

Psychosocial adjustment, experiences and  
views of fathers of sons with  
Duchenne Muscular Dystrophy

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# **Abstract**

## **Background**

Although Duchenne Muscular Dystrophy (DMD) is acknowledged to have an impact on families as a whole, few studies have investigated psychosocial aspects. Investigation of fathers in paediatric psychology literature is also neglected, and available DMD studies focus on maternal adjustment. This study addresses calls for both, research within the area of DMD and inclusion of fathers.

## **Aims**

The overall aim was to investigate psychosocial adjustment, and experiences, of fathers of sons with DMD by studying associations between paternal adjustment and:

- boys' functioning (physical and psychological)
- perceived paternal involvement in condition management
- perception of support
- fathers' experiences of parenting a son with DMD

## **Methods**

A mixed methods approach, incorporating questionnaires evaluating level of boys' functional ability (Functional Disability Inventory) and psychiatric adjustment (Strengths and Difficulties Questionnaire); paternal involvement in condition management (Dads Active Disease Support Scale); paternal ratings of satisfaction, and paternal adjustment (General Health Questionnaire), was used. In-depth

interviews were also undertaken, and written accounts of experiences and views recorded.

## **Results**

50 fathers completed questionnaires and 48 provided written accounts, with a cohort of 15 participating in interviews. Paternal adjustment was comparable to that of mothers, as noted in previous studies, with 38% above cut off for risk of psychological problems. Predictors of paternal adjustment were boys' psychosocial adjustment, perceived amount of involvement in condition management and perceived support from friends. Themes emerging from the qualitative strand were 1) loss and acceptance; 2) support versus isolation; 3) the fight for resources and 4) race against time.

## **Conclusion**

Findings emphasise the need for bio-psychosocial interventions, acknowledging fathers' needs, role, and involvement in their child's condition. Alongside consideration of the family, the psychosocial impact for fathers should be acknowledged as being equally important to dealing with physical issues surrounding DMD. Professional awareness is needed of the emotional implications, and issues fathers face.

**Keywords:** Father; Duchenne Muscular Dystrophy; Psychological adjustment

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# Chapter 1

## Introduction

This thesis addresses psychosocial adjustment, experiences and views of fathers of a son with Duchenne Muscular Dystrophy (DMD). All forms of muscular dystrophy comprise muscle weakness as a result of genetic faults. Duchenne muscular dystrophy is the most severe of the twenty types of MD, and affects only males. It is characterised by behavioural difficulties and terminal prognosis as a result of progressive muscle weakness. For these reasons, the focus of the thesis is DMD and not MD in general.

Chapter one presents an overview of recent legislative developments relating to muscular dystrophy services in the U.K., providing the broader context in which the thesis lies. A rationale is then presented for undertaking a study of fathers, within the field of Duchenne Muscular Dystrophy. Finally, the developmental significance of paternal involvement with the child is considered.

### **1.1. Rationale for choice of research topic**

Following ongoing campaigns by families and professionals, muscular dystrophy services are currently the focus of new developments at U.K. Government level. The All Party Group on Muscular Dystrophy (APGMD) was introduced in 2008, aiming to raise the profile of the condition. As a result of investigations by the group, comprising evidence from NHS commissioners and professionals, families and researchers, the Walton Report (APGMD, 2009) was presented to the National

Institute of Clinical Excellence (NICE) in August 2009. The report included requests for NICE clinical guidance for muscular dystrophy (MD), and Department of Health (DoH) recognition of the need for specialised muscle services.

Throughout the report, the U.K. Government was criticised for lack of attention to the needs of those with MD and their families. Of note was the lack of any records of official MD data across the U.K. Further, the burden placed on charities (such as the Muscular Dystrophy Campaign) was emphasised, due to lack of U.K. Government funding. An additional recent development of relevance is the Department of Health's report 'Healthy lives, brighter futures: the strategy for children and young people's health' (DoH, 2009). This report outlines plans for specialist intervention across life stages, including pledging to support children with ongoing complex health needs and their families, through provision of individual care plans by 2010.

Specific to Scotland, the Scottish Government launched the 'disabled childrens' liaison project' across 2008/2009, seeking parents' views about experiences and barriers to support (Scottish Government, 2008). Key objectives were outlined for disabled children and their families, including practical areas such as increased access to family breaks. In terms of emotional support, also included was improved provision for families during transition where young people progress through difficult phases.

These developments are promising; however, it is evident from parent-led efforts (e.g. Parent Project U.K.), that families perceive some way to go in terms of the condition being accounted for within U.K. Government health-care policy. A call for inclusion of fathers within policy is reflected in other areas. For example, representatives from the foundation for people with learning disabilities requested amendments within the Government's 'carer's strategy' (DoH, 2008) to include the role of fathers. This acknowledges the cultural context in which policy is framed, with a historical focus on mothers.

The previous examples illustrate that focus on muscular dystrophy, and those affected, is lacking to date, with attention to the condition only a recent development. The lack of attention paid to MD per se, has been accompanied by a lack of research on psychosocial adjustment (Puxley and Buchanan, 2009) of individuals within families who may need support. Work contributing towards this, is a step towards raising the profile of MD and associated individual support needs. Specifically, in the area of MD, fathers have been overlooked in both policy and research.

## **1.2. Why study fathers (in the context of DMD)?**

The literature demonstrates a need to investigate factors that may aid the design of interventions, to address needs of specific family members (e.g. Holmbeck, 2002; Robinson, Gerhardt, Vanatta and Noll, 2007). Regardless of the family-focussed approach frequently cited throughout health literature (e.g. Sloper, 2000), this has not led to inclusion of outcomes/perspectives of fathers in terms of their child's

chronic condition. Specifically within the MD literature, whilst there are studies (although few) investigating parental experiences, an emphasis on fathers' experiences alone is lacking.

Phares, Lopez, Fields, Kamboukos and Duhig (2005) examined the proportion of fathers in clinical child and family research. They reviewed major psychology journals covering an eight-year period, finding that of 514 included studies, only 2.1% involved fathers only. It was concluded that there *"continues to be a dearth of research on fathers"* (Phares et al, 2005, p.631). This echoes the observations of other investigators (e.g. Seiffe-Krenke, 2002; Wysocki and Gavin, 2004; Bonner et al, 2007), specifically that fathers' experiences are under-reported in the literature, with sole emphasis on mothers as carers.

On the basis that fathers are under-represented in research and graduates are the researchers of tomorrow, Silverstein and Phares' (1996) review found that 10% of dissertations explored fathers alone, with male graduate students more likely to include fathers in their design. In relation to doctoral research topics, the authors highlighted that fathers were being neglected in doctoral dissertation research. Opinion amongst researchers in the field of paediatric psychology, also identifies a need for work in this area:

*"Paediatric psychology research lags even further behind child clinical research in including fathers in research designs and analysing for maternal and paternal effect separately. There is also a concomitant lack of inclusion of fathers in family-based interventions in paediatric psychology"* (Phares et al, 2005, p.631).

A review of the paediatric literature has highlighted the fact that articles stating 'parents', without elaborating, are focusing on mothers only. In fact, many studies looking at 'caregivers' or 'family' perspectives usually have a majority female sample (e.g. Raina, O'Donnell, Rosenbaum, Brehaut, Walter et al's (2005) study of *caregivers* of children with cerebral palsy, comprised a 95% female sample).

A literature search for this thesis highlighted few studies focused on parents of a child with Duchenne Muscular Dystrophy (DMD), and of those including 'parents' the highest sample including fathers (that could be located) was 33% (n14), (Chen, Chen, Jong, Yang and Chang, 2002) in a quantitative study. Of studies identified, most were quantitative, or with caregiver samples who were predominately female.

Authors have suggested that studies based on coping of parents of disabled children, add valuable information to the research base (e.g. Webb, 2005). This is highly relevant in the context of DMD, where stress is heightened due to the level of dependence of the child, associated learning difficulties in many cases, and continual deterioration (Webb, 2005; Nereo, Fee and Hinton, 2003). Hovey's review of the literature suggested fathers of chronically ill children have parenting needs that differ from fathers of healthy children (Hovey, 2006). Fathers may also cope differently than mothers with the child's condition (Hovey, 2005 and 2006), presenting unique support needs.

By understanding fathers' experiences, health providers can promote relevant support strategies, and anticipate fathers' emotional reactions to caring for a child

with a terminal condition such as DMD. To date, it appears that psychosocial interventions have not been designed specifically for DMD families, or, they are not widely disseminated. Although rare, the few studies that have been done demonstrate the positive impact of interventions aimed at fathers. One such study (Dellve, Samuelsson, Tallborn, Fasth and Lillemore, 2006) used a prospective intervention with mothers and fathers of children with an uncommon condition. Fathers' high stress levels were strongly associated with overall life satisfaction. There was also an impact of the type of disability, with parents of children with progressive disabilities (as in DMD) having high levels of stress due to their own health issues and social isolation. Fathers showed increased active coping and compliance to professionals' advice post intervention (Dellve et al, 2006). Based on positive response to intervention, the author suggested that intensive programmes to develop parental competence may benefit specific groups - including fathers (Dellve et al, 2006).

In summary, a range of literature focuses on child and family perspectives within the context of chronic illness, yet the lack of research concerning fathers' adjustment, and experiences is striking. Dellve et al's (2006) work demonstrated the potential for fathers to benefit from interventions. This area, therefore, merits exploration in order to promote both awareness and appropriate support efforts.

### **1.3. Developmental significance of paternal involvement**

Considering the concept of involvement is important, as studies suggest child coping behaviour is promoted when family members are proactive in caring roles (e.g.

Lamb, 2004; Thompson, Zeman, Fanurick and Sirotkin-Roses, 1992). Research suggests that increased paternal involvement has a beneficial impact on families (Pleck and Masciadrelli, 2004), and on child adjustment and development (Lamb, 2004). Wysocki and Gavin suggest paternal involvement may act as a *"coping resource that influences both mothers' and children's appraisals of their adaptive capacity"* (Wysocki and Gavin, 2006, p.502). Similarly, in a review of the fathers' role in the aetiology of child anxiety, Bogels and Phares (2008) conclude *"there is strong evidence from cross-sectional as well as longitudinal research, to suggest that paternal involvement, more than maternal involvement, promotes competence and protects against psychological distress in adolescents and young adults"* (Bogels and Phares, 2008, p. 543).

In this context, involvement has been conceptualised as *"the degree to which family members provide one another with emotional and instrumental support"* (Gavin and Wysocki, 2006, p.481). Fathers' involvement may include: care-giving, emotional and practical support to mothers, playing and encouraging activities, and provision of guidance and discipline. Gavin and Wysocki (2006) frame paternal involvement within the Wallender, Varni, Babani, DeHann, Wilcox and Banis (1989) 'Risk and Resilience model'. This explains how negative effects of risk factors (e.g. disability) on adaptation (psychosocial) may be mediated by resistance factors (e.g. socio-ecological, such as social support/interpersonal).

Lamb, Pleck, Charnov and Levine's (1987) tri-partite model of fathers' involvement acknowledges the different forms of interaction that may occur. The three areas



include: interaction (one to one with his child); accessibility (physical presence and being emotionally responsive) and responsibility (care and wellbeing of the child) (Lamb et al, 1987). In typically developing children, paternal involvement has demonstrated a positive influence on areas such as child developmental and cognitive/behavioural outcomes (e.g. Garfield and Isacco, 2006; Flouri, 2005). Large-scale longitudinal studies have also identified father involvement as a protective factor, in risk situations (Flouri and Buchanan, 2004)<sup>2</sup> having some benefit in reducing boys' externalising behaviour problems compared to girls' (Carlson, 2006).<sup>3</sup>

Given the benefits of paternal involvement, researchers have highlighted this as an appropriate focus for research and intervention within the context of paediatric chronic illness (Gavin and Wysocki, 2004). Research has also found greater paternal involvement is associated with better quality of life amongst chronically ill adolescents (Wysocki and Gavin, 2006). The authors suggest that both quality and quantity of involvement have a direct impact on areas such as treatment adherence and frequency of reinforcement for condition self-management (Wysocki and Gavin, 2006).

A measure (Dads Active Disease Support Scale: 'D.A.D.S.') was developed by Wysocki and Gavin, 2004, attempting to provide direction in quantifying the amount and helpfulness of fathers' involvement in paediatric disease management. Measures of family, mother and child functioning were completed by 224 couples. Parental

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<sup>2</sup> National Child Development Study (n=7,259).

<sup>3</sup> The 1979 National Longitudinal Survey of Youth (n=2,733).

scores highlighted low levels of paternal involvement- fathers carried out disease related tasks less than half the time, with mothers rating fathers' helpfulness higher than fathers did themselves (Wysocki and Gavin, 2004).

In their later study, Gavin and Wysocki, (2006), investigated associations of paternal involvement in paediatric disease management with maternal and family outcomes (190 couples). Maternal rating for perceptions of greater paternal involvement ratings on D.A.D.S. was associated with fewer maternal psychological problems, and less disease impact on family functioning. Reports from both parents suggested higher levels of paternal involvement were associated with more positive outcomes for marital satisfaction and family adjustment (Gavin and Wysocki, 2006). In light of Wysocki and Gavin's (2004) findings that mothers rated fathers' helpfulness higher than fathers did themselves, it is possible that fathers perceived their 'help' as surplus or not as useful from the mothers' perspective.

Supporting this, studies have demonstrated that men are less confident as care givers and more sensitive to perceived criticism (Gaugler, Given, Linder, Kataria, Tucker and Regine, 2008). Of relevance is the observation of Paediatric Psychologist, Elizabeth Seagull (2000), from her clinical work with families of chronically ill children. She described a vicious circle with mothers becoming comfortable with their child's medical needs and becoming 'expert' in this regard. Fathers, however, felt distanced from treatments and incompetent in relation to care. She emphasised the need to include fathers in a systems approach to intervention with families affected by chronic illness (Seagull, 2000).

Research has also found that fathers of disabled children describe their role as a support for partners who undertake carer responsibilities (Gray, 2003). Dellve et al's (2006) intervention study for parents of children with rare diseases illustrated fathers reporting more stress than mothers, pre-intervention, in relation to perceived incompetence in caring for the child. Post-intervention, fathers' perceived knowledge increased and those with a high level of stress due to their perceived incompetence decreased. This study emphasised that increased involvement of the father may assist with family functioning, acting as a buffer for mother's stress (Dellve et al, 2006).

Findings such as these indicate more paternal involvement in care is associated with higher levels of family, marital and maternal adjustment, and higher quality of life amongst ill adolescents (Wysocki and Gavin, 2006). Various DMD related factors may influence involvement, such as the progressively deteriorating nature of the condition, and adjustment problems in boys. Identified factors that hinder paternal involvement and promote negative attitudes are learning difficulties and behavioural problems (as with DMD) (Bristol, Gallagher and Schloper, 1988).

The fathers' view of his parental role is also suggested to influence involvement (McBride, Brown, Bost, Shin, Vaughn and Korth 2005). Parent role identity (Parke, 2000) provides a basis for defining father involvement, with Parke arguing that in the absence of traditional gender 'norms' for father involvement, role-identity is especially relevant as a precursor to father involvement. Input into the role of

'fathering' may be altered as a result of a major event, such as discovering your son has a chronic condition (Major, 2003).

Also relevant is Gagliardi's (1991) ethnographic study of 3 DMD families, in which she observed that fathers did not have much time to spend with sons. Citing a desire to provide for sons, all worked overtime, resulting in reduced involvement with sons. Gagliardi suggested fathers found it easier to cope by avoiding seeing the child suffer, thus managing their own emotional distress (Gagliardi, 1991).

### ***1.3.1. Father-son relationship (links to involvement)***

An additional factor to consider is the fact that DMD affects only sons. It is suggested that fathers feel a loss of the traditional father-son relationship and have more problems in adjusting expectations for sons compared to daughters (Lee, Miles and Holdich-Davis, 2006; Waite-Jones and Madill, 2008). Frey, Greenberg and Fewell, (1989), also described the impact of having a son with a disability heightening psychological distress in fathers.

According to Social Learning Theory (Bandura 1986), fathers might have greater involvement, with stronger influence on sons due to acting as role models. Supporting this, Trute (1995), used separate interviews to investigate gender differences in psychological adjustment of parents of developmentally disabled children, finding that fathers of boys appeared to be at higher risk of depression (Trute, 1995). The possibility that father involvement has greater influence on boys' behaviour, compared to girls' has been suggested (Carlson, 2006).

Allowing insight into the importance of father-son relationships, Barnett, Marshall and Pleck (1992), found that sons reporting positive paternal relationships had low levels of psychological distress. In this study, measures of both maternal and paternal relationship to the child were entered into a regression equation. Only the father-child relationship was significantly related to the male child's distress (Barnett et al, 1992). Gender effects have also been found in research on paediatric cancer, where boys were identified as more vulnerable to distress compared to girls, when their father was distressed (Robinson et al, 2007).

### ***1.3.2. Summary of involvement***

Paternal involvement is associated with positive outcomes for families, and is seen to have specific benefits for chronically ill children. Characteristics of the child's disability are seen to influence levels of paternal involvement, with a number of studies linking progressive chronic conditions to less involvement. The fact that DMD affects males only, is also a factor to consider, as evidence suggests the father-son relationship in particular is important in relation to adjustment.

### **1.4. Overall summary**

Work raising the profile of DMD, and understanding the impact on fathers is an important step towards promoting research in both father and DMD specific areas. In recent years, researchers (e.g. Bonner, Hardy, Willard and Hutchinson 2007) have highlighted a need for inclusion of fathers in paediatric psychosocial research. Further, researchers have lately questioned the lack of psychosocial investigation into DMD, given the practical and psychological consequences on families (Puxley

and Buchanan, 2009). Overall, the DMD psychosocial literature is lacking with most studies conducted in the 1990s and stemming from the U.S.A. and Canada, but available studies highlight the detrimental impact of the condition on carers. Little is understood about parental experiences and this is lacking in relation to fathers.

A potential obstacle to inclusion of fathers is explained by methodological challenges in father studies, including recruitment/retention and identification issues (Mitchell, See, Tarkow, Cabrera, McFadden and Shannon, 2007). This was identified in a previous DMD interview study, where Firth and Barry (1986) reported that despite inviting fathers to participate, the study mainly relied on mothers. DMD has rarely been investigated in the psychosocial literature, therefore combined with the paucity of work focused on fathers, this thesis aims to address this imbalance.

In addition to the theoretical basis motivating this research, the focus and aims of were also influenced by discussion with clinical and research staff<sup>4</sup> who have (anecdotally) noted issues faced by some fathers. Within the context of recent steps (e.g. introduction of the All Party Committee) towards improving muscular dystrophy services, the choice of research topic stems from a desire to highlight the need for focus on family members affected by DMD.

This thesis addresses a gap in the literature, by allowing insight into the experiences and outcomes of fathers. It may also contribute by highlighting characteristics of those who could benefit from increased support, and clarifying barriers faced. As a

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<sup>4</sup> Contact is maintained with staff at the Dubowitz Neuromuscular Centre at Great Ormond Street Hospital, London (formerly based at Hammersmith Hospital). This is the largest muscle centre in the U.K. (one of 4), representing a high proportion of the U.K. DMD population.

result, specific areas with implications for intervention may be highlighted. The underlying goal is to draw attention to the area, highlighting potential for future relevant psychosocial research.

## Chapter 2

### Literature Review

This chapter comprises an introduction to DMD, followed by a description of DMD specific<sup>5</sup> and additional relevant research, underpinning the current study. An overview of DMD is presented, followed by an exploration of adjustment in DMD carers. This section also considers influences of child characteristics (behaviour and disability), support and parental gender. Following this, a conceptual model to guide the choice of variables considered within the thesis is outlined. Finally, the aims and research questions are stated.

#### **2.1. Duchenne muscular dystrophy as a deteriorating condition**

The muscular dystrophies are genetic conditions that are inherited or may arise without prior symptoms (MD Fact-sheet, Muscular Dystrophy Campaign). These conditions have been described as 'chronic diseases manifesting with progressive muscle weakness' (Grootenhuis, de Boone and Van der Kooi, 2007). DMD is named after the French nineteenth century medic, Dr. Duchenne de Boulogne, who first studied muscular dystrophy. The difference between DMD and other forms of MD lies in the associated behavioural difficulties, severity and terminal prognosis of DMD.

More than 30,000 people within the U.K. have muscular dystrophy or related conditions and 120,000 individuals are indirectly affected as relatives and carers (MD

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<sup>5</sup> All identified DMD psychosocial studies available are included.



Fact-sheet, Muscular Dystrophy Campaign). The condition is usually diagnosed between 2-5 years. There is no national register in Scotland for DMD, however, contact with Muscular Dystrophy Campaign Family Care Advisors, suggests there are approximately 200 boys with DMD in Scotland. Throughout the U.K. there are approximately 1,500 boys at any time with DMD.<sup>6</sup> DMD affects approximately 1 in 3,500 male births (or 100 boys in the U.K. each year). Females rarely show any symptoms, however, may be 'manifesting carriers' of the defective gene, passing the condition to their sons (Dubowitz, 1982). Males are affected via transmission by an altered gene on the x chromosome, in a sex linked (recessive) inheritance pattern, with approximately 50% likelihood of a carrier's son being born with MD (Dubowitz, 1982).

In approximately one third of cases, the condition is not hereditary but due to 'fluke' gene mutation (Murphy and Mutalik, 1989). The result is that affected boys have abnormal levels of the enzyme 'creatine kinase' in their blood, leading to detrimental effects on muscle tissue. The overall impact is a defect in dystrophin, the protein required for healthy growth of muscle fibres, resulting in severe disability, deterioration over time, and terminal prognosis. Progressive bodily weakness leads to respiratory and cardiac muscle failure in the child's early twenties (Kohler, Clarenbach, Boni, Brack, Russi and Bloch, 2005).

Physical problems first occur between 1-3 years of age, when children have difficulties in activities such as running and climbing. With time, boys fall frequently

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<sup>6</sup> Communication with Family Care Officer, Western General Hospital, Edinburgh. Contact was also made with Edinburgh University genetics department, but they were unable to provide an estimate of numbers in Scotland.

and have trouble walking. Around the ages of 8-11, the child cannot walk and there is a downward spiral in condition requiring wheelchair use (Grootenhuis et al, 2007). Condition management involves intensive routines, such as stretches and exercise, use of an apparatus for standing and at later stages, spinal brace and night ventilation.

In DMD, illness stages (diagnosis: signs of muscle weakness; transitional: difficulties walking; loss of walking; adult stage: heart and lung muscle deterioration) are marked by the introduction of interventions such as a spinal brace or callipers. This is very much specific to DMD, as not all conditions are associated with marked illness stages. Boys require ongoing physical interventions, with varying needs throughout the course of the condition (Parent Project U.K., 2006). Focus is placed on managing symptom progression and promoting life quality (Grootenhuis et al, 2007).

In addition to physical problems, DMD is associated with behavioural characteristics, with studies identifying high levels of behaviour problems, including limited social skills, attention deficits and depression (Leibowitz and Dubowitz, 1981; Thompson et al; 1992 Nereo et al, 2003). Some behaviours are thought to result from the condition itself (Donders and Taneja, 2009), whilst others may be reactive responses to the condition, such as frustration. Leibowitz and Dubowitz (1981), in a sample of 57 DMD boys aged between 3-13, confirmed the association of intellectual impairment - especially verbal, with DMD. Subsequent research has also identified a deficit in verbal and performance IQ (Polkaloff, Morton, Koch and Rios, 1988; Hinton, Nereo, Fee and Cyrulnik, 2006). Cognitive function is not thought to be

associated with physical decline (Nereo et al, 2003). Hinton et al (2006) describe DMD as affecting both muscle and brain, however, the neurological basis is unclear. Physiological studies have focused on the role of dystrophin on central nervous system function. Anderson, Rae and Morley (2002), indicate evidence for 'disorganised central nervous system architecture' and loss of neurons, however, a conclusive neurological basis for cognitive impairment remains elusive (Anderson et al, 2002).

Hinton et al (2006) point to the heterogeneity of performance amongst DMD sufferers, however, severe learning difficulties are identified in approximately 19% of boys (compared to 2-3% of general population). The Muscular Dystrophy Campaign have stated that in approximately one third of cases, boys have problems associated with learning, with parent-led organisations stating that boys often experience difficulties with learning that are undiagnosed (Parent Project U.K., 2006).

It has been suggested that emotional and behavioural difficulties interfere with the child's ability to focus in a learning situation (Polkaloff et al, 1989). In turn, this can have an impact on social functioning, making peer acceptance difficult (Charron-Prochownik, 2002). As with Donders and Taneja (2009), Nereo and Hinton (2003) believe observed social deficit is an associated characteristic of DMD (found regardless of age; I.Q.), not a reaction to the disease per se. In comparing a diagnosis of DMD to that of Down's syndrome, Green and Murton (1995) describe doctors delivering a 'death sentence on the child'. Although recent medical

developments are encouraging- including a U.K. study leading towards understanding the role of dystrophin in muscle regeneration (Griffin and Des Rosier, 2009)- related research remains at an exploratory stage (Manzur, Kinali and Muntoni, 2008). There remains no cure and boys have an average life span of 25 years.

In summary, DMD involves much more than muscle wasting. As such, DMD has been labelled a 'complex chronic terminal condition' involving intensive care, with the terminal phase being some time from diagnosis (Gravelle, 1997).

## **2.2. Adjustment and coping in carers of boys with DMD<sup>7</sup>**

The terms 'adjustment', 'adaptation' and 'coping' are used in various contexts throughout the literature. Eiser (1990) noted that researchers tend to 'opt out' of defining the concept, instead referring to a questionnaire score (Eiser, 1990). Adjustment has been defined as: *"terms that refer to emotional and social functioning"* (Wallander and Thompson, 1995, pp. 125-126), with coping resources conceptualised as: *"the capabilities and strengths to manage a stressor while maintaining established patterns of functioning"* (McCubbin and McCubbin 1993, p. 29).

### **2.2.1. General adjustment issues in DMD**

Within family systems theories (e.g. Kazak, 1989, Kazak, Simms and Crump, 2002) parental adjustment is important to consider due to influence on child outcomes

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<sup>7</sup> All available DMD papers were reviewed, however, findings from other relevant conditions are drawn upon as the psychosocial DMD literature is sparse.

(e.g. Robinson et al, 2007), and those of the family (Holmbeck, 2002). Studies concerning associations of family factors with psychological outcomes of disabled children, highlight effects of parental functioning on child adjustment (e.g. Lee et al, 2006). A bi-directional effect has been suggested in work with DMD mothers and sons<sup>8</sup> (Thompson et al, 1992; Nereo et al, 2003).

Generally, levels of stress experienced by parents (usually mothers) of boys with DMD are elevated (e.g. Reid and Renwick, 2001; Chen et al, 2002; Abi Daoud, Dooley and Gordon 2004). Adjusting to caring for a child with DMD draws upon family resources (Chen and Clark, 2007; Chen, 2008), with stress related to practical and emotional adjustment (Polkaloff et al, 1988). Using a questionnaire survey, Abi Daoud et al, (2004) investigated depression, self-esteem and mastery in 35 families (14 fathers) of boys with DMD. Results highlighted one third as being at risk of a major depressive episode, compared to 4% of a national control group. Parents of boys older than 13 years were more likely to experience distress that had a negative impact on functioning, (Abi Daoud et al, 2004).

Similarly, Thompson et al (1992), using parent report measures for 35 parents (3 fathers) of boys with DMD, found poor self-reported psychological adjustment in 57%. The mediational variables of parent appraisal of stress, use of palliative coping methods and level of family conflict, together explained 58% of variance in general distress, 50% in depressive symptoms and 31% of anxiety symptoms. In this study, 89% of boys were classed as having psychological problems (mainly internalising).

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<sup>8</sup> Due to word limitation, throughout the thesis the term 'DMD parents/child/boys', is taken to mean parent of a child with DMD etc.

Parental distress could predict 19% of the variance in these problems (Thompson et al, 1992). Given the prevalence of behavioural problems, recent research, (Chen, 2008), has identified difficulties for parents (8 single fathers, 26 single mothers, 46 couples) in assisting boys' with their emotional adjustment to DMD, and accessing relevant services (Chen, 2008).

Adjustment to DMD may also be understood in terms of loss and grief (Kubler- Ross, 2005). Loss may be experienced in relation to emotional and physical factors (Hinds et al, 2005). Adjustment may incorporate 'anticipatory grief' (stages of denial, anger, despair and acceptance), whilst caring for a terminally ill child (Rini and Loriz, 2007; Holley and Mast, 2009). Research has further suggested that challenges to adjustment result from adapting to the carer role, routines and increased demands on resources (Young, Lynam, Valach, Novak, Brierton and Christopher, 2001; Dellve et al, 2006). These studies illustrate the vulnerability and distress experienced by many parents. They also show that conflict may arise within families, possibly hindering their efforts to adapt, and to facilitate boys' adjustment.

### ***2.2.2. Influences on adjustment and coping***

Further identified challenges include continually explaining to others, and fear of explaining the condition (especially prognosis) to the child (Abi Daoud et al, 2004). Qualitative work has demonstrated that these parents often feel guilt at having possibly 'done something' to cause their child's condition (Webb, 2005). This may especially be the case surrounding mothers who have carrier status for DMD.

Additional factors such as declining physical abilities, especially around adolescence (when children seek independence) may result in heightened parental stress. In addition, decisions that impact the child's life span (e.g. ventilation; back surgery or requesting no intervention) may also represent times of additional stress. Research indicates continual medical intervention and associated uncertainties are found to threaten family adjustment (Sloper, 2000).

Illustrating this, Garralda, Muntoni, Cunniff and Diaz Caneja, (2006) mixed methods investigation of mothers' (n=17) views and adjustment to boys' use of callipers, found their introduction was a trigger for a repeat of the reactions felt at diagnosis. High psychological risk for depression and anxiety was found for 41% of mothers compared to expected 20-30% in the normal population. Garralda et al (2006) reported a trend for higher levels of mental health problems in parents of a boy currently using callipers, highlighting the impact of loss of walking on carer adjustment.

Consistent with this, ongoing stress may be experienced by parents due to constant deterioration in the condition (Gagliardi, 1991). This steady loss of function characterising DMD has been described as a 'cycle of loss' by parents (Kornfield and Siegal, 1979). Abi Doud et al (2004), identified a period of psychosocial transition to DMD, allowing pacing of this process, through use of coping mechanisms such as denial and 'magical thinking' (Abi Doud et al, 2004).

In relation to use of coping strategies, Fitzpatrick and Barry (1990) and Gagliardi (1991) have described the reactions of DMD families (3 families, 3 fathers). Gagliardi (1991) found that families withdrew from others due to challenges of the condition, leading to a 'smaller world'. When efforts were made to interact with other DMD parents with similar issues, however, coping was easier. Fitzpatrick and Barry (1996) investigated the processes of communication in DMD families (number of fathers unknown), linking this to coping. Using interviews with parents of 23 boys with DMD, communication issues amongst families were identified.

The authors found that spouses rarely discussed the deteriorating nature of the child's condition, and this served as a means of coping. In addition, inability for parents and boys to talk together about the condition was noted. Fitzpatrick and Barry (1986) described this as an attempt to take life bit by bit, however, frustration about this lack of communication was perceived as a major stressor, especially where parents differed in their preference to discuss or avoid the condition.

In contrast to this, Webb's (2005) interviews with 16 families (15 mothers, 1 father alone) of a child with DMD, in relation to coping, concluded parents did not report such problems. Overall, studies indicate a range of coping strategies, both adaptive and detrimental to coping.

Acknowledging the tendency for some families to withdraw (e.g. Fitzpatrick and Barry, 1986; Gagliardi, 1991), Soutter, Hamilton, Russell, Russell, Bushby et al, 2004, introduced boys and their families (74 families, 17 including father; 3 father alone) to personal computers and the internet, in the 'Golden Freeway Project'. This



was seen as a means of reducing social isolation and promoting boys' independence. The authors reported that the presence of the computer resulted in increasing family cohesiveness, boosting boys' confidence and reducing isolation, demonstrating the impact of a basic intervention on previously identified family adjustment factors.

In summary, parents of a child with DMD are seen to experience a number of stressors, requiring adjustment as DMD progresses (Witte, 2004). Quality of parental adjustment is also related to quality of child adjustment (Chen, 2008). Adapting to the child's condition may be influenced by factors such as family interaction, communication and involvement (e.g. Taanila et al, 2001; Coleman, 2002). Findings also suggest that parents of DMD boys reporting adjustment problems may have less 'reserves' of emotional coping skills (Abi Doud et al, 2004). Research (e.g. Love, Street, Harris and Lowe, 2005) has indicated that access to support can facilitate more productive coping strategies.

### ***2.2.3. Characteristics of DMD (boys' psychological adjustment and disability) and parental outcomes***

A number of authors have reported an association between boys' behavioural problems and parental adjustment. Thompson et al, (1992) identified a bi-directional effect, whereby, family adjustment problems were identified among children with psychosocial adjustment problems. Good parental functioning also predicted fewer behavioural problems and better psychological adjustment (Thompson et al, 1992).

Reid and Renwick (2001) focused on the period of adolescence to investigate DMD family stress. They found that DMD adolescents presented poor psychosocial

adjustment compared to healthy peers, with family stress significantly associated with boys' adjustment and intellectual ability. As with Thompson et al (1992), levels of parental stress were predicted by child psychosocial adjustment, with socio-demographic factors (age, employment) unrelated to outcomes. Despite high stress levels, the authors found that few belonged to a support group.

Also focusing on adolescence, Witte (1985) found that parents of 13-16 year olds with DMD (adolescents) presented higher levels of stress and high levels of guilt and problems in discussing death issues. Supporting this, Nereo et al's study of mothers of boys with DMD, suggests that stress is raised due to the child's problem behaviours, rather than as a result of physical demands of the condition (Nereo et al, 2003). In this study, disease progression and level of disability were not found to relate to parents' (mothers') stress. In contrast, behaviour was found to predict stress in terms of parent-child interactions. Results indicated that DMD mothers' stress reduced as time progressed (Nereo et al, 2003).

Abi Doud et al (2004) compared parental outcomes in DMD parents with data from a national population health survey (1999). The author concluded DMD parents were more likely to experience clinically significant depression, and lower self-esteem and mastery. Age of parents and child and level of disability were not predictors of these outcomes, however, parents of boys aged over 13 years were more at risk of depression. Supporting earlier findings (Chen et al, 2002), Abi Doud et al (2004), found DMD parents show fewer emotional coping skills compared to healthy controls.

These studies point to teenage years as resulting in specific periods of increased stress. Fitzpatrick and Barry, (1986) found that as boys aged and became more isolated, parents believed psychological input was more important. Further, this study demonstrated that although psychological disorder was identified in 52% of boys, parents were not seeking professional input. Supporting this, a survey of behaviour problems in children with neuromuscular dystrophy, conducted by Darke, Bushby, LeCouteur and McConachie, (2006), found that behaviour, social and communication problems were common. Frequently reported needs included assistance with child behaviour problems and communication skills. Consistent with Fitzpatrick and Barry (1986), families presenting these problems were those reporting high levels of unmet needs for services. The authors suggest clinics should screen for children at risk of such problems and plan for the families' needs. Confirming the issue of unmet needs, Chen (2008) highlighted that DMD parents found access to care challenging.

These findings add to earlier qualitative work, (Buchanan, LaBarbera, Roelofs and Olson, 1979), finding that physical problems resulting from DMD were only mentioned by 4 of 25 families interviewed in relation to reactions to their child's condition. Further corroborating these results, more recent studies (Chen and Clark, 2007; Chen, 2008) found level of disability was not significantly correlated with family function. As such, the combination of both family issues and behavioural problems may be faced by boys and carers, affecting mutual psychological adjustment (Heaton, Noyes and Sloper, 2005).

Relationships of child/ family variables with family function<sup>9</sup> were also investigated by Chen and Clark (2007), with greater family function significantly correlated with earlier age at diagnosis. This was explained by additional time taken to adapt when boys are diagnosed earlier and parents have good access to professional care. Neither income, employment, nor disability level, were correlated with family function. Consistent with Reid and Renwick (2001), the authors suggest that level of disability was not related to family function, or other predictors, due to stress resulting from the distress and emotional reactions, not practical care demands associated with DMD (Chen and Clark, 2007).

Given the intensity of care-taking, contrary to expectations, the previous studies indicate that disability level per se, is not associated with parental adjustment (Chen and Clark, 2007; Chen, 2008). The most recent available DMD study, in Taiwan (Chen, 2008), investigated mediators affecting DMD family function. Again, the author expressed surprise that level of disability was not associated with family function or other predictor variables. He explains this as a result of parents attending support groups. This was suggested to assist with adjustment to the deteriorating nature of the condition (Chen, 2008).

Prospective research provides further justification for targeting a progressive condition. Dellve et al's (2005) investigation of stress and well being in parents of children with rare diseases, demonstrated that compared to other forms of disability, mothers of children with progressive disabilities reported high stress levels, often as

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<sup>9</sup> Defined as problem solving, communication, roles, affective responsiveness/involvement and behaviour control (Chen and Clark, 2007).

a result of social isolation. These parents reported a higher physical and emotional load than parents of non- progressive disabilities. As a progressive condition, DMD involves uncertainty of disease progression, and related treatments, which have been identified as a risk factor by various authors. The deteriorating nature of the condition, for some, may be emphasised through the introduction of physical interventions.<sup>10</sup>

Such unpredictability as to what to expect throughout the child's treatment, has been shown to heighten uncertainty in parents (Cohen, 1999). In light of findings of no association between adjustment and child's level of disability, a key factor may be parents' reactions, not the condition itself. Despite uncertainty, parents may avoid discussion of future treatments. Erby, Rushton and Geller, 2006, focused on 'advanced care planning' with 17 DMD families (fathers unknown), demonstrating avoidance of emotionally difficult aspects of the condition, as boys approached perceived milestones. The authors also identified a lack of communication in relation to advance care planning. Areas relevant to psychological adjustment included swinging between hope of future treatments, and avoiding discussion of treatment related issues (Erby et al, 2006).

Considering the high level of physical intervention required for children with DMD, technology dependence studies may identify areas for interventions with DMD families. For example, Heaton et al's (2005) interviews with families of technology

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<sup>10</sup> Callipers or in physiotherapy terms, 'knee ankle foot orthosis' (KAFOS) are used to prolong walking as muscles weaken. Deciding to use KAFOS requires a high level of decision making, involving a cost-benefit assessment on the part of the parents and child. The introduction of KAFOS has been highlighted as an indicator of the declining health of the child (cessation of walking), and a tendency towards increased maternal mental health problems (Garraida et al, 2006).

dependant children who were cared for at home, concluded that care of technology-dependent-children places high demands on families.

Compared to other conditions (cystic fibrosis/renal disease: Holyroyd and Guthrie, 1986), and healthy controls (Nereo, Fee and Hinton, 2003), DMD parents (usually mothers) present with higher stress levels. Lower stress has been found compared to parents of acutely ill children (fever: Chen et al, 2002); with similar levels to cerebral palsy (Nereo et al, 2003). Boys' behaviour problems were found to have a significant impact on parental adjustment, with DMD parents also found to use less coping strategies.

In summary, from these studies it appears that although factors such as demographics, and level of disability are not generally associated with DMD parental adjustment, key stages in condition progression (e.g. loss of ambulation) may require more intensive support.

#### ***2.2.4. Gender differences in adjustment to DMD (and other chronic conditions)***

Research has shown that parents may cope differently with the child's disability, with support needs for DMD parents differing for fathers compared to mothers (e.g. Chen et al, 2004). Because sample sizes are small and inclusion of fathers is minimal, available DMD research does not usually compare parental differences in coping. Only 2 DMD papers (Firth, Gardner-Medwin, Hosking and Wilkinson, 1983; Chen et al, 2002) specifically refer to gender differences in parental outcomes. A brief

summary follows, with attention paid thereafter, to key findings across other chronic conditions.

