.

APPENDIX

Appendix A: Systematic literature review results

Authors & Title	Type & Aim	Participants (Inc. *Country*)	Methods	Data Analysis	Results & Conclusions	Pros and cons
Webb, C. L. (2005) *Parent's perspectives on coping with Duchene muscular dystrophy*	Qualitative To explore resources and information relating to parents coping with DMD* *Only stated in abstract.	15 families: 7 mothers, 1 father, 7 parents conjointly. 17 sons with DMD overall, aged between 5-23 years . *USA.*	Recruited through 2 websites & conference presentation. Selected based on age of boys with DMD, geography & family demographics. 1-2hr semi-structured interviews with families.	Grounded theory analysis (Glaser & Strauss, 1967)	Six coping themes emerged from the data: 1) Genetics 2) Diagnosis 3) Reactions to diagnosis 4) Treatment 5) Equipment 6) School issues Several implications for professionals proposed to aid parental coping as disease progresses: a) trusting instincts, b) anticipating a grief response to diagnosis, and c) cooperative and collaborative efforts to (e.g.) obtain equipment and/or double-check understanding/retention.	*Pros:* Implications suggested are largely practice-based, and put the imperative for change on services rather than the parents themselves. Provides forum for sharing of information between parents on challenges faced over time. Personal experience interwoven into analysis, adding additional layers to findings. *Cons:* Aims are not explicitly stated in text. Genetics theme not explicitly linked to data. Personal experience sometimes overshadows gathered data. Similarly, 'grief process' model used but not all stages accounted for in data. No discussion of genetics or school

						issues, outside of results section. Analysis is largely descriptive, with no overarching narrative framework, theorising and/or evolving conceptual model.
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Gravelle, A. M. (1997) *Caring for a child with a progressive illness during the complex chronic phase: Parents' experience of facing adversity*	Qualitative To explore the experiences of parents caring for children with life-threatening conditions, at the 'complex	Sample size of 8, totalling 11 parents (8 mothers, 3 fathers). 4 fathers declined to be involved. 1 mother from single-parent family. Sons with life-threatening progressive disease aged between 26	Theoretical sampling used, recruited through Muscular Dystrophy Association of Canada and two community health departments. Two-phased interviews, lasting 35-120 minutes in total: 1.	Phenomenology (Omery, 1983)	'Facing adversity' (central theme) as a successive and cumulative process, comprised of 'defining' and 'managing' adversity, repeated along the disease trajectory. Central concepts of normalisation, loss, and 'women as caregivers' emerged throughout. In *defining adversity*, several themes emerged: 1. Individual condition 2. Individual family: Acceptance, Living with loss, Gaining strength, Magnitude of impact In *managing adversity*:	*Pros:* Implications are both person-focused and informed by disease trajectory. Considers implications for service management and respite provisions. Presents visual conceptualisation of data, presenting progressive nature of disease, alongside ever-changing need to redefine challenges and identify different ways of managing. Discussion reframes findings using common concepts throughout the data. Considers diversity and socio-cultural contexts (e.g. gender roles). *Cons:* The sample reflect a range of different life-threatening conditions

chronic stage' of their disease.	months and 16 years.\n\nDiseases included were (no numbers provided): DMD, spinal muscular atrophy, metachromatic leukodystrophy, Retts syndrome, cerebral palsy.\n\n*Canada*.	Unstructured, using trigger questions; 2. Semi-structured following first interview analysis, clarifying and validating findings*.\n\n*3/8 did not complete second interviews		1. Seeking information\n2. Planning/preparing\n3. Negotiating\n4. Utilizing resources\n\nImplications: assessments *must* be ongoing, how parents define and manage adversity changes over time, as does psychosocial/emotional impact. Community-based case management advocated.	and is not specific to muscular dystrophy.\nThe number of children diagnosed with each particular disease is not provided.\nNo explicit reasons given for why four fathers declined to participate and no hypothesising as to why this was the case.\nNo examples of 'trigger questions' provided, nor how these had been developed.\nEthics procedure not detailed.
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Dawson, S. & Kristjanson, L.	Qualitative\n\nTo	16 carers interviewed overall; 11 family carers	Semi-structured interview, based on	Content analysis (Field & Morse,	Three themes emerged from the data, that were similar across both DMD/MND:\n*Reactions and response*: Grieving	*Pros:* Inclusion of young people with DMD, with questions focused on anticipated challenges, adds a supplementary layer of

(2003) *Mapping the journey: Family carers' perceptions of issues related to end-stage care of individuals with Muscular Dystrophy or Motor Neurone Disease*	identify the needs of families with a child with DMD near the end of their lives. To identify issues for carers and possible solutions. Evaluate suitability of palliative care for	of individuals with DMD, 5 carers of individuals with MND. 6 families were bereaved (3 each from DMD and MND samples). 3 young men with DMD participated. *Australia.*	literature, experience and reference group consultation. Adapted over time to focus further on end-of-life issues in particular. Interviews lasted for 1-2hr each.	1985) And constant comparison techniques (Glaser and Strauss, 1967).	every day; Fearing each crisis may mean the end; Watching life in reverse. *Health system crossing points:* Getting lost in the system; Living with limits; I want to know but who do I ask? *Reaching forward:* Holding on to the big picture; Learning from other carers; Needing help to plan the future; Just getting on with it. These themes were recognised by health professionals, who noted staff challenges include: discussing dying/death; transitions from children to adult services; gaining access to palliative care; and the level/lack of understanding of these conditions in care settings. They did however find funerals and memorial services helpful.	understanding. A multi-perspective reference group has ensured a collaborative and meaningful approach to the research, resulting in credible findings with clinical relevance. Themes highlight the challenges faced by families (responses/reactions), their interaction with services (Health system crossing points), and opportunities for change (reaching forward). This paper suggests the need for physicians to 'cushion' physical/emotional deterioration that will occur with later stages of DMD, with palliative care measures: supporting caregiving processes, rather than just identifying problems. They suggest that this should be done in terms of 'future planning' – in order to anticipate increasing needs and prevent crises. *Cons:* The inclusion of carers of those with MND, introduces a lot of

						difference to the sample – particularly because these individuals were the spouses to adults (which is significantly different to the parents of young men with DMD. The theme 'grieving everyday' could have been further supported with participant quotes. It is not evident from the data that changes in living circumstances are construed as 'losses', which is the emphasized summary of other features of this theme. No limitations are provided. Further demographic information should have been provided (e.g. age of those with DMD vs. those with MND, which could have been quite different). No example questions from the interview schedule are provided.
families of young men with DMD.						
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Bendixen,	Qualitati	15 parents: 3	Recruited from	Thematic	An overarching theme of	*Pros:* The authors indicate the need

R. and Houtrow, A. (2016) *Parental reflections on the diagnostic process for Duchene Muscular Dystrophy: A qualitative study*	ve To explore the shared experience of being in receipt of a diagnosis of DMD. To gather the resources/ guidance provided for post-diagnosis.	men, 12 women. 12 participants were married or in long-term relationships, 3 were divorced/ separated. Relating to 15 boys living with DMD, diagnosed between 3 to 10 years prior. *USA.*	two ongoing studies at large academic facilities. 1hr semi-structured interviews over the telephone.	analysis (Lyons & Coyle, 2016)	'communication breakdown' emerged from the data. This depicted several challenges for parents: 1. Not feeling listened to about the symptoms they report (Dismissive); 2. Healthcare professional's misunderstandings and misperceptions of symptomology (Limited knowledge); 3. Feeling that diagnoses were carelessly delivered (Careless delivery); 4. And that providers do not communicate guidance (Lack of guidance). The authors conclude that despite 'medical progress over several decades', substantial barriers to acquiring a diagnosis and receiving appropriate guidance remain. One key barrier they identified relates to the lack of attention to parental concerns,	for further research that explores the paediatricians' experience of the diagnosis/treatment of DMD, in order to improve this service. The authors suggest some limitations with the study, including critical reflections over retrospective data (e.g. recall bias). Quality assurance checks were used to ensure accurate data during transcription process. Interview transcript reviewed by 'qualitative experts' for quality purposes. The authors expose continued difficulties in delivering timely diagnoses of DMD across services, over the past 30 years. *Cons:* The aims of the study were to explore 'shared' experiences, but it would appear only one parent per family was interviewed. As such, this may not actually represent a shared experience. Interview schedule was only reviewed by 'qualitative experts'

