

**Research decisions: Living with Duchenne Muscular**  
**Dystrophy**

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## **Abstract**

### **Research decisions: Living with Duchenne muscular dystrophy**

Duchenne muscular dystrophy (DMD) is a severe form of muscular dystrophy that affects males. Muscle deterioration leads to increasing levels of disability during childhood and adolescence, with death commonly occurring in the late teens or early twenties, although changes in care and treatment are leading to increasing numbers of boys with DMD living into adulthood. Parents and parent-led charities are raising funds to find effective treatments and a cure, and much of the medical research they promote requires the participation of those with DMD. This raises questions about children and young people's involvement in research, including their role and approach to consent and how willing they are to be involved in the medical research their parents and DMD charities advocate. Through qualitative interviews with nine boys and young men with DMD and one young woman with muscular dystrophy, I explored their thoughts on medical research and the broader issue of how they live and cope with their condition. As part of this discussion I examined how they might make a decision to participate in medical research, focusing on the processes, interactions and individuals they consider important in helping them to decide. My approach privileges the participants' thoughts and opinions, positioning them as able social actors (James & Prout 1997) who can provide insight into their experiences. Currently little is known about the lives of children and young people with a significant, degenerative disability, particularly around their thoughts on medical research participation and decision-making (Dixon-Woods 2006). The views of my participants provide the basis for this research, with work from the sociology of childhood and from disability studies informing and contextualising it. The way in which parents are involved in daily life is discussed to gain an understanding of how the participants work with those they trust. This relationship may provide understandings of how decisions are influenced by family input and how support assists those who are young and have a degenerative condition. It is possible that this model of working with the significant people in their lives promotes agency and independence, aiding the participants towards, rather than away from autonomy.

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# Contents

<b>Abstract</b>	i
<b>Acknowledgments</b>	ii
<b>Contents</b>	iii
<b>Tables</b>	vi
<b>Introduction</b>	1
<u>DMD/MD</u>	3
<u>Chapter structure</u>	6
<b>Chapter One - Textual representations of children and young people: protection, participation, agency and autonomy.</b>	9
Summary of healthcare and medical research policy and legislation relating to children and young people	10
<i>The Children Act and Every Child Matters</i>	12
<i>The United Nations Convention on the Rights of the Child (UNCRC)</i>	14
Informed consent for medical treatment and medical research	15
<u>Consent to medical treatment</u>	18
<u>Consent to medical research</u>	20
Theoretical discussions of competence and consent	26
Sociology of childhood	36
<u>Agency and autonomy in the sociology of childhood and disability studies</u>	42
<i>Relational autonomy</i>	46
<u>Competence and consent in the sociology of childhood and disability studies</u>	49
Conclusion	55
<b>Chapter Two - Methodology chapter</b>	57
Research with children and young people	58
Qualitative semi-structured interviews	61
Conducting the interviews	64
<u>Social researcher and 'polite guest'</u>	65

<u>Follow-up interviews</u>	70
<u>Analysing data</u>	72
Ethical approval	75
Sampling and recruitment	77
<i>NHS Clinic</i>	79
<i>Action Duchenne</i>	80
<i>Wheelchair Football</i>	80
<i>Muscular Dystrophy Campaign (MDC)</i>	82
<u>Ethical issues</u>	83
<u>Limitations</u>	85
Discourse analysis methodology	87
<u>Document search and selection</u>	87
<u>The approach to discourse analysis</u>	91
<u>Foucault and discourse analysis</u>	93
Conclusion	96
<b>Chapter Three - Living with DMD/MD</b>	97
<u>Coping with DMD/MD</u>	97
Isolation	101
<u>Isolation and the impact on selfhood</u>	104
Independence	112
<u>Being independent</u>	114
Finding out about DMD/MD: diagnosis & getting information	121
Parents	127
<u>Supportive carer</u>	128
<u>Advisor and gatekeeper</u>	130
<u>Parents' role in communicating with doctors</u>	135
Doctors and hospitals	137
<u>Communicating with doctors</u>	137
<u>Medical encounters</u>	142
Conclusion	146
<b>Chapter Four - Research decisions</b>	150

Attitudes to medical research: hopes and expectations	151
<u>How should funds be spent?</u>	157
The approach to decision-making	161
<u>Autonomy of choice</u>	161
<u>Participation and motivation</u>	166
Conclusion	180
<b>Chapter Five - Constructions of children and young people in healthcare discourse</b>	183
Discourse analysis: constructions of childhood, children and young people	185
<u>Future beings</u>	185
<u>Constructions of participation</u>	189
Discourse in medical research	198
<u>Discursive absences</u>	202
<u>Contextualised lives</u>	206
Conclusion	210
<b>Chapter Six – Conclusion</b>	214
Independence, agency and relational autonomy	215
<u>Discursive tropes</u>	217
Contextualised decision-making	217
Conclusion	220
Limitations and future research	221
<b>Appendix</b>	224
<b>Bibliography</b>	267

## Tables

Table One: Lead questions for the thesis	4
Table Two: Age range in recruitment strands	83
Table Three: Databases used in discourse analysis	88
Table Four: Reference list of primary documents	90

## Introduction

This research has been funded by the ESRC (Economic and Social Research Council) as a CASE (Collaborative Awards in Science and Industry) studentship. The ESRC fund research on economic and social issues and CASE studentships aim to encourage collaboration between universities and partner organisations. The partner organisation for my research was TREAT-NMD, who also partly funded the research. TREAT-NMD, which stands for Translational Research in Europe – Assessment and Treatment of Neuromuscular Diseases, an EU Framework Six Network of Excellence Project, has been set up to study the tools needed by clinicians, scientists and industry to bring novel therapies into use. The TREAT-NMD project also examined the social and ethical aspects of research in neuromuscular disease, and it is from this context that my research has been developed. Thus, the funding and agenda of my studentship have influenced how the research proposal was set out and how my starting point was shaped by this agenda. My research has been innovative in that it aims to develop better understandings of the lives and experiences of boys and young men with Duchenne muscular dystrophy [DMD], a condition that only affects boys, and muscular dystrophy [MD]. Both these forms of muscular dystrophy are incurable and degenerative and, due to the severe nature of DMD in particular, charities, which are often parent-led, are keen to fund and promote medical research that will find effective treatments and a cure. Some children and young people with DMD/MD may be invited to participate in medical research; raising questions about how the various influences in the participants' lives shape the way decisions to participate are made. It will be seen in Chapter One that children and young people's consent-giving and participation in medical research is governed by strict guidelines and legal rulings. So whilst parents can be highly motivated to raise the profile of medical research for a currently 'untreatable disorder' (Franson & Peay 2013, p12), this is tempered by medical research safety protocols, which some consider are too conservative and risk averse (Franson & Peay 2013).

It has been from within this complex nexus that my studentship was conceived, during discussions between medical researchers involved in Treat-NMD and my



supervisors. From the medical researchers' perspective they could see that developments in the field could lead to more young people with DMD becoming involved in clinical research and, therefore, wanted an exploration of the issues involved. This research, before I began my investigation of it, was framed as an examination of how those with DMD think about and experience participation in medical research. As I began the interview phase it quickly became apparent that my participants had limited experience and poor knowledge of medical research. Few of the participants had been approached to participate, or had participated in, medical research and their knowledge of it was limited as the development of new treatments is focused on much younger boys. For this reason, the boys and their parents did not spend much time considering the value of the research to them or the possibilities of participating. It felt problematic to stay focused on a set of questions of little meaning to the participants and therefore my approach to working with them became centred on a more nuanced response to the questions set-out in the studentship. My approach engages with the key questions, but does so in a way that represents the participants' perspectives, their concerns and what is of importance to them.

Research has been conducted that examines the lives of those with DMD/MD from the perspectives of their parents, their siblings, healthcare professionals and a range of theorists. However, the group are underrepresented as spokespeople on their own lives and little is known regarding their thoughts on participating in medical research. Indeed, it is argued that the recruitment of children into medical research has often taken place in an 'empirical vacuum' (Dixon-Woods et al. 2006 p175), and the research that does exist has found that children are often marginalised in the process (Coyne & Harder 2011). Thus, my research seeks to address this 'vacuum', and the principal motivation is to explore what those living with DMD/MD think about medical research, if they share the keenness of some parents to promote it and how they think they might approach making a decision to participate.

To better understand the dynamics that will be explored in this research, more details on the diagnosis of DMD/MD, its treatment options and the hope medical research offers will be explained in the next section. The primary focus in this

research is on DMD; however, one young woman with a rare muscular dystrophy [MD] which has a less severe prognosis is also included. The reason for her inclusion, due to low recruitment numbers, is more fully explained in Chapter Two, and her views have provided a much valued additional voice in this research.

### DMD/MD

DMD is an X-linked form of muscular dystrophy which means that, almost exclusively, it affects boys. DMD occurs as a result of mutations in the dystrophin gene leading to an absence of or defect in the protein dystrophin (Muscular Dystrophy Campaign [MDC] 2013). Diagnosis is at approximately 5 years of age, and muscle strength deteriorates as the boys mature, requiring the use of a wheelchair before they are in their teens; non-progressive cognitive dysfunction might also be present in some boys (Bushby et al. 2010). Over time, respiratory, orthopaedic, and cardiac complications develop, and without intervention, the average age at death is around 19 years (Bushby et al. 2010). DMD involves a significant level of disability, with upper limb function and good posture becoming increasingly difficult (MDC 2011) and older boys may require spinal surgery to correct scoliosis, a deformity of the spine (Bushby et al. 2010). Respiratory problems occur in older boys, due to a loss of respiratory muscle strength, which needs the careful management of non-invasive ventilation (Bushby et al. 2010). Monitoring is necessary for cardiac care as muscle strength decreases, and for nutritional needs as swallowing becomes more difficult (Bushby et al. 2010). The use of steroids has been proven to have an effect on muscle strength in DMD if used in boys who are still walking; and steroids may stabilise or even improve muscle strength for a period of time (Bushby 2008). The use of steroids to prolong muscle strength, and advances in the care and knowledge of DMD have helped to prolong life expectancy, meaning that a generation of young men with DMD are alive who were not expected to live to adulthood (Abbott & Carpenter 2009; Gibson et al. 2009).

The use of steroids and improvements in care management can improve life expectancy, but DMD remains a severe and deteriorating condition. Effective

treatments for DMD are not yet available in the clinic (Bushby et al. 2010) and current medical research into DMD hopes to slow the disease progression. Trials on Exon skipping agents offer the potential to 'skip' the damaged sections of genes (MDC 2013). Whilst this is not a cure it is expected that it will halt or reverse the symptoms, making them more like Becker muscular dystrophy, which is less severe (MDC 2013). Medical research into the efficacy and safety of treatments requires the recruitment of participants with DMD, and the research sometimes involves procedures such as injections and muscle biopsies. Issues of competence and the capacity to consent to these onerous tests can raise concerns as they involve 'vulnerable' children with a life-limiting condition.

Improved care means that the decisions surrounding boys and young men's possible medical research participation are more likely to directly involve them as they mature and develop decision-making skills. However, the process of maturing is accompanied by progressive muscle weakness and loss of mobility, and the usual pattern of growing up and leaving home is severely disrupted (Jessup & Parkinson 2010) or entirely curtailed because of care needs, which are often met by parents. Medical research holds out the tentative promise of a cure or effective treatments, but this can be overshadowed by the threat of a decline in health (Hodges & Dibb 2010). Because of some improvement in life expectancy, boys and young men with DMD are now able to contribute more directly to social and medical research (Abbott & Carpenter 2009; Rahbek et al 2005). Growing up with a significant disability that emerges in childhood and increases during adolescence raises issues about the role agency takes and how independence can require support from others. Thus, it is timely, and the central focus of my research, that the participants express their thoughts on medical research decision-making and the related question of how they experience agency, independence and autonomy in their lives. There is a potential for healthcare and medical research governance to judge children's abilities according to their age and developmental stage. Yet some argue that children have diverse capacities (Hagger & Woods 2005) and may be adept at decision-making and managing regimes of medication and treatments (Alderson 2003; Berntsson et al. 2007; Dixon-Woods et al 1999). Such observations help to substantiate this research, which privileges the participants' perspectives on decision-making. In

order to contextualise how these decisions are reached my research will reflect on elements of the participants' lives, depicting what it is like to grow up with a degenerative condition in a disablist society that privileges health, independence and progress (McLaughlin 2006). The broader context within which the participants live is also significant, this is made up of their family, friends, educational life and their healthcare, and acknowledging this world helps to situate them as moral actors (Carnevale 2004; Cockburn 2005) who live in relational contexts as part of a family.

Parents may see participation in medical research as a hope for their sons and a source of hope for them (Schaffer et al. 2009) but it is less certain if those with DMD/MD share this hope; are they willing to participate, or do they have other priorities? The lead questions from the project aim to explore the approach taken by boys and young men with DMD to participating in medical research. The project began with these four questions:

1. What factors from the boys' lived experience influence their thoughts and decisions regarding participation in medical research?
2. In what ways does the boys' family, social & cultural setting shape their attitudes to participating in medical research?
3. What key processes & interactions play a part in how the boys consider or decide to participate in medical research?
4. What resources are significant to the boys when considering medical research?

Table One: Lead questions for the thesis

The questions under each of the four headings listed above guided the interview schedule, (see Appendix A for schedule) and the initial development of my research. As well as addressing decision-making, the questions touch on the nature of influence, risk, safety, the recognition of disabled children's competencies, and adult power. These issues may lack a clear point of consensus within ethical and consent discussions, and the participants' contributions will add a useful perspective. As explained earlier, I had to adapt my approach as I began the interview phase of my research. It became apparent that the lead questions detailed above in Table 1 and which helped to shape my interview schedule were too focused on medical research, and during the fieldwork and writing-up phase of my thesis it was necessary to respond to this due to the limited knowledge the participants have of medical

research. Therefore, greater space, for example, was given to them talking about their experiences within healthcare and exploring their family lives and their interests. These factors have been relevant to the understandings I have developed; the participants' lives and the experiences and influences within them all contribute to informing their views on medical research and guide the way in which decisions are likely to be made.

My conceptual approach is informed by the sociology of childhood, which considers children and young people to be able actors in their lives who can demonstrate agency and competence (Jenks 2005; James & Prout 1997). Disability studies will also be drawn on; it promotes the view that disabled children and young people can enact agency in their lives and are able to give informative accounts of their experiences (Connors and Stalker 2007; Davis & Watson 2000; Watson et al. 1999). These two theoretical approaches will help to generate understandings of the lives and decision-making experiences of those with DMD/MD, acknowledging them as competent actors rather than passive patients. Next, I will briefly outline the structure of this thesis and the content of the chapters.

### Chapter structure

In Chapter One the literature that has informed my research will be reviewed and discussed. UK policy and guidelines relating to children and young people's healthcare and participation in medical research will be examined before moving on to cover theoretical discussions on children's competence and consent. The final part of the literature review will explain the relevance of the sociology of childhood and disability studies. Both these theoretical positions complement and inform my approach, as they address children and young people's capacity for agency and autonomy. Relational autonomy will also be discussed as this provides a way to understand the close and trusting relationships in which disabled children can live and make decisions.

Chapter Two, the methodology chapter, will explain the processes employed in conducting the research, including sampling and recruitment and the methodology

for the discourse analysis in Chapter Five. There will be a discussion and validation of qualitative methods and semi-structured interviews, reflecting the sociology of childhood's acknowledgment of children and young people's abilities as reporters of their own lives. This will be followed by an explanation of how the data was analysed and a discussion of the ethical issues involved in the research.

Chapter Three will examine daily life and the challenges of living and coping with DMD/MD, and the way both isolation and independence can impact the participants' lives. The way participants find out information about DMD/MD will be explored, as this may reveal what information they consider is useful and how it is shared within families. In the final section, the role of parents, doctors and the hospital will be discussed, as parents play a large part in the participants' lives, and doctors are likely to have some influence on how they experience their healthcare and on any decisions they might make.

Chapter Four will address the central themes of this research that emerged from discussions with the participants, encompassing their thoughts on medical research, their hopes and expectations of it and how they might approach making a decision to participate. Scenarios were used in order to help the participants contextualise and imagine how medical research could relate to their situation, and their attitudes to risk and the possibility of having a muscle biopsy will also be discussed.

Chapter Five uses discourse analysis to examine the discursive tropes that are present in official healthcare policy and associated documents in order to understand how children and young people are constructed within them. This chapter will highlight the discourse within which the participants receive their healthcare, giving consideration to the ways in which the rhetoric around children's participation may or may not be effectively improving their inclusion in their healthcare and in decision-making.

Chapter Six is the concluding chapter, it will consider how the findings made in the body of the thesis may contribute to better understandings of the contexts within which disabled children and young people live and make decisions. I will draw

together some of the factors that have been discussed, indicating what may shape the participants' approach to making decisions. The conclusion will attempt some resolution of the issues covered through illustrating how agency and autonomy can be thought of as relational, dynamic and contextually dependent concepts that contribute to competence and independence in the participants' lives. Finally, the limitations of this research and the possible direction future research could take will be acknowledged.

In the next chapter, Chapter One, I will discuss the literature that informs this research, starting with a discussion of healthcare policy.

## Chapter One

### **Textual representations of children and young people: protection, participation, agency and autonomy.**

This literature review will examine some of the ways in which children and young people, including those with a disability, are thought about, positioned and treated within a range of texts. The review will begin by examining policy documents that inform and guide children and young people's healthcare and the governance of children and young people's participation in medical research. Examining these areas of healthcare and medical research governance illustrates how children and young people with DMD/MD are positioned in healthcare decision-making and in the giving of consent to medical research. As explained in the Introduction, medical research is currently taking place to develop effective treatments for DMD/MD, and it is patient groups and charities run by parents that are keen to support research and to see drugs becoming available. Whilst children and young people's participation in decision-making is now a central tenet of UK healthcare, elements of protectionism may also be evident in the guidelines. Protectionism and paternalism can limit the ways in which those with DMD/MD are involved in decisions on their healthcare, and this may be more likely in the governance of medical research where specific legal rulings must be observed, despite parents' and/or their child's willingness to participate in research. Therefore, the sense of urgency amongst those promoting medical research into DMD/MD may conflict with the cautious, protectionist way in which children are positioned in healthcare, and particularly in medical research.

After reviewing healthcare and medical research guidelines, the work of various writers will be drawn on to discuss and examine the tensions between children's protection and their rights to participate in decisions on their healthcare and medical research. A review of these matters is salient to my research, which is developing understandings of how children and young people with DMD/MD think about issues relating to consent and autonomy, and how they think they might approach medical research decisions. The specific situation for those with DMD/MD raises questions that are explored within the thesis about how these children and young people may



or may not be positioned as competent, active agents in their care and in the decisions they make. The review will then move on to discuss the sociology of childhood and disability studies, to present a theoretical position wherein competence, agency and autonomy are accepted as integral aspects of children's lives. The theoretical framework of the sociology of childhood and disability studies will inform this thesis; it provides a means of examining and troubling the gap between children and young people's protection, and their agency and autonomy.

### **Summary of healthcare and medical research policy and legislation relating to children and young people**

This opening section will review policy and guidelines on children and young people's participation in their healthcare, and on consent issues relating to medical treatment and medical research. Attention will be paid to how children's competence, their vulnerability, and the need for their ongoing protection are constructed in these documents.

There has been an increasing awareness of the need for children and young people to participate more fully in their healthcare; in the Kennedy report *Learning from Bristol* (Bristol Royal Infirmary Inquiry 2001), the importance of establishing and promoting participation is emphasised. The document was produced in the wake of high death rates amongst babies undergoing cardiac surgery at Bristol Royal Infirmary; this then raised concerns over broader failings in children's care throughout the NHS (Bristol Royal Infirmary Inquiry 2001, p8). *Learning from Bristol* notes that a 'National Service Framework for children is urgently needed and [we] welcome the Government's recent recognition of this' (Bristol Royal Infirmary Inquiry 2001, p414). Hence, a National Service Framework [NSF] was established to improve health and social care services for children, and the document *Getting the Right Start, National Service Framework for Children: Standard for Hospital Services* (Department of Health [DH] 2003) was the first NSF standard to be produced. Implementing the standard for hospital services was regarded as essential, as it entailed children's safety whilst in hospital, thus directly responding to the findings from the Bristol Royal Infirmary. As with the other documents that will be reviewed,

*Getting the right start, National Service Framework for Children: Standard for Hospital Services* (DH 2003), henceforth referred to as the *Standard for Hospital Services*, is likely to have some ongoing impact on how my participants receive their healthcare. The impact of how they receive their healthcare may then shape how they think about and make decisions on their health, and this includes making decisions to participate in medical research.

The *Standard for Hospital Services* (DH 2003) states that its aim is to promote hospitals as being one 'strand in a seamless mesh of services' (DH 2003, p4). It refers to *Learning from Bristol* (Bristol Royal Infirmary Inquiry 2001), observing that a 'worrying' number of observations were made regarding the Bristol Royal Infirmary, including children's rights and vulnerabilities being overlooked, poor quality of care and the fragmentation of services (DH 2003). The *Standard for Hospital Services* (DH 2003) observes in its introduction that it is 'imperative that this NSF provides a robust framework for responding to Professor Kennedy's concerns and... those of... other inquiry reports' (DH 2003, p4). One of those 'other' reports had been Lord Laming's inquiry (TSO 2003) into the murder of Victoria Climbié in 2000 at the hands of her guardians, a tragic death that motivated closer attention to children's safeguarding. The *Standard for Hospital Services* (DH 2003) calls for hospitals to meet their responsibilities both to safeguard and promote the welfare of children, and for a 'shift in culture to gear services to the needs of the individual child' (DH 2003, p5), with staff trained to support children to be active partners in decision-making (DH 2003, p16). It also calls for 'real' choice over treatment and care, which, for younger children, it describes as including 'where to sit during a procedure... or which arm to put a thermometer under' (DH 2003, p18). The document states the importance of child-centred practices, with children and young people situated as 'active partners in decisions about their health and care, and, where possible... able to exercise choice' (DH 2003, p9). Simultaneous to being more involved in their care, young patients' safety is highlighted as they are considered vulnerable, and in need of 'safeguarding' (DH 2003, p10). This document sets out its expectations that children should be more involved in their healthcare, although worries over their safety somewhat limit the parameters of participation.

The *National Service Framework for Children, Young People and Maternity Services: Disabled children and Young People and those with Complex Health Needs* (DH 2004b) sets out the standards and expectations of care for disabled children and young people. Therefore, it will impact upon how my participants are treated and receive their healthcare, due to the influence of the NSF on healthcare professionals' practice. Produced in 2004, the document states that:

this standard locates the experience of disabled children and young people... within the wider community. It promotes their inclusion and their ability to live as ordinary a life as possible through effective partnerships with them. (DH 2004b, p8)

The document observes that often 'disabled children are less actively involved in decision-making than children who are non-disabled' (DH 2004b, p29) and they should routinely be involved and 'supported in making informed decisions about their treatment, care and services' (DH 2004b, p30). Disabled children are also considered more likely to 'experience abuse than non-disabled children' (DH 2004b, p35) and the document aims to develop 'safeguarding guidance and procedures for professional staff working with disabled children' (DH 2004b, p36). As with the *Standard for Hospital Services* (2003 DH), children's involvement in decision-making is promoted, whilst the need for their safeguarding is simultaneously reinforced. These twin roles of involvement and protection are also evidenced in *The Children Act* and in *Every Child Matters*, both of which will be reviewed next. These documents further inform and embed the policies, practices and guidelines that may affect how my participants are positioned in their healthcare.

### *The Children Act and Every Child Matters*

*The Children Act* (TSO 2004) was produced in 2004, and its aim has been to improve the well-being of children (TSO 2004, p7) and, amongst other measures, to ensure the safeguarding and welfare of children in NHS hospitals and facilities (TSO 2004, p9). The Act also establishes a Children's Commissioner whose role is to promote 'awareness of the views and interests of children in England' (TSO 2004, p1). The call for enhanced attention to children's needs and their involvement in

their healthcare, legally underpinned by *The Children Act*, is set out in the policy document *Every Child Matters: Change for Children* (Department for Education and Skills [DfES] 2004). *Every Child Matters* proposes a ten year plan to build services ‘around the needs of children and young people so that we maximise opportunity and minimise risk... preventing things from going wrong in the first place’ (DfES 2004, p2) and to improve ‘children’s health and well-being’ (DfES 2004, p5). Lord Laming’s (TSO 2003) inquiry into the death of Victoria Climbié is cited as a motivation in the plan and reinforces the significance of the NSF and in particular the *Standard for Hospital Services*:

The Government has brought safeguarding within the framework of clinical governance through the new National Service Framework hospital standard, following the recommendations of Lord Laming. (DfES 2004, pp75-6)

*Every Child Matters* states that, after consultation with children and young people, the key outcomes of change include: being healthy, and staying safe (DfES 2004, p4), and these outcomes ‘are given legal force in the Children Act 2004’ (DfES 2004, p8). The impetus for healthcare improvements exemplified in the NSF, and reiterated in both *Every Child Matters* and *The Children Act*, emerges out of failures to protect some children and reflects ongoing concern about children’s welfare. The attitude of these documents and the drive behind their implementation is to avoid more scandals and tragedies through the embedding of safe practices that aim to manage risk and avoid future failings. Simultaneous to these concerns is a push to increase the opportunities for children to be involved in their healthcare. These two intentions, to safeguard children and to encourage their participation, are evident in the documents so far reviewed. The overall strategy of the NSF, as set out in the *National Service Framework for Children, Young People and Maternity Services: Core Standards* (DH 2004a) is to promote children’s inclusion in their healthcare, whilst safeguarding their welfare (DH 2004a, p9). Hence, elements of protectionism alongside elements of partnership with children and young people are given credence by *Every Child Matters* and *The Children Act*, and are reinforced in the NSF. Whilst *Every Child Matters* and *The Children Act* are UK originated and focused; international instruments also have some impact in the UK, leading to the relevance of the next document to be reviewed.

## *The United Nations Convention on the Rights of the Child [UNCRC]*

*The United Nations Convention on the Rights of the Child* (United Nations 1989) is an internationally recognised human rights treaty that grants all children and young people a comprehensive set of rights (DfES 2013). It was ratified by the UK in 1991, but it is not legally binding (Cave 2013) and is yet to be incorporated into UK legislation (James 2008), though it may have some persuasive weight (Liberty 2008). The NSF is, according to Moore and Kirk (2010) explicitly driven by Article 12 of the UNCRC and in the NSF's *Core Standards* (DH 2004a) Article 12 is quoted from, stating the right for children to express their views. The NSF's *Core Standards* asserts that the NHS should design services around the needs of the patient and that 'children have a right to be involved in decisions on their care' (DH 2004a, p90). Likewise the NSF *Standard for Hospital Services* (DH 2003) says that children should be involved in decisions on their care (DH 2003, p11), and as with the NSF *Core Standards* (DH 2004a) it quotes Article 12 of the *UNCRC*. The *UNCRC* emphasises working in partnership with children, and, though it is cited in NHS healthcare policy as a standard for care, the status of the *UNCRC* in the UK is somewhat contingent, and children's involvement is partly dependent on healthcare professionals' assessments of the child's competence (Alderson 2007). This contingency around the *UNCRC*'s legal status is apparent in *The Children Act* (TSO 2004) which observes that the *UNCRC* is 'subject to any reservations, objections or interpretative declarations by the United Kingdom for the time being in force' (TSO 2004, p3). So, whilst the *UNCRC* promotes the increased involvement of children in various kinds of decision-making, and is cited in healthcare policy, children's involvement is somewhat influenced by UK healthcare professionals' attitudes and working practices.

According to the current UK Children's Commissioner, Dr. Maggie Atkinson, her role and that of her office is to promote and protect the rights of children under the *UNCRC* (Children's Commissioner 2012). Point 1 of Article 12 of the *UNCRC* states in full that:

States Parties shall assure to the child who is capable of forming his or her own views the right to express those views freely in all matters affecting the

child, the views of the child being given due weight in accordance with the age and maturity of the child. (United Nations 1989)

This establishes that when adults are making decisions affecting children, children have the right to express themselves and have their opinions taken into account. This places a responsibility on those caring for children and young people to pay attention to their thoughts and opinions. Point 3 of Article 3 in the *UNCRC* says that:

States Parties shall ensure that the institutions, services and facilities responsible for the care or protection of children shall conform with the standards established by competent authorities, particularly in the areas of safety, health, in the number and suitability of their staff, as well as competent supervision. (United Nations 1989)

The priorities set out in the *UNCRC* inform NHS policy, encouraging the participation of children and young people in choices and decisions affecting their healthcare. This participation is expected to be in accordance with their age and maturity, whilst also paying attention to their health and safety and ensuring the competency of their care. However, children have often been regarded as vulnerable individuals in regulation and legislation (Hagger & Woods 2005), and newer working practices do not entirely supersede these presumptions about children's inherent vulnerabilities and need for protection. The *UNCRC* is not fully embedded in UK law and, whilst it has a recognised status, it may be open to the interpretations and attitudes of healthcare professionals and how they balance children's involvement in their care with their safeguarding and protection. These latter two factors are particularly relevant in the next section, which will review the regulation of children and young people's role in informed consent.

### **Informed consent for medical treatment and medical research**

*Consent – The voluntary agreement of an adult or competent child, based on adequate knowledge and understanding of relevant information, to participate in research. (Medical Research Council [MRC] 2004, p21)*

The legal purpose of consent before a medical procedure is to protect the autonomy or right to self-determination of the person, and the giving of consent entails an interactive process between the subject and the healthcare professional to ensure understanding is reached (Meaux & Bell 2001, p243). Consent provides

healthcare professionals with a 'justification to what would otherwise be a battery' (Cave 2013, p2). In the case of medical research and clinical trials, the participation of children and young people is regulated by specific laws, rules of governance and consent procedures. Medical research is a generalised term for research that does not fall into the category of clinic trials. It could involve testing out a type of physiotherapy, drawing blood for analysis or conducting a quality of life study. Clinical trials are conducted to evaluate medicinal products in scientifically controlled settings to ascertain their safety and efficacy, and trials have their own strict protocols. Throughout the thesis I will use the term 'medical research' as a generalised term that refers also to clinical trials, unless I need to be more specific, in which case I will use the latter term where appropriate.

The development of contemporary approaches to informed consent for medical research emerged in the Nuremberg Code which was established after *The Nuremberg Trials*. The Trials dealt with details of brutal and inhumane experiments conducted without consent on detainees in concentration camps during the Second World War. The key principles of The Nuremberg Code established ethical research on human subjects that would produce results which were for the good of society (BMJ 1996). The voluntary consent of all research subjects was deemed essential, with researchers expected to ensure there were no undue risks (Smyth 1999). The requirement for consent precluded children from participating (Botkin 2003) as point 1 of the code requires the person to have legal capacity to give consent (BMJ 1996). The Medical Research Council (MRC 2004) explains how current medical research has evolved from the Nuremberg Code:

Current ethical principles for conducting research involving children have evolved from the Nuremberg Code which emerged after the Second World War. The code set out statements of moral, ethical and legal principles relating to research involving human subjects, and included a bar on research involving children. Research was seen as a potential harm from which vulnerable people should be protected. (MRC Ethics Guide 2004, p9)

The Declaration of Helsinki in 1964, which set out ethical standards for research, established that it is the physician's task to promote the health, well being and rights of patients involved in medical research (World Medical Association 2013). It also stated that research on minors which was of benefit to the population being

represented and which could not be conducted on legally competent individuals was acceptable (MRC, 2004). From the restriction on research with children to the conditional approach adopted at Helsinki, attitudes to minors' participation in medical research have shifted, although in the UK the 'law relating to research on children... has never been clearly established' (Hull 2000, p180). As Hull (2000) observes, there has been less clarity about the status of children in consent issues, in part because children are regarded as a vulnerable group who are not generally considered to be autonomous. Therefore, questions remain around their role in decision-making and consent-giving (Alderson 1993).

Due to children's variable and at times vulnerable status, there have been cases of medical experimentation exploiting the involvement of children being cared for in institutions (Meaux & Bell 2001; Hagger & Woods 2005). Between the 1950s and the 1970s, children with learning disabilities who were being cared for in a state school in New York provided a ready pool of subjects. Some were infected with yellow fever, syphilis, hepatitis and tuberculosis, to test out the efficacy of diagnostic tests and vaccines (Meaux & Bell 2001). These events, amongst others, bring into focus the 'dangers imminent in research and the moral and social desirability of ethical research' (Halse & Honey 2007, p338). Although not specifically related to children's consent, concerns have been raised in the UK over the process of giving and receiving consent after Alder Hey Hospital and Bristol Royal Infirmary were found to have retained children's organs without parental consent (Alderson 2007; Corrigan 2003; English & Sommerville 2003). Following the death of some children, their vital organs were removed and kept in hospital storage without obtaining consent as a consequence of vagueness in the law surrounding permission (English & Sommerville 2003). Not surprisingly this led to a public lack of trust in hospitals and the need for clarification and a stronger emphasis on consent (Alderson 2001), and also to the implementing of governmental policy aimed at protecting children (Moran-Ellis 2010). Not only have the breaches of trust at Alder Hey and Bristol Royal Infirmary caused public mistrust, they have also impacted on some NHS staff, who have expressed concerns over sufficient consent being obtained, and worries about child protection issues after the uncovering of the two hospitals' errors (Stalker et al. 2004). The outcome of these scandals has been to enhance the surveillance



and management of children's healthcare, and this includes the management and observance of consent procedures both for medical treatment and medical research.

As there is more clarity around consent to treatment than there is regarding consent for medical research, the next section will start by reviewing the former aspect of consent, prior to moving on to discussing consent to medical research.

### Consent to medical treatment

Within the NHS, the NSF's *Standard for Hospital Services* (DH 2003) states that hospitals should follow Department of Health guidance on consent for treatment, observing that all 'professionals should be familiar with the concept of "competence" in giving consent' (DH 2003, p17) as competence is a key feature of informed consent. The Department of Health's *Seeking Consent: Working with Children* (DH 2001) provides healthcare professionals with guidance on competence and consent. This document states that once children are 16 they are presumed in law to be competent to consent to their own medical treatment, however, they should be encouraged to include their parents in consent procedures (DH 2001, p3). The document goes on to define competence as the capacity to:

take a particular decision... to comprehend and retain information material to the decision...and use and weigh this information in the decision-making process. (DH 2001, p4)

Those under 16 are not presumed competent to make decisions on their healthcare:

However, the courts have stated that under 16s *will* be competent to give valid consent... if they have "sufficient understanding and intelligence to... understand fully what is proposed" (sometimes known as "Gillick competence")... there is no specific age when a child becomes competent to consent to treatment: it depends both on the child and on the seriousness and complexity of the treatment being proposed. (DH 2001, p5)

Gillick Competence addresses the child's right to decide what treatment or medical procedure they undergo in accordance with their demonstration of competence to comprehend the relevant information. This right was brought into question after a

circular from the Department of Health asserted that advice on contraception could be given to girls under the age of 16 without parental knowledge, according to the discretion of individual practitioners (NSPCC 2012). A concerned parent, Victoria Gillick, contested this, claiming that the parental right to decide was being denied and that the actions should be viewed as treatment without consent. The case went to the House of Lords where Mr. Justice Woolf rejected her argument and gave an account of when a child might be judged competent:

The child must be capable of making a reasonable assessment of the advantages and disadvantages of the treatment proposed, so the consent, if given, can be properly and fairly described as true consent. (Walters 2008, p1)

This ruling permits those under 16 to give consent if they can demonstrate sufficient competence. Thus, Gillick competence rests upon when it is judged that the child has sufficient skill and maturity to understand and make their own decisions about medical treatment. The implied general acceptance is that adolescents are entering a time of more autonomy (InnovAit 2008), but the issue of judging competence under the Gillick ruling is left to the health professional's discretion and their own assessment of each child's context-specific competence.

Despite the recognition of children and young people's potential for competence, if they refuse to consent to a recommended medical procedure this can be unfavourably viewed. According to the Department of Health's *Reference Guide to Consent for Examination or Treatment* (DH 2009), the 'refusal of a competent person aged 16-17 may... be overridden by a person with parental responsibility or a court' (DH 2009, p32). Thus, Gillick competence bestows some opportunity for children and young people to demonstrate competence to make decisions, but this does not necessarily extend to children and young people's refusal of treatment. The implication is that adult decisions about a child or young person's treatment options can influence how children are, or are not, involved in making decisions. There is then, some imbalance, as:

patients whose competence is in question are found rational and able to give consent if they accept the advice of the doctor; but are judged incompetent if

they reject that same advice (Shield & Baum 1994, p1).

Thus, the tensions between children's participation and their protection, which were evidenced in the policies and laws reviewed earlier, are also apparent in competence and consent issues. Children and young people are permitted some freedom in how they are involved in the giving of consent, but this is reliant upon adult judgements and assessments of their competence.

### Consent to medical research

The cautious approach seen in consent to treatment extends into the governance of paediatric medical research, an area of healthcare which the Department of Health has stated it intends to improve on:

The use of medicine in children should be guided by the best available evidence of clinical effectiveness... and safety, ideally derived from clinical trials conducted with children. (DH 2003, p25)

In the above statement, ensuring the efficacy and safety of medicines requires the participation of children in clinical trials, therefore medicinal safety is achieved through children's involvement in clinical trials, and the associated consent issues focus on ensuring children's safety. These issues impact those with DMD/MD, whose future access to effective drugs and treatments is interrelated with the current necessity of involving those with DMD/MD in medical research. The rulings and governance that are being reviewed here concerning consent directly apply to the way those with DMD/MD would be positioned if or when approaching taking part in medical research.

Medical research and clinical trials involving children are recognised as a key strategy for ensuring that paediatric medicines are suitably tested prior to use. Various DMD/MD charities including Action Duchenne, the Duchenne Research Fund, Harrison's Fund, Joining Jack, the Muscular Dystrophy Campaign, Parent Project Muscular Dystrophy and Duchenne Now are fundraising and campaigning to ensure the continuation of medical research to find effective treatments for DMD and

MD. In the charity Action Duchenne's words, there is a determination to find treatments and a cure for DMD:

Action Duchenne was established to find a cure or more effective medicines to treat Duchenne Muscular Dystrophy. Duchenne is a severe and progressive muscle wasting disease for which there is presently no cure. However, clinical trials are now a reality, thanks to the work of organisations like Action Duchenne. <http://www.actionduchenne.org/action>

According to the Royal College of Paediatrics and Child Health [RCPCH] there is also a drive to promote paediatric medical research for the broader paediatric community:

Children's research is needed to define the causal biological mechanisms, alter the development of aberrant trajectories, preserve health, and reduce the costs of healthcare in adult life... the organisational structures of the NHS, the largest universal healthcare system in the world, provide a unique platform to integrate clinical research and patient care... Research harnessing the wealth of post-genomic sciences and the power of the NHS, offers unparalleled opportunity to improve the wellbeing of infants and children, turn the tide of the growing burden of ... diseases that have their origins in early life and lead to premature adult death, and benefit the health of future generations. (RCPCH 2012, p4-5)

The development of paediatric medical research is managed, however, by complex guidance that is viewed as overly restrictive by some who are keen to see more research for and on children. This is particularly so in DMD, where some consider the risk posed by the child's health condition to be as or more risky than the research which might produce a future treatment or cure (Franson & Peay 2013).

In the Department of Health's guidance document for health professionals, *Seeking Consent: Working with Children* (2001), the section discussing research opens with the statement:

The lawfulness of research on children who lack capacity has never explicitly been considered by the English courts. However, parents may often be invited to consent to their child being involved in "therapeutic research", on the basis that a new treatment may be as effective, or more effective, than the standard treatment. (DH 2001, p25)

This precautionary approach places the decision-making with parents, and this is also the case with reference to non-therapeutic research:

The courts have held that people with parental responsibility can consent to a non-therapeutic intervention on a child as long as that intervention is *not against* the interests of the child and imposes only a minimal burden. (DH 2001, p25)

The problem this raises is that what an adult considers is a minimal burden or low risk, such as taking blood or having a series of non-invasive scans, may be differently experienced by a child (Alderson 1993), particularly where the child already faces repeated medical interventions.

The notion of children maturing towards having competence and decisional capabilities, which has to some extent been evidenced in healthcare policy, is absent from the British Medical Association's [BMA] (BMA 2012) guidance on medical research. Unlike the context for medical treatment, where children of 16 are presumed legally competent to consent to their own treatment (DH 2001), the guidance states that, in the context of research, the 'presumption of competence for 16-17 year olds does not apply. A 16-17 year old is only deemed competent if Gillick Competent' (BMA 2010, p16). It goes on to state that the final decision to participate in medical research:

rests with patients (when competent) and with parents.... Depending on the nature of the research, and the research ethics committee's view, parental consent may also be required, even if the child is competent. (BMA 2010, p52-53)

In the UK, common law applies to research which does not involve the *Medicines for Human Use (Clinical Trials) Regulations (2004)*. For research involving investigational medicinal products, the *Medicines for Human Use (Clinical Trials) Regulations (2004)* apply (MRC 2004). Under these regulations a minor is classed as a person under 16 and, whatever their level of competence, they cannot consent to participation and a parent or guardian must give consent (MRC 2004, p25). The developing recognition in general healthcare of children and young people's variable levels of competence and ability are not apparent in the governance of clinical trials. However, points 6 and 7 of the *Clinical Trials Regulations (2004)* do state that the

principles relating to a minor should ensure that:

Point 6: The minor has received information according to his capacity of understanding, from staff with experience with minors, regarding the trial, its risks and its benefits.

Point 7: The explicit wish of a minor who is capable of forming an opinion and assessing the information referred to in the previous paragraph to refuse participation in, or to be withdrawn from, the clinical trial at any time is considered by the investigator. (Medicines for Human Use (Clinical Trials) Regulations 2004, p43)

Point 6 and Point 7 both hinge upon an adult having the ability to make fair and just decisions on a child or young person's abilities and the adult's expertise to competently explain a research protocol to them. The effective and informed involvement of children and young people can be enabled or denied according to the judgements and information-sharing of the adults conducting a trial. The Medical Research Council remarks that the *Clinical Trials Regulations (2004)* afford additional protection to children at a time when more may be asked to participate as part of a drive to provide fully licensed medicines for children (MRC 2004, p25). A considerable proportion of medicines used in paediatric care are not licensed for paediatric use (Brierley & Larcher 2010; RCPCH 2012) and the risks associated with clinical trials are minimised by the close regulation of children's involvement and are offset, or validated, by the risks associated with taking unlicensed drugs. However, as noted, the move towards children and young people's involvement in decision-making in their healthcare is not readily evident in the governance of clinical trials, where strict protocols exist. These rules are in place to protect children, and they reveal the 'tension between respecting and promoting children's autonomy, and recognizing that often children need protecting from harm' (Frith 2006, p179). There may be times, however, when protection also silences children or provides them with only a limited space in which to ask questions and receive suitable answers to their questions about participation.

As noted, parent-led groups and charities strive to fund more medical research on DMD/MD, which, in the case of DMD, often requires the recruitment of younger boys, and this can make some of the issues regarding competence and consent more

complex. Proxy consent may not sufficiently ensure a child's willing participation, as parents' motivation to consent for their child may be based upon their hope of a miracle cure, or of 'carrying on the fight' (Shilling & Young 2009) on behalf of future generations. The issues around proxy consent are not necessarily rational, they may be:

shaped by the particular context and nature of the trial, [and] the hopes, fears, expectations and obligations the parties bring to the trial discussions. (Shilling & Young 2009, p5)

The hope of a cure or the threat posed by a child's health condition (Alderson 2007) can overshadow parents' comprehension of a research project, and parents might see it as being in the child's best interests, although what is considered beneficial varies widely between people (Carnevale 2007b). Whilst parental energy may, understandably, be focused on finding a cure or effective treatments, less is known about what the children and young people who are prospective candidates for research think. In some cases, parents and researchers may regard research participation as an imperative due to the severity of the condition; but this conviction may leave children and young people with less opportunity to decline, or raise questions. Guidelines and legal rulings on informed consent may help to protect children and young people from coercion, but the imbalance of power between adults and children, amidst the impetus to yield clinical results, might influence how some decisions are made. Children generally have less capacity to express their own interests and can be vulnerable 'toward being used or disregarded in favour of adult interests' (Carnevale 2012, p41). Hence, in my research, the voices and opinions of those living with DMD/MD are being sought, to hear what they think about medical research, about the prospect of participation in it and how they view decision-making both as individuals and within the matrix of the family.

The guidelines and legislation on competence and consent explored here reveal that assessments of children's competence are contingent upon the judgements of healthcare practitioners/researchers. Whilst the *Medicines for Human Use (Clinical Trials) Regulations (2004)* pitch a minor as being under the age of 16, the European Compliance Academy [ECA] regards all those under the age of 18 as minors. This

pliable categorisation of maturity illustrates 'how arbitrary it can be' (Alderson 2007, p2273). The ECA promotes information exchange between the pharmaceutical industry, medicines authorities and universities, and composes part of an ad hoc group chaired by the European Commission. They produce recommendations aimed at unifying the rules on clinical trials across the EU and ensuring the ethical involvement of the paediatric population (ECA 2008). The difference between the UK and European definitions of a minor conveys the lack of clarity in defining a 'child'. The mutability of this term and the concomitant complexities in the regulation of children and young people's participation in medical research and clinical trials leads to much debate. The manner in which competence and consent guidelines are set out tend towards presenting them as existing outside of the sociocultural and political contexts from which they emerge. However, they are likely to be assessed according to the presiding practitioner's values, which will be influenced by the professional guidelines and culture in which they are immersed. Chapter Five will, therefore, use discourse analysis to interpret and critique healthcare discourse and the ways in which children are constructed within it. The analysis will provide a different perspective on how children and young people's healthcare is shaped, through troubling the discursive guidelines and culture that healthcare professionals work within.

So far I have detailed how competence and consent are set-out in healthcare and medical research guidelines, which apply age-based definitions alongside context-specific assessments of competence to decide if and when children can give consent. I have raised some concerns with how these guidelines may assess children and young people's competencies. The possibility of subjective judgements being made by healthcare professionals when assessing competence and the problems with the giving of proxy-consent have been observed and it has also been seen that the age at which competence is assumed alters according to the context of the research being conducted. The concerns raised have the potential to impact how those with DMD/MD are positioned if thinking about taking part in medical research. The various influences on their decision-making, from parents, health professionals and parent-led organisations, and the concerns and issues encompassing competence and informed consent raise key questions for this thesis.