Firth et al (1983) found that fathers<sup>11</sup> had more difficulties accepting DMD diagnosis than mothers, suggesting this was due to loss of expectations for their son. Issues surrounding use of coping strategies may also arise, with DMD research (e.g. Chen et al, 2002) highlighting mothers' use of both more, and different types of coping strategies than fathers. In their study of coping in parents of DMD and children with a fever, Chen et al (2002) demonstrated DMD mothers had increased impact, conflict and help needs compared to fathers. Fathers required more information and needed more help from resources, with mothers found to use more emotion focused coping strategies (Chen et al, 2002). Supporting this, Buchanan et al, (1979), described fathers' difficulties in revising expectations for sons as they acknowledged their son would not fulfil expected 'male' roles.

Drawing upon the chronic illness literature, various themes emerge, emphasising differences between mothers and fathers in their quality of adaptation. These include social support, role-identity and coping differences. For example, in relation to support, studies suggest that men are less likely to seek help and in clinical contexts it may be challenging to offer support to fathers (e.g. Oliver, Pearson, Coe and Gunnell, 2005). Researchers have suggested that male identity/masculinity may be threatened by having an ill child, exposing fathers as vulnerable (Chesler and Parry, 2001; Walker, 2004; Seidler, 2007).

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<sup>11</sup> The study refers to 'parents' however, despite commenting that the study mainly relied on mothers does not state numbers of fathers participating.

Research has also shown that distress experienced by wives, relationship problems and concerns for the child's future are key sources of distress for fathers (Gray, 2003). As reported by mothers, fathers may be reluctant to discuss the child's condition or ask for help, taking on this burden alone. This may result from conforming to the need to 'be strong' (Pelchat and Perreault, 2003; Waite-Jones and Madill, 2008). In terms of gender roles, traditional roles, influencing certain tasks and expectations, may be heightened amongst parents whose child has a disability. This has a direct impact on levels of stress, well being and coping mechanisms (Grey 2003). Furthermore, differences in parental priorities have been identified, with fathers' concerns focused on visibility of disability and mothers' on daily living (Britton and Moore, 2002).

Differences in perception of parental reactions have also been identified, for example Oliver et al, (2005), described fathers reporting they spoke frequently with partners about their feelings, however, mothers reported this was rare and that fathers had problems in expressing themselves (Oliver et al, 2005). Hovey, (2006), also identified differences between parenting concerns, relating to children's health and key concerns of wives. She concluded that professional anticipatory guidance, dissemination of information and encouragement in use of informal support systems were needed by fathers (Hovey, 2006).

Paediatric cancer studies allow comparison of parental coping with a chronic, often terminal condition. Bonner et al (2007) compared 23 mothers and fathers who were main carers to a child with cancer, finding no differences on self-report measures of



distress or condition-specific parenting stress. Most parents, however, were above normative levels on measures of psychological distress with a higher number of fathers presenting raised levels of depression (Bonner et al, 2007). Qualitative research with fathers of children with cancer (Neil-Urban and Jones, 2002), has described fathers struggling to accept diagnosis and experiencing role strain, followed by self-doubt, worry and frustration. Focus groups revealed the vulnerability fathers experienced, described by the authors as: *"stupefying and causes self-doubt, general worry, and frustration with the medical care they receive"* (Neil-Urban and Jones, 2002, p.97).

In a prospective study, Goldbeck, (2001), compared maternal and paternal coping styles for parents of a child with cancer to juvenile arthritis/epilepsy. Parents of children with cancer were found to develop more rumination, defence and information, and less social support seeking strategies compared to controls. Mothers reported more frequent and effective coping, compared with fathers. The author suggests interventions should be developed to allow parents to deal with differences that may have a negative impact on the child (Goldbeck, 2001).

Exploring differences in the experiences of parents of children with Downs syndrome, Pelchat and Perreault, (2003), used separate focus groups with 9 parents (four couples). The study focused on actual and expected roles of parents within family sub-systems and perceptions of the normalization/stigmatization experience. In this study, mothers fared better than fathers in terms of interpersonal and group communications. Fathers' expectations were harder to fulfil than mothers. They

were also more attuned to the outside world than within the family. Mothers were found to be less demanding with more self-focused expectations, and spoke of the fathers' discomfort with the child's condition (such as being seen in public).

Needs, values and worries had an impact on fathers' level of involvement with the child, perceived competence, responsibility for activities and areas they did not wish to be involved in. Mothers revealed lack of confidence relating to fathers' parental abilities, suggesting they felt fathers were not capable of caring adequately for the child. For fathers, being faced with what they perceived as bias from professionals and other families when their child was compared to others, was a major stress. They were also reluctant to seek help as this would mean acknowledging the child was different, highlighting their limitations as a parent (Pelchat and Perrault, 2003). Consistent with other research (e.g. Waite-Jones and Madill, 2008), these studies emphasise a tendency for fathers to try to cope alone without help seeking, whilst often experiencing high levels of distress.

#### ***2.2.4.1. Social support- relationship to adjustment***

Research with a variety of chronic illness groups, has demonstrated that carers with more support are more able to use effective coping strategies to meet psychological needs (Love et al, 2005). Support might be especially important for DMD parents, who may have increased demands on their emotional coping skills (Abi Doud et al, 2004). Support has been defined as *"meaningful contact with people through a mutually supportive communication exchange"*, and may include friends, family, health and government services (McCubbin and McCubbin, 1993, p.214). Families of

DMD boys have reported unmet needs relating to services (Darke et al, 2006), indicating that support is best considered in relation to both 1) professional services and 2) friends and family.

Being part of a social network is believed to provide emotional support (McGarry and Arthur, 2001), with a positive relationship between support and parental adjustment (Kazak, 1989; Soutter et al, 2006). Social support derived from a social network is, therefore, believed to act as a protective mechanism and a coping strategy (Taanila et al, 2001). Research on maternal emotional adjustment (Wallender et al, 1989) has found socio-environmental factors (e.g. family support; social support networks), not disability, to be key influences.

Social support needs may vary by gender, with a prospective longitudinal study of parents of children with cancer (Hoekstra-Weebers, Jaspers, Kamps and Klip, 2001) finding social support variables accounted for higher levels of paternal, not maternal, distress. Furthermore, support predicted paternal, not maternal distress. Also, fathers who were less satisfied with support and experienced negative interactions, were at increased long-term risk (Hoekstra-Weebers et al, 2001). Research with parents of a child with rheumatoid arthritis, (McNeill (2004), demonstrated fathers attempting to show strength for others, with over reliance on self-support strategies.

Of relevance may be fathers' perceptions of their expected role. Many described having lost friendships and supportive networks, due to their need to spend time with the family (McNeill, 2004). Of interest is Pelchat and Perreault's (2003) finding

that fathers may be reluctant to seek emotional support. It has also been suggested that the social network<sup>12</sup> may be perceived as a source of emotional burden for some, leading some fathers to isolate themselves to prevent this (Walker, 2004).

In light of Reid and Renwick's (2001) finding that few DMD parents belonged to support groups, of note is a recent national report (Recognising Fathers, 2009) based on a U.K. survey of fathers (n=250) of children with learning disabilities, which identified social isolation as a key problem. The research found 40% felt unable to discuss their situation/concerns with friends, with many having lost access to social networks (Towers and Swift, 2009).

Intervention studies including DMD families are rare, however, using group therapy in the management of fatal childhood disease, Kornfeld and Siegal, (1979) demonstrated DMD parents required longer to feel at ease with other DMD parents, compared to parents of a child with Spinal Muscular Atrophy (SMA) (Kornfeld and Siegal, 1979). These parents were also found to avoid friendships, stating they did not want pity or to make others feel uncomfortable. Group work highlighted a delay in acceptance, due to gradual loss of ability. This may delay mourning and subsequent adjustment.

Two key issues are evident: parenting a child with DMD can lead to isolation, and support can promote better adjustment. The above studies suggest that measures of social support merits inclusion in father related research. In light of these

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<sup>12</sup> A support network may be defined as number of contacts available, with support relating to perception of quality of received support (Hoekstra-Weebers et al, 2001).

findings, it is possible to draw upon the research across paediatric chronic conditions. Key factors are raised stress levels, potential vulnerability of fathers, and influence of factors such as expectations and visibility of the condition. Due to the nature of the 'illness stages', and 'physical markers' perhaps in the case of DMD this will be even more profound. Thus, the impact on fathers is evident across a range of conditions, highlighting the need to redress their neglect in child health research.

### **2.3. Understanding experiences of DMD parents**

A limited number of qualitative DMD studies focus on parental coping reactions. The earliest study explored reactions of DMD families to the condition, using interviews with 25 families, (2 fathers) of a DMD child (Buchanan et al, 1979). Interviews revealed that most parents (76%) reported chronic emotional stress as the most significant problem in condition management, with marital conflict identified in 50% of families. Within this context, anticipation of future stress (including unpredictability of DMD) was the main issue. Physical problems resulting from DMD were only mentioned by 4 of the families. Coping mechanisms included isolation and 'magical thinking', whereby parents believed their son was different and would not decline. 52% showed over-protection (e.g. lack of discipline) towards the child, often as a reaction to guilt and helplessness (Buchanan et al, 1979).

Kornfield and Siegel's (1979) study also identified denial of the reality of the condition, with uncertainty as a further issue. The authors observed parental discussion groups over an eleven-week period, to investigate parental attitudes and to promote coping. They compared two parent groups (5 parent dyads of boys with

SMA and DMD) on key issues, with both raising similar issues as concerns, specifically: death, over-protectiveness, and sexuality. In relation to over-protectiveness, this was noticed in early stages of DMD where parents desired to shield themselves and child from later stages of the condition. Sexuality was a difficult area for all parents, with DMD parents denying this area and withholding information in this area from sons as they felt they would not live to experience this. Often parents of SMA boys had grieved at an early stage compared with DMD parents, who appeared less inclined to talk about death or the future. It was felt these parents were denying the severity of the condition for longer as their child looked normal with slow decline.<sup>13</sup>

Firth et al (1983) interviewed parents in relation to experiences at diagnosis and early stages. Problem areas included service delivery, daily activities, and emotional problems. A key theme was that parents felt they had received poor information from professionals. Psychological issues included boys' depression, parental distress at witnessing decline, and parental isolation. Again, using interviews, Witte (1985) found that mothers bore the brunt of the child's frustration, often reporting feeling a love-hate relationship and anger as a result. The author explained the process as the boys' attempts to communicate defensively by projecting behaviour they could not verbally convey. Witte (1985) also suggested that parents focused on the child's behaviours in an attempt to avoid issues such as death.

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<sup>13</sup> The facilitators encouraged parents to become aware of the impact of their attitudes, work through loss and towards acceptance. The importance of understanding relationships within families, in relation to promoting condition management was emphasised. Group work allowed parents to realise they were not alone, accept the child and the impact on their own wellbeing (Kornfeld and Siegal, 1979).

Parental perspectives were also studied by Bothwell, Dooley, Gordon, MacAuley, Camfield and MacSween (2002), relating to services, health issues and quality of life issues 'now' and 'in future'. In relation to mental health issues, parents (31 families, fathers unknown) reported social isolation, anger and depression as key areas, especially for those with older boys. Parents whose son had been diagnosed over 6 years ago felt psychiatry input was more important compared to parents of younger boys.

Only one study, Morrow (2004), included interviews with the boys themselves.<sup>14</sup> The boys' input was described, however, as extremely limited. Boys felt they could discuss their condition with parents, however, parents avoided certain areas such as death. Well-adjusted families were able to communicate clearly, included parental recreation and received outside support. Communication styles, especially avoidance of end of life discussion, were found to exacerbate grief and increase anxiety.

In attempting to understand the families' experiences, Gagliardi (1991) used a naturalistic enquiry approach. Interaction with different members of three families occurred over 10 weeks, with follow up at 12 months. Six issues emerged as common themes, including: loss of hope for normality; society's confirmation of the impossibility of normality; dynamics of family; a smaller world; letting go/ hanging on and things must change. These factors were compared to a process of adaptation, with families seen to move through stages of recognition, working out

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<sup>14</sup> It is interesting to note the observation of one previous author in this field. In relation to conducting couple interviews with 6 DMD families (n fathers: 3), Morrow (2004) notes: 'fathers on the whole had little to say, but listened intently to their wives, nodding appropriately and interjecting only to paraphrase a particular comment' (Morrow, 2004).

and resolution. In essence, Gagliardi described the family, in contrast to the child or parents in isolation, as experiencing and adjusting to the condition. In relation to adjustment, Gagliardi found mothers to be overprotective and fathers not to spend a lot of time with sons, due to working long hours. One father admitted this was because it was easier to be away from home. Consistent with previous studies (e.g. Fitzpatrick and Barry, 1986) families tended to withdraw and not discuss DMD.

Findings of Fitzpatrick and Barry (1990) also reflect the tendency to avoid discussion of DMD, with most parents reporting difficulties communicating with sons and being troubled by this. Overall, an increased risk of parental adjustment problems is found throughout the above studies. Poor adjustment is not inevitable, however, as contrasting results (Webb, 2005) are reported whereby parents (23 parents: 7 both; 15 mothers; 1 father alone) interviewed about coping have experienced usual reactions of anger and guilt, but overall coped realistically and positively. In this study, parents wished to be perceived as experts and to empower sons to live life to the full (Webb, 2005).<sup>15</sup>

## **2.4. Overall summary of DMD literature**

### ***2.4.1. What is the impact of DMD on parental adjustment and what are their experiences?***

Different insights have been provided by different analytic approaches. Throughout quantitative studies, parents of boys with DMD are consistently reported to present higher levels of psychological distress compared to controls (Thompson, 1992; Chen

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<sup>15</sup> It is worth noting that the author has a son with DMD



et al, 2002 and 2007; Holyroyd and Guthrie, 1986; Abi Doud et al, 2004). Notably, the child's associated behavioural problems, not the condition itself in terms of severity and care demands, leads to a detrimental impact on parental adjustment (Nereo et al, 2003; Reid and Renwick, 2001).

Parental adjustment is affected by witnessing indicators of deterioration in the child, and isolation (Firth et al, 1983) and negative parental attitude towards the child (Buchanan et al, 1979). Quantitative work highlights DMD families using fewer and less adaptive emotional coping strategies (Chen et al, 2002; Firth et al, 1983; Abi Doud et al, 2004). For example, use of palliative coping methods was found to predict depressive symptoms (Thompson et al, 1992). Problems also result from loss of expectations (Firth et al, 1983) resulting from diagnosis. Qualitative studies generally support these results, with parents describing chronic emotional stress as a key problem (Buchanan et al, 1979). Bothwell (2002) also reported parental mental health issues resulting from social isolation, anger and depression. Issues having an impact on coping included guilt, fear, relationship problems (Buchanan et al, 1979) and worry about future stress (Buchanan et al, 1979). Psychological problems result from parental tendency to adopt unhelpful coping strategies. These include denial, overprotection, and avoidance (Kornfeld and Siegal, 1979).

Detrimental coping strategies such as withdrawal are also reported, especially for parents of older boys (Bothwell, 2002). Isolation, along with overprotection was reported in an early study, (Buchanan et al, 1979). Other authors cite strategies of withdrawal (Gagliardi, 1991) and avoidance (Witte, 1985). Where reported

separately, fathers are found to display more problems coping with diagnosis (Firth et al, 1983), and may avoid contact with the child (Gagliardi, 1991).

A number of quantitative papers report communication problems (Darke et al, 2006; Fitzpatrick and Barry, 1990) and avoidance of emotionally painful topics (Erby et al, 2006) between parents and sons. Qualitative work describes parents avoiding certain discussions with sons, such as death (Morrow, 2004) and sexuality (Kornfeld and Segal, 1979). This was an attempt to protect the child and parent from the child's impending death. Denial and overprotection such as this, was also reported as a common parental coping strategy (Buchanan et al, 1979). Poor adjustment was not inevitable, however, with Buchanan et al (1979) finding better-adjusted parents more likely to communicate openly; focus on the present; seek recreation, and gain support outside the family.

Few qualitative studies explore the processes parents go through in attempting to adjust. Gagliardi, (1991),<sup>16</sup> described parents working through a series of stages. Group therapy has also revealed that due to the child looking 'normal' and the slow progression of DMD, a repeat cycle of loss, adjustment and loss was associated with DMD (Kornfeld and Siegal, 1979). Witte (1985) also described a sequence of events in adjustment. Initially, diagnosis led to parents experiencing various stages of grief. Following shock, coping skills developed, involving an attitude change focused on maximising son's quality of life.

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<sup>16</sup> Three stages of adaptation were described: attempts to deal with feeling detached from the world, leading to feeling loss, different and fear; adjusting to DMD to maintain family balance, and recognising life must continue both within and outside the family (Gagliardi, 1991).

Taken together, results of studies overall indicate a number of factors relating to parental adjustment: lack of support at diagnosis and ongoing support issues; boys' problem behaviour and emotions; problems with communication; and use of dysfunctional coping strategies. Unmet needs included support with child's behaviour and communication problems (Darke et al, 2006; Chen, 2008), and emotional problems and daily activities (Firth et al, 1983). However, little information is available to describe the processes involved and fathers' perceptions.

#### ***2.4.2. Which child/condition specific variables relate to DMD parental adjustment?***

The fact that the child's level of disability did not predict parental adjustment was indicated in a number of quantitative studies (Chen and Clarke, 2007; Abi Doud et al, 2004). As suggested by Chen and Clark, 2007, it is possible that this is influenced by the heightened emotional issues surrounding DMD, and importance placed on this. Qualitative work supports this, with few parents in one study citing physical problems as a key factor (Buchanan et al, 1979). Adolescents' emotional and behavioural problems, however, were found to be predictors of parental adjustment (Reid and Renwick, 2001). The reverse (bi-directional) effect was also found in this study (suggesting reciprocal effects), with levels of family stress predicting psychosocial adjustment in boys (Thompson et al, 1991).

Qualitative work highlighted that behavioural problems resulted from frustration at lack of condition improvement. This was often the case when parents had not approached the topic of condition decline with boys, or where they denied the

progressive nature of DMD (Buchanan et al, 1979). Parents of older boys are found to be more at risk of depression (Abi Doud et al, 2004; Garralda et al, 2006). Additional work (Garralda et al, 2006; Erby et al, 2006) has demonstrated the importance of awareness of parental reactions at illness stages characterising DMD.

## **2.5. Conceptual framework to guide the quantitative study**

Throughout the literature, various factors are shown to influence parental stress, including parental characteristics (Reid and Renwick, 2001; Abi Doud et al, 2004), child characteristics (Thompson et al, 1992; Nereo et al, 2003), and social factors (Soutter et al, 2004). Overall, this indicates stress results from more than provision of practical care (Morrow, 2004; Raina et al, 2005). Within the limited DMD psychosocial literature, there remains a lack of inclusion of conceptual models,<sup>17</sup> to guide research and provide an explanatory framework. Only 2 DMD studies refer to a specific framework: Thompson et al, 1992 (*transactional theory of coping and stress*, Lazarus and Folkman, 1984) and Chen and Clark (2007), who included *the resiliency model of family stress, adjustment and adaptation* (McCubbin and McCubbin, 1993).

A number of models, however, are available to guide research, such as the Cognitive Stress and Coping Model (Lazarus and Folkman, 1984) and the Risk Resilience Model, (Wallander et al, 1989). Within these models, background variables, carer characteristics, social factors and care-giving demands may have an influence on adjustment. Chen and Clark (2007), whilst investigating DMD family function,

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<sup>17</sup> A call was made in the early 90s (Thompson et al, 1992), for conceptually driven studies to explore processes associated with outcomes within DMD affected families, but this does not appear to have been followed up.

utilised a conceptual framework derived from McCubbin and McCubbin's (1993) 'Resiliency Model of family stress, adjustment and adaptation.' The Resiliency Model combines ecological and developmental perspectives, placing adjustment within a broad context. From an ecological perspective, problems result from interaction of individual vulnerability and the impact of stressful experiences within specific social contexts. The developmental perspective considers the impact of stress in relation to timing, as resiliency may vary in light of challenges at different stages (Walsh, 2003).

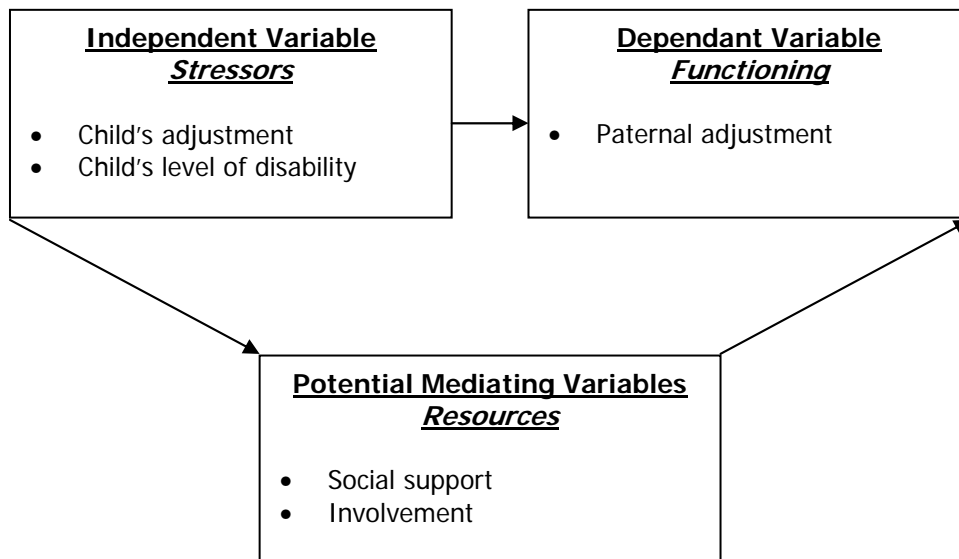
According to the model, response to stress involves adjustment followed by adaptation. In response to a stressor, adaptation is determined by interacting components. The stressor and severity interact with vulnerability that is influenced by the build up of stressors. In turn, vulnerability interacts with patterns of functioning, which subsequently influences resistance resources. Adjustment includes appraisal of the stressor, balancing individual and family members' needs and thereby influencing problem solving and coping strategies. Buffers may include characteristics (individual/family) or resources from support networks, with resilience promoted when resources are perceived as equal to stress (McCubbin and McCubbin, 1993).

The proposed framework has previously been applied to DMD research, investigating family function specific variables, and provides a guide for investigating father

specific factors. For this reason, the model (outlined in Figure 1, below) was chosen as a framework to guide selection of variables and the current study design.<sup>18</sup>

**Figure 1**

*Study variables as related to the Resiliency Model*



The following investigation studies relationships between child's behaviour and disability (stressor variables), social support, involvement, family (resources) and father psychological adjustment (adaptation/functioning). This thesis focuses on fathers in order to contribute to the 'missing' element of available frameworks.

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<sup>18</sup> Brackets below include variables addressed in this study, according to the model.

Shaped by the literature review, the following aims and research questions guided the investigation:

## **2.6. Aims and research questions**

### ***Aims***

#### ***To use both quantitative and qualitative methods to:***

- 1.** Investigate psychosocial adjustment in fathers of a son with DMD (Research Questions: 1 and 2).
- 2.** Explore fathers' perspectives on caring for a son with DMD (Research Questions: 3 and 4).

### ***Research Questions***

- 1.** Is paternal adjustment associated with child's level of physical ability and psychological/ behavioural adjustment?
- 2.** Is paternal adjustment associated with perceived level of involvement and support?
- 3.** What is the experience of parenting a son with a progressive terminal condition?
- 4.** What are fathers' views of, and suggestions for support?

## Chapter 3

### Methodology Rationale

This chapter briefly outlines the rationale for undertaking a mixed methods study to investigate fathers' adjustment and experiences. The process of adopting a combined approach is presented in Appendix 18, (p.243).

#### 3.1. Design

##### ***3.1.1. Mixed methodology***

Drawing upon the theoretical underpinnings of quantitative and qualitative approaches, a cross sectional design, incorporating qualitative methodology, was used to investigate the psychological adjustment and experiences of fathers of a son with DMD. This design was chosen to enhance understanding of the impact of DMD, by using a spectrum of research tools. This incorporated questionnaires, interviews and written accounts to highlight different phenomena. The study therefore comprised two components, consisting of distribution of questionnaire batteries, complemented by written accounts and a series of 15 in-depth interviews. Each component will be described separately in chapters 4 and 5 respectively.

##### ***3.1.2. Rationale and benefits of a mixed methods approach: application to this study***

The rationale for combining methods was based on quantitative methods summarising outcomes, whilst qualitative methods explored context and underlying dynamics. Thus, a complementary approach was adopted. The key advantage of the



design was allowing investigation of a novel area, where a qualitative approach is beneficial (e.g. Pope and Mays, 1995), along with simultaneous application of previously validated research tools. This served as a means of building upon previous quantitative research in other chronic illness contexts, whilst exploring additional areas. As each type of data collection has strengths and weaknesses, the combination allowed access to advantages of each.

Downfalls of quantitative methods, such as lack of contextual information, were accounted for by learning from fathers' experiences. To illustrate- the extent of fathers' involvement or coping with a child's condition was measured using questionnaires. However, this did not describe the experience, therefore, the addition of a qualitative element helped to identify, explore and understand the perspectives of fathers.

The quantitative strand complemented the qualitative approach by using data to answer specific research questions. The addition of qualitative interviews facilitated in-depth understanding, offering a 'real' or valid account of the topic (Greenhalgh and Taylor, 1997). This promoted reliability of the data, offering an inclusive interpretation of the research problem (Matveev, 2002; Foss and Ellefsen, 2002). It also heightened confidence in the validity of the data and subsequent interpretation (e.g. Connor et al, 2001).

### ***3.1.3. Integrating mixed methods***

Various methods of interpreting mixed-methods studies have been described, including use of qualitative data to 'explain' quantitative results, or using qualitative

results to produce hypotheses to test quantitatively (e.g. Foss and Ellefsen, 2002). In this thesis, both sets of results are drawn together in the 'triangulation design model' described by Creswell, Fetters and Ivankova, (2004). This model given equal emphasis to both types of data, with findings brought together in the discussion, as supporting or contradictory evidence for results (Creswell et al, 2004).

#### ***3.1.4. Summary of methodology rationale***

In summary, use of mixed methods offered a complementary methodological approach, drawing upon the strengths of both as appropriate. This allowed an attempt to pinpoint associations between phenomena, and to describe the nature and processes involved with the phenomena being measured. The main benefit of the study design stemmed from highlighting associations between child and fathers' variables, and attempting to explain the processes involved. The design allowed identification and understanding of relevant areas for the development of future interventions.

## Chapter 4

### Methodology I: Quantitative

#### 4.1. Study rationale

Questionnaire choice was underpinned by a theory driven approach, considering variables as outlined in the Resiliency Model (McCubbin and McCubbin, 1993). Research has indicated the influence of factors such as illness related demands (e.g. Hockstra-Weebers et al, 2001; Raina, O'Donnell, Schweltnus, Rosenbaum, King et al, 2004); child variables (Chen, 2008); involvement (Wysocki and Gavin, 2006); and social support (Wijnberg-Williams, Kamps, Klip, Hoekstra-Weebers, 2006; Dewey and Crawford, 2007), for parental adjustment to children with paediatric conditions. However, this is largely derived from research with mothers, thus measures were chosen to investigate these factors in fathers.

#### 4.2. Recruitment

##### *4.2.1. Recruitment via national organisations*

Data collection occurred between February and October 2007. Fathers were recruited via national charity organisations: Muscular Dystrophy Campaign (M.D.C.); Scottish Muscle Network (S.M.N.); Parent Project U.K. (P.P.U.K.), and the Duchenne Family Support Group (D.F.S.G.). In addition a 'snowballing' technique was used once interviews commenced. Relevant organisations were approached via introductory letters and emails. These communications included a description of study aims, reasons for interest in DMD and a copy of the participant information

sheet. Organisations were then contacted by telephone with a view to discussing their potential assistance with recruitment.

All Muscular Dystrophy Campaign family care advisors in the U.K. (n=12) were advised about the study at a national meeting. In this case, one representative agreed to speak on the researcher's behalf to determine interest amongst other care advisors. All agreed to facilitate recruitment, which involved distributing participant packs containing an invitation letter, consent form, information and debrief sheet, demographics proforma, 4 questionnaires and 'comments sheets' (see appendices, p. 196).

Within the Muscular Dystrophy Campaign, contact was made with the Scottish Muscle Network based at Yorkhill Hospital, Glasgow. In the case of Parent Project U.K., contact was made with the Chief Executive of the charity. It was necessary for the research proposal to receive clearance from a Steering Group of experts and parent representatives.<sup>19</sup> Approval was granted, and this allowed access to a Register<sup>20</sup> that had been set up by the charity. The Duchenne Family Support Group circulated an email appealing for participants, and Contact a Family included a mention of the research in an e-newsletter.

Table 1 (p.60) summarises the approach negotiated with each organisation.

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<sup>19</sup> An attempt was made to consider the NHS 'INVOLVE' model of user involvement and this panel served as appropriate initial feedback in terms of relevance to both parents and professionals.

<sup>20</sup> The purpose of the Register was to record cases of DMD and Becker muscular dystrophy (a less severe form of dystrophy) in order for medical scientists to locate boys for clinical trials.

This register represents the only official record of DMD families in the UK, and is seen by the charity as a step towards promoting the involvement of boys into trials.

**Table 1: Process of contacting participants via charities**

	<b>Muscular Dystrophy Campaign</b>	<b>Scottish Muscle Network</b>	<b>Contact a Family</b>	<b>Duchenne Family Support Group</b>	<b>Parent Project U.K.</b>	<b>Snowball technique</b>
<b>Method of contacting participant</b>	-Initial promotion of the study via an advocate at a national meeting -Distribution of packs via care advisors -Advertising in 'DMD News' -Distribution of flyers at Scottish muscle network meetings	Leaflet distribution/ word of mouth at meetings	Advertising of project in e-newsletter	Personal request from Chairman of DFSG, distributed via email network and DFSG newsletter	Distribution of packs to participants with cover letter from PPUK	Contacts made via suggestions of people recruited
<b>Number recruited</b>	N=26	N=5	N=0	N=12	N=4	N=3

#### **4.2.2. Contacting fathers**

Once ethical approval was obtained, fathers were identified and contacted via the methods previously outlined. Organisations usually included a cover letter supporting the study and requesting involvement. The researcher offered to talk to people to address any queries. No financial incentive was offered. Response was via stamped addressed envelope to the researcher.

Regular contact was maintained with those involved in facilitating recruitment, to maximise response rate. Consent to participate from fathers involved contacting the researcher directly via Queen Margaret University, or by returning completed questionnaire packs. Due to the low numbers of children with DMD in Scotland (approximately 200),<sup>21</sup> and low numbers recruited in related projects, recruitment challenges were anticipated. Previous research has demonstrated a number of

<sup>21</sup> There is no official record of DMD cases in the U.K. (although the P.P.U.K. Register is going some way towards establishing this)

challenges when recruiting fathers in DMD and father specific studies (e.g. Firth et al, 1983; Lloyd, O'Brian and Lewis, 2003). This may account for why previous studies have involved modest sample sizes (e.g. Hovey, 2005).

The sample size of 50 in the current study reflects some of the challenges of recruiting fathers and families affected by terminal childhood conditions. From 177 packs distributed, 56 completed packs were returned.<sup>22</sup> Of the 56 completed packs, the final 6 were received late in the study, following data analyses for the target of 50 fathers. Data in these 6 cases were not included. Including late replies, response rate for the study was 32%.

### **4.3. Sample description**

#### ***4.3.1. Inclusion and exclusion criteria***

Fathers of boys aged up to 18 years (n=41) completed the full battery of parent-report questionnaires. Fathers of children over the age of 18 (n=9) completed all questionnaires excluding the Strengths and Difficulties Questionnaire. Exclusion criteria for completing questionnaires were 1) an unconfirmed diagnosis of D.M.D. and 2) fathers of a deceased son.

#### ***4.3.2. Participants***

The sample comprised 50 fathers (age range: 34-63) of children (age range: 3-33 years) diagnosed with DMD from across the U.K: Scotland, England and Wales. The wide age range of children in the sample is acknowledged. In order to have a

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<sup>22</sup> Intention to participate was expressed by a further 10 fathers who contacted the researcher to request packs, and another 3 were 'volunteered' by their wives. In these cases, no completed packs were received.

sufficient sample size and therefore statistical power, a decision was made to include older 'children'. This was in light of 1) a small population to draw upon, and 2) contending with recruitment issues.

The nature of the study concerned paternal adjustment in the context of care-taking perceptions. As children were fully dependent on parents, and all requiring ongoing care, care-taking issues were faced regardless of the child's age. The sample size was comparable to or exceeded previous quantitative DMD focused studies (e.g. Holyroyd and Guthrie, 1986 (43 'parents'); Reid and Renwick, 2001 (36 'families')).

#### **4.4. Procedure**

##### ***4.4.1. Ethical issues***

The research proposal was submitted to the Psychology Ethics Panel at Queen Margaret University in December 2006, with ethical approval received in February 2007. Throughout the research, adherence to the British Psychological Society Guidelines for Ethical Research and Code of Conduct was upheld (British Psychological Society, 2006). This included addressing issues surrounding consent, confidentiality and data protection, and ensuring interviewees were advised of sources of support and distributed a debrief sheet (appendix 3, p.199).

##### ***4.4.2. Issues Arising***

For some participants, it was acknowledged that discussing matters relating to their son's condition would be sensitive. The researcher strove to promote an atmosphere in which participants felt able to express themselves, without feeling awkward. After

each interview, discussion focused on the experience of being interviewed. In general, fathers held favourable perceptions of the interview. A number stated that talking about such issues to the researcher was cathartic. This effect has frequently been noted by researchers in palliative care (e.g. Lowes and Gill, 2006).

During fieldwork, it was noted that a number of fathers scored above 'cut off' for psychological adjustment problems. Relevant ethical issues, such as a desire to intervene, have been considered by other researchers. For example, Sheikh, Hurwitz and Parker (2001), uncovered high levels of psychological morbidity amongst general practice managers, in a questionnaire survey.<sup>23</sup> The authors concluded that confidentiality must be upheld, with subsequent contact of 'at risk' participants deemed inappropriate. They emphasised the roles of researchers as distinct from clinicians, but acknowledge the discomfort this may involve for researchers (Sheikh et al, 2001).

#### ***4.4.3. Confidentiality and informed consent***

To ensure confidentiality of personal data, all participants were identified using an anonymised code (initials and chronological number in order of data collection, e.g. AB01). All named material was held securely, accessed only by the researcher. S.P.S.S. data was on a password protected computer. To ensure informed consent, participants were provided with written information sheets (appendix 1, p.196), and

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<sup>23</sup>The authors found 17% indicating scores of depression. In a reflective report, they questioned the relationship between respondent and researcher, and moral responsibility to take further action. They concluded that in the case of questionnaire respondents, there was a moral obligation to respect confidentiality, and clarified the role of researcher as carrying a different responsibility to that of clinician. In undertaking research with distressed individuals, it was also acknowledged that the researcher takes on an element of 'burden', thereby suggesting the need for appropriate supervision procedures. Supervision was in place throughout, and beyond, the fieldwork for this thesis.



encouraged to ask questions. They were informed of their right to withdraw from the study at any time. All participants provided written consent.

#### **4.5. Completion of questionnaire batteries**

Information was sent by organisations to fathers as follows: 4 standardised questionnaires and 2 measures constructed by the researcher specifically for the study. These were i) demographics proforma, recording ages of father and son, fathers' occupation, address, age of son at diagnosis and willingness to be interviewed; ii) a questionnaire to measure satisfaction with support from friends, professionals and family.

Questionnaires were completed in participants' homes, and returned by post directly to the researcher. According to feedback from participants, and timing of battery completion in test runs, the battery of questionnaires required approximately 40 minutes to complete.

##### ***4.5.1. Description of measures***

The full content of packs sent to participants was as follows: consent form, invitation letter, information sheet, demographics proforma, General Health Questionnaire (G.H.Q.), Functional Disability Inventory (F.D.I.), Strengths and Difficulties Questionnaire (S.D.Q.), Dads Active Disease Support Scale (D.A.D.S.) and Likert rating scales for satisfaction with support. The measures have been widely used in previous research studies in chronic illness contexts, demonstrating their reliability and validity with paediatric populations e.g. S.D.Q: cerebral palsy (Parkes, White-

Koning, Dickinson, Thyen, Arnaud et al, 2008); F.D.I: sickle cell and juvenile arthritis (Palermo et al, 2004); D.A.D.S: cystic fibrosis, and spina bifida (Wysocki and Gavin, 2004). All questionnaires (apart from D.A.D.S) have previously been used with mothers (n=17) in a study investigating user views and adjustment to callipers in DMD (Garraida et al, 2006). The following questionnaires were administered:

#### ***4.5.2. General Health Questionnaire (G.H.Q-12: Goldberg, 1978)***

To assess mental distress and risk for psychological disorder in the father, the 12-item G.H.Q. was used. This measure has demonstrated validity in research with a range of populations, including unemployed men (McKenna and Payne, 1989), and mothers of boys with DMD (Garraida et al, 2006). Different cut-off levels for G.H.Q.-12 (Banks et al, 1983; Goldberg, Gater and Sartorius, 1997) have been cited throughout the literature, with scores above cut off indicating high psychiatric risk. 'Caseness threshold', however, has been recommended as 3/4 for the 12-item G.H.Q., using bimodal scoring ('G.H.Q. score': 0-0-1-1), (Goldberg, 2002, in Manual of the General Health Questionnaire; Jackson, 2007). Additional literature has cited cut-off levels for G.H.Q.-12, as 2-3 (Banks et al, 1983) and 1-2, (Goldberg et al, 1997).

The current study used a conservative cut off point of 4 to indicate 'caseness'.

#### ***4.5.3. Functional Disability Inventory (F.D.I: Walker and Greene, 1991)***

The F.D.I. was used to assess perceived illness impairment (activity limitations and severity of dysfunction) as a result of DMD. Measurement of child functional impairment (difficulty in age-appropriate physical and psychological functioning, due

to physical health status) is important in order to determine impact on child and carer's lives (Palmermo et al, 2004). In this study, it was used to examine associations between functional disability with paternal adjustment.

The measure is described as a *"global measure of functional disability for use in research regarding the impact of illness on children's physical and psychosocial functioning"*, which may be used with a range of paediatric conditions to assess activity limitations and severity of dysfunction (Walker and Greene, 1991, p.40). The F.D.I. has documented stability and sensitivity, and has been validated in a range of paediatric populations. These include abdominal pain (Walker and Greene, 1991), recurrent headaches, juvenile arthritis and sickle-cell disease (Palmero, Zebracki, Cox, Newman and Singer, 2004) and recently, DMD (Garralda et al, 2006). Whilst there is no set cut-off point, higher scores indicate higher impairment and physical limitation.

#### ***4.5.4. Strengths and Difficulties Questionnaire (S.D.Q. Parent-Report: Goodman, 2001)***

The S.D.Q. assesses child and adolescent emotional and behavioural symptoms over the previous 6 months. It comprises a behavioural screening tool of 25 items, rating psychiatric symptoms in five areas: emotions, conduct, hyperactivity, peer problems and pro-social behaviour. The extended version of the questionnaire was used, including an optional 'Impact Supplement' towards the end of the S.D.Q. This assessed the everyday distress experienced by child and family relating to the child's mental health problems. The S.D.Q. has demonstrated validity and reliability, in

research with various U.K. paediatric populations (Goodman, 2001), including cerebral palsy (Parkes et al, 2008), and DMD (Garraida et al, 2006). Cut-off points allow identification of total and sub-scale scores as, 'normal' (N), 'borderline' (B) and 'abnormal' (A). Combined overall scores from sub-scales (excluding pro-social, which gives a 'stand alone' score), present a total difficulties score reflecting the extent of emotional and behavioural symptoms.