					which they suggest leads to diagnostic delays.	and not those with lived experience. Recruitment was from current studies, and so experiences may reflect a particular subset of families (self-selecting bias; e.g. those with a difficult experience only). Quotes were provided to demonstrate responses to questions, but associated themes were not provided.
Authors & Title	**Type & Aim**	**Participants (Inc.** *Country***)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Daack-Hirsch, S., Holtzer, C. and Cunniff, C. (2013) *Parental perspectives on the diagnostic process for Duchene*	Qualitative To identify the factors that influence the diagnostic process,	30 participants overall representing 25 families. 5 parents were interviewed together, 19 involved the mother only, and 1 interview was with a father.	Recruited muscular dystrophy clinics and through the Muscular Dystrophy Association. Six individual interviews over the telephone. Five group	Qualitative descriptive approach: two-staged. Identify themes, then map onto help-seeking model.	This study identified a range of facilitative and inhibitory factors within the three stages of the help-seeking model: problem recognition, decision to seek help, and service selection. In addition to these factors, the authors list further categories: parental explanations for perceived delays, value of earlier diagnosis, and suggestions for primary care providers.	*Pros:* This research elaborates on facilitating/inhibiting factors across all stages of help-seeking and at both the parent and physician level of interaction. The methods and analysis enable a strict focus on identifying pragmatic challenges and opportunities in the diagnostic process, which once identified may be modifiable in practice. Through distinguishing the family history status of participants, the

| *and Becker Muscular Dystrophy* | including which may be modified in order to reduce the time to diagnosis. | The sons with DMD were on average, diagnosed at the age of 4.6, but this differed between those with and without a family history (2.8 and 5.1, respectively).

USA. | interviews in person (two to eight participants). Participants received a list of topics to be covered. | | This research highlights that parents do not necessarily move through the help-seeking stages uni-directionally. Indeed, they found delays were possible at each stage of the model, with impeding factors prolonging progress through a stage or leading to families repeating previous stages. As such, in the case of DMD, the model is not so simple. If others did not corroborate concerns, families would not seek help but monitor for signs instead. When services cast doubts in the mind of parents, parents would regress from service selection to problem-recognition. Importantly, this research also highlights differences in the diagnostic process based on the individual' ethnicity; with black children being provided therapies but diagnosis not being pursued. | authors identify a general pattern of delaying help by those with a family history.
The influence of ethnicity on the diagnostic process is considered, highlighting physicians disproportionately treating but not diagnosing black children, as compared to white children.
The findings are often presented with a frequency for how often these factors were reported.

Cons: It is unclear how the interview schedule questions were designed (e.g. were service-users or experts in the field consulted?).
No example questions are provided in the paper.
Although Figure 1 indicates some hypothesized factors that are thought to influence movement throughout the stages, these are not returned to in the discussion.
Despite finding additional influential factors on how parents and physicians engage in help- |

						seeking that go beyond the current model, the authors do not attempt to produce an adaptation to the help-seeking model. Recruitment letters were on occasion sent by clinical directors, which could have influenced whether participants got involved in the study.
Authors & Title	**Type & Aim**	**Participants (Inc.** ***Country*****)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Samson, A., Tomiak, E., Dimillo, J., Lavigne, R., Miles, S., Choquette, M., Chakraborty, P., and Jacob, P. (2009)	Qualitative To describe the lived experience of hope among parents of a child with DMD	12 parents from nine families. 3 interviews represented both parents. In 7/9 families one boy with DMD. One father reported on experience raising two	Recruited through Neuromuscular Clinic. Staff in regular contact with child, approached parents. Semi-structured interview using a written 'question guide' lasting	Empirical Phenomenological Psychological analysis (Karlsson, 1993)	Three themes were identified within the experience of parental hope: Facing the loss; Learning to adapt; and Beyond the loss. Within these, the authors noted subthemes of Context; Coping; and Fabric of Hope. 1) Through facing the loss, parents were said to perceive of the illness as a threat to their child and way of life (context). This leads them to hope for a medical cure, coping in a passive manner – waiting (coping). Parent's hopes are concrete and specific: that	*Pros:* The findings indicate that healthcare professionals can be involved in developing contexts in which hope can be fostered; aiding adaptability and coping. The findings although not explicitly conceptualised along the disease trajectory, describe observed changes in the conditions of hope at particular developmental stages and thus suggest adaptation to these time/context-specific experiences. The authors detail the number of parents who declined to participate in the research.

The lived experience of hope among parents of a child with Duchene muscular dystrophy: Perceiving the human being beyond the illness		boys with DMD. One interview reflecting raising one daughter with DMD. One interview was retrospective, as son died 2 years prior to interview, aged 11. *Canada.*	approximately 90min.		medicine will provide a complete, definitive and immediate cure (fabric). 2) Through learning to adapt, parents talked of their own and child's sense of increasing difference from others and isolation (context). Despite the losses during this time, parents redefine normality, gain a sense of achievement, and the illness is no longer perceived of as an outside threat, but is incorporated into daily life (coping). Reflecting this, hope becomes focused on the positive and tangible affect one can have on a child's wellbeing in the here and now (fabric). 3) Beyond the illness, the relationship evolves, where parents become facilitators/educators in their child's growth as a person (context). Parents give meaning to the illness experience; more coherent (coping). In this sense, the child themselves become the source of the parent's hope – in	The authors provided a clear rationale for why researchers would interject/interrupt a participant during interview. The language used is emotive and resonates with the reader, reflecting the lived experiences of those children and families in similar situations. *Cons:* Some statements in the results and discussion appear unsubstantiated at times, and would have benefited from direct quotes from the data (e.g. references to the 'miracle'). The discussion unnecessarily repeats content described in the results section. It is unclear as to the composition of the interviews, as it details three interviews were with both parents together, but does not list which parent interviewed for the others (with the exception of the mention of one father). For example, ratios are reported rather than numbers

					their emerging individuality (fabric).	for a relatively small sample size.
Authors & Title	**Type & Aim**	**Participants (Inc.** *Country***)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Tomiak, E., Samson, A., Miles, S., Choquette, M., Chakraborty, P., and Jacob, P. (2007) *Gender-specific differences in the psychosocial adjustment of parents of a child*	Qualitative To explore how each individual parent experiences and adapts to living with a child with DMD. To explore the impact of	11 parents interviewed individually: In one instance, a father did not consent to interview (so 5 couples were interviewed separately, and one mother). Median age of mothers was 40.5, and fathers 47 years. 5 of the 6	Recruited through Neuromuscular Clinic. Staff in regular contact with child, approached parents. Semi-structured interview using a written 'question guide' lasting approximately 90min.	Empirical Phenomenological Psychological analysis (Karlsson, 1993)	This research identified five themes relating to: Care of the child; Coping styles; Partner relationship; Career, and Social support. In general, the parents felt overwhelmed and had little outside supports. Differences tended to relate to the respective roles of primary or secondary caregiver status, which with the exception of one relationship, was the mother as main caregiver. The father would typically oversee family functioning, provide psychological support and moral direction. Differences in relationships at first were seen as threatening the relationship, and	*Pros:* This paper adds to the literature on the lived experience of parents caring for their children with DMD through a closer look at gender-roles and differences in experience. The background literature discussed is comprehensive. The sample is described in great depth, situating individuals in the context of their personal, relational and cultural histories. A full interview schedule is provided. The authors use their data to suggest health professionals focus on the relational aspects of support that can foster an environment in which the child's wellbeing is equally considered.