Are children and young people with DMD/MD vulnerable and in need of protection or can they act as agents in their lives who are able to participate in making medical research decisions? These matters will be addressed through discussions with my participants, and in Chapter Four the focus will be on the key topic of the participants' attitudes to medical research and how they might approach decision-making.

In the next section I will review the variable ways in which competence and consent issues are discussed and theorised by a range of writers. These discussions range across differing opinions on children and young people's competence and capacity to consent, with advocates both for and against recognising their capacities at young ages.

### **Theoretical discussions of competence and consent**

It has been seen that the assessment and governance of children and young people's competence and consent within healthcare and medical research is complex. Yet it is recognised that more medical research involving and on behalf of children is needed (Hagger & Woods 2005; Modi et al. 2012; RCPCH 2012; Smyth & Weindling 1999). Rosalind Smyth, the former director of the National Institute for Health Research Medicines for Children Network, states that there is a 'dearth of high quality paediatric research' (Smyth 2001, p1377). Medical research with children presents unique challenges, and the stated need to improve on recruitment levels is complicated by concerns over ensuring children's protection. To better understand the range of concerns expressed in arguments for and against children's competence and involvement in decision-making and consent processes, debates from the field of bioethics and medical sociology will be examined. The debates portray how a range of writers think the issues of competence and consent could or should be theorised and applied. Bioethics is a branch of inquiry that originated in the 1970s as a response to rapid developments in healthcare, which have raised complex and ongoing ethical issues (Carnevale 2012; Whitty-Rogers et al. 2009). In medical sociology the sociocultural setting within which individuals live is examined and the social effects of medicine and illness are addressed, along with the complex

health related issues that attend the increase in technological healthcare (Nettleton 2006).

Competence is central to the legal approach to consent (Shaw 2001) and informed consent respects the individual's autonomy (Stoljar 2011). A valid consent is one that is based on sufficient information and freedom from the coercion of others (Boulton & Parker 2007, p2189). However, the concept of informed consent has 'tended to be interpreted rather narrowly and largely in terms of respect for autonomy' (Boulton & Parker 2007, p2189). Autonomy is questioned by some, as it favours individualism and an atomistic approach to decision-making, failing to account for joint decision-making, which is recognised and validated in feminist and relational ethics (Carnevale 2012). For disabled children and young people, joint decision-making and supported, relational autonomy are viable options, through which they can reach decisions with parental support. There is the possibility that parents and other adults who hold strong views can 'significantly influence a young person's freedom to choose' (Larcher & Hutchinson 2010, p310). Indeed, the process and dynamic of decision-making within families of disabled children has the potential to be complex due, amongst other reasons, to the requirement for comprehensive adult care and parental fears about losing their child. However, it is also possible that familial contexts may encourage children and young people's competence and their ability to make decisions within a relationally supportive setting.

The lack of clarity around competence seems apparent in the legal positioning of competent under 18 year olds who may not be able to refuse treatment where someone with parental responsibility consents to it. Writing on medical ethics, Dickenson (1998) considers that the ruling on a refusal of treatment, as seen in the Department of Health's *Reference Guide to Consent for Examination or Treatment* (DH 2009) reveals a dichotomy. The ruling states that the 'refusal of a competent person aged 16-17 may... be overridden' (DH 2009, p32) if a parent or the courts consider a refusal to be detrimental to the young person's health and wellbeing; thereby undermining the move towards children's greater involvement in decision-making. Dickenson argues that the 'differential "tariff" for consent and refusal' (1998,

p5) means that competence and consent have variable weightings, according to a child's acceptance or refusal of treatment options. Whilst competent adults can withhold consent, even to life-saving treatments, young patients have less definite rights. As Cave (2011) observes, in 'medical law negative autonomy is generally given higher priority... competent minors, conversely, have a strong right to consent to treatment, but a weaker right to refuse it' (Cave 2011, p437). Thus, competent children's positive decisions are accepted when they accord with adult preferences, although their competence to make decisions can be vetoed when their wishes go against adult judgements. Moores & Pace (2005) observe the conflict between autonomy and protection:

A child may be deemed competent to consent to one treatment but incompetent to refuse another. This appears to be a reflection of society's desire to encourage child autonomy and yet to ensure that children are protected from the consequences of decisions that (in society's view) put them at risk. (Moores & Pace 2005, p245)

Caution is understandable when decisions to stop or refuse treatment might have detrimental outcomes, but it does indicate how children and young people's competence is viewed as conditional.

The protectionism which is seen in rulings relating to refusing treatment also extend into the way children and young people are situated in medical research. Informed consent is a central concept in research ethics (Harris & Holm 2003), and Thong and Harth (1991) discuss the contentious origins of informed consent. They state that the 'dark side' of the informed consent doctrine is its origin in laws relating to battery and negligence; 'its brief [is] to protect members of society from medical malpractice... as such its stance is always adversarial and never conciliatory' (Thong & Harth 1991, p568). Thong and Harth ask how such a 'blunt' instrument can achieve harmony in consent procedures, and they suggest that what is needed is 'conciliation and arbitration' (1991, p569). Gillick competence, as an individual assessment of each child's capacity, could be a means towards 'arbitration', potentially giving minors an opportunity to demonstrate competence. It is possible that through talking with a child or young person, they are able to demonstrate their ability to understand quite complex issues. However, in a discussion of how Gillick

competence could be applied in medical research, Hunter and Pierscionek (2007) argue that Gillick competence should only be used in specific circumstances that are likely to 'bring direct benefits to the participants and pose minimal risks for them' (Hunter & Pierscionek 2007, p661). Concerns around professional competence are raised in Hunter and Pierscionek's argument as they question the skills of medical researchers to gauge the competence of children under 16 years old. They suggest that medical researchers can be influenced by a need to promote recruitment, resulting in 'too ready a determination of Gillick competency' (Hunter & Pierscionek 2007, p661), thus rendering children vulnerable to researchers' agendas (Hunter & Pierscionek 2007).

What Hunter and Pierscionek (2007) are suggesting may or may not be the case. The argument does, however, highlight how protectionism can regard children as vulnerable to adult power, persuasion and motives. It is possible that the partisanship Hunter and Pierscionek (2007) are concerned about does sometimes influence participation in medical research, with children and adults being vulnerable to the 'institutional context and affiliation of researchers' (Boulton & Parker 2007, p2191). However, the protectionism exemplified in Hunter and Pierscionek's argument can be restrictive, as it may unproblematically portray children as vulnerable, thus limiting their involvement in decisions. Managing children's consent processes in medical research calls for a balance between over-protection and under-protection:

There are the risks of protecting children to the extent of silencing and excluding them from research so that their voices are not heard... versus the risks of coercing or exploiting their involvement (Alderson 2007, p2281).

Children and young people's competence and abilities can be measured according to quite fixed ideas about their developmental capacities; Piaget is regarded by some as the 'founding father' of developmental psychology. This theory separates the different stages of childhood which must be passed through prior to reaching adulthood:

Piaget's ideas about children's cognitive and moral development... as they

progress towards adulthood and acquire adult 'competence' - have had a significant influence on professional and public perceptions of childhood. (Morrow 2011, p12)

Alderson (1992) asserts that developmental stages are not reliable indicators of children's competence, although these have 'been used almost as facts on which to found bioethics theories' (Alderson 1992, p121). It could be argued that assumptions of incompetence are a contravention of children's rights to respect as human beings. The recognition and optimising of children's competence is a right that, Fundudis (2003) states, is set out in the UNCRC. Fundudis, a child psychologist, suggests that the question of children's competence is more helpfully considered not as an issue of when they should be treated like adults, 'rather, it should be viewed in terms of treating them with respect as people involved in a process aimed at sensitive and appropriate assessment of their abilities' (2003, p21). Fundudis acknowledges the limitations of age based criteria of competence and notes there is a need for competence to be contextualised. Fundudis considers that, 'children are generally competent but their competency... should be assessed in terms of "the matter requiring decision"' (2003, p20). He argues that each decision should be taken on its own merit, according to the various factors involved in weighing the matter up. Fundudis does, however, go on to state that children's competence needs to be assessed in 'developmental terms as a function of... psychological maturity and competence' (2003, p21). This may be criticised for placing too much emphasis on developmental psychology's measurements of maturity, when a focus on personal experience may be a more sensitive indicator of individual capacity. Age and stage based theory can overlook the contextual and social components of cognitive development (Hagger & Woods 2005), failing to engender constructive acknowledgement of children's variable levels of ability. Alderson (1992) contends that isolating competence from its social context situates it as a mechanistic skill, rather than something that is part of the child's growing development of contextualised abilities.

Alderson is an advocate for children being more involved in healthcare decision-making (Dickenson 1998, p3), and she is vocal in advancing the view that children are not inherently vulnerable. Her views on children's capabilities counter

paternalistic assumptions about children's need for protection, assumptions that can limit children's involvement in decisions on their healthcare. Alderson questions legal discussions on competence, arguing that 'legal debates about minor's consent... are less concerned with children's rights than with adults' freedoms' (2007, p2273). She advocates for recognition of children's competencies at young ages and proposes that they can gain knowledge through personal experience. Her work and, it will be seen later, work emanating from the sociology of childhood, emphasises children's contextualised abilities rather than age-based definitions of competence. Alderson critiques Piaget's theories and she believes that 'contingencies such as experience and ability can be more salient than age to a child's competence' (2007, p2273). She draws on studies of children with type 1 diabetes and cystic fibrosis to illustrate how they can make sophisticated decisions about their healthcare and commit to regimes of treatment, illustrating that their consent can be an 'informed commitment... beyond passive compliance' (Alderson 2007, p2277).

Larcher and Hutchinson (2010) consider that competence is related to cognitive ability, but they also contend that children's experiences of illness can provide them with greater insight and understanding. Larcher, a UK consultant in paediatrics and clinical ethics, and Hutchinson, advocate a nurturing of relationships between professionals, children and their families. Larcher and Hutchinson admit that assessments of Gillick competence can be contentious, 'since value judgements may be involved' (2010, p307), and state that the child's contextualised ability to understand is central to Gillick competence. They suggest competence should be assessed in collaboration with children and their families in order to reduce the dichotomous distinctions between competence and incompetence:

Assessments of competence need to be undertaken within the general dynamic of working in collaboration with children and families, and the fostering of relationships based on trust, mutual respect and information sharing. There are strong ethical and clinical obligations to involve children in decisions about healthcare to the extent that they are willing and able to do so. (Larcher & Hutchinson 2010, p311)

Cave considers children under 16 to be highly dependent on the way information is

shared with them, as doctors are given 'considerable discretion in deciding how much information to give' (Cave 2011, p434). She regards Gillick competence as setting a high threshold for competence, and observes that children's dependence on adults may make informed decision-making complex, with the role of professionals and parents likely to be influential, raising the possibility that collaborations can sometimes be challenging. She argues that a doctor's best interests for the patient can include their desire for the patient to make what they, the doctor, consider to be the 'right' clinical choice. Cave asserts that children need to be given opportunities to develop and demonstrate capacity:

As human rights jurisprudence places increasing emphasis on the autonomy rights of minors, it seems that doctors... will give greater prominence to the child's competent choices. In light of this it is increasingly important to ensure that children are given every opportunity to develop and demonstrate their capacity. (Cave 2011, p448)

Cave (2011) cautions that the dividing line between persuasion and influence is thin, and that information-sharing should be aimed at improving children's understanding. With reference to medical research decision-making, building children's confidence in their capacity to be involved can ensure that they are included in decisions and that they will be listened to, with their choices heeded.

Information-sharing is essential in enabling children and young people to comprehend their healthcare and cope with unpleasant medical treatments and procedures. Drake (2001), a UK nurse writing from her experiences on a hospital ward, argues that it can be insufficient for doctors to give only the amount of information they consider suited to their child patient on proposed medical procedures. She states, 'English medical law... holds that the levels of information to be given to the patient are... dependent on the responsible body of medical opinion' (Drake 2001, p103). This statement implies that children who are considered unable to cope with too much information may be under-informed. Drake describes a situation where a child with Crohn's disease required nasogastric feeding, which she felt was insufficiently explained to the child by the consultant, leading to distress upon the procedure being commenced. Drake contends that differing professional working practices can clash, due to incompatible views on the amount and depth of

information young patients can cope with.

In Drake's example, the doctor gave information he/she considered was in the child's best interests, however, using 'in the best interests of the child' can adhere to a 'paternalistic criterion' (Dickenson 1998, p7). This paternalistic criterion may raise objections to children being involved in decisions, as it is assumed that children will make decisions which situate 'them outside adult protection... and act in ways that place them at risk' (Lansdown 2001, p7). Protecting 'vulnerable' patients may be used as an excuse for poor communication and failure to explain proposed treatments properly (Pearce 1994, p715), yet children are dependent upon information being shared appropriately with them (Alderson 2007). Lansdown asserts that 'where children are encouraged to articulate their concerns and given opportunities to express their views, they will be far better protected' (2001, p7). Children's competence can be positively influenced when time is spent, 'supporting and listening to children, offering them choices and encouraging them to take an active part in their treatment' (Alderson 1992, p122). Ensuring this process takes place shows respect for the child, even if they lack the capacity to be fully involved in decision-making; it involves them in the current decision being made and can better prepare them for involvement in future decisions. Pearce (1994) cautions, however, that whilst it is important to ensure the child is in control of what happens to them, there can be a risk of placing too much responsibility on them to make a decision. Frith (2006) sees this as being resolved by sometimes treating the child as an autonomous being who can make their own decisions, whilst at other times parental consent should be sought. With regard to those with DMD/MD, gradual and respectful involvement in their healthcare can build their confidence in their own abilities. It can help to ensure that as they mature they are able, if they wish, to take more control of healthcare decisions and/or medical research decisions.

These debates on competence and consent demonstrate the gradations in how children may or may not be effectively involved in decisions on their healthcare. Adults might think their influence is benign, and most doctors aim to support children's growing competence. However, the professional status of doctor's advice and the emotional dilemma for parents who want to both help and protect their child



can be such that children are placed in an unequal position when important decisions are being made. Competence may be assumed when all the parties are in agreement, but called into question when difficult decisions (Alderson 1992) and disputes occur. Alderson (1992) argues that involving children in making interim decisions respects and develops competence; this involvement can nurture agency and autonomy in the lives of children and young people with serious, degenerative conditions like DMD/MD.

The nature of DMD/MD and the decisions that have to be made on treatments or on medical research participation draw on many issues and influences. These may include:

the home and family, school and friends, disease and treatment, health professionals and hospital... the current state of medicine, the law, the media and concepts of childhood. (Alderson 1992, p122)

Each or all of these can influence the way in which decisions are reached and the dynamics that contribute to the entire decision-making process. Carnevale (2007b) critiques bioethical frameworks which presume the ethics of children's medical care are resolvable through the weighing of harms and benefits. He contends that this 'modern western narrative construes moral agents as self-determining rational problem solvers' (Carnevale 2007b, p574). The predicament for parents of children with DMD/MD to make the right decisions with and for their child is finely balanced. Doctors and parents want the best for the child, but the layered interactions can make the decision-making process prone to adult proclivities. These processes include the children as patients and as recipients of their parents' caring work for them, and these factors can influence how decisions are reached. Parents may be hopeful that new treatments for DMD/MD are coming closer to fruition, potentially influencing how they think and talk about medical research and decision-making with their child.

The promise of effective treatments for DMD can be a powerful source of hope, as Samson et al. (2009) describe in a discussion of how hope can sustain parents of a child with DMD. The parents they interviewed expressed a belief in the possibility of a cure:

This belief is fuelled by the excitement surrounding technological developments in general and specifically by the advances made in characterising the genetic basis of DMD. (Samson et al. 2009, p108)

This expectation of positive clinical results is a motivational factor in generating funding, of being part of 'community interaction' (Condin 2002, p2) and of 'assisting in the endeavour to "find a cure" for DMD' (Condin 2002, p27). Whilst it is highly unlikely any parent of a child with DMD would force their child to participate in medical research, the subtleties of influence can be hard to identify. Influence may be invisible and unvoiced, and legislation and guidelines, whilst effective, cannot account for the invisible thread that familial allegiance may sometimes exert. Applebaum et al. (2009) state that pressure is probably the most common behaviour to impair the voluntariness of consent to research, and it is possible that children are inclined towards agreeing with their parents and doctors, as they trust them and rely on their support. Nevertheless, the positive aspects of kinship and familial loyalty provide strong and vital bonds for disabled children and young people.

In families with a son who has DMD, the lives of child and parent are often interdependent (Abbott & Carpenter 2009; Abbott 2012; Condin 2002; Samson et al. 2009), with parents and son collaborating to reach decisions, and this can blur what some consider are distinct boundaries around consent, competence and autonomy. As well as collaborating, there may be times when there are subjects which are just too difficult to talk about (Abbott 2012), and protection and parental pain over their son's condition can lead to information and decisions being poorly managed. Defining the best way to inform and involve seriously disabled children and young people in decision-making can fluctuate, as each child and family will have their own preferences (Alderson 1992) and their own concerns. Despite these variables, discussions on the ways in which parents, doctors and children can effectively work together to make complex decisions can enrich understandings of what it means to be a care-dependent individual, who can be enabled towards living well. The role of interdependence and relational autonomy will be further discussed later in the chapter as this offers a way of understanding how supportive relationships can work in positive ways, enabling disabled children and young people's agency.

Discussions on children's competencies and the levels of input they should be afforded or denied stimulate intense debate, and adult attitudes are not neutral or decontextualised, as informed consent is embedded in social processes (Barr 2006; Corrigan 2003). Adults read and assess children's competencies through the filter of their own presumptions, their training and their values, and a key anxiety in building partnerships with children is caused by uncertainties around children's competence (Dixon-Woods et al.1999). As discussed, children in medical research are not necessarily or singularly vulnerable subjects, and their competencies develop not just through age but through personal experience (Larcher & Hutchinson 2010). Recognition of children's competence and the knowledge and experience they accrue through having a chronic illness or disability are concurrent with concepts from the sociology of childhood, and disability studies. These concepts regard children to be capable actors in their lives, with the capacity for agency and autonomy. As my research gives prominence to the thoughts and experiences of children and young people with DMD/MD, the conceptual framework of the sociology of childhood and of disability studies will inform how I approach interviewing, interpreting and theorising the participants' contributions. Hence, the next section will review these concepts.

## **Sociology of childhood**

The sociology of childhood has developed over the last two decades of the 20<sup>th</sup> century as children and childhood have been re-theorised, re-thought and re-positioned (Cocks 2006, p247). James and Prout's (1997) *Constructing and Reconstructing Childhood* has been an influential publication which drew on the contributions of empirical researchers from the UK and other countries (Moran-Ellis 2010), instigating new conceptualisations and studies of childhood. This approach understands childhood to be a sociocultural construct rather than a natural state of being, involving an 'actively negotiated set of social relationships within which the early years of human life are constituted' (Prout & James 1997, p7). Childhood is, therefore, situated and understood through 'a set of beliefs and practices determining how children are treated' (Alderson 1992, p123). The sociology of childhood challenges reductive beliefs about children and argues for them to be

considered as active social agents who, to some extent, can shape the structures and processes around them (Morrow 2011, p6). Prout and James assert that previous theorising about children and childhood dealt with the 'biological facts of life, birth and infancy... with little account taken of any cultural component' (1997, p14). Childhood as an institution has often been treated as a static entity, and this constructed way of thinking about children and childhood has repercussions. In the healthcare domain, attitudes and beliefs about children's abilities can obscure their actual competencies (Alderson 1992), denying them the right to involvement in matters concerning their health.

Children's physical immaturity is acknowledged in the sociology of childhood, but the way in which this immaturity is interpreted in relation to adulthood and 'full' maturity is examined and challenged as being a result of specific social, political and cultural institutions (Shanahan 2007). Thorne (2002) explains the sociology of childhood as focusing on children as social actors, agents and cultural creators; and it is:

intent on "de-naturalizing" age categories and relations, asking, for example, how particular ideas of "the child" are constructed... theoriz[ing] age as a structural and discursive dimension of social life, analogous to and intersecting with gender, racial-ethnicity, and other lines of difference. (2002, no page number)

Tisdall (2012), drawing on James and Prout's ideas for encapsulating how the sociology of childhood views children, states that:

children are and must be seen as active in the construction and determination of their own social lives, the lives of those around them, and of the societies in which they live. (2012, p7)

Through resituating children as active in their lives and the lives of others, they are seen as agents rather than passive, 'empty vessels to be filled with adult wisdom' (Tisdall 2012, p7).

At various historical moments the child as an individual and childhood as a conceptual, collective term have been explained and understood according to influential theories. Children have been considered to be innately innocent and pure

(Hogan 2005), as exemplified by the philosopher Rousseau in the 1700s who idealised the child as developing an inherent capacity for reason (Jenks 2005). The late 1800s, by contrast, saw children to be in need of saving or correcting, an identity that fitted with the values of the 'middle class and the respectable working class' (Hendrick 1997, p35). According to Wyness (2006), the establishment of compulsory schooling created children's economic dependence on adults, whilst also assuming children's ignorance, and failing to value their contextualised knowledge. Current attitudes to children and childhood reflect both the concept of childhood corruption and depictions of childhood innocence, and this is further reinforced through media representations of an 'innately passive child' (Kitzinger 1997, p174). This depiction is regularly used to raise funds for children's charities (Morrow 2011; O'Dell 2008), including those for disabled children (Battersby 2011; Millard 2011). The reinforcing of this discourse can embed notions of children's inferiority and weakness as 'the discourse of innocence conflates notions of innocence and vulnerability' (Meyer 2007, p91). Meyer's summation can be extended to include disabled children who may be regarded as doubly 'burdened' by being both young and physically incapacitated and this can efface or ignore the experiential knowledge children accrue.

As well as problematising constructions of children as innocent and vulnerable, there has also been a questioning of attitudes to childhood shaped by psychology, education, welfare and social policy (Alanen 1994, p27). The sociology of childhood and related empirical studies seek to advance informed understandings of childhood, moving beyond the theorising of developmental psychology and providing 'fresh understanding of the social structuring of childhood and the way in which children make sense of their world' (Banks et al. 2001, p802). This conceptual shift regards children and young people's agency as exemplified through their capacity to define themselves as they make decisions and cope with their lives (Johnson 2001). This conceptual approach is reflected in a number of qualitative studies which highlight children's position as active agents (Leonard 2009), with an emphasis on children's current lives rather than seeing childhood as a prelude or precursor to adulthood (Alderson 1993).

The relevance of children's lives in the present is important, as focusing on the experiences children are facing now validates their current situatedness rather than diminishing these as less important than what children will experience and become in the future (Morrow 2011). This is reflected by proponents of the sociology of childhood who argue for acceptance of:

the social construction of childhood rather than normalised development, a respect for children and childhood in the present rather than a focus on adults and adulthood as the 'gold standard', and recognition of children's agency and rights rather than perceiving them as passive and dependent in the private family. (Tisdall 2012, p2)

Mayall (2000) has argued that a developmental discourse, informed by biological notions of growth towards future maturity, positions children as 'pre-people, outside the polity' (Mayall 2000, p246), regarding them as being in a preparatory rather than a participatory stage of their lives (Mayall 2000). Such constructions support the notion that children are not socially embedded citizens, because they lack maturity and competence and need protection not autonomy (Tisdall 2012). In the healthcare setting this can limit their decision-making opportunities, effectively subordinating their choices (Beresford & Sloper 2003; Coyne & Harder 2011). Whilst their marginalisation may be motivated by adult protectiveness it can be paternalistic and restrictive in its outcomes, with adult theories of childhood infringing upon how children and young people experience their own childhood (James in Grover 2004). The theoretical positioning of children as adults-in-waiting, or as 'socially developing organisms' (Grover 2004, p91), contrasts with the sociology of childhood approach, where children's development is seen as variable, and physical immaturity does not justify children's exclusion from being treated as current members of society (Moran-Ellis 2010).

Situating children as socially engaged agents with individual perspectives (Shanahan 2007) is relevant to my research project, in which children and young people with DMD/MD are being given the opportunity to speak-up about aspects of living with their condition. Acknowledging that children have capabilities which are current, rather than nascent, recognises how they can be 'competent and reflexive in reporting their own experiences' (Balen et al. 2006, p43). When disabled children

and young people are conceptualised as vulnerable this can eclipse 'notions of children's competencies' (Morrow 2007, p9). Such inaccuracies are countered by representing how disabled children and young people can be competent and knowing actors in their lives. However, even in the enlightened conceptual space of the sociology of childhood, there is little work with minority children's groups, including disabled children (Moran-Ellis 2010). This echoes their historical marginalisation and silencing, 'in research and in many other spheres' (Abbott 2012, p242). Permission to conduct research on, or with, disabled children and young people can be problematic due to consent issues, the sensitivity of researching disabled childhoods (Cocks 2006) and institutional inflexibility in granting access permission (Heath et al. 2007). Institutional gatekeepers (Heath et al. 2007) and NHS staff's concerns around consent issues (Stalker et al. 2004) can make access to hearing disabled children and young people's views complex. Hood et al. (1996) observe from their own research that:

The process of gatekeeping has raised problems about how research can work towards reaching informed consent from children. Our study shows that we could not approach children directly; their socio-political positioning means that adults must give permission. (Hood et al. 1996, p126)

The process of gaining permission to conduct some of the interviews for my research will be further discussed in Chapter Two, portraying how gatekeepers and extensive paperwork can make research with disabled children and young people challenging.

The next section will review concepts of agency and autonomy, drawing on perspectives from the sociology of childhood and also from disability studies to better understand how these terms can be applied to disabled children and young people. Whilst the sociology of childhood highlights children's capacities, there is, as noted, less work on disabled children's experiences, so disability studies provides an added perspective to my research. Disability studies coalesced in the late 1980s as a reaction to the dominance of psychological and medical discourses of disability (Barton & Oliver 1997) and the recognition that disability is socially and institutionally marginalised and discriminated against. One of the key elements to come out of this development was the social model of disability, which defines the difference between

impairment and disability; whereby the 'former is individual... the latter is structural and public' (Shakespeare 2010, p268). The social model emphasizes the disabling and oppressive role of the social world and its environs, and it regards disability as being socially created. However, this model has been challenged for failing to acknowledge the significance of impairment in some disabled individual's lives. For instance, those in chronic pain, and those struggling with fatigue or with degenerative conditions may be less able to ignore the impairment effects of their condition (Barnes 1998; Shakespeare 2010). Therefore, models that integrate the social with the personal have been developed, which can respond to the impact of the relationship between bodies and sociocultural contexts (Gibson et al. 2007). Nevertheless, the social model is a useful term for understanding how disabled people can be excluded from society. For disabled children, who may already be considered a minority group due to their immaturity, the experience of social exclusion can be intensified by the way society views their physical differences; both 'children and disabled people have been treated as 'lesser' because they are positioned as dependent on adults or carers' (Tisdall 2012, p6). Tisdall (2012) suggests that there is a link between studies of childhood which have sought to re-conceptualise children and childhood, and disability studies which has sought to re-conceptualise disability and disabled people. Thus, both childhood and disability studies offer 'theoretical and practical reconsiderations of 'normality', competency, independence and dependency' (Tisdall 2012, p7). Similarly, Connors and Stalker (2007) observe that drawing on insights from the sociology of childhood and from disability studies can draw attention to the perceptions and experiences of disabled children and young people. This attention has the potential to develop better understandings of their lived experiences and acknowledgement of their contextualised competence, capacity, agency and autonomy.



## Agency and autonomy in the sociology of childhood and disability studies

Disabled children and young people can be defined by adult assumptions about their abilities, overlooking their capacity to engage with and make sense of their world and its meanings themselves (Islam 2008). Within healthcare, practices that focus on 'disease processes alone ignoring patients' needs for connection and self-determination' (Speraw 2009) can curtail young patients' agency. In Watson et al.'s (1999) report on life as a disabled child, they observed how the children experienced negative consequences because of their disability. However, the children and young people researched also felt 'more capable and independent than they were given credit for' (Watson et al. 1999, p16), and they were able to exercise some agency through opposing and resisting adult discourses. Agency is a key term throughout my research and, in relation to children; agency can be defined as children being capable social actors in their own right (Morrow 2011). In more general terms it is described as the need for all humans to act on their own behalf, and having a capacity for self-determination (Speraw 2009). Speraw enumerates some elements of what it is to be a self-determining human, listing the:

multidimensional, integrated, and autonomous, with biological, psychological, social, and spiritual elements: possessing inner drives toward wellness and development; having worldviews influenced by culture; capable of suffering, compassion, and moral decision making. (2009, p733)

It is recognised that there are limits and tensions regarding agency, and that children can be regarded as 'both constrained by structures and as agents acting in and upon structures' (Prout & James in Morrow 2011). In this context agency is generally understood as the capacity to act and make choices, whilst also being impacted by the actions and choices of others.

Autonomy is another key term in my research, and it can be understood as the right to personal freedom of action (Drake 2001) and self-directed free choice (Larcher & Hutchinson 2010). Autonomy means that the individual is able to act freely in a chosen manner with the 'capacity to accept or refute information or desires independently of another's manipulation or coercion to accept' (Whitty-

Rogers et al. 2009, p747). Some critics contend that an emphasis on the autonomous individual that defines 'the self as individualistic, isolated and ahistorical... [and] independent of the social context in which they are situated' (Ho 2008, p128) ignores the relational dimensions of autonomy. For those living with a significant disability, atomised notions of autonomy may be unhelpful; their experience of agency and autonomy can be better understood using a language of relationship and support, which does not override autonomy but supports and engenders it. For example, Dreyer et al. (2010) interviewed Danish men who have DMD, and Dreyer et al. found that the participants, despite their profound impairment, described themselves as living autonomous, outgoing lives. The men reported feeling strong and independent, and the meaning of autonomy for them was 'autonomy of... thought' (2010, p2). This sense of autonomy does not mean that those with DMD are free from the physical and social constraints of their condition. Dreyer et al. (2010), Gibson et al. (2009), Abbott and Carpenter (2009) and Abbott (2012) all comment on the sense of isolation experienced by young and adult men with DMD, due to 'social isolation... [and] exclusion from adult relationships (Abbott 2012, p241). However, despite these negative experiences, the men in Dreyer et al.'s (2010) research considered themselves to lead autonomous lives and they valued making their own decisions, which were central to their sense of agency and autonomy.

Tisdall (2012) questions an unproblematic theorising of children's agency and independence without also acknowledging the limitations of some children's contexts. She acknowledges that early leaders of both disability and childhood studies had to fight the dominant paradigms of psychologically focused development, and this required a strident approach in asserting emerging concepts such as children's agency. As Tisdall (2012) suggests, whilst it is right to speak of and champion agency, it is also appropriate to recognise that agency may, in some children's lives, be marginal because of social inequalities and the failure of adults to attribute agency to children. This marginalisation can occur when adults do not recognise self-determination in disabled children's lives. Davis and Watson (2000) recount how a care worker described the disabled children they cared for. The worker observed that the children 'find it very difficult to make up their own minds.

We have to tell them what they want to do, help them decide' (Davis & Watson 2000, p214). After observing the care workers' interactions with the children Davis and Watson (2000) comment that:

We soon came to learn that very often children were denied agency not because they were incapable of making choices, but simply because their ability to make choices went unrecognised. (Davis & Watson 2000, p214)

The Viper Project, which has involved the direct participation of young disabled people in its research activities, is a partnership project carried out by the Alliance for Inclusive Education, the Council for Disabled Children, NCB Research Centre and the Children's Society. It has explored the levels of participation disabled children and young people experience in a range of decision-making situations. In their findings the researchers comment that some groups with a disability were notably excluded:

Some groups of disabled young people are less likely to participate than others... those in care, disabled young people from black and minority ethnic groups and those with significant support needs. Some reasons given for excluding such young people were lack of staff time to facilitate communication and assumptions by parents, carers or staff about their ability to participate. (Viper 2012, p22)

It is apparent that, in some instances, adult assumptions and practices can restrict disabled children's agency and limit their participation in decision-making. Adults may assume that disabled children will react in specific and pre-determined ways, and as was seen in Davis and Watson's (2000) example above, disabled children can be treated as passive and incapable of change, growth or adaptation.

Child development theories frequently stress that a stable, secure life is necessary for children to flourish; however, this ideal is problematised by childhood adversities, including disability (Boyden 2003). Boyden (2003) asserts that children can cope and develop despite adversities, and she comments that they do not react in a universal way to difficulties, and nor are they inherently vulnerable. Innocence and vulnerability may be regarded as innate characteristics of children and childhood, denoted by children's smaller or weaker bodies, or their lack of social skills, and these constructions can lead to further marginalisation:

Children's practices, decisions and ways of reasoning are generally not awarded the same status as those of adults because they are considered immature. These structures make children relatively powerless and structurally vulnerable... they can produce the kind of personal lack of social experience and social vulnerability that the discourse of innocence portrays as innate to children. (Meyer 2007, p90)

Vulnerability can be experienced by children and young people at times, for instance when they are in pain, ill or undergoing a medical intervention; but being informed about their treatment plan can help children to 'appreciate the logic and good intentions underlying painful, frightening procedures' (Alderson 1993, p134). Vulnerability is not a global property of children and childhood; it is context-specific and can be addressed in ways that do not undermine the child's potential and capacity for agency.

Boyden (2003), a professor of international development and director of 'Young Lives', a study of childhood poverty, challenges ideas of children's innate vulnerability. As suggested above, she attests that agency can be present despite acute or challenging circumstances, and she develops some engaging ideas highlighting children's adaptability and capacities. Boyden comments that research focusing on 'psychopathology tends to omit important environmental and relational dimensions of young people's wellbeing' (2003, p7). Boyden discusses the resilience and coping strategies of children living in areas of armed conflict, although her observations have a salience beyond this. She argues for children to be positioned as capable 'agents of their own development' (Boyden 2003, p1) who can foster coping mechanisms despite living in unstable conditions caused by a range of problems. Boyden draws from the sociology of childhood to illuminate the point that children can be active agents, involved in interpreting the 'world about them, making choices... [and] reaching decisions' (Boyden 2003, p18) which, for disabled children and young people can be a significant means of taking control (Guell 2007; Speraw 2009). The points raised by Boyden are important, as doctors and parents may feel it their duty to protect children from the hard facts of their health condition. However, children may wish or should at least be given the option to be more involved, providing them with opportunities to enact some agency and autonomy over the decisions and procedures necessitated by their condition. Blue-Bond Langner's

(1978) research on hospitalised children dying of cancer revealed how children can be acutely aware of their health status even when excluded from information on it. Blue-Bond Langner, an anthropologist, documented how the hospitalised children acted within their world as knowing agents who could read and respond to adult distress, through maintaining a mutual pretence regarding their expected death (Blue-Bond Langner 1978, p204). Children can absorb information, both verbal and non-verbal, from the adults and the circumstances that surround them, and, extrapolating from Blue-Bond Langner, adults' silence on serious issues might also silence children. When information is withheld from children and they comply with this behaviour it may indicate children's 'motivation to conform to socially desired behavioural norms for children, rather than [be] a simple demonstration of moral immaturity' (Carnevale 2004, p403).

The role of parents is significant in their ill or disabled child's life and they can both enable or deny children information about their condition; where they work with children to keep them informed, this can support the child's comprehension of information regarding their health (Coyne & Harder 2011). Collaborative approaches to decision-making, that involve the child's parents and doctors, can be complex due to multiple contributing views, but they can also be beneficial, with the anxiety of decision-making shared between children and trusted adults (Alderson & Montgomery 1996). Many of the children Alderson and Montgomery interviewed identified their own maturity through having more equal discussions with adults, rather than decision-making in 'lonely autonomy' (Alderson & Montgomery 1996, p157). In this setting, autonomy becomes a shared, relational form of autonomy, and this has relevance to the context within which the participants in my research are likely to manage their daily lives. It is a useful concept for understanding how disabled children and young people work closely with trusted adults. As will be seen in the next section, relational autonomy is a way of managing the demands of decision-making within a familial context.

### *Relational autonomy*

Relational autonomy is based on the valuing of relationships and interconnections

between people in which they often share decision-making with others in their social network. Gibbons (2008), a researcher in law, describes relational autonomy as being built around notions of interdependence, and it has been informed by, amongst others, the work of feminist writers. Feminists have criticised the ideals of autonomy as being atomised, individualised and rationalistic (Mackenzie & Stoljar 2000). One of the effects of atomised notions of autonomy is an absence of the 'inherent significance of intimate relationships, which are characterized by collectivity' (Ho 2008, p129). Gibbons (2008) does caution that an over-emphasis on interdependency may overlook the individual's right to self-determination; nevertheless, relational autonomy provides a means to understand how individuals draw on the support of others. Its exponents argue that self-government and the individual freedom to choose does not give 'sufficient weight... to the fact that people make decisions within the context of... relationships' (Gibbons 2008, p1-2). Gibbons states that:

we are not constantly clashing rights with those we live with, rather our interests are intertwined. People in close relationships seek a compromise which is good for "us" and do not see it as a matter of weighing up competing interests. (2008, p2)

In families with a son who has DMD, Abbott and Carpenter (2009) observe that the rights, autonomy and voice of the young men are bound up with their parents. This shared identity is partly because the families have to fight for support, which can develop an 'us' and 'them' outlook (Abbott & Carpenter 2009). In such families the parents and children regularly collaborate to reach decisions and this collaborative approach has evolved within 'the family's history and contributes dynamically towards its future' (Ho 2008, p130).

Dreyer et al. (2010) discuss the nature of independent dependency as a lived experience for those growing up with DMD and, as noted earlier, they observe that the young men they interviewed recognise they are 'physically dependent, but they describe themselves as independent human beings' (Dreyer et al. 2010, p5). The care they receive has had to intensify as disability has increased, but this does not alter the men's sense of selfhood, or their sense of autonomy. Dreyer et al. (2010)

argue that:

The lived experience of physical impairment is found to be “independent dependency”... the findings in our study indicate that physically impaired people experience dependency but in a manner which nevertheless is not regarded by themselves as being dependent. It is something known and accepted and as far as possible disregarded. (Dreyer et al. 2010, p6)

For disabled individuals, having the physical and emotional care and support of their family and ‘the belief of others in their value and worth as persons’ (Speraw 2009) can help to sustain and enrich their life. Ho (2008) discusses relational autonomy for adults who are ill and making serious decisions on their care. She observes that ‘family involvement can be integral in promoting patients’ overall agency’ (2008, p128). Healthcare is often fragmented and impersonal, and against this backdrop the role of familial support can preserve or restore ‘autonomous agency’ (Ho 2008, p131) and a sense of self. Protective parenting may negatively influence relational autonomy at times, as Carnevale et al. (2006) found, parents of children who are ventilator-dependent thought that they should protect their children from being directly involved in decision-making. This reveals how parents face a ‘complex relationship between striving for increased autonomy for their children and providing protection’ (Barron 2001, p439). Relational autonomy and shared decision-making are, despite the problems of over-protection, likely to be the approach used by many families who have an ill or disabled child (Whitty-Rogers et al. 2009), with the child being supported as they make hard and occasionally life-changing decisions.

Disabled children and young people’s acceptance of their parents’ help can demonstrate their competence as social actors who value support; it is observed that even quite independent young people may ‘wish their parents to retain formal responsibility for decisions’ (Alderson & Montgomery 1996, p90). Delmar et al. (2006) assert that, in a relational view of living and coping, we are all dependent on each other and that living in dignity can incorporate others into decision-making. They state that ‘including other people into one’s life situation can be an important sign of self-management’ (Delmar et al. 2006, p266). Delmar et al. conclude that:

Independence, self-control and taking responsibility for one’s own life can

evolve as significant values... within the framework of a relational view of the human being. (2006, p267)

Disabled children and young people can develop decision-making capacities within a supportive, connected setting where 'lived autonomy is related to individuals' connection with others' (Whitty-Rogers et al. 2009, p747). Sometimes the line between support and undue influence may be hard to identify, and this may be notably so with a severe, degenerative condition such as DMD. Yet relational autonomy offers a space for negotiation and collaboration (Whitty-Rogers et al. 2009) wherein children and young people are treated as actors in their lives (Cockburn 2005), negotiating and engaging in complex issues and decisions.

Relational autonomy recognises disabled children and young people's competencies and abilities which, with support, can be nurtured and encouraged. However, in the discourse of social and healthcare policy and practice, children can be unproblematically positioned as vulnerable, immature and lacking in competence:

In social welfare policies, children are frequently seen as being at risk of abuse... The dominant idea is that children are vulnerable which may lead to an overemphasis on 'rescuing' children, and a lack of emphasis on enabling them to 'participate'. (Morrow 2011, p7)

There are alternative ways of considering and positioning disabled children and young people, and discussions of competence and consent emanating from the sociology of childhood and disability studies provide constructive ways of thinking about such matters, which will be discussed in the next section.

### Competence and consent in the sociology of childhood and disability studies

As has been seen, children's competence can be questioned by adults, and in informed consent procedures children are regarded as needing protection and adult intervention. When children have a significant disability, informed consent can be considered even more risky than with a 'normal' or 'mainstream child' (Cocks 2006). Whilst the sociology of childhood has promoted children as competent agents in their own lives there has been less constructive recognition of the difficulties in assessing



agency and competence, and managing consent issues, with children who have disabilities. Disability studies generates knowledge on the experiences of children who have contextualised vulnerabilities and who may be 'omitted from explorations of children's demonstrations of agency' (Cocks 2006, p254-5). Difficulties in accessing and working with ill and disabled children in order to evaluate their thoughts and experiences of their healthcare provision can lead to their views being marginalised (Clavering & McLaughlin 2010). The result can be that healthcare research is less fairly representative of children than it should be, due to concerns over consent issues. Carter (2009) discusses some children's marginalisation in healthcare research, whereby the vulnerability that is ascribed to children, and particularly those with significant disabilities, is readily linked to the notion that they lack competence. She observes that children who are perceived to be 'the most vulnerable are most under-represented within health-care research even though they are high consumers of services' (Carter 2009, p859). Because some disabled children's voices are underrepresented in healthcare research, assumptions about their competencies can go unchallenged or even be reinforced. The corollary is that poor understandings of disabled children and young people's competence can impact how they are involved in their healthcare and in medical research decision-making.

Davis and Watson (2000) discuss how disabled children who are already positioned as vulnerable are further isolated by the way in which their competencies are overlooked by the adults who care for them, despite legislation:

Most of the guidance and legislation... that covers the rights of disabled children... are also covered by a proviso that the children should exhibit competency. In our experience competency is denied to many disabled children in different settings by a variety of adults. (Davis & Watson 2000, p214)

Adults may consider children to be 'incomplete people' (Mayall 2001, p255), thus compromising children's opportunities to demonstrate competence, because adults are not prepared to question their own assumptions. Adults' assumptions can 'stem from a lack of dialogue which occurs because an adult refuses to adapt their first impressions of a child' (Davis & Watson 2000, p216), which may be formed using

inflexible methods of determining a child's abilities:

Many disabled children are aware that adults pathologise them on the basis of apparently objective biological, educational, social or cultural criteria... Hence, as a result of this it should not be assumed that disabled children's non-compliance, silence, or resistance is incompetence. (Davis & Watson 2000, p219)

As Davis and Watson observe above, adult judgements of children's abilities are 'very often 'measured' by their educational attainment' (Morrow 2011, p5), and further judged according to sociocultural and 'objective' criteria; these presumptions can be difficult for disabled children to disprove.

Valuing disabled children and young people's unique perspectives and viewpoints (Ireland & Holloway 1996; Minkes et al. 1994; Speraw 2009) can raise 'new insight into children's worlds and into their relationship with the adult world around them' (Clavering & McLaughlin 2010, p606). Adults may assume that disabled children lead marred or damaged lives (Morrow 2012), which impacts how they think about and interact with disabled children, although these same children may think of themselves as ordinary, with their health condition being normal to them. Children with cystic fibrosis in Williams et al.'s (2009) study demonstrated competence in managing their condition whilst maintaining school relationships and a social life. This involved complex negotiating between compliance with their health needs and a desire to develop independence and maintain 'normality' in the face of a disruptive condition, demonstrating contextualised competence (Williams et al.'s 2009). Children with ongoing experience of ill health or disability can develop sophisticated decision-making skills through the experience of managing their physical condition (Alderson et al. 2006; Dixon-Woods et al. 1999). Alderson (1993) suggests that 'children become competent by first being treated as if they are competent' (p173), indicating how competence is not a static entity but is situated in sociocultural and infrastructural contexts that together impact on lives (Watson et al.1999).

In a discussion on informed consent, Young et al. (2010) were part of a larger project at a children's research hospital, investigating communication problems within the hospital. Teenage cancer patients were interviewed for their perspectives

on informed consent in paediatric cancer research. Young et al. (2010), quoting Corrigan, state that most theorists write about informed consent from a bioethics standpoint, and they were concerned to understand how the teenage patients experienced and perceived the process of consent. They discovered that teenagers with cancer often felt that decisions were being made for them, albeit in their best interests. Some thought that the purpose of the informed consent process was to explain to the teenager and their family what was going to happen to them; the 'only power that these teens felt they could exercise was the ability to sign the consent form' (Young et al. 2010, p637). This last comment demonstrates how the purpose and function of the informed consent process may be poorly explained, or understood, by some young patients. In another example, an ill young woman who was old enough to give her own consent is quoted as saying that she approached making a specific decision by asking her parents 'what they thought and what they felt we should do' (Young et al. 2010, p637). This latter example suggests how even quite mature young adults with DMD/MD may approach decision-making and consent processes. They have a life-limiting condition and they live in close relationships with their parents and they may sometimes elect to make supported decisions. As was discussed in the section on relational autonomy, close relationships can enhance competence and the approach to consent. Thus, for disabled children and young people, whose competencies may be poorly understood or questioned by some healthcare professionals, the presence of parental support during decision-making can be an asset. Alderson and Montgomery (1996) assert that, during decision-making, parental support and advice enshrines 'the usual and ideal position that most decisions should be taken by parents and children together' (Alderson & Montgomery 1996, p90).