Cut-off scores identify possible 'symptom caseness', defined as follows: Total: N:<sup>24</sup> 0-13; B: 14-16; A: 17-40; Emotional: N: 0-3; B: 4; A: 5-10; Conduct: N: 0-2; B: 3; A: 4-10; Hyperactivity: N: 0-5; B: 6; A: 7-10; Peers: N: 0-2; B: 3; A: 4-10; Pro-social: N: 6-10; B: 5; A: 0-4; Impact: (0-10) N: 0; B: 1; A: 2+. Scores of 2+ for the 'impact score', indicate significant impact relating to chronicity of child's problems, distress to the child and burden on family.

#### ***4.5.5. Dads Active Disease Support Scale (D.A.D.S: Wysocki and Gavin, 2004)***

The D.A.D.S. was used to explore perceived paternal contribution to disease management. The authors based the measure on the social support literature focusing on supportive actions and social cognition surrounding support. They describe D.A.D.S. as a "*measure of amount and helpfulness of father's contribution to family adaptation to conditions*" (Wysocki and Gavin, 2004, p.232). The questionnaire comprises two 24-item sub-scales: *amount* of involvement offered and perceived *helpfulness* of involvement. These sub-scales yield separate scores for

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<sup>24</sup> N= normal; B= borderline; A = abnormal, according to cut off scores on the SDQ (Goodman, 2000)

each aspect of paternal involvement. Each item assesses perceptions of involvement in common management tasks (emotional and practical support). For each item, respondents are requested to rate on a 5-point scale, the amount and helpfulness of paternal involvement in common management tasks. 'Amount' items are scored: 1=0%; 2= 25%; 3=50%; 4=75%, and 5=100%. 'Helpfulness' items are scored using a 5 point Likert-scale (1= harder; 2= neither harder or easier; 3= slightly easier; 4=easier and 5=much easier).

The measure has been used with various paediatric populations requiring intensive medical regimes, including cystic fibrosis, phenylketonuria (P.K.U.) and spina bifida (Wysocki and Gavin, 2004), showing validity with paediatric populations.

#### ***4.5.6. Satisfaction with support***

A satisfaction with support scale was designed to explore associations between paternal adjustment and perceived satisfaction with support. This measure was adapted from a previous DMD study, where it was used to document attitudes of mothers towards boys' calliper use (Garralda et al, 2006). Perceptions of satisfaction with support received from family, friends and clinical staff respectively were rated, using a 6 point Likert scale, 0 (poor) – 5 (excellent).

Table 2 (p.69) summarises properties of the measures, including sub-scales, and Chronbach's alpha for normative samples. Guidelines for interpretation of questionnaires are presented in Appendix 11 (p.213).

**Table 2: Summary of questionnaires: areas measured, sub scales and Cronbach's alpha<sup>25</sup> for normative samples.**

Measure and author	Area	Sub-Scales	Cronbach (normative)
<b>General Health questionnaire (G.H.Q.-12)</b>	Parental mental distress and risk for psychiatric disorder	Somatic, anxiety, social dysfunction, depressive symptoms.	<u>Chronbach a</u> = .83 Sensitivity: 84.6%
<b>Goldberg (1997).</b>	[Cut off: 3 = risk, using bimodal scoring system]	4 point Likert. Score: 0 – 12. Bimodal scoring = 0-0-1-1	Specificity: 89.3% Reliability: .78- .95
<b>Functional Disability Inventory (F.D.I.)</b>	Child's physical difficulties with daily activities: Illness impairment;	<u>12 items:4 scales.</u> General tasks – parent completed based on child's physical abilities	<u>Cronbach r</u> = .86 -.91 <u>Test-retest:</u> parent-report .64.
<b>Walker and Greene (1991)</b>	psychosocial functioning.  Assesses activity limitations in children and adolescents with a variety of pediatric conditions.  [Cut off: n/a. Increased scores = greater level of disability]	<u>15 items.</u>  1 - 5 (1: no trouble; 2: little; 3: some; 4: a lot; 5: impossible)  (total score 0-60)	
<b>Strengths and Difficulties Questionnaire (S.D.Q., parent completion)</b>	General functioning and adaptation of child.  [Cut-offs for parent report: 'Normal', 'Borderline', 'Abnormal' for each scale]	Peer problems; conduct; emotional and pro-social behaviour scales. Total family impact score.	<u>Cronbach a</u> = .73 <u>Re-test:</u> r =65
<b>Goodman et al (2001).</b>	Total: N: 0-13; B: 14-16; A: 17-40 Emotional: N: 0-3; B: 4; A: 5-10 Conduct: N: 0-2; B: 3; A: 4-10 Hyperactivity: N: 0-5; B: 6; A: 7-10 Peers: N: 0-2; B: 3; A: 4-10 Pro-social: N: 6-10; B: 5; A: 0-4 Impact: (0-10) N: 0; B: 1; A: 2+	<u>25 items: 5 scales.</u>  Pro-social not added to total score.	

<sup>25</sup> In all cases, Chronbach's alpha indicates good levels of reliability

<b>Dads Active Disease Support Scale (D.A.D.S.)</b>	Amount and helpfulness of paternal involvement in paediatric disease management	Level of involvement – emotional and instrumental support tasks regarding illness management.	<u>Cronbach a</u> = .92 for scores for amount, helpfulness, and total.
<b>Wysocki and Gavin (2004)</b>	[Cut-off: n/a]	<u>24 items; Likert scale 2.</u>	<u>Test-retest: r</u> = Range .75 (fathers 'amount') to .82
<b>Likert scales to record satisfaction, recorded on comments sheet designed specifically for study Format was based on structure used in a previous DMD study</b>	Perceived support [cut off: n/a]	Likert scales in order to rate levels of satisfaction with support from family; clinic; friends  Score: 0-5	N/a
<b>(Garralda et al, 2006)</b>			

#### 4.6. Data Analyses

The study aims, and research questions, determined the choice of analyses. A power calculation (Cohen, 1992) informed the minimum sample size for the quantitative component. This was based on the number of independent variables, research questions and method of analysis.

Data analysis was conducted using SPSS for descriptive, correlation and multiple regression analyses. Guided by the Resiliency Model (McCubbin and McCubbin, 1993), multiple variables were measured. Descriptive analyses were followed by correlation analyses to identify variables of relevance for the regression analyses. Subsequent multiple regression analyses allowed investigation of the strength of association between variables. Relationships between possible risk factors (independent variables such as child's emotional and behavioural problems) and outcome measure (fathers' mental health status) were investigated in this way.

Potential predictors were child-related variables (condition specific and adjustment), and perceived paternal involvement and support.

Before applying univariate analyses, normality, kurtosis and homogeneity of variance were examined. Similarly, prior to regression analysis, checks were made to ensure underlying assumptions were met.



## Chapter 5

### Methodology II: Qualitative

#### 5.1. Study rationale

As previously outlined, the aim of the qualitative study was to explore fathers' experiences and views. Recording of participants' experiences, in their own words, allowed insight into phenomena that could not be understood using solely quantitative methods. Incorporating interviews into the design facilitated a collaborative approach with participants, attempting to place quantitative findings in context and remaining true to participants' perspectives.

#### 5.2. Recruitment

##### *5.2.1. Selection via the quantitative study*

Participants completing questionnaires were requested to note interest in being interviewed. Of the 50 participants in the quantitative study, 2 did not wish to be interviewed, 8 did not state any preference and 40 expressed willingness to participate in an interview.

Quantitative data from completed questionnaires were available before interviews, allowing access to information relating to, for example, area of residence, boys' ages and fathers' mental health. As completed questionnaire packs were received, interviews were arranged with consenting fathers across Scotland, England and Wales until a proportion was interviewed in each. Of those agreeing, 15 fathers from across the U.K. were interviewed.

Attempt was made to represent a roughly equal divide of Scottish and English participants. This included fathers from Scotland (n=7), England (n=6: 3 North, 3 South) and Wales (n=2). Selection of interviewees was partly dictated by logistics. Due to financial and time limitations, face to face interviews (interviews 1-8) were conducted in Scotland and Northern England. Interviews further afield were conducted by telephone.

### **5.3. Sample description**

#### ***5.3.1. Inclusion and exclusion criteria***

For the qualitative component, there was no restriction on the age of the child as exploration of a range of experiences was sought.

#### ***5.3.2. Profile of fathers interviewed***

A total of 15 fathers (mean age 48.4, age range: 34-60), of sons aged 8-32 (mean age 16.1) were recruited for the qualitative study.<sup>26</sup> The sample size was comparable to previous qualitative studies of DMD mothers (e.g. Garralda et al, 2006, n=17). Interviewees represented fathers of sons at different ages, allowing exploration of potential associations between child's stage of disability and care-taking issues. The sample of 15 participants represented a broad range of experiences. This included varied perspectives, covering early childhood before the condition deteriorates, to adolescence, early adulthood and losing a son. The cohort of interviewees included a father as the sole carer, a father who had lost a child to DMD and a father of 2 boys with DMD.

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<sup>26</sup> One father of a deceased son was also included at his request. All but this individual also completed questionnaire batteries for the quantitative strand.

A summary of participants is presented in Table 3 below.

**Table 3: Summary of interviewees**

Interview No	Region	Domestic situation	Age of father	Son's age at diagnosis	Age of Son	Years since diagnosis	Mode of interview
1	Scotland	With partner	46	2	12	10	Face to face
2	Scotland	With partner	60	10	25	15	Face to face
3	Scotland	With partner	57	In utero	21	20	Face to face
4	Scotland	With partner	51	3	13	10	Face to face
5	Scotland	With partner	46	At birth	15	15	Face to face
6	England	With partner	51	6	8	2	Face to face
7	Scotland	Single father- sole carer	34	5	15	10	Face to face
8	Scotland	With partner	missing	3	15	12	Face to face
9	England	With partner (full time carer, whilst wife works)	52	6	13	7	Telephone
10	England	With partner	60	6	32	26	Telephone
11	England	With partner (own son: 13 and step son: 2 both with DMD)	39	1 month	13 (based experience s on 13 year old)	1	Telephone
12	England	With partner	46	4	8	4	Telephone
13	Wales	With partner	missing	N/a	DEAD	N/a	Telephone
14	England	With partner	50	4.5	26	21	Telephone
15	Wales	With partner	38	4	9	5	Telephone
<b>Mean</b>			48.4 (34-60)	4.8	16.1 (8-32-deceased)	11 (1-26)	

## **5.4. Procedure**

### ***5.4.1. Ethical issues and informed consent***

As with the quantitative strand, written informed consent was obtained prior to conducting interviews (section 4.4.3, p. 63). Due to the nature of the topic, guidelines of sensitive interviewing were followed (e.g. Britten, 1995). Names and identifiers were removed from transcripts, and data were held in a locked cabinet accessed only by the researcher. As noted, confidentiality was maintained and mechanisms were in place to ensure data protection.

### ***5.4.2. Conducting interviews***

Interviews were conducted between May and September 2007. Eight of the 15 interviews (n1-8) were conducted face-to-face, taking an average of 1.4 hours, and ranging from 1 to 2 hours. The remaining seven interviews (n9-15) were conducted by telephone. Telephone interviews of approximately 45-minute duration were used, ranging from 30 minutes to one hour. Average duration for all interviews was 1 hour, 12 minutes. Interviews were conducted in either the participants' home or workplace.

Interviews concerned experiences and perceptions of specific areas including diagnosis, coping/ adjustment, involvement, support, needs and services. Collins (1998) defines interviews as '*dynamic social interactions wherein multiple dialogues are constructed*' (Collins, 1998, p.1). It has been highlighted that interviews are vulnerable to influence by interviewer beliefs (e.g. Greenhalgh and Taylor, 1997). As

such, the researcher attempted a reflexive approach, considering interactions with participants.<sup>27</sup>

### **5.4.3. Written information**

Written information was recorded on an optional 'comments sheets' (appendix 5, p.204) distributed with questionnaire batteries. This was designed to cover similar areas to the interview, and comprised a summarised version of the interview guide. A section was included for fathers to write about additional issues they felt were important.

This technique allowed fathers to respond to sensitive issues at their own pace, including experiences and perceived needs. The rationale for this was that for some men it might be easier to write about experiences 'anonymously', rather than talk directly about them. Psychology researchers have suggested written accounts may be more reflective and focused than interview transcripts, thus assisting data analysis (Handy and Ross, 2005). Although optional, only 2 of 50 men completing questionnaires did not complete comments sheets.

## **5.5. Description of interview guide**

A structured interview guide was devised (appendix 4, p.201), with the general format based on previous work with mothers of a child with DMD (Garralda et al, 2006).<sup>28</sup>

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<sup>27</sup> A reflective diary and field notes were maintained throughout the research process

<sup>28</sup> The researcher had previously co-designed a semi-structured interview guide for a study investigating carer satisfaction with knee ankle foot orthosis (KAFOs), within the context of DMD (Garralda et al, 2006). This guide had been piloted and used in a study with families (participants were mother and child) affected by DMD, and the general format was adapted for use in this study.

### 5.5.1. Development of the interview guide and written comments sheet

The research questions provided a framework from which to identify general areas to explore in interviews. The semi-structured interview guide was used as a flexible tool to allow a degree of focus but facilitate generation of in-depth data around different topics. The aim was to understand fathers' perceptions or "framework of meanings" (Britten, 1995, p.252) whilst maintaining awareness of the effect of the researcher's viewpoint on the focus of the research. The resulting interview guide covered the general areas of diagnosis, coping/ adjustment, involvement, support, needs and services. Each area was covered using open-ended questions, allowing exploration of participants' views. At the end of interviews, participants were asked if there were any other issues they would like to discuss.

Table 4 below summarises the general structure of the interview schedule.

**Table 4:** Summary of interview schedule

Semi -Structured Interview Schedule	General interview topics	Questionnaire used to measure corresponding area
<b>Semi structured interview and comments sheet designed specifically for the study</b>  <b>(Based on format co-designed by the author in a previous study (Garralda et al, 2006))</b>	• Diagnosis	-
	• Coping and adjustment	• General Health Questionnaire
	• Involvement	• Dads Active Disease Support Scale
	• Needs	-
	• Perceived support	• Likert Satisfaction with Support Scale
	• Any other area interviewee wishes to discuss	-

### 5.6. Data analyses<sup>29</sup>

Interviews were transcribed and anonymised, then imported into NVivo7 (QSR International, 2006). NVivo7 was used for the storage and analysis of interview and

<sup>29</sup> See personal reflection for more detail (Appendix 18, p.243)

written material from comments sheets. Analysis was an ongoing process, informed by Grounded Theory principles (Glaser and Strauss, 1967; Charmaz, 2006). Grounded Theory has been conceptualised as *"a way to learn about the worlds we study and a method for developing theories to understand them"* (Charmaz, 2006, p.10). In light of the research questions, Grounded Theory was considered the most appropriate approach to analysis as it allowed a bottom-up method to make meaning of participants' experiences, whilst promoting theory development. It also facilitated a flexible response to developing theory- for example by addressing emerging issues.

A number of researchers (e.g. Charmaz, 2006) have proposed modified versions of grounded theory as originally proposed by Glaser and Strauss, 1967. Barbour (2000), argues that *"grounded theory is invoked with greater frequency than it is practised"*, suggesting that it is unrealistic for researchers to undertake research in a *"theoretical vacuum"* (Barbour, 2000. P.87). This refers to the pre defined ideas and understanding each researcher brings to their project, which impacts upon 'pure' emerging theory as originally defined by Glaser and Strauss (1967). This broader approach was adopted within the current study. Accounting for aspects such as use of an interview guide (partly pre-determining themes), and undertaking the literature review *prior* to data collection, it is acknowledged that the current study undertook a 'critical approach' to grounded theory (e.g Barbour, 2000; Charmaz, 2006).

Using a Constructivist interpretation of grounded theory (Charmaz, 2006)<sup>30</sup>, text was coded to form core categories, in order to generate themes (key concepts). As themes were identified within the data, a coding frame was developed and expanded, leading to categories that illustrated key findings. The aim was to generate categories and explanations, with a view to answering research questions, and exploring emerging themes. This procedure involved ongoing comparisons-similarities and differences, throughout the interview process. This method of continual comparison allowed evaluation of themes as they arose, and consideration of developing themes in light of new data (Pidgeon and Henwood, 1997).

As such, analysis and data collection was simultaneous, as a conceptual framework was developed and refined. Categories were incorporated as they arose and the process was repeated until 'saturation' or apparent lack of new themes was reached (Barbour, 2000). According to this method, theory may be understood as an explanation of categories that have emerged (Cooligan, 2004; Pope, Ziebald and Mays, 2000). In this context, the use of 'theoretical' sampling (Glaser and Strauss, 1967), exploring further individual experiences to elaborate upon themes, facilitated this process. As such, use of an interview guide served to some extent to pre-define general themes to explore. This did not restrict data generation, but served as a flexible guide, with themes explored as they arose.<sup>31</sup> To maximise validity, negative cases were accounted for.

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<sup>30</sup> Charmaz describes 'conceptual understandings developing from an inductive, thematic analysis of textual material'. Practically, this involved line-by-line study of transcripts, identification of 'meaning units' and ongoing comparison with later units. This was followed by grouping categories containing related meanings and labelling these as themes (see Appendix 18, section 1.4. for full details).

<sup>31</sup> Charmaz (2006) notes an unresolved 'tension' between data collection and 'forcing' ideas onto data, acknowledging that use of a semi-structured interview guide does not constitute imposing codes onto data! This does, however, emphasise the importance of not being restricted by set research questions.



Throughout the research process, attention was paid to evaluative criteria according to Charmaz (2006), including credibility, originality, resonance and usefulness. Acknowledging a criticism of emphasis on post hoc 'reflection' as opposed to ongoing evaluation (Morse, Barrett, Mayan, Olson and Spiers, 2002), active attempts were made to ensure credibility of data.<sup>32</sup> Attempts were made to address 'verification strategies' for validity and reliability as outlined by Morse et al (2002), including methodological coherence, sampling sufficiency, development of a dynamic relationship between sampling, data collection and analysis, theoretical thinking and theory development. To further ensure dependability, an experienced qualitative researcher<sup>33</sup> read the analysis within the context of the emerging coding frame, with general agreement overall.

### ***5.6.1. Transcribing and recording context***

Interviews were recorded and transcribed verbatim. Transcribing and writing of field notes took place within one day of conducting the interview. The researcher undertook all transcribing, with each interview taking an average of 4 hours and 40 minutes to transcribe verbatim. Field notes (appendix 17, p.234) were made after interviews, in order to facilitate and provide context for subsequent analysis. The purpose of writing field notes was to reflect on emerging issues, and to record the context of interviews and reactions of interviewees. In addition, maintaining a reflective diary allowed identification of initial thoughts, considered as the initial stage of data processing and providing context for analysis (Tilley, 2003;

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<sup>32</sup> Morse et al (2002) criticised the tendency for qualitative researchers to focus on research post hoc reflection rather than accounting for the methods used to ensure rigour throughout the research process. For example, they do not consider member checks to be a verification strategy per se.

<sup>33</sup> Dr Jo Hockley, Department of General Practice, University of Edinburgh (currently at St Bartholomew's, London).

Etherington, 2007). It has been suggested that these practices are an integral element of reflexive practice (Pidgeon and Henwood, 1997).

Awareness of the need for reflexive practice was maintained, throughout the research process. This facilitated transparency in relation to practicalities of conducting the research at each stage. Acknowledging this, a critical personal reflection is presented in Appendix 18 (p.243).

## Chapter 6

### Results I: Quantitative

This chapter presents results from the quantitative strand of the study. The purpose of this component of the study was to examine associations of paternal adjustment with 1) child's level of physical ability and psychological/ behavioural adjustment; 2) perceived level of involvement and 3) perceived support. Using the Resiliency Model (McCubbin and McCubbin, 1993) as an analytic framework, the aim was to examine which of these factors were associated with adjustment (as measured by the G.H.Q.).

Descriptive statistics are presented according to measures used, followed by analysis of relations between independent and dependent variables using bivariate and multivariate analyses. Included in the analysis were a series of t-tests to investigate differences in scores based on demographic variables. Following descriptive analysis, correlation analyses were used to determine associations amongst variables. Multiple linear regression analyses were conducted to identify variables contributing to variance in G.H.Q. scores.

As low numbers of cases were included, the 'simultaneous' method of regression analysis was used. This procedure is recommended where theory is being developed, and/or with few included cases (Coolican, 2004; Field, 2005), as was the case with this study.

## 6.1. Descriptive statistics

Table 14 (appendix 12, p.215) presents participant characteristics for the quantitative study. The mean age of fathers was 46 years (range: 34-63 years; s.d. 7.5), with the mean age of sons 14.1 years (range: 3-33 years; s.d. 6.9). Fathers with children aged over 18 (n=9) did not complete the Strengths and Difficulties Questionnaire. The majority (76%, n=38) of the sample resided in England, 4% in Wales (n=2), 18% in Scotland (n=9) and 2% in Northern Ireland (n=1). National Statistics Socio Economic Classification (2005) data, according to profession, was available for 37 participants. 57% (n=21) were in the 'higher managerial/professional' bracket; 13% (n=5) 'lower professional/higher technical'; 16% (n=6) 'intermediate clerical/technical'; 8% (n=3) 'semi routine' and 5% (n=2) 'unemployed'.

Questionnaires were analysed for 50 fathers for measures: Functional Disability Inventory (F.D.I.); General Health Questionnaire (G.H.Q.) and Dads Active Disease Support Scale (D.A.D.S.). Data were missing in 2 cases for the Support Scale, and the Strengths and Difficulties Questionnaire (S.D.Q.) was not applicable in 9 cases. Numbers included in all analyses are stated.

The current sample size compares favourably to previous quantitative father only studies (e.g. Wiener et al, 2001). The few DMD studies available have also included modest sample sizes (e.g. Chen et al, 2002; Chen and Clarke, 2007), reflecting the challenges of recruitment (e.g. Phares et al, 2005; Mitchell et al, 2007). A minimum sample of n=38 was required for large effect size power=.80 for alpha .05 (Cohen,

1988), and the sample size for all analyses in this study met or exceeded this number.

Table 5 below summarises the means, standard deviations, and score ranges for all study variables.

**Table 5:** Means, standard deviations and score ranges for all questionnaires

Questionnaire	Mean	Standard deviation	Score range	Norms <sup>34</sup> Mean (sd)
General Health Questionnaire (n:50)	3.5	3.8	0-12	-
Functional Disability Inventory (n:50)	29.7	11.7	3-57	-
Strengths and Difficulties Questionnaire: Total score (n:41)	11.5	6.8	0-29	8.4 (5.8)
Strengths and Difficulties Questionnaire: Emotions (n:41)	2.9	2.7	0-10	1.9 (2.0)
Strengths and Difficulties Questionnaire: Peer problems (n:41)	2.6	2.1	0-9	1.5 (1.7)
Strengths and Difficulties Questionnaire: Conduct (n:41)	1.8	1.8	0-6	1.7 (1.8)
Strengths and Difficulties Questionnaire: Pro-social (n:41)	7.8	1.9	2-12	8.4 (1.7)
Strengths and Difficulties Questionnaire: Hyperactivity (n:41)	4.2	2.4	0-9	4.0 (2.7)
Strengths and Difficulties Questionnaire: Impact on Family score (n:41)	1.9	2.6	0-9	0.5 (1.2)
Dads Active Disease Support Scale: Perceived amount of involvement (n:50)	2.7	.71	1.5- 4.6	-
Dads Active Disease Support Scale: Perceived helpfulness of involvement (n:50)	2.4	.68	1.1-4.2	-
Satisfaction with support: Hospital/ Staff (n:48)	3.3	1.7	0-5	-
Satisfaction with support: Friends (n:48)	3.4	1.4	0-5	-
Satisfaction with support: Family (n:48)	3.5	1.5	0-5	-

<sup>34</sup> Normative UK data for SDQ (Meltzer, Gatward, Goodman and Ford, 2000)

Table 15, Appendix 13 (p. 217), presents scores for each participant on all measures. Details of interpretation of questionnaire data and scoring are presented in Appendix 11 (p. 213).

### ***6.1.1. Psychiatric adjustment in fathers (G.H.Q.) (n=50)***

The mean G.H.Q.-12 score was 3.5 (s.d. 3.8), with a range of 0-12. According to recommendations for defining 'threshold for case definition' (Araya, Wynn and Lewis, 1992), a score of 4 indicates 'caseness'. 32% (n=16) presented above cut off with scores of 5 or more, and 6% (n=3) with scores of 4. Thus, 38%<sup>35</sup> of the sample were 'at risk', according to scoring protocols, for clinically significant problems. According to boy's ages, 40% (6/16) of fathers of boys aged 3-9 years were at risk for mental health problems, with 33% (8/16) in the 10-20 year group. At risk scores were found in 22% (2/9) of fathers of the older boys (aged 20-30+). Scores tended to be higher for fathers of younger children. Table 16c (appendix 14, p.222) outlines sub-scale and total scores on the G.H.Q.

Independent t-tests indicated *no significant differences* ( $p > .05$ ) between fathers scoring above cut off for 'risk' on G.H.Q. and those below cut-off, for the variables: total S.D.Q; total F.D.I. and D.A.D.S. amount of involvement. This suggests that fathers with higher reported mental health problems were not over-reporting child disability, child adjustment problems or their amount of involvement. *Significant differences* were found for *family impact* ( $t = -1.787$ , 37 degrees of freedom (df),  $p < .05$  (2-tailed) and *DADS helpfulness* ( $t = 2.96$ , 43 degrees of freedom (df),  $p < .05$

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<sup>35</sup> In the normal population the expected rate would be 20-30%

(2-tailed)). Regarding perceived helpfulness of involvement and impact of child related problems on the family, it is possible that those with higher G.H.Q. scores interpreted their input into child-care as less valuable.

### ***6.1.2. Child's functional ability (F.D.I.) (n=50)***

Functional Disability Inventory scores indicated that all sons were impaired, with a mean raw score of: 29.7 (s.d. 11.7). Scores ranged from 3 to 57, with scores increasing with age due to progressive deterioration. There are no standard cut off points for the F.D.I. (Walker and Greene, 1991), higher scores indicate greater disability and increased levels of impairment. Scores of 4-5 indicated 'a lot of trouble', with scores of 2-3 indicating 'a little or some trouble'.

Motor activities were impaired, with 80% (n=40) of boys finding it 'a lot of trouble/impossible' to walk upstairs, do sports (66%, n=33), or go to the bathroom (68%, n=34). Regarding social activities, 32% (n=16) found activities with a friend to be 'a lot of trouble/impossible'. Meal times were 'a lot of trouble/impossible' for 20% (n=10) and 'a little/some trouble' for 40% (n=20). 60% had 'no trouble' attending school. Minimal difficulties were found with less physical activities, such as watching television (98%, n=49, 'no trouble' or 'some trouble') and going to sleep (48%, n=24).

Tables 16a&b (appendix 14, p. 222), outline F.D.I. scores, indicating levels of difficulty faced for various daily tasks.

### 6.1.3. Child psychosocial adjustment (S.D.Q.) (n=41)

The mean total S.D.Q. score was 11.5 (s.d. 6.8), with a range of 0-29. Psychiatric risk according to cut-off scores (Goodman, Ford, Simmons, Gatward and Meltzer, 2000), was above normative levels for 22% of boys, with most problems reported in relation to emotional (borderline + abnormal: 32%) and peer related problems (borderline + abnormal: 45%). Table 6 allows comparison with normative data for the U.K. (Goodman et al, 2000).

Examining S.D.Q. scores according to level of functional ability, children presenting 'normal' scores on S.D.Q. emotions had a mean F.D.I. score of 14 (score range was 3-57 for the boys), indicating less disability. For those with 'borderline' and 'abnormal' S.D.Q. scores, the F.D.I. scores were 22 and 36 respectively. For S.D.Q. 'peer problems', those within the normal range presented a mean F.D.I. score of 30, with 32 for borderline and 34 for abnormal.

Table 6 contains percentages scoring in the 'borderline' and 'abnormal' ranges for each sub scale.

**Table 6:** Emotional and behavioural adjustment in children. Numbers and % of boys above cut off for psychiatric risk according to S.D.Q. (n= 41)

Sub scale	Borderline	Abnormal	Mean (s.d.)	Normative UK <sup>36</sup> Mean (s.d.)
<b>Emotions</b>	8% (n:4)	24% (n:10)	2.9 (2.7)	1.9 (2.0)
<b>Peer Problems</b>	24% (n:10)	21% (n:9)	2.6 (2.1)	1.5 (1.7)
<b>Conduct</b>	8% (n:4)	17% (n:7)	1.8 (1.8)	1.6 (1.7)
<b>Prosocial</b>	0% (n:0)	7% (n:3)	7.8 (1.9)	8.6 (1.6)
<b>Hyperactivity</b>	5% (n:2)	19% (n: 8)	4.1 (2.5)	3.5 (2.6)
<b>Total Score</b>	7% (n:3)	15% (n:6)	11.54 (6.8)	8.4 (5.8)

<sup>36</sup> Normative SDQ data for UK (Meltzer et al, 2000)



Table 6 shows that most problems were found for emotional (24% 'abnormal')<sup>37</sup> and peer related problems (21% 'abnormal').

**6.1.4. Impact of child's problems on family (optional 'Impact' section in extended S.D.Q. section) (n=41)**

Mean impact score was 1.9 (s.d. 2.6), range 0-9. 'Normal' scores, according to the S.D.Q. scoring protocol were found for 52% (n=21), with 'abnormal' scores for 36% (n=15), and 'borderline' for 12% (n=5). These results highlight a detrimental impact of child adjustment on family functioning in 48% of families.

Table 7 compares Impact scores with normative U.K. mean.

**Table 7: Total scores: impact on family (n=41)**

Variable	Min	Max	Mean (s.d.)	Normative UK Mean (s.d.)
Total impact on family total score	0	9	1.9 (2.6)	0.5 (1.2)

**6.1.5. Involvement (amount and perceived helpfulness) in child's medical and emotional care (D.A.D.S.) (n=50)**

According to the scoring protocol for this measure, described by Wysocki and Gavin (2004), mean item scores for perceived amount of involvement (2.7; s.d.70; range 1.5-4.6) suggests amount of involvement in disease management took place in 25-50% of opportunities over the previous 6 month period. Mean item scores for perceived helpfulness of involvement were, 2.4 (s.d. 68; range 1.1-4.2), indicating that involvement was perceived as making management of childcare tasks 'neither harder nor easier'.

<sup>37</sup> SDQ categories as defined by Goodman (2001)

**6.1.6. Satisfaction with support from hospital, family and friends (n=48)**

With a maximum score of 5 (range: 0-5), support was rated as 'good' from hospital staff by 56% (mean: 3.3; s.d. 1.7), family 59% (mean: 3.5; s.d. 1.5) with lower perceived support from friends, 50% (mean: 3.4; s.d. 1.4). Table 8 presents a summary of Support scores.

**Table 8:** Total scores on satisfaction with support scales (n=48)

Variable	Min	Max	Mean (sd)
Hospital/staff	0	5	3.3 (1.7)
Family	0	5	3.5 (1.5)
Friends	0	5	3.4 (1.4)

Table 9 summarises percentages of satisfaction with support in each area.

**Table 9:** Percentages: satisfaction with support (n=48)

Variable	Poor 0-1	Average 2-3	Good 4-5
Hospital/ staff (n: 48)	27% (n: 13)	16% (n: 8)	56% (n: 27)
Family (n: 49)	23% (n: 11)	18% (n: 9)	59% (n: 29)
Friends (n: 48)	23% (n: 11)	27% (n: 13)	50% (n: 24)

**6.1.7. Summary of descriptive statistics**

Regarding *fathers' mental health*, 38%, (n=19) of fathers were within the clinical 'risk' bracket for mental health problems, with a trend for increased problems for those with younger children. Descriptive results from the measures of *child functional ability* indicated challenges for most boys with physical activities (66-80% depending on activity). Basic social activities, such as activities with friends were also impaired for 32% boys.

Using the parent rated S.D.Q. to explore *child psychosocial adjustment* with DMD boys, most problems were found with emotional and peer problems. Descriptive results suggest a trend towards increasing psychosocial problems with increasing physical limitations. Almost half (48%, n=20) of fathers reported a detrimental *impact* of child related adjustment problems on the family.

D.A.D.S. data show a mean *amount* rating of 2.7, demonstrating *involvement* in condition management was taking place at most on half of available occasions. With a mean *helpfulness* rating of 2.4, fathers generally perceived their involvement in child-related care as making 'no difference'. Regarding *support*, fathers were mostly satisfied with the support they had received from hospital/ clinic (56%, n=27) and family (59%, n=29). Half the fathers rated support from friends as good (50%, n=24).

## **6.2. Correlations** (associations with paternal adjustment)

In the second stage of analysis, a correlation matrix was used to examine associations amongst variables. All significant correlations are presented in Table 10 (P.91).

**Table 10: Significant Pearson Correlations between measures**

	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.	16.
1. Fathers' Age	1															
2. Sons' Age	.81**	1														
3. SDQ Child Emotion			1													
4. SDQ Child Conduct	-.44**	-.41**	.32*	1												
5. SDQ Child Hyperactivity			.44**	.32*	1											
6. SDQ Child Peer Problem			.49**	.49**	.49**	1										
7. SDQ Child Prosocial Beh			-.32*				1									
8. SDQ Family impact			.64**	.44**	.57**	.63**		1								
9. SDQ Total Score			.79**	.62**	.77**	.76**	.78**		1							
10. FDI Disability	.49**	.66**	.41**	.44**	.44**	.44**	.33*	.33*		1						
11. GHQ Father s' Mental Health			.38*	.32*	.31*	.31*	.39*	.41*			1					
12. DADS Help									-.38**			1				
13. DAD Amount									.51**				1			
14. Support from Hospital														1		
15. Support from Family			-.33*						-.32*					.31*	1	
16. Support from Friends									-.44**						.36*	1

\*\* = significant at the 0.01 level; \* significant at the 0.05 level

**6.2.1. Relationships between paternal adjustment and child functional ability, demographics, child adjustment, family impact, involvement and support.**

Fathers' adjustment was positively associated with perceived amount of involvement, S.D.Q. total and a number of S.D.Q. sub-scales, as described below. Significant associations were found for child emotional symptoms ( $r= 0.382$ ,  $p<.05$ ,  $n=41$ ); child conduct ( $r=.312$ ,  $p=<.05$ ,  $n=41$ ), and child peer problems ( $r=.310$ ,  $p<0.05$ ,  $n=41$ ). This indicates that fathers' adjustment was negatively associated with increasing child emotional and social problems. Fathers' adjustment was positively associated with both S.D.Q. family impact ( $r=.395$ ,  $p=<.05$ ,  $n=41$ ) and S.D.Q. total ( $r=.409$ ,  $p<.05$ ,  $n=41$ ), highlighting problems with fathers' mental health with child's overall increase in (internalising and externalising) behavioural and emotional problems and the impact of child related problems on the family.

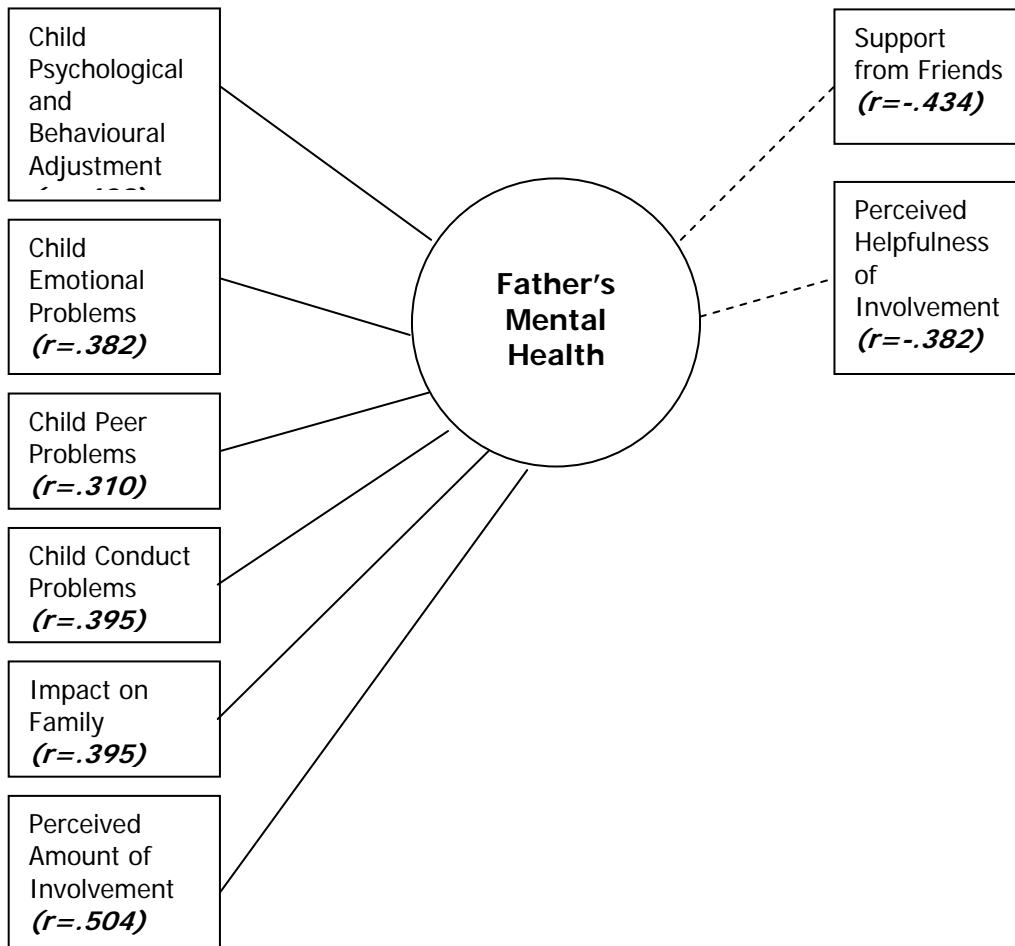
Father's adjustment was also positively associated with perception of amount of involvement with the child ( $r=.504$ ,  $p<.01$ ,  $n=50$ ), and negatively associated with perception of helpfulness of involvement ( $r=-.382$ ,  $p<.01$ ,  $n=50$ ). This suggests that fathers who are better adjusted are more involved, and those with poorer adjustment feel their involvement is less helpful.

Finally, fathers' adjustment was negatively associated with support from friends ( $r=.434$ ,  $p>.01$ ,  $n=50$ ) indicating that those perceiving less support from friendships were less well adjusted. No associations were found for level of disability, support from hospital or family.

Figure 2 illustrates relationships between paternal adjustment and child adjustment, family impact, involvement and support.