with Duchene muscular dystrophy	the illness on career develop ment in mothers and fathers.	children affected by DMD were boys, one child was girl (aged 10-14, the median age of diagnosis was 4.5). 2/6 children were adopted. At time of interview, on average 8 years has elapsed since time of diagnosis. *Canada.*			served to isolate each parent from one another. Over time however, parents would learn to live with these differences, that could enrich their lives over time. Dialogue appeared to be the key to balancing these differences. Suggestions for practice include a focus on communication and the quality of parental relationships, as these were deemed to impact the wellbeing of the child. Additionally, practical support at time would free up the primary caregiver to network socially and time to nurture their couple relationship.	*Cons:* The explicit focus on the differences between parents appears problem-saturated at times, and experiences of commonality appear to be left under-researched or at least, under-recognised in this context. The sample is restricted in the sense that all families except one abode to traditional gender-roles scripts. Experiences are likely to be different depending on the stage of the disease (e.g. at the point of diagnosis, as opposed to 8 years of caring). The procedures and data presentation could be argued to proliferate gender role discourses, as divided interviews could well prime a context in which difference/disagreement is expected; not necessarily reflecting the lived experience. Limited sample to parents in the 'early and late wheelchair phase' but the rationale is unclear, particularly

						as no definition is provided as to what these phases represent. Findings relating to communication with child's sibling shared in discussion, but no data presented in findings (although this is acknowledged by the authors). Some unnecessary discussion on methods not used within the research project.
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Witte, R. (1985) *The psychosocial impact of a progressive physical handicap and terminal illness (Duchenne*	Qualitative To explore the communication process between children and their parents regardin	Six families of boys diagnosed with DMD, aged 13-16. The child were not diagnosed with any other physical or intellectual disability.	Recruited through a school for special educational needs. 'Extensive guided interview schedule' used. No further details	No information provided on how data presented was analysed. The paper appears to use a descriptive approach, with	This paper explores the parents' perceptions of their child's psychosocial development through their teenage years. The author describes an insecurity about unfamiliar others; a withdrawal and fear The author considers the concept of shame as key to the child's engagement with wider social networks. The author suggests that "the parents also contended with insensitivity issuing from the	*Pros:* This paper appears to be the first UK study to address the needs of parents to young people with DMD. The experience of the child is explored through the psychosocial and relational impact on parents, eliciting psychological hypotheses about function and adaptability. The paper provides several case examples, with associated quotes, in order to demonstrate the family's experience and contextualise the author's interpretations.

muscular dystrophy) on adolescents and their families	g DMD and death (though no aim explicitly listed)	*UK.*	provided.	psychoanalytic theorising to link the experiences of the families.	physically normal world" (p.181). In short, Witte notes that parents' enjoyment of the outside world is limited, and they are 'overtaken' by depression and guilt. Parents were then said to 'project' this guilt into anger directed at the outside world. As such, guilt was said to bind the family together. The author describes the importance becoming the caregiver for the mother's identity, and that the terminal nature of the disease meant a death of any meaningful future. As such, parents were said to often 'detour' any conversations around their child's future, and closed down dialogues.	*Cons:* Unconventional research paper format which omits significant details. For example, there is an inadequate description of the recruitment, sample, methods and analysis procedure. The researcher appears to apply a particular psychoanalytic lens to their discussion and analysis, but do not address this or reference this in the paper. An acknowledgement of the researcher's epistemological stance will have aided the paper, particularly as language appears to be used in an assumed and non-critical manner (e.g. 'normal peers'; 'replacement children').
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Cunniff,	Qualitati	15 fathers,	The sample	Grounded	Four key themes (and several	*Pros:* This study used an inductive

A., Chisholm, V., and Chouliara, Z. (2015) *Listening to fathers of sons with Duchene muscular dystrophy*	ve To explore father's perspective on caring for a son with DMD.	aged 34-60 (mean 48.4), of sons aged 8-32 (mean 16.1). This sample included one father who had two sons with DMD, one whose son has died, and another who was described as the sole-carer. 55 fathers also contributed through a written comments sheet. *Uk.*	was recruited through four different national charity organisations. Eight interviews were conducted in person lasting approximately 80 minutes. Seven interviews were completed over the telephone, lasting approximately 45 minutes. An interview schedule was developed around experiences	theory (constructivist interpretation) (Glaser & Strauss, 1967)	subthemes) emerged relating to fathering sons with muscular dystrophy: 1) Loss and acceptance; Loss, expectations, guilt, adaptive coping and acceptance vs. maladaptive coping. 2) Support versus isolation; Identity issues, strained friendships, family/marital stress, barriers to involvement; 3) Fight for resources; Frustration, spare part/exclusion, needs and suggestions; and 4) Race against time; Images of next stages: transition to adulthood: comparison with other children, deterioration and making the most of life, decisions, and talking about death. Fathers found transition periods particularly challenging, particularly in the context of previously held hopes and expectations around becoming a father. 'Issue fatigue' captured the 'perpetual stream of things to	approach to understand the experience of fathers to children with muscular dystrophy, who have often been overlooked in research due to primary caregiver roles. This is the first study to look specifically at the experience of fathers, and one of few studies to be conducted within the UK. The research evidences the need for family interventions that encourage paternal involvement, anticipate transitions and offer support at critical periods. This research not only describes the challenges faced by fathers in 'the fight for resources', but also the suggestions they made. *Cons:* It was not made clear what findings/quotes were evidenced in the interviews or written comments. No conceptual framework in order to understand the data was developed (which grounded theory is well suited). There is no discussion about how

| | | | and perceptions of specific areas. A comments sheet was used to collect data from the 55 fathers not interviewed. This sheet was a simplified adaptation of the interview guide. | | deal with' and with little opportunity to talk about these things with friends or peers (due to gender perceptions). They felt isolated from routines and from professionals; feeling surplus to requirement by staff. Within the family, conflict could arise due to these differences. There was also a desire for speed, in which appointments were not offered quickly enough, and fathers wanted to 'cram in' as many activities for their sons within their shortened timeframe. Some fathers addressed the dying process with their sons, whereas others did not feel equipped/read to discuss this – and so avoided the issue.

In the context of a cycle of loss model, this research adds an additional loss: A re-evaluation of previous expectations for continuing 'the family name'. They also note that some fathers | the sample size was chosen or concluded (e.g. at the point of data saturation). Some of the demographic information was missing for participant 13. |

185

					lived in anticipation of the next loss, and so as means of self-protection would not become too close to their sons.	
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Yamaguchi, M. and Suzuki, M. (2015) *Becoming a back-up carer: Parenting sons with Duchene muscular dystrophy transitioning into adulthood*	Qualitative To examine the process of change in parental behaviours during the child-adult transition of their sons, diagnosed with	14 mothers and four fathers (mostly aged 50-59), all with one son with DMD (Median age 22.1). 17 of these boys started using mechanical ventilators at point of transition. Six of these families also had other	Theoretical sampling approach: two MD associations, two independent living centres, association for wheelchair-football, and specialised MD hospital. Semi-structured interviews, lasting 34-106 minutes.	Grounded theory (Glaser & Strauss, 1967)	This paper describes three superordinate themes that relate to 1) emotional / 2) physical / 3) determination domain behaviours, as mapped onto time (age). 1) Parents tried to *sympathise* with their sons, recognising that they were susceptible to embarrassment, disappointment, irritation and anxieties. Parents were *encouraging* at every available opportunity, eager to build their son's 'spiritual strength' to overcome obstacles/barriers. 2) Parents needed to *provide care* at first, but as their sons grew older, four other behaviours	*Pros:* The authors developed a conceptual framework that maps themes onto age. The researchers employed quality checks to ensure themes generated were reflective of the data, which included revisiting interviewees for member feedback. An example theme is clearly demonstrated through a series illustrative quotes in Table 4. Full interview schedule provided. The researchers used a 'forward and back-translation' approach to ensure that the participant's intended meanings were maintained. *Cons:* The model is designed around

DMD.	children. *Japan.*	Purposeful to invite *one* parent only (main caregiver). Second interviews were held if disagreements on coding (15-48 minutes). 14 participants had second interviews. Observations of parent-child interactions at least once, looking at provided care, support and communication. Field notes were taken. In combination, over 200 hours		became more common. This involved *handing over* complete and substantial management of physical and medical support to their sons over time. As sons became more independent, this meant that parents needed to teach and *entrust* others who would follow their sons (e.g. care-worker). Parents lived with a sense of 'impending crisis' and so were ready to *provide backup* at any point. Parents did not wish to intrude upon their sons care, but had to maintain a degree of *understanding* around changes to his health (due to being a backup). 3) *Advocating* their child's needs, became particularly important in their son's teenage years. Despite this, parents also felt it was important to *sway* their sons decisions when necessary. *Supporting self-determination* from a young age was important to the parents, particularly as	timeframes and transitions, but this is based on the theoretical sampling of 15-30 year olds – transitions in other families may take place at different time points, including outside of these age ranges. This is acknowledged. The researcher took the perspective that they would gather all the information necessary, from only inviting one parent to interview – thus it could be argued that this study only captures 'one story' of parenting (rather than the couples story). The findings relate to a Japanese sample, and so questions could be asked as to how comparable cultural and social aspects of family life are to UK and USA samples. However, at face value, these findings appear consistent with much of the other literature.