However, the relationship between parent and disabled child may sometimes lead to competence and consent issues being difficult to clearly identify as either supportive or overly influenced by parents. Abbott (2012) represents something of the intertwined nature of relationships between parents and their sons with DMD in the following dialogue. Taken from Abbott's research, the mother of a son with DMD has been talking about a cure for DMD, which she does not envisage happening. Neither, she adds, will 'they' be taking part in any 'silly trials', as she then explains,

with reference to her son:

Mother: And we decided then didn't we, it's not worth doing. If you've got it you've got it... we just let nature take its course really haven't we?

Young person: I used to take morphine didn't I?

Mother: Yeah

Young person: Now I'm on Fentanyl. It helps a bit doesn't it? But I won't up the dose.

Mother: No

Young person: Will I?

Mother: No. (Abbott 2012, p245-6)

Abbott observes that he is slightly unsettled by this interaction and wonders:

Does he [the son] remain unsure or convinced that his pain management is right? The mother here is telling a story, which unites her and her son in common purpose, but the interaction between them... throw this into question. (Abbott 2012, p246)

The dynamics of parental relationships with their disabled children highlight issues for my research about how competence, agency and autonomy are viewed, and how informed consent can be achieved in ways that appropriately involve children and young people. The question of what the children and young people themselves think about the decision-making process is germane; are they in agreement with their parents on various issues around medical research or is there the potential for conflict? These questions can challenge assumptions about disabled children and young people's agency and capacities, and in the main chapters of my research the participants' views may help to inform better understandings of how agency and competence are achievable and demonstrable despite having a serious disability.

Broome et al. (2001) interviewed children and their families on their experiences of taking part in medical research. They discovered that most of the children and teenagers in the study 'had a very complex understanding of their disease, the treatment and how this was going to affect their life' (Broome et al. 2001, p46). They

also summarise their findings by noting that:

informed consent is not a process that begins and ends with a signature on a... document.... Investigators should be cognizant of the multiple factors influencing children and adolescents involved in research and strive to develop processes of including them during... informed consent in specific and meaningful ways. (Broome et al. 2001, p47)

The 'multiple factors' influencing children and adolescents can include the power differentials between them and adults, and parental experiences of hope, denial, isolation and over-protection (McLaughlin 2006; Webb et al. 2005), which can all impact how children and young people are positioned in decision-making.

Recognition of disabled children and young people's contextualised and situation-dependent competence (Davis & Watson 2001) can help to build their confidence that they will be listened to. Morris (1999) cites the ways in which disabled people are discriminated against and ignored. She quotes from a group of young disabled people who criticise the medical model of disability:

For many years doctors, social workers and other people have told disabled people that they are disabled because of 'what is wrong with them'... this is known as the medical model of disability. It says that it is the person's 'individual problem' that they are disabled. (Morris 1999, p5)

When disabled children and young people are measured against standards of normative physical development and well-being, their personhood and competencies are diminished and their rights compromised (Davis & Watson 2001).

Healthcare and medical research decisions may be reached with the welcome help of parents, although in some instances parental influence may be unwelcome and the giving of proxy consent can sometimes fail to sufficiently include children's own preferences and capacity for agency and autonomy (Alderson et al. 2006; Davis & Watson 2000; Whitty-Rogers 2009). Whilst informed consent is regarded as upholding autonomous decisions, for some disabled children and young people autonomy can be enabled and supported by the involvement of others, drawing on models of relational autonomy. There is little literature dealing directly, or in depth, with disabled children and young people's own values and perceptions related to the

informed consent and medical research process (Broome et al. 2001). Therefore, my research hopes to build on understandings of how decision-making, competence and consent are thought about and experienced by those living with DMD/MD and how the influence of others may guide their approach.

## **Conclusion**

This review has covered some of the issues relating to children and young people's participation in their healthcare and medical research. A review of healthcare policy, guidelines and legal rulings has revealed elements of protection and paternalism alongside calls for children's increased participation in their healthcare and medical research. The protectionist stance taken in guidelines implicitly and sometimes explicitly suggests that children and young people are vulnerable and in need of adult protection. In light of this, my research will question how disabled children and young people can make decisions within the contextual setting of their lives. To further explore this question, issues of competence, informed consent and the related matters of agency and autonomy have been reviewed. These terms merit scrutiny regarding how they are realised as living concepts and processes in disabled children and young people's lives. The shape competence, consent, agency and autonomy take may differ from generalised conceptualisations of these terms, potentially uncovering the inadequate or limiting ways in which disabled children and young people are understood. Being young and having a significant and progressive condition creates unique challenges for how participation in decision-making is enacted. The role of adults in the lives of disabled children and young people can be vital, and also influential, with parents and doctors likely to contribute to how decisions are approached and made.

As observed, the drive to produce effective drugs for DMD/MD may cause some parent-led charities to promote research and research participation in persuasive ways. To address the questions and issues raised, I will explore, in the main body of this research, the participants' own unique views and experiences to gain an understanding of how they think about these terms within the context of their lives. As explained, I am approaching the research from the perspective of the sociology of

childhood which accepts children and young people as being competent and able actors, and the participants' thoughts on medical research decision-making will help to address a gap in research. Contextualised discussions on the experiences of those with DMD/MD can help to improve their sociocultural and healthcare status through a deeper understanding of how they live, their hopes and their presence as citizens and individuals.

In the next chapter I will explain the methodology for my research and my approach to recruiting and interviewing the participants; the specific methodology for the discourse analysis that is conducted in Chapter Five will also be explained.

## Chapter Two

### Methodology chapter

The research has focused on the opinions and experiences of children and young people with a degenerative disability, and in this chapter I will describe the process I worked through to conduct my research. Although there were challenges in getting to the point of hearing from the participants, their contributions are the central concern of the research which is based around their experiences and thoughts. Popular media depictions and public perceptions of children and young people facing challenging situations can portray the child as a victim of their circumstances (Boyden 2003). These portrayals often draw on rhetoric of the child's bravery and of parental sacrifice and this can misrepresent the reality of living with a disability. As was established in the literature review, my research positions the participants as socially engaged and active contributors who can inform how we think about and understand the ways in which decisions to take part in medical research are made. In the process of gaining this understanding it has become apparent that the children and young people work together with their parents, yet also achieve some measure of independence. Therefore, this research is not just about medical research decisions; it is also about the broader context in which collaboration and supported autonomy are lived out. The participants' contributions add another dimension to discussions on what it is to be social beings and the varying shapes that agency and autonomy can take.

This chapter will begin by discussing research with children and young people, before moving on to explain qualitative semi-structured interviewing techniques and my preparations for conducting the interviews, including developing my ice-breaker questions and my research schedule. Then I will explain some of the practicalities of how I conducted the interviews and the follow-up interviews. There will then be a discussion of analysing data, ethics approval, sampling and recruitment, ethical issues and the limitations of my research; in the final section I outline the methodology for the discourse analysis.



## Research with children and young people

Assumptions that able-bodied and disabled children and young people are not competent to be interviewed are, clearly, detrimental to promoting their participation in research (Cocks 2006, Coyne et al., 2009, Franklin & Sloper 2005). There has been a recognition that children themselves, including those with a disability, should be asked for their opinions, with a shift of emphasis from information about children to information from children (Christensen & Prout 2002; Docherty & Sandelowski 1999). This attitude has motivated this research, which considers disabled children and young people to be the experts in narrating their own perspectives (Christensen 2004). Due to their immaturity, my participants may be thought of as a minority group (Hood et al. 1996) who need protection (Balen et al. 2006); and hearing from them, whilst revealing more about their lives, may also reveal more about the attitudes, rhetoric and disparities of power present in the adult world. The participants' perspectives can bring insight to how adult practices are experienced by those who are frequently in a less powerful position.

Issues of power also relate to the process of interviewing, where my own presence will have had some bearing on the interviews:

the dimension of power in interviews is most important... neither party leaves behind any asymmetries of power in the wider society when they... enter a room for an 'interview'. (Wengraf 2001, p196)

However, the sociology of childhood advocates children's potential for agency and action (Boyden 2003; James & Christensen 2008; Mayall 1996), therefore, overly stressing the interviewee's vulnerability may wrongly assume that interviews are potentially harmful, whilst failing to regard the young participant's agency in the interaction (Corbin & Morse 2003). Valid concerns about the participants' vulnerability are balanced by the possibility that they may appreciate an opportunity 'to not only name their reality, but to begin to look at that reality in different, and more critical, ways' (Berman 2003, p111). Therefore, whilst conscious that my status as a mature woman from a university might be considered intimidating, dull or 'other' to them, I was mindful that the participants had agreed to meet with me. Their agency

in making this choice was not, to the best of my knowledge, coerced by their parents, and I tried to ensure during initial contact with parents that their child was keen to take part; therefore, I respected the participants' contributions as being willingly given.

As a means to dissipate power inequalities, social researchers can try taking the 'least adult role' (Harden et al. 2000), by attempting to become more child-like. However, Harden et al. (2000) question this as a viable strategy when interviewing children and young people. They suggest it may be more productive to acknowledge that the interview relationship is one of inherent inequality. It could be construed that taking the 'least adult role' is disingenuous, setting up a relationship with the interviewee that is falsely premised. Christensen (2004) proposes seeing children as fellow human beings, with the researcher approaching the interview as:

one who is seriously interested in understanding how the social world looks from children's perspective but without making a dubious attempt to be a child. Through this the researcher emerges first and foremost as a social person and secondly as a professional with a distinctive and genuine purpose. (2004, p174)

Thus, I sought to avoid being overly friendly with the participants but equally, I avoided 'talking down' to them. I tried, within the confines of the interview, to remain my natural self and hoped the participants would do likewise. I listened carefully to the personal accounts of my participants, and to their reasoning, my attitude of listening was as an acceptance of different beliefs and of lives in which those beliefs make sense (Frank 2000, p361).

The interview is inevitably a more formalised setting than is the case with a spontaneous discussion, yet to overly problematise it does the interviewee a disservice and I have assumed that they are 'competent and reflexive in reporting their own experiences' (Balen et al. 2006, p43). My anxiety regarding my, relatively, more powerful position as an adult is somewhat minimised by considering the interview to be a collaborative one. In the interview, knowledge is constructed in a two-way relationship, viewing both interviewer and interviewee as active participants (Holstein & Gubrium 1997). Harden et al. (2000) contend that these co-

constructions are valid observations on aspects of the participants' social setting however partial they may be. Whilst mindful of the interview as a co-construction, I was, ultimately, the one guiding the encounter and I endeavored to work creatively and sensitively, using various approaches to engage with methodological issues as appropriate to the interviewee and their abilities (Clavering & McLaughlin 2010; Darbyshire et al. 2005). I was aware that my approach as a social researcher called upon my moral obligation to maintain ethical sensitivity despite my need for data (Ireland & Holloway 1996).

In some cases, interviewing children and young people can be different as they are infrequently asked their opinions because 'in a variety of everyday contexts adults speak for children' (Harden et al. 2000, p10). Certainly, some of the boys did not willingly enter into prolonged discussion with me and this could be for a variety of reasons including the quite commonly held view that boys can be less forthcoming. Mahon et al. (1996) report from their interviews with a range of children that those with boys aged from 9 upwards were less successful than other interviews. Although, Harden et al. (2000) state that even partial or truncated answers are useful forms of data. The brevity of some participant's answers could be due to my own lack of experience, their lack of interest, or their lack of energy on the day; any of these reasons, plus many others may have hindered discussions. Several of the boys are at the stage of coping with respiratory problems caused by weakening muscle strength, and this can result in tiredness, headaches and shortness of breath (Action Duchenne 2010).

It has been complex to gain access to the participants due to the challenges of ethical clearance, gatekeeping and recruitment, and these will be further discussed in the following sections. However, hearing directly from the participants, rather than adult proxies (Valentine 1999) is timely in highlighting their presence in health, social care and society. The next section will explore the qualitative approach to data gathering and further explain how I conducted the research.

## Qualitative semi-structured interviews

Quantitative methods such as questionnaires can:

preclude children from constructing and reporting their own reality, as reality is already defined by a limited and predetermined set of responses... the complex realities of children's lives are reduced to scores on instruments. (Woodgate 2001, p151-2)

Interpretive research methods are 'more likely than 'hard' methods to elicit children's views' (Alderson 1993, p67) and for my research, qualitative semi-structured interviews were responsive to the interplay of each encounter. In conducting semi-structured interviews I was able to explore the participants' lives as they are, accepting their accounts as being their active response to my questions and prompts. This situates their reasoning and their experiences as those of contributing social actors (Mayall 2000), rather than thinking of children and young people as incompetent or unreliable reporters. It also acknowledges that life with a disability places even young children in a position of experientially gained knowledge that may not be limited by their age or developmental stage (Coyne & Harder 2011; Woodgate 2001). If provided with an appropriate opportunity to vocalise their knowledge and skills, children and young people are capable of sharing some of the depth of their experiences. This assists our understandings of their health related experiences, with qualitative methods offering a viable medium for exploring these. However, as in any social interaction there is the possibility for silences and gaps in conversation, which adults can be more adept at filling, as silences can be uncomfortable. In some of my interviews there were times when I had to stop myself from anxiously filling the silence, particularly with one participant who was very quiet and shy. Abbott & Carpenter (2009), in their interviews with young men with DMD note that there were moments when discussion drifted and they would, for example, play a computer game with their participant. Although I cannot play computer games, I did try to leave time for participants to think, sometimes giving prompts to help them, or if one line of questioning was not helpful then using alternative phrasing or moving on to other topics. Ireland & Holloway (1996) suggest that in rigid interviewing techniques there is a tendency to use prolonged, repeated questioning which departs from

normal conversational conventions. Qualitative methods, in using a more conversational approach, are a viable means to further our understandings of children and young people's experiences as 'embodied health care actors' (Mayall 1998, p270) whose insights matter.

As an ESRC CASE studentship, the main focus of the research was already established prior to my starting the project, although this focus was refined as I took up and thought about the research from my own perspective and as I sought to make it into a personalised body of work. To begin with I produced some 'ice-breakers' (see Appendix B) that would initiate the research encounter, the aim was to build a rapport with each participant and to overcome any nerves, both for me and them (Beresford 1997). Over time I did not need to refer to the ice-breakers, as naturally occurring talk spontaneously presented itself, as it often does in everyday life. An interview schedule (see Appendix A) was devised with supervisory guidance and this gave me bullet-point reminders of key questions and prompts. The questions for the interview schedule developed from the lead questions in the primary research proposal; these lead questions were itemised in Table 1 in the Introductory Chapter, and they were the starting point for generating the schedule questions. The initial questions covered general issues around the participants' daily lives and then moved closer to more direct questions about decision-making. This format gave me flexible and open questions and prompts if needed, whilst offering the possibility of exploring interesting points in more detail, without straying from the topic's central themes.

It took several attempts to develop appropriate questions and once in place this schedule offered a dynamic means of monitoring and improving on my interview questions. After conducting 3 interviews it was apparent that I needed to refine my techniques in exploring medical research as a topic with the participants, they seemed unsure what I meant by medical research, or were struggling to comprehend the issue as it related to their lives and condition. This could be considered an initial research finding, as many of the participants appeared not to be overly familiar with the processes and conduct of medical research. With my supervisors' support I devised some scenarios (see Appendix C) to engender tangible discussion with my

participants, and these scenarios provided a flexible structure and 'talking-point' for my interviews. The scenarios depicted situations where a fictional young person is thinking about taking part in some medical research. Where time permitted, these scenarios were mailed-out prior to my visit and they mostly proved useful by engaging with the participants' imaginative skills and placing them in another's situation. I thought that by putting an imaginary boy in the scenario it might give my participants some critical distance to explore the setting. Two participants commented that it was hard, or illogical to answer for someone else, although the scenarios still provided them with a means to make some useful comments. Overall, the scenarios contextualised medical research, making it readable to the participant's own setting and life.

Using a flexible approach to the interviews worked well with Tim, my youngest participant (aged 10) who did not respond to some of my questions and was perhaps unfamiliar with the format and purpose of the research interview (Irwin & Johnson 2005). If a more structured schedule had been employed Tim's comments and thoughts would not have been given the opportunity to develop, and qualitative methods enabled me to adapt to his style of communicating. Ollie (14), who has Asperger's syndrome, did not always cope well with direct questions, and this could have been due to a lack of confidence when we touched on issues he was less sure of, although with other topics he was confident to express himself clearly and uniquely. As will be seen in the data chapters, he produced some ideas that were unconventional and highly imaginative. He had a speech impediment that made it hard to understand him at times, but I avoided asking him to repeat himself too much by trying different approaches to the question, or moving on to other questions. With Tim and Ollie I judged that it was not helpful to try and keep them too much on topic as they did not reply to some questions or they gave answers that were related to other subjects. I found these two interviews the most demanding, but I felt that however off-topic we were going this was hard to avoid without being overly didactic in my approach, which would not be in the spirit of my research.

## **Conducting the interviews**

As I prepared for the interview phase of my research, I consulted, via email, two social science researchers who had separately conducted research into aspects of DMD and they offered useful advice on recruitment and interviewing. Both commented on the challenges they faced, noting that participants may be hard to recruit, and that there can be problems with engaging participants in conversation. It was apparent that parents may try to do most of the talking if they are present at the interview, and I was also advised to give space for participants to answer if they have impaired respiratory function. Prior to conducting the interviews I held a rehearsal interview with a local man who has Duchenne; this gave me an opportunity to try out my questions on someone with DMD and to ask him for advice on how I could improve my questions and approach. I asked him to think how a boy or younger man with DMD might respond to the questions and he made some interesting observations about his own experiences when he was a teenager. Pilot interviews would have been useful, but as there were so few potential participants it was more practical to conduct a rehearsal interview. The interviewee responded well to the questions, although I was conscious that as he speaks at conferences, he has developed skills as a communicator that may be less apparent in my participants. This interview helped me to prepare for my main interviews and to practice positioning my digital recorder correctly, coping with noisy pets, making field notes and transcribing the interview.

Parents mainly contacted me by telephone or email to express an interest in their child being interviewed, with one parent contacting me by post. I was then able to initiate further contact with them by telephone to discuss the interview in more depth and to set up an interview date and time. All interviews were conducted in the participants' homes at a time to suit them. At the start of each interview I ensured the participant had read the information sheet which I had sent and I explained a little more about my project and answered any questions they had. I then worked through the consent and assent forms with them, asking them to sign the form if they were happy to continue. For those under 16, they could sign the Competent Minor's and Over 16's Consent Form (see Appendix D), if, after a brief discussion with them and

their parents we all agreed this was acceptable. The 10 year old I interviewed signed the Assent Form (see Appendix D) and his mother signed the Parent/Guardian Consent form (see Appendix D). Consent and assent forms had been included in the mail-out packs to each address so that participants could read them ahead of time. I explained that if, during the interview, the participant said anything that suggested a risk of harm to them or someone else, then I may have to stop the interview and decide on a course of action. Although some of the participants had very little hand movement, they were all able to sign their form. I asked permission from each participant to use a digital tape recorder, a matter that had been mentioned in the information sheet, and I confirmed that the recording would be stored and encrypted on a university computer. I informed the participants that they would receive a transcript of their interview when I had typed it out and explained that I would replace their name with an alternative (pseudonym) in my research. I noticed that almost all the participants and their parents used the word 'condition' to describe their DMD/MD and so I too have adopted the word.

Silverman (2005) considers how the taking of field notes is a first stage of analysis in qualitative research, during which an initial reading of the setting and the activities observed and experienced are noted down and worked with as part of the process of analysis. As soon as I left each home and boarded public transport or located a nearby café, I would write down in field-note form my initial thoughts and impressions from the interview encounter. Not only are these useful to look back on, but they also sealed those early thoughts in my memory through the act of writing them. This has helped me to recall the atmosphere, sensations and the 'feel' of each research encounter as an added source of data.

### Social researcher and 'polite guest'

I was aware the participants may want to avoid hurting their parents' feelings by not discussing some issues in front of them; this was mostly avoided as the majority of parents left the room during the interview. During my initial contact by phone or, upon meeting each parent at their home, I would clarify if the parent was to be present during the interview. In 3 of the interviews parents opted to remain in the



room. In one instance this clearly gave the participant courage and he was reassured by his mother's presence, this reassurance has also been observed in interviews with younger children (Coyne et al. 2009; Irwin & Johnson 2005). In the other cases the parents had a more authoritarian, protective role, with one mother frequently answering for her son. This in itself was insightful, suggesting how these parents helped their sons to manage their lives and the challenges they encounter. As a guest in her home I could not ask the mother who answered for her son to leave the room, I was very aware that I was both a researcher and 'polite guest with the "rules" and obligations connected with this' (Yee & Andrew 2006, p407). To try to fully include her son I directed questions at him, making eye contact and using his name, but it would have been impolite to ignore his mother's input. The mother may have considered the interview an opportunity to voice her own account (Guell 2007), which to some extent she did. I think she also wanted to support her son, and it could be that what he perceived to be the formality of the interview was a little intimidating. This is possible as he had been very personable in the kitchen as hot drinks were prepared, becoming quieter when the actual interview was conducted. He then returned to his more talkative self when I said that we had covered the main questions. Fortunately I made it clear the tape was still running as, at this point, he visibly relaxed and added some very interesting comments that have enriched my insights on living with DMD. This was my first interview, and contact with the mother had been brief, I wrongly assumed that her stated keenness for her son's voice to be heard equated to her not being present during the interview. This made me aware that in subsequent interviews I needed to manage more clearly the transition from being invited into each home and chatting with parents, to starting the interview and establishing courteously if parents were to stay or leave the room.

In one other interview, both mother and father remained present, and they let their son answer for himself, just occasionally interjecting information. It is unclear whether he would have been more forthcoming if interviewed alone. His father waited until the formal interview was over before voicing his own very detailed views on medical research and DMD. The complexity of young disabled people's relationships with their parents may make them unwilling to express personal views which oppose or challenge those of their parents, who they rely on for care (Skär

2003). It would be improper to cajole information from any participant, and yet the researcher must also create a situation in which the participant feels they can talk openly. These young people and their parents are coping with extraordinary demands on their material and inner resources and living these complex lives in as ordinary a way as possible. Thus, I followed the participant's lead in the depth and tenor with which topics were covered, and I avoided asking questions on sensitive issues, as this was not the focus of my research. The one young woman (21) who was interviewed and who has a rare form of muscular dystrophy was very easy to talk with, raising matters I had not mentioned or even thought of, and she provided some varied and candid observations. There is not room here to explore the gendered issues of young women versus young men as interviewees; however, her input was a different experience from the other interviews. Her readiness to explore topics demonstrates that qualitative interviews are uniquely influenced by the dynamic interplay between those present. Her different diagnosis and prognosis may also be contributing factors, her disability is significant and will deteriorate over time, but she has much more physical mobility, and the possibility of a more open future.

As all of the participants were interviewed in their homes, this provided familiar surroundings and a comfortable atmosphere; the participants were close to their assistive devices and to their parents, and the setting allowed them greater control of their environment (Ireland & Holloway 1996). From my own experience with my three sons and their friends I know that being overly direct or formal is unlikely to produce a good rapport. Indeed, Beresford (2004) suggests that minimising face-to-face contact can help alleviate social anxiety or nervousness. As interviews were conducted in a lounge or the participant's bedroom, I was able to check with the participants where they wanted me to sit.

During two interviews the participants became restless as their energy levels or interest waned. This was signaled by shorter answers or by the loss of eye contact and these can be ways in which consent is withdrawn or withheld by children and young people (Mahon et al. 1996). Hood et al. (1996) observe that although young participants may have given consent to be interviewed, they may nevertheless

exercise a withdrawal from interaction whilst still apparently participating, through lapsing into monosyllabic answers. In my interviews I regarded these as signs the interview should either change pace or be drawn to a close. In one instance, the participant, who was the youngest in my sample, directly asked when the interview would end. He had looked bored and tired, and so I said we would finish immediately if he wished. In the other example, the participant began shifting his wheelchair and commented that his father looked like he was checking if the interview was over, so I took my cue and said that we would move into the final stages of the interview. In retrospect, I wonder if it would have been appropriate to give each participant a small item, a red card, for instance, which they could have pointed to or held up, if able, when they wanted 'time out' or to end the interview (Alderson et al. 2006). This could have given the participants more control of the process without having to negotiate the tricky requirements of telling an adult they wanted the interview to end. I did ensure that none of the interviews went over an hour in length, with 40-45 minutes being the average.

In all of the homes I visited, the interview had to be fitted around day-to-day life for the participants, such as school, college, physiotherapy, sports activities and in one case a visit by an occupational health worker. Being in the participants' homes was informative; highlighting how much equipment is required and how their homes must be adapted to provide smooth flooring, accessible doorways, hoists, and modified bathrooms. For the two homes where there are two boys in the family with DMD, much space had to be devoted to their accessible bedrooms, and garden space had been encroached on to extend the houses and to accommodate large, adapted vehicles. Despite being busy, each household warmly welcomed me and, in the time before and after the interview, the parents gave varying details of how they coped. They spoke of their frustrations with poor health and social services, and the elusive nature of a cure or effective treatment for their growing child. As a single parent of a similar age to the parents, I was able to grasp some very small element of their struggles and fears, whilst feeling deep respect for their abilities to cope both at a physical and emotional level. I was always aware that my presence came at some cost to each home where they had, obviously, consented to the interview, but where they had to incorporate it into daily life.

Whilst I was interviewing a young man, his mother called me through to the lounge as there was a report on the television news about some promising findings from medical research on DMD. She commented to me, as we watched the news item that she did not keep up-to-date on news about research as it was too late for her two sons who had both gone into wheelchairs early. This incident was painful and insightful for me; I recognised her own resolve in managing her life and avoiding exposure to information that was irrelevant to her boys. This concurs with Condin's (2002) findings on parents of children with DMD for whom 'research news is perceived as capable of causing harm if exposure to it is not carefully controlled' (2002, p28). This was a finding in itself, showing how the parents must regulate their levels of hope and anxiety, and how their own practices around the flow and management of information on DMD/MD may significantly infuse their child's knowledge base. In some homes it is apparent that parents work to accrue information on DMD/MD as their way of coping, whilst for others, as with this example, information has the potential to hurt and cause despondency, therefore parents may manage the information they give their child (Young et al, 2003). Although I am not writing about the parents' experiences, they are, as will be seen in later chapters, key elements in their child's life. They were my first contact in each research encounter and they shared something of their daily experiences with me, and I could not help but see how their attitudes are likely to be connected with their child's. As I visited the previously mentioned home twice, and also met this mother a third time at a wheelchair football session, I saw how hard she and her husband worked in fitting their own lives around the needs of their sons. Without making direct connections between this family and the others, it seemed apparent to me that all the families balanced the many demands on their time, and worked very hard to keep their homes running as smoothly as possible. These unexpected observations give my data analysis added depth, and my findings are enhanced, and influenced, by the emotional, sensory as well as intellectual nature of some of my observations. These observations are then mediated by rethinking and interpreting them as data that informs and shapes my research:

Every [research] encounter is interpreted in terms of acquired understandings, shaped by previous experiences and the cultural system within which such experiences are lived. An interpretive approach seeks to understand human

experience through thick contextual description, complimented with comparative analyses that elucidate significant patterns and similarities across cases. (Carnevale 2006, p50)

The validity of interviewing the participants in their own homes where they were comfortable and safe meant that I was able to better appreciate life with a disability and the 'mundane and often gruelling schedule of families' (Stevens et al. 2010, p502). However, as noted, this placed me in a position as both guest and social researcher, with the sociocultural balancing of appropriate behaviour and sensitivity required in entering each home with attentiveness to the family and participant's style of communicating. Even the most well prepared researcher lacks complete 'expert status', as research can often be a case of 'muddling through' or feeling 'lost and out of place' (Gallacher & Gallagher 2008, p512). This was certainly sometimes the case for me, I did not feel powerful, nor an expert and each interview encounter was a challenge that drew on my inner resources as I strove to respond appropriately.

Due to the geographical spread of participants I needed to plan for quite extensive travel, with the inevitable frustrations of using public transport and locating the venues, but it was worth the struggle as visiting the participants' homes enriched my research. As will be seen, the first ethics committee turned me down over concerns for my safety as a lone worker travelling large distances, but visiting the participants in their homes was an invaluable element in developing my findings.

### Follow-up interviews

I interviewed 4 participants a second time, selecting those who had responded well in their first interview, who had raised some interesting points and who I thought would be amenable to a follow-up interview. 2 were face-to-face as the participants lived in Northern England and 2 were by telephone as they lived in Southern England, I had met these latter 2 face-to-face for their first interviews. It was intended that the second interviews would be shorter and that I would ask the participants to clarify or extend their thoughts on some topics we had covered or

where their comments had provoked my own thoughts after I read back the first transcripts. For each participant, I typed out the relevant quotes from their first interview and then posed the new questions these raised for me underneath, I then posted these individualised prompt sheets to each participant's home to give them an opportunity to read them through and think of their responses (see Appendix E for example).

I had been apprehensive about the telephone interviews, I thought the participants might find this style of remote interviewing strange; but in fact they went very well. As with the second face-to-face interviews, I confirmed that the participant's previous consent was still valid and I checked that they were happy to have the interview recorded. Both participants had obviously run through the questions prior to the interview and gave confident responses without appearing stilted or awkward.

Of the 2 face-to-face interviews, one participant looked unwell or was perhaps just tired, and his responses were limited. It did not help that my train made me an hour late, although the family were happy to have the interview go ahead and we agreed a time to conclude it, before I conducted the interview. At the other interview, there had been some recent issues at school, with an upset between the participant and a school colleague. The participant was a little preoccupied by this during the opening minutes of discussion, although as we talked he became more settled and relaxed.

The follow-up interviews were successful overall, giving me an opportunity to check and expand on important points. The process of typing out their previous quotes and adding my new questions underneath and then posting/emailing these ahead of time proved a very useful strategy, suggested by my supervisors, which worked well. I will indicate in the data chapters when the participants' comments are from second interviews, and I will also indicate if the interview was by telephone. Once all the interviews had been conducted, I entered the next phase of my research, which was analysing the data and this will be discussed in the next section.

## Analysing data

Woodgate (2001) cautions that the power to interpret data lies in the researcher's hands, and this suggests that no research can be entirely symmetrical as the researcher's own perspectives influence the inclusion and interpretation of data. At the initial stage of analysis, Richards (2009) suggests a close re-reading of transcripts, working up from individual documents and data to gradually identifying themes and concepts that spread across the data records. The researcher is looking for patterns in the data, bringing the accounts together and identifying different individual's responses to issues and conflicts. As I began to critically re-read my transcripts and analyse them, I sought the advice of various colleagues who have undertaken qualitative research, each had taken a slightly different course, but I observed that they had worked in an ordered and well-managed way. I took some of their advice and implemented my own practical mechanisms for storing and managing the coded and categorised data in both physical and electronic form. It was important that I developed clarity in my working practice as I was not opting to use a software package for my data analysis; this was a viable decision due to my small sample size and may have needed rethinking had there been a larger sample.

Saldana (2009) explains the procedure for effective data analysis, moving from reading the transcripts to beginning the task of coding them, coding takes place through the identification of words or phrases that convey significant or useable meanings. These meanings are then categorised under headings, indexing the material into groups which may alter over time, with codes merging, emerging or being revised as understanding develops. Categories are then consigned to the main themes that will form the structure and content of the data chapters. Crabtree and Miller (1992) describe using an editing style, which is similar to the approach I adopted; this involves highlighting words and phrases during a systematic reading of each transcript. These are then organised into codes and categories that are reread for further interpretations which may alter the coding. The interpreter then explores the categories and determines the patterns and themes that connect them; in this approach, data analysis is an iterative process that begins shortly after the first data is collected, with analysis creating new understandings that may bring changes until

understanding is complete enough to stop this process (Crabtree & Miller 1992).

My analysis began as I wrote my fieldnotes and returned to my desk to transcribe my interviews, some words and phrases stood out immediately during transcription, leading me to note down my initial thoughts in the margin of the transcript. Phrases and statements used by the participants such as 'I just want to get on with my life', raised my awareness of the ways in which they manage elements of their lives. These words and phrases provided initial analytical codes and categories to work with, expanding upon or adjusting them as the data was studied (Pope 2000). Working with my supervisors' help I developed these codes and the categories they suggested, cutting and pasting relevant sentences identified by page and transcript number into separate documents broadly encompassing the themes emerging from the data. The initial focus of the research proposal was significant in forming my interview schedule, which then guided my identification and development of codes, categories and themes. The analytical challenge has been to identify categories and themes within and across the interviews, bringing these segments together as a step to opening up meaning, even as codes have been adapted (Silverman 2005). The process just described took time to develop, but the words the participants spoke and their trust in sharing those with me were instrumental in shaping my findings. As Christensen (2004) has documented:

Working together with children in research requires attention to the trust and loyalty that accompanies it. In my attempt to unravel particular parts of my fieldwork, my engagement with the children would provoke new lines of thinking and demonstrate in a quite practical way how data production and analysis are inextricably linked together. (2004, p172)

As an example of how I worked, under the general category of 'Doctors and hospitals', the initial sub-categories that emerged were: 'Sleep'; 'the hospice'; 'physiotherapy'; 'being at the hospital'; 'talking to doctors'. Some of these categories proved to have too few quotes to make them relevant as distinct entities and this led to the condensing of categories. I frequently drew spidergrams in my notebook and these worked very well for me as a strong visual and creative guide to how I saw these categories and themes connecting up. The sketched out spidergrams



condensed into a PowerPoint document that helped me and my supervisors to clearly see the categories and themes I had developed (see Appendix F for PowerPoint themes). Spidergrams provided an opportunity to work intuitively and helped me to recognise content that strayed beyond the boundaries of my research as I gradually worked the categories into my main themes (Basit 2003). I repeatedly returned to the transcripts and re-read them during the analysis phase, checking that my themes were logical and workable, and importantly, that they remained faithful to the participants' accounts. Thus, whilst I have interpreted their contributions using my own perspective which is shaped by the research agenda, I have attempted to centre my interpretations on the participants' intended meanings as I understood them.

Silverman (2005) considers how ambiguity in analysing qualitative data can be viewed as a resource rather than a problem; in my research this ambiguity, where the participants' answers were occasionally limited, hesitant or blunt, could be due to a range of unseen and unvoiced possibilities, some of which have already been mentioned. For some participants, 'living in the moment' may have precluded them from previous speculation on medical research. Due to the limited promise of medical research that can directly help them, they may have adapted to a life with little or no reference to such activities. Bearing these factors in mind, interpreting the data evolves from engaging with the transcripts and field notes towards a tentative analysis that is mindful of varying interpretive possibilities. It is tempting to keep a coherent thread within the analysis; however, the process should also remain open to deviant cases (Silverman 2005). My own sample has not provided examples of hugely varying opinions, but there have been differing accounts of similar themes both within participant's accounts and also across the accounts given by the group. For example, one participant stated that boys should make their own decisions on taking part in medical research, and then in another section he says that the decision is for the boy and his parents to make together. In another example, two participants in the research use the same hospital for their care but they differed in what they thought of the doctors and services provided there. The presence of variance does not render the findings and the analysis invalid, but it points to the need for a critical awareness from me as the researcher to engage with the analysis at an intellectual,

interpretive, emotional and analytical level.

I was aware during the interviews that what people say in an interview can be dependent upon what is currently happening in the participant's life. This was noticeably so with the two follow-up interviews which were conducted face-to-face. As mentioned, one participant had been upset about a situation at school and the other participant was tired, or unwell, and not as talkative as previously. Their responses on the second occasion were somewhat subdued, as was the overall mood, but this does not mean that the resultant data is worth less, it points to the way in which we all respond to the daily occurrences of life, and the data analysis should reflect this rather than hide or smooth over it.

The initial research proposal and the resulting interview schedule provided a boundary for what was to be discussed without being too rigid, and there was room to cover any important points the participants raised. None of my observations have been wasted and I found the entire process absorbing, compelling and a privilege. Several people have commented that I must have found the interviews depressing, an attitude that reflects the generally held view that disabled peoples' lives are tragic and that they are victims (McLaughlin & Goodley 2008). I did not find any part of the experience of interviewing them depressing, because, just as with any life, there are difficult times for the participants but there are also good times and, further to this I am deeply interested in lives that are lived differently. Alongside my primary research focus I have seen how disability, marginalisation and the altered boundaries of hope and 'normality' can develop alternative ways of living and coping (Carnevale et al., 2006; Condin 2002; Rehm & Bradley 2005). These alternative ways of living were not easily achieved, but denial was not possible for the participants and their families (Rehm & Bradley 2005) who have had to deal with and adapt to the tough realities they face.

### **Ethical approval**

Gaining permission to undertake research with children and young people who are ill or disabled can be problematic and restrictive (Carter 2009; Heath et al. 2007).

High-profile breaches of informed consent at Alder Hey Hospital and Bristol Children's Hospital have intensified debates on the increasingly legal orientation of how social research is conducted (Heath et al. 2007), because of concerns with consent, confidentiality and protection (Alderson 2007; Stalker et al. 2004). Responding to these concerns needs a balance 'between measures that ensure high standards are in place and measures that may impose unnecessary restrictions on potentially worthwhile research' (Stalker et al. 2004, p380). The establishment of national policies and codes has generated a discursive system requiring guidelines, practices and 'systematic modes of surveillance to govern the enterprise of ethical research' (Halse & Honey 2007, p339). Researchers can face challenges in securing permission to conduct some research, and this can be notably so when the research is on or with children (Carter 2009). It is observed that 'within the context of research governance children are categorized as a vulnerable population requiring special consideration' (Carter 2009, p859). As a major part of my recruitment was through an NHS Clinic, approval from a Research Ethics Committee (REC) was required, along with obtaining enhanced Criminal Records Bureau clearance and the securing of an NHS Honorary Research Contract. I was aware that as I would be recruiting from a population considered to be 'vulnerable' it was important to stress to the REC my commitment to work sensitively and ethically at all times. Therefore I took great care as I prepared my documentation and recruitment materials in readiness to gain ethical approval.

Despite careful preparation, my first application for approval was turned down by the REC, and surprisingly, given the issues noted above regarding the recruitment of children; this refusal was not to do with concerns for the participants' welfare, but my own. The REC were unhappy with my lone worker status; I had included in my protocol a document on safe conduct for lone workers (see Appendix G for Lone Worker protocol), but the REC had concerns for my safety as I would need to travel extensively. They felt it was 'a bit much' to be travelling the length of the country to conduct interviews and they did not like the idea of my going into strangers' homes. The implication was that my safety would be at risk, and they suggested, somewhat impractically, a central venue would be the best place to conduct interviews. I do appreciate their concerns for me, but my sample population are broadly dispersed

and it would have made it difficult to recruit if I stipulated centralised venues for interviews. However, a favourable REC opinion was granted on appeal and I was able to conduct my study.

As stated above, I followed approved protocol for lone workers, observing measures to ensure my personal safety as I was travelling large distances and visiting the homes of people I had not met before. I took the precaution of conducting all interviews in the day time, and this fitted with the participants' routines. I ensured that my supervisors and a friend or family member knew I was conducting an interview and texted or phoned when I had left the interview to inform them I was heading home. Advanced planning of my transport links also minimised any safety issues and arriving on foot or by taxi gave me a sense of the area each participant lived in.

## **Sampling and recruitment**

The original plan was to recruit between 20-25 boys and young men aged 12-18 and who have DMD, this age range was chosen as, whilst research has been conducted with parents of children with DMD, little research has involved the young people themselves. This age range also recognises that the boys will have a growing appreciation of their condition and may be aware of medical research into DMD or may have taken part in some. As the condition almost exclusively affects boys and men, the recruitment was by necessity aimed at boys and young men. The geographical recruitment area was to start with the North-East/West of England and Southern Scotland, widening this area if needed. The recruitment material was adapted slightly for the recruitment routes used, which were: an NHS Clinic; a charity called Action Duchenne, and a wheelchair football team (see Appendix H for recruitment materials) and latterly, the Muscular Dystrophy Campaign [MDC] (see Appendix J for MDC recruitment materials). The recruitment material was designed in accordance with advice from my supervisors, colleagues, the National Research Ethics Service and the British Dyslexia Association's website, and formatted to be as simple and legible as possible. Over time, and as recruitment proved problematic, the inclusion criteria was enlarged to include individuals with any muscular dystrophy

and with an age range extending from 12-21. It was decided that including individuals up to the age of 21 still kept the focus on young people and that opening the sample to other forms of muscular dystrophy kept the research emphasis on neuromuscular disease. This suited the range of literature and information I had drawn from in my preparations. Opening the sample to other muscular dystrophies also raised the possibility of girls and young women being recruited. The recruitment problems I faced, and which precipitated extending my sample range are further explained in the 'NHS Clinic' section.

In DMD there can be delays with language development, with some boys having autistic-spectrum disorders (Action Duchenne 2010). The NHS Clinic vetted their own contact list, ruling out mailing information to families where the son had communication problems or where it was thought the family would be burdened by extra demands. It is noted that children with communication impairments and complex needs can be further marginalised from inclusion in research (Noyes 2000, p1207), but for my project it was not viable that the extra support needs required could be met. This limited the sample number for my project, although one participant did have Asperger's syndrome and he coped very well, and his inclusion suggests that restricting who should take part may miss some valid contributions. However, I trusted the NHS Clinic's guidance on who they sent recruitment materials to, and as the other recruitment routes sent out or advertised recruitment information to all potential participants in the age range regardless of communication impairments, it was up to the families and children to decide if they contacted me or not. It also depended on my assessment of a participant's suitability to take part, which was confirmed after talking with their parent and meeting the child or young person. As recruitment was complex, with take-up very slow, I concluded recruitment when I had 9 boys with DMD and 1 young woman with muscular dystrophy. In order to protect the young woman's anonymity, I have not disclosed her diagnosis, which is rare and less severe than DMD, but which is variable in long-term impact, meaning a wheelchair may be needed later in life. As I began to analyse my data, it became apparent to me that the inclusion of this young woman with a different, but related diagnosis to the boys was salient; she provided insight, and a sensitive reading of her own condition and how she imagined life to be from

the boys' perspectives. Her inclusion and opinions acted as a contrast and comparator to the boys' interviews, and helped me to think creatively about my data.

### *NHS Clinic*

Initially, recruitment was to be facilitated through nurses at an NHS Clinic handing out leaflets about my project to boys and their parents when they visited the clinic (see Appendix K for original flow chart). It was pointed out by my gatekeeper, a specialist nurse at the clinic, that it would take a long time to recruit enough participants as the boys only visit the clinic once every six months. As a result of this observation I applied to a REC for permission to make substantial amendments to my recruitment process and extended the recruitment age-range from 12-18 to 12-21. The amended recruitment process entailed the NHS Clinic staff mailing-out my recruitment packs to those on their database who fitted the age criteria and who the staff considered would be able to communicate effectively. This route fulfilled confidentiality needs as I would have no sight of contact details and parents would make first contact with me if interested. This amendment was accepted by the REC and so I prepared and left the recruitment material in envelopes at the Clinic for them to address and mail-out. Throughout this time, my gatekeeper at the Clinic, who had been recommended to me by the clinic's consultant, became very hard to reach; she did not turn up for meetings with me or respond to emails and was off work for some time with ill health. This made it very difficult for me to keep track of the recruitment process which, for reasons of confidentiality, had to be through a gatekeeper. I found someone else to help me but eight weeks after dropping the recruitment envelopes off at the clinic for her to address and post, I went back to find this had not been done. I ensured that this was dealt with and the first mail-out of 24 envelopes was completed. As response was minimal, a second mail-out went to a wider area; this took several more weeks to set-up. Throughout this recruitment phase, during which time I lost all contact with my gatekeeper, the clinic staff were hard to track-down or engage with as they had so many duties without the additional work of helping me.

Out of approximately 40 packs sent out in total, only 2 participants were recruited through the Clinic. Despite the difficulties in finding and keeping a gatekeeper it was

informative to attend Clinic team meetings and to speak with the physiotherapists, specialist nurses and advisors. In the course of time I met with a member of staff from the clinic to talk over my first thoughts from my interviews, she gave a much appreciated and unexpected apology on behalf of the clinic for the difficulties I had encountered and I really appreciated this and her observations on my initial findings.

### *Action Duchenne*

Action Duchenne, a charity set-up to promote fund-raising for more research into DMD was my second recruitment route; this had been explained to the REC as being a recruitment option that would be used if the Muscle Clinic route was insufficient, as was the case. This second route went smoothly and I had excellent communications with the database curator (see Appendix L for flow chart). I sent an email to the curator who contacted me rapidly to say they would be pleased to help, suggesting a mail-out would be the best way to recruit. I sent a research proposal to the board of trustees and some sample recruitment materials, to check they were happy with the wording on the information sheets. Then I sent them 51 recruitment packs in accordance with their estimation of how many individuals they had on their database that fitted the criteria and they then addressed and mailed the packs out for me. This mail-out was to North-East/West England, Southern Scotland, Yorkshire and Humberside. The charity also placed an item about my project on their website, in their newsletter and sent out emails to their members detailing my recruitment needs. I attended their annual conference and set up a stall to raise the project's profile, I met many interesting and informative people and attended various seminars but, despite my efforts, only 2 participants were directly recruited through Action Duchenne, although as will be seen, a snowballing effect was instigated through this route.

### *Wheelchair Football*

A father who had responded to my recruitment pack sent through Action Duchenne contacted me by telephone to say his son was interested in being interviewed and the father added that he ran a wheelchair football team which several boys with DMD used. I had already seen this football team online and had wondered about making contact with them as they looked well organised and had a strong online presence.

The father felt sure that I could recruit more through this route so when I went to interview his son I took some recruitment packs for the father to hand out at the next wheelchair football meeting. I also attended one of the wheelchair football games to watch the boys in action and to meet them and their parents and to hand out recruitment leaflets. Attending the game was absorbing, I was a similar age to the mothers who were there watching their sons and it was easy to chat and to hear about their lives. One mother in particular, took me 'under her wing' and seemed to trust me enough to describe her experiences as the mother of a son with DMD. The other mothers were equally open with me, and managed to chat whilst also doing 'running repairs' on their son's adapted wheelchairs, which took quite a few knocks during the football session. Their comments, which were not instigated by me, but offered during free-ranging conversation, covered the issues of grief, one mother's non-disclosure to her son of his full diagnosis, poor funding, resource and equipment needs and fears for the future. I eventually recruited 3 boys through wheelchair football; 2 were brothers, one of whom at 10 years old was 2 years below my age range. However, with recruitment being so slow it was decided that as he was keen to be interviewed and as he had not been recruited through the NHS clinic, which only had permission for the 12-21 age range, I would interview him. I sought assent from this boy, and his mother gave her consent; as this participant was younger than my original age range, seeking his assent and his mother's consent ensured an acceptable level of permission was reached.