**Figure 2**<sup>38</sup>

*Relationship between paternal adjustment and above variables*



**6.2.2. Relationship between child functional ability and demographics, adjustment, family impact, involvement and support.**

Correlation analyses were conducted to determine associations between child functional ability and child and father demographics (age), child and paternal adjustment, family impact, involvement and support. A number of significant associations were identified between variables. The total F.D.I. score was positively

<sup>38</sup> For all figures, solid lines represent a positive association and broken lines negative

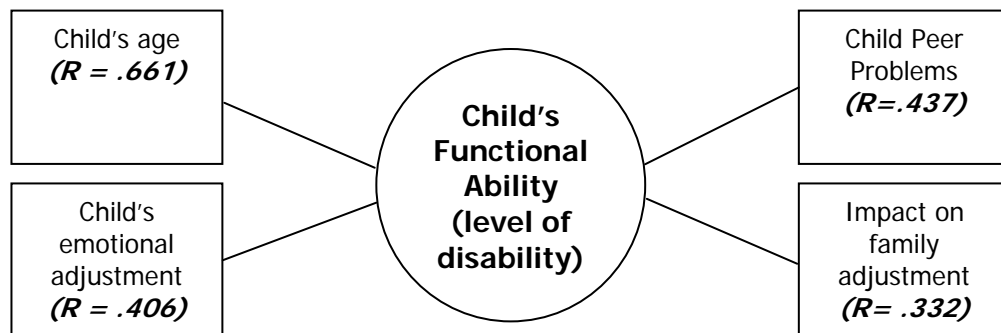
associated with son's age ( $r = .661$ ,  $p < .01$ ,  $n = 41$ ), indicating as expected that increasing age correlated with impaired functional ability.

Significant positive associations were also found for a number of S.D.Q. sub-scales. F.D.I. total score was positively associated with S.D.Q. emotions ( $r = .406$ ,  $p < .01$ ,  $n = 41$ ); S.D.Q. peer problems ( $r = .437$ ,  $p < .01$ ,  $n = 41$ ) and S.D.Q. Impact on Family ( $r = .332$ ,  $p < .01$ ,  $n = 41$ ). In sum, increasing child disability is associated with problems with emotions, peer problems and the impact on the family. No associations were found with fathers' mental health, S.D.Q. sub-scales: conduct, hyperactivity and pro-social.

Figure 3 illustrates relationships between child functional ability and child age, child emotional and peer problems and family impact.

**Figure 3**

*Relationships between Child Functional Ability and above variables*



**6.2.3. Relationships between child psychosocial adjustment and child functional ability, demographics, paternal adjustment, family impact, involvement and support.**

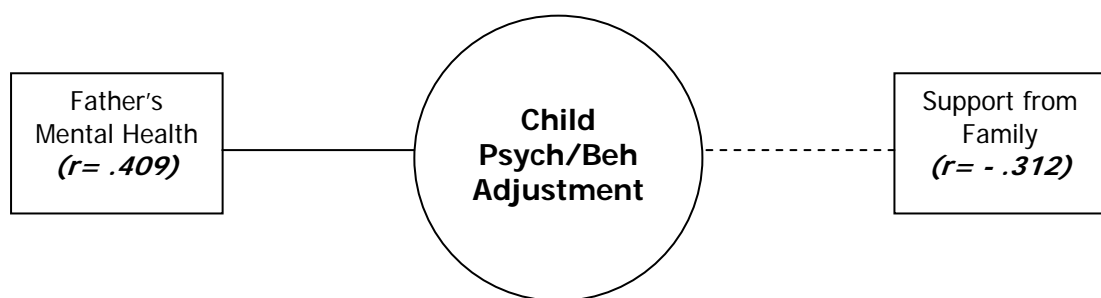
The total S.D.Q. score was significantly associated with 2 variables, fathers mental health ( $r = .409$ ,  $p < .01$ ,  $n = 41$ ) and family support ( $r = -.312$ ,  $p = < .05$ ,  $n = 41$ ). A positive association with G.H.Q. total, indicated that more perceived child

adjustment problems were associated with increasing mental health problems in fathers. The negative association with satisfaction with family support ( $r = -.312$ ,  $p < .05$ ,  $n = 41$ ), suggested that fathers perceiving less family support reported more child adjustment problems. No significant associations were found for age or involvement variables.

Figure 4 illustrates relationships between child psychosocial adjustment and paternal adjustment and support from family.

**Figure 4**

*Relationships between child adjustment and above variables*



**6.2.4. Relationships between family impact and child functional ability, demographics, adjustment, involvement and support**

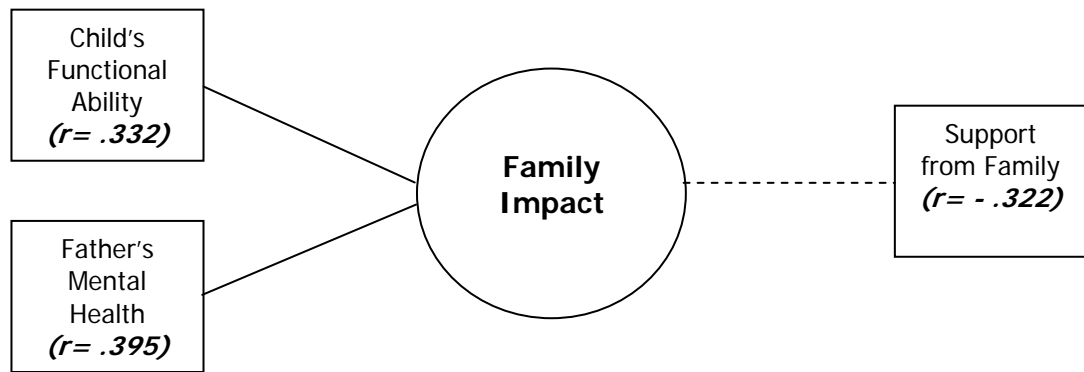
Family impact score was positively associated with F.D.I. score ( $r = .332$ ,  $p < .05$ ,  $n = 41$ ), G.H.Q. score ( $r = .395$ ,  $p < .05$ ,  $n = 41$ ) and negatively associated with satisfaction with support from family ( $r = -.322$ ,  $p < .05$ ,  $n = 41$ ). This indicates that greater child's disability was associated with greater stress on the family. The negative association with support from family indicates that increased impact on family related to lower perceived support from family.



Figure 5 illustrates the relationship between family impact and child functional ability, paternal adjustment, and support.

**Figure 5**

*Relationships between Family Impact and above variables*



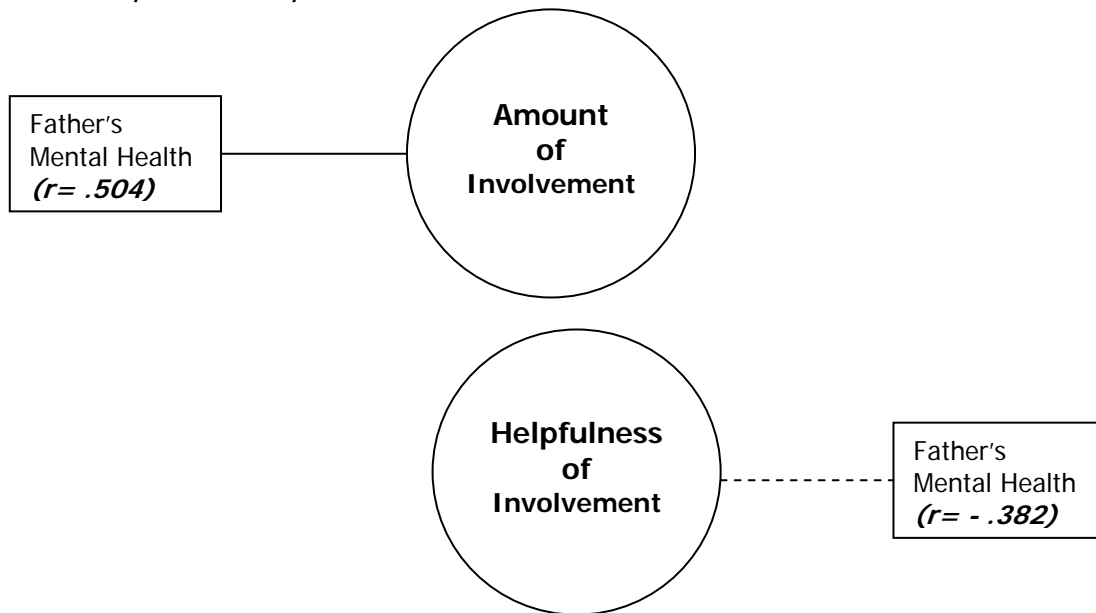
**6.2.5. Relationships between father's perception of involvement (amount and helpfulness) and child functional ability, demographics, adjustment, family impact and support.**

The D.A.D.S. sub-scale, amount of involvement, was positively related to fathers' mental health ( $r=.504$ ,  $p<.01$ ,  $n=50$ ). This suggests that increased involvement is associated with better mental health. Perceived helpfulness of involvement was negatively associated with father's mental health ( $r=-.382$ ),  $p<.01$ ,  $n=50$ ). This indicates that poorer mental health is associated with feeling less helpful when involved with the child. No other associations were identified.

Figure 6 (p.97) illustrates relationships between father's perception of involvement (amount and helpfulness) and paternal adjustment.

**Figure 6**

*Relationship between paternal involvement and above variables*



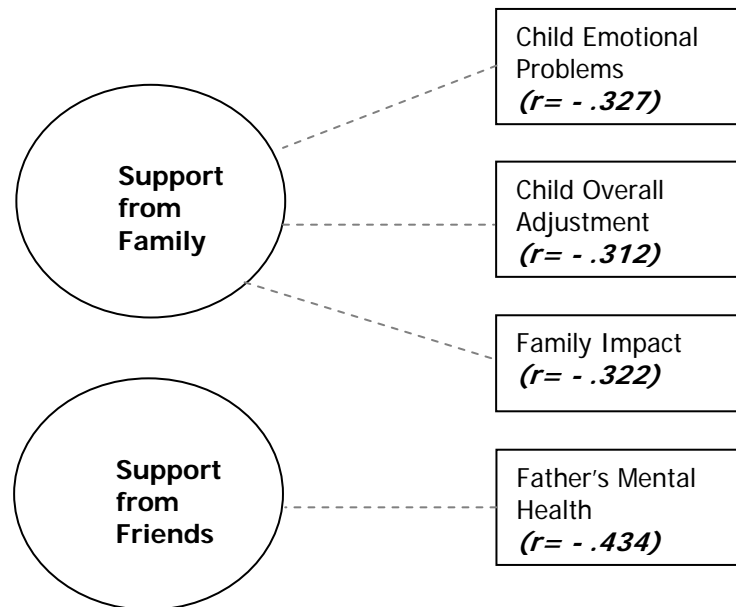
**6.2.6. Relationships between satisfaction with support and child functional ability, demographics, adjustment, family impact, and involvement.**

Satisfaction with support was found to be related to a number of S.D.Q. sub-scales and fathers mental health. Satisfaction with support from family was negatively related to child emotions ( $r=-.327$ ,  $p<.05$ ,  $n=41$ ), S.D.Q. total ( $-.312$ ,  $p<.05$ ,  $n=41$ ) and family impact ( $r=-.322$ ,  $p<.05$ ,  $n=41$ ). Support from friends ( $r=-.434$ ,  $p=<.01$ ,  $n=48$ ) was negatively related to father's mental health. This indicates that lower perceived satisfaction with support from both friends and family was associated with poorer father's mental health and child's emotional and behavioural problems.

Figure 7 (p.98) illustrates the relationship between satisfaction with support and child emotional and overall adjustment, family impact, and paternal adjustment.

**Figure 7**

*Relationships between Satisfaction with Support and above variables*



### **6.2.7. Summary of correlation analyses**

Addressing the research questions, 1) is paternal adjustment associated with child's level of disability and child adjustment? and 2) is paternal adjustment associated with perceived involvement and support?, results showed that paternal adjustment was significantly associated with child adjustment, support and perceived amount and helpfulness of involvement variables. The variables most strongly related to paternal adjustment were amount of involvement; support from friends and child's adjustment.

### **6.3. Regression analyses**

Following the investigation of univariate interrelationships, a series of multiple regressions were conducted. Prior to analysis, normality distributions,<sup>39</sup>

<sup>39</sup> Having inspected normality distributions, analysis was conducted on raw (non-transformed) data

heteroscedascity<sup>40</sup> and collinearity<sup>41</sup> were explored using scatterplots, histograms and normal p-plots<sup>42</sup>. Sub-scales of the S.D.Q. were not entered into the equation for Research Question 1, as inter-correlations indicated collinearity.

An outlier (case 14) was removed prior to analysis for Research Question 2. This analysis addresses research questions 1 and 2.

### **6.3.1. Question 1: Is paternal adjustment associated with child's level of disability and child adjustment?**

Addressing Research Question 1, simultaneous multiple regression was conducted to determine the contribution of 2 predictor variables to paternal adjustment. Overall S.D.Q. total score and F.D.I. total score were entered as predictors. According to the Resiliency Model (McCubbin and McCubbin, 1993), variables 'support' and 'involvement' are potential mediating variables. Correlation analyses indicated a significant association between these variables and paternal adjustment, therefore they were entered into the regression model as predictor (independent) variables.

#### Summary of variables

**y= G.H.Q. total (paternal adjustment)**

x1= S.D.Q. total (child adjustment)

x2= F.D.I. total (child functional ability)

Using the simultaneous entry method, a significant regression model emerged,  $F(2, 38) = 3.84, p = <.05. R^2 = .168$  (adjusted  $R^2 = .124$ ), indicating 17% (13% adjusted)

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<sup>40</sup> Lack of similarity of residual variance across predicted levels of the dependant (criterion) variable: GHQ

<sup>41</sup> Correlations amongst predictor variables

<sup>42</sup> Investigates relationship between predicted and residual values- none found

of the variance is accounted for by child adjustment (beta = .418,  $p = .01$ ). Child functional ability was not a significant predictor ( $p > .05$ ).

### **6.3.2. Question 2: Is paternal adjustment associated with perceived involvement and support?**

Addressing Research Question 2, simultaneous multiple regression was again conducted to determine the contribution of 5 predictor variables to paternal adjustment. D.A.D.S. amount, D.A.D.S. helpfulness and support from friends, clinic and family were entered as predictors. It is acknowledged that the number of predictor variables exceeds the typical recommendation for regression analysis, and accordingly the regression model may be under-powered as a result.

#### Summary of variables

#### **y= G.H.Q. total (paternal adjustment)**

x1=D.A.D.S. amount (perceived amount of involvement in child's care)

x2=D.A.D.S. helpfulness (perceived level of helpfulness of involvement)

x3=Support from friends

x4=Support from clinic

x5=Support from family

A significant regression model emerged,  $F(5, 39) = 5.71$ ,  $p = .000$ .  $R^2 = .423$  (adjusted  $R^2 = .349$ ), indicating 43% (35% adjusted) of the variance is accounted for by the model. D.A.D.S. amount (beta = .421,  $p = .002$ ) and perceived support from friends (beta = -.374,  $p = .007$ ) contributed significantly to the model, with amount of

involvement as the strongest predictor. Perceived helpfulness of involvement, support from clinic and family, were not significant predictors in the model ( $p > .05$ ). Significant predictor variables are summarised in Table 11 below.

**Table 11:** *Regression analysis for D.A.D.S. (amount and helpfulness) and Support (family, clinic, friends) variables predicting fathers' adjustment (n =46)*

Predictor Variable	Beta	P
DADS Amount (perceived amount of involvement)	.421	P = .002
Satisfaction with support from friends	-.374	P = .007

### **6.3.3. Summary of regression analyses**

In sum, results show that child psychological and behavioural adjustment, perceived amount of involvement in son's care and support from friends, were significant predictors of fathers' adjustment as measured by G.H.Q. scores. Of these variables, D.A.D.S. amount was the strongest predictor.

## Chapter 7

### Results II: Qualitative

Fifteen fathers, aged 34-60 (mean 48.4), of sons aged 8-32<sup>43</sup> (mean 16.1), participated in interviews and 48 fathers provided written accounts. Grounded theory methods (Charmaz, 2006), facilitated the development of a framework from which to understand participants' perspectives. Participants represented a range of views, with results highlighting a number of key issues surrounding fathers' experiences. Characteristics of participants selected for interview are presented in table 3 (p.74).

Appendix 15, (p.228), presents examples of extracts within the context of themes, illustrating the development of a coding frame. From the analysis 4 key themes were identified: 1) loss and acceptance; 2) support versus isolation; 3) the fight for resources and 4) race against time.

Table 12 below contains themes and sub-themes:

**Table 12:** *Themes and sub-themes*

Main theme	Sub-themes
<b>1. Loss and acceptance</b>	<ul style="list-style-type: none"><li>• Loss</li><li>• Expectations</li><li>• Guilt</li></ul>
<b>2. Support versus isolation</b>	<ul style="list-style-type: none"><li>• Adaptive coping and acceptance versus maladaptive coping</li><li>• Identity issues</li><li>• Strained friendships</li><li>• Family/marital stress</li><li>• Barriers to involvement</li></ul>
<b>3. The fight for resources</b>	<ul style="list-style-type: none"><li>• Frustration</li><li>• Spare part/exclusion</li><li>• Needs and suggestions</li></ul>
<b>4. Race against time</b>	<ul style="list-style-type: none"><li>• Images of next stages: transition to adulthood: comparison with other children</li><li>• Deterioration and making the most of life</li><li>• Decisions</li><li>• Talking about death</li></ul>

<sup>43</sup> Of the 15 interviewees one father of a deceased son was included

Each theme is described below, and illustrated using verbatim quotes.<sup>44</sup>

### **7.1. Theme 1: Loss and Acceptance** (sub-themes: loss; expectations; guilt; adaptive versus maladaptive coping)

This theme concerned fathers' reactions to their son's condition, where several losses were experienced. Initial diagnosis was described as a devastating time for all, frequently expressed in terms of loss, bereavement, and challenging previously held ideals, as written by the following fathers:

*"Your child's diagnosis is like a bombshell- it's a sentence of death on your child which you are powerless to change" (CS: 47)*

*"The most challenging time was the first two weeks after diagnosis. It was a lot to get your head around, it brings sadness to you and challenges your outlook on life" (CS: 26)*

Fathers generally said they found the diagnosis, and following weeks to be the most gruelling time. Many described feeling helpless, without knowing what the future held or understanding the condition properly, as illustrated by the following quote:

*"It was the kind of case where, when we got diagnosed with it, it was a case of, you know 'there's nothing we can do for you. There's no cure or anything for this kind of thing'. It's just a case of, at that time we felt we had to sit and for the next few years, just sit and watch your son waste away" (SSI: 7)*

An array of emotions, including anger was reported by some, in relation to the manner of finding out the diagnosis. In some cases, fathers said they felt they had struggled for a diagnosis or their concerns had previously been minimised by medical staff.

*"The way we were told by Mr X was disgusting. If we had been told people are trying to find a cure we may have dealt with it a bit better. He told us nobody is doing nothing, end of" (CS:14)*

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<sup>44</sup> At the end of each quote, the abbreviations 'SSI' (semi structured interview) and 'CS' (comments sheet) and participant number, are used to identify the source of information. All written answers are recorded in full.



*"We thought he had flat feet for 3 years, and they kept telling us we were overprotective parents, and this that and the other. So we kept pushing and pushing and pushing. Until eventually a physio noticed what was wrong with him...we went into this room, we thought it was for a blood test. We thought it was strange but went along with it. He literally handed my wife a piece of paper- literally said nothing, just handed my wife a piece of paper. The piece of paper said 'DMD- in a wheelchair between 9 and 12, and dead by 19'. Then he just left the room- absolutely nothing else. We left the hospital with that piece of paper" (SSI: 9)*

After diagnosis, various reactions were described, including delayed shock, wanting to know the full picture, and finding practical ways of moving forward such as researching and seeking information. Many described an initial period of grieving, to absorb the reality of diagnosis, often before moving forward positively.

*"I just got on with it, I just thought every day we put him to bed is another day off his life, let's get cracking here, let's get the whip going. Y' know, it took me a good 6 weeks before I started to shake the dust off, before the dark side left me. Before I started realising 'hey, we need to crack on here, we've got to do something" (SSI: 6)*

Impact of diagnosis was often framed within the context of previous hopes and expectations for their son's future and the difficulties in realising these would not be achieved. A number of fathers described a feeling of both themselves and sons losing out, and revising their 'life plan' following diagnosis.

*"If you've got two boys, you think 'great' and you plan the next 20 years. You've got this plan, then that's it, they say one word and you just kick it out the window. Well, you just think you can plan your life don't you. You think you can go cycling together. I don't do fishing or anything like that, but you can go away for camping holidays. You know, those are the sort of expectations that most men have got. If you've got two boys, you think 'great' (laughs)" (SSI: 11)*

The fact that DMD is not diagnosed immediately meant revising such expectations for physical father-son activities, and as such loss of aspects of a typical father-son relationship, was often a painful process which some found hard to deal with. From

fathers' reports, loss of expectations appeared harder to cope with, than the disability itself.

*"You go along, and at first, you see, you have all these aspirations for your son, and you don't know until he was actually diagnosed that he would never really kick a ball. Y' know and you couldn't really go out and have a robust play with, with your son. You know, eh, and that hurts, hurts. Because you feel they're losing out on something and the father's losing out on something as well. Something everyone else has. You just try and (sigh), well maybe I don't deal with it well enough, y 'know, probably at times" (SSI: 3)*

One father commented that although most parents of DMD boys he knew were together, he thought the reason some fathers left their families was due to inability to deal with this loss of their 'ideal' family.

*"All the couples I know, all the boys we know- the families are all together. But I can actually empathise with them because, you know (sighs), again it goes back to expectations. Everybody wants the perfect family. Of course the perfect family can disintegrate. I should imagine that what they feel is they need, they just want out because they realise what's involved. It's not 'oh, we'll get over it in sort of 5 years down the line'. It's basically until death do us apart" (SSI :14)*

Encapsulating others' views, another father referred to the challenge of fathers acknowledging their son would not be able to fulfil previous aspirations. He said he felt this was the reason why, following their son's diagnosis, some fathers he knew of had left the family:

*"Fathers who are not, well, [present in family] I think it's probably the fact, I probably would say that it's down to having a son who's not perfect. That, they seems to think. I think he's perfect, but y'know, he is perfect. They're mostly not there. They don't go to anything, they don't sort of think about anything or anything like that. It's not something they want to..well, they're not there, so you can't really ask them. . I suppose for men, it's to think about that and think that their son is not going to fulfil the aspirations. I don't think they can deal with that. It's difficult to deal with, but you've still got to deal with it" (SSI: 3)*

After diagnosis, further perceived 'losses' were reported as boys approached teenage years. This transition was a major challenge for most fathers, as it seemed

to represent their son being 'left behind'. The comparison with able-bodied children emphasised differences, indicating a period fathers said they found difficult. The boys' increasing disability as they entered adolescence was highlighted in light of the increasing independence of other teenagers. Two fathers described this as follows:

*"When they're younger you can sit them in their chair or whatever, they can play with their toys and stuff like that but when they're getting to a teenager, they'd like to do all the things others do. So, when you start trying to let them do things, then you see a big difference" (SSI: 8)*

*"When they're younger it's easier and you can do things. A lot of times when they're younger, they've not got the spinal fusion, so you can lift them and do different things. When they get into teenage years, and especially [son], after the spinal thing you can't lift them. The fear is there that if you lift him, you're going to stretch his spine and damage the rods that's in it or the bone graft or something. And at that age, he's missing out on a hell of a lot. That's the stage where I'm at just now, where if he's only got me for another 15-20 years or whatever and I'd like him to see some of the world before anything happens you know" (SSI:7)*

An important issue for fathers concerned their son's friendships. Fathers tended to place much importance on their son leading a normal life and having close relationships with other boys.

*"He has his friend X who is very, very, special. X is so sensitive to [son] he seems to know when he needs something before [son] has even asked for something. They just seem to have a very good relationship" (SSI: 6)*

As their son aged, some fathers described how they felt he was often losing out socially compared to other boys. In light of impaired physical abilities and restricted social activity, one father was particularly aware that this included areas such as sexuality. The following quotes illustrate these issues:

*"He went through a stage after his operation and that. He wasn't eating, but now he's fine again, back to his old self. I don't know if it was the operation or just he's a teenager. He was 14 and seeing all his pals starting to spread their wings. That's what I've noticed in the last year or so, all his wee pals are starting to live their lives. Starting to..they're growing up" (SSI: 5)*

*“As a father, you’re a male and your son is a male, so there are certain things that’s going to be coming up for him that you’ve done but he’s not able to. If you’re into your own sexuality so to speak, or things that are supposed to be masculine you will have insight into things that may be bothering him” (SSI: 4)*

Perceived isolation of their son was marked at times when most young men were gaining independence in contrast to their son who increasingly required more intensive care. Concern was expressed when it appeared that their son was socially isolated or had trouble sustaining friendships, and fathers said they worried about the impact of isolation as boys grew older.

*“The most challenging thing was friends leaving as they got older” (CS: 46)*

*“Apart from school, he doesn’t really have any friends. He’s really quite a solitary boy...there isn’t really anybody else here he can call a friend- a real friend for him if you know what I mean. So that’s interesting to see how the boys who are married do the social aspect. You do have to think about things, and it’s really when you get to the teens and especially with X just going into puberty. So you worry as he’s changing, and you do start to think more” (SSI:7)*

The child’s attitude affected how fathers dealt with the condition, with sons coping positively making it easier to cope with ongoing challenges, and facilitating fathers’ own adjustment.

*“If he was a youngster who would grizzle and moan it would make life extremely difficult. But by and large he is cheerful most of the time. It helps us cope better with it I think” (SSI: 10)*

Boys’ frustration at their physical limitations were reported, with teen years especially challenging.

*“His own frustration does come out sometimes. He’ll just sort of take a strop. You can tell the difference if it’s a teenage strop or if it’s part of his thing. It is getting harder and harder just now for him” (SSI 7)*

Issues of guilt underpinned some participants’ reports of the impact of DMD on family life. This was apparent within various contexts, including diagnosis;

restrictions on the boys' and siblings' quality of life; the need, but inability, to take a break, and through genetic issues that affected the wider family.

A number of fathers reported that upon acknowledging the terminal nature of DMD, they struggled to accept that their son would die before them. As such, thoughts of the future were reported to present emotional challenges, including guilt that they would outlive their sons:

*"Dads are not supposed to outlive their sons" (CS: 28)*

*"Knowing that he hasn't a normal future. Knowing I will bury my own son" (CS: 21)*

The impact of the condition on siblings was also referred to frequently, involving stress in trying to balance the focus between the son and other children. This was the case especially when focus was placed on carrying out 'normal' activities with siblings, whereby the son with DMD was unable to participate:

*"They two [siblings] are missing out on a hell of a lot because we don't like to leave [son] out. There's not a lot of places we can go out and get X to. I don't like to go to places where X is just sitting watching. He likes to do that, but I don't like to watch him do that, because you know he's sitting watching and he wants to be involved. So, that's taking a big pull on us at the moment. The likes of last week I was off and made the decision I was going to take the other two away for a couple of days camping for their first camp trip, and he stayed with his Gran... it was the first time I've ever went away and left him. It made me feel really guilty" (SSI:7)*

Meeting the needs of both siblings and sons could be challenging, as often fathers said they perceived one or the other as being left out. When this was the case with siblings, it could lead to attention seeking behaviour, placing increasing stress on the family.

*"Other kids are almost left out as all attention is focused on the other one. That can be a massive strain.. you've got to spread everything very carefully" (SSI: 14)*

*"Our second son, we've had a lot of trouble out of him. I think through attention seeking because obviously [son] is the centre of attention...you try your best to treat them evenly, but I've seen [son's] face where [sibling] is off on his bike and he's just sat there, sat down" (SSI: 15)*

The impact of 'carrier' or genetic issues, for some, was said to lead to additional stress, and a degree of guilt in relation to grown-up daughters' relationships. Genetic implications of DMD were reported, for example, when plans were made to marry.

*"My daughter was planning on getting married and we had to tell her and her boyfriend together. We had to explain the possibility she might be a carrier and we would understand if he changed his mind... these are the kinds of things you have to put up with" (SSI: 13)*

Further issues concerned mothers' carrier status, with a number of fathers commenting on mothers' guilt due to their carrier status:

*"Because it doesn't come from men, I think the women find that- well my partner says 'I've given you that beautiful boy you always wanted, look at how beautiful a little boy I've given you but he's damaged'. She feels it's all her fault, as she's given me this lovely little boy who is damaged". Because of the XY, XX chromosome problem, because it's her genes that's damaged" (SSI: 6)*

Acknowledging guilt and removing blame in relation to genetic issues, was necessary in order to move forward as a family in dealing with the condition:

*"At that time [wife] was into the business of that she was a carrier or it was a rogue gene or something. But she's not a carrier and it doesn't run in the family, so I think what we had to do very, very, quickly was say 'this is nobody's fault, so can you please just- see the business of blame, can you just take it away and [expletive]' because it's no part of this" (SSI: 4)*

Overall, reports oscillated between acceptance and despair at the situation, with a roughly even split between those who appeared to accept their circumstances and cope well, and those for whom an ongoing grieving process in terms of loss was apparent. The fact that there is no cure was difficult to accept:

*"Any parent will find it difficult especially knowing that there is no cure for DMD and over time he will lost all muscle strength. It is more difficult than a cancer diagnosis as cancer can be cured but with DMD there is no cure" (CS: 17)*

Additional emotional reactions described by fathers, included fear and concern. Many reported experiencing stress that interfered with quality of life for themselves and families to some degree. Some reported having time off work due to depression, and being prescribed medication to help cope with the situation. Often this stress was described as ongoing, without relief. A number of fathers referred to others who had not been able to cope, sometimes describing them as resenting the situation:

*"I've heard it time and time again...for some reason they resent the situation. They just can't take it and they've got to a stage where they've got to get away. They do a runner" (SSI: 13)*

A number had come to a point of extreme stress, being diagnosed with clinical depression and a few had experienced total breakdown. One father reported drinking more than he did before. They commented on how the condition had affected their mental health and outlook on life:

*"Difficult- have suffered two bouts of depression over the last 5 years- has affected work and general outlook on life" (CS: 19)*

*"We weren't focused, things didn't get paid.. stupid things like my partner got arrested for parking tickets, as we're always late coming back to the car. A lot of things that would have been tiny specs in the ocean, become massive mountains to get over" (SSI: 6)*

A variety of coping strategies were reported, with many becoming involved in gathering information and researching the condition. Echoing others, one father said he coped through information seeking, and dealing with the situation on a detached level:

*"A certain amount of disbelief and my ability to go out there to find out the information and deal with on an academic level rather than an emotional level" (SSI: 1)*

Others said that maintaining a work routine helped them to cope. A number of men said they found support in the workplace, whilst they did not talk about it in detail at home. Many felt they had coped well, despite difficulties for some in accepting the condition, and usually this became easier over time. Fathers described learning to live with the situation after initial shock and the need to move ahead, incorporating boys' needs into the daily routine, which became a normal part of life. They described how the family adjusted to the child's medical needs, until this became routine.

*"He is so much part of our daily lives we don't feel we are looking after a disabled child" (CS: 47)*

Adjustment involved altering expectations that were held prior to diagnosis, being realistic and accepting that no one was to blame, allowing fathers to adopt a positive attitude.

*"I actually got the advice from a colleague to say 'no one's to blame'. But when he said that it was freeing and being able to say 'it's nobody's fault'...that helped set it and I think the attitude is most important. If you get advice about attitude from the beginning it helps" (SSI: 4)*

Fathers who grew to accept the situation described attempts to focus on the positive and to give the child the best possible experiences in life. Some commented that they did not want to lose sight of their son, or normality, in light of diagnosis. Frequently mentioned was a desire for sons to experience life to the full. Furthermore, aspects such as discipline changed, with reports of becoming more lenient.

*"We responded by making a decision to give him the best experiences we could. This meant we enjoyed some good times and appreciated them-something we may not have done with a 'healthy' child" (CS: 11)*



*"I think your attitude does change, because things like discipline changes. Because we both work full time and we want to shower him with as much as we can. We try not to spoil him, although it's so, so difficult. But, allow him to do as many things as he would enjoy. I mean, almost immediately when he was diagnosed- he's really into cars.. so we said 'let's just book a stretch limo for him" (SSI:12)*

A number of fathers were involved in organisations that aimed to find a cure or fund raise for DMD. This was described as a proactive way of dealing with the situation, and many found positive action a distraction. Fathers often mentioned their hope that a cure would be found in their son's lifetime. Active involvement in fundraising and campaigning was referred to as both distraction and working for the cause, as illustrated below:

*"I don't know, I mean from talking to other people and my experience, I find it [DMD] puts a lot of pressure on families, especially marriages. I don't know, there's always a strong one in the family. I think strong is the wrong word. I do what I do with the charity, and I'm sure the reason I do what I do is to stop me thinking about anything else. When I'm not talking to you here, I'm emailing people, I'm doing research" (SSI: 9)*

In time, fathers described a need to deal with the deteriorating condition. An element of loss of control was reported due to lack of predictability, with an emphasis on the need to constantly adjust to new situations as they arose. The degenerative nature of DMD served as a constant reminder, with fathers reporting adjustment in light of this as an ongoing, or impossible, process.

*"It doesn't get any easier so you have to keep adjusting" (CS: 44)*

*"As the child's needs constantly change as the condition worsens, adjustment is not really possible" (CS: 47)*

The constant moving of milestones, in cases where the child's progression and therefore timing of death were not as predicted, made coping challenging.

*"Now we're getting kids coming into an older stage, hitting 30s. So your mindset is having to change now. There was a time I thought 'I'm going to have to prepare for X dying in the 20 mark, or before that" (SSI: 4)*

It was reported by some that they felt a constant barrage of events was ongoing, with little chance to adjust. One father described a phenomenon he termed 'issue fatigue'. This captures many of the views of fathers in relation to facing ongoing challenges:

*"Issue fatigue is more a sapping mental state that we and I believe others recognise as being simply a perpetual stream of things to deal with and to be addressed, little can be parked for later...the shifting sands of DMD" (CS: 18)*

Frequently expressed was a need to appear to be coping, whereby fathers concealed their distress, in contrast to how they actually felt. They felt both family and professionals assumed they would deal with things without needing support. Many talked about the expectation to be seen as strong and support others within the family.

*"On the face of it, we cope better but it's still very difficult and emotions are kept under the surface. Frustration, anger, pity, guilt- could I do more? Am I somehow responsible?" (CS: 6)*

*"The family think I will soldier on and be strong. Professionals have no idea that I have to work to get on with things to keep the status quo and be a provider" (CS: 23)*

Some fathers reported appearing to cope better on the surface, but found it difficult to keep emotions such as frustrations, anger, pity and guilt, hidden. One father described keeping his feelings hidden in an attempt to cope:

*"I am able to compartmentalise my feelings about my son's condition and cope in spite of them, this can be a logical coping mechanism" (CS: 56)*

Fathers commented on hiding their emotions from partners. They moved forward and tried to keep positive, but with an element of reluctance for some to share their worries. They also reported thinking a lot about things, but often keeping these thoughts to themselves.

**7.2. Theme 2: Support versus Isolation** (sub-themes: identity; strained friendships; family/marital stress; barriers to involvement)

Frequently, fathers described losing support from friendships and the impact of this in terms of identity. This continuous sub-theme of identity issues, both as a person and a father, appeared to underpin fathers' experiences of readjusting expectations in light of diagnosis, to reappraisals of their 'father/friend/partner role' and the need to adopt a protective attitude on behalf of the family. One father felt the diagnosis had changed his perception of others and his own character, whilst others commented on general loss of friendships:

*"I found that now I have got the whole world on my back, there's not many [friends] around any more. They dropped me like a brick... I've totally gone off the scene, like I said, I was a colourful character about me home town getting into a lot of music and entertainment because of the industry I work in.. and all that entourage. 'Sunshine friends' I call them.. well to be honest.. I'm a man, and I haven't got a 'soul bro'. I don't have that any more. That's what this condition has done- it's made me so protective of my family that outside people who I can't rely on, I've dropped them because they've done the same to me. They've dropped me as a friend and where they need me before I need them, I've just cut the chase and just says 'I'm not going down that road with you where if I turn round crying on your doorstep, I'm just going to make a fool and get the door slammed in my face (laughs). I'm not going to give you that opportunity" (SSI: 6)*

As a result of the strains associated with DMD a number of fathers described withdrawing from others, especially around the time of diagnosis. Others referred to people they knew of, who had reacted this way.

*"When I heard about [diagnosis], I just stopped going out. I stopped going to all sorts of things, with it being progressive I just stayed in and tried to get focused" (SSI: 11)*

*"It [the fight] just makes you wish you could hibernate in your own wee world. People do that, I know people who are not involved in PPUK and don't want anything to do with MD. Even the physio that comes to the house, and the OT, they say 'there's people out there who won't allow us through the door, they just won't accept it. Just won't accept the diagnosis'. I think this is early on. I'm not saying they're wrong, but there are people who want*

*nothing to do with services, who are maybe in denial or whatever. People like that need counselling. People like that need help. There are 2 or 3 I know who have shut the door- they're just trying to pretend it's not going to happen" (SSI: 12)*

This sense of isolation linked to a lack of opportunity for fathers to seek support and talk about issues affecting them. For some, resulting loss of self-confidence, and feeling depressed, was reported to have an impact on socialising.

*"You lose a lot of self confidence, at times you feel 'down', but still have to work and your social life is severely impacted" (CS: 53)*

Few had been offered psychological interventions, however, often reported their partner being offered this type of help. A small number had been prescribed antidepressants and counselling:

*"Prescription drugs have helped heal paper over wounds I suppose" (CS: 18)*

*"At the time I guess it [the diagnosis] took the bottom out of my world. It's difficult now, because that was a long time ago and for the most part I've managed to deal with a lot of issues that have come up. But it's not been easy and at times has involved therapy for me certainly, and in dealing with it in other ways as well. But yeah, it's been hard to deal with. I had a couple of episodes where I had to go long term sick from work. The second time, I had to spend some time in psychotherapy just to deal with it" (SSI: 1)*

For many, the main support system was their partner followed by immediate family. Since diagnosis, a number commented that they had become very protective of their family, sometimes having a negative effect on friendships. In some cases 'dropping' people before they expected to be ostracised by friends was described. With friends, most fathers did not generally talk about their son's condition, and their social networks generally appeared not to encourage this.

*"Fathers tend not to interact or seek out other fathers. There doesn't seem a need to interact with other DMD dads. You can do it via the internet" (CS:13)*

A number felt a need to prove they could deal with things themselves, and reluctance for others to discuss DMD. Some fathers said they found it difficult to talk to others about the condition. Other responses ranged from being direct, to avoiding talking about it. A number of fathers said that they found it easier to talk about away from home, and that talking to work colleagues helped.

*"We never talk about it [at home] really, other than symptoms and treatment" (SSI: 9)*

*"I spoke to a lot of people at work about it and that seemed to help a lot" (SSI: 7)*

One father described a general avoidance- on a par with that of bereavement, on the part of friends to discuss the condition, and the difference between his friends' reactions compared to his wife's friends:

*"It tends to revolve around mothers. I mean friends, the first thing they said when they found out is 'how is [wife] taking it?', no-one ever says 'how's [participant's name] taking it?' (laughs). They tend not to talk about it at all- my friends don't. I mean, the girls do, [wife] and her friends. I suppose girls are more open and used to discussing things. But nobody speaks about it, nobody mentions it. I certainly don't. I just don't think it's ever mentioned. None of my friends ever mention it to me. It's ok with me. I suppose at the beginning, I wouldn't have minded if people had come up and said 'oh, I'm sorry, is there anything we can do?'. But people seem to ignore it. I suppose it's a bit like somebody dies in the family and you just don't mention it" (SSI: 9)*

There was a general perception that the condition and associated stress would highlight any problems that were already present in a relationship. For some, since diagnosis it was felt that the relationship was in the background.