					their son becomes more independent, yet his condition more dependent on others. Finally, parents also talked about *respecting independent determination* in their sons. In their analysis then, the authors identified three problems in parents 'becoming backup carers': psychological support, the aging of parents (in relation to changing/complex health needs), and parents intervention in the self-determination of their child (e.g. medical advocates for when son's opt for end-of-life care and parents struggle with this, as research suggests the parents will try and *sway* their sons).	
of data was collected.						
Authors & Title	**Type & Aim**	**Participants (Inc. *Country*)**	**Methods**	**Data Analysis**	**Results & Conclusions**	**Pros and cons**
Gagliardi, B. (1991)	Qualitative	Three families Fathers were	Ethnographic / naturalistic case study	Although not named, but referenced,	Six themes emerged that describe the families experience, under three 'stages':	*Pros:* Triangulation, using a mixed methods of ethnographic observation, participation and

*The family's experience of living with a child with Duchene muscular dystrophy** AND *The Impact of Duchene Muscular Dystrophy on Families** *Both papers report the same findings	To explore the social interactions within families, the problems associated with DMD, and the family's adaptive responses to it.	all 30 years old, the mothers were aged 26-30. Only one family had a history of DMD. Their sons were diagnosed with DMD, aged between 7 years and 10 months to 9 years and 11 months. Only one had any siblings (3 sisters). All had similar age of onset.	method. Weekly visits over a 10-week period. The research was involved in a multitude of activities with families and kept a reflective account, acknowledging her own emotional experiences also. Interviews were also conducted, twice over the 10 week period and then a third time 1 year later.	much of the Grounded Theory approach to analysis was used (e.g. comparative method, saturation, memo-taking, etc.)	1) Recognition. For all parties, the inevitability of progressive physical deterioration and increasing pain, led to a *disillusionment*; the illusion of normality fades, leaving people disappointed, left out, sad, angry, cheated and frustrated. Families also found that *society confirms the impossibility of normalcy* (through ineffective access strategies, or the reminders of difference in social gatherings with non-disabled children). 2) Working out. There were particular *dynamics of the family (Who's disabled anyway?)* whereby mother's encouraged dependency, potentially due to the guilt of being a carrier of the DMD gene – and fathers would work, for financial aids and to avoid seeing their son in pain. As the disease progressed, *a smaller world* emerged – whereby self-imposed boundaries looked to control as much as possible; e.g.	interview – including a follow-up interview over a year later. This was an extensive piece of work that purposely used a small sample in order to focus on the experience of the family. There was a degree of member-checking, whereby themes identified in earlier interviews or engagement could shape subsequent interview schedules. The authors sought independent audit of their coding schemes, reaching over 90% agreement on the emerging themes. The authors consider issues of credibility in the data. For example, a search for 'negative cases' that were the exception to the identified themes. *Cons:* Although ethnography acknowledges the participation of the researcher within the observations, there is no reflection on epistemological assumptions in order to position the researcher's

| | | | The ethnographic emersion and interviews were not prefixed, instead, capped once data saturation was reached. | | families did not talk about the disease, and sons would withdraw to their rooms, engaging in isolative activities. 3) Resolution. During this phase, families recognised that life must go on within and outside of the family; *letting go or hanging on* – boys became more self-sufficient, which ran counter to their parent's efforts to protect them. At this point, parents became much more involved with outside activities for themselves. *Things must change:* parents became much more accepting of the disability – immersing their sons into related activities, special educational-needs schools, etc. Overall, the families became much more involved and linked in with other families in similar situations.

Several suggestions are made: disease-phased consultations/counselling, | use of reflexivity throughout. The sample size is small, but as described above, methodologically justified. However, this cannot be considered as generalizable to other family's situations (but does not claim to). The provided example 'analytic memo' could have been expanded upon, to elaborate on some of the latter interpretations made in the findings section. The coding scheme is quite descriptive and does not reflect the presenting analysis (different themes are used). It is unclear how these themes progressed and developed into the more analytical and relational themes discussed. |

| | | | | | greater education for nurses, a role in educating the public, a need to develop mechanisms that maximise similarities rather than differences between disabled children and their nondisabled peers, at the same time as groups/initiatives that strengthen the bonds within any disability-orientated networks, and for peer support groups for parents (to share and manage many of their struggles). | |
Authors & Title	Type & Aim	Participants (Inc. *Country*)	Methods	Data Analysis	Results & Conclusions	Pros and cons
Buchanan, D., LaBarbera, C., Roelofs, R., and Olson, W. (1979) *Reactions of families*	Qualitative To determine the nature of psychological distress and	23 mothers (aged 24-40: 32.9 mean) with a child diagnosed with DMD (aged 4-14, mean 7.9). 1 mother was a stepparent. Two fathers	Recruited from DMD clinic and interviews conducted within the clinic, lasting for approximately one hour. No details on	No reference to analysis approach, however some descriptive analysis is clear within the results	Unclear as to the analysis approach, but the subheadings relate to *Major identified problem* - 76% of the families identified as having a psychological problem, and another 16% identified having physical difficulties relating to care procedures, such as lifting. Although they did not regret having their child, 84% said that they would recommend	*Pros:* This was one of the earliest papers to address the family experience, and did so in a comprehensive manner. Despite a focus on the family overall, this study also sought to collate individual experiences within the family (i.e. child, siblings), and other systemic factors (i.e. marital relationship, external services). This paper makes some initial links

to children with Duchene muscular dystrophy	coping styles in families living with DMD.	were also interviewed. *USA.*	how interview schedule was developed or the questions asked.		other carriers not having children. *Family history* was said to allow parents to anticipate diagnosis and thus reduce the psychological impact, as contrasted by those without history, who expressed (e.g.) shock, numbness. Some of latter group coped through use of psychiatric meds (x5), psychotherapy (x1). One father developed heart disease, another had psychiatric admission, one mother attempted suicide, and two other mothers 'abandoned' their families. 6 parents reported *marital conflicts*: with arguments based on caring, discipline, constant fatigue and interference from the respective partner. Another 6 were divorced, with three assigning the reasons as caring for a son with DMD. Four of the parents reported a strengthening of their family, whereas the other	in the described experiences, for example, 'not talking' about the disease was linked to the child's internalised blame for being 'weak' – yet the proposal for support in disclosure is suggested as a means to communicate that such weakness is beyond the child's control. From the research, practical changes were made to the participant's situations in order to assist (e.g. parental education, school placements, marriage counselling). *Cons:* There is no description of the method of analysis used. There are no details on how the interview schedule was designed. The study interviewed predominantly mothers, but generalises all findings to the 'family experience' rather than question whether this is equally applicable to the father's experience. This could have been done through a preliminary comparison with the

					12 reported that things were reasonable. Fathers were said to experience diagnosis as "a crushing blow to their expectations of their son".	two accounts made by fathers within the study or through data checking with respective interviewee's spouses.
					The core *coping strategies* noted included defensive manoeuvres; refusing to accept the diagnosis and seeking second-opinions; magical thinking (son as different to other DMD cases, would not progress like others, maybe recover completely). Thirteen families also used over-protection to cope (e.g. isolating the child; a conscious lack of discipline). *Siblings* were also invested in this overprotection and experienced a degree of emotional distress themselves. It was noted that there was often competitiveness between male siblings. *Personal:* Only one child from the study knew that DMD was a terminal condition and tried his best to philosophize, but	Very biomedical and non-person centred language at times, such as "DMD children…are new mutants". There was a range of different experiences reported throughout, and so reporting on the percentages of the relatively small sample may not have been necessary.

					was often moody and irritable: reflecting the lack of sexual expression, impending death, lack of attractiveness, physical immobility, and perception of critical and negative attitudes from others towards him. Parents tended to allow children to believe that they would get better in the future.	
					In general the *extended family* were a great resource, but on two occasions, grandparents were perceived as overly intrusive. All 18 children who were in school, experienced problems in obtaining their *education* based on their condition. Mothers felt that special educational needs schools were more individualised, but orientated towards play rather than academia.	
					The most 'well-adjusted' families were a) open communication	

					between mother-father, b) orientation towards present activities and accomplishments, c) organised/routine form of recreation for parents, and d) supportive institution outside of family.	