The comments of one father at wheelchair football were somewhat telling, as I handed him a recruitment leaflet he said, not unkindly, that he was only interested in things that would directly help his son, who had a different but deteriorating form of MD. I took this to mean that he would not be interested in an interview for his son. Several contacts from charities and some parents I met all commented that parents are key gatekeepers in their child's life and various issues were cited as potentially impeding recruitment. These included parents' lives being too busy to accommodate me interviewing their child, or parents being fearful that I could upset their child, or concern their child may find out more about their condition than they already know. Abbott and Carpenter (2009) conducted interviews with young men with DMD, at which parents and other family members were sometimes also present, occasionally



it was apparent that 'brand new information was coming to light within a family, for example, what a young person did or did not know about DMD and its prognosis' (Abbott & Carpenter 2009, p20). This did not happen in any of my interviews, but anecdotal evidence and the comments of a mother at the football session suggest that in some homes, diagnosis and prognosis are not shared with the child. It is also likely that many families, understandably, just wish to get on with their lives and keep DMD in its place. I gained so much from being at the football session, and the information I gathered happened spontaneously as I chatted with the mums. These unplanned occurrences made me feel that I had an interactive role in a recruitment process that had, at times, been frustratingly remote and bureaucratic.

### *Muscular Dystrophy Campaign [MDC]*

To try and bring in more participants, I extended my recruitment to the MDC, and in so doing broadened my recruitment to any form of muscular dystrophy. The MDC placed my recruitment information on their website in several different formats to reach as broad an audience as possible, including putting an advert on their Trailblazers site which is for young people with muscular dystrophy (see Appendix J for recruitment materials). I attended their annual conference where I set up a recruitment stall and the only female participant was directly recruited through this method. The MDC website brought in my last 2 participants, and both have DMD; a mother saw my article online and contacted me to say her son would be very interested to be interviewed and she helpfully set up an additional interview with her son's friend who lived nearby. These 3 interviews through the MDC all proved vital to my project at a time when recruitment was slow; they were key factors that enriched my data. The age range of the participants from each recruitment route is represented in the next table.

<u>Muscle Clinic</u>	<u>Action Duchenne</u>	<u>Wheelchair Football</u>	<u>Muscular Dystrophy Campaign</u>
18 years old	14 years old	14 years old	21 years old
18 years old	17 years old	10 years old	16 years old
		16 years old	12 years old

Table Two: age range in recruitment strands

At this point it was decided to conclude the recruitment phase, it took 9 months from recruiting and interviewing the first participant to conducting the final interview, and recruitment was complicated and slow. This was a similar experience to those I had been in contact with who had previously conducted interviews with DMD participants. The reasons for this are varied, including, but not limited to: parents being busy, parents wanting to protect their son from prying questions, and parents and/or their child may see social research as irrelevant compared to the need for a cure for DMD. Despite the low numbers recruited, each interview enriched my findings; there were commonalities amongst the participants, bringing a consensus to some elements of the data, but there was also much that was personal to each participant. Therefore, whilst the sample was small the data was rich; however, the lower than expected recruitment levels meant that additional data was required to strengthen and corroborate the research. Hence I discussed with my supervisors the possibility of exploring policies and regulations relating to children's participation in their healthcare, and to do so through conducting a discourse analysis. This would inform the research, through examining the discursive tropes policy documents adopt when discussing children and their healthcare. The discourse analysis is the subject matter of Chapter Five and the methodology for the analysis is explained in the final section of this chapter.

### Ethical issues

Ethical issues have been touched on at various points in this chapter, but here I will note some specific issues that emerged. Eventually, after all the ethical

permissions are in place, the research interview is an interaction between researcher and participant, and this relies on the researcher's skills and a strong code of ethics to guide them in collecting data, while upholding the rights of the participant (Corbin & Morse 2003). As this research has faced quite stringent checks on its ethical approach I have been highly aware of the need for sensitivity and a robust, ethically sound sensibility in preparing for and conducting the interviews. All names were changed for anonymity, and contact details and other identifying information were stored on a password protected university computer to which only I have access. Full consent, or assent in the case of the participant aged 10 was secured before each interview, and age appropriate information sheets were sent in advance of the interview to help participants and their parents understand the nature of the interview and my project.

In the 2 situations where the participants became restless, it may have been wise to conclude the interview sooner and, as observed earlier, giving the participant a red card or other device would allow them to pause or end the interview. When I sent out the verbatim transcripts to each house, in one instance I felt a certain discomfort as the participant had mentioned a personal matter, nothing to do with harm, about a parent's physical health condition, which I did not use in my data chapters. In the transcript's margin, next to the sensitive comments the participant had made, I wrote that I would not be using that data, in order to reassure the parent when they read it. After I had sent off the transcript to the family, the mother texted to thank me for sending it and she added that it was a great way for her to better understand her son, and she was very proud of him. This helped me to feel much more comfortable with the situation; I also greatly appreciated the feedback. This incident does reveal the issue of confidentiality as, invariably, parents will have opened and looked at the transcripts, even if they were addressed to their son. This highlights how disability impacts agency and autonomy and it also indicates the inevitable closeness within these families. Independence is mingled with interdependence and the parents act as facilitators in their son's life (Samson et al. 2009) as well as primary caregiver. For my research, none of the subject matter covered sensitive issues or raised the potential for uncomfortable disclosures, so if parents read the transcript I did not regard this as a problem. Indeed, many parents,

whilst being out of the room during interviews, were within hearing range; it does, however, point to the fact that it is hard to send out items that are for the boys without drawing their parents into that process as facilitators.

As I only had one female participant and as her diagnosis is rare I have been careful to protect her anonymity, it would have been interesting to say a little more about her type of muscular dystrophy but this may have identified her to some people. My final ethical concern was that I was relying on parents to help me set up the interviews and that I was offering nothing in direct return to the parents, who had been instrumental in helping me. I was also taking time away from each participant's day, and drawing on their precious energy reserves. It was hard to leave one mum who was isolated and very conscious that her neighbours may see her as 'living on benefits' because she has 2 sons with DMD, she gave me a long hug as I said goodbye to her and I wished I could have stayed longer. As each family had made first contact with me I trusted that they understood enough from my information sheets to comprehend the transactional nature of the situation. I also hoped that the participants and their families appreciated an opportunity to share their thoughts and experiences and that they enjoyed being involved.

The varying problems I faced, including a 'hierarchy of gatekeeping' (Hood et al. 1996) have demonstrated the ways in which children and young people can be difficult to reach and hear from. The difficulties are not just due to overprotection, but also to bureaucracy in gaining NHS ethics approval, the demands on clinical staff and parents' time and, understandably, the status of sociological research in comparison to finding effective treatments for DMD. When one considers these matters, it helps to contextualise my small sample size, but it also makes what the participants have each contributed to my research a meaningful step in the privileging of their voices and experiences.

### Limitations

The interviews only covered the accounts of 10 disabled children and young people, and a bigger sample may have revealed more divergent experiences.

Recruitment was slow as varying attempts were made to bring in more participants, although the extra time spent on recruitment gave me the opportunity to read extensively and to gather varying opinions from parents and personnel involved in DMD/MD organisations on why recruitment was so problematic. These comments included the observation that those with DMD may not want to dwell upon their condition. Or they may be more interested in the things that other adolescents are interested in, like a love life and cars or university, rather than focusing on their healthcare needs. Any or all of these factors could have been key points that impacted recruitment; and I certainly found that parents are key gatekeepers, who are instrumental in enabling access to recruiting participants.

As mentioned earlier, some parents may not share full details of diagnosis and prognosis with their disabled child, and these families are unlikely to get involved in social research. There will also be parents who are not coping well and who have no desire for their lives to be observed or recorded by others, and my research is in some ways unrepresentative, as it cannot easily reach those families and children. Indeed, in a social encounter, I mentioned what I was researching to someone, and he recounted that he and his wife had once fostered a teenage child with DMD whose family had not wanted to care for him. My research covers the worlds of children and young people in homes where life is quite well-ordered and DMD/MD is present, but is competently managed; and it is highly likely there are less successful situations where effective coping strategies have not developed.

My research methods may draw criticism because of the subjectivity of semi-structured qualitative research which focuses on individual accounts in some detail, rather than giving more generalised findings. It does, however, examine the insights of a small group of 'experts' on living with their condition, providing a platform for further research which may generate broader findings. I could have used other methods to work with the participants; for instance, visual methods may have worked, using short video clips or images relating to DMD/MD and decision-making. Or a storytelling technique might have worked well, although a narrative approach may have its own limitations when working, as I was, with participants who can be reserved or who tire easily. Finally, my own health was a limitation as I became

unwell during my studies and this developed into a chronic condition needing time for adjustment and adaptation. Eventually, this experience has helped to inform my knowledge in some small way, of the NHS and the role of doctors in a patient's life, showing the importance of good and effective personalised information and communication between doctors, nurses and their patients.

It was explained earlier that Chapter Five will use discourse analysis to explore policies and regulations relating to children's healthcare participation, and in the final section of this chapter I will discuss the methodology I used for the discourse analysis.

### **Discourse analysis methodology**

The purpose of the discourse analysis in Chapter Five will be to explore and question the range of attitudes expressed in official documents relating to children and young people's participation in their healthcare and medical research. I will examine the ways in which participation and consent are represented, and how constructions of children and young people, including those that are disabled, may be reiterated and reinforced within official accounts in an unchallenged and taken for granted way. High profile institutions like the Department of Health have a formative influence on attitudes and practices regarding children's capacities, and this influence can be seen in many of the Department's guidelines and policy statements. Therefore, the need to find documents pertinent to my analysis led to a thorough search and selection process that is described next.

#### Document search and selection

Sources for this discourse analysis have been identified from the reference lists of texts in closely related documents relating to healthcare, as well as searching the university's library catalogues and databases. To limit my search to a manageable size I conducted an online search for documents produced in the last 15 years, that is, from 1998-2013, in order to provide enough data whilst ensuring that the data is

representative of recent and current attitudes. Although it would be interesting to look further back, this approach has proved sufficient to situate current practices and attitudes around children's healthcare participation, consent issues and medical research. Therefore it resonates with my own empirical research as the participants are at an age where such issues and attitudes may impact on their healthcare and decision-making. Additionally, this 15 year period is post-Gillick competence guidelines, the details of Gillick competence were discussed in Chapter One, and current discourses to some extent reflect or at least reference a more subtle, if complex attitude to children and young people's competence. During the research process, close attention has been paid to following the threads of discourse and the referencing amongst and between documents. It was recognised that documents cite one another, demonstrating how discourse is produced, reproduced and reinforced. Thus, items which were referred to by the original documents were read if they related to current or recent policy practices and discussions. These additional documents included those produced by charities such as the National Children's Bureau [NCB] and the umbrella body the Council for Disabled Children [CDC]. The table below shows the databases that have been used in my discourse analysis:

<p><a href="http://www.legislation.gov.uk/search">http://www.legislation.gov.uk/search</a> as a generic site to start from;</p> <p><a href="http://www.official-documents.gov.uk/">http://www.official-documents.gov.uk/</a> which holds Command papers, House of Commons papers and key departmental papers that cover amendments and discussions surrounding Acts of Parliament,</p> <p><a href="http://www.ukop.co.uk">http://www.ukop.co.uk</a> the catalogue of UK official publications, and;</p> <p><a href="http://www.dh.gov.uk">http://www.dh.gov.uk</a> the Department of Health's site.</p> <p>Web of Knowledge, Ovid/Medline and PubMed.</p>
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Table Three: Databases used in discourse analysis

In addition to these databases, UK healthcare organisations' websites, including the British Medical Association and the General Medical Council have been accessed to understand the general tenor of discussions around children and young

people's participation, attention was also paid to the related sites and organisations that were mentioned. These healthcare organisations tend to refer back to documents from the Department of Health, thus a referential link between documents emerges, showing both the source and the ongoing, iterative nature of discursive forms.

Within each database a key word search was used to identify references I was interested in, relating to children and their participation in healthcare and medical research. The key words and terms searched for were:

- *Autonomy*
- *Participation*
- *Clinical trials*
- *Medical research*
- *Children*
- *Disabled/Disability*
- *Consent*
- *Gillick competence*
- *Adolescents/Adolescence*
- *Capacity*
- *Competence*
- *Young people/person*
- *Agency*

Only items within policy documents and closely related texts have been used, rather than using a broader repository such as newspapers and other forms of print and popular media. This strategy helped to keep the focus on documents relating to healthcare policy and practice. Influential journals that discuss and reflect current policy and practice amongst clinicians, healthcare professionals and social researchers were studied, these journals included: *BMJ*, *Paediatrics and Child Health*, *Pediatrics*, *Archives of Disease in Childhood*, *The Lancet*, *Childhood*, and *Social Science and Medicine*. These provided background knowledge and covered discussions on and constructions of children and young people relating to participation, competence and consent over the 15 year period. Attention was paid to the presence of any 'discursive events' involving a change in tone and positioning from the earlier to more current texts. The focus has been on how children and young people are constructed in the articles, how childhood is spoken of and situated, and what is not said around these issues but is based on assumptions. The documents used are listed in alphabetic order below:



- Aynsley-Green A., Barker M., Burr S., Macfarlane A., Morgan J., Silbert J., Turner T., Viner R., Waterston T., & Hall D. (2000) Who is speaking for children and adolescents and for their health at the policy level? *BMJ*. v321, pp229-232.
- BRI Secretariat (1999) *Bristol Royal Infirmary Inquiry Paper on Informed Consent: Concept, Guidelines and Practice with Reference to Children Undergoing Complex Heart Surgery*. Bristol: BRI Secretariat
- Bristol Royal Infirmary Inquiry (2001) *Report of the Public Inquiry into Children's Heart Surgery at the Royal Bristol Infirmary 1984-1995: Learning from Bristol*. London: TSO.
- Burke T. (2010) *Anyone Listening?* London: NCB.
- Department of Health (2002) *Listening, Hearing, Responding*. London: DH.
- Department of Health (2003) *Getting the Right Start: National Service Framework for Children: Standard for Hospital Services*. London: DH.
- Department of Health (2004c) *National Service Framework for Children, Young People and Maternity Services: Executive Summary*. London: DH.
- Department of Health (2004a) *National Service Framework for Children, Young People and Maternity Services: Core Standards*. London: DH.
- Department of Health (2006) *Research Governance Framework for Health and Social Care*. London: DH.
- Department of Health (2008) *Involving Children and Young People in Healthcare: A Planning Tool*. London: DH.
- Department of Health (2012) *Report of the Children and Young People's Health Outcomes Forum*. London: DH.
- European Compliance Academy [ECA] (2008) *Ethical Considerations for Clinical Trials on Medicinal Products Conducted with the Paediatric Population*. European Medicines Agency.
- Hull D. (2000) Guidelines for the ethical conduct of medical research involving children. *Archives of Disease in Childhood*. v82, pp177-182.
- Kennedy I. (2010) *Getting it Right for Children and Young People: Overcoming Cultural Barriers in the NHS so as to Meet their Needs*. Department of Health. London: DH.
- La Valle I., & Payne L. (2012) *Listening to Children's Views on Health Provision*. London: National Children's Bureau.
- Medicines for Human Use (Clinical trials) Regulations* (2004).
- Medical Research Council. (2004) *MRC Ethics Guide: Medical Research Involving Children*. London: MRC.
- Modi N., Clark H., Wolfe I., Costello A., & Budge H. (2012) Royal College of Paediatrics and Child Health. A Healthy Nation: Strengthening child health research in the UK. *The Lancet*.
- Royal College of Paediatrics and Child Health. (2003) *Bridging the Gaps: Health Care for Adolescents*. London: RCPCH
- Royal College of Paediatrics and Child Health. (2012) *Turning the Tide: Harnessing the Power of Child Health Research- Executive Summary*. London: RCPCH
- Royal College of Paediatrics and Child Health. (2012:a) *Involving Children and Young People in Health Services*. London: RCPCH

Table Four: Reference list of primary documents

## The approach to discourse analysis

To prepare for conducting the analysis, some general advice was gained from a discourse analysis workshop I attended at Manchester University in 2012. The workshop gave useful examples and practical tips on the starting point from which to conduct an analysis and a framework for questions to address including: *What role do the texts play? Who are they for? Who do they inform? Do new discourses impact the powerful institutional forces that hold the power base? Is there intertextuality in the discourse, are other voices brought in?* At the workshop, and in the literature I have used, it is observed that there is no one agreed method for conducting discourse analysis, nor is there a singular perspective (Hewitt 2009; Powers 2007). Analysis is more an approach and a philosophical mindset than an agreed upon or generalised set of practices, with the analysis being open to the interpretation and the conceptual understanding of the individual undertaking it. Lupton (1992) gives a useful definition of discourse:

Discourse... is defined as a patterned system of texts, messages, talk, dialogue or conversation which can both be identified in.... communications and located in social structures. (1992, p145)

Discourse analysis focuses on the reproduction of ideology, in which a particular view and presentation of issues, usually by those with the power and influence to be heard, forms a consistent set of linked ideas and logic. The emphasis is not so much on the message itself as is analysis of the elements and influences in the text, such as subtle forms of persuasion and manipulation (Lupton 1992). Discourse analysis responds to the texts being studied, engaging with them at a deeper level than an initial reading might reveal, and paying consideration to the 'constitutive role played by language in creating meaning and notions of reality' (Lupton 1999, p260). It examines the way in which patterns of knowledge and practice can be produced and reproduced through discourse, providing insight into how institutions function (Powers 2007). Pervasive discourses can frame the way that problems and their solutions are looked at (Shaw & Greenhalgh 2008) and the role of discourse analysis is to disrupt or challenge the 'truths' being produced, through looking for gaps and omissions in their official narratives (Hewitt 2009).

In relation to children, young people and their capacity, it is possible that a close reading of documents will identify elisions or taken for granted assumptions that are reiterated amongst a range of 'experts'. These documents are produced for professionals and they are about children, rather than for children, and they are somewhat dissociated from the subjects they discuss. The various documents will be analysed from a poststructuralist viewpoint, in which there is a rejection of any singular perspective, grand narrative or source of original logic or truth (Goodman & Ritzer 2003). Discourse analysis can be used to dissect and disrupt narratives, thereby de-familiarising what might be presented as familiar (Graham 2005). There is then, no singular, overarching truth, rather, there may be multiple discourses, each representing a version of reality according to the values of the institution from which they emerge.

There can be an absence of human agency in documents produced by bureaucratic organisations, which may not readily be challenged due to their tendency to report rather than engage in exposition (Fairclough 2003). Thus, the documents for analysis may appear to be straight-forward and unproblematic statements, in some cases about problems or issues existing 'out there' (Shaw & Greenhalgh 2008), offering solutions without an exploration of cause, context or recourse to dialogue. Stenson and Watt (1999) observe, in their analysis of local government texts, how the documents can entail experts addressing experts in an inner world; they also note that there are absences in who is included in the documents. This can efface complexity, whilst removing issues from their social setting, with bureaucracy reducing or omitting nuanced representations of social phenomena. Discourse can order reality in certain ways, determining who can speak, how and with what authority, representing a version of reality and playing a part in:

the very construction and maintenance of that reality itself. There is a dynamic relationship between the text and the context in which the text is produced. Texts are both constitutive of and, in turn, constructed by their context. (Cheek 2004, p1144)

Discourse is productive and circular in nature and it is not neutral, even though actorless, passive verbs may be used (Porter 2007), which tend towards removing

the agency of individuals and the accountability of the powerful by obscuring cause and effect.

Shaw (2010) describes using discourse analysis to scrutinise health related policy, and observes that a rationalist approach to policy formation persists in administrative and bureaucratic processes. Shaw contends that this rational approach entails the gathering of evidence which then informs policy decisions. However, more complex patterns of social problems and issues that are hard to articulate may be 'rendered invisible by the analytical processes in play' (Shaw 2010, p199). The analysis of discourse in policy documents contends that no one, including those who formulate policies, stands outside of the policy process (Shaw 2010). Policy formulations can identify and create their own issues (Stenson & Watt 1999), constructing objects of concern and agendas for action (Stenson & Watt, 1999) and impacting how they can be thought about and acted upon:

Policy documents are significant social mechanisms that can be analysed in their own right (rather than windows on the reality they claim to represent). They might be thought of as legitimating devices, with the official nature of a policy document having possible bearing on its staying power. (Shaw 2010, pp205-6.

Shaw asserts that policy is ideological and that using a poststructural approach which draws, amongst others, on the work of Foucault, can support considerations of how 'knowledge might be intertwined with mechanisms of power' (Shaw 2010, p203). Foucault's approach to discourse analysis does not follow or stipulate a step-by-step guide, although he indicates pointers towards how to conduct an analysis, and his approach will be examined next.

### Foucault and discourse analysis

Foucault has been influential in informing the process of discourse analysis, but it is 'difficult to find coherent descriptions of how one might go about discourse analysis using Foucault' (Graham 2005, p2). Hewitt (2009) suggests that to prescribe a methodology for discourse analysis would not be in the spirit of Foucault's work. To do so would imply a position of truth when no such claim can

legitimately be made, as truth is always conditional:

there are many methods employed and at the same time no methods... Truth is constructed within a discourse and, therefore, is relational to the knowledge and practices of that discourse. The relational nature of truth means that methodological choices made in any research project are driven by the problem at the centre of the research. (Hewitt 2009, p3)

Foucault's treatment of power is regarded by some as 'the most important notion... because it forms the basis for the analysis of discourse' (Powers 2007, p28).

Foucault regards power not as emanating from a particular source, or as being owned by specific groups, but existing through and in the production and execution of the discourses espoused by them (Foucault 1972). Knowledge and language are, from this perspective, embedded in the workings and outcomes of power:

Power... which is assumed to exist universally in a concentrated or diffused form, does not exist. Power exists only when it is put into action. (Foucault in Hewitt 2009, p7)

Foucault does not view power as a conflict between authorities and target groups (Powers 2007); rather he situates it as contextual, present in bureaucracy and institutions without being easily recognisable. For Foucault, power is not conspiratorial then, but exists as a historical phenomenon that has occurred without an overall strategy; it is present in discourse and can shape how and what is known and thought about (Foucault 1972). Drawing on the idea that discourse produced by institutions has power effects, van Dijk (2008) observes that official discourses can perpetuate a disparity of power in which one group can make claims about another group as they have the 'discursive and communicative scope and resources' (van Dijk 1993, p255). One of the key features of power and dominance is the privileged access to discourse and communicative utterances that this positioning facilitates (van Dijk 1993). This suggests that, in the context of disability and illness, health professionals and policy makers regularly speak from a platform unavailable to patient groups, including those who are young and disabled.

Foucault (1972) is concerned with understanding how the history of a discourse relates to the present, exploring the domain of knowledge, its selectivity and

absences. He proposes that statements have a different function from the linguistic utility of a sentence. The statement can construct and reify that of which it speaks, operating in a communicative void and rising 'above the flux of everyday oral discourse' (Stenson & Watt 1999, p192). As Foucault puts it, 'in analysing discourses themselves, one sees the loosening of the embrace of words and things' (Foucault 1972, p49), a discourse can uncouple statements from the things they impact upon. Graham (2005) indicates how statements can speak subjects into being; 'thus borrowing from Foucault, I interpret the statement as an articulation that functions with constitutive effects' (Graham 2005, p8). For Graham's own research on Attention Deficit Hyperactivity Disorder [ADHD], this means that children may be diagnosed with ADHD if they fit specific criteria of behavioural traits produced by a 'pedagogical discourse' (Graham 2005, p11). This can make statements and the discourse from which they emerge powerful and persuasive forces, representing a body of anonymous rules that impact upon individuals.

Foucault and others have demonstrated elements of their methodology in the descriptions they provide of their philosophical approach to analysis, and these accounts guide the mindset that I am applying to my analysis. However, Porter (2006) cautions that discourse analysis is a tool mainly available to and practised by a privileged, academic group that can exclude the less vocal from taking issue with its assertions. Whilst this is a possibility, awareness by the researcher of the potential power inequalities in research may help to reduce, if not remove disparities. Bearing these points in mind, the application of discourse analysis remains appropriate as a means of questioning powerful and widespread statements and practices in the field of young patients' healthcare.

The above section has discussed the methodological approach for conducting discourse analysis, and the analysis itself constitutes Chapter Five; in it I will draw, amongst others, on the work quoted above to inform my interpretation of the discursive construction of children and young people in healthcare.

## Conclusion

This research project has been challenging and innovative in seeking out the thoughts and opinions of children and young people with a degenerative condition, and the accompanying methodological and ethical issues have been prepared for and dealt with. Recruitment and NHS ethics approval demanded persistence and commitment in overcoming the barriers to hearing from potential participants. Once research was underway, concern for each participant's well-being during the interviews was minimised by staying within the bounds of my research questions and working sensitively. The semi-structured interviews proved an appropriate technique, revealing some interesting perspectives that could not have been covered effectively through proxy accounts or through quantitative methods. There were moments when the presence of parents called for a balancing of my role as a social researcher, with their role as a protective parent, and in those instances, common courtesy and respect were prioritised before research needs.

The input from the participants has been greatly valued and I have endeavoured to be faithful to their accounts as I analysed and integrated their contributions into my data chapters. The interview phase was, at times, a difficult but vital point to reach, and this shaped my approach to each research encounter. I felt a sense of privilege and a duty to work ethically and I valued the participant and parental trust in me. There were experiences in the fieldwork phase that I could not fully prepare for, such as unexpected comments and highly personal lines of reasoning, but these have contributed to the specifics of my findings and to the bigger picture within which my research sits. I am aware that the research continues on into the future as the findings are disseminated and taken up or reinterpreted. In some ways I cannot control that process but I trust that the spirit in which the research has been conducted and written remains, and that the core purpose, to enrich understandings of the lives and healthcare experiences of disabled children and young people continues. This chapter has explained my methodology and it has also touched upon aspects of the participants' lives. In the next chapter I will begin to explore in more depth the participants' lives, how they cope with the daily challenges their condition brings and the role of their parents and doctors.

## **Chapter Three**

### **Living with DMD/MD**

This chapter will help to contextualise Chapter Four, which will discuss how the participants might approach making a decision to take part in medical research. Chapter Four is linked to this scene-setting account, representing how decision-making does not occur in a void, but emerges from familial practices and the realities of living with DMD/MD. These practices encompass the values, hopes, expectations and limitations the participants live with, both as individuals and also as social beings within the setting of their family and sociocultural community. I will explore how making decisions and thinking about medical research evolves, in part, from each family's way of managing a condition that requires constant care and resilience. This chapter will examine a range of experiences relating to living with DMD/MD, before moving on in Chapter Four to discussing the central issues of decision-making and medical research.

It is apparent that having DMD/MD inevitably has an impact on the participants' way of living. For most, this impact is incorporated into their life as part of coping and, as several participants expressed it, of 'just getting on with it'. Whilst parents may experience their child's condition differently, for the young people there was a sense that their disability was normal for them, despite their awareness that their 'normal' was different to that of their able-bodied peers. Williams et al. (2009), in their study of children and young people living with Cystic Fibrosis suggest that achieving normalcy was a joint family endeavour whereby the regularity of treatments and care reinforced a pattern of coping. This would point to families having their own internal logic and parameters of just what their 'normal' is; enabling them to cope despite a challenging set of circumstances, as will be discussed in the next section.

#### Coping with DMD/MD

Most of the participants reported coping reasonably well with their condition on a daily basis, however, the difficulties and inconveniences encountered could be



troublesome. Jay (14) had some health issues that resulted in him missing his favourite sports activities:

*Jay: I haven't been for nearly 6 months.*

Whilst Ollie's (14) family holiday had logistical problems due to the snow:

*Ollie: We went up to Scotland, it was snowing everywhere, what they had to do they had to lift me out of my wheelchair and carry me all the way to the sofa and get the chair in.*

And the snow also interfered with his school life:

*Ollie: Last year, November, I had chilblains on my legs... so I wasn't allowed out [of school at break-time]... if it's snowing I might stay out if I can get wrapped up in a coat and a scarf and gloves.*

Despite problems, the participants obviously valued the activities they could take part in and which give their lives meaning, and for the most part avoided thinking about their condition too much, as Jon (16) demonstrated. For him, DMD was not at the front of his mind:

*Sarah [S]: In your day to day life is Duchenne something that you think about a lot?*

*Jon: Not really, just get on with it really.*

Ed (18), however, was unhappy about his current situation. When asked what might make his life with DMD better he responded, *'if I didn't have it'*. Ed has transitional DMD which means that, at the age of 18, he can still walk and has functional use of his arms and hands. Although he is mobile, Ed finds it hard to leave the house or be with people outside of his family, and he appears to regret having a disability. It is also possible that Ed was prepared to more openly acknowledge and express his negative feelings about his condition. This acknowledgment of struggling to cope with a serious condition was also addressed by Emily (21); the only person interviewed who did not have DMD and also the only female participant. She has a rare form of muscular dystrophy [MD] that, as with Ed's transitional DMD, is not immediately apparent:

*Emily: I found it, I went through a bit of a bad stage when I was 12, I didn't really accept, I still find it hard now, to accept that I've got anything wrong.*

Although Emily and Ed have more mobility than the rest of the group, they struggle with their condition, openly expressing the troublesome issues that they face. Both are able to move about unaided, but they have much less stability when mobile than is the case for their able-bodied counterparts, and this dual positioning appears to create its own stressors. Emily wonders if, in part, the struggles she encounters would be ameliorated by the symbolic social cues a wheelchair gives:

*Emily: When someone's in a wheelchair then you know they've got something wrong with them and people know how to treat you, but when you've got something wrong with you people don't know, like especially with me 'cos I don't like telling people that I've got a disability so if I'm out somewhere and something happens to me then like, if I fell over or something I wouldn't be able to get up, I'd be stuck because I wouldn't want to tell anyone so it's quite scary to think like if that actually happened I don't know what I'd do.*

The other interviewees frequently commented that they just got on with things, which may be due to an orientation to living in the present due to an uncertain future (Gibson et al. 2009). For Emily, her dual status as looking 'normal' and yet knowing that she may not be considered 'normal' appears to increase the pressure on her when in a social setting. As she says above, she does not like telling people about her disability, but knows the risk she faces, if she fell over, of revealing her condition and the limitations it places on her. As most of the participants have experience of using a wheelchair, either permanently or for part of their day, their attitude is less speculative than Emily who, due to the uncertainty around her muscular dystrophy, is not sure if or when she may go into a wheelchair. The participants with DMD appear to have accepted their physical restrictions, living with their condition on a daily basis and tending not to dwell on the subject too much (Abbott 2012, p246). Berntsson et al. (2007) propose that, for the adolescents they studied, who all live with a long-term condition, those who developed ways of coping with and accepting their limits, regarding them as a normal part of their life, appeared to be more resilient to difficulties.

Ollie (14), who has a mild learning disability, obviously valued the positive skills he

has gained as a school mentor and he thought his coping strategies could be usefully passed on to pupils at school:

*Ollie: I'm a mentor at school so it means I help out... kids with their problems, at home and stuff and I'm hoping to become a prefect in year 11.*

Tim (10) associated a positive attribute with having DMD as he recounted how he could beat his friends at computer games:

*S: So, why do you think you're so good at games?*

*Tim: I don't know, I think it's because I've got Duchenne muscular dystrophy, it might help me be better on this, but I don't know.*

Tim is aware of his differences but also of what he thinks might be his heightened ability due to having DMD; computer games provide a competitive space within which boys with DMD can compete with able-bodied peers and even beat them (Abbott 2012). Tim was representative of most of the boys who reported having an X-Box or similar games consol; as Jay (14) commented when asked what his favoured pastime was:

*Jay: I just like, go on my X-Box with my friends.*

Although Jay sometimes uses a manual wheelchair he still has good use of his upper limbs and is keen not to be seen as different when with his able-bodied friends. He recounted winning an award for his wheelchair sports but he only took one school friend with him to the ceremony as he tended not to talk about his wheelchair sports activities with them. Carl (18) has friends over to his house, which is more practical as his home has been adapted for wheelchair use, yet, both Jay and Carl expressed some awareness of the potential disparity between themselves and their friends, with Carl stating that his friends '*put up with me*'. Games consoles provide a source of challenge and entertainment that the boys can engage with on equal terms, but for Carl and Jay there is also an awareness of difference from others.

Adam (16) was less interested in computer games and reported that getting out,

keeping busy and maintaining his social life was important to him:

*Adam: I like to see my friends, go to the pub for instance and watch like a football match with a couple of friends or just go out anywhere and just see my friends, that's my favourite.*

Rick (12) commented that seeing his friends was more important to him than playing on his X-Box, and he takes part in a busy and varied range of activities with his local scout troop. Most of the participants have developed ways of coping with their condition, and their lives evidently have substance and meaning for them, however, elements of isolation and difference did surface in the interviews. Some of the participants had times when they felt isolated and different to their peers, and these more negative experiences will be considered further in the next section.

## **Isolation**

The nature of dealing with a disability and of being different can impinge upon individuals, creating physical and social barriers and leading to isolation (Dreyer et al. 2010). Maintaining a stable sense of self can be difficult, as the condition and attitudes towards it impact the disabled individual's life, causing feelings of hopelessness, fear and anxiety (Delmar et al. 2006). In the context of my research, isolation matters because it may be a contributory factor in how the participants approach making decisions, including those on medical research. Gibson et al. (2007) have observed that the lives of those with DMD are compromised by the way in which society is arranged, limiting their social participation and structuring their identities, and they are 'profoundly marginalized, isolated and excluded' (Gibson et al. 2007, p515). The marginalisation the participants experience can impact their confidence in multiple ways, which may then, for some individuals, influence how they think about and experience their role when decisions are being made.

The experience of isolation extends to parents who have a child with DMD, and who have expressed a sense of loss caused by their child's deteriorating condition and decreasing mobility, leading to social isolation (Samson et al. 2009) both for parent and child. This isolation reflects the dominant attitudes of society, where

disability is often poorly understood, situating those with DMD/MD as objects of concern (Noyes 2000) rather than agents imbuing their lives with meaning. These negative views are reinforced by inadequate social and medical support that sometimes fails to meet the needs of a maturing group of individuals with DMD (Gibson et al. 2009; Parker et al. 2005). Where possible, some individuals may hide their disability from their peers (Berntsson et al. 2007), others may not be able to hide their condition, but nevertheless wrestle with their difference and the absence of terms for sharing that difference with others. Emily (21) frequently returned to this conflict, expressing her struggles with physical and existential matters even as we discussed varying subjects. In the extract below, she describes one part of her dilemma:

*Emily: If there was more people with it [her condition] it wouldn't be so different because I don't want to be different, and then I could talk to people as well and see how they feel, 'cos I feel like no one understands.*

Amongst most of the participants there was some awareness of being different. Tim (10) described the swimming sessions he took part in:

*Tim: I go swimming at my brother's old school,*

*S: So do you go swimming with all the other kids in your class?*

*Tim: No it's not at my school.*

*S: Right, it's nothing to do with school then?*

*Tim: No, 'cos I'm the only one that's sort of disabled... there's like, about one other, but there's only me in a wheelchair.*

*S: Oh right, erm how do you feel then about being the only boy in a wheelchair?*

*Tim: Erm fine... people think it's good because I don't have to walk, but I don't find it very good, because it's boring... but I'm alright with it.*

Tim also mentioned how his social life is shaped by his mobility problems:

*S: Do you ever get to go to your friends' houses?*

*Tim: No, not really.*

S: *Why is that?*

Tim: *Because a lot of them have steps.*

Tim uses a wheelchair permanently, meaning that access to homes without ramps is problematic, a situation that Carl mentioned earlier and which Jay also commented on, stating that *'friends normally come round here'*. Disabled adolescents frequently face barriers to meeting with friends outside of school, for instance at cinemas, because of difficulties with physical access (Dreyer et al. 2010), impeding friendships on equal terms (Skär 2003). I asked Jay (14) how he spent his free-time and he described visiting the cinema with friends, which was enabled with his mum's help; she would get him settled at the cinema, and then visit the nearby shops until the film ended, although as Jay commented, *'You're texting us constantly [says this to his mum], you're waiting for me to text when the film's over!'* His mum's concern for Jay's well-being understandably impact the manner in which he conducts his social life; his mum both enables his mobility and sociability but must also monitor his whereabouts because of her awareness of his support needs.

Tom (17) is a sports fan and he described how wheelchair football filled a need for disabled young people in his area:

*Tom: Well we've been doing that [wheelchair football] about 3 years, started at secondary school and 4 of us have Duchenne and there wasn't much for us to do and that's how it started... simply just with a ball and now we have tournaments.*

The lack of suitable activities, until wheelchair football was organised, resulted in Tom and his friends with DMD missing out on the benefits of socialising and having a focus for their interests. Tom's father helped to set-up the wheelchair football and it is a vital source of interest and friendship:

S: *If you were hanging out with friends how does that work?*

*Tom: It's mainly at [wheelchair] football; it's mainly where most of my friends are; sometimes they come round here and play Playstation.*

At a follow-up interview I asked Tom more about the wheelchair football, I wondered

what he would be doing if his dad had not helped to organise it, *'well nothing... there'd be no sport to do or anything like that'*. This sporting activity offers a space for the boys to meet and chat, helping to combat the lack of social and sporting provision for severely disabled young people. Ollie (14) attends the same wheelchair football sessions as Tom and it provides a sense of camaraderie:

*S: When you're at wheelchair football do you get a chance to talk with other lads or are you too busy with the football?*

*Ollie: Sometimes I do shout at them... like to help them out or to see what they're doing 'cos you've got to talk to each other during the game in order to win it... my emotional confidence goes up with David, he's my, well not really my best mate but he's a team mate that helps me out a lot.*

Jay (14) enjoys wheelchair sports too, but as mentioned earlier, he has been missing one of his favourite activities:

*Jay: I do wheelchair basketball... Well I haven't been going for a while 'cos I had my cataracts done and I've got a stitch in my eye... I had both eyes done but I've just got a stitch in that one.*

He has also had to avoid swimming whilst his eyes heal, missing out on sociable and physically viable activities that can enhance wellbeing (Gibson et al. 2009).

Feeling different from others and missing out on social activities can impinge upon the participants' sense of selfhood and identity in various ways and these will be examined next, as they can contribute to the diverse manner in which isolation is apparent in their lives.

### Isolation and the impact on selfhood

Emily (21) is concerned about the possibility of going into a wheelchair, and people's attitudes towards wheelchair users:

*Emily: I think that's the most scariest thing like for me anyway, being in a wheelchair, I think that is what is the most out of the whole thing is to be in a wheelchair, and obviously I'd be scared of dying as well, but for me I've been in a wheelchair before like when I had my operation and I lost all my strength and stuff and... when people look at you as well I just hate it so much.*

Emily's worry is not just with losing mobility, it is also about how people will perceive her in a wheelchair, and the social isolation that may be experienced as a result of this visible difference. Jay (14) explained that when he broke his leg he needed to use a wheelchair full-time for a while which was not a great experience:

*Jay: When I broke my leg and had to be pushed around all the time people would come out and go 'ahh, how is he?' to my mam and dad and I was sitting there and like 'ask me!'; I used to get quite annoyed by it.*

This was an unpleasant experience because of people's patronising attitudes, an experience common to disabled children (Connors & Stalker 2007).

*Jay: They talk to you like you're stupid, quite condescending...there's nothing wrong with me it's just my muscles.*

Whilst he used the wheelchair, people talked over his head or adopted a paternalistic tone of voice, leading to an experience of feeling ostracised. Together we contemplated the social awkwardness some people have towards wheelchair users, and Jay notes the problems disabled people face when utilising a device that visually indicates a disability. The wheelchair provides functional support and promotes independent mobility, yet can represent the negative connotations that surround having a disability in a society that does not project positive values on to what some regard as defective bodies (Hughes et al. 2005). Jay, like others with DMD experiences marginalisation as 'both "normal" and troubling' (Gibson et al. 2007, p514). Not surprisingly, young disabled people may act to minimise visible difference from others when outside the home (Carnevale 2007a), although, as Emily describes, not using a wheelchair is, at times, also problematic:

*Emily: I've had so many times when, I've got a disabled badge and when I've parked... people have looked... and sometimes they've even said stuff to my mum... in a way I'm glad you can't tell I've got a disability 'cos then it is good but at the same time it's really hard because it's always hidden...Just 'cos you're not in a wheelchair doesn't mean that you're not disabled.*

Rick (12) recounted being stared at when he was at primary school and, as Jay found, using a wheelchair can draw comments which may not be meant as unkind, but which underline difference (Connors & Stalker 2007):



*Rick: At my old school everyone used to stare... 'cos of the wheelchair, but now, they just don't really give it that much attention.*

Fortunately, Rick is having a more rounded experience at high school; *'at my school there's loads of people with... disabilities and they mix together with everyone else'*.

Carl (18) struggled at primary school too; he was not diagnosed with DMD until he was 7 and the attitude of some teachers to his apparent clumsiness, caused anxiety and a sense of being different. This sense of difference is troubling at a time when children can find affirmation through their similarity to others (Skär 2003):

*Carl: I used to think it was just you [being clumsy], is it your fault and it turns out to be something, and you're like 'oh thank god it's that!'*

Ollie (14) had different reasons for feeling that primary school was not a great time for him; he missed out on some education and he is still catching up:

*Ollie: I've been in [high] school 3 years and... at primary, unfortunately they didn't get to me as much and I didn't understand... at high school I'm still catching up ... I went to a [primary] school where I only did one SAT [standard assessment test] and that's it and while they did SATs I went off with my teachers and we had some fun but I missed out a lot of things 'cos if I'd done my SATs I'd probably be clever, I probably might have been great at High School.*

Ollie did not always get the same treatment as his peers at primary school, due to his disability and learning needs. However, high school also presents some problems for Ollie as his personalised PE [physical education] sessions, which require a private room, cannot always be conducted due to a lack of space:

*Ollie: We've only done it [PE] about twice this year due to exams or meetings.*

The sessions have also shaken his confidence:

*Ollie: I do physio but sometimes it's hard with my knee getting onto the bed... I'm a bit worried that they'll hurt me, my confidence was jolted a bit one day, my teaching assistant in PE hurt my knee.*

Ollie requires some specialised treatment at school, and this takes him away from the classroom, occasionally reinforcing how having a disability shapes his experiences and identity. For all the participants, the regular problems associated

with growing up can be exacerbated when combined with their physical differences. In Ed's (18) case he feels unable to cope with the rigours of college and a social life:

*S: Are you at college?*

*Ed: No, I never went... don't really like being around people or anything.*

And,

*S: Are you involved in anything, any activities... outside of the home?*

*Ed: I don't really like to go out.*

*S: Right, why's that?*

*Ed: 'Cos every person always bullies me most of the time.*

Ed is isolated as he feels victimised by others, and he prefers to stay home where his disability is accepted and understood.

Jay (14) balances his time between mixing with his school friends and his wheelchair football friends, and as noted earlier, he keeps these groups separate, to avoid the negative reactions of his able-bodied peers (Berntsson et al. 2007):

*Jay: I don't really tell my friends about my condition, I don't want to be treated differently, 'cos they might feel sorry for us or something, which I don't like.*

Like Jay, Emily (21) finds it hard to talk about her condition with her able-bodied friends and, as she has already commented, this could then bring problems if something was to go wrong or she had an accident:

*Emily: I don't like telling a lot of people... but when I do tell them they don't know what it is and then I find it really hard because if I was to go out somewhere and say there were stairs or if something happened I feel like no one will understand and then I don't know what I'd do so then it puts me off doing things because no one really knows what it is, they can't really understand.*

Emily can walk, but her muscle weakness means that she lacks strength and stamina, activities are not always easy and she misses out on 'doing things' as she

does not want something to go wrong whilst she is out. She goes on to explain how the rarity of her muscular dystrophy and the absence of others her age with the condition is a problem:

*Emily: I think it's harder, well, for me in a way because I don't know anyone that's got XX [name of condition]... there's no one my age so there's no one I can really talk to about it as well to see how they feel about it, it's quite hard really, not having anyone.*

She feels the alienation of her situation, with isolation impacting her life in complex ways due to the duality of her identity as both able and not able-bodied and the lack of commonality this creates. The problem of having physical differences but not wanting those differences to define her, limits Emily's adherence to wearing splints; splints help to stabilise joints and slow down the tightening and shortening of muscles (MDC 2009):

*Emily: I've got splints for my arms, hands and my feet, and I've not worn them in probably years... if I wear them I can't sleep and... I don't like telling people I've got a disability... if someone walks in and I've got my splints on, I won't do it, so there's not a time when I can do it, and also because I can't put them on myself... so I just don't wear them.*

Emily finds the splints uncomfortable and they stop her sleeping, reinforcing the negative, relentlessly embodied aspects of having a disability. The splints are not only physically, but socially uncomfortable, setting the wearer apart as different (Carnevale 2007). For the user of such visible devices there can be an intensified awareness of how disability is judged by normalised standards, 'bodies coupled with medical devices do not comply with pervasive standards of legitimate bodies' (Gibson 2007, p507). As Gibson (2007) observes, men with DMD who are completely reliant on visible technological devices are likely to be negatively identified and categorised when outside of their home environment. Marginalisation and prejudice are apparent in society, and are exemplified by the participants' awareness of this prejudice as they seek to manage their disability. Both visible and invisible disabilities can have negative social impacts that shape the 'complex process of self-identity formation' (Carnevale 2007, p11). Ed (18) is not so concerned with how splints look, as he tends not to socialise beyond the family circle. However, he resists wearing night splints as they are uncomfortable:

*Ed: People think they know what they're like... [saying] 'I would wear them if I was like you' but they don't know what it's like to be me.*

Ed expresses something of the ambiguity faced by many of the young people interviewed; their experience is specific to them, they get help and support but also have to live with their condition in a way that those around them do not. Ed struggles with well meaning hospital staff who have experience with DMD and who know the value of splints, yet for Ed, at a personal and embodied level, splints are uncomfortable and he chooses not to wear them.