*"It's totally spazzed my partner and my relationship. We were a happy, sexy couple, progressive, avant garde. Now we know we are in for a term and it's spoilt a really good thing. It's- the light's gone off. All the plans and expectations we had for a great education, passing on all this kind of colourful, cultural kind of like, experiences onto them. It's not gone, but we were just a really good family. Now it's put distance between us. We haven't got time for partnership while we're living under this thing. We're waiting for a bomb to explode" (SSI: 6)*

In terms of marital relationships, participants generally reported positive, supportive relationships, however, the impact on families of stress resulting from DMD was apparent throughout participants' accounts:

*"Just about everybody I know has come close- including myself, to splitting. It's usually a feeling that one partner is taking it better than the other. One partner feels that the other is not pushing their corner or fighting their corner. I don't think it goes with either sex [anecdote about Rangers player and wife].. I think if there's any weakness in the relationship, it brings it right to a head. A lot of relationships will have weaknesses anyway, and this just piles on top of it. Maybe it's just giving people an excuse to do a runner, I don't know. I know a lot of families, two guys I know who just walked out of the family. They just couldn't take it- just let the wife deal with it" (SSI: 9)*

Although fathers shared mothers' concerns, their responses and coping strategies differed in some ways. A number of fathers talked about differences in coping within the context of gender, and many knew of families who had split as a result:

*"[anecdote about fathers who left families] The whole thing got on top of the father and to cut a long story short, he had a heart attack and died. The other father..he just got up and walked and she hasn't seen him from that day onwards. He just walked away from it. Because of the diagnosis, he couldn't hack it. He couldn't take it and walked. I don't really know, I've heard it time and time again, not just with husbands but with wives aswell. For some reason they resent the situation. They just can't take it, can't face up to it and they've got to a stage where they've got to get away. They do a runner" (SSI:13)*

Frequently fathers reported the number of families they knew where the father had left after diagnosis, and often felt this was due to challenges in dealing with wider issues surrounding DMD.

*"Through PPUK I've been in contact with a lot of people, and it's amazing the number of single female parents that are left with the boy. I don't know why that is" (SSI:12)*

*"That's why families break up. It's not what they've got, it's the strain of the fight" (SSI: 9)*

Differences in dealing with the child, where one parent had not accepted the prognosis also led to difficulties. Often one partner would want to talk about DMD, resulting in conflict when their partner avoided, or discouraged this:

*"She won't tell him the truth and I will...he came back and said 'am I going to die young?'. His mam just went (mimics running), she bolted...and I went 'everybody is going to die" (SSI: 6)*

*"Another thing I'd better tell you as well, because it's part of it, my wife is...she bottles things up. She would hate me to go and talk to other people about it. If one wants to let it out and the other wants to bottle it up, then you've got a bit of a mix up" (SSI: 13)*

*"No-one has ever asked 'how do you feel about having this?' like, it's like having a ball round your neck but you've still got to go on. If you don't go on, then the whole fabric of family life y' know.. but sometimes I get migraines and things like that. Eh, She'll not talk about it. She'll not go to meetings... I try to talk to other people. I mean it's not often, and probably I'm not open enough in that respect myself maybe" (SSI: 3)*

In some cases this led to problems within personal relationships. In order to deal with such issues, some men described attempting to get on with normal life, and found work to be a means of doing this:

*"I think I probably behaved with more autism than my wife has. When [son] was diagnosed it was a Thursday and I was back at work on Monday. [wife] was off work the whole week. [wife] actually felt I was going back very quickly, and I was 'no, no'. I think it was just different ways of coping" (SSI: 4)*

*"She took antidepressants, I just don't talk about it. I just went to work" (SSI: 5)*

Although challenges were often reported, for many others there was no major difference in coping.

*"I think we coped pretty much the same, although I must admit I am sometimes more negative. You have to go over all these hurdles" (SSI: 8)*

Within the context of partnership/family adjustment, fathers described their role in terms of gender, often being involved in physical and practical areas, with mothers generally being involved more in emotional and personal care:

*"We take on different roles so the family can function. She does more than me, dressing, washing etc. I do other chores, in particular care of the wheelchair, medical equipment etc" (CS: 34)*

*"I take more of a supporting role to my wife. I'm at work all day, my wife has more contact with our son" (CS: 26)*

At times, this led to frustration and further feelings of being isolated from their son's life and routine. Although the majority described being involved with their son, an issue for some was a sense of detachment from certain aspects of the child's life. This led to a sense of frustration and isolation from the child's routine and decision making, resulted in feeling left out. Often, this isolation was due to practicalities such as work commitments:

*"It seems to be that all meetings, decisions or whatever are made during the day time which is obviously when I'm working. I come home and everything is set in stone. 'Here's what time the appointment is going to be. Here's what wheelchair he's getting, here's what sling he's getting. Here's the plan of what we're going to do for the house'. So, it's like 'right, ok'.. it's like my voice doesn't really count" (SSI: 11)*

This perception of being removed from close involvement in the child's routine was distressing for some. One father said he found it upsetting that he felt somehow distanced from his teenage son, describing this as an attempt not to become too attached before losing him:

*"Well, I find it strange. I sometimes find myself trying to stay remote from my son, because I don't want to get too attached. You know if you get close to somebody, and then something happens you feel worse. I know it's a strange thing, you know that you're frightened of. He's very close to his mum and he talks to his mum more than he talks to me. I mean [wife] does most of his personal care although I'll be around... so he tends to sit in the bath and talk to her. You know, they've always been very close since he was a baby. That's not to say that I'm not close but that...but I sometimes find myself, I don't know, trying to be slightly aloof so I don't get too close. I don't know, maybe.. it's not that I don't love my son. I love him very much. I can actually feel it happening at times, and I have to overcome it and try to do what's best. I don't know how to explain it, but it's a strange feeling. I find that upsetting, because I feel 'why can't I get closer?'" (SSI: 9)*



Due to working in the day, fathers tended to be involved in physiotherapy and physical routines at night. They reported mothers dealing with different elements of care, resulting for some in the son becoming closer emotionally to the mother as a result of her more intense involvement in personal care:

*"As I go to work, I don't see our son as much as my wife. Therefore our son is much closer to his mam" (CS: 3)*

Having to hold down employment was often described as challenging in light of dealing and bonding with sons, and being able to spend less time with him:

*"Due to work, I put in less one to one time with my son. Whilst my son and I love each other dearly, our relationship will never come anywhere near the bond between my wife and son" (CS: 31)*

Some said they felt bombarded with information when they returned from work:

*"I come back from work and it's all waiting for me and I have been out all day" (SSI: 5)*

*"I work full time and my wife [name] doesn't. She seems to spend most of her time caring for the boys, organising appointments, ringing hospitals, with local social services trying to get wheelchair appointments or whatever, so she doesn't work. I work, and I tend to come home and be hit with all the day's events in one go" (SSI: 11)*

Most fathers commented that being involved was important, but also said they felt there were barriers to becoming more involved:

*"If I had the choice, I would spend less time working and more time enjoying our son's life" (CS: 50)*

*"I start work at 6. I could be home at 5,6,or 7. It varies but she's the one here with him all the time. Really I'm only with him at weekends" (SSI: 15)*

A sub-theme of exclusion underpinned this theme. Fathers tried to be involved but did not do enough; although they were willing, they were unable, due to work commitments.

**7.3. Theme 3: The fight for resources** (sub-themes: frustration; spare part/exclusion; needs and solutions)

There was a roughly even split between those who said they were satisfied and unhappy with social and general medical provision. Often, it was felt that support was patchy, due to DMD being relatively uncommon within general practice. Generally, a high level of frustration was reported in relation to experiences with services.

*“They’re very slow and they get it more often wrong. With the best will in the world, they get it more wrong than they do right” (SSI: 8)*

*“Before we got in touch with PPUK [DMD charity], we were told ‘take him home there’s nothing anybody can do. Now we know it’s not true” (CS: 14)*

As most doctors only see a couple of DMD cases in a lifetime, this was a frequent problem, whereby fathers felt they were teaching the professional and facing frustration at having to do this with new staff. Some also commented that there was little interest in DMD, as a ‘niche’ condition. Frustration at professionals not appearing to understand the specifics of DMD was frequently referred to:

*“What we’ve had to do often, is to educate people we’re talking to- the medical professionals we’re talking to, about the condition. We’ve had to educate them” (SSI: 1)*

*“The staff had little to no knowledge of DMD and therefore didn’t understand my son’s needs” (CS: 16)*

A number felt people generally did not understand the nature of DMD, and found this testing, whilst others reported they felt they were being treated differently because of the child’s condition.

*“It’s also having to tell people about DMD. They don’t get it. They don’t know about Duchennes so you’ve got to keep telling them” (SSI: 5)*

*“Professionals sometimes treat the parents of disabled children as lesser people” (CS: 33)*

When talking about services, the 'fight' or 'battle' was often referred to. It was often stated that nothing came easily, but needed to be pushed for. Fathers repeatedly reported fighting for their sons, often as part of their 'duty' to ensure they were receiving the best care possible.

*"We're not aggressive people, and we're not argumentative but how many people can get through all those hoops. I mean, everybody will give up. I've said to people before at the MD. There was one boy who had difficulty coughing and we said 'can he not get a cough machine'? The lady said 'look [name], I've fought for years and years and years and I just don't have any fight left'. But I said 'you could get one of those from the internet. You can order that yourself. But I forget, some people cannot afford £100 to buy a cough machine but because we're both working we're fortunate. But it does take the fight out of you, and it makes you absolutely exhausted" (SSI: 12)*

*"There's so many things that you have got to look at that are needed and it's difficult when it's first diagnosed. It's difficult for a father to actually come to terms with that and say 'I need to get this'. It's when you need help and it's not always there and you have to fight. It's a fight and that, the fights that I've had with people and social work places like that. I'll go in there and because [wife] will let them off with it whereas I won't. I'll go in there and fight my corner, and make sure that.. and I think that's important for fathers to do" (SSI: 3)*

Constant chasing and delays reportedly led to feelings of lack of control. Many fathers said they felt let down by social services, and talked about experiencing numerous delays with medical equipment and the constant need to pursue providers.

*"It would be nice for once for somebody in a professional position to act on what they are told in the first instance...the constant following up of say, planning permission, just eventually tires out the already tired carer" (CS: 23)*

One father commented that he was surprised when things went unexpectedly smoothly:

*"All the years of asking for everything and this girl came over. She came up to the house to see US! She says 'no problem, I'll see what I can do'. It was totally different from what we're used to" (SSI: 5)*

Many talked about feeling dismissed in relation to attitudes from others, whilst the focus was placed on mothers.

*"You feel envious of other people. They don't understand. No-one seems to understand the father, it's always the mother. How the father really hurts. It hurts, it hurts that your son is probably going to be away before you. That hurts. No-one has ever asked 'how do you feel about having this' like, it's like having a ball round your neck but you've still got to go on. If you don't go on, then the whole fabric of family life y' know. but sometimes I get migraine headaches and things like that" (SSI:3)*

*"It tends to revolve around mothers. I mean friends, the first thing they said when they found out is 'how is [wife] taking it'. No one ever says 'how are you taking it?'" (SSI: 7)*

Some reported that they felt left out in relation to dealing with professionals, with focus placed on their partner, often due to work commitments:

*"They tend to focus on treatments and supporting the wife" (CS: 13)*

*"The problem is not having the time through work commitments to meet the professionals" (CS: 44)*

This perceived 'neglect' from professionals often started around the time of diagnosis. In some cases talking about their own needs was seen as irrelevant in contrast to their son's issues.

*"My needs are not relevant compared with that of my sons. It's hard to discuss my needs when I can get up and walk across a room. He can't" (CS: 34)*

A number said they felt professionals viewed them negatively or as a 'spare part' at appointments and that their role was questioned:

*"Fathers have an equal role to play in child health. When I sometimes take my son for a hospital appointment by myself, I feel health professionals are querying why father is attending and not the mother. I feel it should not matter who is attending or involved" (CS: 17)*

Others, however, were satisfied with support from health services and stated that their needs had been met:

*"We have received excellent support and advice from health professionals"  
(CS: 16)*

*"it's not been plain sailing but anything we've needed we've got it without a great deal of hassle" (SSI: 7)*

The importance of having a good relationship with professionals was reported by a number of fathers:

*"Because see if you get on with them [service providers], and you're a reasonable person, like not wanting to.. and I don't mean not that you don't want the best for your kid, it's better that you negotiate with them, and have a relationship with them rather than going to war. Then, if you see your child having a disability as something that you must constantly fight for.. then you're in with the attitude that you're waiting for things to go wrong and I don't know what kind of a message that gives" (SSI: 4)*

*"If any parent involves themselves, professionals tend to welcome that. You have to be approachable in order for the relationship to work" (CS: 38)*

Since the diagnosis, the parallel process of being assessed for disability benefits and adaptations led to a feeling of being invaded/humiliated for some:

*"Now [son] is in his new bedroom and that took 4 years of fighting to get the extension on the garage. We went through hell because it was means tested. You were treated like a piece of manure we felt. It was horrible. That took ages, you felt violated, because everything in your personal life is gone into" (SSI: 15)*

*"I went down to the DHSS or whatever it is and said 'I have a disabled son, what can I have?'. They went 'you have to tell me'. I said 'I don't know, I've never been in the system'. I left home at 15, joined the army. I've never claimed a penny, any benefit in my life. So I had no idea...I never claimed a penny, any benefit in my life....you feel like you're begging. You really feel like you are begging and you're not. That's why they need a co-ordinating centre. Once you're diagnosed, you can go to the centre with everything and people saying what you're entitled to. I think that's what puts the biggest strain on. That's why families break up. It's not what they've got, it's the strain of the fight" (SSI: 9)*

There was a roughly equal division between those who believed their needs had been met or not, by professionals. In terms of suggestions for improvements and key stages necessitating support, fathers preferred professionals to be honest, and

clear about what they could achieve. In this context, the need for others to acknowledge the time limitations of their sons' life-span was important.

*"Listen to parents. Not everything might be done that we want done...but say 'sorry, we can't do this, but we can do this" (SSI: 15)*

*"It's alright for them saying 'we can get that in 6 months', but 6 months is a long time in a boys.. we have to have it now" (SSI: 3)*

Some reported that they would have liked support from professionals in relation to emotional issues, especially at early stages. This was often described in the context of the isolation felt by working fathers.

*"Early stages: emotional support, coming to terms, being honest with child" (CS: 38)*

*"Emotional help- the mental strains are difficult to deal with without help from professionals" (CS: 7)*

*"More acceptance and awareness of the isolation and alienation that the working/ home carer father is faced with" (CS: 2)*

Dealing with re-evaluating their own expectations and knowing how to move forward, was also mentioned in terms of support needs:

*"The expectations- there will be things that come and go. Then what you need is advice as he gets older on what's the best way to look at things" (SSI: 4)*

Specific times where fathers felt extra support was required were diagnosis, times of change and coping with associated feelings of helplessness/loss of expectations:

*"I would like to have seen more emotional support when changes happen and a friendly face when things are hard" (CS: 27)*

*"As the main carer my partner gets focus from professionals. I have to assert my presence and ask lots of questions to feel engaged!" (CS: 50)*

A key factor included wanting to know what they would be able to do with their sons, instead of only limitations associated with DMD.

*"Fathers want to provide solutions, get things done. Fathers need to know what they will be able to do with the son, not just left to think on what he will never do" (CS: 26)*

Further suggestions for provision of better support included: opportunities for confidential one to one discussions, knowing they were not the only father of a DMD son, and the need to know there was hope.

*"We feel from day 1, our Consultant has been very negative. Well, I think at diagnosis we obviously had said 'is there no cure?', and his words to us were 'well, 20 years ago they discovered the DMD genes and they said there'd be a cure round the corner. That was 20 years ago, and they've never found anything. So, I can't see them finding anything in the next 20 years'. That devastated us, I was devastated. We are realists, I mean, we know what's ahead of us....we know there's not going to be a miracle cure...you just live in hope that if [son] has another 10-15 years, that maybe something will come along. Maybe allow him to live a bit longer. But, we know that and we just feel this consultant is so negative" (SSI: 12)*

In addition, many referred to the strain on relationships, and how some kind of help would benefit this impact:

*"I feel help for fathers would greatly reduce the amount of marriage break-ups. If I had understood what I was going through it would have helped. I am fortunate enough to have a strong marriage" (CS: 31)*

One father, however, stated that he would not respond positively to counselling:

*"The last thing I'd want is some counsellor whose job it is to make eye contact, and telling me she knows how I feel" (CS: 34)*

Written information, aimed at fathers was also suggested. Fathers commented that this would result from the issues raised by participants in this research:

*"If it's there for future dads to read and say 'actually these are the issues about fathers' then someone would think, right ok I can agree with that, I can see where they're coming from now... and actually I don't feel so bad for feeling angry, annoyed" (SSI: 4)*

Others reported a need to know how to practically care for, and talk to, their sons about DMD.

*“Basic support with regard to caring for a disabled child i.e. respite care, help in the home, help at school, support for siblings, a listening ear, access to counselling” (CS: 47)*

*“Talking to your son, and understanding him and his condition” (CS: 35)*

For older boys, fathers felt support needs included the option of someone to talk to independently; a need to address boys' frustration at being physically restricted, and somewhere appropriate for boys to mix socially. Fathers of older boys also reported that it proved hard to seek guidance:

*“Our son is 33, consultants say he is re-writing the text books. We are guiding pathfinders so it's hard to get help” (CS: 1)*

Discussing needs with employers, and a need for flexibility, was a further issue that was repeatedly mentioned. Male only support groups or practical seminars where practical issues would be discussed out-with the family, with opportunity for emotional support if required, were suggested as a means of meeting needs:

*“Women tend to share experiences more with other mums. Men go and do sport etc to forget! Support groups could be for dads only?” (CS: 20)*

*“Support groups to discuss issues with fathers.. little is known or understood about DMD. This causes stress due to continual explanation” (CS: 33)*

*“There should be something there for fathers as well, because like I said I've given up my career” (SSI: 6)*

One man described his disappointment at finding that his local support network consisted of families talking, with out any professional support:

*“We both thought it [family support group], would be somewhere you went where people would sit and speak to you, and give you counselling...give you counselling and ask you how you are coping with this and maybe ‘this is what you should do’...I was all for someone sitting analysing me! [laughs]” (SSI: 12)*

Although good practice was also reported, a more integrated support system of professional services was an overriding theme. A majority of fathers felt services



would benefit from a more cohesive system, which would remove the stress of contacting a range of organisations:

*"A more integrated system of professional support that works within reasonable time scales. A team that works hand in hand to support the family rather than a collection of individuals pulling in different directions" (CS: 2)*

*"Specialist appointments ALL ask the same questions in triplicate at least- it smacks of inefficiency, wastes time, achieves little" (CS: 18)*

Knowing what to expect was a further important issue, and it was felt they would benefit from additional guidance. Provision of a schedule of needs/contacts, corresponding to each stage, was also felt to be beneficial:

*"I think in the early stages around the 7-8 year mark it would have been far better for us as a family to get organised if we knew from other families what they needed, what the boys needed going into sort or early teens.. so all those kinds of things, more information and somebody to say 'look, this is what's going to happen, this is what you're going to need'. That's the kind of information you really need as there is a lot of stuff at the time" (SSI:7)*

One father described a situation where he felt best practice had been achieved:

*"All the sort of professionals came together, rather than going off to different professionals all the time. It's an excellent way of doing it" (SSI: 8)*

Overall, however, an often-chaotic picture of services was reported which led to frustration and increased stress. Many felt more co-ordinated help in areas such as dealing with social services, physical therapies and welfare benefits, was needed. The interactions of professionals with parents had a significant impact on fathers. In general, it was felt that more awareness was required on behalf of professionals in relation to communicating with DMD families.

*"No one in the health service has asked how I am coping since my son was diagnosed 4 years ago. I feel really disappointed in a lack of support from family and friends" (CS: 45)*

Communication problems with professionals were also reported, with some fathers being unclear about what they were told by doctors. Many felt they still did not

properly understand the child's condition, often because they did not fully understand what professionals told them. They believed support was needed in this respect.

*"The doctors tell you so much, but they tell you in their language. You look up your books and get a wee bit more and you understand better" (SSI: 7)*

*"It would be nice to speak to someone to explain exactly what [son's] illness is. Split-up fathers are left to guess what mothers are told. Fathers (not living with son) get no help, that's a fact" (CS: 48)*

Also reported was a requirement for training professionals, and the perception this should be encouraged and developed as a career path for young professionals:

*"Having workshops for the GPs to make sure that especially it would be the ones who had someone in their practice. That then could go forward to other things" (SSI: 13)*

*"I think the money should be there for young doctors, make it a high prospect job to get this thing sorted out. We're dealing with DNA here, everyone loves DNA" (SSI: 6)*

One father felt a national standard for healthcare/ social care professionals dealing with DMD was required. Again, isolation of the father and a feeling of being avoided or not listened to by family and professionals were reported. These were additional areas where fathers thought awareness could be raised and changes made:

*"Speaking to parents as a whole and not 'avoiding' fathers by speaking through them at appointments" (CS: 19)*

*"Until these questions, my thoughts as a father have never been asked. I presume had I shouted someone would have listened" (CS: 31)*

In meeting the needs of fathers, acceptance, awareness of the isolation and alienation some working/ carer fathers faced, were key issues reported as important.

Table 13 below summarises a number of key challenges described by fathers, illustrating needs and suggestions for support

**Table 13:** *Key challenges: needs and fathers' suggestions for support*

Key challenges	Fathers' suggestions for addressing needs/good practice
<b>Early stage of diagnosis</b>	<ul style="list-style-type: none"> <li>• Emotional support and confidential discussions one to one</li> <li>• At the early stages of diagnosis, help with fathers' perceived inability to help their sons</li> </ul>
<b>Acknowledging fathers perceptions of being excluded and encouraging involvement</b> <b>Social activities and support for older sons</b>	<ul style="list-style-type: none"> <li>• Father only support groups</li> <li>• Provide an element of hope</li> <li>• Key person to support and explain what will happen</li> <li>• Speak to parents as a whole and don't 'avoid' fathers by speaking through them at appointments</li> <li>• Appointments outside of 9-5pm hours</li> <li>• Improved access to respite care</li> <li>• Provision of clubs where boys can mix with other boys with DMD and those without any health problems</li> <li>• For older boys, assistance in finding out about the level of support/financial assistance from authorities</li> </ul>
<b>Integrated system and professional training</b>	<ul style="list-style-type: none"> <li>• Address lack of social provision for boys</li> <li>• Suitable organisations where boys can go and mix with other people their own, with physical rather than mental disabilities.</li> <li>• Reduce the amount of chasing people up</li> <li>• Reduce the need to 'fight' for services</li> <li>• A schedule that outlines needs at each stage</li> <li>• Streamline and review processes, to remove stress and facilitate preparation for when deterioration begins</li> <li>• More information about processes, planning for mid to long term future</li> <li>• Encouragement of young doctors into DMD related fields, to develop understanding and expertise</li> <li>• Workshops for the GPs and improved training of professionals</li> <li>• Co-ordinated care packages, to promote greater awareness across multidisciplinary teams</li> </ul>

**7.4. Theme 4: Race against time (deterioration and death)** (sub-themes: images of next stages; transition to adulthood- comparison; talking about death; decisions)

Some fathers described the challenge of DMD confronting their previous concept of an ongoing family line. The following quotes illustrate this expectation for continuation of the family:

*"I think fathers in general see family differently- certainly I see family as a form of immortality if you like. This is my son, he's going to continue after I'm gone. That's the first thing you have to accept, and that's a difficult emotional hurdle to cross" (SSI: 1)*

*"My family name runs out with [son], that's it. I've got no brothers or sisters. Like I say, my family line ends with [son]. That was another thing, one of the things I said when I first heard about it. I says, one of my first questions was 'will he ever be able to have kids?' and they went 'no, no chance'. Now I'm down the line, I find that there's prostitutes in Amsterdam and he can go and have sex if he chooses to, there's different ways he can have kids. It's a possibility" (SSI: 6)*

These fathers reported feeling sadness that their family name would not be continued. The limited life-span of their son was an underlying theme throughout, with fathers conveying a strong sense of urgency. This focus on time limitations included obtaining best medical treatment and ensuring the child had lived as full and rich a life as possible. The need for speed also related to delays with medical procedures, especially when the son's condition was declining:

*"They're dying all the time and we could do something about it and it wouldn't take a lot of money" (SSI: 12)*

*"It took us nearly a year to get an appointment. In that whole year his spinal curvature had increased dramatically. He was on the verge of not getting (the operation)" (SSI: 7)*

One father said he believed he had 'a lend of his child', and also described the challenge of not knowing for sure the life expectancy:

*"You have a lend of this child, who is going to go fairly quickly. At the time they were saying to you 16, 17, 18, if you get beyond 21 you're doing well."*

*But 10 years later now we're getting kids in their thirties. So, your mindset is having to change now" (SSI: 4)*

A number of fathers were angered that others did not appear to share their sense of urgency. One felt campaigns were holding back for reasons such as fear of including boys in trials, and that this delayed progress. In relation to treatment, others felt that researchers were being too cautious and thereby time was running out for a cure within their son's lifetime:

*"It's just not fast enough for me. I need something much more positive" (SSI: 12)*

*"Researchers have to take a risk- instead of years of mice trials let's get them into the clinic into treatments" (CS: 5)*

*"They can cure every mouse in the world but get it out of mice and into the boys. We're just forming a group of guys we call the young men with DMD forum. We've organised their own group so they can go and fight their own battles. They have their own lobby for parliament. I know a guy who is 20. He said 'just get it and inject into me, just do it', do it now, what have I got to lose?" (SSI: 9)*

This desire for speed also involved exposing the child to life experiences and often appearing to 'cram in' as many of these as possible, before time ran out:

*"I'd like him to see some of the world before anything happens you know" (CS: 50)*

*"I get them up at 4am and take them to the airport and don't even tell them. I let them try and guess where they're going. It's like 'Disneyland'. It's just that kind of thing, special little things like that" (SSI: 6)*

Generally, fathers wanted to make life as good as it could be, whilst making every day count. Again, the sense of urgency was felt here. Fathers strove to ensure their son had as many positive experiences as possible:

*"We're showering him with as many things as we can. Taking him on as many holidays as possible" (SSI: 12)*

*"Make every day count...you have to count because time is so short that you probably might even sometimes regret not being there for them. OK you're seeing them grow up and stuff like that but there's so many things that you*

*have got to look at that are needed and it's difficult when it's first diagnosed. It's difficult for a father to actually come to terms with that and say 'I need to get this'. It's when you need help and it's not always there and you have to fight. It's a fight and that, the fights that I've had with people and social work places like that. I'll go in there and because [wife] will let them off with it whereas I won't. I'll go in there and fight my corner, and make sure that..and I think that's important for fathers to do" (SSI: 3)*

*"It's made me realise the importance of life and what my role is. Our house motto is 'no regrets' and this keeps me motivated. The most challenging times have yet to come" (CS: 27)*

In relation to accepting their son would die before them, a reported fear was that of seeing the child in later stages of decline. Many were scared that their son would be rejected when he began to deteriorate. It was also upsetting for some to be reminded of future stages:

*"We just don't want to see him deteriorate too much. I think we would be happy if he could have a 21<sup>st</sup> birthday party" (SSI: 5)*

*"You just live in hope that if X has another 10-15 years, that maybe something will come along. Maybe allow him to live a bit longer" (SSI: 12)*

*"I think when people will see X deteriorate, they probably won't touch him because they think they will catch it" (SSI: 6)*

The progressive nature of DMD was generally described in stages:

*"You just get to a point where you think, life's settling down a bit and then you seem to enter the next stage" (SSI: 11)*

*"You do notice it's a degenerative disease. When you go to hospital you know it's not going to be good news" (SSI: 3)*

For some, it was difficult to accept each stage often due to feeling unprepared for sudden change, as illustrated below:

*"There will be long periods of very little change and then all of a sudden there will be a very dramatic change" (SSI: 10)*

*"Coping with each stage of deterioration is difficult e.g. can no longer walk, cannot feed himself" (CS: 16)*

One of the most challenging milestones was when the child stopped walking, and started wheelchair use. As there was some variation in timing for each child, some found it difficult not being able to confirm this:

*"The worst time was when he stopped walking completely" (CS: 35)*

*"Now [son] is using a wheelchair it makes it more obvious. It is depressing" (CS: 42)*

*"The lack of being able to say to your son 'well, by this time it will be like this'. Because for different children, it's different times" (SSI: 9)*

The ongoing deterioration resulted in a continual process of physical and emotional parental stress:

*"Due to the nature of the condition, I believe that there is no let up in the 'most challenging point or time'- it remains continuously 'the most challenging time' as the disease progressively steals your child's physical abilities and you have to do more for them" (CS: 55)*

The move from childhood to adulthood was also reported as a key challenge, both due to deterioration of the condition and also in relation to gaps in services.

*"The hardest period was when at 16 the hospital could not see [medic] anymore, but gave no indication as to where to go for advice" (CS: 43)*

As their son's condition declined, watching other children grow up was often described as being difficult. This was especially the case where the child was compared to healthy siblings:

*"Watching one grow up and mature whilst the other (physically) moves in the other direction" (CS: 22)*

*"Watching the agonising deterioration since I've been 35, whilst two younger brothers grow up past him- truly sad" (CS: 18)*

Related to the progressive nature of the condition and transition to adulthood, a sub-theme included decision making. This was in light of deterioration, in terms of who led decisions involving treatment, and the actual process of decision making. In relation to treatment, rapid decisions were often required, in the face of time

restrictions on the child's life. Joint decision making with the child in relation to operations such as spinal fusion,<sup>45</sup> achillies tendon release<sup>46</sup> was important for fathers. Fathers described the process of decision making as involving information gathering, talking to parents and boys:

*"I could not make up my mind to say yes. Then suddenly one day a little voice came from the room and said 'oi dad, that operation, I want it'. I breathed a sigh of relief and said 'great, we're doing it" (SSI: 13)*

*"You take a note of everything, but listen as closely as you can to what certain people are telling you. We also spoke to parents of older boys, who'd been through it. Also those who haven't gone through it. All of this is rattling round in your head, but you have to.. We took time in a quiet room with [son] and he was emotional, he was crying. He's quite a young boy for fifteen, he's very academic, but he's quite a young boy for fifteen. At the end of the day, we were probably trying to guide him towards going for it. But you sense when it's not his want or wish for it, that you can't force anyone into that position. And that's how we came to that decision. So you're weighing up all the information. It's all milling about. You're jumping one way, you're jumping the other. There's no cast iron process you go through that you get the right decision" (SSI: 8)*

Making treatment decisions was often described as challenging, as there were many factors to consider including child's quality of life. This was especially the case where conflicting advice was given:

*"Having to decide yes, no, whatever, that was the hardest time" (SSI: 8)*

*"It's stressful in case it's wrong, but you've got to make the decisions" (SSI: 5)*

*"Medicine is famous for that, so why should MD be any different? That's what you have to cope with, so our dilemma now is what happens, as his posture is very good. Do you put him through an operation on this advice that's running against advice you receive? That's our dilemma" (SSI: 4)*

Again, weighting up pros and cons was not easy. Some fathers also talked about finding it hard to put their son through various operations, when the outcome may

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<sup>45</sup> To prevent scoliosis of the spine.

<sup>46</sup> Operation necessary prior to wearing callipers (used to prolong walking as muscles weaken). Each operation is at the discretion of child and family.



not be worth the ongoing pain. One father worried that if his son had a spinal operation and a cure was found, his son would not benefit.

*“Getting the spinal fusion done meant that if they miraculously come up with a cure tomorrow, he still wouldn’t be able to walk again” (SSI: 7)*

Another father, whose older son had died from DMD, recalled the decline in condition and did not want his living son to go through the same:

*“That was a decision which was very, very difficult to make. Well, it wasn’t really difficult to make because when I saw (dead son) and scoliosis had that rib making an indentation, that was... terrible” (SSI: 3)*

The final sub theme concerned death related issues. A number of fathers reported finding it difficult to talk about death with the child, and sometimes expressed relief that this was avoided or dealt with by the mother:

*“He knows he’s going to die. He will ask questions about that, he’s not afraid to. Fortunately for me it’s his mother he asks more than me” (SSI: 4)*

Some fathers avoided dealing with the issue, and worried about how to handle this. It was also difficult wondering how much the child already knew, and fearing having to face something the father did not feel equipped or ready to discuss:

*“The other problem I avoid basically is.. dying. I just wouldn’t know what to say. I’d be like ‘uh-oh, it’s that time [laughs]’. If he asks me directly, that’s ok. I don’t know what to say. I worry about that” (SSI:5)*

A number reported dealing with child’s death related queries directly. In these cases, the importance of being honest, and dealing directly with questions, was emphasised:

*“The only way to do it is to be honest. So, if you’re asked a difficult question and have to give a difficult answer then give it” (SSI: 4)*

*“We don’t hold any punches, we’ll tell him everything” (SSI: 7)*

Others derived comfort from knowing their son would die before them, but found it hard on occasion in relation to care needs, when they thought the son might outlive them:

*"He said 'am I going to die young?...'I went 'everybody is going to die...anyone might die tomorrow or be here 100 years'" (SSI: 6)*

*"There is some comfort from the fact he will die before you.. so when that seems to be turning round a bit you've got to say 'theoretically now age wise I'm going to go first' but who is going to be there for him?" (SSI: 4)*

For some, there were issues in knowing how and when to tell their son about the prognosis. Often, this was led by the child initiating the discussion.

*"He asks me every now and again why. Because we haven't told...well, how do you tell a nine year old?" (SSI: 15)*

*"He talks more to his mother. But I say 'if there's anything just let me know' and he'll tell me. He hasn't really talked to us about the big things" (SSI: 8)*

Some felt the best way was to leave the child to discover things at his own pace.

One father believed it was a good sign that his son had not asked questions.

Another left books around so that his son could find out himself, but found this difficult.

*"He doesn't really talk about it and he's never really asked any questions. So that's a good sign I think" (SSI: 11)*

*"What we used to do was leave books lying around. If he wants to talk about it, we're here. We left him to look into it himself, so he could discover what went on, at his own pace. I think that's one of the hardest things, to let them find out. How to sort of break it to them. But, we haven't actually done that, [son] does give hints. Unfortunately, [son] lost 3 of his friends in the last year. But he very much thinks for himself, if he wants to talk to us" (SSI: 12)*

Accepting the fact he would lose their son, and viewing any time with him as enriching life, was described by one father of a young son:

*"The bottom line is if X dies, my life will have been richer for knowing X the way he is. If he doesn't die it will continue to be richer. So, I'm winning either way" (SSI: 4)*

Fathers thought a lot about how the child would cope at that time of his death:

*"You think about how he will cope, dying- that's what you think about" (SSI: 5)*

*"You would have thoughts like I wonder how he will die, and how I'll be when it happens, and will it be one of those deaths where I can encourage him to let go if he needs" (SSI: 4)*

One father whose child had died years before, clearly recalled a conversation where the child had used humour to let his family know he was prepared for death:

*"He said (to his) 'Grandad, you know you had your party when you were 90', he says 'yeah, well you were wrong, you're not supposed to have it until you're 100'. He said 'yeah, but suppose I don't live to 100, I've had my party when I'm 90 so I can enjoy it. If I live to 100 we'll have another'. He said 'yes Grandad, if we're still here!'" (SSI: 13)*

This father found the son's comment reassuring, as it confirmed that he was aware of his prognosis, although this had never been discussed overtly as a family.

## Chapter 8

### Discussion and Conclusion

This is the first known U.K. study to investigate the adjustment of fathers with sons with DMD. The quantitative study, drawing upon conceptual strands of adjustment, involvement and support; and the resiliency model of adjustment (McCubbin and McCubbin, 1993) to identify variables, assessed correlates and predictors of fathers' adjustment, whilst the qualitative strand explored fathers' perspectives. The discussion is organised under the original research questions, illustrating findings from each component of the study. A critique of the study methodology follows the discussion. Consideration of the implication of findings for interventions concludes the chapter.

#### **8.1. Is paternal adjustment associated with child's level of functional ability and psychological/behavioural adjustment?**

Quantitative results indicated overall risk of elevated psychological distress in this group of fathers. A key finding was that 38% of fathers scored within the range for clinically significant problems. This is in line with DMD studies with mothers, for example Garralda et al's (2006) sample of 17 mothers presented 41% within a clinically 'at risk' (scoring above cut-off using the G.H.Q.) bracket.<sup>47</sup> Results also parallel those of Abi Daoud et al (2004), who identified 31% of DMD parents (mothers) compared to 4% controls, 'at risk' for probability of a major depressive episode. Various studies have identified mothers as being vulnerable to increased

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<sup>47</sup> Caution is needed in interpretation, however, due to the small sample size.

mental health problems, suggesting this is due to being the main carer. The present study indicates that similarly high rates may also be present for fathers.

In relation to child disability and adjustment variables, the study identified a number of factors associated with paternal adjustment. In summary, boys' physical impairment had a significant impact on their social activities, for example 32% found 'a lot of difficulty' or that it was 'impossible' to play with friend. Garralda et al (2006) reported similar findings, identifying most problems surrounding gross motor tasks, with one third having difficulties with social activities. Overall, 22% of boys presented significant psychological adjustment problems according to the Strengths and Difficulties Questionnaire. Consistent with qualitative findings, and in keeping with previous DMD research (Garralda et al, 2006), areas resulting in the majority of problems for boys related to emotional (32%) and peer (45%) problems.

Increasing disability was associated with boys' poorer overall adjustment, with a trend towards increasing emotional, conduct and peer problems, however functional ability was not associated with paternal adjustment. Although functional ability was not associated with paternal adjustment, the Strengths and Difficulties Questionnaire sub-scales: emotions, conduct and peer problems, however, were positively associated with paternal adjustment. This indicates a possible 'knock-on effect' of the relationship between boys' increasing disability and parallel adjustment problems increasing paternal stress. Boys' problems in these areas increased, as the child's condition became more disabling, indicated by a significant association with higher disability scores for those with more difficulties with peers. This illustrates the impact

of disability on the child's ability to socialise. The association between poorer functional ability and more psychosocial problems (namely peer and emotional problems) suggests boys who are less able to interact with peers, or who lack the skills to do so, experience adjustment difficulties. Previous research with parents of intellectually disabled children, has demonstrated that fathers, more than mothers, have concerns about their child being socially included (Saloviita, Italinna and Leinonem, 2003).

Given the lack of association between child's disability and paternal adjustment suggests, it appears that the impact of boy's psychosocial adjustment problems is a more important factor in paternal adjustment. Supporting previous findings with mothers (e.g. Nereo, Fee, and Hinton, 2003), it is possible that non-condition specific variables, rather than actual condition demands are more closely associated with paternal adjustment. It may be, therefore, that in addition to the impact of behavioural/emotional problems of sons, concerns regarding the impact of boys' adjustment on their ability to interact with peers is an influence on paternal adjustment.

Child adjustment was a significant predictor of paternal adjustment, accounting for 17% of the variance. Similar findings have been reported (Nereo et al, 2003; Reid and Renwick, 2001), in research with mothers, finding predictors of maternal stress were related to child variables. As with previous DMD studies (e.g. Chen and Clark, 2007), disability alone was not found to predict paternal adjustment. Similarly, Abi

Daoud et al (2004) found no association between child's ambulatory status and adjustment outcomes.

Findings also support those of Perrin, Lewkowitz and Young (2000), who studied parental needs in relation to services, reporting that unmet needs were seen as more important than severity of child's condition. This may be true for the boys themselves, as illustrated by a Department of Health (2003) needs assessment of 13 DMD boys, which found that boys desired information about how to cope with the impact of DMD on emotions, and social aspects (Beresford, 2003).

No demographic factors were associated with paternal adjustment. Results support those of previous DMD researchers (e.g. Chen and Clark, 2007; Reid and Renwick, 2001), who also found that familial stress was not related to socio-demographic variables but was related to psychosocial adjustment in the adolescent. In sum, data reflect previously reported findings of increased behavioural problems in boys influencing their own and paternal adjustment, and that condition specific variables do not solely account for resulting stress.