Appendix B: Examples of quality evaluation of literature reviewed

Included in this appendix is an extract from a detailed table summarising my quality evaluation of the literature.

This is followed by text that presents an overview and summary.

Authors	a)	Publishability guidelines shared by both qualitative and quantitative approaches					
Title	b)	Publishability guidelines especially pertinent to qualitative research					
	Explicit scientific content & method	**Appropriate method**	**Respect for participants**	**Specification of method**	**Appropriate discussion**	**Presentation**	**Contribution of knowledge**
Gravelle, A. M. (1997) *Caring for a child with a progressive illness during the complex chronic phase: Parents' experience of facing adversity*	Yes: Explicit reference to background literature, aims, and introduction to methods. Conventional format in terms of journal article structure used.	Yes: An inductive approach that addresses 'experiences' was used in this case, consistent with research aims.	No: Ethics procedure not detailed, inc. issues relating to consent and anonymity. No information on how stakeholders were involved in research design or monitoring (e.g. service-users providing feedback on information sheets, consulting on trigger questions).	Yes: Methods are clearly defined, described and operationalised. Sampling and recruitment described (MD Association, community teams). Methods described (two phased interviews: unstructured to semi-structured). Analysis described and referenced: Phenomenological	Yes: The discussion is appropriate, based on the findings, alongside some reflexive critiques (e.g. gender roles). No discussion of limitations/future research however.	Yes: Thematic data is visually conceptualised in two figures; representing a dynamic flux of defining and managing challenges, along a disease trajectory.	Yes: This paper adds to the research literature, making similar findings, alongside novel and useful contributions to the field. A critical lens is also applied to existing knowledge, enabling one to reframe understanding of 'coping' as facing adversity.

			approach.			
Owning perspective	**Situating sample**	**Grounding in examples**	**Providing credibility checks**	**Coherence**	**General vs specific**	**Resonating with readers**
No explicit reference to epistemology or researcher role within this (e.g. reflexivity).	Sample is described in terms of marital status, sex (i.e. mothers, fathers), age of children, and associated conditions (though not frequencies). Exception noted in on single-parent (divorced) family. Also noted that four fathers of participating families,	The author frequently refers themes back to participant quotes.	The second-interviews (5/8 parents) "served purpose of clarifying information from initial interviews, and validating findings" (p739). One possible route to checking research credibility is to return to interviewers and to assess the researcher's understanding of the data. Triangulation was not used, no other analysts were	The findings are presented within a clear and coherent conceptual framework, with a convincing overall narrative of 'facing adversity'. Themes are presented with multiple layers of analysis, with noticeable and important threads been developing in the discussion. The research narrative is extended and complemented by	This study represents a general focus in so much as it considers families facing adversity as parents, in the context of a range of life-threatening diseases. The specificity of this research is in its focus on the 'complex chronic' stage of disease trajectory. Despite this, there is no	Many aspects of the paper may resonate with the reader; particularly in terms of the proposed successive and cumulative dynamic in 'facing adversity'. These themes appear to accurately reflect the transcript content, and yet expands an appreciation for what it must be like to have the lived experience described in the text.

		declined involvement in interviews. Local geography briefly noted (e.g. British Columbia mainlands). No other diversity demographics reported.		used for credibility checks, and only one method of data analysis was used.	Figures 2 and 3.	recognition of the limits to this research – which could have highlighted new directions for research to take (e.g. the impact of mothers sharing caring vs. not having a choice).	
Dawson, S. & Kristjanson, L. (2003) *Mapping the journey: Family carers' perceptions of issues related to end-stage*	**Explicit scientific content & method**	**Appropriate method**	**Respect for participants**	**Specification of method**	**Appropriate discussion**	**Presentation**	**Contribution of knowledge**
	Yes: Explicit reference to background literature and a clear emphasis on the aims of the study. Clear discussion on sampling/recruitment, data collection and ethics. However, no details on how the literature search was	Yes: A content analysis was used, with credibility checks from Grounded Theory – enabling for both content and	Yes: The researchers sought ethical approval from three different boards. Participants were reassured that they have the right to withdraw from the study at any point, and guaranteed anonymity	Yes: Methods are clearly defined, described and operationalised. Sampling and recruitment described (MD/MND Associations, primary care facilities).	Yes: The discussion is appropriate, with a clear focus on implications for practice. This section, although brief, summarised some of the key findings in the context of	Yes: The content analysis provides an opportunity for a broad range of themes to emerge, including pragmatic and	Yes: This paper adds to the research literature, highlighting similar findings to the literature, but appropriating this to the context of end of life care. This is multifaceted, yet

care of individuals with Muscular Dystrophy or Motor Neurone Disease	conducted. Conventional format in terms of journal article structure used.	experience to be analysed in the study, with practical conclusions. Due to the qualitative nature of the research, appropriate checks were put in place, such as the reference group and second analyser.	throughout all dissemination. There was clearly significant stakeholder involvement throughout the process, in terms of the reference group – and so the research was designed around a sensitivity and respect for those with lived experience.	Methods described: semi-structured, and how these were developed. However, examples are omitted. Analysis described and referenced: Content analysis and 'constant comparison techniques (Grounded Theory).	practice and some literature (highlighting similarities and differences). No discussion of limitations/future research however.	experiential elements of caring. This is presented clearly through the core three themes. The thematic data is not conceptualised in relation one another, and so no common underlying theory is used to present these themes in conjunction with one another. However, based on the content of these findings and the	practically-orientated, through the inclusion of those already involved in a palliative care process (MND) and healthcare professionals, in order to compare and contextualise the findings. Of particular interest, is the finding that timing is essential in order to 'cushion' emotional and physical deterioration in a family unit during these times of significant change (e.g. conceived losses, listed on page 39 of the paper).

	Owning perspective	Situating sample	Grounding in examples	Providing credibility checks	Coherence	General vs specific	Resonating with readers
						purposes for evaluative feedback to an already existing process (i.e. palliative care), this seems justified and appropriate.	
	No explicit reference to epistemology or researcher role within this (e.g. reflexivity).	The sample is described in relation to their role in relation to the individual diagnosed with DMD or MND. Namely, whether a family carer, parent, spouse or	The authors frequently refer themes back to participant quotes throughout the text, with the exception of the interviews with the healthcare professionals. The data relating to the healthcare professionals is descriptive, rather	To aid claims of credibility, an eight-membered reference group of service-users, health professionals and representatives from MD/MND associations was used. This group was used to help enhance	The findings are presented using three themes that depict the family unit, their interaction with services and what the future opportunities and challenges may be for them. These are presented in	This study represents a specific focus in so much as it considers end of life care for families of those with DMD and MND. It is however general, in the sense that it	This paper presents three themes pertinent to a family's journey to considering end of life care for their loved ones. These themes when read in succession with one another, depict a struggle for families across multiple domains

| | | young person themselves. Their situation is described in terms of time: current situation or a retrospective account due to bereavement. The sample is situated further by the context of recruitment through a primary care facility and DMD/MND Associations.

No further details are provided (e.g. socio- | than exploratory and no quotes are used to support these statements. | trustworthiness of findings; making results more meaningful and comprehensive. They assisted with issues of sampling, interview guide design and synthesis of results. For example, one reference group member coded the interviews independently to ascertain closeness of coding by two researchers.

Although, triangulation was not used, the authors | succession to one another, but could have benefits from an overall conceptual framework – particularly in the context of a 'journey' as described in the title of the paper. Having said this, there is a coherent narrative through the paper that leads to practical implications. | explores and describes the families experiences, alongside perceived and pragmatic concerns. | and will resonate with the reader as struggles internal to the family, specific to the services, and related to uncertain futures are presented. |
|---|---|---|---|---|---|---|

		economic status, ethnicity).		complemented the content analysis with comparison checks used in Grounded Theory – aimed at testing the credibility of the findings at the level of themes and corresponding raw data.			