Tom's (17) frustration at a lack of physiotherapy suggests a sense of marginalisation and neglect. He describes a meeting he attended, at which he felt DMD was not prioritised:

*Tom: The one [meeting] we went to, it was about muscular dystrophy, quite a lot of older people there and they were complaining about stuff, most of it was like physio and ... we're young and we don't get physio and they're like older and complaining.*

Tom's family pay for private physiotherapy, but their sense of injustice at poor provision was apparent, also leading his father to pay for other healthcare, including a private cardiologist. Ollie's (14) struggles with isolation related more to his closeness to his mum, who he missed during respite visits to the hospice:

*Ollie: It's alright, in the day, it's just at night I miss my mum a lot so it's kind of like pretty hard.*

He also missed some of his older friends, who have moved to the hospice's young adult section, he mentioned this as I asked him what he liked about the hospice:

*Ollie: Well having to meet a lot of new people in wheelchairs there, teenagers my age, but a lot of them, my old friends have gone to the other end of XX [the hospice], which is the older [section], 16 to 24 [year olds], and then; well I do get entertained by DVDs and video games.*

Ollie benefits from visiting the hospice, and it gives his family a rest, but his reliance on his mum and the graduation of friends to the hospice's young adult section can make it hard for him to cope sometimes. Many able-bodied adolescents have

challenging times; however, for those with DMD/MD, the complexities of their situation create specific experiences which may sometimes be hard to overcome.

Most of the participants had some aspects of their lives facilitated for them by others, and this does not directly lead to isolation and is vital to their well-being. However, time spent at hospital and the hospice, needing assistance, and having visible and invisible physical differences contours their attitudes, imbuing them with an experiential knowledge of disability. Living with a degenerative, incurable condition creates distinct experiences:

Life... is one of uncertain health... overshadowed with the documented certainties of the outcomes for others. It is an existence in which energy is expended in the constant negotiation of normal and different. (Jessup & Parkinson 2010, pp360-1)

A minority of the participants had limited interactions with others, due to factors such as temperament, the limitations of their disability and physical and social barriers. As Dreyer et al. (2005) observe from their study of people living with DMD:

They try to live as normally as possible, but especially in their teens they have trouble with physical and social accessibility... [and their] social relations are complicated by prejudice. (Dreyer et al. 2005, p5)

The problems with socialising and accessing the venues where their able-bodied friends meet can make it difficult to overcome isolation, and relying on the support of their parents to redress some of the imbalance created by isolation may, concomitantly, serve to highlight differences. The level of support needs amongst most of the participants is very visible, signifying their ongoing need for physical help at a time when their able-bodied friends may be experiencing greater physical independence. Seemingly disparate issues like Jay's operations that kept him away from sports activities, Emily's concerns due to limited medical knowledge about her condition and the duality of seeming to be able-bodied, and Tom's sense of injustice at the lack of physiotherapy emphasise the isolation and anxiety disability produces.

At times, the participants' experiences of isolation may diminish their sense of active engagement in their lives or limit the ways in which they can express

themselves, and the effects of isolation may influence the participants' attitudes and approach to decision-making. Their attitudes, in part, emanate from the experience of living with a serious condition and from a sense of being different and outside of social 'norms', which can be a disempowering experience. Alongside the participants' experiences of isolation and difference, the assumptions adults hold about 'vulnerable' disabled children and young people can also isolate them. Such assumptions can position them as needing additional protection from responsibilities, including the responsibility to make, or be involved in making, decisions on their healthcare (Alderson & Montgomery 1996). As has been observed above, the participants do not dwell too much on their situation. However, it is a possibility that there are times when they live with a sense of being resigned to their circumstances (Gibson et al. 2007), and this sense of resignation may constrain their involvement in decisions for various reasons. What may be read by adults as a child or young person's passivity, vulnerability or disinterest in decision-making may, in fact, be much more complex than that. The child or young person may indeed be passive, vulnerable and/or disinterested, but they may also be influenced by how others identify them. It is possible that 'how persons are identified as members of social groups and positioned within group hierarchies manifests in their embodied understandings of themselves' (Gibson et al. 2007, p516). For the participants, isolation is experienced in ways that are not always easy to define, but which can impact the way they express themselves and their own wishes and preferences.

The negative experiences caused by isolation are not the only ways in which the participants are defined, as they also experience agency and independence, which contribute to enriching their quality of life (Bushby et al. 2010; Miller et al. 1990). The next section will examine the role of independence and how it is thought about and experienced by the participants. Noyes (2000) observes that involving disabled children and young people as active participants rather than passive recipients in treatments and decisions on their care can be an influential factor in building a sense of independence and agency. Whilst the point has been raised above that isolation may contribute to how medical research decisions are made, it is probable that the development of independence can also influence decision-making.

## Independence

The participants experience independence in differing ways, with parents playing some part in how it is realised, as having a disability and needing support can cause independence to be impacted by the attitudes and actions of others. In this setting, important matters may be dealt with by the parents and child together in a relational context; and exploring these contexts provides an understanding of how the participants experience independence. As introduced in Chapter One, Dreyer et al. (2010) have described the ways in which autonomy and independence can be developed in the lives of those with DMD. They contend that growing up with DMD is a different experience to acquiring impairment later in life; with independent dependency being a part of coping. They explain this as being dependent on physical help in order to live, but also being able to exert independence through pursuing friendships, education, sport and using the computer (Dreyer et al. 2010). They also consider how decision-making contributes to making independence 'a real experience... we call this "independent dependency"' (Dreyer et al. 2010, p7). This notion of independent dependency may help to explain the partnerships that were apparent between child and parent in the families I visited. Each family expressed this commitment differently, but it was noticeable from the participants' accounts that there was a commonality of reliance and trust. Out of this relationship of trust, the child can enact independence as they define what they need, rather than having their needs defined by someone else (Cockburn 2005). There are constraints, due to physical dependency, but the participants spoke of the ways in which their own values were important when formulating decisions. Whilst other people play a significant role in their lives the participants can also experience self-directed independence. This will cause the decisions they make to be formulated not just with the help of their parents but also with attention to their own directives, which are developed as they experience and grow in independent thought.

Independence can be expressed in the autonomous ability to do things without help from others, and equipment is vital in enabling this. Rick (12) made a point of emphasising how valuable his wheelchairs are to him:

*Rick: I forgot to mention this 4x4 one that I've got... they're like pneumatic tyres, that's quite a good wheelchair, it also tilts to allow you to get up and down slopes and stuff so it tilts backwards if it's quite steep going down.*

*S: So like if the family were going out for a walk?*

*Rick: Yeah I can come too.*

Ollie (14), in summing up his life noted that having all the equipment he needed was beneficial to him as it enables his mobility:

*Ollie: I've got everything really, I've got loads of equipment I need and, yeah I'm fine, I'm just a normal young adult, I've just got something wrong.*

Ollie describes how equipment helps him towards being a 'normal young adult', and like Rick, his all-terrain wheelchair enables him to enjoy family walks. A parent in Carnevale et al.'s (2006) research recounted the value her sons placed on their electric wheelchairs; they were 'very, very upset when their chair breaks... and they're dependent on us ... they get very depressed, they can't move' (Carnevale et al. 2006, p56). Tom (17) recounts the distress he felt at his sudden loss of independence after spinal surgery; and he was enthusiastic about his arm supports which enabled him to regain some independence:

*Tom: My arm supports, I'll tell you about these, I got these when I had my operation on my back [for scoliosis] ... it meant I couldn't lift my arms high enough to reach my mouth... I wanted to feed myself, it's not nice being fed and so what happened was my dad rung this guy and he just came out and did it and we paid for it, it was such a desperate situation, not desperate but... I wanted it then... they're amazing [he demonstrates them] I can move side to side... so they didn't recommend it or anything, they'd never heard of it [in hospital].*

*S: So how did you hear about it?*

*Tom: We just saw it once, a demonstration of it.*

Emily (21) recalled how she had to stay in hospital longer than was necessary after an operation, due to a lack of equipment; she remarks how vital her mum was in bridging the gap in resources:

*Emily: I had to have an operation when I was about 14, 15, and I was in*



*hospital for quite a long time and I got to a stage where, because, if I don't move around and keep active I just become really weak so because I was in bed all the time I couldn't walk ... and I had to have, there was this machine that got me out of bed and moved me to a chair, and I couldn't go home until I had that because I couldn't do anything and my mum couldn't, the weight, so and my mum was trying to sort it out with... [2 towns were arguing over funding Emily] and both of them were arguing over who's going to fund it, and I was in hospital and I didn't need to be in hospital because they wouldn't agree who was to supply this machine ... but I needed my mum to help me, she lifted me and stuff, I couldn't have done it, I wouldn't have been able to come home without her.*

Emily's physical independence and her return home were compromised by the lack of coordinated care, illustrating how equipment is vital in supporting independence (Noyes 2000). Emily was denied access to options and self-direction due to the absence of the equipment that would enable her independence. Independence is actualised through making decisions and directing one's care (Dreyer et al. 2010) but this can be problematic when the material means are not in place to facilitate this. Emily's experience of hospital care was troubled and she required continued parental support to overcome inadequate provision and regain some independence (Parker et al. 2005; Sloper 1999). Parents often step into the breach due to inadequate support, providing assistance and helping their child towards independence, meaning that their life is closely enmeshed with their child's. Occasionally, and sometimes in extremis, the parents must act not only as an advocate for their child, but also as a facilitator of the most basic necessities for living.

### Being independent

As Ed (18) has good mobility he is not so reliant on equipment, but he nevertheless feels his disability compromises his independence, stating that '*my disability stops us doing things I want to do*'. However, he did not want his disability to define him, and he wanted recognition of his independence and personhood, as was apparent when I asked him how he thinks young people should be spoken to at the hospital:

*Ed: Treat them like a person... just because they've got a disability doesn't*

*mean they can't talk back to you properly.*

Ed was not always happy to be given advice at the hospital, implying the problematic nature of some hospital encounters, although Rick (12) was much more positive in his views on hospital visits. Rick felt that as he matures it would be good to have some appointments on his own:

*S: Why would it be good to have some appointments alone?*

*Rick: Probably saves my parents' time and stuff, and probably you feel more confident about doing it 'cos you're more independent.*

In a second interview (by telephone) I revisited this discussion with Rick:

*S: Why do you feel it's important to be heard at the hospital?*

*Rick: Well I think as you get older it becomes more important, they're talking to you not your parents.*

Rick viewed being able to speak with clinicians as a part of confidence-building and gaining independence, and he recognised that ultimately the doctors are addressing him. Jon (16) was very quiet and shy in sharing his thoughts with me, but he made the point that his opinions should be sought at hospital appointments:

*Jon: They [doctors] probably should ask me questions if it's something about me instead of asking my mum.*

Jon felt his own voice should become more prominent as part of a move towards independence, and independence was also on Jon's mind when I asked him why he decided to participate in my research project and how he would like to see my findings used:

*Jon: Probably to help us [boys with DMD] be more independent... 'cos like if you talk to someone that didn't know about it or something, they probably wouldn't really know, so if you ask people that have the condition it probably helps the research more.*

Jon recognises his expert understanding of living with DMD and how his contribution may improve independence for those with DMD through giving others a more

thorough understanding of their lives. When I moved on to asking what his motive might be for taking part in some medical research, independence was again a primary concern:

*S: What would be your motive [for taking part]?*

*Jon: Probably just to help people with the condition*

*S: In what sort of ways?*

*Jon: Probably like I said, to be more independent.*

*S: So being more independent in what sort of ways then?*

*Jon: Erm like going out to places.*

Jon's poor mobility curtails his access to some levels of independent choice about what he can do and when, and his version of independence relates to 'going out' more, and he regards independence as a worthy goal for social and medical research. He uses the local hospice for respite care and a local charity takes him out for recreational visits, but spontaneous activities are harder to realise due to his support needs. This emphasis has also been noted by Young et al. (2007) in their research with young disabled people; they observed that self-direction was particularly important for the older boys, 'often taking the form of a desire to visit friends or to go out unaccompanied' (Young et al. 2007, p664). Thus, the question of independence, which in my research contributes to understandings of the way decision-making is approached, also has broader implications. The broader reach of independence encompasses helping young disabled people to live well through making their own choices; having a level of self-determination and, with reference to equipment, having the material conditions in place to support a good life.

Hopes and plans for the future are significant in maintaining an independent outlook as the participants cope with their disability and work beyond it to 'enjoy life and fulfil their dreams' (Dreyer et al. 2010, p6). Ollie (14) spoke of what he hopes to achieve; *'I've got loads of ambitious dreams for the future and let's hope I get to the place I want to be'*, and one of those dreams relates to physical mobility and independence:

*Ollie: [I'm] hoping to drive as well, I think I'll try and I'm hoping to get a job, might go to college and stuff and hopefully get enough money for a car... 'Cos I admit... you don't really want your mum or dad driving you, you'd rather drive yourself.*

Ollie also has some hopes for the more immediate future, centred on school activities and his expertise with disability:

*Ollie: There's meant to be a [disabled] girl or guy coming [to school] so hopefully I can give them advice as well because I've been there.*

As we discussed current and future treatments for DMD, Ollie reflected that there was little in the way of treatments that could help him, but he felt he could still invest in his social skills, which he clearly values:

*Ollie: After those years you can't really do much can you [regarding treating DMD] but I can at least improve my social skills and stuff and make me more confident.*

Helping others at school provides Ollie with a means to develop his skills, and his hope of becoming a school prefect gives him a realistic goal, exercising independence within the frame of his condition. Ollie's frank acceptance of the limited treatment options for DMD will be discussed further in the next chapter, where, as will be seen, this influences his views on medical research.

Adam (16) exerts his abilities and independence as he makes time to go to London to campaign for a future that would see better services and funding for DMD:

*Adam: I do go up to London to lobby Parliament to give more money; we go and stand near Downing Street with a load of banners and stuff.*

*S: Right, was that with, as part of a charity group or..?*

*Adam: That's with XX which is like the charity that's sort of trying to get stuff.*

*S: Yeah, right, so was that your decision to go or was that?*

*Adam: Yeah I like to go, 'cos I could stay here if I wanted to and mum just go, because I could get a carer but I like to go and show my support.*

*S: Do you feel it's good to be involved in, in your case lobbying, is that something you feel positive about?*

*Adam: Yeah I really do, I think it's a really good idea because erm me and other boys are the ones we're trying to lobby for so we might as well show them that we're there and sort of helping.*

In a second interview (by telephone) I asked him more about what his motivation would be for getting involved in lobbying and attending meetings:

*Adam: It would be mainly personal learning to learn about the treatments and get involved in how I could raise money and stuff and understanding what we're raising money for.*

It is Adam's preference to have his voice heard, exerting his developing independence and autonomy (Kirk 2008), and he hopes to remain a visible, vocal presence on disability issues as he matures.

Rick (12) saw developing his independence as being an investment for the future:

*S: Is it important to you to be independent as you mature?*

*Rick: Yeah I think so... as my disability progresses I get weaker so I just try to be independent as much as possible.*

*S: Could you tell me a bit more about what you mean by independence?*

*Rick: Like, you don't have someone to help you all the time and you can go off and do things by yourself.*

Rick experiences independence as he journeys out on his own in his electric wheelchair:

*S: Can you remember when you first ventured out on your own in the wheelchair... is it gradually, that you've gradually built up?*

*Rick: I have kind of built up 'cos erm, yeah my mum, we used to walk there and back to school, we used to take the car but then mum walked me up there and back quite a lot and taught me how to be safe on the road and stuff and then gradually I just built it up and just managed to be able to do it by myself.*

*S: And that's been, that's while you were still 11 isn't it, when you started to do that?*

*Rick: Well I think it was before that actually it was while I was still 10 I used to do it a couple of times. 2<sup>nd</sup> interview (by telephone)*

Rick's independence has been facilitated by his mother who has supported him in venturing out unassisted, and his ability to achieve this skill does not appear to be directly related to his age. Not every parent would give their wheelchair-using child such freedom, but for Rick it is valued as part of his growing autonomy. It reflects his competency in accessing the physical environment and making decisions, such as about crossing the road, whilst also having the broader experience of self-determination. Rick also mentioned the value he places on being articulate, and I asked him to further explain this:

*S: So for you are social skills something that are important to you... in your life?*

*Rick: I think so because I just think friends are the most important thing or the most important people that you have in your life and it's really good to be able to, be able to speak to people and have more friends. 2<sup>nd</sup> interview (by telephone)*

He enjoys mixing with students with a range of different disabilities and also with non-disabled people at school; *'there's loads of people with loads of different disabilities and they mix together with everyone else'*. For Rick, investing in social interactions and sociability is valued as an activity that builds bonds despite his physical limitations, developing a sense of selfhood and agency as part of a group of disabled and able-bodied peers (Williams et al. 2009).

The participants each manage ways, within their familial setting, of realising independence. Some have adapted better than others to their physical situation, and most are building their hopes for the present and future into the reality of their physical limitations. Dreyer et al. (2010) report how a participant in their study who used home mechanical ventilation explained that even though he was dependent upon others, he considered himself to be an independent person living his own life. Dreyer et al. (2010) suggest that those with DMD adjust as their condition alters, with their view of independence being modified by experience and expectations, although they observe that the teenage years can be challenging due to social isolation. Jon

(16) and Rick's (12) comments that, for them, independence is linked to going out and doing things may have specific relevance to their age group, where independence and agency often relate to the freedom to go where they want when they want. Able-bodied adolescents can act out their maturing selfhood through absenting themselves from their parents' presence, but this is harder to achieve when poor mobility requires adult help, therefore a desire for distance from parents and home can be challenging to realise, and the shape of independence inevitably differs. The constraints imposed by DMD/MD mean that the participants must, paradoxically, rely on support to achieve some independence (Briscoe & Woodgate 2010), and parents may need to actively promote their child's independence (Alderson 1993). Thus, issues of independence, dependency and the development of personal agency and autonomy intersect in multiple ways, with each family attempting to manage the tensions.

Independence can be difficult for the participants where physical barriers, poor support, and stigma persist (Carnevale 2007; Connors & Stalker 2007; Gibson et al. 2009). The participants are embedded within the contextual, relational dimensions of their life (Carnevale 2004) where there is no seamless access to independence but nor is there only dependency. Independence may evolve in a different arc for those with DMD, with periods of adjustment as the condition changes (Dreyer et al. 2010), but this does not make their experiences of situated agency (McLaughlin & Goodley 2008) less valid. Common discourse, exemplified in media portrayals, regularly depicts disabled children as tragic or heroic, embodying identities that are subsumed within their disability. This ignores the fact that disabled children and young people occupy a broad spectrum of abilities and talents that reflect their complex humanity and identities (Goodley & Tregaskis 2006). There have been commonalities across the interviews, but the participants have also dealt with issues according to their own disposition and values. For example, Emily (21) has considered both the merits and disadvantages of her dual positioning; sometimes it is good to 'pass' as normal but at other times it is less helpful. This alternating identity indicates the context dependent nature of agency for individuals, who, whilst sometimes experiencing marginality and isolation, should not be regarded as victims (McLaughlin & Goodley 2008). The participants are also capable of independence

and agency as they work to cope with their constraints, whilst focusing on the goals many young people have for their lives. They have demonstrated elements of independence through sharing their views with me, as we discussed some of their experiences and explored the scenarios, showing to various degrees the ability for independent thought.

An integral factor in the participants' lives and sense of self is their knowledge about their condition, the process through which they have found out about it and the manner in which they accrue knowledge on it. Parental attitudes coupled with that of the clinicians providing care, alongside each participant's own outlook, will mediate how information is dealt with and used and this is likely to create specific competencies and attitudes to medical research and decision-making. In the next section consideration will be given to how the participants learn about DMD/MD, from what sources, and how this information is incorporated into their lives.

### **Finding out about DMD/MD: diagnosis & getting information**

This section will examine two different types of information gathering; how the participants came to find out about and understand their diagnosis, and how they approach finding out a range of information about DMD/MD. These issues help to identify how the participants are given information and what motivates them to find out more about their condition; these processes may also inform and motivate how decisions are made. Carl (18) was relieved to find out his diagnosis, which was relatively late compared to other boys with DMD. He had, as mentioned earlier, been labelled as clumsy by teachers at primary school and his reaction to hearing he had DMD was relief:

*Carl: Well I was glad it was that... so I didn't know until I was 7 so I was wondering when I was in infants like why I was slow, why I was slower than everybody else.*

Obviously this is a different reaction to that of parents, who can face a difficult time of adjustment to their child's diagnosis (Webb 2005). What it does indicate is how



getting a diagnosis may help individuals deal with a health condition. I discussed with Carl his thoughts on a situation in which a parent withholds information about diagnosis from their child. Carl was emphatic that withholding knowledge from children was not a good idea:

*Carl: He should have been told years ago, but she should really tell him as quickly as possible... 'cos he's going to be wondering why he can't do all that and getting upset.*

The layered emotional, practical and psychological factors parents must deal with can impact the way in which they share information with their child, who may, as with Carl, benefit from knowledge which situates physical limitations as being part of the condition, rather than a personal failure. Yet some parents may respond to their parental role as protector by withholding or adjusting information they think is too negative for their child (Carnevale 2004).

Some of the participants could not recall when they first knew they had DMD as it has always been a part of their lives. Adam (16) was representative of this approach, and his mum has gradually helped him understand more about his condition. He regarded having this information as a useful factor as we discussed an imaginary young person with DMD who has not been told their diagnosis:

*Adam: I think he should be allowed to sort of, he should definitely be told what he has... but definitely be told that that doesn't change what he could do because it doesn't... at that age when he knows he's obviously got a disability 'cos I mean obviously you're like still walking but walking badly then you might not know you've got a severe disability and you might not have been told by then but once you're in that wheelchair... I think you should know.*

Adam expresses the value he places on having and using information to his benefit, and of using this knowledge as part of his investment in his life and getting the most out of it. Rick (12) had a similar attitude; he had recently found out the full facts of his condition after he asked his mum to explain more about DMD. Rick reflected that it was the right time to get detailed information, such as about life expectancy, and I asked him how he felt about these interactions:

*S: Can it be useful then, [when] your mum talks to you about it, is it useful to*

*have those sort of conversations?*

*Rick: Yeah I think so.*

*S: Not necessarily fun but,*

*Rick: Well yeah, I still don't mind.*

Carl (18), possibly due to his own experience of getting a late diagnosis, felt that boys should be told about DMD in an age appropriate way:

*Carl: Well you tell them in a way they understand so if you talk about a lot of stuff a 7 year old's not going to understand it, but if you try and dumb it down as much as you can so they understand the basic idea and then explaining it more as they get older as they start to understand more.*

Simultaneous to the young people dealing with information about their condition, their parents also encounter challenges as they find out and deal with information.

Emily (21) explains how her mum has at times found it hard to attend meetings to do with muscular dystrophy:

*Emily: My mum was saying when she went that even she didn't, like she heard about all these muscular dystrophy conferences and stuff but she couldn't hack going either because like the same thing as me, as if you don't want to see people and then it's scary especially as we didn't know that much about XX [her condition] at the time, there was nothing on it, there's still not much on it now either.*

Emily faces problems due to the rare form of her condition and both she and her parents have found it hard to attend conferences as they don't know how Emily will fare in the future. This uncertainty is difficult to deal with when they encounter other young people at events who have a range of muscular dystrophies that are better understood and have a clearer diagnosis and prognosis. Tim (10) knows some of the information about DMD from overhearing his mum's conversations, some of which are quite unguarded:

*S: How do you find out about things to do with DMD?*

*Tim: Err I don't know... I just know what it is,*

*S: So how have you found out?*

*Tim: 'Cos like I've asked my mum... well other people have asked her and I've heard what my mum says.*

*S: So she's the expert?*

*Tim: Yeah; my mum, she only found out about Jon [his diagnosis of DMD] when he was about 5 and if she would have found out earlier she wouldn't of had me.*

Jon (16), Tim's older brother with DMD, recounted how he found out information along with his mum:

*S: As you've been growing up and you've maybe had a question about Duchenne is that something that you try to find out about on your own?*

*Jon: Not really, probably mostly with my mum.*

Jon, as the eldest son, may need to ask his mum questions directly, and he and his mum are learning together as his condition progresses, whereas Tim appears to receive some information indirectly from his mum, in her conversations relating to DMD. Tim's comments highlight the ways in which children pick up on snippets of information, as Carl (18) notes:

*Carl: I tend to know what's going on 'cos I tend to overhear conversations, [laughs] phone calls and stuff so I know what goes on and stuff like that.*

*S: If you had a direct question about DMD... might you ask mum and dad first or might you do your own bit..?*

*Carl: Well I ask and if they don't know I kind of research it myself... but I've never actually had to do that yet... but I would if I needed to.*

In line with most of the participants, Carl uses his parents as a source of information about DMD/MD; they are a trusted and reliable point for verifying and supplying knowledge. Jay (14) commented that he used the internet to find out details about DMD in conjunction with his mum's support:

*Jay: I'd just have a chat with my mam; have a look on the XX [charity's] website and stuff.*

At this point his mother, who was present during the interview, intervened to check with Jay if he would be more likely to ask her first, prior to doing a web search; she had earlier commented to me on the unreliability of some information on the internet:

*Jay: No; I'd have a chat with you first [says this to his mum], 'cos my mam knows quite a bit about it, so I'd just ask my mam.*

Emily and Jay are aware that the internet contains some poorly informed and researched information and they have learnt that it is good practice to use trusted sites:

*Emily: When I first found out I did find it really, really hard and I did look on the internet which made me feel worse and my mum told me not to because she said there's stuff on the internet that might not even be true... now I think I'd look on the XX [charity's] website.*

With some of the older boys, who are losing hand and arm function, using their computer is difficult and none reported having speech activated software; therefore problems with using the computer may reduce independent searches for information. Tom (17) was representative of many of the participants when asked how he went about finding out details on DMD:

*Tom: I don't know, I don't really look for much information really.*

*S: Like if you just had a question?*

*Tom: I usually ask my parents or, I don't know I don't really ever have that much to ask... there's nothing really, I just take it as it comes really.*

He was not unduly concerned about finding out information, wanting instead to get on with his life:

*S: Do you ever feel there are things you'd like to talk about or ask about if you had the chance?*

*Tom: No... I already know things,*

*S: You know enough?*

*Tom: Yeah... I know quite a bit about it.*

Tom has many years experience of living with DMD and this experience may be why

he has few questions he needs answering. When I asked him what he talked about with his friends he mentioned that occasionally someone may bring something up regarding DMD, but that mostly their conversations were around 'normal stuff':

*Tom: Football erm, college and school, [football] tactics, [laughs] just about anything that's going on, just normal stuff.*

Tom also mentioned using the hospital and doctors as a source of information, as did some of the other participants, and these comments will be explored, along with a broader discussion on medical care, later in the chapter.

Perhaps reflecting his inquisitive nature and youth in comparison to the other participants, when I asked Tim (10) if he had anything to add at the end of our time together he responded with a big question:

*Tim: Who created it [DMD]... like, how it happened, when it first started?*

This question is beyond the reach of most adults' understanding and expresses a desire not just for the details about DMD, but for a grasp of the greater picture within which it sits. Some of the older boys commented that they already have most of the information they need, this was exemplified in Tom's earlier comment that he '*already knows things*', and their attitude seems to be to accept their condition and get on with life. Some participants reported looking for information, but were careful where they gathered this from, only using reputable sources, and amongst the majority of participants there was an attitude of focusing on the things that bring meaning to daily life. It was not apparent that any of the participants spent a long time researching their condition online or through other mediums, although key figures for information when mentioned were parents, doctors and trusted websites. This does not imply that the participants were indifferent but, perhaps, that they sought not just to live with DMD/MD but to live beyond it; they know what needs to be known, and their interests and activities are their main focus. This was exemplified by Ollie's (14) response when I asked him if he used the internet for information on DMD; he responded that he did not use the computer much as a research tool '*because to be honest I only want to find out the facts on the net*'. With

regard to information, it is not lack of interest that appears to cause this group to report a minor amount of knowledge-seeking; rather, most reported operating in an integrated way with the adults in their lives. Such an approach may be specific to this group, who, due to their condition, do not achieve a stable period in their health trajectory during childhood or adolescence. This is in contrast to young people who have a more consistent health condition (Giarelli et al. 2008); as a result, my participants generally continue to work with their parents in seeking information as they mature into adulthood.

Whitty-Rogers et al. (2009) discuss the importance of keeping children who are seriously ill informed about their care, enabling them to ask questions and 'voice' their opinions. Parents may think it right to shelter their child from painful information, but this can be detrimental to the child's autonomy (Whitty-Rogers et al. 2009). Amongst my participants, there appeared to be a consensus that even serious news should be shared:

*S: Do you feel it's good to know as much as possible?*

*Rick: Erm yeah I think it is because the more you know the better you know how to handle it. 2<sup>nd</sup> interview (by telephone)*

Here, Rick (12) expresses a mature attention to the utility of being well informed as a means of coping, and it helps him to 'handle' his condition. Amongst the participants there did not seem to be a direct correlation between age and the amount of knowledge they had, and for most, there has been a gradual release of information as it is needed. The role of parents in sharing information and providing support is a central factor in the participants' lives and, due to the integral part they play, parents will be further discussed in the next section.

## **Parents**

This section will discuss how parents are involved in the lives of the participants, wherein the dynamics of the relationship with their child can influence and inform how decisions are approached and made.

## Supportive carer

Most of the participants are physically dependent on their parents at a time when their able-bodied peers are extending their physical and emotional independence, and DMD/MD is not grown out of but grown into (Jessup & Parkinson 2010). The complexity of requiring enhanced care as physical dependency increases can modify independence (Skär 2003), with parents providing support that places them in a central role of care. This was apparent as I discussed the local children's hospice with Ollie (14). He has already talked about missing his mum when he stays there overnight, and he also misses the familiarity of his parents managing his physical care:

*Ollie: I'm quite used to my mum and dad doing it [providing care] all the time so I'm not used to other people doing it.*

Emily (21) has more physical independence than Ollie, but she relies on familial support and on knowing that her mum is there for her. As Emily explains, her mother has continually fought to ensure adequate provision for her daughter:

*Emily: Yeah my mum's had to fight for so much for me... I wouldn't have been able to... my mum's helped me so much.*

Emily recognises the challenges her mum has encountered and how hard she has had to 'fight' on Emily's behalf because of limited healthcare provision (Sloper 1998). This terminology of having to 'fight' is repeatedly mentioned at DMD/MD conferences and by parent groups in the UK and other countries. It signifies the poor provision parents contend with and which contributes to parental stress (Sloper 1999), and it situates parents as their child's advocate, pressing for improved services and better funding for treatments and a cure. Jessup & Parkinson (2010) describe the struggle for parents who have a child with cystic fibrosis, with their life being a fight 'in which parents are engaged whatever the cost, emotionally and financially' (2010, p357). Parents must push for what their child needs from inconsistent and labyrinthine services, whilst also meeting their child's physical care needs.

Ollie (14) was aware how hard his mum worked, and this was reflected in his

imaginative response when I asked him what equipment or gadget to assist him he would like to see created if possible:

*S: Is there anything you could think of that would make your life a bit easier?*

*Ollie: I really don't know; if I could create a robot servant.*

*S: A what?*

*Ollie: You know, to help my mum out, just do everything.*

*S: Yeah, to help your mum out.*

*Ollie: Yeah and it would know all the complications and figure out what... it was programmed for what my mum usually does.*

*S: Ahh, so something to help mum, that's a really nice way of looking at it.*

*Ollie: I made that up with my inventions.*

*S: So you're not just thinking about yourself are you, you're thinking about mum and you're thinking about what could help?*

*Ollie: Yeah, a robot wouldn't get tired at all. 2<sup>nd</sup> interview*

Although I expected Ollie to come up with something that would assist him, he recognises how integral his mum is to him. He is sensitive to the 'complications' she faces in managing her growing son's physical needs. He also mentioned how his parents act as a resource in thinking up ways to keep him active:

*S: So you sound like you've got quite a busy life really.*

*Ollie: Yeah... but if mum and dad can't think of something I'd probably get bored indoors so you need stuff to do don't you?*

Ollie's family also enjoy holidays away from the stress of daily life and work:

*Ollie: It's been quite stressful, my mum's back got [bad] and my dad's been quite stressed at work so it was nice to get away just for a few days.*

Ollie is an only child and is aware how hard his parents work. He frequently spoke of himself and his parents as 'we' and 'us', implying the closeness of his three-way



family unit. Tim (10) described the logistical challenges for his mum, who has to coordinate two sets of hospital appointments, as both her sons have DMD:

*S: Do you and Jon go in together [to hospital appointments]?*

*Tim: Sometimes, once... the doctors sent my mum a letter saying I had an appointment, just me... When we got there they asked us 'where's Jon?', but my mum said it only said my name on it, but she was right.*

Tim and Jon's mum has a full-time job caring for the boys and ensuring they attend all their appointments. She drives a large adapted van to accommodate two electric wheelchairs. When I asked Tim about visits to the hospital he mentioned a practical problem his mum has; '*well it's quite hard to get parking spaces*', and he describes how she misread the time of a dental appointment, thus missing their time slot:

*S: Oh! was she annoyed?*

*Tim: Sort of... with herself.*

Tim gives a glimpse above into the daily work his parents face. His mum, as with other parents of a disabled child, must field a range of health professionals who are vital but whose presence could be regarded as an invasive necessity (Jessup & Parkinson 2010, p358).

Not only must parents provide care, but they also need to absorb and share relevant information on current muscular dystrophy treatments, care and research. This situates parents as educators and advocates (Carnevale 2007), and as advisors and gatekeepers for their child, and these latter two roles will be discussed next.

### Advisor and gatekeeper

Several parents used meetings run by charities to follow current treatment and care advice for DMD/MD, often feeding back to their child any relevant news. Webb (2005), herself the mother of an adult son with DMD, discusses how parents of a child with DMD become experts on their child's health. Parents equip themselves with advice and information via the internet and support groups (Blackburn & Read 2005; Webb 2005), whilst also dealing with the knowledge they may outlive their son

(Hamby et al. 2006). Hodges and Dibbs (2010), in their study of how parents used a DMD support group, observed that many parents sought, through experience, to focus on the positives in their child's life, using 'active coping'. They found that parents modified their use of the group according to the stage of their son's disease, with involvement plateauing once parents had developed experience both from the group and from being the parent to a disabled child. Indeed, Adam's (16) mum appeared to be 'actively coping'; she was instrumental in keeping up to date on DMD news, whilst realistically sifting that information:

*Adam: [Mum] checks up on like emails saying what's happening and things and reads magazines like that one [on the coffee table].*

*S: Yeah and then does mum tend to share it with you a bit or?*

*Adam: If she finds something interesting yeah but if it's just the sort of same thing then it's not much sort of interesting then, not particularly.*

Rick's (12) mum attended meetings run by a DMD charity:

*S: And what's the focus of the meetings?*

*Rick: Giving parents advice on how to do things better with disability and stuff.*

Rick was aware of the utility in having an articulate, well informed mother and how this helped him to develop his own communicative skills:

*Rick: My mum's really good at like language and stuff like speaking and reading and stuff, so I read quite a lot myself so I understand quite a few words, it's a bit easier for me to understand them [the doctors].*

Ed (18) regards his parents as his primary source of guidance:

*S: So do you have anybody like, an advisor, do you have anybody who gives you any advice or..?*

*Ed: Me mam and dad.*

Although Ed's (18) dislike of mixing with other people may cause a more intense reliance on his parents, all the participants voiced their trust in parental advice. Jay (14) consulted his parents when he had questions:

*S: So who in the family, or is it a mix of mum and dad, who's the one you go to with questions?*

*Jay: Both my mam and dad, sometimes all three of us, we'll discuss [it].*

As we considered a scenario where a young disabled person is making up their mind about taking part in medical research, Emily (21) explored how reliance on a parent could, she thinks, have broader implications on the decisions a young disabled person makes:

*Emily: I don't know, if it was my mum, because, I don't know what it is, if my mum was really, really pushy about it [making a decision to take part in medical research]... I don't know why I'd just feel guilty like I was letting her down or, and because she does so much for me I'd feel a bit like, don't know why, I think I would feel guilty though, just because my mum's telling me to do something and if I'm not doing it and she really wants me to and then I don't, but I think my mum now at my age wouldn't... she has pushed me before though, with certain things.*

In the extract above, Emily is conscious of the nuanced dynamics of the parent/child relationship. The participants' need for parental care, compared to their able-bodied peers, may create a differently weighted relationship. Emily's assertion seemed to be speculative; although she is conscious of the ways in which a close relationship with parents gives their advice a persuasive influence. Skär's (2003) research with disabled adolescents revealed a similar dichotomy:

The relationships were described as close and mutual at the same time as neither close nor mutual, because the adolescents perceived that their parents controlled them... the adolescents were concerned that they might come into conflict with their parents if they did not openly express their appreciation and satisfaction with the help they received from them. (Skär 2003, pp641-2)

It is apparent that some parents strive to protect their children, through gatekeeping the information they are privy to and acting as 'communication executives' (Young et al. 2003). They may filter what their child is told and shield them from being involved directly with decision-making. With Rick (12) I explored an imaginary scenario in which a parent withholds information on DMD from their disabled child. I asked him what he thought of that situation:

*Rick: Well I don't know I just erm... I don't know, I find it quite strange that people would like withhold information and stuff like that.*

*S: Why would you feel it was strange?*

*Rick: You can't really not tell people things, like 'cos in order to have a more successful life you need to know kind of those things.*

Rick appreciates knowing as much as possible about DMD, and information has been shared with apparent openness; *'when I was younger I had it explained [DMD]... I've just known it as we find it out'*. His surprise at a parent withholding details reflects his own experiences, where knowledge is part of a shared interaction.

Carl (18) places trust in his parents' advice regarding his physical health and treatment options. His parents have bought a machine to use at home that stimulates blood flow to the muscles, and Carl recounted how he decided to try the treatment. It was apparent that there was a family commitment to Carl's well-being through regularly using it:

*S: Thinking about this treatment that you're having erm, what made you decide to give that a go, what was like the process that you went through?*

*Carl: Well my dad told me about it, and I just said I'd try it 'cos he recommended it and I might as well try it and I tried it and I stuck with it.*

*S: So you tried it and it's an hour and a half in the morning and,*

*Carl: An hour and a half at night, round about.*

Carl's dad acted as an advisor, spending time and money in practically addressing his son's health needs due to the paucity of DMD treatments. He also acted as a gatekeeper, enabling his son's access to what he considers a beneficial treatment. In a differing way, Emily's mother has acted as a gatekeeper, securing support for her daughter's well-being:

*Emily: My mum's on the phone for hours... she just doesn't give up and she'll push and push and I think you have to be like that to get anywhere.*

Emily's mum is not only a gatekeeper but also acts as a firebreak, defending and securing Emily's rights, and this was common amongst the participants' accounts,

with parents assisting their child towards some autonomy and simultaneously providing protection (Barron 2001).

Carl and Ed, who are both 18, and Emily, who is 21, voiced a continued desire for parental input into aspects of their lives, illustrating the nuanced implications of developing autonomy. The notion that once a young person is 18 they can make their own decisions is blurred by the attitudes of these participants; it places parents in a continuing role as advisor and counsel that does not see a clear demarcation as the individual matures. By expressing a wish for parental support, these young adults could be considered to be demonstrating agency in this choice (Alderson & Montgomery 1996). It would appear to be a prudent and logical preference, as their parents have followed their child's health from the earliest stages and they have a chronological knowledge of their child's condition. Parents play a vital and positive role in their child's life, but there can be negative elements too. The bond between parent and child may, as Emily observed, create a close relationship where the child feels they cannot act independently due to the perceived debt of care (Skär 2003). For many disabled adolescents, relationships with their parents provide a safe space, but there can be asymmetry, with parents controlling decision-making and acting as 'obstacles to... independence' (Skär 2003, p642). No other participant, apart from Emily, mentioned a sense of debt as a factor in decision-making, although support needs and the intimate family setting from which decisions emerge can complicate the ideal of informed consent as an autonomous process. The approach to making a decision about medical research participation may be formed by diverse and hard to identify influences and factors, and the close involvement of parents in their child's life is likely to have some effect (Alderson 1992).

It is understandable that, in the role of advisor and gatekeeper, parents guard their child, and this protective role may be useful during the regular hospital visits the participants must make. The hospital is a site where it was apparent the young people appreciated being accompanied by their parents, and in the next section this supportive role when talking to doctors and being at hospital will be discussed.

## Parents' role in communicating with doctors

During hospital visits parents act as moral support for their child, a role that will have been assimilated at the time leading up to and after diagnosis and which continues with their child's developing maturity. Physical deterioration in DMD/MD means that constant surveillance is required, as what is 'normal' is always changing (Samson et al. 2009). Samson et al. (2009) consider that in this altering terrain the parent's role persists, but in a changed configuration as their child matures, with parents acting as a facilitator. Ed (18) considered the hospital a source of information not just for him but for his mum too; '*well, she only knows what I know ... what the hospital tell us*'. He finds visits to the hospital can be intimidating, and it is reassuring to have his mum with him:

*S: Do you like the idea of having some appointments without mum in the room?*

*Ed: [shakes his head] Don't like to be on my own around people.*

*S: So it's reassuring to have mum there?*

*Ed: Just in case I say anything wrong.*

Amongst the other participants there were assertions of trust in their parents' presence and guidance when interacting with clinicians, even though, like Ed, they are young adults, as Carl (18) explains here:

*S: I know that some boys have said that they, when they're in a hospital appointment, they're happy for mum and dad to do all the talking.*

*Carl: Most usually they'll give me... and I'll speak up if I need to, usually I do it yeah, 'cos they know the science stuff and I just kind of sit there watching.*

*S: Yeah, yeah, and I think that's the sort of impression I've been getting, that the boys speak up if they need to... so perhaps when you're a bit older would you perhaps want to have appointments on your own or...?*

*Carl: Well obviously, eventually yeah I will do appointments on my own... I never say 'yes' [to having one alone] I always say I want you [parents] in, I never say 'no'.*

Emily (21) lets her mum speak on her behalf, acting as a 'communication buffer'

(Young et al. 2003) during medical encounters and co-managing some exchanges:

*Emily: I do like the fact that the doctors speak to me, I wouldn't like it if they ignored me, like I couldn't talk or something, but I do like to have my mum there as well and then she can talk to them if I don't really want to.*

Adam (16) was content for his mum to take some control, but he would speak up if he judged it was needed:

*Adam: I'd be happy for mum to say everything at appointments but if I didn't agree... I'd be inclined to interrupt and say 'that's not quite how I see it'.*

Tom (17) expressed his continued need for his parents' presence during consultations:

*S: When you're a little bit older sort of 18,19, would you like to have some appointments on your own do you think or would you still like,*

*Tom: No, no, there's never that many... to be honest.*

*S: So you think you'd probably still want...[your parents there]?*

*Tom: Yeah; I don't know really, it's just the way it is I suppose.*

Tom prefers to have his parents there, acting as a resource in his communications with doctors (Young et al. 2003); and, as will be seen in the next section, the apparent lack of trust Tom has in his doctors may contribute to his unease at being unaccompanied. In describing the roles parents can play in doctor/patient interactions, Young et al. (2003) note parents sometimes act as an 'envoy', with the young people briefing parents to seek information on their behalf or as with Emily earlier, acting as a buffer. Young et al. (2003) also describe parents as 'human databases' who store information, or as 'communication brokers' who customise and reiterate information into manageable forms for their child. They observed that young people's preferences for parental support in interactions were fluid and contextual; however, they also contend that parents may constrain some interactions with doctors (Young et al. 2003). This implies the complexity of the three-way communicative process, in which parents may feel acutely responsible for their child (Alderson & Montgomery 1996; Young et al. 2003), occasionally limiting their child's

participation. This dynamic could shape how decision-making proceeds in some families, with parents feeling that they must protect their child from making the wrong decision, whilst also wanting to guide their child towards some level of self-direction. The participants' views on medical research and how parents are or should be involved in decision-making will be further discussed in the next chapter.

This section has looked at how parents figure in hospital encounters, whilst the next section will explore more directly how the participants themselves experience talking with doctors and being at the hospital.

## **Doctors and hospitals**

The participants' relationships with a range of healthcare professionals rely on a level of trust in which the participants must comprehend their treatment needs and engage with small and occasionally major decisions on their healthcare. The participants have first-person knowledge of the health service, and their time at hospital and interacting with health professionals contributes to their web of experiences. Alderson et al. (2006) note in their study of children with diabetes that consent and refusal was a part of daily life due to the regularity of the children's treatment regimes, and the children developed competencies in line with their experiences. Alderson et al. (2006) add that their research addresses a gap in the literature that needs to be filled with the voices and medical experiences of children. So too with this research, which, in the next section explores the participants' thoughts and their experiences on communicating with doctors.

### Communicating with doctors

Effective communication with doctors is vital in ensuring the participants' health needs are met and that they are increasingly involved in their own care through making incremental choices and decisions. The importance of being able to effectively communicate with doctors is exemplified by Adam (16) as we discussed how he approaches talking with doctors:



*S: If you had a question you wanted to ask the doctor, would you feel you could address them yourself?*

*Adam: With the ones that don't talk to me probably not, but a lot of them do talk to me as well; most about 90% of them do, well at least a little bit.*

Adam is confident and eloquent, yet occasionally he feels that he is not treated with adequate respect, and at his second interview (by telephone), he explained the importance of being fully included in consultations:

*S: Is it important that doctors do talk to you?*

*Adam: Yeah it's very important to me that doctors talk to me because when they do talk over my head it seems quite rude, and I do understand what they're saying and so that means they can talk to me 'cos I know what they're talking about.*

Adam is prepared to seek clarifications when needed as he recognises the doctors' expertise:

*S: Is it ever hard to understand what the doctor has said, I mean are they using technical phrases that, or do they break it down?*

*Adam: Erm, they normally break it down enough because like my mum for instance might not understand the technical ones because she isn't a doctor so I think they've got to break it down enough for parents anyway so I think it's kind of OK... But if like I didn't understand I could ask most of the doctors to make it a bit simpler for me to understand because I don't understand all the doctor terms.*

*S: Yeah, ideally they're kind of putting it in terms that you can understand or explain in English,*

*Adam: Yeah 'cos not being a doctor I wouldn't know it all... that's why they're doctors.*

Tom (17) was unsure how he felt about his experiences as a patient:

*S: Thinking about medical appointments... and your dad was saying you'd gone private for some stuff?*

*Tom: Yeah,*

*S: Erm, so when you go to a typical appointment,*

*Tom: Yeah, sometimes you go and they just talk rubbish!*

*S: Yeah, and how do you find them... there was a report about young men with Duchenne and they said they don't always feel included, like the doctor's not necessarily talking to them, how do you feel?*

*Tom: No it's not, it seems OK... I never know exactly what to ask them, they're OK with me, it's just that I don't know...I think that they're OK.*

In the extract above, Tom was light-heartedly ambivalent about his hospital encounters and this is also reflected in his next comment:

*S: Do you have someone you tend to trust and listen to more on your condition in general?*

*Tom: If it was somebody medical then I think that they'd [the doctors] possibly know what they're talking about... they probably don't!*

This ambivalence, conveyed through Tom's darkly humorous comments, implies a lack of complete trust in doctors.