The qualitative study identified a number of factors to add to the above quantitative findings. High importance was placed on sons living as normal a life as possible. This was particularly in relation to acceptance and friendships with other boys. Concern and loss were felt when sons were socially isolated or experienced trouble with friendships. Teen years were especially challenging due to comparison with healthy peers and boys' inability to participate in usual teen activities, which often led to

frustration. Consistent with quantitative findings, fathers' inability to help their sons made coping harder.

Also consistent with quantitative findings, deterioration itself was not generally perceived as a key issue as physical aspects of care became routine, but distress resulted from seeing peers 'overtake' and become independent, whilst their son grew increasingly dependant. Fathers described a need to deal with ongoing decline and adjust to each situation, as there was no alternative. Supporting previous findings (e.g. Gagliardi, 1991; Witte, 1985), some described long periods of little change, followed by dramatic change and needing to cope with each stage. Sons' ageing introduced issues surrounding later stages of the condition. This included fear of their son being rejected and seeing him reach the final stage of deterioration. A specific need mentioned by fathers was dedicated health professionals to look after the various needs of older boys, not just the physical aspects.

Not knowing for sure when the next stage would arise led to anxiety for some. This seemed to activate continual uncertainty for the future, framed by re-evaluation of prior expectations and 'reminders' of the future. Findings echo those of Buchanan et al (1979), Witte (1985) and Bothwell et al (2002), who identified anticipation of future stress and future needs as key issues for DMD parents.

The finding that child adjustment problems, but not functional ability was a predictor of paternal adjustment problems is consistent with previous research with mothers (e.g. Nereo, 2003). Rather, stress results from emotional challenges surrounding the



condition for the boys themselves and this may affect fathers' adjustment. Findings support past research indicating child adjustment as more important than functional ability (e.g. Cohen, 1999). Stress also appears to result from the contrast in expectations, the reality of deterioration and the impact this has on boys' social adjustment, often having a detrimental impact on boys' behaviour.

Findings support those of Darke et al, (2006), who identified unmet needs in relation to dealing with boys' social problems. Fathers in the present study indicated a need for help with communication and being honest with boys, especially around adolescence.

### **Key points**

- Functional ability was not associated with paternal adjustment
- Increasing disability was positively associated with boys' peer and emotional problems
- Boys' psychosocial adjustment was a predictor of paternal adjustment
- Fathers were concerned with sons' loss of friendships and isolation
- Uncertainty about the future had a negative impact on paternal adjustment

### **8.2. Is paternal adjustment associated with perceived amount/helpfulness of involvement and perceived level of support received?**

The variables most strongly related to paternal adjustment were D.A.D.S. amount and support from friends. Poorer levels of adjustment were associated with less perceived helpfulness of involvement and lower perceived levels of support from

friends. Fathers reported involvement in child-care in 25-50% of opportunities over the previous 6 months. This is in line with findings reported by Wysocki and Gavin (2004), who stated this level of involvement in child-care leaves *'substantial room for improvement'* in paternal involvement (Wysocki and Gavin, 2004, p.231). Amount of involvement was positively associated with adjustment, indicating increased provision of emotional and instrumental support may be a factor in successful adaptation. Fathers perceived their involvement as making the situation *'neither harder nor easier'*. Similar to the findings of Gavin and Wysocki (2006), it is possible that fathers may not appreciate the benefit of their involvement.

Consistent with Gavin and Wysocki (2006), no significant associations were found between paternal involvement and impact of the condition on the family. It may be that helpfulness to mothers is associated with family adjustment but amount may be more relevant to paternal adjustment, and in turn, boys' adjustment. Previous research (e.g. Maurer and Pleck, 2006) has identified the impact of maternal appraisals on fathering identity, and in this study, some fathers described feeling distanced or awkward around the child's routine. Lower helpfulness scores were associated with poor adjustment, suggesting that less well-adjusted fathers interpreted their helpfulness as less valuable. Research has indicated that mothers often do not wish for fathers to be involved and had low confidence in their ability (Lloyd and Lewis, 2003), and perhaps this is communicated indirectly in DMD families.

Regarding satisfaction with perceived support, most were satisfied with hospital (56%) and family (59%); and half were satisfied with support from friends (50%). Support variables overall were associated with boys' adjustment and paternal adjustment. Support from family was negatively associated with child emotions, S.D.Q. total and family impact. It is possible that lower perceived support from family was associated with increased child emotional problems and overall poorer family adjustment. Paternal adjustment was negatively associated with support from friends, indicating less support reported by less well-adjusted fathers. The lack of association between paternal adjustment and other areas of support (i.e. hospital) indicates the importance placed on friendships. It is acknowledged, however, that correlation analyses does not allow insight into the direction of associations between variables.

Amount of involvement and support from friends, were both significant contributors to paternal adjustment, accounting for 43% of variance in G.H.Q. scores, with amount being the strongest predictor. Support from family and clinic, were not significant, suggesting a key role for fathers' friendships in relation to adjustment. Of note is McNeill's (2004) suggestion that the social network may be an emotional burden for fathers, resulting in isolation. Interestingly, Wijnberg-Williams et al's (2006) research investigating psychological distress in parents and social support (using G.H.Q. and support measures) over a 5-year period, demonstrated dissatisfaction with support and negative interactions as significantly affecting fathers, and not mothers' adjustment.

In terms of involvement, the qualitative study indicated that fathers often described work as a barrier leading to feeling removed from the routine to an extent, although one father had taken early retirement to care for his son. As a result, their partners' bond was perceived as stronger in some cases. Most fathers<sup>48</sup> felt their experience of caring for their child differed from that of mothers. Fathers were more involved in research and practical matters, whilst mothers were more involved in physical care. As a result, wives were usually perceived as more engaged in emotional aspects, with fathers describing their roles in relation to more physical and practical areas. This could lead to a sense of frustration and isolation.

There was a balance to be struck between focusing on both career and family issues, and this was often challenging. Fathers wanted to be involved in aspects such as physiotherapy and meetings. Often after work, fathers would carry out physiotherapy routines with their son, however, others felt isolated from aspects of care due to work commitments. When asked about involvement, the main barriers were timing due to work, and sometimes being neglected in relation to care-taking. This reflects the findings of Grey (2003), who identified fathers' perceptions of their role as a secondary support to care-taking partners. In relation to Lamb's (1987) tripartite involvement model (see p.15), results identified issues surrounding *interaction*, including communication with partner, professionals and son. *Accessibility* related to perceived barriers to involvement and *responsibility* involved a perceived expectation to appear to be strong and coping, to fight for services and

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<sup>48</sup> This observation from the qualitative study was supported by a content count of comment sheet responses, with 76% (n38) citing differences

to protect the family. The model offers a useful method of conceptualising key factors central to paternal involvement.

Within paediatric settings, communication with professionals and information provision<sup>49</sup> have previously been identified as inadequate from parents' perspectives (Hummelinck and Pollock, 2006). With regard to professional support, although good practice was reported, there was a general perception of having to fight for professional help, especially with benefits and housing, leading to increased stress in an already challenging situation. Where needs had not been met by either family or professionals, key issues were families not asking about the emotional needs of fathers and professionals not enquiring about coping. It was also challenging to find guidance for parents of older boys, as they were 'writing the rule book'.

In relation to support needs, the main areas where help was needed were: emotional support (including professional awareness of the potential for fathers to feel 'isolated' and the need for father only support groups), and advice about how to communicate with sons. Education about the condition (including best and worst case scenarios), help for older boys and respite were further issues. Loss of friendship support and self-confidence resulted for some, since diagnosis. This sense of isolation sometimes resulted from social withdrawal as an attempt to protect the family. Difficulty or a desire not to talk about DMD was reported by some, further impacting upon friendships. There was a wish to talk to a professional outside the family, but not many had taken this further.

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<sup>49</sup> It is noteworthy that parents may resist information (due to its negative impact), as a coping mechanism (Hummelinck and Pollock, 2006).

Although seeking support from other DMD families helped, as with Erby et al (2006), there was sometimes a need to distance themselves from others with the condition. This was especially the case when it proved upsetting to witness older boys' decline. Again, this may result in loss of support. Similar to Firth et al (1983) who described negative effects on the marital relationship, some felt since diagnosis their relationship was in the background due to challenges in having a break from care. In some cases problems resulted from different methods of coping compared to partners.

### **Key points**

- Perceived amount of involvement was a predictor of paternal adjustment
- Fathers wished to be involved with sons but were under-involved in sons' condition specific care
- Fathers may not appreciate the benefits of their involvement
- Barriers to involvement included feeling isolated from care routines and work commitments
- Fathers may be vulnerable to social isolation
- Support from friends was a predictor of paternal adjustment

### **8.3. What is the experience of parenting a son with a progressive terminal condition?**

The qualitative analysis illustrated the emotional impact of parenting a son with DMD. All experienced the extent of the condition at certain stages, with a range of reactions from those who coped well to those who found most days challenging.

Fathers described an array of perspectives, however, a number of common themes linked their experiences. Many coped positively, although often described the unrelenting influence of DMD on the whole family. The first major challenge was dealing with diagnosis, particularly revising previous expectations held for sons. This was a distressing time, involving loss of the father-son activities hoped for and parenting ideals fathers held.

At this time, anger, frustration, guilt and shock, similar to previous findings (Webb, 2005; Buchanan et al, 1979) were reported, along with a perception, for many, of having received poor information or not being able to understand information received. This finding supports Firth et al's (1983) study of DMD parent's experiences of diagnosis, where one third were not satisfied with how they were told. Around this time, attention may be focused on mothers and children, and fathers may feel a sense of expectation to be strong for others. The manner in which diagnosis was conveyed remained vivid in fathers' memories, and when this was perceived to be handled badly, this was dwelled on.

Following the shock of diagnosis, fathers generally perceived a number of 'losses' in relation to expectations, their own and sons' isolation. In keeping with Kornfeld and Siegal's (1979) reported 'cycle of loss', an underlying theme of loss, due to limited life-span, was obvious throughout fathers' descriptions. In addition to areas previously described, as with Lee et al (2006), this extended to re-evaluation of previous expectations for continuing the family name.

Challenges included seeing siblings and friends growing up, and becoming independent as their sons' condition declined. This was painful to witness, and fathers often did not discuss this with others. Some described a loss of everything 'normal' families take for granted. Specific times, such as approaching stages of DMD and perceiving sons being isolated from healthy peers as the boys aged, were particularly hard to deal with. Concern included boys being socially accepted, whilst comparison with peers emphasised their sons' condition, reinforcing a sense of overall loss. There was uncertainty as to how to meet their sons' needs and where to seek emotional support for boys.

Many felt it was not possible to adjust fully due to- as described by one father, the '*shifting sands of DMD*'. Because of repeatedly experienced loss, with no set milestone, stress was felt to increase in light of uncertainty. Some lived in anticipation of next 'stages', and through fear of this, felt an inability to become too close to their sons as a possible means of self-protection. In keeping with Kornfeld and Siegal, (1979), a key factor may be that DMD boys look normal in their younger years, and loss of function (resulting in obvious disfigurement) is slow. This may lead to a repeated cycle of loss, adaptation and loss, creating more stress. Absence of boys' friendships also contributed to this loss, with fathers often described feeling helpless, useless and angry at their sons missing out.

Adaptive coping was achieved through proactive attempts to make the most of life, whilst not looking too far ahead. Many fathers coped well, maintaining a sense of hope for a cure and using charity work or fundraising as both a distraction and



coping mechanism. As also identified by Erby et al (2006) in discussions of advanced care planning with DMD parents, maintaining an element of hope was important and it helped when professionals provided this, whilst remaining realistic. Despite the terminal prognosis, fathers wanted to know they could still do normal activities to some extent.

Many moved forward after an initial mourning period and coped through practical efforts with DMD campaigns. Others found help through the structure of work and returned to work quickly. With time, boys' routines and needs became a part of family life, although involvement with sons' medical regime was often prevented through working, and mothers took on the brunt of personal care. Fathers sometimes felt isolated, both from routines and in relation to attitudes and interactions with professionals.

Complete adjustment was often described as impossible due to constant changes associated with the condition, leaving no time to 'recover'. Some reported a sense of duty to appear to be coping and be strong for the family, when they actually felt especially vulnerable. They described a need to conceal their own support needs, feeling they were expected to cope and be strong for the family. They noted emotional support being offered to mothers but not fathers.

In contrast to Buchanan et al (1979), and Chen et al (2002), coping strategies including self blame, wish fulfilling fantasy and 'magical thinking' were not described. However, defensive coping mechanisms reported as attempts to cope, included

withdrawing, or working overtime to avoid family contact. Most were realistic, however, and often made attempts to over-compensate through providing 'amazing' experiences their son would remember. This appeared to be a form of over-protection, also found by Kornfield and Siegal, (1979). Coping was generally described in terms of being less emotional and more practical than the mothers' care role. There may be a perceived expectation for fathers to attempt to counteract mothers' more emotional focused approach, as has been described previously (McNeill, 2004).

Friendships were described as an important support, and in a number of cases these had been affected by fathers' own reactions to the diagnosis. A perception was held that people outside the family could not fully understand the impact of the condition. They often did not wish to, or did not feel others wanted to talk about the child's condition.

Fathers described the whole family as affected, including maternal guilt, testing daughters for the gene and problem behaviour from siblings due to attention placed on the boy with DMD. Consistent with findings of Firth et al (1983) and Fitzpatrick and Barry (1986), communication difficulties emerged as an important area for fathers. Communication within relationships also led to challenges, especially with partners, where lack of agreement occurred, or no desire to discuss relevant issues, was desired by one party. In line with previous work (e.g. Pelchat and Perreault, 2003) interviews identified that coping dissimilarities as reported by fathers, often exacerbated problems within the family.

Similar to Erby et al (2006), avoidance of emotionally sensitive issues was reported. Communication with sons was highlighted, in particular discussing issues surrounding death, and lack of awareness of how much the child already knew, were causes of distress. Witte (1985) has previously identified problems regarding discussion of death issues in DMD families. Knowing how to approach this topic and how best to deal with it, emerged as an important need. In a number of cases, avoidance of discussion surrounding death arose. The significance of the sex of parents and awareness of child dying is understudied, with recent authors suggesting more research may guide care efforts to promote well being (Hinds, 2007).

Decision-making around treatments was often a cause of stress, made worse in some cases by conflicting advice and a perception of time running out and therefore pressure to decide between options. Making decisions was also challenging, in light of the pain procedures may cause the child for unpredictable gain. A need was voiced by some for better information about trials and treatments. As various treatments are involved in slowing the progression of DMD, fathers often felt the pressure of balancing the child's future prospects (such as ability to walk if a cure was found) with invasive and painful treatments (e.g. spinal operation to prevent curvature of the spine). They felt their children generally coped with treatment well, and this made things easier.

### **Key points**

- Adjustment was difficult due to unpredictable changes in the condition

- Expectations to be seen to cope resulted in reluctance to disclose distress
- Fathers wanted more support around decision making for treatment options
- Emotional effects and practicalities (treatments; housing adjustments; grants; schooling; benefits) were ongoing stressors
- Support from friendships may be protective for fathers' distress but fathers may not seek social support

#### **8.4. What are fathers' views of, and suggestions for improved support?**

Although cases of excellent practice were reported, some felt support services did not account for families' let alone fathers' needs. Two key issues arose regarding services: firstly, fathers often felt overlooked or isolated from involvement, and secondly, partnerships and communication with professionals could lead to frustration.

The need to protect and fight was repeatedly referred to, and without understanding this reactive need and expectation, professionals may simply view some fathers as aggressive or difficult. Similar to Fitzpatrick and Barry (1986), communication with both professionals and within the family was a key issue. Frustration at having to educate professionals about DMD, and communication issues within the medical profession, was described. Similar to research investigating the psychosocial impact of a genetic X-linked condition- Allport Syndrome, (Pajari and Sinkkonen, 2000), having to 'educate' professionals and constantly explaining the condition specifics was stressful. Health workers have previously been found not to acknowledge parents' need for information about the implications of the condition (Perrin et al,

2000). Acknowledgement by professionals of the impact of treatment delays and time scales was an important issue, in light of fathers' heightened awareness of their sons' limited life span. Research in the field of childhood cancer has shown that at later stages, more detailed information is required to steer parents through treatment procedures (Earle, Clarke, Eiser and Sheppard, 2007).

Negative experiences included a feeling of being viewed as surplus to requirements by staff, perceived as having less involvement with the child compared to the mother and perception of receiving a lower quality of service without a fight. Many fathers felt a need to 'fight' for their child's care, and co-ordinated a large number of agencies. A more cohesive support package was felt to be a step forward. Dissatisfaction with support and negative interactions that fathers experienced with professional services had an impact on levels of distress. Previous research has demonstrated such a lack of awareness amongst health providers, about the impact of emotional issues on parents (McKay and Hensey, 1990).

Specific needs, especially around diagnosis included information about how to communicate with their son, details about the condition, what to expect at various stages and activities they would be able to do with their son. Fathers also wanted an opportunity to talk in a confidential setting with professionals who could help them understand this process and come to terms with loss and adjustment issues. Fathers also wanted to be listened to and advised realistically, whilst maintaining hope. Additional areas requiring support were coming to terms with the effects of disability and being able to talk about it. Being honest with their child was another area in

which advice was needed. Longer appointments allowing opportunity to explore concerns, was suggested as a means of meeting needs. There was frustration at the lack of father-related health service awareness. Specific times where this was deemed most relevant included post diagnosis, at times of decision-making and as boys reached adolescence.

Acknowledging the boys' restricted life span in the context of treatments was also identified. Additional needs included provision of optional emotional support to deal with diagnosis, inability to 'mend' the situation and advice about talking about DMD with sons. As with the work of Firth et al, (1983), a number of fathers felt they had not been able to understand or process information given by professionals. This was often due to the heightened emotions surrounding interactions with medics. This is consistent with Chen et al's (2002) finding that fathers needed more help from resources and information.

Awareness of professionals that fathers often feel surplus to requirements, and feel there may be barriers to involvement in care was needed. Fathers also worried about transition from child to adult services and lack of opportunity for sons to attend social activities where they could actively be involved and not simply watch others. Frequently they expressed a need for a more cohesive service, with one contact point. Previous work (Heller and Solomon, 2005) has found that consistent staff and co-ordinated continuity of care results in less anxiety in parents and a belief the child is receiving good care (Heller and Solomon, 2005).

Such continuity appeared to be lacking in the current study, resulting in increased levels of frustration and 'chasing' services. Dealing with numerous appointments, especially at an early stage placed added stress on families. In terms of support from professionals, fathers wanted an opportunity for support if required but only a few had received psychological intervention, despite a number suggesting a need for this. A further need was to know that they were not alone in their current situation. Fathers only support groups were suggested as a way of meeting fathers' needs. Liaising with employers, and a negotiating for working flexibility, was a further issue that was repeatedly mentioned.

Fathers have previously demonstrated high stress in relation to perceived incompetence (Dellve et al, 2006). A number of the fathers in the current study felt ignored or 'talked over', when attending appointments. Often, they did not feel able to discuss some of the emotional implications. Qualitative results also illustrated that fathers often perceived their support needs as less important (valid) than those of their son and partner. Some even felt guilt at considering their own needs, in light of their sons' disability. Previous research has found similar results, indicating a lack of acknowledgement of fathers needs (Bailey, 1991). As with the work of McNeill (2004), fathers in this study attempted to demonstrate strength for others and often over relied on self-support strategies.

Many described having lost supportive networks, sometimes due to their need to spend time with the family. Fathers' reluctance to seek emotional support has been described previously (e.g Pelchat and Perreault, 2003). Researchers have previously

suggested that fathers are at risk as a result of isolation due to lack of social support and a need to be in control (Sabbeth, 1984). It has been suggested that the social network is potentially a source of emotional burden (e.g. McNeill, 2004), for some, perhaps leading some fathers to isolate themselves to prevent this.

In the current study, isolation and loss of friendships were key issues raised by fathers. Similarly, Firth et al, (1983) found that social isolation for both parents and sons was a main concern. Further, this was also associated with an increase in child's emotional problems. These findings echo previous work with parents of a child with cancer, where social support variables accounted for increased levels of father but not mothers' distress (Hoekstra-Weebers et al, 1999 and 2001).

In terms of personal support, most stated their partner and immediate family provided support, with needs met often within the family. It is possible, however, that those close to fathers may be too upset to provide appropriate support. Sometimes this caused problems, for example, when reluctance of one partner to discuss ongoing issues, led to lack of opportunity to discuss the impact of DMD. This situation was highlighted when coping dissimilarities were described. Similar communication problems were identified by Fitzpatrick and Barry (1990), and highlighted as one of the main stressors within the family.

Results show that perceived availability of social support in accordance with relevant needs is an important issue for fathers. Carers with more support are more able to use productive coping strategies and meet psychological needs (Love et al, 2005).



Although social networks provide emotional support (McGarry and Arthur, 2001), demands of caring for a son with DMD may have a negative effect on these relationships. As proposed in the theory of stress and coping (Lazarus and Folkman, 1984) a number of interactions lead to adaptive coping, including condition specific, appraisal of the situation, and available resources to cope. Social support fits into this model as a positive way of coping and reducing perceived stress.

### **Key points**

- Fathers may perceive themselves as distanced/ overlooked from home and clinic based care
- Perception of services as 'chaotic' with multiple professionals involved suggests a need for a point of contact where services (e.g. benefit advice; paediatric services; physiotherapy) are brought together
- Emotional support was needed around diagnosis, decision-making, and stages of change
- Help communicating with and supporting sons (socially and emotionally) was needed

### **8.5. Summary**

The impact of DMD on fathers is evident from the findings. Overall, psychosocial factors- child adjustment, involvement and social support were predictive of paternal adjustment. Psychosocial determinants may therefore be more important to adjustment, in comparison to condition and socio-demographic variables. Together, the findings from both strands of the study indicate influences on fathers'

adjustment were 1) child-related factors (boys' emotional and peer problems) and 2) socio-ecological factors: involvement and friendships.

Results may be interpreted within the Resiliency Model (McCubbin and McCubbin, 1993) as outlined in Chapter 2 (p.51) as an investigation of specific stressors and resources. According to the model, less involvement with the child and loss of friendships are potential risk factors for paternal adjustment problems. Disability and socio-demographic variables were not associated with adjustment. The lack of association between disability and paternal adjustment indicates the impact of emotional/behavioural, not condition specific variables in relation to paternal adjustment. Findings indicate, however, that these areas are not routinely addressed by professionals. The qualitative study also highlighted the magnitude of distress and perceived isolation some fathers experience, with unmet needs resulting in increased frustration. Together, results highlight the importance of greater mental health input and a need for professional awareness.

In sum, the study provides initial information about paternal stress in DMD families. As with previous work (Raina, O'Donnell, Rosenbaum, Brehaut, Walter et al, 2005; Hinton et al, 2006; Nereo et al, 2003; Chen, 2008) results suggest interventions should be aimed at supporting parents to cope with boys' emotional and behavioural problems, with provision of support integrating practical strategies for fathers to promote adjustment.

Findings complement the work of Government agencies such as Sure Start, which aims to support families through integration of education, health and family. In evaluating Sure Start, Lloyd et al (2003), recommend developing a coherent plan for involving fathers. The first step towards helping people necessitates understanding the problems they face, how they make sense of events and how they adapt (Dewey and Crawford, 2007). It is hoped that this thesis has made an initial exploratory step towards this goal.<sup>50</sup>

### ***8.6. Methodology critique***

Weaknesses of the study include the cross-sectional design and the relatively small sample size for the quantitative component, which has the potential to inflate predictor effects (Coolican, 2004). Regarding measures, reliance on father-only self-report introduces the possibility of response bias due to lack of objectivity (Howard, 1994). In relying solely on father report, it could be argued that mental health issues coloured their perceptions of child related problems. Father's distress, for example, may bias reporting of child physical or psychological symptoms. A related methodological issue is that of 'source variance', as fathers provide information about their own mental health and their child's problems, which may artificially inflate resulting associations (Hastings, 2003).<sup>51</sup> Methods of obtaining supplementary information might have included teacher report of child emotions/behaviour.<sup>52</sup> In

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<sup>50</sup> Of note, is that mid way through recruitment for this study, an article was printed in the muscular dystrophy campaign magazine, 'MD Matters'. The article, written by a care adviser entitled 'how dads cope' (Stein, 2007), described two fathers' accounts of how diagnosis and differences in coping placed a huge strain on relationships. The neglect of fathers and need to understand their experience was mentioned in this article. This article confirmed that the current research represented a real need.

<sup>51</sup> Interestingly, research examining relationships between parent's reporting of their own and their child's health and illness (e.g. Waters, Doyle, Wolfe, Wright, Wake and Salmon, 2000) has demonstrated that, although parents self reporting poor health were more likely to report poor child health, this may be affected by parent gender. Mothers' self-reported and child health were strongly associated, but this effect was not found for fathers (Waters et al, 2000).

<sup>52</sup> Future work might include teacher/sibling report, however in the current study, time and resource limitations necessitated reliance on father report.

this exploratory study, however, self-report offered a feasible approach to measurement as focus was placed on perceived problems that impair adjustment.

A further limitation is the unknown extent to which fathers in the quantitative study were representative of families with DMD. Although the sampling frame involved recruiting from various organisations, they were all, by definition some type of support. The research may also under represent lower income, less educated parents as highlighted by the nature of fathers' employment, indicating higher socio-economic status for 57%<sup>53</sup> of participants.

Regarding statistics, use of correlation in this study allowed a degree of insight into associations between variables, but it has been highlighted (e.g. Raina et al, 2005) that this does not lead to full examination of 'multidimensional pathways'. This is due to no insight being provided into the direction of associations. Thus, to address this as far as possible, a combination of quantitative and qualitative techniques allowed some insight into relationships surrounding predictor variables and outcomes.

Despite identified weaknesses, a key strength of the study was allowing insight into fathers' experiences, in an under-researched area. This added a previously untapped perspective into the topic. Inclusion of fathers from across the U.K., representing a range of perspectives, strengthened the study further. Use of standardised measures

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<sup>53</sup> Socio-economic status data available for 37 of 50 participants.

to assess psychiatric function, functional ability and involvement, and adherence to appropriate evaluative criteria also heightened confidence in the study findings.

### **8.7. Conclusion and recommendations<sup>54</sup>**

The picture is not a wholly negative one, with many fathers coping well despite the challenges. The decision to give their child the best possible experiences, led to an appreciation of life, which some felt may not have been the case with a 'healthy' child.

#### ***What do the findings contribute? Implications for the design of clinical interventions***

Guided by the Resiliency Model (McCubbin and McCubbin, 1993), (p.51) this study has identified issues surrounding adjustment, involvement and support and related experiences in caring for a son with DMD. Barriers have been uncovered, along with an indication of stages of greatest support needs. The results contribute knowledge to this area by providing health care professionals with a starting point to aid understanding of fathers' perceptions and improved information provision. Findings may inform basic interventions, for example by involving fathers, promoting supportive networks and targeting parents' understanding of each other's reactions.

Professionals might anticipate the reactions of some fathers, specifically issues surrounding loss/expectations; involvement and withdrawal from social support. In

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<sup>54</sup> Dissemination: participants and relevant organisations will be informed as to the key outcomes of the work, and encouraged in accordance with the NHS 'Involve' model, to act as contributors (as appropriate) to publications that may result from the research. In addition the Medical Director of Children's Hospice Association Scotland, physiotherapists at Great Ormond Street Hospital and research practitioners at Bristol and Birmingham Universities have requested the results. Professor Carpenter, the author of 2009 Department of Health Report '*Transition to adulthood for DMD boys and their families*', refers to the lack of empirical work with parents and carers of young men with DMD in the UK. He also mentions the fact studies rely on maternal report, and notes awaiting the results of this thesis.

addition this includes the need to be practical and perception of the expectation to 'fight' for their son. Family interventions (e.g. Fiese, 2005) could encourage mothers' and professionals' awareness of the importance of supporting paternal involvement in medical routines and appointments. Encouragement of appropriate support seeking and insight into fathers' potential to isolate themselves could also be integrated into basic interventions.

The fact that many feel a sense of isolation might be addressed through encouraging male-sensitive communication (e.g. accounting for perceptions of others' expectations) and involvement with others in a similar position. For some fathers, simply being told these perceptions are not unique may relieve a degree of the stress associated with caring for their son. Due to the progressive nature of DMD and related challenges, anticipatory guidance could be available. In addition, availability of father specific support at critical periods, such as decision making in relation to operations or milestones such as preparing boys for wheelchair use, may prove beneficial.

At early stages of working with families, these issues, common responses and preparatory coping strategies may be discussed. According to theoretical models (e.g. McCubbin and McCubbin, 1993), coping is dependant on finding equilibrium between having access to resources and negative impact of DMD. In increasing such positive resources, this research suggests some benefit in fathers being made aware, for example, of the importance of maintaining social networks and of

focusing on the positive aspects of caring for their son, such as opportunities to seek new experiences which otherwise may not have happened.

In relation to coping resources (e.g. Lazarus and Folkman, 1984; Lazarus, 2000), maintaining hope may be essential. Fathers reported here echoed this need for hope, often in light of professionals not wishing to be overly optimistic. Some fathers felt that communication from professionals and information provided was inadequate. This draws attention to the possible role of health psychology in assisting clinical staff to develop an awareness of how parents think of and experience DMD. Findings reported here emphasise the need for bio-psychosocial interventions, moving focus from physical interventions required by boys. The psychosocial impact, for fathers and families, should be acknowledged as being equally important as medical interventions in DMD.

As DMD is the most common of childhood neuromuscular disorders (two boys are born with Duchenne every week in the U.K.), it has been suggested that attention should be placed on the wide- ranging implications for all who are affected (Morrow, 2004), as boys are now living longer. In addition to the focus on physical and genetic aspects within DMD literature, this thesis highlights a need to promote research into understanding and tackling the emotional impact on family members. Identifying factors impacting on fathers and related experiences, allows insight for improved service provision.

The Resiliency Model (McCubbin and McCubbin, 1993), (see p.51) provides a useful explanatory guide for understanding factors associated with quality of adjustment in fathers of sons with DMD. Within the model, support from friends and involvement may be potential resources, with child adjustment problems acting as a stressor in relation to paternal adjustment to their sons' condition. According to the model, loss of social support and perceiving professional support as inadequate or to 'fight for', may be a stressor for some fathers. The model may be applied to future research with fathers to understand needs, and identify areas that may benefit from intervention.

Drawing upon findings, it is proposed that future conceptual models incorporate factors such as dealing with issues surrounding expectations; management of child related adjustment problems; communication with sons; involvement; and social support, in order to promote a wider approach to subsequent interventions. Awareness by professionals of the emotional impact, reactions and issues they face, may reduce frustration amongst fathers. Health psychologists are in a position to make a positive difference, by improving awareness amongst health providers, highlighting needs, devising interventions and evaluating them. The results of this thesis and other studies emphasise the importance of a broad approach to family centred support. This is in contrast to emphasising physical interventions that focus on the child.

Health professionals require understanding of fathers' adaptation to their son's condition, so that they can ensure a collaborative, effective approach to working



together. The National Service Framework for Children (Department of Health, 2003) requires the development of services that account for needs of children and families. Findings of this research emphasise the potential for broadening traditional models of family adaptation to include the experiences of fathers. Researchers might note the lack of relevant work in this area, whilst including fathers within relevant study designs.

This thesis serves as a starting point for future research to enhance understanding of DMD families' needs and to further improve both the type and amount of available support.

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Department of Health Online: <http://www.dh.gov.uk>

Duchenne Family Support Group: <http://www.dfsg.org.uk>

Muscular Dystrophy Campaign: <http://www.muscular-dystrophy.org/research>

National Health Service Online: <http://www.nhs.uk/conditions>

Parent Project UK: <http://www.parentprojectmd.org>

## **Appendix 1**



Queen Margaret University College

EDINBURGH

### **Experiences and views of fathers of a child with Duchenne muscular dystrophy**

#### **An Information Sheet for Participants**

My name is Anna Cunniff and I am a post graduate student from the Centre for Health Psychology at Queen Margaret University, Edinburgh. As part of my doctoral degree, I am undertaking a research project. The title of the project is: 'experiences and views of fathers of a child with Duchenne muscular dystrophy'.

#### **Why am I undertaking this work?**

In comparison with other conditions, the impact of neuromuscular disorders- in particular Duchenne, has remained largely under-researched. There are few studies investigating parents' experiences of caring for a child with Duchenne, and available studies tend to focus on mothers as the main caregiver. As a result our understanding of the impact of Duchenne on families is quite limited.

Research with mothers has highlighted the care-taking challenges experienced by carers from a female perspective. The needs and views of fathers of a child with Duchenne have not yet been fully identified. However, the views of fathers could give a deeper understanding of issues faced by parents and thereby enable healthcare professionals to provide support that more effectively meets the needs of families who care for a young child with DMD.

#### **Relevant work:**

I previously worked as a researcher, under the supervision of Professor Muntoni at the Dubowitz Neuromuscular Centre, Hammersmith Hospital London. The study involved working closely with families and children with Duchenne. The work is now published, and demonstrated the effects on carers (mothers) of the child's condition.

As a result I developed an understanding of the considerable responsibilities involved in caring for a child with a progressive condition. I also gained an appreciation of the need for further work in this area. I am now keen to pursue my interest in this area, in an attempt to address the lack of research in both the areas of neuromuscular disease and reports of fathers' views.

This work will serve to raise the profile of neuromuscular conditions and the impact on carers, amongst health and research professionals.

The outcome of the research will also contribute towards enabling the appropriate provision of support services for families who have a child with Duchenne.

Previous work I have been involved with covers a study looking at user views and adjustment to the use of KAFOs, and an article aimed at health professionals highlighting issues facing carers of children with Duchenne.

#### **Funding:**

I am self-funding this project as part of my doctoral degree.

**What will I have to do if I take part?**

The research consists of two parts: questionnaires and interviews. The first part involves postal completion of questionnaires. The questionnaires cover your perceptions of your child's ability to engage in everyday tasks, and the effects of muscular dystrophy on family life and parental coping. A pre-paid envelope will be provided for return of questionnaires. Return of questionnaires is requested within two weeks, in order to ensure the study remains within schedule.

The second part involves asking a small sample of fathers if they would be willing to be interviewed about experiences, and support needs, in parenting a son with Duchenne. The interview, which will take approximately 45 minutes, will take place at a location convenient to you.

Participation in this research is completely **voluntary**. Any information you tell the researcher will be treated in strict confidence and used only for the purposes of this research.

**Will my taking part in this study be kept confidential?**

If you consent to taking part, all information collected during the course of the study, will be kept **strictly confidential**.

The information from the interview will be taped and transcribed, that from questionnaires coded: it will then be held on a computer. You and your child cannot be identified from this information, as you are identified by a number only.

**What will happen to the results of the study?**

The results will be written up in a research paper, which will be presented for publication in journals for health care professionals such as the British Journal of Health Psychology.

The study will take place over a 6-month period, so results are likely to be published in 2008. The findings of the research will be presented at conferences, and workshops for families and health professionals involved in the care of children with Duchenne.

No individuals or families will be identified in the published work.

**What do I do now?**

If you wish to take part in the study, please let me know and I will be happy to answer any questions or concerns you have before asking for your written agreement to take part.

If you would like to contact an independent person, who is aware of the project but not involved in it, you are welcome to contact the course Director: Dr Joyce Willock. Contact details: (0131) 3173610; j.willock@qmuc.ac.uk.

**Contact details:**

**Researcher:** Anna Cunniff.

**Email:** [05008550@student.qmuc.ac.uk](mailto:05008550@student.qmuc.ac.uk)

**Address:** Anna Cunniff (Student: Year 2, Doctor of Health Psychology course), Centre for Health Psychology, School of Social Sciences, Media and Communication, Queen Margaret University, Clerwood Terrace, Edinburgh, EH12 8TS.

**Contact details of Supervisor:**

**Supervisor:** Dr Vivienne Chisholm (Senior Lecturer).

**Telephone:** (0131) 317 3613 (answer machine)

**Email:** [v.chisolm@qmuc.ac.uk](mailto:v.chisolm@qmuc.ac.uk)

**Address:** as above.

Many thanks for taking the time to help with this important study.

## **Appendix 2**



Queen Margaret University College  
EDINBURGH

### **PARTICIPANT CONSENT FORM**

#### **AGREEMENT TO PARTICIPATE IN RESEARCH PROJECT**

I (name) \_\_\_\_\_

Of (address) \_\_\_\_\_

#### **Agree to take part in the research study:**

##### **‘Experiences and views of fathers of a child with Duchenne muscular dystrophy’**

I confirm that the nature of the demands of the research have been explained to me (information sheet) and I understand and accept them. I understand that my consent is entirely voluntary, and that I may withdraw from the research project if I find I am unable to continue for any reason.

I have read and understood the information sheet and this consent form. I have had an opportunity to ask questions about my participation.

I agree to participate in this study.

**Signed:** \_\_\_\_\_ **Print Name:** \_\_\_\_\_

**Date:** \_\_\_\_\_

#### **Investigator’s Statement:**

I have explained the nature, and demands of the above research to the participant:

**Signature:** \_\_\_\_\_ **Date:** \_\_\_\_\_

**Anna L. Cunniff**

Postgraduate Student, Centre for Health Psychology, School of Social Sciences, Media and Communication.

Queen Margaret University, Clerwood Terrace, Edinburgh, EH12 8TS.

**Email:** [05008550@student.qmuc.ac.uk](mailto:05008550@student.qmuc.ac.uk)

## Appendix 3



Queen Margaret University College  
EDINBURGH

### Debriefing sheet for participants

#### **Experiences and views of fathers of a child with Duchenne muscular dystrophy.**

Thank you for participating in this study.

One of the main aims in this study is to examine the experiences and views of fathers of a child with Duchenne. Understanding the views of specific family members can give a deeper understanding of issues faced by parents and thereby enable healthcare professionals to provide support that more effectively meets the needs of families who care for a young child with DMD.

The reason for including only fathers in this study is because there are very few studies investigating parents' experiences of caring for a child with Duchenne, and available studies tend to focus on mothers as the main caregiver. As a result our understanding of the impact of Duchenne on families from both perspectives is quite limited.

Your contribution to this study is therefore extremely important and greatly appreciated. Your responses will be used to help answer the questions of what challenges parents (from the fathers' point of view) face, and how health professionals can work to improve this.

This study will also contribute towards raising the profile of neuromuscular conditions amongst different health professionals, and hopefully promote further work to improve services and support for families.

If you feel you would like information on support or services available, the following table summarises relevant organisations:

<b>Name of organisation</b>	<b>Area of support/ information provided</b>	<b>Contact details</b>
<b>Muscular Dystrophy Campaign</b>	Awareness raising; support to parents via care advisors/ range of information; genetic research	<u>Headquarters:</u> 7-11 Prescott Place, London, SW4 6BS <u>Tel:</u> 020 7720 8055 <u>Email:</u> <a href="mailto:info@muscular-dystrophy.org">info@muscular-dystrophy.org</a> <a href="http://www.muscular-dystrophy.org">www.muscular-dystrophy.org</a> <u>Scottish Branch:</u> PO Box 14813, Bonnybridge, FK4 2YD. <u>Tel:</u> 01324 810958.
<b>Contact a family Scotland</b>	Support and wide range of information for families with disabled children.  Area for fathers on website.	<u>Contact a family Scotland:</u> Norton Park, 57 Albion road, Edinburgh, EH7 5QY. <u>Tel:</u> 0131 4752608 <u>Email:</u> <a href="mailto:scotland.office@cafamily.org.uk">scotland.office@cafamily.org.uk</a> <u>Web:</u> <a href="http://www.cafamily.org.uk/scotland">www.cafamily.org.uk/scotland</a>



**Parent Project  
UK/ Scotland**

Family support and awareness raising organisation specifically aimed at parents of a child with Duchenne.