Guidelines especially pertinent to qualitative research

Owning one's perspective

All of the papers included in the systematic review failed to explicitly detail their epistemological assumptions in writing. This includes Gagliardi (Gagliardi, 1991a, 1991b), whose ethnographic study involved her own participation in activities with families and Witte's (1985) paper that appears to use a psychoanalytical lens throughout, but without the transparency of naming this. This lack of 'owning one's perspective' was also the case in Webb's (2005) paper, whereby she includes her own personal experiences in relation to MD in the findings and discussion. In this paper, Webb does not explicitly present her epistemology or personal relationship to MD for the reader to assess in what ways this has influenced the design, implementation and analysis. Overall, none of the papers adequately addressed the self of the researcher.

Situating the sample

Most of the research included adequately situated sample descriptions. Witte's (1985) paper is the exception in this case, as he only provides the age range, sex, diagnoses and geographic location of the sample. There was variation in the extent to which authors situated their sample, but most detailed the age of parents, the age and number of children in the household, sex, familial role (i.e. mother, father, sibling), and geography. Daack-Hirsch and colleagues (2013), Gagliardi (Gagliardi, 1991a, 1991b) and Bendixen and Houtrow (2016) were the only authors to report on ethnicity. Only two studies reported on location and household income, and educational background of participants (Bendixen & Houtrow, 2016; Samson et al., 2009). Other diagnosis-specific information was also presented, such as age at the point of diagnosis and family history. All papers detailed where the sample were recruited from and so provide the context in which

individuals were approached to participate. Interview configurations were often omitted from descriptions, meaning that it was unclear as to whether parents were interviewed conjointly, separately or just one parent deemed representative of the couple perspective. However, Samson and colleagues (2009), and Tomiak and colleagues (2007) explicitly detailed this.

Grounding in examples

All of the papers included in this review used quotes to illustrate points throughout. Witte (1985) focuses on six case studies in his paper and uses quotes from all families. One paper did not use any quotes for a theme entitled 'genetics' and no explanation for this is provided, and instead, the author uses their own personal experience to discuss this (C. Webb, 2005). Overall, the selected papers used direct quotes in an appropriate manner and themes were substantiated by reference to this 'raw content' (i.e. talk within the interview).

Providing credibility checks

There were a variety of credibility checks reported by the range of authors. This included member-checking following interview (Gravelle, 1997); stakeholder consultation groups (Bendixen & Houtrow, 2016; Dawson & Kristjanson, 2003); two or more coders (Daack-Hirsch et al., 2013; Dawson & Kristjanson, 2003; Tomiak et al., 2007); data-saturation methods (Bendixen & Houtrow, 2016; Gagliardi, 1991a, 1991b); constant-comparative method (Cunniff et al., 2015; Gagliardi, 1991a, 1991b; C. Webb, 2005); triangulation (Gagliardi, 1991a, 1991b); and searching for exceptions to the coding schemes (Gagliardi, 1991a, 1991b). No credibility checks were either employed or not reported by Witte (1985), Buchanan and colleagues, (1979), Webb (2005), nor Samson (2009).

Coherence

Most papers presented their findings coherently using the themes that emerged from the data to lead into further discussion around the personal and clinical implications of these. All papers were satisfactory in terms of presenting a coherent narrative thread to their findings, with notable strengths in Gravelle's (1997) conceptualisation of 'facing adversity', Samson and Kristjanson's (2009) 'mapping of [the] journey', and Waldboth and colleagues' (2016) conceptualisation of the individual/parent/sibling perspectives following a systemic literature review.

Accomplishing general vs. specific research tasks

Most of the papers reviewed accomplished general research tasks, in that they explored a target population in particular situations, but aimed to apply their findings to other similar contexts (the applicability of the findings to other contexts). Where authors described the implications of the findings on clinical practice and proposed improvements, there was no explicit caveat about the possible over-generalising of these findings – particularly considering the relatively small sample size of most of the studies. However, most of these findings were corroborated across the different papers (e.g. parental interviews at key transitions/stages of the disease trajectory), suggesting that the findings have not been generalised inappropriately.

Resonating with readers

I believe that the quality assessment of resonation should be read in conjunction with the epistemological assumptions and position of the researcher exploring this quality. What resonates for one reader, may be very different for others. As such, this aspect of

evaluation is proposed to be assessed in conjunction with all other domains of quality. Most studies raised a multitude of concerns that captured the complexity of parenting and caring for children living with MD. I found the papers that were more collaborative in nature (e.g. the mixed-methods ethnographic study by Gagliardi, 1991a, 1991b), that conceptualised beyond loss (e.g. Gravelle, 1997), were the papers that resonated with me the most for they had clear implications for all audiences.

Appendix C: Evidence of ethical approval

UNIVERSITY OF HERTFORDSHIRE
HEALTH & HUMAN SCIENCES

ETHICS APPROVAL NOTIFICATION

TO

CC

FROM   Dr Richard Southern, Health and Human Sciences ECDA Chairman

DATE   07/04/2016

Protocol number: cLMS/PGR/UH/02353

Title of study: Narrative accounts of being a parent following the death of a child to muscular dystrophy

Your application for ethics approval has been accepted and approved with the following conditions by the ECDA for your School.

Approval Conditions:

Please provide a brief protocol for how potential distress during the interview(s) will be dealt with to hhsecda@herts.ac.uk for approval by the Chairman, prior to recruitment and data collection.

This approval is valid:

From: 07/04/2016

To: 31/1/02017

Confirmation following conditions met:

Thank you for your submission. The Chairman has confirmed that the conditions for Protocol number cLMS/PGR/UH/02353 *Narrative accounts of being a parent following the death of a child to muscular dystrophy* have been met for full approval of your project.

Please retain this email together with your approval notification (attached for your reference).

Appendix D: Participant information sheet

BEING PART OF THE MUSCLE DREAM

The Parents' Story

PARTICIPANT INFORMATION LEAFLET

An invitation to participate in a research project from the University of Hertfordshire, working with The Muscle Help Foundation (Registered Charity No. 1096716).

What are the aims of this research?

We want to look at how families share their stories and tell their accounts of parenting beneficiaries of Muscle Dreams.

We are looking to interview the parents of beneficiaries, asking questions about their entire life (so across the 'lifespan').

We want to provide the opportunity for parents to retell as much of their story as possible, including their narratives that go beyond the passing of their loved one.

In retelling their accounts of parenting a child with muscular dystrophy throughout the lifespan:

- We start to understand how parenting a child with muscular dystrophy is storied through detailed and unique accounts

- We can provide a platform for sharing one's story as a resource for other parents on their own journeys

- We can identify narratives around the role of *Muscle Dreams* and understand how parents use this chapter of their lives within the retelling of parenthood

Why is there a need for this research?

Lots of research on how young people with muscular dystrophy talk about their experiences, understand their health and develop their identities over time

Some research addresses the experience of caring for someone with muscular dystrophy

Very little research looks at how parents tell their accounts throughout the lifespan (for example, from getting a diagnosis to their loved ones passing)

No research looks at the retelling and storying of events like Muscle Dreams.

MUSCLE DREAMS

The experienced research team work in accordance to their professional code of conduct, which includes ethical practice.

If participants have a question, comment or complaint, they can contact one of the research team directly or contact the ethics committee on XX, who are independent from the team.

Meet the research team…

STRONGER TOGETHER

James

David

James is the lead researcher on this project and will interview you, should you choose to participate. Email James on: j.randall-james@herts.ac.uk

David and Lizette are the primary & secondary supervisors, respectively. Email David on d.m.wellsted@herts.ac.uk & Lizette on l.nolte@herts.ac.uk

Who can participate in this research?

The research project is looking to recruit 4-6 parents to interview in order to explore their stories in some depth. We will interview people twice, lasting approximately 1.5hr per interview. This is to allow individuals the time and space to tell their stories and for these to be fully heard.

We use the term parents inclusive of all the many different shapes and forms parental units can take.

What will happen if I decide to take part in this research?

James will interview participants using a number of questions agreed upon by the research team. However, this approach does allow some freedom to ask questions unique to the interviewee and their experiences.

▮▮▮▮▮ will liaise with you in order to arrange a time and place that is best suited to you. This means that James can visit you in your home or arrange for a convenient alternative. The interviews will be videoed or audio-recorded, depending on your informed decision.

We discuss this further on the next page under 'What will you do with my data?'

What will you do with my data?

All interviews will need to be recorded in some manner, as these become transcribed for analysis later on in the research.

Interview transcripts will not be published in their entirety, but extracts selected during the analysis process will be published alongside other accounts in an academic journal.