Emily (21) considered some appointments to be pointless, involving little more than a reiteration of established facts, partly as there is not much knowledge on her rare condition:

*Emily: It's hard as well because it's really only Dr X who actually knows [about her condition] like when I go to hospital now it's really annoying, I go for an appointment... and sometimes it's like a different doctor... there's no point going because they're basically asking me what's wrong with me... just them finding out what's wrong with me and not telling me anything and it's pointless, really annoying.*

Adam (16) was also unsure if all his appointments were productive:

*Adam: Some appointments they kind of see you and nothing really happens and it feels a bit pointless sometimes, they have to be done but they're just a bit pointless, a lot of them are good but some of them are just, they kind of say stuff we've just said.*

Adam accepts that the appointments have to be done but mirrors Emily's comments that some of them seem 'pointless', covering matters that have already been discussed. In contrast to Emily and Adam's comments on the futility of some

appointments, Rick (12), who attends the same hospital as Adam, found talking to doctors a positive experience:

*S: How do you feel about visiting the hospital?*

*Rick: Yeah, quite good I think, because, I do rely on the hospital quite a bit so I don't mind going.*

And he felt confident talking directly to doctors himself:

*S: Some boys have said that they feel left out, like the doctor's just talking to the parents.*

*Rick: I don't really 'cos I usually talk myself as well... they usually talk to me instead of my parents.*

Rick felt included in interactions and was confident that he could follow what was being discussed:

*S: Do you ever find it hard to understand what the doctor or nurse is saying?*

*Rick: Not really, I just know most of the things.*

He also regarded hospital appointments as efficient:

*S: If you could tell doctors how you feel about hospital appointments, what would you want them to know, is there anything that could make them better?*

*Rick: Erm, don't really think so no 'cos they're quite efficient at doing it so they're usually quite quick.*

Despite using the same hospital, there is a marked contrast between Adam (16) and Rick's (12) attitudes. Adam is 4 years older than Rick and therefore has paid more visits to the hospital, and there is a sense of boredom brought on by the long journey and time spent waiting around:

*Adam: Erm, a typical appointment, I'm already a bit bored because I've been in the hospital transport for a couple of hours up to XX [name of hospital] and I'm kind of like want to be home really... and I don't really like waiting around, you have to wait around in hospitals, get bored.*

He also wanted discussions to be concise and focused on health matters:

*Adam: ...because they're kind of your doctor not your mate sort of thing, you want to get going, want to go home.*

Ed (18) did not mention boredom as a factor, but still wanted hospital visits to be as short as possible, '*I just want to be in and out to be honest*', and, as has already been observed, he is not entirely comfortable in his role as a patient:

*Ed: Like I've got enough to deal with and like they're telling us to gan [go] out and mix with people, and I dinna [don't] want to; it's the way I am.*

*S: Yeah, do you think that they could talk to you in a better way than they are doing?*

*Ed: No not really, I think they do an alright job just, I don't really like doing all that stuff... and they have stuff to do with my health but they're going on about like other stuff like mixing, and going to college.*

He recognises the hospital are doing an 'alright job', but struggles with reconciling his desire to just get on with his life with the need for health monitoring and interventions. As he has an older brother with DMD there is a reminder that his condition will not improve and the certainty of continued hospital visits seems to be a source of some stress, rather than an informative and useful event.

The participants demonstrated varied responses to communicating with doctors; Rick (12) wants to take a lead in consultations, whilst others, such as Emily (21), are happy for parents to co-communicate. This does not, however, equate to a lack of competence or poor communication skills amongst the participants, who all appear to take their healthcare seriously (Alderson & Montgomery 1996). Rather, it may be a response to the repetitiveness and seeming futility of some appointments, or to how marginal they might feel in consultations where they feel that their priorities are of little interest to medicine (Young et al. 2003). Adam's (16) knowing and poignant comment about his interactions with doctors when visiting the highly respected hospital he uses expresses a dilemma experienced amongst the participants:

*Adam: The doctors do sometimes... speak over your head a bit...and I'm sort of thinking 'I am here'.*

Adam appreciates his mum's support when at the hospital, but he also appreciates

his own active presence being acknowledged. He does not want to do all the talking, but neither does he want to be ignored.

### Medical encounters

As well as routine appointments, the participants all have experiences of medical procedures, ranging from injections and blood tests to major spinal surgery. I asked Jon (16) to give me an overall idea of his hospital visits:

*S: Could you give me like an idea of the sort of, a typical month say, what range of appointments you have?*

*Jon: Erm I don't think it's that many, it's a couple, there's like the heart scan one and I went for the ones for my back operation but I think I've been discharged from that one, then I had my ventilator thing err I had to go in for a sleep study for that... and I had to go back in to see if it was doing what it should be doing.*

In Jon's estimation, there were not that many appointments, and yet, as he enumerates them, the list appears quite varied, drawing upon the expert knowledge of a range of clinicians. Like Jon, Tom (17) has had spinal surgery to correct his scoliosis, and his attitude, that it needed to be done, helped him through it without needing to deliberate the decision:

*Tom: With me well they did an X-ray on my back and I could tell I was leaning a bit in my chair... and I went in and the Doctor doing the operation spoke to me and explained it... then I asked if I could look at the X-ray [laughs] and it was just really bad, I saw it and I thought, I couldn't tell it was that bad... so when he showed me how bad it was I thought... right I've got to have this done.*

*S: And did you discuss that with your mum and dad?*

*Tom: Yeah a bit, there wasn't much to discuss.*

Ollie's (14) experience after surgery for scoliosis was not as straightforward, possibly as he was not communicated with appropriately, and he did not feel cared for by his doctors in the aftermath of surgery:

*Ollie: I remember I woke up in HDU [high dependency unit], but there the doctors weren't good, I had to have 2 blood transfusions ... which was bad and after I was sick and they were 'no, no he's fine' but I needed to take sickness tablets 'cos I'm quite allergic to like tablets.*

Ollie felt the doctors did not attend to his needs and were not listening to him, and later, as we were discussing Ollie's attitude to repeated trips to the hospital for appointments, he returned to his experience of the operation:

*Ollie: Yeah 'cos I'm used to the hospital now because I've been there so many times ... but also when I was in HDU I got really angry... I had a back catheter in but that really hurts it digged in and I was crying my eyes out and they wasn't listening.*

At our second interview, I wondered how Ollie felt things could have been improved in that situation:

*Ollie: Well if the nurses stayed a bit more and they can tell the doctors 'cos they went 'oh no no no, he'll be fine', they wouldn't know what to do with me, 'cos of all the patients.*

*S: So did you feel that the doctors and nurses were listening to you or,*

*Ollie: No I felt quite left out.*

Ollie states above that he is 'used to the hospital now', but his time in HDU implies that he has had some problematic experiences there, his surgical needs were managed, but his distress was not adequately dealt with. Alderson (1993) reports similar issues with young people who had regular hospital admissions; their fear was generated by bumpy trolley rides or physical handling which left them feeling unsafe. Emily (21) had a negative experience when she was younger where there appeared, as with Ollie, to be a communication breakdown which was badly handled:

*Emily: I've had a few doctors though who have been not very nice ... there's this doctor, and I hate going, because it's the same one every time and he's just so rude and speaks to me like I'm just a piece of dirt... and I remember I went there and I was young at the time so I wasn't like very, and you know when you're young and he was like 'take your top off' and there was like 5 other men in the room... and I was like 'I don't really want to, I feel uncomfortable' and he was like 'well how am I supposed to see your back then' and he did it really, really rudely and I was, I didn't know what to do and I looked at my mum and there was a nurse there and she said 'I'll get you one of those [gowns]'.*

*S: So it's important that they have respect?*

*Emily: Yeah 'cos it's like, he's above me and he speaks to me like I'm some kind of...*

Emily's right to respectful care was undermined by the doctor's attitude of unconcern about her wish for modesty, and in her comments Emily is conscious of the unequal relationship wherein the doctor is the more powerful figure (Barron 2001). As a young person she was reliant upon him to take the initiative and act in her best interests, which he failed to do.

In the course of a general discussion on visiting the hospital Ollie (14) recalled having an X-Ray, which produced some safety concerns for him:

*Ollie: I'd say everything's fine at the hospital, I don't mind going to appointments, I always miss school and the only main thing I had before was at the X-Ray room they only had a stool and I was worried about falling off it but now what they do is get a hoist and then I'd be able to sit on a chair so I got more confident 'cos I have to have another X-Ray next year.*

*S: OK, so, sorry just going back to what you were just saying so they use a hoist to sit you in a chair?*

*Ollie: No well my mum and dad used to lift me on but they do hoist me in a kind of chair and instead of moving you around they move the chair around.*

His need to feel safe when being handled is important, staff may have had the situation under control, but for Ollie it felt precarious. As Ollie made a point of raising this issue, it suggests that attending to each patient's needs is an integral part of their care and well-being. Ollie and Emily's problems may seem incidental to major health concerns, however, young disabled people should feel they can voice their worries in the confidence they will be validated (Alderson & Montgomery 1996); when healthcare professionals do not respond to young people appropriately this can lead to anxiety. Emily felt vulnerable and Ollie felt uncared for and insecure in the two incidents he recalled. Young people's compliance is often assumed by hospital staff (Alderson 1993), yet each experience involves the patient's personal safety and dignity, which requires staff to be vigilant and courteous in their practice.

Alderson and Montgomery (1996) observe that young people can feel powerless when they have stitches removed or injections given as these are potent symbols of other people being in control. Emily (21) talked about her fear of injections which has increased over the years, a common complaint amongst young patients regarding what they consider are the worst aspects of being in hospital (Alderson & Montgomery 1996):

*Emily: Now that I've got older I've got worse with like injections and stuff, when I was younger I had like a biopsy... and loads of blood tests... and it didn't bother me as much when I was really young it's like as I've got older 'cos I've had bad experiences with it and stuff and now I hate them.*

Due to these 'bad experiences', Emily now 'hates' some medical procedures, and as will be seen in Chapter Four, this impacts on her criteria for taking part in medical research. Regarding a decision Jay (14) had taken to start testosterone injections to help him grow, when it came to starting treatment he needed time to think it over:

*Jay: Yeah well when we were actually there... 'are you actually sure you want to'... I had to think it over.*

He took some time and decided he did want to go ahead with the treatment, giving him a sense of control over the decision, which he is now happy to have taken. Unlike Emily, Rick (12) was pragmatic about injections and did not regard them as a problem:

*Rick: I just let them do what they've got to do, it takes a shorter amount of time to just let them do what they've got to do.*

As has already been seen, Rick does not mind visits to the hospital, and he approaches medical encounters as a source of help and support. The participants are all accustomed to going to the hospital, but they express a range of attitudes to treatments and interactions, with bad experiences undermining confidence in their care. The variety and quality of their experiences and the attitudes of hospital staff contribute to each participant's personalised view of the healthcare system.

The participants must cope with the multiple strands converging in their lives



regarding their healthcare and its demands, and these experiences differ in distinct ways from those of their able-bodied peers. Young people with a chronic condition can feel isolated due to repeated hospital trips, through missing out on the social elements of school (Nicholas et al. 2011). They may find the hospital environment to be a place of 'strange medical surroundings' (Grinyer 2007, p274), or a boring place that keeps them from normal life. The need for vigilance with their health means the participants must continue in a relationship with the hospital and its staff, and in those relationships there can be an inequality of power (Sartain et al. 2000). Whilst attendance at the hospital is not forced, it does involve compliance and consent to care and treatment, which is a necessity; with agency and autonomy shaped within this necessity of care. The participants' attitudes and comments on their medical encounters reflect their level of confidence in their care, and this may then influence how they draw on such experiences if thinking about medical research participation. What is apparent is that the participants appreciate being respected and talked to with a mix of compassion, constructive purpose and honesty, as Emily (21) states; *'with doctors, just for them to treat you like they would treat their daughter or their son'*.

Relationships between doctors and patients can be productive where doctors work responsively, recognising that each patient's needs differ, with young patients variously wanting to 'control, share in or refer decision-making' (Alderson & Montgomery 1996). The wish to 'control' decision-making may reflect the young patient's growing sense of autonomy, whilst 'sharing' appears to be the favoured option for the participants. 'Referring' might also be an option if the participants are overwhelmed by choices or not well enough to decide for themselves, and at such times parents are relied on to help. Parents can act as spokesperson and advocate, sharing the burden of being a patient and helping to manage the tripartite relationship between patient, parent and health professional (Alderson & Montgomery 1996).

## **Conclusion**

Prevailing attitudes to disabled children and young people can reflect outdated

assumptions and stereotypes that position them as dependent (Alderson & Montgomery 1996), vulnerable and ill-suited to making important decisions about their health care. The problems and issues the participants face may not be welcome, and occasionally can be very limiting, but it was not apparent that they found their limits completely insurmountable (Berntsson et al. 2007) and this may be as they have adjusted, over time, to the lived reality of DMD/MD (Grootenhuis et al. 2007). This is not to minimise their experiences of inconvenience and pain at a physical, social and emotional level, but to surmise that, as Adam earlier stated, they are 'here', present in the world and living lives which are far from tragic or one-dimensional. The participants have expressed a range of opinions that carry a weight and gravitas borne out of long experience, despite their youth. They are, through various means, living and coping with DMD/MD, and their parents and to some extent their doctors help them to negotiate the tensions between the need for care and the growing need for independence.

Some of the participants have times when isolation causes concern; being different from their able-bodied peers, having physical support needs that their parents or carers must meet, and their inability to spontaneously undertake activities can each contribute to isolation. The individual's temperament, circumstances and context also have an impact on how isolation is coped with, and interventions which aim to diminish social isolation and promote inclusivity would help improve life for boys and young men with DMD (Bray et al. 2010). Some of the participants seemed to have accepted their situation and were 'getting on with life'; this was noticeable amongst those with less mobility, perhaps as they have fewer options open to them. Dissatisfaction and annoyance were also expressed, as the participants react to the imperfections of health care and the poor social positioning of disability, which contributes to a sense of being marginalised.

Independence was differently experienced in each family, as the participants live in individually specific circumstances, and the abilities they demonstrated during discussions with me appeared to be somewhat aligned to their familial context. However, their familial context is counter-posed by the participants' own preferences, which may temper the ways in which their parents help them. Broadening out from

home life, the participants live in networks where their agency is acted out but is also acted upon by social and physical barriers and by the prejudice that disability can provoke. In this environment the participants and their families work to redefine and live beyond mainstream definitions of independence. These definitions often portray adolescence as a time of growth and physical autonomy that is expressed through mobility and separation from home. For the participants this separation is not readily viable, yet there are ways in which they can develop independence and volition, as they make decisions and cope with the lived reality of their condition.

As discussed, the participants' lives are interconnected with their parents and doctors, who each play a part in ensuring the young person has a voice and participates in decisions about their care (Whitty-Rogers et al. 2009). Medical treatment and medical research decisions evolve from communicative processes that are not undertaken in isolation (Alderson & Montgomery 1996) but are part of an interpersonal, supportive process (Ho 2008; Tan et al. 2010). For the participants, family input and well-judged guidance from doctors may help to nurture agency and decision-making capabilities, and the participants have described something of this process. Ho (2008) states that even for adults, their identity is often rooted in their social and familial affinity to others and that the family's role in medical decision-making should not be overlooked. Disabled children and young people live in social networks of engagement, where they draw upon the knowledge and opinions of those they trust, and where their choices can also reflect the broader reach of their cultural communities (Carnevale 2005).

What was notable from the participants' accounts was how they worked with their parents to gather and comprehend information on DMD/MD and how parents also co-managed communications with doctors. Parents act as repositories of knowledge, and this model of supported independence provides an adaptable strategy in communicating at the hospital. Because communication at the hospital may at times be problematic, parents remain involved, protecting the best interests of their child but stepping back as their child matures and taking a watchful stance. By implication, the role of being a watchful guardian may persist in the process of making a decision to participate in medical research; with child and adult working

towards the 'best or least harmful decisions' (Alderson & Montgomery 1996, p63).

In the next chapter, the issues that have been discussed so far will help to contextualise the participants' comments on how they might make a decision to take part in medical research. The relationally contingent way in which the participants manage their healthcare, and the effects of both isolation and independence that have been examined in this chapter, help us to understand how these experiences feed into what the participants say next about medical research.

## Chapter Four

### Research decisions

The previous chapter has considered how the participants live and cope with DMD/MD, some challenges they encounter, and the impact of isolation and independence; and each of these factors contributes towards shaping their identities. The participants' identities are also shaped by the way information about DMD/MD is given to them, and by how they and their parents think about and use that information. Experiences at hospital and during communications with doctors were explored, and these experiences, alongside their familial and sociocultural setting help to contextualise this chapter's exploration of the participants' attitudes to medical research. Living with DMD/MD may affect how the participants, as a distinct group with a chronic progressive condition, approach thinking about medical research, although the views to be explored are also specific to each participant, reflecting their individuality. This chapter will explore some of those views to help identify the approach to decision-making. For some, thinking about and discussing medical research with me was a speculative process, whilst others were able to draw on real experiences as they have taken part or are considering taking part in research. The attitudes and thoughts shared assist in developing understandings of how and why decisions are made; emerging as they do from the participants' experiences as socially embedded beings. Through engaging with me in these discussions on medical research, the participants have demonstrated their capacity for reasoning and evaluation, and contributed vital knowledge to my research.

In understanding the participants' attitudes to medical research it is useful to establish how their thoughts may be informed by the fact that a cure or viable treatments are still some way from availability. Condin's (2002) observation, partly cited in Chapter Two is more fully referenced here, depicting how some parents manage exposure to medical research news because it is too painful, disappointing or irrelevant to them, due to their child's advanced disease progression:

This trepid or hesitant approach was a learned one; parents adopted it after

the ensuing disappointment of having had hopes raised before. Research news is perceived as capable of causing harm if exposure to it is not carefully controlled, because it confronts parents with the terminal nature of their child's condition. (Condin 2002, p28)

It was mentioned in Chapter Two that after the first interviews it became apparent I needed to devise scenarios (see Appendix C) in order to connect medical research to possible real-life settings for the participants. There appears to be under-exposure to information about medical research, or of minimal family discussions on it, as there is, of yet, no effective treatment or cure; notwithstanding steroids which, whilst having significant side-effects, can prolong muscle strength for a period of time in DMD. Developing the scenarios proved useful in relating medical research to the participants' own experiences, contexts and understandings. In families where the child and parent work together to build knowledge on DMD/MD, parental attitudes and their way of dealing with information may guide and influence the participants' thoughts. Alternatively, some of the participants may follow their own interests and concerns on medical research information irrespective of family practices, and the scenarios offer a means of exploring these various possibilities.

Before more directly examining the participants' thoughts on decision-making it is useful to begin by discussing any hopes and expectations they have for medical research and how they think it may help those with DMD/MD. The latter parts of this chapter will then consider how the participants think research decisions should be made, what their attitudes and motivation for participating might be, and what they think about the risks and benefits of research participation.

### **Attitudes to medical research: hopes and expectations**

Ollie (14) has previously commented that there is little in the way of drug treatments that could help him at his stage of DMD, and his brief experience of taking steroids when he was younger was not a positive experience:

*S: Are you on steroids?*

*Ollie: No, I used to be, in year 2 but I was getting a bit aggressive towards teachers and pupils and stuff... Which wasn't my fault it was down to the*

*drugs so we gave up on it [steroids], within about probably 2 or 3 months.*

*S: I think a few boys have found that.*

*Ollie: That's the thing with steroids it can make you a bit aggressive... I swore at the teacher, I used to knock kids off their chairs but it wasn't my fault... it was the tablet's fault for doing it.*

Steroids are advised as a standard treatment for DMD (Bushby et al. 2010), but Ollie, as with some of the other boys, has opted not to continue with them due to side-effects. The wish Ollie expressed in the previous chapter, for a robot servant to help his mother, articulates something of his very low, or non-existent expectations of drug treatments that can help him. Ollie's thoughts as we moved on to discussing medical research imply his hopes are not so much grounded in reality, where he realistically assesses there is little hope for him; instead he considers some imaginative solutions. His response comes as we discuss if he might be prepared to participate in medical research that would not bring any physical benefits to him. He thinks he would be happy to take part, but then he also adds:

*Ollie: If I thought there would be a goal for making me walk again but I don't think there's no such thing really... to a point in the future when they make a lot more extreme chemicals that could probably make me, I'm just thinking of a machine that can just, could probably, in the future, I'm looking forward to inventing stuff like, things that can make my legs walk and stuff and hopefully create a time machine that, it's not really a time machine but it can put you back to where you were, imagine if you want to walk and you needed the toilet or something and you press back and then go back and walk which is kind of cool.*

*S: That would be amazing!*

*Ollie: And get down stairs and stuff... I want to do it but I can't.*

*S: So, ideally, if medical research could create a time machine [laughs].*

*Ollie: I don't think it will, but maybe one of my inventions, but I'm hoping they can make something like that.*

In the above quote, Ollie refers back to a time when he could still walk and negotiate stairs and visit the bathroom unaided, and his imagination offers him a glimpse of this prior experience of physical autonomy. This is, he acknowledges, now in the past, and only reachable with a time machine.

Tom (17), in a different way, seemed conscious that there are few viable options for him:

*S. Is that [medical research] something you discuss as a family?*

*Tom: Not often no... not really,*

*S. So it's more keeping up to date...?*

*Tom: Yeah with how things are going, there's not much to discuss really, I don't know,*

*S. And to your knowledge, has mum or dad ever said 'would you like to get involved with some medical research'?*

*Tom: There's never been any.*

At a second interview I revisited some points with Tom:

*S: I'd noticed that you erm... you seem not to spend too long thinking about, researching stuff on Duchenne... could you tell me a bit more about why you don't tend to do much research on it?*

*Tom: 'Cos I don't see what there is to research [laughs].*

*S: You don't see what there is?*

*Tom: No, I don't know what there could be.*

Ollie and Tom live with the physical reality of their condition, dealing with decreasing mobility and having few workable options for minimising their ongoing physical decline. They express their losses in differing ways, Ollie, who is a film buff, applies his imagination to how he would like things to be, with his time machine taking him back to when he could walk. Obviously he knows this is impossible, but his response portrays the losses he lives with and has had to adapt to. Tom recounts the rapidity with which the disease has progressed as he describes how he did not think he would lose mobility in his arms. When he first saw arm raisers demonstrated he could not imagine ever needing them, *'we saw a demonstration of these about 2 or 3 years ago, I thought 'oh I'll never need them''*. As he described in Chapter Three, spinal surgery impeded his ability to lean forwards to feed himself, necessitating the rapid purchase of the arm raisers. These comments, in differing



ways, convey how the participants' lived experiences bring with them the knowledge that medical research is unlikely to slow or halt their loss of mobility; however, their attitudes do not appear to indicate an absence of hope so much as a realistic acceptance of how things are. Adam (16) was pragmatic in assessing the progress and possibilities of medical research:

*S: So is medical research something that you kind of spend much time thinking about?*

*Adam: Erm, not too much, I mean if there's any developments yeah I would think about it, but at the time; but then it's, there's no point sort of pondering it when nothing's happening, when there's nothing major happening... Just got to wait for it to happen, wait for them to make another stage.*

As Adam states above, he would give consideration to medical research 'at the time', however, until such a time, his focus is not particularly on the subject, and he manages his expectations by not 'pondering' the subject much. Tim (10) would like to see medical research effectively resolving his mobility problems:

*S: If you were a doctor or scientist working in medical research what would you like to invent or create to help boys with Duchenne?*

*Tim: Err, an operation to sort them out.*

*S: Right, an operation, yeah erm, so what sort of operation... if you were the most brilliant doctor in the world... maybe something to help you walk?*

*Tim: Yeah and sort of like, sort of, not have the problem anymore... and you can walk again.*

Tim mentioned in the previous chapter that being in a wheelchair can be boring, which he went on to explain was 'because I can't walk', and he would appreciate independent mobility being restored to him. Tim's brother Jon (16) expressed more modest hopes from medical research as I asked him what he knew about the subject:

*Jon: Don't know, not much really,*

*S: Right, is that something that interests you, is it something you think about?*

*Jon: Err...maybe a bit.*

*S: What sort of things would you be looking for drugs to do for you... or for other boys with Duchenne?*

*Jon: I don't know, like I said something that you could move your arms like without anything or... like to be able to itch my head or something.*

Jon is 6 years older than his brother Tim and his wish to be able to scratch his head may denote how, over time, the participants modify their expectations because of their embodied experience of their condition. Tom has spoken about his loss of arm mobility and for Jon it is also a problem, it is partially solved through the use of the arm-raiser device, however, it is noteworthy that both boys mentioned this loss rather than recalling when they went into a wheelchair. Reduced arm movement may be experienced as a more serious marker of muscle loss, as arm mobility is vital in facilitating a range of tasks related to personal volition. Adults with DMD report that their independence is significantly enhanced by the continuing use, however limited, that they have in their hands. One man with DMD, interviewed in another piece of research, commented that controlling a wheelchair independently enables individuals to get out of the house. Hand strength also:

vastly improves an individual's ability to use a computer, which is often a window to the world for people who may be isolated, and is a social lifeline. The ability to use a mouse or keyboard can enable individuals to engage in gaming, one of the few interactive leisure opportunities available to adults with DMD which can have a huge impact on wellbeing. (TREAT-NMD 2013)

Both Tom (17) and Jon (16) have limited arm movement, which has happened at a younger age than with some of the other participants, and this was a notable indicator of loss for them. Their comments illustrate the deteriorations the participants deal with and adjust to, and how their focus on what medical research may be able to do for them is modified as mobility decreases.

Carl (18) accepts that there is unlikely to be an instant fix from medical research:

*S: What would you describe as medical research?*

*Carl: Like... it's weird, it's weird medical research, well you'd want both, finding how to fix it and to have like, 'cos they can't fix it, like, so ways to make it better and ways to make you feel better, that kind of thing,*

*S: So even if they can't fix it, so even if they can't find a cure it's ways of making it more manageable?*

*Carl: Freezing it or... making it a bit better.*

Freezing the disease would be a viable option, and Carl is aware this might be the best he can hope for; it was noted in the Introduction to this thesis that clinical trials on exon skipping hope to halt or reverse the symptoms of DMD, making them more like the less severe Becker muscular dystrophy (MDC 2013). However, exon skipping is still at the trial stage and Carl's quote about not yet being able to 'fix' DMD and Tom's (17) earlier assessment that he does not know what there is to research portray how expectations, for the older boys, are constrained by their experiences. Their thoughts on medical research are not just formed through external knowledge of the subject, but through their physical losses, which manifest the realities and limits of medical research.

Condin (2002) speaks of how parents who have a child with DMD adjust their hopes for research, particularly as potential therapies will be more effective if given to younger children; and 'time and chronicity were salient components' (Condin 2002, p33) of their narratives. The parents in Condin's study, and in my research the young people themselves, must manage the tensions of dealing with the hopes and disappointments of research. Condin (2002) surmises that the parents he interviewed do not give up hope but that it shifts in response to their child's changing physical status, with some 'reconfiguring what hope is – from hoping for the future to hoping for the present' (2002, p35). So too with the participants, the regular need to adjust to their diminishing physical abilities draws upon, or develops, a realistic assessment of how applicable medical research is for them. Ollie (14) and Tim's (10) imaginative hopes are also wishes about how they would like things to be, which may appear improbable, but which are not altogether dissimilar to the hopes adults have when their child is diagnosed with DMD. Samson et al. (2009) observe that the parents of a child with DMD can initially 'have a profound belief that ongoing gene and stem cell therapies may produce a cure' (2009, p108), a belief which alters as the parents find ways to cope without any real hope that research will help their child. Similarly, the participants in my research have adapted to the lack of progress in

medical research that can assist them. Their comments in the previous chapter portrayed how they are busy living their lives in the present and for the immediate future, and this current discussion suggests they do not envisage a cure or 'freezing' of their condition. The participants seem aware, although they did not state it as an outright assertion, that they are unlikely to be the recipients of a medical breakthrough.

### How should funds be spent?

I explored with the participants how they thought funds raised by DMD/MD charities should be spent, to gain an insight into their personal priorities from research. Some families are active in their support for fundraising and in promoting the public profile of DMD/MD as a means to fund more medical research; it can foster feelings of excitement and empowerment at being involved (Hodges & Dibbs 2010). With this in mind I approached asking the participants their own opinions on fundraising and how the money should be distributed. I asked Rick (12) if he had been involved in fundraising activities and he described how his presence may have helped to stimulate charitable giving by passers-by:

*Rick: I did before for the XX [charity] I think where they fund wheelchairs, me and my mum collected about a 100 pounds for them.*

*S: And what activity was that, just holding the [collecting box]?*

*Rick: Yeah mum was doing that and she wanted me to come 'cos if they see me in the wheelchair they probably want to donate.*

At his second interview (by telephone) I asked Rick how he thought the money raised should be spent:

*S: Do you think they [funds] should be spent on equipment as well as finding drugs for Duchenne?*

*Rick: Yeah I think it should be 'cos some of the equipment can make your life better so I think yeah balance that out as well.*

Emily (21) enjoyed the feeling of being involved in raising money for a muscular dystrophy charity, she has organised a coffee morning and has also held a collecting

box outside a sporting event, and she hopes to do more in the future:

*Emily: It's just nice and makes you feel involved; it's nice it makes me feel good, especially when I get quite a bit of money.*

She was not so much focused on a specific goal for fundraising, but enjoyed doing something sociable and positive that used her organisational skills. Rick and Emily were the only participants to mention being directly involved in collecting public donations of money for charities, although Adam lobbies for more funding on DMD/MD. Two participants mentioned that their parents had taken part in bike rides for charity, whilst Carl's father is closely involved in the organisation and fundraising activities of a DMD charity.

Tom (17) values physiotherapy as a direct benefit to him, and he identified this as a funding need when I asked him how he would prioritise the distribution of funding:

*Tom: Good question that, a tough one that... would I say research or would I say physio...? I don't know, I don't know which would be best, they'd both be useful.*

*S. Both be useful... so like you say, research, or the other side of it, more physio.*

*Tom: I wouldn't mind really, a bit on each.*

As discussed earlier, his dad has paid for private physiotherapy, and Tom envisaged some balance between these two needs, one being future focused and one bringing immediate benefits. Carl (18) was more tentative in what he considered to be worthy of funding:

*Carl: Well not for silly things, but I don't know what you'd class as silly things, I'm not a charity person so I don't know what they use money for.*

I gave a few examples of how funds might be spent and of what other boys had suggested:

*Carl: Well it should be done mainly on research and a bit on equipment 'cos fixing it is a bit more of a priority, is a priority as well as the equipment, but they need to put more money into like research and then some into equipment.*

As noted, Carl's family and particularly his dad are committed to raising funds towards a cure for DMD, and whilst Carl did not appear to regard himself as part of that endeavour, his assertion that research is a priority is perhaps shaped by his family's dedication to funding medical research. Jon (16), although 2 years younger than Carl, has less mobility, hence his earlier comment that he would like to be able to scratch his head, and his related comment below about moving his arms. He values equipment, which is of vital importance to his independence, and he sees there being a need for funding both on equipment and on medical research when I ask him how funds should be allocated:

*Jon: Could be like equipment and wheelchairs and things that we need... like [and] could help with research or something.*

*S: What sort of research would you... be interested in the funds going towards?*

*Jon: Er ... like something to help me move my arms.*

*S: To help you move your arms.*

*Jon: Yeah, I know I've got my arm raiser things but like, without them.*

Adam (16) gave careful consideration to the issue of fundraising and research budgets, and as explained in Chapter Three, he is actively involved in pushing for more funding and in promoting the rights of those with DMD:

*S: How would you like to see the government improving the situation for people with disabilities?*

*Adam: I'd like to see them funding more to sort of help find treatments for quite an obvious disability with lots of people with it in this country.*

*S: Yeah erm, if they could be persuaded to give more money, which is unlikely, what would you, how would you like to see it spent?*

*Adam: Into research for cures and treatments, for Duchenne.*

*S: Yeah... erm what about other things like equipment... or would you prefer it just to go on drugs and treatments?*

*Adam: I think you need some sort of balance because obviously the treatments are going to be for the boys in the future but the boys now need care and stuff so some of it needs to be put to them for care and equipment and stuff.*

*S: Moving back to thinking about lobbying, could you tell me why that's something that you value?*

*Adam: Because it can raise awareness of DMD and help to raise funds for sufferers in the future.*

Adam's personal interest and investment in lobbying and fund-raising for future 'sufferers' suggests his low expectations for more immediate medical research that will assist him.

Beyond the hope of finding a cure the participants used their own experiences of need and what they most valued in assisting them as ideas for how funds should be allocated. The participants' modest expectations from funding may reflect how few options are open to them, or how few they are aware of, and it may also reflect the way in which charitable funding is an embedded norm in their lives. This was illustrated by Ollie's (14) listing out of some equipment he was provided with through funding, including his bath and a sleeping system, which turns him during the night to avoid stiffness; this saves his parents from having to wake multiple times to manually turn Ollie. I asked him how the money had been raised:

*Ollie: Well we just did charities really, just did it with a number of charities... they helped us along, get towards it... it was actually through, there was through, a cycle ride around XX [a park] and they gave us the money... so yeah, we've earned a lot of money doing that.*

Whilst some parents focus their hopes and fundraising activities towards finding a cure and treatments for DMD, young people with DMD/MD may have a different focus that is more attuned to addressing their life now. Several participants used the word 'balance' to describe how they thought funds should be allocated between research and equipment, and hopes for an imminent cure or effective treatments as a result of fundraising were not apparent from their comments. From the participants' accounts, fundraising is an issue they are aware of and support, but they also focus their energies on getting on with and investing in their lives.

## The approach to decision-making

Although the participants appear to have limited expectations that medical research can directly help them, this did not stop them from expressing their hopes for future research. In some instances as we explored how they might respond to a speculative research situation, the participants could draw on their real-life experiences, whilst also imaginatively engaging with the theme. Their ideas and comments help illustrate how the participants view and experience agency and autonomy in their lives, revealing what they regard as important and how they think children and young people with DMD/MD could approach making decisions. The discussion on decision-making begins with the issue of autonomy of choice.

### Autonomy of choice

It was introduced in Chapter Two that for those with DMD, despite their physical dependencies, autonomy can be experienced in decision-making (Dreyer et al. 2010), and that this decision-making is often approached from within the context of a supportive relationship. Hence, relational autonomy provides a way to understand how the burden of decisions and choices can be shared in the lives of some disabled children and young people. The various ways in which the participants think about decision-making and how it is or can be approached were explored as I discussed medical research decisions with my participants.

I used the scenarios I devised (see Appendix C) to explore the participants' thoughts on personal choice and how independence, agency and autonomy are experienced and thought about in their lives. With Adam I discussed a scenario where the parents are making a decision to involve their son in some medical research without giving their child a say. Adam (16) felt that this infringed the child's right to independent choice:

*S: If you heard of a family where the parents are kind of 'tough' [the child has to do what the parents say]?*

*Adam: It would be a bit harsh on the child... they should be allowed to make their own decisions if they want to or not.*



He observed that where a child isn't fully consulted about participating in some medical research, this level of disempowerment could be detrimental:

*S: Do you think it's important for the young man to make up his own mind about taking part?*

*Adam: Yeah definitely because if they don't want to do it then they'll be more worried about it and more sort of scared about it if they haven't been able to make up their own mind sort of thing, they might not have control over the situation.*

Adam's comments identify how not being fully involved in decisions may cause fear and a loss of control over the situation, indicating how anxiety is generated when young people are not appropriately consulted. Using the same scenario Jon's (16) opinion was also that the decision should be guided by the young person, in an age appropriate way:

*Jon: Probably let the boy make some decision about whether or not to do it.*

*S: Right, why would you say that that was the case?*

*Jon: 'Cos it's up to him not his parents,*

*S: Yeah, so it's up to him, erm and do you think that, thinking about different ages ... you know, you've got your younger brother [Tim]... do you see the balance changing as boys get older?*

*Jon: Yeah, probably if they're a bit younger the parents might have to make the decision, but if they're a bit older they might want to make their own decision about it.*

Tom (17) felt that whatever the child's age they should remain the central figure in how decisions were made:

*S: Do you think it's important for the [imaginary] boy to be able to make up his own mind about taking part?*

*Tom: Yeah,*

*S: Uhuh, tell me a bit more about that, why you feel that way.*

*Tom: Well because it's them that it's happening to.*

*S: Them that it's happening to yeah... I think that's a good comment.*

*Tom: I just think that, yeah.*

*S: What about if it was a much younger boy, if he was 12 would you still feel that?*

*Tom: Well yeah, I don't think it matters what age they are. 2<sup>nd</sup> interview*

Rick (12) gave consideration to a similar discussion where the parents were not fully including their child in a decision about participating in medical research:

*S: What if the [imaginary] parents were being really pushy, do you think that would be wrong?*

*Rick: Yeah 'cos it's up to the child to make up their own mind whatever they want to do... it's like your freedom about what you want to do, I don't think it would be very good if he wasn't allowed to make his own decision. 2<sup>nd</sup> interview by telephone*

Rick believed the parents should attend to their child's wishes in decision-making, indicating that this was to do with the child's 'freedom' and, by extension, their right to independent choice and thought. This does not suggest to me that the participants wanted to make decisions in isolation from the trusted adults in their life, but to acknowledge that they want to be seen as central actors in choices. Indeed, several participants stated that they would want to make decisions that were right for them, but to make these decisions in consultation with their parents. Tom (17) describes this process, in response to a scenario where an imaginary boy is interested in getting involved in some medical research but is a bit unsure if he should or not:

*Tom: Well I think they need to be told... all the information... The parents and the lad to make a decision, they've got to make a decision between them.*

Tom regards having all the information as being a vital part of the process, with the boy working together with his parents to reach a well-informed decision. There does not appear to be a conflict, for Tom, between his earlier comments that the imaginary boy should make up his own mind, with his view here on the merits of parental input. The two strands work together, in Tom's account, and are complimentary to each other.

Emily (21) saw the potential for there to be problems, as we discussed a scenario where the parents were being pushy with their son about his participation in medical research:

*S: Do you think it's important he [the imaginary boy] makes up his own mind?*

*Emily: Yeah, I can understand though, if his mum, if my mum was even now at my age if my mum was really really pushing me to do something it would make me think I need to do it and it would make me feel kind of guilty in a way if I didn't do it.*

In the above response, Emily extended the discussion by imagining herself in the scenario, where her closeness to her mum and the emotional power (Barron 2001) in the relationship could give Emily's decision a bias motivated by wanting to please her mum; something she also touched on in the previous chapter. This was an idea rather than an assertion, but it illustrates how decisions are influenced by a range of factors including familial relationships. For Emily, the relational aspects of making a decision are potentially bounded by feelings of debt to her parents or 'guilt', thereby impinging upon autonomous decisions. By contrast, Tom (17) views parental support as a practical and positive benefit that, in his explanation, maintains clear boundaries of selfhood, this clear demarcation does not see making a decision that is right for the boy as being in conflict with having parental input. Emily and Tom display the complexities of informed decision-making, where family setting and the dynamics of relationships impact how and why some decisions are made.

Adam (16) was clear what his attitude would be if he had a son with DMD who was deciding whether to take part in some medical research:

*S: As the parent would you want to make the final decision?*

*Adam: Erm no, I would definitely leave the final decision to the child, that's up to them, if they don't do it then other people would, but obviously if they want to do it then they can definitely do it.*

Rick (12) also clearly expressed how those with DMD should make decisions:

*S: If you had a son with Duchenne how would you help him make a decision about taking part in medical research?*

*Rick: Well I'd say if you don't want to do it, don't and if you do, go for it...let him choose what he wanted to do. 2<sup>nd</sup> interview (by telephone)*

Ollie (14) had some advice for an imaginary friend, who is unsure about participating in medical research, even though the parents are keen for the boy to get involved:

*S: Thinking about this pretend friend, what do you think is the most important things he should think about as he makes up his mind?*

*Ollie: Confess to his mum... and dad of course, more reasons, he can just be more confident and make [up] his own mind.*

Ollie saw autonomous decision-making as being a part of maturing, when I asked him if he thought it was important the imaginary friend makes up his own mind he responded; 'yeah, with a young adult or teenager, like me'. At his second interview he made a similar comment about the value he places on making his own mind up when I enquired:

*S: What's helped you to... become the young adult you are?*

*O: Well just be myself really, make my mind up myself, I make my mind up about a lot of things.*

When I asked Ollie to imagine what his motivation would be if he took part in a trial, he had some thoughts on taking part that, again, were related to proving his skills as a young adult who can make mature decisions:

*S: So if you did decide to do the [imaginary] trial what would be your motivation for doing it, what would be your,*

*Ollie: To show other Duchenne boys that it's OK to have it done and it would be great for them to let them have it done, to check what it does.*

*S: That's a good answer, yeah, nobody's said that before, what about the time involved and the hassle, how would you feel about that?*

*Ollie: Yeah it would be a long while but at least it's worth it, it would help me prove my skills as a young adult.*

Ollie regards participation as being something that could prove his growing capacity to make decisions and to cooperate in research. He does not see the speculative

research as benefitting him physically but rather of being socially helpful, and this is reflective of his earlier summation that research will not directly help his condition. Ollie has developed some useful strategies, particularly at school, for demonstrating his maturing abilities and he considers decisional capabilities as marking his move into young adulthood. In his comments Ollie implies that openness between a young person and their parents is helpful, suggesting the imaginary friend 'confesses' to his parents if he does not want to commit to medical research. His use of the word 'confess' may, however, highlight the dilemma of making decisions in conjunction with parents. A desire to include parents in decisions may require negotiation between the sometimes divergent attitudes of child and adult, where the child needs to speak up about their own reasons for or against taking part. The problems Emily noted, of a sense of debt or guilt, may also occasionally problematise the decision-making process. Candidates for medical research may find their personal preferences more difficult to express where these differ from those of their parents. Decision-making is a process through which disabled adolescents can attain some independence, but concerns about conflicts with the trusted adults in their lives can induce anxiety (Skär 2003). This sense of anxiety could, in some instances, impede individual decision-making skills and the freedom to make personally appropriate choices.

### Participation and motivation

Some of the participants have taken part in some medical research or are seriously considering it, and I was interested to know more about their experiences, Jay (14) has taken part in some research that entailed having four magnetic resonance imaging (MRI) scans. Initially he was approached at his clinic appointment about participating and then someone came out to his house to explain more about the research:

*S: So you sat there and listened to her, and if you'd suddenly thought 'no, I don't want to be part of that' would you have felt you could say no?*

*Jay: Yeah, well she said... 'if you want to say no [then] say no'.*

And,

*S: So when you were thinking about taking part in that MRI [scan] did it take you a while to make up your mind and think it through?*

*Jay: Not really, I knew that it would help, like, researching the condition.*

The scans were quite arduous and a commitment of time and courage, with Jay finding one of the nurses a bit 'bossy', he appreciated having his grandma there, as well as his mum, to help him through the final two scans. However, he was open to taking part in some new research if this was offered to him:

*S: If you were approached to take part in some new research would you think about it?*

*Jay: Yeah I'd discuss it with my parents, and then, get information on it, and... go from there.*

*S: Some people say that taking part helps them to feel that they're doing something, what would you say?*

*Jay: Yeah, you feel like, included in what's happening, into the research into the condition, yeah.*

Jay is motivated by being included in the process of building knowledge on his condition, and I asked him to further explain how he would make his mind up about participating in another research procedure:

*S: If there was some new medical research that they asked you to take part in, not MRI, something different, would it be good to have your mum and dad's help in making your mind up?*

*Jay: Yeah ... I'd like to have information on it, like I've had for the MRI and this [the information sheets for my research].*

*S: And if you had any questions, how would you go about getting those questions answered?*

*Jay: Probably just ask my mam and dad and then sort it out from that, yes.*

*S: In maybe a couple of years time, if there was something, would you perhaps like to make more of a decision on your own or?*

*Jay: Probably still want to make it all together with me mam and dad.*

Rick (12) is actively thinking of taking part in a clinical trial he has heard about and we talked about the decision-making process:

*Rick: Yeah, well I'm going to take part in this one, I'm thinking about taking part.*

*S: You're thinking about it, so how did you hear about it?*

*Rick: Don't know really, my mum, people kind of send her kind of stuff about it... people in the hospital and stuff.*

*S: Yeah, so your mum got the letter, read it and then did she,*

*Rick: She explained to me, it was on the internet and she printed a copy off.*

And,

*S: So what questions as you're kind of going through thinking this through and making that final decision, what things are coming up into your mind?*

*Rick: Well I don't know I just think it might help other people and maybe myself and stuff, to, yeah to try and help.*

Rick thinks his participation might be a benefit to others and to him, and his mum was involved in informing and helping him learn more about the research. Ed (18) has, in a less overt way, taken part in some medical research, as samples of his blood, along with those from his brother who also has DMD, have been taken and sent to London:

*S: Right, so that's sort of, that's a tiny bit of medical research that you've been a part of hasn't it? And your blood's gone off [to London],*

*Ed: Yeah.*

*S: Erm, how would you feel, about, say a tiny bit of that blood being sent off to... I don't know, say Switzerland or to some clinic there for them to test... is that something you're happy about?*

*Ed: Not really bothered... You have to do what you have to do.*

*S: So you wouldn't mind that blood going off to various places to be tested?*

*Ed: As long as it makes people better and that, I'm not really bothered.*

Ed apparently has no qualms with his blood being used to improve understandings about DMD, and despite his misgivings about hospital appointments, this was a procedure he was happy to assist with. His last comment, that he does not mind the blood sample being taken 'as long as it makes people better' perhaps illustrates Ed's misunderstandings about how medical research can involve many phases, prior to reaching the point of treatments. Initial trials will test a drug's safety on a small group of people before later phases include larger groups, with the final phase used to learn more about the side-effects and long-term risks and benefits of a newly licensed drug (Cancer Research UK 2013). However, Ed did not give the impression he was concerned what happened to the sample and it was his mother, who was present during the interview, who mentioned it. For Ed, having the blood taken may just be one more procedure which does not bother him or raise ethical issues, and is part of being at the hospital and doing 'what you have to do'.