PPUK:  
Epicentre  
41 West Street  
London  
E11 4LJ  
Tel: 02085569955  
Email: [info@ppuk.org](mailto:info@ppuk.org)  
PPUK (Scotland):  
Email:  
[sarahfidelo@yahoo.co.uk](mailto:sarahfidelo@yahoo.co.uk)

Thank you again for participating and helping with this important study.

If you would like more information, or have any further questions about any aspect of this study, then please feel free to contact Anna Cunniff.

**Email:** [05008550@student.qmuc.ac.uk](mailto:05008550@student.qmuc.ac.uk)

**Address:** Anna Cunniff (Student: Year 2, Doctor of Health Psychology course), Centre for Health Psychology, School of Social Sciences, Media and Communication, Queen Margaret University, Clerwood Terrace, Edinburgh, EH12 8TS.

## Appendix 4



Queen Margaret University College  
EDINBURGH

### SEMI-STRUCTURED INTERVIEW GUIDE

*Date:*    /    /  
*Time at start:*    :

*The purpose of this study is to explore the views and experiences fathers of a child with Duchenne Muscular Dystrophy. This will include general information about your role in caring for your child, how you cope and experiences with services.*

*The interview will take about 60 minutes. Any information you tell the researcher will be treated in strict confidence and used only for the purpose of this research.*

*The information from the interview will be audio-taped and transcribed, that from the questionnaires coded: it will then be held in a computer. You and your child cannot be identified from this information as you are identified by a number only.*

*(For all questions participants will be asked to explain their answers)*

- Start with broad question- how would you describe your experience of parenting a child with DMD? Any particular areas you feel are important to talk about?

#### **1. DIAGNOSIS OF DUCHENNE**

*I would like to ask you some questions about your child's condition.*

- How old was your child when the diagnosis was made?
- Can you explain your initial feelings on learning diagnosis?
- How much support did you have at that time? From?
- How did the diagnosis affect your child/family? (gender differences)
- What has helped you cope? **Particular ways of coping?** Recommendations to others?

#### **2. TREATMENT**

*I would like to ask some questions about the treatment that your child has had or is having at present.*

- Can you tell me about how you have dealt with progression of DMD.
- Do you see the condition as being in 'stages'? Did you have to make any treatment decisions? How make these?
- Does anything **help you/ child cope** with the treatment?

- Did anything make coping harder/ easier?

### **3. SERVICES and SUPPORT:**

- What is your overall view of services for your child/ family/ you?
- Do you think parents/ fathers' needs are important/considered?
- Thinking about **services**, what are the main areas parents/ fathers need help with?
- Do you feel fathers are acknowledged by professionals, as having a valid role?
- What could be done to improve, if necessary?
- Can you tell me about the type of **support** you received initially and are currently receiving?
- How did you feel about the **support** received?
- What kind of **support/information** would you find **more helpful**? What could be done differently?
- What has **been most useful** for you in terms of support/ help?
- Do you think the **support needs of parents differ?** Why?
- On reflection, how would you summarise the support available to families?

### **4. HELP**

- Differ between **mother/father with kinds of help preferred?**
- Are there any other areas where you feel fathers/ family need help?
- How could the needs of fathers be met?

### **5. INVOLVEMENT:**

- Since diagnosis, how would you describe your involvement in your child's care?
- Has this varied with different stages or how his health was?
- Is involvement important?
- Have you ever felt isolated or not as involved as liked? Why?

### **6. COPING**

- How has DMD **affected you/ impact on family, emotionally, practically?** (**Gender differences**)?
- How did your **child's ability to cope** affect you / as a family?
- How would you describe your role in the family?
- Overall, how would you describe how you have coped with/ adjusted to (DMD; treatment).

- Can you talk to your son about any concerns?
- Overall, what would you say has been the most challenging time for you?

#### **7. GENERAL**

- If you could make one main change within the health care/ support system what would it be?
- Opinion this type of research?
- Finally, any other comments/ areas I have not mentioned that you would like to talk about?

*Time at end of interview:    :*

**Appendix 5**



Queen Margaret University

EDINBURGH

**'Comments Sheet'**

**Your views and experiences**

1. From your perspective, do you feel that your experience as a father caring for your child differs from that of mothers? If so, how?

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2. As a father, do you feel your needs (as a parent in caring for your child) have been met by family/ professional/ other? How have these needs been met?

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3. Do you feel fathers are acknowledged by professionals, as having a valid role? What could be done to improve, if necessary?

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4. What are the main areas that fathers (and families) might benefit from help with?

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5. How could the needs of fathers (and families) be met? Your recommendations?

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6. Do you think fathers respond differently to other family members, in terms of needs, coping etc?

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7. How did your child's diagnosis *affect you*? What would you say has been the most challenging time for you?

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8. Overall, how would you describe how you have coped with/ adjusted to your child's diagnosis and treatment)?

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9. Do you have any other comments? (This could be your views about the research, issues you feel are important or any general points you feel are relevant for researchers or health care providers).

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10. On reflection, how would you summarise the support available to fathers?

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11. How satisfied do you feel about the *support* given to **you** since your child's diagnosis, from:

<b><u>Hospital staff</u></b>	<i>Poor 0 ---1---2---3---4---5 Good</i>
<b><u>Family</u></b>	<i>Poor 0 ---1---2---3---4---5 Good</i>
<b><u>Friends</u></b>	<i>Poor 0 ---1---2---3---4---5 Good</i>
<b><u>Other support (state)</u></b>	<i>Poor 0 ---1---2---3---4---5 Good</i>

## Appendix 6

### Flyer designed for advertising at Scottish Muscle Network meeting

 <p>Queen Margaret University College EDINBURGH</p>	<p><u><a href="#">Why are you doing the study?</a></u></p> <ul style="list-style-type: none"><li>• the impact of neuromuscular disorders- in particular Duchenne, is under-researched.</li><li>• very few studies have investigated parents' experiences of caring for a child with Duchenne.</li><li>• available studies tend to focus on mothers as the main caregiver.</li><li>• as a result our understanding of the impact of Duchenne on families is quite limited.</li><li>• this study aims to investigate the fathers' experience.</li></ul> <p><u><a href="#">What will I be asked to do?</a></u></p> <ol style="list-style-type: none"><li>1) Complete 4 short questionnaires and briefly write down your opinion (by post).</li><li>2) If you agree - an interview to ask your views.</li></ol> <p><u><a href="#">Can I complete the questionnaires only?</a></u> Yes, any help is valuable for the study and much appreciated.</p>
<p><b><u>Research Project</u></b></p> <p>'Views and experiences of fathers of a child with Duchenne muscular dystrophy'</p>	
<p><u>Please contact: Anna Cunniff</u></p> <p><u>Email: <a href="mailto:05008550@student.qmuc.ac.uk">05008550@student.qmuc.ac.uk</a></u></p> <p><b>Address:</b> Centre for Health Psychology, School of Social Sciences, Media and Communication, Queen Margaret University, Clerwood Terrace, Edinburgh, EH12 8TS.</p>	

## Appendix 7

### General Health Questionnaire

For each item, please mark the answer that has applied to **you** over the **past few weeks**. Please answer **all** the questions.

Have you recently?.....

Question	Please mark one response for each question			
<b>Been able to concentrate on what you're doing?</b>	Better than usual	Same as usual	Less than usual	Much less than usual
<b>Lost much sleep over worry?</b>	Not at all	No more than usual	Rather more than usual	Much more than usual
<b>Felt you were playing a useful part in things?</b>	More so than usual	Same as usual	Less useful than usual	Much less useful
<b>Felt capable of making decisions about things?</b>	More so than usual	Same as usual	Less so than usual	Much less capable
<b>Felt constantly under strain?</b>	Not at all	No more than usual	Rather more than usual	Much more than usual
<b>Felt you couldn't overcome your difficulties?</b>	Not at all	No more than usual	Rather more than usual	Much more than usual
<b>Been able to enjoy your normal day-to-day activities?</b>	More so than usual	Same as usual	Less so than usual	Much less than usual
<b>Been able to face up to your problems?</b>	More so than usual	Same as usual	Less so than usual	Much less able
<b>Been feeling unhappy and depressed?</b>	Not at all	No more than usual	Rather more than usual	Much more than usual
<b>Been losing confidence in yourself?</b>	Not at all	No more than usual	Rather more than usual	Much more than usual
<b>Been thinking of yourself as a worthless person?</b>	Not at all	No more than usual	Rather more than usual	Much more than usual
<b>Been feeling reasonably happy, all things considered?</b>	More so than usual	About same as usual	Less so than usual	Much less than usual



## Appendix 8

### Functional Disability Inventory

In the **past 2 weeks**, how has your child coped **physically** with the following practical activities?

<b>Walking to the bathroom.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Walking up stairs.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Doing something with a friend (for example playing a game).</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Doing chores at home.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Eating regular meals.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Being up all day without a nap or a rest.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Riding the school bus or travelling in the car.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Being at school all day.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Doing activities in gym class (or playing sports).</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Reading or doing homework.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Watching TV.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Walking the length of a football field.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Running the length of a football field.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Going shopping.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
<b>Getting to sleep at night and staying asleep.</b>	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible

## Appendix 9

### Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. Please try to answer all items even if you are not certain or if the question seems silly! Please answer on the basis of your child's behaviour over the ***last 6 months*** or this school year.

Age of child: \_\_\_\_\_

**Not  
True**

**Somewhat  
True**

**Certainly**

Considerate of other's feelings

Restless, overactive, cannot stay still for long

Often complains of headaches, stomach aches or sickness

Shares readily with other children (treats, toys, pencils etc)

Often has temper tantrums or hot tempers

Rather solitary, tends to play alone

Generally obedient, usually does what adults request

Many worries, often seems worried

Helpful if someone is hurt, upset or feeling ill

Constantly fidgeting or squirming

Has at least one good friend

Often fights with other children or bullies them

Often unhappy, downhearted or tearful

Generally liked by other children

Easily distracted, concentration wanders

Nervous or clingy in new situations, easily loses confidence

Kind to younger children

Often lies or cheats  
Picked on or bullied by other children

Often volunteers to help others  
(parents, teachers, other children)

Thinks things out before acting

Steals from home, school or elsewhere

Gets on better with adults than other children

Many fears, easily scared

Sees tasks through to the end,  
good attention span

**Do you have any other comments or concerns that you feel are relevant to the placement?**

---

---

Overall, do you think your child has difficulties in one or more of the following areas: emotions, concentration, behaviour or being able to get on with other people?

No ; Yes, minor difficulties ; Yes, definite difficulties; Yes, severe

**If you answered 'yes', please answer the following questions about these difficulties:**

- **How long have these difficulties been present?**

Less than a month ; 1-5 months ; 5-12 months ; over a year

- **Do the difficulties upset or distress your child?**

Not at all ; Only a little ; Quite a lot ; A great deal

- **Do the difficulties interfere with your child's everyday life in the following areas?**

**Not at all      Only a little      Quite a lot      Great deal**

HOMELIFE

FRIENDSHIPS

CLASSROOM LEARNING

LEISURE ACTIVITIES

- **Do the difficulties put a burden on you or the family as a whole?**

**Not at all      Only a little      Quite a lot      Great deal**

## Appendix 10

### Dads' Involvement Scale

This scale measures how much you are involved in tasks relating to your child's medical condition and how your involvement affects your family's coping with Duchenne and its treatment. After reading each item, please think about how many times that task was needed in the past 6 months. Then, rate how much you have done that behaviour when it was needed and how your level of involvement has affected/ helped your family. Please put a check mark next to the answer that best matches your view of each statement.

*It is important that you try to respond to every task below. If there was absolutely no need for the task described in an item within the past 6 months, please write 'N/A'.*

<b>TASK</b>	<b>AMOUNT</b> (When it was needed, how much have you done this in the past 6 months?)					<b>HELPFULNESS</b> (Has this made family coping with Duchenne harder or easier?)				
	<i>1 = hardly ever; 2 = sometimes; 3 = often; 4 = very often; 5 = always.</i>					<i>1 = harder; 2 = neither harder nor easier; 3 = slightly easier; 4 = easier; 5 = much easier.</i>				
Check to see if there is enough medication and other supplies; call clinic to request repeat prescriptions.	1	2	3	4	5	1	2	3	4	5
Pick up prescriptions.	1	2	3	4	5	1	2	3	4	5
Administer medication to child at prescribed times.	1	2	3	4	5	1	2	3	4	5
Prepare supplies or equipment for required medical procedures.	1	2	3	4	5	1	2	3	4	5
Make medical appointments.	1	2	3	4	5	1	2	3	4	5
Come to child's medical appointments, hospital appointments.	1	2	3	4	5	1	2	3	4	5
Talk with teachers, and other caregivers to help them understand your child's condition and its treatment.	1	2	3	4	5	1	2	3	4	5
Talk to health professionals about child's symptoms.	1	2	3	4	5	1	2	3	4	5
Recognise and respond appropriately to child's symptoms that require attention.	1	2	3	4	5	1	2	3	4	5
Share leisure activities with your child or supervise these activities.	1	2	3	4	5	1	2	3	4	5
Remind your child or yourself when it is time to take medication or perform other tasks related to the medical condition.	1	2	3	4	5	1	2	3	4	5

Perform or supervise required medical monitoring (e.g. splints/ KAFOS).	1	2	3	4	5	1	2	3	4	5
Pay medical bills or straighten out related problems.	1	2	3	4	5	1	2	3	4	5
Gather information about your child's medical condition and share it with your family.	1	2	3	4	5	1	2	3	4	5
Attend a support group or educational workshop about your child's condition.	1	2	3	4	5	1	2	3	4	5
Talk with your child to understand how the condition affects him socially or emotionally.	1	2	3	4	5	1	2	3	4	5
Talk about how the medical condition affects you or your child socially or emotionally.	1	2	3	4	5	1	2	3	4	5
Reward or praise your child for co-operating with treatment.	1	2	3	4	5	1	2	3	4	5
Discipline your child for poor co-operation with treatment.	1	2	3	4	5	1	2	3	4	5
Help relatives, neighbours, friends, or other children to understand your child's medical condition and its treatment.	1	2	3	4	5	1	2	3	4	5
Take over other household tasks to give you more time to attend to the medical condition.	1	2	3	4	5	1	2	3	4	5
Give up sleep if your child's condition requires it.	1	2	3	4	5	1	2	3	4	5
Stay home from work if necessary when your child is sick.	1	2	3	4	5	1	2	3	4	5
Take care of your child so that you can go out for recreation.	1	2	3	4	5	1	2	3	4	5

## **Appendix 11**

### **Interpretation guidelines for questionnaires**

#### **G.H.Q. Interpretation**

G.H.Q. comprises items relating to symptoms and behaviours, asking if respondents have experienced these recently. Areas such as concentration, loss of sleep and feeling unhappy are rated, with response options: not at all, no more than usual, rather more than usual, and much more than usual.

Total scores for G.H.Q.-12, range from 0-12, with items scored according to the bimodal system, 0-0-1-1 known as the 'G.H.Q. score'. Scores indicate severity of psychological disturbance on a continuum, with higher scores indicating greater perceived dysfunction.

#### **F.D.I. Interpretation**

Covering the previous 2-week period, 15 items are rated in terms of ability to carry out physical activities. The F.D.I. includes categories of sleep and rest (6,15), eating (5), home management (4), school (8,10), ambulation (1,2,12,13), mobility (7), and social interaction and recreation (3,9,11,14).

Items relating to physical difficulties with a range of tasks are rated from 0-4, 'no trouble', 'a little trouble', 'some trouble', 'a lot of trouble' and 'impossible'. Scores are (0) no trouble to (4) impossible, giving a maximum score of 60 (0-60).

Whilst there is no set cut-off point, higher scores indicate higher impairment and physical limitation.

### **S.D.Q. Interpretation**

Cut-off points allow identification of scores within each sub-scale as, 'normal', 'borderline' and 'abnormal'. Combined overall scores from sub-scales (excluding pro-social, which gives a 'stand alone' score), present a total difficulties score reflecting the extent of emotional and behavioural symptoms. Cut-off scores identify possible 'symptom caseness' (see section 4.5.4. in thesis).

A separate 'impact score' may also be obtained, with cut off 2+ indicating significant impact relating to chronicity, distress to child and burden on family.

### **D.A.D.S. Interpretation**

'Amount' items question how much specific tasks were carried out in the past 6 months. 'Helpfulness' items ask whether this made family coping harder or easier. For 'amount' items, responses options are, 1 (0%), 2 (25%), 3 (50%), 4 (75%), and 5 (100%). For 'helpfulness' items, responses are, 1 (harder), 2 (neither harder nor easier), 3 (slightly easier), 4 (easier) and 5 (much easier). Following guidelines (Gavin and Wysocki, 2004), where a task was not needed, a mean score for 'amount' and a score of 2 (neither harder nor easier) is recorded for 'helpfulness'.

Normative D.A.D.S. data are not available for DMD, however, in a study exploring psychometric properties of the measure, 224 parents of children with various chronic conditions completed D.A.D.S. (Gavin and Wysocki, 2004).

## Appendix 12

**Table 14:** *Participant characteristics (quantitative study)*

N <sup>55</sup>	Age of father	Age of child	Fathers Occupation	Residence	National Statistics Socio-Economic Classification
<b>1</b>	63	33	Computer engineer	England	HM
<b>2</b>	39	13	Mechanical engineer	England	HM
<b>3</b>	40	6	Teacher	England	HM
<b>4</b>	46	12	Medicine	England	HM
<b>5</b>	52	13	Retired fire officer	England	LP
<b>6</b>	45	8	Banking	N. Ireland	HM
<b>7</b>	51	15	Finance	England	HM
<b>8</b>	42	4	Snr. Manager Education	Isle of Man	HM
<b>9</b>	46	12	IT	Scotland	HM
<b>10</b>	Missing	22	Missing	England	-
<b>11</b>	Missing	25	Missing	England	-
<b>12</b>	Missing	17	Software sales	England	-
<b>13</b>	40	6	IT	England	LP
<b>14</b>	43	6	Missing	England	-
<b>15</b>	40	9	Health and Safety Officer	England	LP
<b>16</b>	49	18	Unemployed civil servant	South Wales	U
<b>17</b>	35	8	Finance	England	HM
<b>18</b>	44	10	Cattery owner ex IT banking	England	HM
<b>19</b>	50	23	Retail in IT	England	-
<b>20</b>	39	6	Missing	England	-
<b>21</b>	38	8	HGV driver	South Wales	SR
<b>22</b>	Missing	Missing	Missing	England	-
<b>23</b>	51	8	Graphic designer	England	HM
<b>24</b>	49	15	GP	England	HM
<b>25</b>	46	14	Missing	England	-
<b>26</b>	35	3	Missing	England	-
<b>27</b>	40	10	Royal mail	England	I
<b>28</b>	34	7	IT	England	LP
<b>29</b>	60	27	Retired postmaster	England	I
<b>30</b>	40	11	Missing	England	-
<b>31</b>	46	15	Finance	England	HM
<b>32</b>	Missing	Missing	Missing	England	-
<b>33</b>	51	12	Operations director	England	HM
<b>34</b>	47	14	GP	England	HM
<b>35</b>	49	17	Unemployed civil servant	England	U
<b>36</b>	57	21	Missing	Scotland	-
<b>37</b>	57	18	Carer	England	SR
<b>38</b>	51	13	Social worker	Scotland	LP

<sup>55</sup> Chronological number of data sheet entry on SPSS, used for identification purposes



<b>39</b>	46	15	Bus driver	Scotland	SR
<b>40</b>	34	15	Operations management	Scotland	HM
<b>41</b>	43	15	Manager airport operator	Scotland	HM
<b>42</b>	40	9	Gardner	England	I
<b>43</b>	60	32	Missing	England	-
<b>44</b>	63	25	Accountant	Scotland	HM
<b>45</b>	46	8	Housing	Scotland	I
<b>46</b>	43	16	Missing	Scotland	-
<b>47</b>	Missing	Missing	Accountant	England	HM
<b>48</b>	50	15	Missing	England	H-
<b>49</b>	45	15	Accountant	England	HM
<b>50</b>	39	8	Teaching and coaching	England	HM
<b>Mean</b>	46 (34-63)	14.1 (3-33)			

HR= higher managerial/professional: Social Classes I and II  
LP= lower professional/ higher technical;  
SR= semi routine; I= intermediate: clerical, sales, technical  
U= unemployed.

## Appendix 13

**Table 15:** *Individual scores on all questionnaire items*

N <sup>56</sup>	Age of father	Age of child	SDQ	Impact on family	FDI	GHQ	DADS	Agree to interview	Written account received
1	63	33	N/a	N/a	45	N	Help:2.6 Amount: 4.1	Y	Y
2	39	13	E= A C=A H=A PP= A P= N Total= A	A	38	A	Help:3.1 Amount: 2.5	Y	Y
3	40	6	E=N C=B H=N PP=N P=N Total=N	N	4	N	Help:2.0 Amount: 2.2	Not stated	Y
4	46	12	E=N C=N H=N PP=A P=N Total=N	A	30	N	Help:mis sing Amount: 1.5	N (medicin e)	Y
5	52	13	E=A C=N H=N PP=B P=N Total=B	A	36	A	Help:1.3 Amount: 3.6	Y	Y
6	45	8	E=N C=N H=N PP=N P=N Total=N	B	16	N	Help:2.7 Amount: 2.7	Y	Y
7	51	15	E=N C=N H=N PP=N P=N Total=N	A	28	N	Help:2.3 Amount: 3.1	Y	Y
8	42	4	E=N C=N H=N PP=N P=A Total=N	N	16	N	Help:3.1 Amount: 2.9	Y	Y
9	46	12	E=N C=N H=B PP=B P=N Total=N	A	32	N	Help:1.7 Amount: 2.4	Y	Y
10	Missing	22	N/a	N/a	39	N	Help:1.8 Amount: 3.1	Y	Y
11	Missing	25	N/a	N/a	39	N	Help:4.2 Amount: 2.5	Not stated	Y
12	Missing	17	E=N C=N	N	31	N	Help:2.0 Amount:	Not stated	Y

<sup>56</sup> Chronological number of data sheet entry on SPSS, used for identification purposes

			H=N PP=N P=N Total=N				2.2		
13	40	6	E=N C=N H=N PP=N P=N Total=N	missing	14	N	Help:2.9 Amount: 2.1	Y	Y
14	43	6	E=B C=A H=N PP=N P=N Total=N	N	3	A	Help:1.1 Amount: 4.6	Y	Y
15	40	9	E=N C=A H=N PP=N P=N Total=B	A	27	N	Help:3.8 Amount: 3.6	Y	Y
16	49	18	E=A C=N H=A PP=A P=N Total=A	A	35	A	Help:2.9 Amount: 3.6	Y	Y
17	35	8	E=N C=N H=N PP=B P=N Total=N	N	23	A	Help:1.4 Amount: 3.2	Y	Y
18	44	10	E=N C=N H=N PP=B P=N Total=N	A	40	A	Help:2.6 Amount: 2.8	Y	Y
19	50	23	N/a	N/a	57	N	Help:2.3 Amount: 1.8	Y	Y
20	39	6	E=B C=N H=A PP=A P=N Total=N	A	21	N	Help:2.9 Amount: 2.4	Y	Y
21	38	8	E=N C=A H=N PP=N P=N Total=N	B	15	A	Help:3.0 Amount: 3.3	Y	Y
22	Missing	Missing	E=A C=B H=N PP=N P=N Total=A	N	28	N	Help:2.9 Amount: 2.3	Not stated	Y
23	51	8	E=N C=N H=N PP=N P=N Total=N	N	17	A	Help:1.5 Amount: 4.5	Y	Y
24	49	15	E=N C=N H=N	N	35	N	Help:3.0 Amount: 2.7	Y	Y

25	46	14	PP=N P=N Total=N E=N C=B H=B PP=B P=N Total=N	B	38	N	Help:2.2 Amount: 1.8	Y	Y
26	35	3	Total=N E=N C=B H=N PP=N P=N Total=N	N	18	N	Help:2.5 Amount: 2.3	Y	Y
27	40	10	E=A C=A H=A PP=A P=N Total=A	A	36	N	Help:3.5 Amount: 2.5	Y	Y
28	34	7	E=N C=N H=N PP=N P=N Total=N	N	9	N	Help:2.2 Amount: 3.3	Y	Y
29	60	27	N/a	N/a	37	A	Help:1.6 Amount: 3.3	Y	Y
30	40	11	E=A C=A H=A PP=A P=N Total=A	A	33	A	Help:1.6 Amount: 4.1	Y	Y
31	46	15	E=B C=N H=N PP=A P=N Total=N	B	34	N	Help:2.5 Amount: 1.8	Y	Y
32	Missing	Missing	E=A C=N H=N PP=N P=N Total=B	N	28	N	Help:2.4 Amount: 1.7	Not stated	Y
33	51	12	E=A C=N H=N PP=B P=N Total=N	N	44	A	Help:1.5 Amount: 2.9	Y	Y
34	47	14	E=N C=N H=N PP=B P=N Total=N	N	37	N	Help:2.1 Amount: 2.5	N (GP)	Y
35	49	17	E= C= H=A PP=B P=A Total=	N	31	N	Help:2.0 Amount: 3.2	Y	Y
36	57	21	N/a	N/a	22	A	Help:1.7 Amount: 3.5	Y	N

37	57	18	E=n C=N H=n PP=B P=N Total=N	N	44	N	Help:3.5 Amount: 3.1	Y	Y
38	51	13	E=N C=N H=N PP=N P=N Total=N	N	30	N	Help:3.6 Amount: 2.8	Y	Y
39	46	15	E=A C= H= PP= P= Total=	B	47	N	Help:2.6 Amount: 2.5	Y	N
40	34	15	E=A C= H= PP=A P= Total=	A	33	A	Help:2.5 Amount: 2.3	Y	Y
41	43	15	E=N C=N H=N PP=N P=N Total=N	N	33	A	Help:2.2 Amount: 1.8	Y	Y
42	40	9	E=B C=A H=A PP=A P=A Total=A	A	31	A	Help:1.9 Amount: 2.6	Not stated	Y
43	60	32	N/a	N/a	37	N	Help:3.3 Amount: 2.9	Y	Y
44	63	25	N/a	N/a	45	N	Help:1.9 Amount: 2.4	Y	Y
45	46	8	E=N C=B H=N PP=A P=N Total=N	A	12	A	Help:2.8 Amount: 3.1	Y	Y
46	43	16	E=N C=N H=N PP=A P=N Total=N	A	34	N	Help:2.8 Amount: 2.7	Y	Y
47	Missing	24	N/a	Missing	44	N	Help:2.4 Amount: 2.6	Not stated	Y
48	50	15	E=N C=N H=A PP=N P=N Total=N	N	28	N	Help:mis sing Amount: 2.4	Y	Y
49	45	15	E=N C=N H=N PP=N P=N Total=N	N	33	N	Help:2.2 Amount: 2.1	Y	Y

50	39	8	E=N C=N H=N PP=B P=N Total=N	N	8	N	Help:2.3 Amount: 2.1	Not stated	Y
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Total number with child aged up to 18 years: n=41

Total number with child aged over 18 years: n=9

### **SDQ**

**E**= emotions

**C**= conduct

**H**= hyperactivity

**PP**= peer problems

**P**= prosocial

**Total** = total SDQ score range N= normal; B= borderline; A= abnormal

### **DADS**

**Help** = evaluation of whether performance of the task made illness management easier or harder

### **Interviews**

Refused: n=2

No preference stated: n=8

Agreed: n=40

### **Written accounts received**

48 of 50

## Appendix 14

### Tables 16a-16k. Group total and sub-scale scores for each questionnaire

#### 1. Functional Disability Inventory

**Table 16a:** *Sub-scale and total scores on the F.D.I.*

*Mean F.D.I. scores (n=50)*

Activity	Min	Max	Mean (s.d.)
Walk to bathroom	1	5	3.98 (1.5)
Walk up stairs	2	5	4.36 (1.1)
Activity with friend	1	5	3.04 (1.3)
Chores at home	1	5	4.1 (1.3)
Eat regular meals	1	5	2.36 (1.4)
Up all day without a nap	1	5	1.62 (1.2)
Ride school bus or travel in car	1	5	2.38 (1.5)
Attend school all day	1	5	1.80 (1.3)
Do sports	1	5	3.98 (1.2)
Read/ do homework	1	5	2.5 (1.3)
Watch TV	1	5	1.20 (.67)
Walk length of football field	1	5	4.16 (1.4)
Go shopping	2	5	2.98 (1.3)
Go to sleep and stay asleep all night	1	5	2.16 (1.2)
<b>Total FDI score</b>	<b>3</b>	<b>57</b>	<b>29.7 (11.7)</b>

Table 16b below outlines the percentages, and collapsed scores,<sup>57</sup> for children having difficulties with tasks in each area.

**Table 16b:** *Level of functional ability (%) (n=50)*

Activity	No trouble	A little trouble	Some trouble	A lot of trouble	Impossible
Walk to bathroom	14% (n:7)	8% (n:4)	10% (n:5)	2% (n:1)	66% (n:33)
Walk up stairs	0% (n:0)	14% (n:7)	6% (n:3)	10% (n:5)	70% (n:35)
Activity with friend	10% (n:5)	26% (n:13)	27% (n:14)	14% (n:7)	18% (n:9)
Chores at home	6% (n:3)	8% (n:4)	12% (n:6)	20% (n:10)	50% (n:25)
Eat regular meals	40% (n:20)	18% (n:9)	22% (n:11)	6% (n:3)	14% (n:7)
Up all day without a nap	68% (n:34)	18% (n:9)	6% (n:3)	0% (n:0)	8% (n:4)

<sup>57</sup> 'Collapsed' scores were calculated by the author to create total percentages for those having trouble in different areas. To do this 'no/a little/ some trouble' and 'a lot/impossible' percentages were totalled in the column below percentages for each response.

Ride school bus or travel in car	44% (n:22)	24% (n:12) 12% (n:6)	20% (n:10)	10% (n: 5)	8% (n:4) 14% (n: 7)
Attend school all day	60% (n:30)	32% (n:16) 12% (n:6)	6% (n:3)	6% (n:3)	24% (n:12) 8% (n:4)
Do sports	6% (n:3)	18% (n:9) 4% (n:2)	22% (n:11)	20% (n:10)	14% (n:7) 46% (n:23)
Read/ do homework	30% (n: 15)	26% (n:13) 22% (n:11)	20% (n:10)	14% (n:7)	66% (n:33) 8% (n:4)
Watch TV	88% (n: 44)	44% (n:21) 8% (n:4)	2% (n:1)	0% (n:0)	22% (n:11) 2% (n:1)
Walk length of football field	10% (n:5)	10% (n:5) 6% (n:3)	10% (n:5)	6% (n:3)	2% (n:1) 68% (n:34)
Go shopping	14% (n:7)	16% (n:8) 20% (n:10)	38% (n:19)	10% (n:5)	74% (n:37) 18% (n:9)
Go to sleep and stay asleep all night	36% (n:18)	58% (n:29) 30% (n:15)	18% (n:9)	14% (n:7)	28% (n:14) 2% (n:1)
		48% (n:24)			16% (n:8)

## 2. General Health Questionnaire

Table 16c presents a summary of G.H.Q. scores.

**Table 16c:** *Sub-scale and total scores on the G.H.Q. (n=50)*

Variable	Min	Max	Mean (sd)
Been able to concentrate	1	4	2.4 (.66)
Lost much sleep	1	4	2.3 (.78)
Felt playing useful part in things	1	3	1.9 (.53)
Felt capable of making decisions about things	1	3	2.1 (.47)
Felt constantly under strain	2	4	2.6 (.81)
Felt couldn't overcome difficulties	1	4	2.1 (.81)
Been able to enjoy day to day activities	1	4	2.5 (.76)
Been able to face problems	1	4	2.4 (.63)
Been feeling unhappy or depressed	1	4	2.4 (.97)
Been losing self confidence	1	4	1.9 (.90)
Been thinking of yourself as worthless	1	4	1.6 (.81)



Been feeling reasonably happy all things considered	2	4	2.4 (.67)
Total GHQ score	0	12	3.5 (3.8)

### **3. Dads Active Disease Support Scale**

Tables 16d and 16e present summaries of D.A.D.S. scores:

**Table 16d:** *Sub-scale and total scores on the D.A.D.S. amount of involvement*

*(n=50)*

Variable	Min	Max	Mean (sd)
When needed, how much have you...			
Checked to see if enough medication/supplies; call clinic to request repeat prescriptions?	1	5	2.4 (1.6)
Picked up prescriptions	1	5	2.4 (1.5)
Administered medication at prescribed times	1	5	2.9 (1.4)
Prepared supplies or equipment for required medical procedures	1	5	2.5 (1.3)
Made medical appointments	1	5	2.3 (1.5)
Attended child's medical or hospital appointments	1	5	3.9 (1.3)
Talked with teachers or other carers to help them understand your child's condition and treatment	1	5	3.3 (1.3)
Talked to health professionals about your child's symptoms	1	5	3.5 (1.4)
Recognised and responded appropriately to child's symptoms that require attention	1	5	3.7 (1.2)
Shared leisure activities with your child or supervised these activities	1	5	3.6 (1.1)
Reminded yourself or child when it's time to take medication or perform other medical activities	1	5	3.3 (1.4)
Performed or supervised required medical monitoring	1	5	2.8 (1.4)
Paid medical bills or straightened out related problems	1	5	2.3 (1.5)
Gathered information about child's condition and shared it with your family	1	5	2.9 (1.3)
Attended a support group or educational workshop about child's condition	0	5	1.9 (1.5)
Talked with your child to understand how the condition affects him socially or emotionally	1	5	2.7 (1.3)
Rewarded or praised child for co-operating with treatment	1	5	3.7 (1.3)
Disciplined your child for poor co-operation with treatment	1	5	1.7 (1.1)
Helped relatives, neighbours, friends or other children to understand your child's condition and treatment	1	5	2.9 (1.4)
Taken over other household tasks to give you more time to attend the condition	1	5	2.5 (1.5)
Given up sleep if the condition requires it	1	5	3.2 (1.6)
Stayed home from work if necessary when your child is unwell	1	5	2.6 (1.6)
Taken care of your child so that you can go out for recreation	1	5	2.6 (1.3)
Total DADS perceived amount of involvement score	1.5	4.6	2.7 (.71)

**Table 16e:** *Sub-scale and total scores on the D.A.D.S. perceived helpfulness of involvement (n=50)*

Variable	Min	Max	Mean (sd)
Has .....made family coping harder or easier?			
Checking to see if enough medication/supplies; call clinic to request repeat prescriptions?	1	5	2.4 (1.1)
Picking up prescriptions	1	5	2.4 (1.0)
Administering medication at prescribed times	1	5	2.6 (1.1)
Preparing supplies or equipment for required medical procedures	1	5	2.4 (1.1)
Making medical appointments	1	5	2.1 (.98)
Attending child's medical or hospital appointments	1	5	2.9 (1.4)
Talking with teachers or other carers to help them understand your child's condition and treatment	1	5	2.7 (1.3)
Talking to health professionals about your child's symptoms	1	5	2.7 (1.3)
Recognising and responding appropriately to child's symptoms that require attention	1	5	2.8 (1.3)
Sharing leisure activities with your child or supervising these activities	1	5	2.9 (1.3)
Reminding yourself or child when it's time to take medication or perform other medical activities	1	5	2.6 (1.1)
Performing or supervising required medical monitoring	1	5	2.4 (1.1)
Paying medical bills or straightened out related problems	1	4	.2.2 (.81)
Gathering information about child's condition and shared it with your family	1	5	2.4 (1.2)
Attending a support group or educational workshop about child's condition	0	5	2.2 (1.1)
Talking with your child to understand how the condition affects him socially or emotionally	1	5	2.5 (1.1)
Talking about how DMD affects you or your child	1	5	2.1(1.1)
Rewarding or praising child for co-operating with treatment	1	5	2.8 (1.1)
Disciplining your child for poor co-operation with treatment	1	5	2.2 (.88)
Talking to relatives, neighbours, friends or other children to help them understand your child's condition and treatment	1	5	2.5 (1.2)
Taking over other household tasks to give you more time to attend the condition	1	5	2.3 (.97)
Giving up sleep if the condition requires it	1	5	2.3 (1.3)
Staying home from work if necessary when your child is unwell	1	5	2.1 (1.2)
Taking care of your child so that you can go out for recreation	1	5	2.4 (1.1)
Total DADS perceived helpfulness of involvement score	1.1	4.2	2.4 (.68)

#### **4. Strengths and Difficulties Questionnaire**

Tables 16f and g present summaries of S.D.Q. scores.

**Table 16f:** *Sub-scale and total scores on the S.D.Q. (n=41)*

Variable	Min	Max	Mean (sd)
Emotional	0	10	2.9 (2.7)
Conduct	0	6	1.8 (1.8)
Hyperactivity	0	9	4.2 (2.4)
Peers	0	9	2.6 (2.1)
Prosocial	2	12	7.8 (1.9)
Total SDQ score	0	29	11.5 (6.8)

**Table 16g:** *Emotional and behavioural adjustment in children. Numbers and % of boys above cut off for psychiatric risk (n=41)*

Sub scale	Borderline	Abnormal	Mean (s.d.)	Normative UK Mean (s.d.)
Emotions	8% (n:4)	24% (n:10)	2.9 (2.7)	1.9 (2.0)
Peer Problems	24% (n:10)	21% (n:9)	2.6 (2.1)	1.5 (1.7)
Conduct	8% (n:4)	17% (n:7)	1.8 (1.8)	1.6 (1.7)
Prosocial	0% (n:0)	7% (n:3)	7.8 (1.9)	8.6 (1.6)
Hyperactivity	5% (n:2)	19% (n: 8)	4.1 (2.5)	3.5 (2.6)
Total Score	7% (n:3)	15% (n:6)	11.54 (6.8)	8.4 (5.8)

Table 16h presents scores on the S.D.Q. Impact on Family scale

**Table 16h:** *Total scores impact on family (n=41)*

Variable	Min	Max	Mean (s.d.)	Normative UK Mean (s.d.)
Total impact on family total score	0	9	1.9 (2.6)	0.5 (1.2)

Table 16i presents percentages scoring above cut off for Impact on Family scale.

**Table 16i:** *Percentages scoring above cut-off (n=41)*

<b>Normal</b>	<b>Borderline</b>	<b>Abnormal</b>
<b>52% (n:21)</b>	12% (n:10)	36% (n:15)

## **5. Support Scales**

Table 16j presents a summary of Support scores.

**Table 16j:** *Total scores on satisfaction with support scales (n=48)*

<b>Variable</b>	<b>Min</b>	<b>Max</b>	<b>Mean (sd)</b>
<b>Hospital/staff</b>	0	5	3.3 (1.7)
<b>Family</b>	0	5	3.5 (1.5)
<b>Friends</b>	0	5	3.4 (1.4)

Table 16k summarises percentages of satisfaction with support in each area.