If participants are happy for their interviews to be videoed, then we anticipate these recordings could be used as a resource for parents in similar circumstances through publication on the internet. These videos could also be used for educational purposes in health settings and academia.

Are there any costs to taking part?

It is possible that individuals could become distressed in sharing their accounts, particularly in the context of loss and bereavement. James will be sensitive to this in his interview technique and delivery, but has also designed a debrief pack with the relevant support networks provided for participants. It is also important to know that interviewees will be free to withdraw from their interview at any point, should they need to.

CONSENT FORM

I, the undersigned *[please give your name here, in BLOCK CAPITALS]* ………………………..

………………………………………….hereby freely agree to take part in the study entitled:

BEING PART OF THE MUSCLE DREAM: PARENTAL ACCOUNTS

1 I confirm that I have been given a Participant Information Sheet (a copy of which is attached to this form) giving particulars of the study, including its aim(s), methods and design, the names and contact details of key people and, as appropriate, the risks and potential benefits, and any plans for follow-up studies that might involve further approaches to participants. I have been given details of my involvement in the study. I have been told that in the event of any significant change to the aim(s) or design of the study I will be informed, and asked to renew my consent to participate in it.

2 I have been assured that I may withdraw from the study at any time without disadvantage or having to give a reason.

3 In giving my consent to participate in this study, I understand that voice, video or photo-recording will take place.

4 I hereby give my consent for my transcript, video and audio-recordings to be used for the following purposes:

4.1 -An online resource for other parents researching muscular dystrophy and/or muscle dreams

Signature: _____

4.2 -For educational purposes in clinical training and academia (e.g. research methods training)

Signature: _____

5 I have been given information about the risks of my suffering harm or adverse effects. I have been told about the aftercare and support that will be offered to me in the event of this happening.

6 I have been told how information relating to me (data obtained in the course of the study, and data provided by me about myself) will be handled: how it will be kept secure, who will have access to it, and how it will or may be used.

7 I understand that my participation in this study may reveal findings that could indicate that I might require medical advice. In that event, I will be informed and advised to consult my GP. If, during the study, evidence comes to light that I may have a

pre-existing medical condition that may put others at risk, I understand that the University will refer me to the appropriate authorities and that I will not be allowed to take any further part in the study.

Signature of participant …………………………………… Date…………………………..

Signature of investigator …………………………   Date…………………………

Appendix F: Confidentiality agreement with transcription company

Doctorate in Clinical Psychology

University of Hertfordshire

Transcription confidentiality/ non-disclosure agreement

This non-disclosure agreement is in reference to the following parties:

James Randall-James (discloser)

And

Transcribe It / Nicki Brown

The recipient agrees to not divulge any information to a third party with regards to the transcription of audio recordings, as recorded by the discloser. The information shared will therefore remain confidential. If the recipient is able to identify and knows any participant in the recording, the recipient agrees to cease transcription, inform the discloser and destroy any copies of the recording.

The recipient also agrees to destroy the transcripts as soon as they have been provided to the discloser.

The recipient agrees to return and or destroy any copies of the recordings they were able to access provided by the discloser.

Signed:

Name: Nicki Brown

Date: 4/10/16

Appendix G: The use of videography (beyond this thesis)

Consent for the use of the interview accounts was sought for multiple purposes. Namely, participants' videos could be used as public resources for other parents in similar situations, and for training and teaching purposes. Although, this approach removes anonymity and so warrants a more comprehensive consent checking procedure, I believe the approach remains ethical. Narrative research not only has the potential to provide a platform for accounts often unheard or witnessed, but also has the potential to influence the collective sharing and shaping of narratives in a public domain. This sharing of accounts creates the opportunity for new and informed choices to develop for others. The "process of narrative sharing thus becomes a means of empowerment and also potentially leads to forms of social action and change" (Murray & Sargeant, 2011, p. 165).

As described prior, there were three points in which consent was checked throughout the recruitment process, to try and gain fully-informed consent. In relation to the use of video accounts, further safeguards seemed necessary. At the time of writing, these accounts have not been edited, but from the point of submission, follow-up telephone calls will take place to double-check the participant's consent for the video recording to be used outside of the academic publication route. If they do not consent, then the video footage will be destroyed. Any video footage produced will be shared with participants prior to publication to gain final consent prior to publication. All possible uses for these videos, who 'owns' these videos and issues relating to timescales of usage are listed in the below participant information sheet.

The use of my video footage: Additional information sheet

This project has the potential to the potential to create a valuable resource for those living or working with Muscle Dreams more widely in either text of film format, with the aim to.:

- Firstly, illustrate the widespread impact of living with Muscular Dystrophy and having a Muscle Dream experience, not only for the beneficiaries, but those around them (i.e. their families, guardians and carers).

- Secondly, offer a resource for parents researching their child's condition and wishing to consider living with and managing the difficult prognosis of deteriorating health.

- Thirdly, illustrate the charity's impact to individuals looking to invest, support or use the services.

- And finally, for teaching and training of future clinicians and researchers.

[For an example of what this could look like, please take a look at the website http://www.healthtalk.org – where you can find videos of individuals talking about their experiences of disability and wellbeing. It is hoped that we may be able to produce some videos in a similar light.]

It is important that you know how these videos will be used, how they will be edited and who will use these and in what way. It is important to us that you feel able to say yes or no to these uses in an informed way, so if anything is unclear or you do have some questions- please do not hesitate to get in contact.

Please consider the different uses for your video, listed below. Following this, we also list our promises to you. Please read this in full before agreeing to these uses.

1. ***Research publication only.*** We will still need to audio-record your interview and this will then be transcribed. All personal data will be anonymised at the point of transcribing. We will delete your audio data on completion of the study to insure confidentiality. The transcribed interviews will be used in our write-up of the findings of the research and presented in published journal articles. We will need individuals to consent to this use in order to participate in the research, as our methods of interviewing and analysis require this.

2. ***Use of my audio and/or video recording as a public resource.*** Your interview will be video-taped, or we will store and use the audio from your interview. These audio and/or video recordings will be edited and you will be asked if you are happy with a draft version. Audio and video recordings may be cut and/or edited to highlight certain points,, rather than covering *your entire* interview. These recordings may be posted in the Muscle Help Foundation website or their social media outlets and/or may be used in promotion of or fundraising for the Foundation. By consenting to this option, you are also consenting for your interview data to be used for the research publication route described above (1).

3. ***Use of my audio and/or video recording as a public resource <u>and a research resource</u>.*** Your interview will be video-taped, or we will store and use the audio from your interview. These video recordings will be stored electronically on an

encrypted USB stick (password protected) by James Randall-James. These videos may be used in part or their entirety for teaching or research purposes, and secondary analysis for future studies (for example, an analysis of the interviewer style and approach).

Our promises to you

- We will ask you whether you are happy for us to use your interview data in each way described and you will be free to say no to any of the options described.

- There will not be any costs for you not agreeing to any or some of these options.

- If you agree to your video being used for any of the named purposes, then the research team will contact you a month following your interview to ask for confirmation of your consent. This is to double-check that you have had the time and space to consider this properly.

- You will be provided with a copy of the video before it is published or finalised. You will be asked whether there are any segments that you would wish to be edited and the research team will liaise with you to ensure the video content is agreeable to you.

- If you do not wish for the video to be published at this stage, we will work with you to try and adapt the video appropriately. However, the video will be your story and we will respect your right to choose against publishing it, even at this later stage.

- The rights to all videos will be owned by the lead researcher, James Randall-James.

- For those videos relevant to, and edited for, the impact of the Muscle Dream experience and the story of parenting young people with muscular dystrophy: Once published,   will be permitted to use and store these videos on his YouTube channel, Muscle Help Foundation website and the Muscle Dreams website.

- Videos will be kept indefinitely unless participants request otherwise.

- If people agree to their videos being used for teaching and research purposes, then these videos will be kept indefinitely in hard-copy form (DVD) by James Randall-James. These hard-copies can be destroyed if individuals change their minds at a later date if individuals contact James via the research team.

Appendix H: Ethics protocol for distressed participants

Protocol ref: cLMS/PGR/UH/02353

Conditional acceptance requirement: Please provide a brief protocol for how potential distress during the interview(s) will be dealt with to hhsecda@herts.ac.uk for approval by the Chairman, prior to recruitment and data collection.