Emily (21) is thinking about taking part in a clinical trial that she has heard about, and her motivation is partly as a result of so little being known about her condition and how her future health will be affected:

*Emily: If there's not many people and there's not much research on it I don't actually know how my future's going to be... I just want to know, I don't like being clueless about my future.*

She describes the process she is going through:

*Emily: I don't know if I'm going to do it but it was about doing this trial thing, it's a tablet, I haven't read that much about it, it was just on the XX [charity's] website actually, 'cos they sent an email to my mum and then my mum forwarded it to me and there was a link on it... and it didn't say too much but it said it was a tablet... and I am interested in doing it I'm just a bit wary of it, just a bit scared of it, I'd want to speak to someone first.*

Emily is frustrated at the lack of research on her muscular dystrophy and this is causing her to seriously consider participation; as she explains when I ask her if she is on steroids for her condition:

*Emily: No I'm not on anything yet, there's nothing for it at the moment, that's why I really want to do this thing... I want them to hurry up and find something*



*for it... because it's so rare there's not going to be so much research on it... it makes me think they're never going to find something... obviously they're going to put more money into something where it's more common ... And because there's not many other people with it as well it makes it harder for them to find people as well.*

Her motivation is quickened by the fact that few people have her condition and, therefore, few people will enrol on the trial, and she cannot rely on others taking up the opportunity to get involved.

Amongst more speculative discussions there was evidence of motivation to take part in medical research even if it brought no direct physical benefits. Jon (16) commented in Chapter Three that he would take part in some medical research to help others with DMD to become more independent. He would also approach participation in medical research as helping '*to give them [doctors] more ideas what works and what doesn't*'; he understands that research develops knowledge of a drug or treatment through eliminating ineffective options. Adam (16) was also prepared to consider participation without personal benefit, proposing a realistic attitude that was future orientated:

*S: Do you think it's good to be involved in research even if it brings no physical health benefits to you?*

*Adam: Yeah, because it will benefit people in the future, and help them and then, or if the experiment doesn't work like they want it to they'll be able to rule that out and move onto something else quicker, yeah.*

Adam, like Jon, is aware that medical research can be a process of elimination, with findings that may rule a drug or treatment as unsuitable for further development, but this does not deter him, although he would want a good understanding of the aims of the research. He described what would inform and motivate him in deciding to participate, he felt that it would '*help if the staff... talk to me about it and explain to me how it might help and what the research might do*' (second interview by telephone). This suggests that Adam, in principal, is prepared to participate in medical research that will help future boys with DMD; and helping others, despite little hope of personal gain was also a reason Tom (17) might take part in medical research:

*S: So do you think it's good to be involved in research even if it brings no benefits to you?*

*Tom: Hmm... yeah if it helps other people,*

*S: If it helps other people?*

*Tom: Yeah... possibly.*

*S: And why would helping other people be important to you?*

*Tom: Hmm... I don't know really... obviously the fact that it helped you, overall... but if it helps, it just helps in any way. 2nd interview*

However, in this instance we were talking speculatively about motivations for taking part in medical research; when I discussed with Tom and the other participants how they might respond if they found that they had been given a placebo, this produced a more tentative attitude. I explained the term 'placebo' to the participants as being a harmless, pretend drug or sugar pill which has no clinical effect. Placebo is used in research to compare whether trial results are due to real clinical change or if they may also be due to the 'placebo effect' of the patient perceiving they have improved, despite only taking a false drug.

Several participants commented that their motivation for taking part in medical research would be to help others; but the issue of being on the placebo arm of a trial or of having a biopsy produced a more cautious response. In the previous quote from Tom (17), he saw participation in medical research as being of benefit to others, but in his next comments on taking part in some imaginary research where he may or may not take the real drug he was less sure:

*S: Would it bother you that you wouldn't know if you were getting the real drug or the placebo?*

*Tom: Yeah I think so, 'cos you're wasting your time a bit if you get the placebo... and there was something about taking some muscle or something [biopsy]... I wouldn't really want that done ... I don't think I'd want to have that done just, just for no reason.*

*S: Yes, if you had the placebo and then they [did a biopsy]... yeah.*

*Tom: Yeah, you'd think 'what's the point?'*

*S: What's the point yeah... erm some people would say, or have said, it's the taking part that counts, whether you get the placebo or the,*

*Tom: Yeah I suppose. 2nd interview*

This response from Tom contrasts with his previous comment that 'if it helps, it just helps in any way' and may represent a distinction between generalised discussions on medical research and more personalised discussions, which can produce differently balanced considerations. Emily (21) commented on how she would feel if she found out she had been taking a placebo in a trial, particularly if injections had been used to administer the drug:

*S: So how would you feel, say you'd committed to it and you found out 'oh I was on the placebo'?*

*Emily: I would be a bit gutted [upset] 'cos I would be like I'd wasted my time... if it was just a tablet then I wouldn't mind but if it was an injection... that would put me off because I hate injections so if it was any kind of injection that would really,*

*S: That would be a big issue?*

*Emily: Yeah 'cos I really, really have a phobia of injections and stuff.*

Emily mirrors Tom's attitude in her assertion that the placebo is a waste of time, this perhaps reflects the quite unsophisticated understandings some of the participants have on the testing of new drugs. Ed (18) appeared unsure about the role of placebo in trials, not so much seeing it as a waste of time but as a wasted procedure. When I asked him what his reaction would be if he found out he had been taking a placebo he was dismissive of using a 'pretend' drug:

*Ed: I don't know why they would want to try with a pretend drug that's not doing anything.*

Ed and his family did not report following news and updates on medical research, and most of their information comes on a need to know basis through the hospital. A lack of knowledge about medical research may contribute to Ed's dismissal of the idea of using a placebo instead of the real drug, although Carl (18), whose family are closely involved in a DMD charity, also expressed surprise at the use of placebo in trials:

*Carl: It's a bit cruel making a bunch of kids have injections that's not going to do anything and people know it... I think that's a bit cruel... But if it was just the real one [drug] and not the placebo one then,*

*S: The real one, right.*

Carl's mum and dad who were present during the interview demonstrated their knowledge on drugs trials as they interjected to explain to Carl that the placebo is considered a necessary control in trials:

*Carl: Yeah but I still think that's a bit mean though,*

*S: Yeah if you'd committed to 24 weeks and were really hoping you were getting this new drug and then you found out oh [you had been getting the placebo].*

*Carl: You'd be a bit angry wouldn't you?*

Carl was uncertain about the purpose of placebo in trials, and he felt it was 'cruel' and 'mean' to put children through many weeks of injections with no chance of benefit, although he saw the worth of participating if the real drug was administered.

By contrast, Adam (16) commented that 'having 2 biopsies wouldn't be that nice' but he understood the role of placebo in trials, and how medical research is a form of experimentation:

*S: Would it bother you that you might be getting the placebo?*

*Adam: Erm I'd probably wonder about it at the time, but then when the trial had been completed I'd probably want to be told just so I knew if there were like any changes, so if I knew there were any changes then they could be because of that.*

*S: Would you be annoyed if you'd gone through the whole thing and discovered you had got the placebo?*

*Adam: I probably would be slightly but then on the other hand you've got to have some people who get the placebo otherwise you can't have the experiment.*

Rick (12), although being one of the younger participants also had an understanding of the experimental nature of placebo:

*S: If it was you taking part in the [imaginary] trial, would it bother you that you wouldn't know if you were getting the real drug or the placebo?*

*Rick: Erm, not really sure, I wouldn't really mind that much 'cos it's just... to experiment with what would happen.*

*S: Some people get confused and they think they're going to get better if they take the drug.*

*Rick: Yeah, but you might not as well.*

Rick works with his mum, whose career is in the field of science, to build his knowledge on DMD, and he understands that being in a trial might not make him better.

Emily (21) has already commented on her fear of needles, and her probable decision to take part in a real clinical trial on her condition is encouraged by the fact that the test drug would be administered as a tablet. However, she is conscious of the risks of participation and is concerned that she may be the next person after a mouse to be testing the drug:

*Emily: I read something about them testing it on a mouse,*

*S: Yeah, they do test them on mice.*

*Emily: So if I was the next person from a mouse I think I'd be a bit scared [laughs].*

Emily's awareness of the risks in trialling a new drug were not shared by Ed (18), who took the view that it was worth trying:

*S: What would you think about risk? That it's a drug that's just being tested out?*

*Ed: Still do it... It might be getting rid of your disease.*

And,

*S: So, thinking it through... say I was here today, I wasn't here to do this [interview him for my research], I'd come and said 'Ed there's this drug I want you to test out' erm, which I'm not obviously, what would help you as you really thought it through and made up your mind, [and] you were weighing up the risks?*

*Ed: There'd be nothing to think about I'd just get on and do it.*

*S: So I'm getting from you mainly that your attitude is 'go for it'?*

*Ed: I think everyone should.*

Ed was not inclined to weigh the risks, he thinks that 'everyone' should get involved, and the possibility he might 'get rid of his disease' was enough motivation for him. Ed also thought that being rid of the disease would make life easier for his parents, and this was obviously something that Ed had discussed with them, this became apparent as we discussed a scenario:

*S: What do you think your mum would reckon, having to drive you to the hospital every week for 24 weeks?*

*Ed: She would do it, they already said they would, me mam and dad... And they'd want us to get better too 'cos it makes it easier for them an all.*

Ed sees how his older brother with DMD is affected by the condition and Ed knows he too will go into a wheelchair at some point, albeit later than other boys with DMD as Ed and his brother have a less severe form of the condition. Ed's attitude reflects the relational aspects of living with a deteriorating condition, and how decision-making can emerge from considerations of what is best not just for the individual, but for the family too. Ed does not spend time thinking about the risks of research, but he sees it as a solution both to his physical situation and to his family's ongoing commitment in caring for him and his brother.

Rick (12) was aware, but also philosophical, about the risks of trying a new drug if he does go ahead and participate in the clinical trial he mentioned earlier. He has gained experience of some of the drawbacks of taking steroids for his condition; *'there are a few side-effects about the drugs [steroids] that I'm taking at the moment, like it kind of makes you shorter, 'cos I'm quite small'*, and one brand of steroid he took for a while made him a bit 'grumpy', and caused him to gain weight. This experience may be helping him to take a practical view of any risks with the new drug:

*S: So thinking about taking this possible new drug would you be concerned about risks with that, would you want to investigate?*

*Rick: Probably, I'm not really that concerned about too many.*

*S: Right, that's interesting, if you had any questions about this drug that you're thinking of taking how would you get those questions answered?*

*Rick: Probably if I looked deeper into it, contact the person who sent us that [information] first to get more, to get further information on it.*

In a second interview [by telephone] with Rick, which took place a few weeks later, he told me that he has decided to participate after talking with his doctors at hospital about it, and he described the process:

*Rick: One of the doctors was showing me how it will work, like a diagram of what the drug does and stuff and how it will help you.*

*S: And did you have any questions or did your mum have questions?*

*Rick: I had 2 I think... would they make me like really drowsy and I think and the other one was how it kind of works a bit more... just broadening my knowledge a bit.*

*S: Broadening your knowledge, yeah that's a good way to put it, and was it kind of after you'd had that discussion you sort of moved toward making your mind up?*

*Rick: Yeah.*

*S: Or had you already made your mind up?*

*Rick: Er well yeah, I think that pushed me towards it a little bit more.*

*S: Yeah, and was it helpful the way they discussed it with you, was it sort of in a way you could follow?*

*Rick: Yeah they're really good at helping me understand and everything.*

It was seen in Chapter Three that Rick is confident in addressing doctors, and he has 'broadened his knowledge' on the proposed research drug through asking direct questions that have guided him towards his decision. I briefly asked him what he might do if, when he is taking part, he experienced some side-effects which were quite unpleasant:

*Rick: Well I'd probably talk to my mum about it and talk to like erm there's people that you can ask questions like I think on this sheet where it's*

*explaining about the trials, it's got the numbers of three doctors who are involved with it so I'd probably talk to my mum and sort something out and maybe pull out if it carried on.*

Rick's first line of action would be to consult with his mum, and then to contact the doctors, with the implication being that he would draw his own conclusions, with guidance, about whether to continue in the trial. Whilst Rick is not overly concerned about side-effects and possible risks, Emily (21) has already expressed her concerns over being the first person after a mouse that the drug is tested on. This concern is apparent in her approach to making a decision about participation, *'it's just scary... I think you have to be quite brave to do it'*. She might find out more about any side effects by speaking with the doctors involved to assess the risks:

*Emily: I think also if I spoke to a doctor about it and they told me all the, like what could happen and if you get any bad side effects from it, and if it seemed quite safe and my condition couldn't get any worse from it and it would just be small side effects that could be for anything and it wasn't that high of a risk then I probably would do it.*

Emily is closer to making her mind up, but her worries and the need to be 'brave' are valid causes for her hesitance; like Rick, she would seek guidance from doctors and the information they provide would inform her decision. Adam (16) mentioned a specific drawback of participating in medical research, reflecting his current commitment to school work:

*Adam: I'd probably be weighing up the pros and cons of the thing and for instance, now I wouldn't do it because I'd miss too much school for instance... probably if I wasn't in that situation obviously I'd probably... so yeah it's weighing up the advantages against the sort of things that...*

As Adam comments, the process of thinking about participation involves weighing up the various advantages and disadvantages, and for each participant these are going to be influenced by the values and practices embedded in their personal and family life.

There was a general consensus that taking part in medical research is a positive contribution to helping others. The issue of a placebo, however, raised differing levels of comprehension and concern amongst the participants, some of whom were



unfamiliar with the nature of clinical research (Young et al. 2010). Some of the participants were unclear on the point of having a placebo and on the process of experimentation, and this was not age defined but seemed related to their knowledge acquisition. Carl (18) did not understand the role of placebo in trials, despite his parent's close involvement in a DMD charity and their evident knowledge of trial protocol, which was demonstrated during the interview when they intervened to explain to Carl why placebo is used. This example suggests that information gathering within families is influential, but it can also be contingent upon the child or young person's needs, their personal level of interest, and what parents do and do not choose to discuss with their child. Rick (12) and Emily (21) have been considering participation in clinical trials, and they each mentioned questions they needed to address as part of making their mind up. Rick was, in part, drawing on his experience of taking steroids in how he thought about any risks with the experimental drug, whilst Emily has no experience of taking medication for her condition and is more tentative about testing a drug. There was an overall consensus that medical research is a valid process, but there were more individualised responses regarding risks, biopsies and placebos. Concern over the use of placebo is not just restricted to the participants; it is also expressed by some who are promoting more research into DMD:

For pediatric patients with progressively fatal rare diseases, who have no other alternatives for care, the notion of enrolling in randomized trials with the likelihood of receiving placebo creates challenging ethical and practical dilemmas for patients, parents, and study sponsors (i.e. restricting access to hope). (Franson & Peay 2013, p11)

And,

Ethical dilemmas imposed by placebo-controlled trials... create an imperative to consider new approaches... for patients who otherwise must face physical decline without hope for reversal. (Franson & Peay 2013, p13)

Franson and Peay (2013) propose that risks and benefits have a differential weight when measured against the relentlessness of DMD progression and they call for more participation from those directly affected by DMD in deciding what risk/benefit ratio they may be prepared to tolerate. Such discussions merit their own considerations that are beyond the scope of my research, however, some of the participants' comments on placebos correlate with the concerns of others about the

time 'wasted' in administering a fake drug to an already burdened and seriously disabled young patient.

Although the participants have a chronic progressive condition, they assert their individuality and personal interests, and to some extent, they deal with their limitations as a normal part of life (Berntsson et al. 2007). They do not appear to spend more time than is necessary researching DMD/MD or 'pondering it' and their variable knowledge indicates how life in the 'now' is where much of their energies are spent. There are a range of factors and influences working together to form the participants' opinions on medical research; Tom (17), as a sports fan, values physiotherapy and keeping his joints mobile. Adam (16) is studious and thoughtful, but also sociable, and his knowledge on medical research is not gathered through hours of research but is achieved in tandem with his mum and his developing interest in lobbying. Whilst these young people are joined by their condition, they are also singular in their capacities, interests and the focus of their lives. Each participant displayed some measure of conviction that if choosing to take part in medical research, their own opinion is the ultimate arbiter. However, they also had different ideas on how to reach that point, with some having a closer relationship with their parents where decisions are concerned, whilst others drew more upon their own innate sense of what was right for them. Nurturing an interest and knowledge on their condition and related topics can, for some participants, be a way of acting upon their life as agents of their own development (Boyden 2003). Others, such as Ed, appear to have less breadth of knowledge about DMD, and this may be an active coping strategy for minimising anxiety; it is also possible that Ed's lack of in-depth knowledge is caused by his isolation and his reluctance to engage in discussion with doctors when at the hospital. Most of the participants were prepared to discuss medical research, and using the scenarios created valuable discussion and comments. However, it was also clear that medical research is not a central concern for the participants, and although living with DMD/MD is challenging, it is not all that life is (Carnevale, 2011).

Recognition of disabled children and young people's growing capacity is central to ensuring they are given sufficient information on their condition and the opportunity to be involved in decisions; 'children need opportunities to learn how to participate in

decision-making over a period of time' (Coyne & Harder 2011, p316). Accounts on quality of life given by individuals with DMD/MD are, in some areas of their life, higher than expected (Bray et al. 2010; Vuillerot et al. 2010; Grootenhuis et al. 2007). This suggests that the severity of a disability does not always significantly alter life satisfaction (Grootenhuis et al. 2007), despite external assessments. Such assessments may cause some parents to look to medical research as the best and only hope for their severely disabled child; 'parents are vulnerable both to misunderstanding and overestimating the potential of medical research to benefit their children' (Schaffer et al. 2009).

Drawing upon the accounts given by my participants relating to their lives, their health status, and how they want to be involved in decisions, has demonstrated their abilities for cogent discussion. In my interviews I contextualised my questions for the participants, and they responded to this approach. Alderson (1992) observes that children's ability to consent can draw on their contextualised understandings of risk and benefit which 'concern actual or potential experiences, and are understood through the imagination, a potent skill in children' (Alderson 1992, p121). This implies that when children and young people are making decisions about medical research, they can draw on their own health related experiences, and also, as with my scenarios, they can apply their imaginative skills and their experiences to weighing-up their options. Paying attention to these abilities can help to ensure that disabled children and young people are listened to and accepted as socially and morally involved individuals when decisions are being made.

## **Conclusion**

When youthfulness and disability are combined, assumptions made by adults about children's capacities can detract from children's opportunities for self-determination (Lind et al. 2003). Negative attitudes can be prejudicial to the development of selfhood and agency and impact how children and young people are positioned in policy formations that affect them:

Interpretations of children as weak and incompetent tend to justify... policies and interventions that treat children as the objects of adult decisions, rather

than social subjects with valid insights and perspectives. (Boyden 2003, p22)

Treating disabled children and young people as socially engaged subjects assists their move to independence and greater involvement in decision-making (Coyne & Harder 2011), positioning them not just as objects of care, but as agents who are actively involved in determining their life. My research looks at the approach to medical research decision-making, but the background to these decisions, such as the participants' sociocultural setting and the entire impetus of their care and treatment are implicated in the decision-making process.

It is apparent that tensions and complexity are sometimes present in the lives of young people with a chronic progressive condition, they need parental support in order to flourish, and their capacity for independence may be differently configured than that of their able-bodied peers. Oppositional behaviour (Vuillerot et al. 2010) such as ignoring or refusing to take parental advice may be less apparent, and their sense of loyalty to their parents can impact how decisions are made. Disabled children and young people may feel that their parents are one of several stakeholders in their healthcare, and that parental dedication gives them a right to partake in their child's decision-making, which indeed it may. The benefits of a close relationship with the meaningful adults in their lives can provide support when disabled children and young people are thinking about decisions, and these will rarely be made without some help from family and doctors (Coyne & Harder 2011).

What has been evident from their comments is that the participants want to be involved in meaningful ways in managing their lives, making decisions about their healthcare and taking some responsibility for themselves (Berntsson et al. 2007). This was a point that came through in all of the participants' narratives; there was a commonality of feeling that certain boundaries of selfhood should not be overstepped by others. However, beyond this common attitude amongst the participants about their primary role in decision-making, the shading of opinions was more personalised. This reflects the individualised process of decision-making, which draws on a broad repository of views, family dynamics and experiences. As Young et al. (2003) discuss, there may be critical times when parents of an ill or

disabled child must take an executive role in decisions, and it is possible that some parents find it hard to step back from this role. Over time, with experience, and during more stable periods of health, some families may develop a partnership model of working (Young et al. 2003). In this situation, more open and equal communication can take place between parent and child, positioning the child as a central figure in discussions and decisions. Samson et al. (2009) explore this transition in the lives of parents of a child with DMD, where there is a shift towards assisting their growing child in an atmosphere of mutual respect, recognising that their child is becoming a young adult with a sense of their own identity. Boyden (2003) speaks of children's resilience, proposing that competence contributes to the wellbeing of children and that they should, wherever possible, have an active role in decisions and processes that affect them. Nurturing this capacity for resilience in disabled children and young people can be a positive step, enabling them to mature as engaged agents in decisions affecting them.

Widely held assumptions and the normative views of society (Boyden 2003) and of some in the medical profession can perpetuate misleading notions of disabled children and young people as inherently vulnerable. In the next chapter, attention will be paid to the framework of medical knowledge and the discourse that emanates from it. A protectionist stance and statements about children's vulnerability (Boyden 2003) are likely to be evident in some healthcare documents and circulated in their discourse as self-evident facts. There may be some dissonance between the capabilities of disabled children and young people such as my participants, and the views disseminated by those in a position to influence and produce policy regarding them. These official policy documents will be discussed in the next chapter using discourse analysis to problematise their claims and assertions.

## **Chapter Five**

# **Constructions of children and young people in healthcare discourse**

The previous chapters have considered the experiences and views of children and young people who are living with DMD/MD, and who are in various ways developing independence and agency. Drawing on concepts from the sociology of childhood and from disability studies, consideration has been given to their thoughts as socially engaged and competent healthcare actors. However, their competence and ability to participate in their healthcare may not be fully acknowledged within the discourse produced by healthcare institutions. This chapter will, therefore, use discourse analysis to examine and question the ways in which children and young people's participation in their healthcare is constructed and spoken of in a range of documents.

As outlined in Chapter One, the atrocities committed in the name of medical experimentation during the Second World War, abuses involving experimentation with children (Meaux & Bell 2001), and failures in clinical care (NICE 2002), such as at Bristol and Alder Hey, have all contributed to highly protective approaches to children's participation in healthcare and medical research. However, the landscape has changed, and there is now a move towards children and young people being more involved in decision-making on their healthcare and on medical research participation. This discourse analysis will examine how children and young people are constructed in healthcare documents, raising the possibility that these constructions impact their participation in decisions on their health. The analysis will look for the fit between calls for the increased participation of children and young people, alongside how healthcare documents discuss and construct them, and how guidance is given to healthcare professionals. The methodology for this discourse analysis was explained in the final section of Chapter Two, and in this opening section I will contextualise how this discourse analysis applies to children and young people's healthcare.

Discursive practices can involve the maintenance and reproduction of institutions (Shaw 2010) and the language of children's participation expressed by some institutions, may, in some cases, be rhetorical. The rhetoric may not engender real change, reflecting instead a concern with 'goals and plans' (Stenson & Watt 1999, p194), therefore, the power structure of an institution such as the NHS remains intact, whilst statements of intent or 'tokenism' (RCPCH 2003) have been paid to initiatives aimed at children and young people's participation. Institutions might not display their power and influence overtly, they may be an integral part of their presence as sites and producers of statements and practices. Thus, the authority of discourse can uphold, but also render invisible, the 'structures of domination and authority of the specific clinical institution' (Houghton in Crowe 2000, p75); and discourse analysis can challenge these hidden or difficult to identify discourses, troubling and questioning their dominance.

In Department of Health documents, there is evidence of a move towards more involvement of young people in their healthcare. This is partly in response to Article 12 of the *UN Convention on the Rights of the Child*, which states the child's right to express themselves freely. According to a UNICEF report *Promoting Children's Participation in Democratic Decision-Making* (Lansdown 2001), those caring for children should not only listen but also respect and give proper consideration to their views. As Lansdown (2001) observes, the way in which children and young people are involved in their healthcare can be affected by the attitudes of the adults supporting them. This situation has a corollary for young patients, as good healthcare and treatment depend on effective communication, where the child is encouraged to express their own thoughts and preferences:

It is through learning to question, to express views and having their opinions taken seriously, that children will acquire the skills and competence to develop their thinking and to exercise judgement. (Lansdown 2001, p12)

Through examining language in use (McNeish 1999, p193) this discourse analysis will explore how children and young people's participation in their healthcare is situated and presented and what the language used may signify. Texts can use language in subtle but powerful ways to convey meanings (Lupton 1992), and these

meanings can have ongoing effects as they create and uphold certain versions of reality (Lupton 1999). Patterns of knowledge and practice are reproduced in and through discourse, and analysing these patterns and the underlying messages and values they contain can uncover how institutions function (Powers 2007).

This analysis will proceed by discussing how dominant voices (Lupton 1992) speak of children's worth as future adults rather than present actors. Children and young people's participation in their healthcare and constructions of vulnerability will then be discussed, before moving on to explore how they are frequently regarded as vulnerable and in need of protection in medical research. The final section of this chapter will contextualise some of the participants' comments, illustrating how these comments undermine the homogenising and decontextualised tendencies of discourse.

### **Discourse analysis: constructions of childhood, children and young people**

My participants have each demonstrated their ability to make intelligent observations about their healthcare and medical research decisions, and they have proven to be able narrators of their lives. It is also apparent that their abilities and their levels of knowledge are diverse, and that each participant's approach to decision-making emerges from their own specific circumstances. This diversity may not be evident, however, in the healthcare documents that are going to be analysed. Many of the constructions of childhood participation and decision-making that will be examined emanate from a broadly similar perspective. This perspective supports certain approaches and institutions, thereby excluding others (Shaw & Greenhalgh 2008), tending towards generalising children and young people in ways that evade more complex representations of them.

#### Future beings

There is a tendency in some documents to speak of children and young people as future beings, whereby discourse displaces them from their current experiences and concerns. The document *Bridging the Gaps* (RCPCH 2003) is produced by the



Royal College of Paediatrics and Child Health, who have an influential voice and role in advocating for children and young people's participation in their healthcare.

*Bridging the Gaps* (RCPCH 2003), as will be seen later, has some positive contributions to make to promoting children and young people's effective participation. However, the document also displays elements of discursive tropes as it discusses the value of ensuring children and young people's health in terms of future benefits and their economic worth, as this paragraph demonstrates:

It is clearly important that young people are nurtured so that they may become healthy adults and contributors to society. This is increasingly important for sound economic reasons in an ageing society. (RCPCH 2003, p18)

This adult-centred (Alderson 1993) thinking is a recurring theme. It regards children as 'socially developing organisms; not yet fully actualized' (Grover 2004, p91) and is repeated in various iterations across several documents. It situates young people as future agents in society, diminishing the worth of their presence as current actors whose lives have relevance now, with as much validity as their tentative adult selves. This can perpetuate a compelling, if flawed and politically motivated logic, situating health benefits within the blueprint to build the nation's future financial security, rather than simply nurturing young people's health and rights as an end in and of itself. *Bridging the Gaps* (RCPCH 2003) states, probably with some degree of accuracy, that 'problems in adolescence... indicate the likelihood of long term adverse health and social consequences' (2003, p18). Adolescent healthcare is presented not only as a present concern but as a potential problem for future society; by various 'routes... children have been linked in thought and practice to the destiny of the nation and the responsibilities of the state' (Rose in Jenks 2005, p60). Therefore, healthcare may be delivered from an adult perspective, informed by adults' social and economic goals (Mayall 2000), instead of being tailored to the individual patient's needs in the present.

A future, economically focused attitude is replicated in *Getting it Right for Children and Young People* (Kennedy 2010); a review of NHS children's services that was conducted by Professor Sir Ian Kennedy on behalf of the NHS. Kennedy, who also chaired the inquiry into the failings of paediatric cardiac surgery at the Bristol Royal

Infirmery, states:

We fail... if we do not do our best to provide them [children & young people] with the best possible opportunity by way of health and healthcare... to grow into adulthood able to flourish and fulfil themselves... it makes good economic sense to invest in the welfare of children and young people. (Kennedy 2010, p52)

This attitude is also apparent in the *Report of the Children and Young People's Health Outcomes Forum* (DH 2012) which was set up at the request of the Secretary of State to respond to Kennedy's 2010 findings:

The focus on children and young people is not only an emotionally driven one. It is also based on economic reality: children and young people are crucial to the future well-being and prosperity of our nation. (DH 2012, p9)

And,

The focus on... children and young people is so critical because the consequences of poor health affect individuals throughout their lives (DH 2012, p38)

And again;

Children and young people are crucial to the future wellbeing and prosperity of our nation. Improvements in their health and healthcare will result in healthier and more productive adults. (DH 2012, pp38-39)

This repeated assumption, that healthy children become productive, healthy and economically viable adults, does not speak for those young patients whose futures are uncertain. The lives they are leading now matter, and for those with a shortened lifespan, or who are struggling with current deteriorations in health, future benefits may be of less concern than their more immediate quality of life.

This forward-looking discourse also appears in the Department of Health's *National Framework for Children, Young People and Maternity Services: Executive Summary* (DH 2004c). As explained in Chapter One, the National Service Framework [NSF] was established in the wake of the Bristol Inquiry, which investigated the failings of paediatric cardiac services at the Bristol Royal Infirmery. One of the results of the Inquiry was that the importance of children's healthcare participation was recognised (Donnelly & Kilkelly 2011). Professor Aynsley-Green, in the NSF's *Executive Summary* states:

Children and young people are important. They are the living message we send to a time we will not see... they deserve the best care because they are the life-blood of the nation and are vital for our future economic survival and prosperity. (DH 2004c, p4)

Aynsley-Green is a significant figure who strongly advocates for the healthcare of children, although in the above statement he draws children into a discourse of economic 'survival', whilst in the BMJ [British Medical Journal] he describes children as 'our most precious resource' (Aynsley-Green et al. 2000, p231). Such statements echo the 'predominant neo-liberal and rational economic ethos, which permeates most contemporary western cultures' (Crowe 2000, p73), an ethos that situates individuals as contributors to the economic wealth of society. In this discursive context, children are, in part, viewed as the future workforce and as economic actors; 'thereby reframing health benefits in terms of labour' (Shaw & Greenhalgh 2008, p2512). But, as noted, this limits the status of those young people who may never be able to take part in work-related activities and whose future is bounded by health limitations. It also conflicts with the sociology of childhood view that children are not just in a process of maturing towards adulthood but have 'experiential significance' (Balen et al. 2006, p31) in the present. Thinking of children as yet-to-be-adults reflects 'the absence of value and status placed on children as children rather than on what they will become' (Viper 2013, p43). Children are drawn into national projects, and childhood imbued with adult meanings and values linking the 'welfare of human society to the well-being of children...[with] childhood a project of the modern state' (Shanahan 2007, pp417-18).

Various constructions of childhood - as a hope for the future, as a preparatory and vulnerable stage, or as a national project - could each be considered emblematic of the role of discourse and of the 'institutional colonizations of childhood' (Moran-Ellis 2010, p189). The idea that children need to be 'saved from harm and the concerns of the adult world is strongly embedded in (modern) Western ideology and practice' (O'Dell 2008, p385), stemming in part from the Victorian view that children are a 'national resource' (Foley in O'Dell 2008, p385). Lupton, quoted in Willard (2005) asserts that metaphors applied to the medical setting are 'commonly used in ideological struggles around a contested site of meaning' (Willard 2005, p120).

Thus, the statements made in documents can reflect certain perspectives, spin and the interests of groups and/or institutions. In one reading, Aynsely-Green's (DH 2004c) introductory statement that children are 'the lifeblood of the nation' fits comfortably into commonly held beliefs, it sounds reasonable and stirring. But, it could also be decontextualised as a metaphor based as much in cultural or even nationalistic discourse and rhetoric as it is based in healthcare or on the needs of young patients.

### Constructions of participation

*Bridging the Gaps* (RCPCH 2003), as discussed above, does at times use rhetorical language in which children are positioned as future agents rather than current actors. However, it also recognises the tokenistic failings of encouraging young patients' participation in decisions on their healthcare:

More consultation, "involving young people in decision making", is the politically correct thing to do but it is often complete tokenism or, at worst, an excuse for not acting on the information we already have. (RCPCH 2003, p9)

The document suggests this is because consultations with children and young people have not always brought the answers the medical establishment wants to hear or accommodate. Consultations and attempts to improve participation with children and young people are regularly itemised in Department of Health documents, in what Kennedy (2010) describes as a 'never-ending treadmill of policies' (Kennedy 2010, p52). This is apparent in the document, *Listening, Hearing and Responding* (DH 2002), which outlines an action plan for increasing the participation of children and young people. The document describes the processes put in place to promote 'citizenship and greater involvement by patients... which includes children, young people and their carers' (DH 2002, p8). It cites PALS [Patient Advice Liaison Service] and ICAS [Independent Complaints Advocacy] as services through which young patients can make complaints and influence service provision; a route that is not without challenges. PALS was assessed some years later in the document *Anyone Listening?* (Burke 2010) and it was 'found that children and young people were low users of PALS' (Burke 2010, p42). *Anyone Listening?*

(Burke 2010) investigated evidence of children's effective participation in a range of decision-making situations in England, including participation in healthcare. It undertook to examine how Article 12 of the UNCRC was being implemented, as it is a key right in the Convention that children should be able to 'express views in all matters that affect them, and for 'due weight' to be given to their views' (Alderson et al. 2006, pp301-2). However, in *Learning from Bristol* (Bristol Royal Infirmary Inquiry 2001) it is suggested that recognition of the UN Convention in UK children's healthcare 'may appear to some to be a triumph of gesture over action' (2001, p427). Indeed, 9 years after the previous comment was made, it was surmised in *Anyone Listening?* (Burke 2010) that effective communication and being listened to by health professionals is variable:

A major issue was that health professionals do not always speak directly to children or explain the illness or treatment plans to children. This caused children to feel anxious and stressed (Burke 2010, p44).

Thus, the nurturing of participation, the implementation of PALS, and the standards itemised in the UNCRC have not comprehensively addressed the development of children's participation in their healthcare.

*Listening, Hearing and Responding* (DH 2002), much as it promotes participation, also relies on 'the use of audit as a technology of governance' (Powers in Stenson & Watt 1999, p194). This can be seen in the way the document sets out its plans for the future including:

- Patient Surveys – developed for children and young people;
- Working models in parent partnership developed for the Patient Advice and Liaison Services (PALS);
- Development of the Independent Complaints and Advocacy Service (ICAS);
- Development of the Children's National Service Framework (NSF) and some national roadshows for children and young people;
- Improving the hospital environment and facilities to meet the needs of teenagers and young children. (DH 2002, p25)

*Listening, Hearing and Responding* (DH 2002) makes claims of intent that appear to act as evidence of activity, irrespective of their outcomes, as discourse can set up 'boundaries between 'policy' and 'delivery'... so that debate about means supplants

debate about ends' (Clarke & Newman in Stenson & Watt 1999, p194). The document dates from 2002 and, as noted, the outcomes of both PALS and ICAS have not proven to be entirely successful in promoting young patient's participation in their care, partly as access to these services is problematic for children. This is notably so for disabled children, who are reported as less likely to be involved in approaching services and in decision-making or participation than are their non-disabled peers; 'the active involvement of disabled children in multi-agency services remains underdeveloped' (Burke 2010, p48). Thus, the instrumentality of providing more services in order to implement participation appears to be rhetorical; the setting up of services becomes an act in and of itself, it is a 'means' without clear evidence of any 'ends'.

A more recent document produced by the now defunct NHS Centre for Involvement, *Involving Children and Young People in Healthcare: A Planning Tool* (DH 2008), was developed to support staff in building on engagement and participation with young patients. It was produced in collaboration with the charity *Action for Sick Children*, and it encourages NHS staff to develop 'involvement activities' with children and young people, such as seeking their input in the redecorating of a children's hospital ward. It was designed because:

Whilst there were many areas that were developing a culture of participation and engagement... it was also apparent... there was still some way to go for this to be embedded into NHS organisations and their practice. (DH 2008, p4)

The document calls for NHS staff to create participatory methods for improving children and young people's participation, but there is a sense of needing to do this activity in order to function in and as a bureaucracy as it states that 'organisations need to be able to provide evidence of PPI [patient and public involvement] activity' (DH 2008, p12). The document extends the technocratic development of participation through a series of tick boxes; this process has been commented upon by those working with young disabled people:

Staff from... voluntary organisations suggested that local authorities did not always seem to genuinely listen to the messages from disabled young people. They suspected that participation was something of a 'tick-box' exercise.

(Viper 2012, p29)

Leading to the question:

Does this model of delivery allow a local authority to 'tick the participation box' without changing their own culture? (Viper 2012, p54)

The Viper Project, which researched the participation of disabled children and young people in decision-making within their lives, uncovered how disabled young people were poorly involved:

We found only a few examples where disabled young people had any real control over what decisions they participated in. Usually it is adults who decide what decisions they can participate in. (Viper 2012, p31)

The repeated underrepresentation of disabled children and young people at all levels of healthcare mirrors the broader exclusions apparent in the sociocultural world, further minimising their presence as actors in their own lives. Children and young people, including those with a disability, are situated, in part, as tokenistic consumers, who are consulted on aspects of healthcare provision without bringing about real change.

Although the planning tool responds to initiatives to involve children and young people at all levels of healthcare, their input can be intermittent and unrepresentative (RCPCH 2012). Professor Kennedy in *Getting it Right for Children and Young People* (2010) commented on this discrepancy:

As regards children and young people, not only was I told of occasions when they felt that they were not really involved in their care, but there is also another deeper cultural point. DH and the NHS are anxious to suggest that the NHS's services respond to the needs expressed by the public. The only catch is that the only members of the public who are routinely surveyed are in fact adults. There is no regular survey of children and young people. Not asking means not knowing. (2010, p39)

Whilst some children and young people may be canvassed for their views, these are often on adults' terms and in relation to their agendas, using those considered 'the least vulnerable/most adult-like children' (Carter 2009, p858). For those with disabilities or long-term conditions, they may be excluded from participation in

service provision activities and also silenced at the point where service provision matters most - their health and well-being. Although these exclusions may be unintentional, they can be reinforced by a discourse of vulnerability, creating a cycle of assumptions, and conflicting with the agenda of children and young people's enhanced involvement in their healthcare.

A discourse of vulnerability runs through healthcare discourse relating to children and young people. It is expressed in comments that are regularly posited as common sense, and are repeated in earlier and later documents. In this, they build on their ubiquity and historical presence, persisting as a body of statements that are 'systematically related to each other' (Stenson & Watt 1999, p192). This discourse of vulnerability can be seen in the *National Service Framework for Children, Young People and Maternity Services: Core Standards* (DH 2004a) in which Professor Aynsley-Green, who was then the National Clinical Director for Children at the Department of Health, provides the introduction in which he states:

Children's vulnerability and the inability when young or disabled to articulate what they feel pose a challenge for all those involved in delivering health and social care services to meet their individual needs and those of their carers. (DH 2004a, p4)

Even as healthcare services strive to improve their care of young patients and build on effective communication with them, assumptions and well-meaning but paternalistic attitudes persistently underline the vulnerability inherent in both being young and in being disabled. Kennedy is quoted in the *National Service Framework for Children, Young People and Maternity services: Core Standards* (DH 2004a), where he comments on children's 'relative physical and emotional immaturity, in comparison with adults' (DH 2004a, p8). Whilst his statement is, in some ways, a fair assessment, it does also set up a comparator with adults, implying children's lack through the use of the term 'immaturity'. The weight given to assertions that are made by highly regarded individuals such as Aynsley-Green and Kennedy can entrench and replicate their attitudes, conflicting with the goal of participatory engagement with young patients.



In the document *Bridging the Gaps* (RCPCH 2003), some useful points are raised, however, as already demonstrated, it occasionally makes over-generalised and reductive statements. This is seen in the comment that ‘adolescents, in establishing their own self-identity, lack confidence to challenge services, or to develop ways of using them that meet their needs’ (RCPCH 2003, p22). This statement fails to admit the variability in how adolescents approach healthcare interactions; a variability that has been evidenced amongst my participants in their comments on communicating with doctors.

In *Listening to Children’s Views on Health Provision* (La Valle & Payne 2012) disabled children are unproblematically grouped together as members of a vulnerable group. This is seen as La Valle and Payne (2012) explain the literature review they undertook for their document; ‘we specifically searched for literature on a wide range of vulnerable groups... those with a disability, learning difficulties and long term health problems’ (2012, p12). Disabled and ill children and young people can indeed be vulnerable, but the uncomplicated way in which this is repeatedly asserted does so without any qualifying of these claims. Moreover, when these claims are made without explanation or caveats, they may become further embedded and relatively untroubled by counter-claims as to their veracity. Additionally, if researchers adopt La Valle and Payne’s (2012) technique when conducting literature reviews and related research activities, their ready acceptance of vulnerability may determine their findings rather than troubling and questioning such notions. Even as disabled children and young people’s participation is written into policy it is often circumscribed by caution over their ‘vulnerability’, marginalising them still further as they are subsumed within a ‘hierarchy of marginalisation’ (Carter 2009, p861).

The Department of Health’s *Getting the Right Start: National Service Framework for Children – Standard for Hospital Services* (DH 2003) comments that more needs to be done to improve children and young people’s participation in how the NHS is structured. It cites various reports, including the findings from the Bristol Inquiry, and asserts that services should be focused on the needs of the individual child. It also states that ‘children are more vulnerable than most adults, and have a greater need

for safeguarding their welfare' (DH 2003, p10). This statement reveals the anxiety that exists around the establishing of participation in the NHS, when viewed through the lens of widely reported failings. In *Learning from Bristol* (Bristol Royal Infirmary Inquiry 2001), it is stated that:

Safety is fundamental to achieving a high quality service. As is apparent from the lessons of Bristol, it must be the responsibility of all staff... to create a culture of safety within the NHS. (2001, p274)

In *Getting the Right Start: National Service Framework for Children – Standard for Hospital Services* (DH 2003) it notes that:

The NSF will also need to respond to the recommendations on child protection set out in *The Victoria Climbié Inquiry*. (DH 2003, p5)

The language of participation and the ongoing policy statements which respond to the need for more involvement by young patients conflicts with the cautious and risk-averse underpinnings inherent in the NHS that perpetuate constructions of children's vulnerability and their need for safeguarding. This duality is apparent in a section on clinical governance, which, whilst not about participation, implies the overall caution that informs hospital procedures. It states that:

Clinical governance systems do not always explicitly recognise children and young people as a separate and vulnerable client group. In addition to the particular issues of child protection, hospital procedures and systems themselves can jeopardise the safety and wellbeing of children. (DH 2003, p21)

Caution is apparent in the document, which both proposes greater involvement for young patients, who 'should be encouraged to be active partners in decisions about their health and care' (DH 2003, p9), but which limits these proposals due to concerns over the welfare of young patients. Notably, a risk of harm from the service's own 'procedures and systems' is mentioned; the health service is not fully secure in its own role as a reliable site of care and wellbeing. Safeguarding concerns are also apparent in *Listening, Hearing and Responding* (DH 2002); the document describes how the Department of Health is dedicated to promoting citizenship for children and young people by listening to them and increasing their

influence in decision-making. However, the listening process appears to have the dual role of ensuring young patients' safety; a duality that is evidenced in the assertion that 'listening to children helps protect them from harm and keeps them safe' (DH 2002, p12). The document further elaborates on the linked themes of safety and participation:

It is crucial when working directly with young people that the Department ensures their safety and supports them to encourage positive participation. It is the responsibility of the Department to make sure that young people are not placed at any risk of harm. (DH 2002, p20)

The Department of Health's promotion of participation exists alongside fears about children's vulnerability and safety.

As part of the call to include children in their care, *Getting the Right Start: National Service Framework for Children – Standard for Hospital Services* (DH 2003) states that those working with children require training in how to support children to be active partners in decision-making. This includes training in how to listen to and communicate with them and the 'need to understand the extent and the limits of children's comprehension at various stages of development' (DH 2003, p16). However, as discussed in Chapter One, and demonstrated in my participants' varied comments in Chapters Three and Four, age and stage are not clear indicators of competency (Alderson 1993), and some of the variance amongst my participants will be further discussed in the final section of this chapter. Incorrect assumptions can distort adult readings of children's abilities:

A powerful distorting pressure when assessing children's abilities is the tradition of the child negatively defined as not adult: not wise, informed or experienced. (Alderson 1993, p193)

To speak of the 'extent and the limits' of a child's comprehension can be less constructive than to assume a level of understanding and an ability to be involved. The promotion and discourse of participation is compromised, even as its facilitation is written into Department of Health documents. Change in healthcare is troubled by the embedded nature of attitudes and practices, so that discursive shifts are not

'neatly sequential and older agendas cannot be wholly displaced' (Stenson & Watt 1999).