**Table 16k:** *Percentages: satisfaction with support (n=48)*

<b>Variable</b>	<b>Poor 0-1</b>	<b>Average 2-3</b>	<b>Good 4-5</b>
<b>Hospital/ staff (n: 48)</b>	27% (n: 13)	16% (n: 8)	56% (n: 27)
<b>Family (n: 49)</b>	23% (n: 11)	18% (n: 9)	59% (n: 29)
<b>Friends (n: 48)</b>	23% (n: 11)	27% (n: 13)	50% (n: 24)

## Appendix 15

### Examples of extracts illustrating development of coding frame for themes

#### 1 and 4

Main theme	Sub-themes	Example quotes
<b>Theme 1: Loss and adjustment</b>	<ul style="list-style-type: none"> <li>• Loss (diagnosis; friends)</li>   <li>• Loss (in light of expectations)</li>   <li>• Adaptive coping and acceptance</li> </ul>	<ul style="list-style-type: none"> <li>• "Your child's diagnosis is a bombshell. It's a sentence of death on your child which you are powerless to change"</li> <li>• "The initial reaction was as if we had suffered a bereavement"</li> <li>• "I haven't got a soul bro. I don't have that anymore. That's what this condition of X's has done. It's made me so protective of my family that outside people who I can't rely on I've dropped because they've done the same to me"</li> <li>• "I suppose for men, it's hard to think their son is not going to fulfil the aspirations. I don't think they can deal with that. It's difficult to deal with but you've still got to deal with it"</li> <li>• "All these expectations we had...it's not gone but it's put a distance between us"</li> <li>• "Knowing he has DMD-the feeling of despair never leaves you. Never giving up hope, wondering how mum, son and brother will cope as time goes on. Most challenging time was at the beginning-thinking things like 'dads are not supposed to outlive their sons"</li> <li>• "We responded by making a decision to give him the best experiences we could. This meant we enjoyed some good times and appreciated something we may not have done with a 'healthy child"</li> <li>• "I like to think I've got a good faith.. it's like something else you've got to believe in and we pray there will be a cure"</li> <li>• "I do what I do with the charity and I'm sure the reason is to stop me thinking about anything else"</li> <li>• "I actually got the advice from a colleague to say 'no-one's to blame'. But when he said that, it was freeing and being able to say 'it's nobody's fault'.. that helped set it and I think the attitude is most important. If you get advice about attitude from the beginning it helps"</li> <li>• "I've adapted my life around it. From coming into work, taking him to school just being there from the minute he sleeps until he wakes up"</li> <li>• "He is so much part of our daily lives we don't feel we are looking after a disabled child"</li> <li>• "If he was a youngster who would grizzle and moan, it would make life extremely difficult. But by and large, he is cheerful most of the time. It helps us cope better I think"</li> <li>• "My son helped me to come to terms with it because he makes us laugh a lot and always wakes up with a smile"</li> <li>• "When we first found out X was 5 and</li> </ul>

**Theme 4: Race against time**

- Maladaptive coping
  - told take his home and watch him "die". Through us finding PPUK this has given us hope, and whatever happens in life you take hope away you have nothing to live for "thank God for PPUK"
  - "I sometimes find myself trying to stay remote from my son, because I don't want to get too attached. You know if you get close to somebody, then something happens you feel worse. I know it's a strange things to say, you know you're frightened of..' .....I sometimes find myself, I don't know, trying to be slightly aloof so I don't get too close... it's not that I don't love my son... I can actually feel it happening at times and I have to overcome it. I find that very upsetting"
  - "It makes you wish you could just hibernate in your own wee world2"
- Images of next stages
  - "We just don't want to see him deteriorate too much. I think we would be happy if he could have a 21<sup>st</sup> birthday party"
  - "They send out some horrific photographs. ..my partner was that and I found her upstairs in the corner crying. But, I says 'we are going to be in for that"
- Transition to adulthood and comparison with other children
  - "Watching the agonising deterioration since I've been 35, whilst two younger brothers grow up past him- truly sad"
  - "The hardest period was when at 16 the hospital could not see X anymore, but gave no indication as to where to go for advice"
  - "Even with the wheelchair, he is still a teenager and wants to do normal stuff. The problem is he can't do it. He will try to do it. That's the hard bit, all the stuff he can't do"
- Deterioration
  - "There will be long periods of very little change and then all of a sudden there will be a very dramatic change"
  - "Now we're getting kids coming into an older stage, hitting 30s. so your mindset is having to change now. There was a time I thought 'I'm going to have to prepare for X dying in the 20 mark, or before that"
  - "The diagnosis totally crushed me. Getting through each day is a huge challenge. Coping with each stage of deterioration is difficult e.g. no longer able to walk, cannot feed himself etc"
- Making the most of life
  - "We're showering him with as many things as we can. Taking him on as many holidays as possible"
  - "Make every day count...you have to count because time is so short that you probably might even sometimes regret not being there for them"
- Decisions
  - "Having to decide yes, no, whatever, that was the hardest time"
  - "It's stressful in case it's wrong, but you've got to make the decisions"

- Talking about death

- "He knows he's going to die. He will ask questions about that, he's not afraid to. Fortunately for me it's his mother he asks more than me"
- "You would have thoughts like I wonder how he will die, and how I'll be when it happens, and will it be one of those deaths where I can encourage him to let go if he needs"
- "The other problem I avoid basically is.. dying. I just wouldn't know what to say. I'd be like 'uh-oh, it's that time (laughs)'. If he asks me directly that's ok. I don't know what to say. I worry about that"
- "It's the teenage stuff- answering things I don't want to talk about. I don't know how much he knows"
- "He doesn't really talk about it and he's never really asked any questions. So that's a good sign I think"

## Appendix 16

### Collated written responses from comments sheets

1. From your perspective, do you feel that your experience as a father caring for your child differs from that of mothers? If so, how?

Yes	No	Possibly
N38 (76%)	N9 (18%)	N3 (6%)
<b>Key examples</b>		
<ul style="list-style-type: none"><li>• Father involved in research and practical areas, mother more in physical care and emotional aspect.</li><li>• Others think fathers are immune</li></ul>		

2. As a father, do you feel your needs (as a parent caring for your child) have been met by family, professional, other? How have these needs been met?

Yes	No	Not clear
N19 (38%)	N25 (50%)	N6 (12%)
<b>Key examples</b>		
<ul style="list-style-type: none"><li>• Family may not acknowledge emotional needs of father</li><li>• Health professionals don't ask fathers about coping</li></ul>		

3. Do you feel fathers are acknowledged by professionals, as having a valid role? What could be done to improve if necessary?

Yes	No	Sometimes/ not clear
N26 (52%)	N10 (20%)	N14 (28%)
<b>Key examples</b>		
<ul style="list-style-type: none"><li>• Problems for fathers meeting professionals due to work</li><li>• Professionals need to acknowledge role of fathers</li></ul>		



**4. What are the main areas that father might benefit from help with?**

**6 areas**

- Emotional (most frequent)
- More integrated system
- Education about condition (best/ worst case scenarios)
- Respite
- Work
- Help for older children

**5. How could the needs of fathers be met? Your recommendations?**

**6 areas**

- Acceptance and awareness of isolation
- Accept needs as valid
- Support groups for fathers only
- Dedicated health professionals to look after all needs of boys, would help parents
- Supportive friendships
- Practical aspects (house alterations; benefits)

**6. Do you think fathers respond differently to other family members, in terms of needs, coping etc?**

**Key themes**

- Don't show emotions, bottle them up
- Male ego in proving can deal with things
- At early stage of diagnosis inability to help son

**7. How did your child's diagnosis affect you? What would you say has been the most challenging time for you?**

**Key themes**

- Diagnosis most challenging time
- Like a death only worse
- Reaction as if suffered a bereavement
- Coping with each stage of deterioration
- Loss of expectations

**8. Overall, how would you describe how you have coped with/ adjusted to your child's diagnosis and treatment?**

**Key themes**

- Adjusted and coped as no alternative
- Keep thinking positive in hope that one day they will find a cure
- You lose virtually everything 'normal' families take for granted
- Had to cope. Family need someone who is strong
- Son is special, lucky to have him
- Importance of family and friend support
- As child's needs change as condition worsens and grows older adjustment not really possible

**9. Do you have any other comments? This could be views about the research, issues you feel are important or any general points you feel are relevant for researchers or health providers).**

**Key themes**

- Manner of diagnosis being communicated dissatisfactory for many and professionals need to give hope of cure
- Researchers need to take a risk- get treatments out of mice into boys
- New medical breakthroughs often disappoint parents
- Problems with complex DLA forms
- This type of research is long overdue
- Poor staff knowledge of DMD, don't understand boys' needs
- Need better information about trials/ possible treatments. Information is found by parents
- Little interest by researchers in DMD
- Ongoing challenges, no time to 'recover'
- Ensuring best possible treatment
- Introduction of national standard for healthcare/ social care professionals dealing with DMD

## Appendix 17

### Interview field notes

Interview N	Date	Participant initials and (number in SPSS)	GHQ score	Age of child (dad)	SDQ Impact on family score	Reflection
1	14/05/07	O W (9)	Normal	12 (46)	Abnormal	<p>The interview took place at the workplace of the participant, within a business park. As I was later attending an academic meeting at a university, I was dressed in a suit. I felt this was appropriate for the setting and would be more appropriate so that attention would not be drawn to me or OW within the workplace and make the situation awkward for OW. I wanted to fit into his environment so that he would not feel embarrassed. I presented myself both as a professional who had experience of working with DMD families, and also emphasised my student status as a learner and researcher. Initially, OW appeared quite business like but once in an interview room was more at ease. He appeared to be 'bursting' to talk about experiences and emphasised his feeling that every thing needed to be 'fought for'.</p> <p>At one stage, he described watching healthy children playing and how painful it can be. I became slightly choked and turned off the tape. He stated that it was refreshing to a professional displaying some emotion.</p> <p>I felt very much at ease throughout the interview, as OW appeared to do. There was laughter throughout, almost as each 'heavy' topic was discussed.</p>

The interview lasted around 1 hour, and OW seemed very reluctant to finish talking. I felt he was enjoying the opportunity to have some one actually to talk to and who understood issues such as KAFOs and wheelchairs.

OW was interested if I had personal experience within my own family of DMD. I explained that my interest came from working with DMD boys and families, but that we did not have personal experiences of this. I shared with him the fact that my mother has a muscle condition called 'fibromyalgia' which involves heightened pain receptors. We discussed the similarities and the fact that he had not heard of the condition- much the same as most people have not heard about DMD.

The end of the interview was jovial and OW invited me to meet his son if I was in Aberdeen in future. He repeated that he had a lot to say and couldn't talk for a long time on the DMD topic. I had to leave for a work-related meeting, but emphasised the value of all the information and insight he had shared with me.

Again, before I left, he reiterated that it was refreshing to see a health professional displaying emotion and he acknowledged that he can talk about it without being affected as he has done so for years.

02/07/07 J McD (44) Normal 25 (63) [SDQ not completed]

2

The interview took place in the family home in Lanarkshire, after I had been collected at the train station. Very welcoming- perception of the research as important in raising awareness. Son not diagnosed until the age of ten- problems with medical knowledge gaps and oversight. I could see that talking about issues was both helpful (perceived as furthering the 'cause') but also not easy to talk about. Described the 'stages' of DMD in detail and saw the progression of condition very much in terms of stages. Coping- wanted to know each stage at a time, and not any further ahead.

Tearful when talking about diagnosis- said it brought it back. Became tearful a few times- usually talking about how son copes so well.

Tape off when wife came in with tea, and seemed to be 'checking' that he was ok. I got the feeling that the diagnosis has not been talked about, or perhaps the impact of the condition itself between the parents. Described a supportive family unit as being the way he has coped. They have not sought help out of the family- but do attend MN meetings. Described offering help to someone who did not attend meetings.

I met son and wife after interview. Talked about studying (son had done 2 years of a PhD and left it) football teams in Glasgow and laughed about 'men and football' and my lack of knowledge. Ended in good humour. I was given a lift to the next interview, where both men knew each other. Before I was in the door they both mentioned that the condition puts such a lot of stress on relationships and that 50% + of dads leave the relationship. They also mentioned that they knew of 1 mother who couldn't cope who had left the family home.

30/06/07 J O'H (36) Abnormal 25 (57) [SDQ not completed]

Interview took place in family home in East Kilbridge. Made very welcome and immediately introduced to son. Dad sighed heavily throughout interview, but appeared to benefit from 'sharing' as he stated throughout that no- one had ever asked him his views and how he feels. He talked in a low voice, as son was in the next room. Kept looking over at his son's silhouette throughout, and looked very tearful when talking about the son who had died just before his 16<sup>th</sup> birthday (also had DMD).

Wife arrived home half way into the interview- voice lowered further, did not want her to hear?

Almost as if he was now embarrassed. He had mentioned earlier that she does not want to talk about it- so this would explain. I met her briefly before I left and seemed friendly towards me.

I mentioned the low response in Scotland, and was told this may be due to people thinking I represented a certain group (MDC) even though I was from a university and no group mentioned. This may be

<b>4</b>	02/07/07	R W (38)	Normal	51 (13)	Normal	<p>because people are often frustrated with the groups. The MN is seen as 'neutral' and the head medic appears to be held in high esteem (Dr DW).</p> <p>Drove me to the station and mentioned he is involved in charity work. Also repeated the view of the importance of work such as mine.</p> <p>Interviewed in workplace- team leader in social work. Immediately showed me pictures of son on PC, along with famous football player who had met him. Humour</p> <p>Jokes and laughter throughout</p> <p>Not the moaning type. Talked of others who just moan. Everyone has something to deal with.</p> <p>Talked of kindness of others due to son who seems to generate it.</p> <p>Very positive towards other professionals.</p> <p>Interviewed in family home. Was very welcoming although appeared a little bemused at first. Made frequent comments about the Irish connection.</p> <p>Laughed frequently and tried to appear jovial, but was upset (eyes watered) when talking of his son's death and how he would like him to have a 21<sup>st</sup> birthday party. Family support and close knit support form extended family were the most important types of support. He was very happy that his son had a good friend who he cold visit in a normal way as the family had installed a ramp. He was upset for his son thinking about missing out on normal 'boy' things as his friends were growing up and spreading their wings.</p> <p>Interviewed in family home. Very keen initially that I got to know son, went for coffee and watched him playing on beach. Like all the dads so far, I felt he wanted to put the interview into context of his son as a person.</p> <p>He became very angry at times, when talking about being let down by professionals and at times I had to steer the conversation to diffuse this. There appeared to be an element of blame towards the</p>
<b>5</b>	07/07/07	D Q (39)	Normal	46 (15)	Normal	
<b>6</b>	14/07/07	P B (23)	Abnormal	8 (51 )	Normal	

7	21/07/07	A R (40)	Abnormal	15 (34)	Abnormal	<p>partner regarding the genetic element 'it was something wrong with her', but later stated this question wasn't an issue.</p> <p>An interesting issue was the 'end of the line', and the end of the 'family name'. A major issue was partner's lack of coping and inability to talk about it, compared to his need to talk about it and deal proactively with it.</p> <p>The loss of a close male friendship, and previous carefree lifestyle was another significant point. There has been a total change in identity.</p> <p>On the way to the station, the son said 'I'm going to die' to me. His father said 'we're all going to die'. I felt sad as he looks so perfect and cheeky. The children are allowed to do anything, they are obviously very spoilt- compensation?</p> <p>He was very angry that DMD is seen as low profile.</p> <p>Interviewed in family home. Very friendly. Feels now is the most challenging time as his son is changing into a man. He raised concerns about his other children. Also raised the issue of genetic testing of his daughter- feels he has had no information about this such as what age she needs to be etc. and he has concerns about this. Another father mentioned this issue.</p> <p>He seems to cope positively as the sole guardian of the children. His parents are actively involved in childcare also and the mother sees the children every 2<sup>nd</sup> weekend. He is friends with his ex-wife and did not elaborate about the impact of the condition on the relationship.</p> <p>He wants to learn from DMD men or older boys about how younger boys can do normal activities.</p> <p>Interviewed at Park and Ride near workplace, in family car. Shortened half hour interview due to time related work restrictions.</p> <p>Very willing to be involved and friendly genuine. Throughout the interview, eyes watered especially when discussing the latest treatment decision making (spinal fusion) and the considerable stress</p>
8	02/08/07	B. M (41)	Abnormal	43 (15)	Normal	

involved in this- this made me realise a strong theme of time-pressured 'window of opportunity' decisions relating to DMD treatment. e.g. do it now or his spine will collapse etc. Referred to faith throughout, he has a very strong Christian faith. Also, the other son was tested for DMD at the same time as other son was diagnosed. The younger son was not positive for DMD, this was very hard to deal with.

The mother had previously been tested for DMD, as her brother had the condition. She was told she was a non-carrier, but the father believes the test was flawed as earlier versions of the test were not very accurate. So, many issues involved here.

He was very positive about the research, and his involvement was seen to promote DMD cause.

This was a telephone interview lasting half an hour. Very agreeable and easy to engage over the phone. Emphasised anger about always having to fight and co-ordinate services them selves. Frustration. Mentioned the huge impact of the condition on families especially marital relationships. He was the first person to mention- unprompted, that he finds himself withdrawing from his own son almost as if he is trying not to become too close to him. This is a protective mechanism.

Talked about frustration of witnessing other parents who do not take a proactive stance. He seemed to summarise the 'fast pace' do things now type approach I have noticed in many of the fathers. I suppose time has a different meaning when your child's life is limited.

Interesting perception of MDC- feels they make things 'easy' for the Government as they fund themselves whereas PPUK campaigns hard for funding. I previously noticed that some parents feel PPUK is a bit aggressive, for example in use of graphic pictures of end stage boys within promotional campaigns.



17/08/07	M B (43)	Normal	32 (60)	[SDQ not completed]	<p>A 30-minute telephone interview. Easy to talk to and appeared relieved to be able to talk about the challenges faced by families. He emphasised throughout the point that his son has been employed in the past. Again, the 'fight' theme emerged although there was praise for council services. Much criticism of the way the NHS deals with families affected by DMD.</p> <p>A main point that he wanted to get across was the fact that there are no facilities (appropriate) for older boys. The fathers all mention they don't like their son to sit and watch others doing things as they feel they will be left out and want to join in when this is not possible.</p> <p>He mentioned that he was glad that someone was asking families as this would mean that something would be written about it and something may be done. I am left with a huge impression of the frustration facing both boys and families.</p>
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18/08/07	M J H (2)	Abnormal	39 (13)	[Missing]	<p>30 minute telephone interview. I left this interview with a feeling of sadness as the family in this case had been through such a lot. He mentioned that his wife was a manifesting carrier and already had a son with DMD when she became pregnant with the son who is now 2. The step son is 13. The fathers are all keen for me to know something about the son almost as if they are ensuring that they are not talking about the son as a 'case'. Names are used, and in this case I was asked to say hi to the 2-year old over the phone! He was a lovely little boy who sang to me!</p> <p>He mentioned that his wife was previously diagnosed with manic depression (he mentioned a paper that is out linking depression to being a carrier) but this proved not to be the case. He seemed to want to talk 'more' than he could (perhaps with wife in house) and said 'it's like having 3 kids', when referring to wife's depression. Again, extremely critical of services. He received counselling via the genetics clinic initially. I feel slightly as if I wish I could have offered something concrete to improve his situation, but all I can do is</p>
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forward the debrief sheet and know that at least I'm trying to do something positive for families.  
 I confess I forgot to write field notes for this interview at the time!  
 The son of this man died aged 19 10 years ago. This was a 1 hour plus phone interview, with a father who has dedicated his life to fighting for other DMD boys as he felt his son was let down by services etc. He was very angry at times, recounting the battle and constant fighting to ensure his son was provided with decent services. He recalled many stories 'he said, I said' and traced from diagnosis to death. He recalled many conversations with his son, and became tearful at times when doing so.  
 Although his son died a decade ago, he talked about him in the present tense as if he was present. I wanted to emphasise that he has helped others (he is involved heavily in charity work). I think talking to me was another way of remembering his son and honouring him. He was really happy to talk to me, but I really felt he was telling me 10 years worth of bottled up grief and frustration. He talked a lot about fathers not coping and leaving the mother as they couldn't face the child.  
 Compared to early interviews and transcribing comments sheets, I felt sad for this man and his family, but have learned not to become too emotional about a situation I cannot change. The valuable thing that I can do is to reinforce the fact that helping with the project is positive for others and for helping future fathers.  
 When the tape was off, this participant mentioned areas needing research to include psychological effects of tests on the boys. The wide- ranging impact on families, including siblings and extended family came up throughout the interview.  
 He talked a lot about losing his own parents, as they had not been able to cope with the situation and had be no help at all. He was quite harsh even laughing about his mother's recent death. I suspect

**12**

27/08/07 J H (45) Abnormal 46 (8) Abnormal  
 31/08/07 B T Not completed DEAD (60s) [SDQ not completed]  
 (n/a: not included in questionnaire study- no SPSS number)

**14**

03/09/07 S B (19) Normal 50 (13) [SDQ not completed]

he was very hurt by the situation and deals with this by maintaining a harsh stance against them.

The issue of his son's impending death, and how to cope with this, arose. Work seems to be the main stability for fathers, as many of the fathers have talked about or written in comments sheets about this. IT seems to offer a support network and reference frame for normality? This man described in detail, the expectations fathers have for sons, and how these are shattered with a DMD diagnosis. He talked about fathers being second place after a child is born, then by the time things settle it is usually age 4 when they are diagnosed. From this point, everyone else in the family comes second to this. He feels that siblings and fathers are neglected, and in the shadow of the diagnosis.

I feel there is huge scope here for family interventions.

This man was very, very keen to talk about his experiences and I felt he had been 'saving up' anecdotes to tell me. His overall impression appeared to be the constant fight for services and the feeling that parents are put to the back of things. Again, like many other dads he talked about the difficulties in having one son with DMD and a younger brother who is overtaking him physically whilst the DMD son becomes frustrated at his own decline.

Throughout this interview, the wife chipped in. This is the first time a wife has been present, but I felt like they had a 'dual' story to tell and wanted me to know everything if something was about to be 'left out'.

At the end of the interview, the father asked me if I thought anything would change because of the research. I replied that I believed so, even though this is a relatively small study. I told him about the interest in my own academic department and how colleagues have become informed and interested in this area. I emphasised the importance of all the information he had shared with me.

## **Appendix 18**

### **Personal Reflection**

This section considers key stages throughout the project, within the context of a reflective critique of the research process, management and evaluation. Influences on choice of thesis topic and the resulting design are considered along with fieldwork, analysis and writing up components of the project. Finally, a general overview summarises the research journey and lessons learned.

#### **1. Process**

##### ***1.1. Choice of topic***

In identifying a research topic, I hoped to build upon my interest in working with families of chronically ill children, and to develop a project that would be perceived as useful to both families and practitioners. From February 2006, one month into the course, I narrowed down areas of interest: developmental psychology; chronic illness and working with families. Having considered a range of options, I opted to target fathers of a son with Duchenne.

I decided upon this for 3 reasons; having previously worked on a project with DMD mothers, the topic of fathers' issues had arisen frequently. Further, I identified a lack of inclusion of fathers in psychosocial research. Finally, I hoped to undertake research that would go some way towards promoting adjustment for families. Having identified the lack of father related research, and overall dearth of psychosocial DMD studies, it felt natural to undertake my thesis within this area. A review of the literature indicated the need for this type of study, and I began to

consider specific research questions and areas of investigation that may be beneficial to the development of interventions. A systematic review of overall parental adjustment to DMD was undertaken as the first step. This served to illustrate previous study designs and summarise findings, whilst considering the strength of evidence based on critical appraisal of study designs.

Having identified gaps in previous research, discussion of the literature with supervisors, and other researchers, allowed me to think laterally about which aspects to target.

### ***1.2. Design***

In considering the study design, I acknowledged that I was more comfortable with quantitative methods, as I had more experience in this area. I felt comfortable within the context of measuring a 'construct' (e.g. depressed) as this was familiar to me. I identified unease with qualitative methods, and my leaning towards positivism as a result of lack of experience and insight into alternatives. At the time of deciding the study, I was receiving training through my employer in qualitative methods, including analysis of qualitative data.

This served to provide me with essential skills and confidence to broaden my methodological skill set, enabling me to consider alternative approaches. In order to provide best answers to the research questions, I started to study various methodological approaches. Prior to, and throughout undertaking the project, I read widely around the topic of qualitative research (e.g. Ziebald and McPherson, 2006);

data collection (e.g. Pawson, 1986; Barbour and Featherstone, 2000) and analysis (e.g. Pope et al, 2000; Barbour, 2000; Morse et al, 2002). I also attended training in both interviewing and analysis.<sup>58</sup>

As a result, I developed insight into the debate over approaches and the nature and purpose of research. I was drawn to the advantages of both quantitative and qualitative methodologies, and preferred a dual approach as it challenged me to undertake work in an area I would learn from, in addition to being advantageous to the research questions. Keeping project aims in mind, the methodology chosen was therefore underpinned by two philosophical approaches. I understood that each implied differing assumptions, and attempted to understand both the nature and purpose of each.

Reading also highlighted the importance of the relationship between researcher and participants and the resulting 'knowledge'/data. I gained understanding of Realist and Constructivist debates (e.g. Adams, 2006; Barbour and Featherstone, 2000; Charmaz, 2006) and methods underpinned by ontological and epistemological assumptions about 'reality'. In considering my research approach, I thought about my own philosophical stance, and was drawn to an Interpretivist ontology (acknowledging data as mediated by the thinking of the researcher). I felt that as the topic was a little known area, it would benefit from the addition of an exploratory qualitative approach. Consideration of sensitive issues (i.e. talking about

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<sup>58</sup> Trainers: Dr M. Kendall and Ms. R. Pratt, Edinburgh University. Training consisted of an introduction to qualitative methods, interview skills, and applications and use of the qualitative data management package NVivo.

sons' terminal illness) pointed to the benefits of an interactive relationship comprising interviews.

According to Glaser and Strauss, (1967), Grounded Theory (GT) is an 'inductive approach of identifying analytical categories as they emerge from the data'. Pope et al, (2000), define this as 'developing hypotheses from the ground up rather than defining then a priori'. In pure Grounded Theory, theories are derived from the data rather than from the researcher's prior theoretical viewpoint (Barbour, 2000). Researchers such as Barbour, (2000), argue that in reality, it is rare to work in this way. For example, not conducting a literature review before interviewing is uncommon. With this in mind, I felt the qualitative element was based in grounded theory principles, but did not adhere to 'pure' GT as described by Glaser and Strauss (1967).

My personal understanding of theory was making meaning of individual experiences, evolving from broad areas to subsequent refined but 'grounded' themes. Through interaction with participants and transcribing of all tapes myself, I aimed to immerse myself in their perspectives (engaging with the data) to make sure data were grounded in people's experiences and their interpretation of them. My approach, therefore, reflected GT (it was grounded and inductive) but unlike GT, it started deductively from pre-set aims and objectives.

The approach of Charmaz (2006) appealed to me as a relevant means of drawing upon GT methods to strengthen my study. Charmaz views data and 'theories' as

constructed rather than 'discovered' (Charmaz, 2006, p10) through the researcher's interactions and perspectives. As such the researcher's interpretation plays a key role in data construction. According to Charmaz' Constructivist revision of Glaser and Strauss' (1967) classic GT, the approach assumes a Relativist stance; acknowledges multiple views and realities (researcher and participants) whilst maintaining a reflexive mindset.

Again, this appealed to me in terms of relevance to the research questions and subject matter. She also asserts that GT methods can complement other approaches, and should not be viewed as 'opposing' them (Charmaz, 2006, p9). In order to best answer the research questions, this confirmed for me the possibility for conducting a mixed methods study. In combining methods, I felt that, should contradictions arise from each data set, this would in fact help to refine, not detract from, any evolving theory.

An alternative approach to analysis was Interpretative Phenomenological Analysis (IPA) (Smith, 1996). Although there are similarities, such as capturing meanings and experiences, GT methods were chosen over IPA for 2 main reasons. Firstly, within GT, the capacity for theory generation was relevant in light of lack of prior work in the area. Secondly, in contrast to IPA, GT allowed the interview schedule to be tailored in light of emic issues. Thus, in the context of exploratory research, GT facilitated a more flexible approach to data collection and analysis.



### ***1.3. Fieldwork***

I found the fieldwork component to be a rewarding and interesting part of the project. I enjoyed hearing people's experiences, and was pleased that many said they felt talking to me was cathartic. I felt positive that my topic choice was going some way towards helping families affected by DMD. In reflecting on interviews, I felt a great sadness at the struggles many faced in dealing with DMD as a family. As interviews progressed, I became 'acclimatised' but remained sensitive to much of the subject matter. I took the advice of a colleague who worked in palliative care, to use a 'switching off' technique (simple visualisation) after each interview. This helped me to cope personally, and to approach each participant with a clean slate for interviewing.

As I interviewed participants and heard about their experiences, I felt a sense of responsibility towards them. During a conversation with the Chief Executive of Parent Project UK (a parent led charity), I was told the study represented the first approach from the psychology profession to request volunteers. Also, on comments sheets, statements such as 'this type of research is long overdue'; 'this is the first time anyone has asked my views', reinforced the feeling of responsibility towards participants. I was aware that my 'attachment' to the topic would influence my approach, and kept this in mind when interviewing.

Throughout fieldwork, awareness of the importance of the 'reflective practitioner' approach and an overall reflexive account of my interactions with participants was maintained. Researchers (e.g. Britten, 1995) have indicated the importance of considering the relationship between methodological approach and the information

this generates, emphasising this requirement for 'reflexivity'. Throughout interviews, I was aware of how I presented myself, how the research was perceived and influences on the nature of information shared with me. I introduced myself to as post-graduate student who had worked in the area of DMD, however, if I had presented myself in another manner this may have changed how participants related to me.

As interviews progressed, there was repetition of issues and themes and I felt that by interview 15, I had gathered a range of experiences leading to 'saturation' of categories. Although this number of interviews has been cited as 'sufficient' for qualitative studies (Guest et al, 2006), according to theoretical sampling there is no requisite sample size (Glaser and Strauss, 1967). The question of sample size is addressed by theoretical saturation whereby data collection ceases to reveal new data (Glaser and Strauss, 1967). I acknowledge that other groups- such as those from ethnic minority groups, may have other issues, but it was not possible to identify these groups (out-with the respondents) due to time limitations.

In terms of skills, I developed my interview skills further and strengthened my understanding of issues within this area of research. In conducting the interviews, clear and effective communication was required. In thinking about improvements, from transcribing all interviews, I identified my interview technique 'flaw' as interrupting and not being comfortable with silence. I acknowledge this is something I will be aware of in future.

Despite finding fieldwork a positive experience, I also found it to be lonely and frustrating. I sometimes regretted choosing a topic that was so emotionally draining, especially concerning terminally ill children. However, these times were minimal and part of the course of the role of a Researcher.

#### ***1.4. Analysis***

I was aware that throughout analysis, my task was to make sense of participants' experiences from their perspectives. As such, I understood the importance of remaining 'grounded' in the data. Analysis was conducted in parallel with the interview process. Although this involved an inductive process, the analysis was also guided by the nature of the research questions. As with the interviews themselves, I maintained an awareness of the importance of a reflexive stance when coding and analysing. For example, I was aware that I needed to remain true to the data and ensure theory was truly 'grounded', and not simply a projection of my specific interests.

Practically, the first step in the analysis was familiarisation with the interview content, leading to early ('process') analysis. I undertook all transcribing myself, as I felt this was beneficial to immersing myself in the data and to completing the life cycle of the project. Many hours were spent transcribing interviews, listening and re-listening to recordings and making notes, leading me to feel immersed in participants' accounts.

My analysis followed Charmaz' description of initial, followed by focused coding (Charmaz, 2006). During initial coding, I aimed to keep an open mind as to the direction of the analysis, but I also sought answers to research questions. In practice, I commenced with line by line coding to 'reveal' initial data. Line by line coding as advocated by Charmaz (2006) included sorting data into properties, looking for assumptions, comparing data with data and identifying gaps. Initially, each interview was coded broadly under (but not restricted to) general question headings, using NVivo as a tool to help organise the data. This process ensured I felt familiar with the data and my interpretations of participants' meanings were fresh in my mind. I then started to ask questions (e.g. what is emerging and which category does this fit or not?) of the data and to compare stories (using the constant comparative approach of GT) and experiences reflected in the data.

Following the more descriptive initial step of analysis, I moved towards more intricate analysis of meaningful concepts and themes. In the second phase of coding- focused coding, I aimed to create more conceptual codes. Initial codes were grouped into larger components and given a title to illustrate content. Groups of similar meaning were merged into more explanatory themes (or categories), whilst seeking connections as the coding process continued. Through comparison of categories at this level, focused coding allowed me to consider differences/ similarities amongst participants' experiences and how they perceived them. Essentially, focused coding led me to aggregate earlier codes and make sense of them.

Whilst conducting the analysis, I was aware that the process needed to fulfil criteria for rigour in qualitative research. Reading around the topic (e.g. Barbour, 2001; Golafshani, 2003; Morse et al, 2002), I identified a school of thought criticising checklists that cite, for example, respondent validation as confirming 'rigour' (Barbour, 2001). Morse et al (2002) asserted that *"the literature on validity has become muddled to the point of making it unrecognisable"* (Morse et al, 2002, p.4). The authors point to reliance on 'evidence' such as triangulation, audit trails and memos, and argue that these processes are not verification strategies and are of little relevance to reliability and validity in qualitative research.

Similarly, Barbour (2002) argued that this results in the *"tail wagging the dog"* and stated that these measures can only strengthen research *if 'embedded in a broader understanding of design and analysis"* (Barbour, 2002, p.1115). In attempting to address such criticisms, I attempted 1) to adhere to accepted criteria for rigour within GT studies, whilst understanding these needed to be incorporated into the research process not added in retrospect, and 2) understand reasons for data collection and analysis choices in relation to initial research questions.

In researching evaluative criteria, various methods were available, for example Lincoln and Guba (1985) propose: credibility, transferability, dependability and confirmability. Morse et al (2002) consider key 'verification strategies' to be: methodological coherence; sampling sufficiency; developing a dynamic relationship between sampling, data collection and analysis, thinking theoretically and theory development. I aimed to choose criteria of relevance to the research, and adopting

the above strategies involved ensuring my research questions 'fitted' my data collection and analysis procedures.

Appropriateness of the sample required participants who had close knowledge of the topic (theoretical sampling), to achieve quality data and saturation of categories. The interactive process of data collection and analysis allowed me to identify potential gaps and, where necessary, seek additional data to explain these. In striving to 'think theoretically', I sought to remain open to emerging ideas whilst constantly comparing against collected data. In developing theory, I moved from initial codes to making meaning of others' experiences.

Throughout analysis, in dealing with the subject matter, I found transcribing many of the initial transcripts and comments sheets upsetting. This became easier over time, but the nature of the topic, and strength of fathers' emotions had a stronger impact on me than anticipated.

### ***1.5. Writing up***

In writing up the qualitative results, I was again aware that themes I 'uncovered' could be influenced by my own ideas/interests and attempted to maintain an open mind. I did not want to impose (even subconsciously) my own biases onto the data- for example areas I deemed more important. In writing up, I knew there was a danger of fragmenting people's experiences and I tried not to do this.

The actual writing up process evolved section by section, starting with methodology- with feedback at each stage. Links to each chapter were developed until the body of the thesis felt coherent and integrated. The importance of keeping the original research questions in mind was reinforced whilst writing up. In particular, I realised the importance of keeping focused on the initial aims and objectives. Whilst writing, I thought about applications of the work to other areas. This led to writing an article about coping with indicators of deterioration of illness, which was peer reviewed and printed in Health Psychology Update. I found writing a slow process, but at the same time rewarding to see the project take shape and produce results.

## **2. Management**

In relation to project management, an area of strength was my organisational skills. I devised systems for recording returns and data management before data collection began and this served me well. From the start, I maintained a reflective diary of the research process and a log of all events relevant to progress. The diary was both cathartic and useful as a reflective learning tool, whilst the log of events allowed me to monitor and manage progress efficiently.

Conducting the thesis took longer than anticipated, although I had drafted timelines and Gantt charts prior to the proposal being submitted. In devising a time scale I had not accounted for other commitments, feedback and re-drafting, which I now understand takes a substantial amount of time. Having gained experience, when next required to undertake the writing up phase, I would allow more time. In managing and evaluating progress of the work, I requested meetings with

supervisors to discuss arising issues and to seek feedback. On reflection, I feel this was of great assistance in maintaining motivation and monitoring progress.

### **3. Evaluation and reflection**

Having previously worked with DMD mothers and sons, I was aware of the emotional nature of researching this area. I felt a large responsibility in undertaking the topic, due to lack of previous research and a desire to represent participants' experiences to the best of my ability. I did not want participants to feel that I was simply undertaking the project for the sake of a novel topic, but that I had a genuine interest in the needs of DMD families and wished to apply my skills to help in some way.

Throughout the research process, I was aware that the interviews had an emotional impact on me. Transcribing the interviews was also challenging, as this served to reinforce some emotional details. I was able to discuss these issues throughout supervision, and used various techniques to 'switch off'. This was important, as at the time of thesis data collection, I was also employed in a project involving interviews with prostate cancer patients. Maintaining an objective awareness of the impact of this type of research, and my own mental health (burnout issues), was a crucial element of undertaking the work, and working within British Psychological Society competency guidelines for good practice. A further issue concerned my perception of the topic, in light of my first (and current) pregnancy corresponding with the thesis write up. I had a heightened awareness of genetic issues, and



developed unfounded concerns that my child might be affected by a similar condition.

Positive aspects of the research included undertaking work in an under-researched area with a client group (fathers) I had no experience of. This was satisfying and allowed me to use my skills to contribute to the field. I also had the opportunity to learn new research skills in qualitative analysis, gained understanding of different epistemologies, and had the chance to design and undertake a mixed methods study. In adopting a rigorous, critical approach to the study at all stages, I feel I can have confidence in the findings and have contributed to the best of my ability.

Other positives included undertaking regular supervision, which served both as a learning tool and the opportunity to discuss any issues arising from the research. Maintaining clear communication with supervisors, and receiving regular feedback allowed me to feel motivated and supported throughout. I feel my key strengths were my autonomy and initiative throughout the work. I also feel I used supervision well and maintained my motivation throughout the research process.

Possible changes to my approach to future research projects could include closer links to clinicians in the field to facilitate recruitment and create networks of interested parties. In future, I would ensure that I plan a more realistic timeframe, given other commitments. Throughout most of the Doctorate, I was employed in an unrelated research post, then embarked on full time motherhood and a second

pregnancy. Despite careful planning, I did not account for the amount of time that I realistically had to dedicate to the thesis

Overall, I learned that carrying out a doctoral thesis is a complex task, with multiple components. The process was made easier by tackling each stage, whilst not losing sight of the bigger picture and aims. I found this was facilitated by good supervision, good communication and seeking feedback at every stage. As a result of undertaking the thesis- from ethical approval to writing up, I feel that I have improved my research skills and had the opportunity to undertake the complete 'lifecycle' of a challenging research project.

On reflection, professionally I have gained a strong empathy for families affected by chronic and terminal disease. I developed an understanding of family dynamics (from participants' descriptions) and individual reactions, both positive and those that may hinder adjustment. As a health psychologist, I am better equipped to work with affected families and would like to develop this interest clinically. Personally, I found the experience fulfilling but highly emotionally taxing. I feel this type of study is best undertaken as a full-time project, without other major commitments. I learned that I have the resilience to cope with emotionally demanding research and the patience to follow through the, often complex, path to completion. I also identified a need to distance myself from my work at times, as I tend to become immersed to the detriment of life quality.

Areas where my practice could be improved would include further improving my interview technique; drawing up a more realistic plan of action and learning to enjoy the process more. I would seek to integrate more with others working in the field where possible, both for personal and professional support. I would now also have more confidence in my own appraisals of my work- whilst being realistically, not overly, critical.

I feel that final 'closure' from the work will come in the form of developing papers from the thesis, knowing that I have contributed towards highlighting the condition and the potential for involvement of Health Psychologists. Feeding results back to fathers will also allow this.

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