Protocol for distressed participants:

- The researcher is a trainee clinical psychologist with over 7 years of experience of working with vulnerable people and distress. He will draw on these experiences when conducting interviews within this research.

- The researcher will initially allow some space for the participant/s.

- He will be empathetic, offering them time to process their experiences or the discussion.

  o E.g. "this cannot be an easy thing to talk about. Take as long as you need."

- The researcher will insure that there are tissues on site, in case these are needed.

- Prior to the interview he will familiarise himself with the interview setting, offering the participant a break from the interview if they wish – possibly in a separate room.

- Gauging the level of distress, the researcher will also remind the participant that they can discontinue the interview if they wish (as empathically as possible).

  o E.g. "if you feel that it is going to be difficult for you to continue today, then that is okay. We can finish now if you feel you need to?"

- If it seems appropriate (and is during the first interview), then participants can also be reassured that the interview can be stopped at this point, and we consider continued at the second appointment.

- o The researcher will offer to phone the participant prior to the second appointment to check whether they still wish to participate.

- Again if it seems appropriate, the interviewer can remind participants that their involvement is voluntary and they can withdraw from the study if they wish.

- The researcher will provide the participant with a participant debrief sheet at the end of this interview, and discuss some of these services available if they feel that they need to/wish to know more about what they can offer.

- If the researcher becomes concerned about the participant, he will encourage them to contact their GP to seek further support. He will also contact his supervisory team as soon as possible if support is needed.

Appendix I: Debrief sheets

BEING PART OF THE MUSCLE DREAM

What if I need support?

A DEBRIEF PACK FOR PARTICIPANTS
Retelling accounts of bereavement can be distressing for anyone. We hope that some of the resources within this pack will be helpful should you find yourself needing some extra support.

Introduction to this pack

This pack has been developed to support those wishing to continue conversations about their loved ones, and those who have felt that they could do with some additional emotional support.

Firstly, consider visiting your GP for advice if you are feeling low or experiencing difficulties relating to your loss.

This debrief pack should not be considered equivalent to consultation with a professional – *please do* seek support if you are worried.

Cruse Bereavement Care
Somewhere to turn when someone dies

CRUSE Hertfordshire have their own dedicated helpline to talk, listen & reflect on people's experiences.

Tel: 01707 278 389

Bereavement Friendship groups are held monthly in Chestnut, Baldock & Hatfield.

For further information, visit: www.cruse-hertfordshire.org.uk/

Duchenne Family Support Group

The Duchenne Family Support Group is a partner organization to the Muscle Help Foundation, who provide social and emotional support for parents and carers of young people with muscular dystrophy.

For further information & support call them on
0800 121 4518

Or visit www.dfsg.org.uk

MUSCLE DREAMS

If you want more information on how to support a loved one through bereavement and loss, from where to start and how to talk, perhaps download this informative leaflet: http://goo.gl/9E8PqJ

Or call 'Dying Matters' on
08000 21 44 66

Meet the research team…

STRONGER TOGETHER

James

David

Lizette

The research team have compiled this debrief pack to go alongside an information leaflet relating to our research project.

Our professional code of conduct and ethics approval for this study means that we cannot personally support individuals with support beyond the remit of the study. Therefore, we have spent some time compiling these resources together to support you as best as we can.

together for short lives

You may already be aware of the national charity called Together for Short Lives, but if not, you may find their resources that map out 'the family journey' (inc. bereavement support) particularly helpful. Additionally, they host a community forum across multiple conditions, where you can talk to others in similar circumstances.

Visit: www.togetherforshortlives.org.uk
And look under the tab 'Help for Families'

If you feel that you would benefit from engaging in a talking therapy (such as cognitive-behavioural therapy), then you may wish to consider self-referring to the Wellbeing Services in Hertfordshire. Staff will work with you to assess your needs and identify what would approaches may be helpful for you at this time.

To self-refer call on:
0300 777 0707

For self-help leaflets visit:
www.hpft.nhs.uk/help-for-adults

stand by me bereavement support

Stand-by-Me Bereavement Support are a North-Hertfordshire based charity, who support children and families through bereavement and loss.

They may be particularly helpful if you have other children struggling with the loss of their sibling as they host a helpline on Tuesdays from 6-8.30pm on:

01438 730580

Stand-by-Me also provide a selection of information leaflets on bereavement support in general (from Dying Matters):

www.stand-by-me.org.uk/leaflets

We sincerely wish you all the best and hope that you have found this information leaflet helpful.

If you have a question, comment or complaint, you can contact one of the research team directly or contact the ethics committee on XX, who are independent from the team. Thank you.

Appendix J: Press release text

James hopes to interview parents of *Muscle Dream* beneficiaries in some detail to provide an opportunity for people to share their accounts of caring, living and dreaming in the context of muscular dystrophy. In some cases, James will also be interested in how families experience the loss of their loved one, and how that experience changes after death.

This detailed look at the transformational nature of *Muscle Dreams* within a conversation across the lifespan of the young person (from birth to beyond their death) will unearth how the parents of beneficiaries describe and retell their journeys together. In doing so, it is hoped that the impact of *Muscle Dreams* within the family will become clearer and detailed stories from those who have lived alongside these young people living their dream will be given a platform in which they can be fully understood.

Once all the data has been collected and analysed, James will look to publish the results in a scientific journal. However, the charity ███████████████ and James Randall-James are also keen on looking at other ways in which the findings can be used to further support the work of The Muscle Help Foundation and those who engage in *Muscle Dreams.* This will include making user-friendly summaries of the results and with the consent of the person/s involved, possibly using some of the accounts as a health resource for others going through similar experiences.

Appendix K: Interview schedule

"I am interested in hearing about your lives as parents and how you would describe your journey to me. I will be interested to listen to the stories about what has happened to you and so will want to give you the time and space to tell me about this as fully as you want to. I will try and sit back and listen as much as I can, without interrupting. Is that okay?"

OPENING QUESTION:

"Tell me about a time in your life when your role as parents really changed?"

Finding symptoms and the pre-diagnostic stories

"Tell me about the time before ___ got diagnosed with Muscular dystrophy."

The diagnosis stories

"What changed when ___ was diagnosed with Muscular dystrophy?"

"Could you tell me about that very first day, of waking up, knowing that your child now had Muscular Dystrophy?"

The living stories

"How did you change personally, as a family, and as parents, because of the condition?"

The Muscle Dream story

"Starting from the beginning, how did Muscle Dreams come into your lives?"

"What happened when the Muscle Dream ended?"

"Did the conversations you had after the Muscle Dream change at all?"

The story of life beyond dying and death

"If you feel able to, please do tell me about the time you found out that ____ was near the end of their life."

"What conversations did you have? Perhaps they were conversations to yourself, or to each other, or as a family?"

"Tell me about life since ___'s death."

Appendix M: Reflective diary / quote extract

In anticipation of my role as a reflexive-researcher, I also sought quotes that would challenge my thinking in terms of my role and support me to become more attuned to the influence of self in research. Below, in Figure 2.7.1, I include an example annotated quote demonstrating the reflections they inspired through brief examples. What these initial reflections show is that with a particular lens, my behaviour could influence a situation quite differently than if I used some other discourse available to me. Beyond the scope of this research, is a more relational-reflexive approach that can member-check interviewees and explore the influence of the researcher, not too dissimilar to Abbott's reflections in the paper *Other Voices, Other Rooms* (2012).

Figure 2.7.1. Annotated reflections inspired by my reading.

Duty to critique narrative? Consider alt. contexts?

What are people's first impressions of me?

Rather than just note influence, wouldn't ethics propose I purposely use myself – say to contain distress?

"We must go beyond language both in our thinking and in our techniques. There is scope for a development of a whole repertoire of using ourselves, what we wear and how, how we sit and stand, how we interact with our clients, what imagery we invoke, whether or not we use objects and things, and whether or not we touch"

(Malik & Krause, 2004, p. 106)

Should I 'join' families? Do I accept coffee? What is the impact of saying no?

In the context of death, would the use of touch become less taboo in research? Is it unethical and distressing to decline a hug or to avoid a hand on the shoulder?

Who is 'my client'? The late young person? The charity? Science? The University? How does this change how I present myself?

How do objects carry stories on behalf of us? How could I capture this in the analysis, if so?