The reiterative nature of discourse and its institutional rootedness makes it resistant to new approaches, and newer discourses that appear to be promoting change may represent an 'appeal to progressive change rather than an effective mechanism to achieve such change' (Davis & Watson 2000, p225). Kennedy (2010) asserts that operational frameworks 'shape the culture in which the NHS delivers its services' (Kennedy 2010, p47) and some of the issues Kennedy is addressing convey how discourse is instilled in institutional frameworks, in their culture and practices. Documents speak from within the confines of discourse so that change is restricted by the institutional system in which the discourse operates, revealing 'the conflict between rhetoric and reality' (Kennedy 2010, p84), and aspiring to a 'possible future reality rather than one that actually exists' (Shaw 2010, p206). In *Getting the Right Start: National Service Framework for Children - Standard for Hospital Services* (DH 2003), Kennedy found:

Hospitals operating in self-contained worlds, as if the prior experience of the child... had no bearing; and as if what happened to them afterwards, or outside the hospital was of no concern. (DH 2003, p14)

Kennedy's assertion implies how young patients' contexts may be overlooked in the 'self-contained world' of the hospital. The hospital can, according to Foucault, be a site of 'systematic, homogeneous observations... of frequencies and probabilities' (Foucault 1973, p52), wherein there is an 'annulation of variants' (Foucault 1973, p52). Thus, the specifics of each child's circumstances, their abilities and their life experiences beyond the confines of the hospital may not be recognised in healthcare working practices.

In the next section, the participation of children and young people in medical research will be discussed. In this setting there is a likelihood of particular caution, with children's circumstances and contexts overlooked due to legislative and discursive practices.

## Discourse in medical research

As established in Chapter One, failures in NHS care that have attracted national publicity and public inquiries (NICE 2002), and worldwide 'violations of human rights in the name of biomedical research' (Boulton & Parker 2007; Yeung 2007, p88) contribute to the shaping of the current discourse of ethical research and protection of the 'vulnerable' in medical research guidelines:

Well-documented abuses of human subjects in medical experimentation including research with children, created concern... this led to the perception that children were vulnerable, given their inability to protect their own interests. (Oberman & Frader 2003, p301)

The regulation of medical research on children and young people has developed from the position taken after the Second World War when the Nuremberg Code 'effectively forbade research with children' (Brierly & Larcher 2010, p2). This has somewhat influenced current ethical practices and discourse relating to children and young people's participation in research:

As ethics expanded from a way of thinking about research into a system of governmentality, it generated its own discursive systems, meanings and representations of the world, evolving into a particular sort of institutional discourse. (Halse & Honey 2007, p339)

The intention to develop new approaches to working with children and young people, including those who are disabled, can be limited and shaped by 'what can be thought about and acted upon' (Shaw 2010, p201) within the risk-averse discourse of medical research. One of the outcomes of young patients being effectively involved and communicated with would be that they are better able to make decisions on their healthcare. This has significance if they are thinking about or going ahead and taking part in medical research, an activity that is encouraged in the Department of Health's *Research Governance Framework for Health and Social Care* (DH 2006) which states that:

All those using HPSS [health and personal social services] should give serious consideration to invitations to become involved in the development or

undertaking of research studies. (2006, p25)

In this statement, taking part in research is constructed as an activity for ‘all those’ using health and social services in the UK. However, whilst the participation of minors is encouraged as a worthwhile endeavour (RCPCH 2012), it is limited by the risky status often attributed to children’s involvement. The European Compliance Academy [ECA], as discussed in Chapter One, aims to build harmony in clinical trial rulings across the EU, and its guidelines for paediatric research state that:

Protection against the risks of research in such a vulnerable population is paramount, whilst this should not lead to denying them the benefits of research. (ECA 2008, p4)

The ECA define all children under the age of 18 as minors and they state that where research with a minor is necessary, the least vulnerable children should be used:

If research with children proves necessary, the least vulnerable among them should usually be included (ie older children) if there is a necessity to subject children to a clinical trial, the choice of subsets of the paediatric population to be included should be made on the likely target population for the medicine being tested. (ECA 2008, p5)

The framing of the term ‘vulnerable’ in the above passage implies that age is specifically tied to gradations of vulnerability, and this is sometimes the case. However, children and young people’s long-term experiences of illness or disability can inform their approach to medical research decision-making, with their competence to consent developing ‘through experience rather than by age’ (Alderson et al. 2006, p302). The ECA guidance could be contributing to unproblematised notions in UK healthcare discourse of inherent childhood vulnerabilities. The guidelines exemplify both the requirement for more children to be involved in research and the ways in which this is complicated by a highly cautious approach to the inclusion of a ‘vulnerable’ group:

The recommendations in the document aim to contribute to the promotion and protection of the dignity, the well-being and the rights of children (minors) all of whom are vulnerable and unable to give informed consent. (ECA 2008, p5)

Consent processes involving children are considered difficult by many in the medical community; 'as is well known, the role of a child... in the process of gaining informed consent... is problematic' (Bristol Royal Infirmary Inquiry 2001, p420).

Unsurprisingly, this is even more contentious in medical research.

The *Medicines for Human Use (Clinical trials) Regulations* (2004), already quoted from in Chapter One, and which regulate trials in the UK, refer to consent involving minors, which in their definition means those under the age of 16. Point 6 states that the minor should have:

received information according to his capacity of understanding, from staff with experience of minors. (p43)

Point 7 states that:

the explicit wish of a minor who is capable of forming an opinion and assessing the information... to refuse participation in, or to be withdrawn from, the clinical trial at any time is considered by an investigator. (p43)

Point 13 states that:

informed consent given by a person with parental responsibility or a legal representative to a minor taking part in a trial shall represent the minor's presumed will. (p43)

The above regulations, designed to protect the research subject, may impinge upon the participatory rights of those who are being protected; rights that are being nurtured in other parts of the health service as part of the transition in how services are delivered (RCPCH 2012). In one reading, the regulations are, rightly, cautious in how the minor is involved in clinical trials. However, their depiction is one of a relatively passive, voiceless subject, dependent upon adult assessment. Point 6 refers to their 'capacity' to understand the information given, Point 7 states that any 'explicit wish... is considered by an investigator', and Point 13 refers to their 'presumed will' during the giving of informed consent; these all hinge upon the mediating role of adults. This mediating role may be a positive and supportive factor even for very competent children, and the guidance of trusted adults can lift the

anxiety and burden of decision-making (Alderson & Montgomery 1996). However, the way in which the guidelines are framed situates all minors as vulnerable and unable to consent, leaving differences in competence unacknowledged or dependent upon adult 'presumptions'. As pointed out in Chapter One, Point 7 does call for attention to be paid to a minor's 'explicit wish' to refuse or withdraw from participation, however the minor's decisions are not, apparently, central to the consent process. This reveals some divergence from the broader rhetoric on the participation of young patients in their care and treatment; point 5 of the regulations states that those with 'parental responsibility... may... withdraw the minor from the trial at any time by revoking his informed consent' (p43). The minor may be listened to and given information according to their capacity, but the adult consent-giver has the final say in the decision to participate or to withdraw from participation.

In *Guidelines for the Ethical Conduct of Medical Research Involving Children* (Hull 2000), issued by the Ethics Advisory Committee of the RCPCH, children's vulnerability as research subjects persists:

The urgent desire to offer babies and children the potential benefits of medical research is laudable. Yet childhood is a vulnerable, formative time, when harms can have serious impact as well as being potentially long lasting. (Hull 2000, p178)

Hull (2000) is supportive of promoting research involving children, but the repetitious notion of children's vulnerability misses more nuanced depictions of children's capabilities. Furthermore, Hull (2000) states that, 'despite careful selection, children in clinical trials have social and emotional problems that are mainly unpredictable' (Hull 2000, p179). These problems may occur, although they are not necessarily exclusive to children, yet the 'vulnerability of child patients' (Hull 2000, p178) is reinforced in a discursive and decontextualized loop. Repeated use of the term 'vulnerable' and the subjective assessments made by clinicians could render:

any apparent autonomy the child may have in health care as ultimately meaningless, leading to the repression of the child hidden under the acceptable guise of protection. (Moore & Kirk 2010, p2216)



In the following statement from the document *MRC Ethics Guide: Medical Research Involving Children* (2004) the Medical Research Council [MRC] situates children as now being more actively involved in medical research participation:

Earlier research tended to be on children, with children regarded as subjects of the research. Increasingly, research is conducted with children, who are involved as active participants. (MRC 2004, p10)

However, the MRC uses the language of vulnerability in a later passage as it discusses the value of assessing the benefits and harms of research:

In the past, the concern to protect children from the potential harms of research may have denied them potential benefits. To ensure that this vulnerable group are not exploited, the General Medical Council advises that it is important to assess carefully the potential benefits and harm to children at all stages of any research. (MRC 2004, p15)

Protection of all candidates for medical research is of the utmost importance, but the undifferentiated language used, positioning children 'as defenceless research subjects' (Carter 2009, p860) avoids the complexities of accommodating individual competence and maturity:

The perception of the child as a vulnerable individual has been the metric for regulation and legislation... we regard this approach as leading to regulatory approaches that are blunt instruments. (Hagger & Woods, 2005, p59)

Ideally, protection is balanced with children and young people's active involvement (Nuffield Council on Bioethics 2011), a possibility that is not yet readily integrated into the documents being analysed.

### Discursive absences

In *Turning the Tide: Harnessing the Power of Child Health Research* (2012) the RCPCH argues that the translation of research evidence into practice and policy is integral to improving children's health as:

current health indicators suggest that we have fallen from our position as a

leader in child health to one where our outcomes are poorer than most of our European neighbours. (RCPCH 2012, p4)

The RCPCH also note in another 2012 document, *A Healthy Nation* (Modi et al. 2012) how:

A striking dissociation exists between high-level national policy statements that acknowledge the importance of children's health and wellbeing such as the... national service framework for children, young people and maternity services and the poor recognition of the need for research in children... to improve health care. (Modi et al. 2012, p10)

There is a lack of a coordinated means of promoting research in children, and a clash of purposes and discourses. The call, cited earlier, in the *Research Governance Framework for Health and Social Care* (DH 2006) for the population's diversity to be represented in medical research is problematic when a discourse of vulnerability can define an individual's positioning and the 'situation that it is possible for him to occupy' (Foucault 1973, p52). In *Ethical Considerations for Clinical Trials on Medicinal Products Conducted with the Paediatric Population* (ECA 2008) all children, both healthy and disabled or ill, are constructed as 'vulnerable' with regard to clinical trials:

In principle, healthy children should not be enrolled as healthy volunteers, because they cannot consent and are vulnerable like children with a disease or condition. (ECA 2008, p20)

The rights and developing responsibilities as a young social citizen (Hagger & Woods 2005) to be involved in a clinical trial or medical research are challenged by the circumspect attitude of the research community. Some of these attitudes may be based on valid concerns, as Boulton and Parker (2007) observe:

The progressive tightening of research governance... including consent [has] regularly followed the exposure of abuses of medical power. This is at least in part a reflection of ... attempts to protect research participants from technological and other imperatives, and the power, of medical research. (2007, p2189)

Whilst it is reasonable to protect all medical research candidates, including children,

the way in which children's vulnerability is reiterated often goes unchallenged. The exclusionary subtext of the language used and the lack of context repeats notions of vulnerability without explanation, and those considered most vulnerable are given little opportunity to disprove claims made about them. Protectionist and exclusionary language persists in the ECA's (2008) document and the recommendations it makes:

The clinical trials performed in children should be carried out under conditions providing the best possible protection for this vulnerable population whilst recognising children have the right to benefit from research. (ECA 2008, p5)

At times the document speaks of children and young people constructively and as having evolving capacities:

Whenever appropriate, the child should participate in the (informed) consent process together with the parents. Involving children in discussion and the decision-making process respects their emerging maturity. (ECA 2008, p11)

The language is, however, frequently based upon assumptions of vulnerability and the importance of conducting research on the least vulnerable children. Therefore, the opportunities for disabled children and young people to participate are problematic (Hayward & Kuhn 2010, p5) and constructions of their vulnerability can reinforce professionals' attitudes:

the discourse this creates impacts upon professionals' attitudes towards disabled children and is often reflected in the power adults exert over children. (Viper 2013, p44)

The development and practice of medical research procedures can create 'discursive absences' (Stenson & Watt 1999, p196), effacing the presence of those considered the most vulnerable. The discrete treatment of competence as either present or absent does not allow for the possibility of individual gradations of capacity, and disabled children's ready positioning as 'vulnerable' may be intensified in the discourse of medical research. This can result in their participation being managed by others, with adults making decisions for them, due to perceptions about their lack of competence. Although discussing qualitative research, Halse and Honey (2007) observe how the institutional discourse of ethical research tends

towards generalising:

Herein lies the ontological dissonance with the practice of ethical research. In the 'real world' of research, identities are more fluid, mutable, and difficult to pin down. (Halse and Honey 2007, p345)

Amongst my participants there are a range of capabilities and contextual vulnerabilities, but a persistent and unquestioned discourse of vulnerability can efface recognition of a child or young person's competence. 'The notion of vulnerability is all too often closely seen as interchangeable with the notion of lacking competence' (Carter 2009, p861). McNeish (1999) observes the limitations discursive assumptions can place on certain groups:

Negative assumptions and stereotypes which can be applied to young people generally may be even stronger for certain groups... Disabled children and young people can experience particular difficulties in participating in decisions affecting their lives. (McNeish 1999, p200)

The issue of proving competence and being effectively involved in consent-giving can be acutely so for disabled children and young people. Generalised assumptions and homogeneous notions of children and young people's vulnerability in medical research can place those with disabilities at a particular disadvantage. In some cases this may result in them having a limited role in decision-making and in the giving of consent prior to taking part in medical research. The value of discourse analysis is in the way that it can question assumptions, through emphasising the contextual and the heterogeneous (Lupton 1992).

The risk of harm has contributed to shaping the way children and young people are thought of and treated in medical research, where broad measures have led to current constraints:

There has traditionally been a reluctance to include children in clinical studies... In large part, this situation arose from a desire to protect children from unethical research, the potential dangers of experimental drugs, and invasive investigative techniques. These historical constraints no longer serve children well. (Modi et al. 2012, p1)

This is not to suggest that children and young people should make complex decisions on taking part in medical research alone. As has been examined in my research, each of the participants deeply valued and trusted their parents' support and advice, and all expressed some level of wanting their continued help into young adulthood. What was also apparent was that the participants thought decisions on medical research participation should be guided and made according to the child or young person's preferences.

In the final section of this chapter I will move away from discourse analysis and discuss some of the participants' comments, these belie the generalisations in the discursive framings I have highlighted here:

Children's research participation is set within competing - primarily adult-oriented - discourses which view children as a homogenous and naturally vulnerable group. (Carter 2009, p863)

The restrictions and omissions in discourse, and the propensity to de-contextualise on issues relating to children and young people's status and capacities are challenged by the capabilities amongst the participants I interviewed.

### Contextualised lives

In this final section I address the limitations of discourse and the absence within it of heterogeneous possibilities. I will draw on the participants' thoughts to counter and fill the gap between discursive tropes and the variety of lived experience.

Rick (12) put it well when I asked him if an imaginary boy should be free to make his own decision about participating in medical research without being pushed into it by his parents. He responded; *'yeah 'cos it's up to the child to make up their own mind whatever they want to do'*. At his second interview [by telephone] Rick explained the motivation behind his recent decision to take part in some medical research, as I asked him if he was happy to be getting involved. He commented, *'yeah... if I can make other people's lives better with the same disability, yeah it would be good'*. Rick sees participation as a personal benefit to him and also

potentially beneficial to others, and this situates medical research participation as contributing to a sense of purpose in some disabled young people's lives. Like Rick, Emily (21) felt that a young person should be guided by their own preferences, even though, in earlier comments, she has spoken of how a child may feel they should make the decision their parents want them to make. Here, Emily focuses on giving an imaginary 14 year old boy some advice on deciding about medical research participation:

*I would say if you really don't want to do it then don't do it, he's 14 so it's not like he's really young, if he really doesn't want to do it, you don't have to do it, there are other people, he's not the only one with Duchenne, there are other people that can take part and if he's scared to do it I don't think he should be pushed into doing it really. I would say it's a good thing to do but if you aren't happy with it don't do it.*

Emily is clear in the above comment about how a decision should be directed by the boy's preferences without him being 'pushed into doing it'. However, when communicating with doctors at hospital appointments Emily prefers this to be a shared interaction involving her mum. This does not indicate a lack of competence, or that she cannot make decisions, it suggests a relational and contextualised approach that requires the child to effectively communicate with and trust their parents to help them. Emily explained this approach:

*S: So it's important they [doctors] get a balance, sort of talking to you and your mum when that's appropriate?*

*Emily: Yeah, like if, for them to talk to me but then for it to be Ok if I say 'oh mum can talk now' and he talks to my mum as well so it's like just to talk to both of us not one or the other.*

Doctors might read this as vulnerability or passivity, but it can be a demonstration of a mature ability to delegate (Alderson & Montgomery 1996), sharing the responsibility, and at times the overwhelming nature of coping with a serious, untreatable condition. The approach Emily discusses calls on doctors to remain focused on the patient, whilst also working with the parent who is acting, at times, as their child's spokesperson. This approach reflects the dynamic that can also shape decision-making, with the child and parent working together in a supportive setting.

Tom (17) feels decisions should be approached with the child and parents making 'a decision between them' and this concurs with a relational view of autonomy which is a common approach when young people make decisions on their healthcare; 'in most cases choices are likely to be made within a family setting' (Cave 2011, p443). Adam (16) gave some thought to medical research decision-making and why it is vital that the child is listened to:

*If the parents are sort of really pro-research then they might not sort of stop to think about what the child might be feeling, whether they feel scared or they don't like injections or they just don't want to be at hospital all day, because the parents aren't like doing it themselves they're not thinking in the same depth as a child would be, I'd have thought... specially if they're very pro-research, and they'd be thinking sort of the good points from it, of the research points... if the research is what they've got in their head then that's what they'll kind of think about, the research parts of it.*

The subtleties in decision-making that are suggested by the participants' comments are often absent or poorly developed in consent procedures; and each comment shows a slightly different emphasis. Rick feels the boy should make his own mind up, Emily too felt that, certainly for a 14 year old, the decision should be their own to make. Tom thought the boy and parents should make the decision together, and Adam considered how parents who are 'pro-research' may overlook the child's wishes. The participants' comments encompass aspects of their own preferences and their sense of a just and fair approach to decision-making. Adam is aware that there can be influential elements in decision-making, with some parents' ideals and motives effacing their child's wishes. He commented during his second interview [by telephone] on how he would give advice if he had a son with DMD who was making a medical research decision:

*Adam: I'd probably talk to them about the different aspects from different points, from both sides but I wouldn't force them into doing what they didn't want, just sort of let them weigh up the options for themselves.*

In the above quote, Adam recognises the child's ability to 'weigh up the options', with Adam, as the parent, being careful not to force the child into reaching a particular decision. This approach depicts how listening to children and young people and acknowledging their thoughts, abilities and decisions makes space for their views to be exercised and heard, thus contributing to the process of ethically conducted

medical research.

The participants also demonstrated that they are busy living in the 'now', enjoying friendships and doing the things that give life meaning. Jay (14) described some of the activities he enjoys; '*swimming, playing on my X-Box, I like playing tennis as well... [and] when I'm on holiday I go fishing*'. Rick (12) too spoke of his current priorities as he recounted his daily life:

*Rick: I find my friends most important out of all that... I do see them quite often I try to see them like, as often as I can.*

And,

*Rick: I usually have a lie in at the weekend; usually try to put effort into doing things as a family and seeing my friends and stuff, that's what I mainly do at the weekend.*

Ollie (14) meets with friends and has a life that, with his parents' help, is focused on keeping busy:

*Ollie: Yeah I go out to the cinema a lot, I'm a big fan of movies I'm a movie buff, yeah I go bowling quite a bit and I sometimes go for walks and sometimes I have friends and people round... sometimes I go to football matches... I go to a lot of games I go to loads of games.*

Even Ed (18), who does not like to be away from home, enjoys his older brother's company, '*I always talk to my older brother... 'cos we basically like the same music and films*'. The child can, in healthcare discourse, be viewed through a future-oriented lens, and in the context of medical research they are invariably positioned as vulnerable and passive, with the approach to decision-making being adult-led and bounded by caution. What came through strongly in the participants' accounts was that they are living and investing effort in the present, but they also demonstrated that they can cope with and discuss serious issues, in appropriately contextualised ways. They live with the realities of their condition but they are not subsumed by their disability or by the challenges relating to it. They are leading lives that have meaning for them, in which they are socially engaged individuals who maintain friendships and take part in family life, and the limited promise of medical research for these specific individuals means that it is not something they overly dwell upon.



Tom (17) illustrated his low expectations of a research breakthrough when I asked him what he would do if he heard about a new drug for DMD. He commented; '*I think I'd be in shock, it's such a big thing*', he said this with humour, and his knowledge about DMD and the slow pace of research is experiential. Discourse can represent childhood as a time that is free from the cares of adulthood (Meyer 2007), but this does not speak for children and young people with significant disabilities who have adapted to their situation. Boyden (2003) contends that policy and practice can decontextualise childhood and ignore the resourcefulness of children, positioning those facing hardships as vulnerable and as victims. Emily (21) showed how she has adapted to her disability and is a resourceful actor in her life as she spoke of her plans for the future:

*I wouldn't want to live at home all my life... I will move out and live on my own with help, 'cos I don't need someone there all the time it's just certain things like with food shopping carrying loads of bags or doing my washing.*

She knows she will need help but this is incorporated into her plan and is 'normalised' within the life she leads. What has come through from the interviews is that the participants have tough times, but they have developed insights because of this. They wisely choose to invest in the lives they are leading now but they also have intelligent contributions to make on the healthcare issues and decisions affecting them. Considering disabled children and young people to be helpless denies them the opportunity to express themselves against 'adult decisions and actions' (Boyden 2003, p26). Therefore, attention should be given to children's competencies in decision-making, something that is not readily evident in the discourse emerging from healthcare institutions and medical research guidelines, yet something that is evident from my participants' contributions.

## **Conclusion**

Discourse can work at micro and macro levels (van Dijk 2008), the micro level occurring in, for example, the interactions between a doctor and child patient. The macro level is the overarching legislation, code of conduct or rules adhered to by doctors that inform their role through a combination of knowledge, information and/or

authority (van Dijk 2008). The practices and authority of doctors can appear to be a natural outcome of their role, yet in their micro level discourse doctors may reflect the macro level of the medical institution:

The norms of the professional culture are internalized by the clinician during their acculturation into the profession and the clinical culture. (Crowe 2000, p72)

Thus, it is possible that the macro level discourse may reinforce and perpetuate the other; in this case through shaping the way doctors regard and treat their young patients because of the discourse and culture within which doctors and healthcare professionals work:

Cultures develop a consensus of meaning for certain actions which individuals within the culture take for granted as the common sense way of interpreting the world.... These cultural norms are internalised as constructions of reality by individuals within the culture. Discourses that have the power to categorize behavior as within or outside the norms tend to do this by situating the individual as faulty if they do not adhere to these norms. (Crowe 2000, p71)

In some instances, paediatricians hold 'stereotypical and patronizing views about adolescents' (Beresford & Sloper 2003, p178) and, whilst this is not across the whole community of paediatricians, other reports also document the way in which professional attitudes, institutional practices and an 'anti-holistic categorisation' (Kennedy 2010, p70) of children and young people can negatively impact them.

A paternalistic standpoint that constructs children and young people, particularly those who are disabled, as vulnerable and in need of protection may go unchallenged:

Pervasive structural relationships of power and powerlessness between groups tend to foster ideological justifications for the maintenance of such relationships. Care discourses can run the risk of being used for ideological purposes where 'differences' are defined in the service of the dominant group. (Cockburn 2005, p80)

It is also apparent that there is a gradual culture shift in attitudes, with a greater

emphasis on shared decision-making between practitioners, children and young people, and their families (Berntsson et al. 2007; Jessup & Parkinson 2010). However, the cautious, risk-averse and paternalistic discourse has a strong precedence that is also evident in newer ways of working. As discussed in my research, disabled children and young people may not wish to make decisions on treatments or medical research alone, but this does not preclude clinicians and healthcare staff from addressing them as respected and fully present individuals whose views should be listened to. Failing to do so may lead to children and young people missing out on developing essential decision-making skills (Viper 2012) and lacking the confidence to communicate their own preferences:

This is particularly pertinent for many disabled children and young people who through social exclusion and the disabling attitudes of others may have been denied opportunities to participate in decision-making and have little experience of choice making within their own life. (Viper 2013, p3)

Discourse has various manifestations of its power, some of which may be hard to detect because of 'prevailing cultural norms' (Corrigan 2003, p789). The assumptions present in discourse can diffuse and normalise the workings of paternalism behind a screen of being in the young patient's 'best interests', which may fail to give them a say in decisions, leaving them powerless:

Disabled children are frequently disempowered. They experience discrimination and oppression on the grounds of being not only a child, but also of being disabled. (Viper 2013, p44)

Amongst my participants, their comments imply a contextualised grasp of their condition, and they have demonstrated an experientially gained knowledge of their healthcare. These components indicate that the participants are moral beings (Carnevale 2004) who can effectively be involved in decision-making on their healthcare and on medical research. They may not want to make decisions in 'lonely autonomy' (Alderson & Montgomery 1996, p157), choosing instead to draw on their parents' support and this choice suggests they know their own limits and welcome the advice of others. This signifies a mature competence amongst many of the participants, where healthcare and medical research decisions are dealt with in a

supportive setting, emerging from contexts within which incremental decisions have been made over the course of time.

## Chapter Six

### Conclusion

The primary objective of my research has been to explore what influences from the participants' lived experience and sociocultural setting shape their attitudes as they think about or make a decision to take part in medical research. The research has considered how the participants can demonstrate agency and independence in their lives despite having a chronic, progressive condition and has examined whether they are vulnerable and in need of protection, or if they can make decisions on their healthcare. Healthcare guidance and legislation promote the enhanced involvement of children and young people in their healthcare, although this involvement can be limited by concerns for their safety. Risk management in the more closely controlled area of medical research can further limit the involvement of children and young people, who are regarded as vulnerable and in need of adult interventions when decisions are made. However, I propose and will further explain how disabled children and young people have distinctive knowledge and experiences that can help them to be actively involved in decision-making on their healthcare and medical research participation. Recognising the participants' developing maturity and individuality, despite their condition (Rosenbaum 2008; Vuillerot et al. 2010) acknowledges how they can approach decision-making as active agents, countering assumptions about their vulnerability and immaturity. The participants act in and upon their lives, and their perspectives and experiences can contribute to the way in which they are involved in the complex dynamic of making decisions. Disabled children are often denied the right to communicate their views (Morris 1999), and giving consideration to the participants' thoughts and experiences contributes vital understandings on the role of agency, autonomy, competence, and shared decision-making (Dixon-Woods et al. 1999) in the lives of disabled children and young people.

The participants described the way in which the responsibility for decision-making is likely to be shared with their parents, whilst also being directed by their own preferences; and relational autonomy helps us to understand how this joint means of reaching decisions operates, without undermining the participants' independence and agency.

## **Independence, agency and relational autonomy**

The sociology of childhood and disability studies both advocate for contextualised competencies and this perspective opens up possibilities for disabled children and young people. They can be able partners in their healthcare (Alderson 1993), as children with long-term conditions can know far more than people who have had short-term or emergency conditions (Alderson 2007). Healthcare staff may judge competence using rigid age-based criteria, but competence is also experience and context dependent (Alderson et al. 2006; Hagger & Woods 2005). Therefore, the inclusion of young patients in the ongoing continuum of their care can further the development of independence, agency and autonomy as these are, to some extent, contingent upon how adults inform and involve them in decision-making (Alderson 2007). The specific forms of agency and independence that are experienced in severely disabled children's lives may go unrecognised or be devalued if compared to generalised understandings of these terms. This can be rebutted; however, when it is acknowledged that the self is constituted in relation to others, with agency and autonomy being socially contextualised (Stoljar 2011) and relationally contingent. Relational affiliation gives the participants their sense of identity and orientates them within a field of caring concern, and their decisions may be reached with the help of parental advice. This influence can positively contribute to the participants' overall sense of agency and independence, as parents can give contextualised guidance based on a knowledgeable and caring relationship.

Relational autonomy is a viable means of cooperation, and working together does not automatically equate to the participants' wishes being subsumed by adult motivations; although this process may be poorly understood by those who associate physical dependency with other forms of dependency. The participants' experiences suggest that it is possible for physically impaired people to manage independent dependency (Dreyer et al. 2010), wherein requesting and receiving support creates a sense of control and independence in their lives (Delmar et al. 2006). For disabled children and young people, the 'preservation of an overall sense of identity, agency and selfhood' (Ho 2008, p131) that is achieved in relationship with their parents appears preferable to making decisions alone. Drawing from what the participants

have said, parents variously act as their child's spokesperson, gatekeeper and advisor. This relationship was portrayed as the participants spoke of working with their parents when communicating with doctors and how this would also be the case if making medical research decisions. Out of these shared interactions, the participants emerge as individuals with their own unique ways of being in the world, who are assisted by trusting partnerships that help them to reach decisions and feel involved in their care (Tan et al. 2010).

It has been seen that parental support remains significant as the participants move into adulthood, and healthcare professionals need to recognise and respect disabled children and young people's agency, which is exercised within this supportive network of relational autonomy. It is an effective means of coping with the challenges of having a significant disability, and helps us to understand how these relationships can work and be productive. There is, however, a possibility that parental pressure can cause conflict (Gibbons 2008), with the parents' decisions being prioritised instead of the child's preferences being accounted for. It is also possible that child and parent appear to agree, but this agreement may be due to the subtle yet influential role some parents have in their child's life, detracting from the child being given the space to voice a conflicting view. The participants who expressed an opinion were clear on the point that coercion and undue pressure from parents was unhelpful, and when discussing the scenarios, they stated a belief that children's views and wishes should be respected. They reported wanting parental support during decision-making, but they also indicated that their own selfhood should be recognised by parents and doctors. For this reason, healthcare professionals must listen to and communicate effectively with their young patients, helping to ensure that well informed decisions are being made (Alderson 1993). Clinicians should respect the preferences of capable children who sometimes elect their parents to speak for them (Alderson et al. 2006), whilst avoiding the assumption that the child and their parents are 'synonymous' (James 2008, p56). Whilst there is a possibility of coercion (Gibbons 2008), the role of relational autonomy can help to mitigate the impersonal aspects of being a patient, giving disabled children and young people support when making decisions (Ho 2008).

## Discursive tropes

The layers of interaction, support and relational autonomy that occur in the participants' lives call for expanded notions of agency, independence and autonomy, particularly when healthcare practice tends towards overlooking young patients' abilities and contexts. Chapter Five discussed how healthcare discourse can minimise complexity and heterogeneity, impacting the ways in which disabled children and young people are spoken of and treated. Discourse is not all encompassing, but shared decision-making and the presence of independence and agency in the participants' lives can be constrained when healthcare professionals hold stereotyped attitudes about their young patients. Discursive rhetoric can 'idealize childhood, or imbue childhood with an implicit politics' (Shanahan 2007, p423), considering children to be future adults, rather than currently engaged agents. My findings challenge the homogenising constructions of disabled children and young people that are generated by tropes and assumptions about their futures, their limited competencies and their ascribed vulnerabilities. Considering children to be in a state of maturing towards the wholeness and rationality of adulthood can represent them as lacking moral worth in the present (Cockburn 2005, p82). Yet disabled children and young people are a diverse group, who should each be recognised as having some measure of competence; their diversity and their ways of enacting agency must be reflected in healthcare practice. This is encouraged when young patients are given ample and varied opportunities to express themselves and make decisions in ways that complement their own, specific needs and priorities.

### **Contextualised decision-making**

Doctors and hospitals are an inevitable part of the participants' lives, and this relationship is based on a contractual rather than an emotional obligation, as care is provided in a professional capacity, operating outside the bonds of familial relationships. By contrast, relationships between parents and children are often based on trust and loving support (Ho 2008) and relational ethics helps us to understand how the dynamics of the relationship between children, parents and healthcare professional are interconnected (Whitty-Rogers et al. 2009). Allowing for



this dynamic provides a productive space of mutual respect, where child, parents and professional can work towards appropriate decisions being made. It is possible that at times shared decision-making can be challenging; adults may want to shelter children and young people from negative information regarding their condition, and doctors may struggle to give young, 'vulnerable' patients a real voice in their care. However, those with serious conditions such as DMD/MD may be in a better position to cope than external assessors presume (Vuillerot et al. 2010), and their perspectives should be sought, even, or particularly when difficult decisions are being made. The subtleties inherent in the way opinions and choices can be weighed and evaluated between child and parent means that doctors should be aware of how some children and young people make decisions in contextualised ways (Coyne & Harder 2011). Healthcare professionals must attend to their patients as individuals who sometimes require the support and advice of their parents and doctors, whilst at other times they can make their own decisions (Young et al. 2003).

It has been seen that agency and autonomy are relational and social, and young patients' decisions can be understood as developing within social contexts (Gibbons 2008). The process of giving informed consent prior to medical research can be removed from its social and cultural context (Corrigan 2003), and when informed consent is treated as an isolated activity it is disassociated from the rest of a disabled child or young person's healthcare and from their experiential knowledge. However, 'human capacities... are intrinsically social and relational' (Stoljar (2011, p5), and, therefore, healthcare providers 'must be alert to the social conditions that affect patients' capacities' (Stoljar 2011, p9). Drawing on young patients' personally accrued knowledge, and keeping them well informed, can ensure that they are effectively involved in decision-making (Alderson 1993; Alderson et al. 2006). This personally accrued knowledge may be prevalent in the lives of children and young people with various long-term conditions who are 'likely to have experienced [the] limits and failures of healthcare, so that their consent may be highly informed' (Alderson 2007, p2282). When health professionals acknowledge and work with this personalised knowledge, they can give their young patients the confidence that they are being listened to and included as healthcare actors with the ability for self-determination. This then creates a positive cycle in which disabled children and

young people experience and further build their capacity for decision-making.

As discussed in this thesis, 'lived autonomy is related to individuals' connections with others' (Whitty-Rogers et al. 2009, p747), and informed consent emerges from that lived autonomy as part of a 'dynamic, social and emotional process' (Shilling & Young 2009, p5). Parents may act as mediators in the process, with joint decisions often being preferred as 'children's ability to make autonomous decisions differs from their desire to do so' (Alderson and Montgomery 1996, p44). Healthcare rules and guidelines do not readily accommodate contextualised decision-making; however, it has been illustrated amongst my participants that children and young people with DMD/MD are unlikely to reach decisions without the support of their parents. This collaborative relationship can, itself, be a sign of competence if 'competence is regarded as a way of relating to others [and] not simply an individual skill' (Alderson 1992, p122). Thus the skill they demonstrate when interacting with their parents, as they weigh-up options and consider the risks and benefits of potential research participation can be considered as signs of the maturity and competence that are a requirement in consent-giving.

It is observed that an 'investment in time and resources, training and support may be necessary to enable staff to listen to young people in their care' (Alderson & Montgomery 1996, p58), and this is vital if they are to be better involved in decision-making. Alongside listening to young patients, healthcare professionals could practice interactive methods of encouraging active reflection on decisional options, creating the 'conditions in which patients feel authorized to speak for themselves' (Stoljar 2011, p9). The techniques I employed during my interviews with the participants, using scenarios to instigate discussions, appeared to assist them in thinking about decision-making approaches. This activity opened up a space for exploring ideas, and the participants demonstrated their ability for intelligent, morally engaged discussion when given appropriately defined information. Through providing them with a way and a means to discuss complex matters, the capacities my participants demonstrated during discussions undermine assumptions about the vulnerability, immaturity and dependence of disabled children and young people. The ability to imaginatively engage with and comment on the scenarios I created

suggests that children and young people with severe and degenerative conditions can develop contextualised competence and knowledge through their experiences. This can be a valuable asset as they approach making medical research decisions, it counters notions of their innate vulnerability, demonstrating that they can be involved in decision-making as engaged actors in their healthcare and lives.

It has been explained how the scenarios I devised were a necessary addition to my research, they facilitated the participants' engagement with medical research in terms they could understand, and provided a basis on which they could develop their own thoughts. The need to adjust my approach reflects the uncertain and tentative nature of social research, particularly in this project, which has emerged from a medical context, but has been pursued within a social context. This was a challenging point from which to start, however, my findings generate understandings of the social elements of medical research, and how it is thought about and made sense of within the participants' daily lives and experiences. It was apparent that medical research is not often discussed by the participants, and my thesis represents the broader setting of the participants' lives. In this broader context, their hopes and energies are invested in daily life, in the setting and achievement of realistic goals, and in staying as healthy as possible.

## **Conclusion**

Healthcare professionals may have concerns about giving disabled children and young people too much freedom in decision-making, however, ensuring they have a stake in treatment options and the capacity to develop a 'voice' (Hayward & Kuhn 2010) are vital components in ensuring and developing active participation. Responding to the competencies of disabled children and young people, whilst also giving support when it is needed, creates opportunities for them to develop and flourish within a caring web. By contrast, conflating their need for help with notions of helplessness, and of their situated vulnerability with overall vulnerability is inappropriate. Marginalising disabled children and young people in decision-making may mean that they fail to learn how to make decisions affecting their lives until they become young adults (Viper 2013a), and this absence of experience in decision-

making may lead to vulnerability, rather than avoiding it. By contrast, encouraging participation in decision-making and providing the means for a supported move towards autonomy can help to establish disabled children and young people as capable agents.

The participants have provided accounts that help to explain how contextualised, relationally involved decision-making occurs in the lives of disabled children and young people. They challenge us to think differently about how those with DMD/MD and their families can live and cope despite huge challenges, destabilising notions of lost childhoods and parental valour through offering different, if hard won, versions of living with a disability (Carnevale et al. 2008; McLaughlin 2006). Agency and autonomy are experienced as developing, lived concepts in the participants' lives; and they are dynamic processes which are influenced both by familial values and practices and by each participant's own preferences. The nuanced, relationally contingent answers to my research questions that have been generated point to the way DMD/MD is lived with, indicating the intuitive, responsive way parents and their disabled child work together. Children and young people with DMD/MD and their parents have years of experience in sharing the child's healthcare management, and if making a decision to take part in medical research they are likely to draw on those experiences. Extrapolating from the participants' accounts, it is apparent that those with DMD/MD may reach decisions by weighing-up the relevant information and giving regard to parental views, whilst also being guided by their own choices and directives.

### **Limitations and future research**

In this final section I will discuss some of the limitations that are apparent from my research approach and its scope, and I will also briefly point to possible future research that could further our understandings of living with DMD/MD.

The research proposal was optimistic in terms of recruitment size, the sample has been small and a bigger sample may have brought different or more diverse findings; and as acknowledged in Chapter Two, in homes where DMD/MD is poorly adapted

to it is unlikely that recruitment would be successful. Another limitation is that whilst much medical research is being conducted, which holds a lot of promise, it does not offer the participants any realistic hope of a cure or 'freezing' of their condition, therefore the discussion was somewhat speculative. However, 3 participants have, or were close to taking part in some research, and better management of DMD does mean that children are often living well into adult years.

None of those interviewed reported struggling with making decisions in partnership with their parents and, whilst unsubstantiated, this could be because they tend to make the decisions they know their parents will agree with. If this is the case, my research has not definitively captured this dynamic in the accounts shared with me. However, the possibility of making the decision a parent wants, due to a sense of debt or guilt, was mentioned in speculative comments made by one of the participants. Leading on from this, all of the participants have limited mobility, with some severely so, and their independence is differently lived than with more mobile children and young people. They cannot reject their parents' help and must receive that care in order to survive; were their disability less severe, different results, including more discord with parents may have been uncovered in the interviews.

My findings apply quite specifically to those with a progressive, degenerative condition and may not apply to those with a more stable, treatable diagnosis, although there are some points that could usefully be applied to other disabled children, young people and adults.

The way parents spoke to me, what they said to me and the way they spoke of and to their child provided valuable background information, it was apparent that some families were very protective, demonstrating how they managed access to their child (Carnevale et al. 2006). This, however, was not the focus of my research and so these observations, whilst informative to me, could not be included in this thesis, although they have helped to create contextualised insights that have informed this research.

There was much that was said by the parents I met with that would merit further

analysis; the parents live with an accelerated version of the life cycle as they raise their child, and they must face the fact they could outlive their child. They live, therefore, in ways that may be considered marginal and which are overshadowed by the probability that they will lose their child too soon, but it is also apparent that there are ways to live with loss, making lives that are different, and where hope is still present (Condin 2002; Samson et al. 2009). In this creative negotiation there is much to learn, the media and popular discourse too readily speak of disabled childhoods as damaged and parents as selfless. But in living outside of the norms the families demonstrate that they can live with different valuations of what counts as important, and where the condition does not define them or their child.

Future research could explore what young adults with DMD/MD think about their childhoods using retrospective accounts; these may unearth more complex explanations than were apparent in my interviews. This has, until recently, been a limited option due to the early death of most boys with DMD, and there is now more scope to explore this line of enquiry.

This research has contributed valuable, if at times subtle knowledge that extends beyond medical research questions; it has explored how cooperation, adaptation and familial ties can work in challenging circumstances. It has shown how children and young people are capable of debate and speculative discussion as morally engaged agents. My findings move beyond ethical debates about competence and capacity and into the realm of empirical research, asking disabled children and young people what they think and feel and offering vital contributions to the overall endeavour of social research.

## Appendix A Interview schedule

### ***Heading 1: What factors from the boys' lived experience influence their thoughts and decisions regarding participation in medical research?***

1. Can you tell me about a typical day and/or week?- what do you do?

- What are you interested in/good at?
- What's next at school/college, or interests?
- How do you spend your free time? & who with eg friends, siblings, parent?
- Do you get out much with them?
- Could you tell me a bit more about that?
- Are you involved in much for boys with DMD? Such as; can you describe that?
- Or do you tend to hang out with other friends? Could you tell me a little more?

2. Are you/have you been involved in any social group(s)?

- Wheelchair sports and activities, IT, DMD groups, drama, scouts, church?
- Can you tell me more about it?

3. What medical appointments do you go to most?

- Could you describe a typical app, who do you see at them? Who comes with you? eg mum &/or dad, siblings?
- Who seems to do most of the talking?
- Can you give me an example of that?
- If you want to ask a question or speak up about something that's bothering you, do you feel that you can?
- And do mum/dad ask questions much?
- What can be good about apps? helpful, friendly?
- What's not so good? boring, injections etc?
- Do you like the idea of having some apps without a parent being in the room?
- Is it something you've experienced? ...
- Some boys have said they feel left out of conversations at apps, what do you think?
- Is it ever hard to understand what the doctor/nurse has said?
- How do you deal with that?

***Heading 2: In what ways does the boys' family, social & cultural setting shape their attitudes to participating in MR?***

4.a. Do you go to DMD groups/events etc?

...and do your parents? What about your sibs?

- Are some groups just for you?
- What kinds of things are talked about?
- Is MR talked about? ... Such as?

4.b. If you don't go to meetings are you interested in hearing about DMD, or not too bothered?

- And your parents, do they keep up to date with any news on DMD? ... Where from?

5. Do you take part in fundraising activities for DMD? ... Such as?

- What kinds of things is the money for?
- Can you describe an event?
- Did you know what that event was raising money for?
- Was it good to be involved? ... Why?
- Were your family and/or friends there too?
- Are your parents interested in fundraising?
- What would they like funds to go towards?

- *How come you don't take part?*

- Not interested, too busy?
- & your parents thoughts on fundraising?

6. If you wanted to find out something about DMD where would you look for info?

- Do you ever find things out on your own?
- Where from? What like?
- Do you find out things that you hadn't heard elsewhere? ... Is it useful to you?
- Would you talk about that with anyone? ... Such as your parents/other? ...Why?
- Does anyone in the family keep an eye on DMD information? ... Such as?



Where from? Is it interesting?

- Can it be useful talking about it?

***Heading 3: What key processes & interactions play a part in how the boys consider or decide to participate in MR?***

7. As this project is about MR could you tell me in your own words what you think it is about?

- Have you ever thought about taking part in MR and decided not to?

- How come you decided not to?.. Parents/risk?

- What sort of MR might you consider ?

- Have you taken part in any MR?

- How did you hear about it?

- Who explained the MR to you? How did they explain it to you?

- Did that help, was it clear?

- Did you ask any questions? Such as?

- And did your parents? ... Risk/safety?

- Did you ask anyone else what they thought?

- Did you look up any information?

- Was it easy to make up your mind?

- Could you tell me anything more about the experience in your own words?

- Was it how you thought it would be?

8. If you were thinking about taking part in some new MR would it be good to have your parents help to make up your mind?

- Thinking about how you made your mind up to take part in this project, did you discuss it?

- Who with? How did you make up your mind?

- Thinking about MR, would you like to make your own decisions? ... or with parents?

- Imagine someone was making a MR decision;

their parents were keen but the boy isn't sure, what do you think might help everyone decide?

- Do you think boys should be able to make the final decision to take part in MR?

or ... Should it be a joint decision?

**Heading 4: What resources are significant to the boys when considering MR?**

9. Do you talk about MR much?

- Who do you chat with most? Is it helpful?
- What kinds of things might you talk about?
- Do you have someone you tend to trust or listen to more? ... Why them?
- Is there someone in the family who is interested in MR?
- Do you talk with them?
- Are there things you would like to talk about or ask about if you had the chance?

10. If you wanted to find out more information about MR who might you ask?

a parent, nurse, doctor? Why them?

- What sorts of things might you ask?
- How might you approach them... on your own, or with a parent or others help?
- Do you have a group or meeting where you could ask some questions? Such as?

11. How do you keep informed or hear about things to do with MR?

- Can you tell me a bit more about the kinds of things you hear or know about it?
  - Is it something that interests you?
  - Is it easy for you to find out things for yourself? ... Tell me a bit more.
  - Do you get enough time to go on a computer on your own?
  - Do you think boys with DMD should get involved in MR? ...
  - If you were approached to participate in some new MR, might you think about it? .
  - What might make you decide?
  - Some people say taking part helps them feel they're doing something? Do you?
- Is there anything you would like to add? or ask me?

## Appendix B Ice-breakers

- Opening questions and informal chat with the boys will be generated by picking up ideas from the home environment such as;  
Posters, computer games and consoles, books, chess boards, pets, etc.
- Other possible questions may cover music, television programmes, films, pets, social networking, gaming sites and sports interests.
- This will lead into covering some informal demographics such as...
  - Tell me a bit about your family, about your siblings, and your mum and dad, how many people live here?
  - Tell me about mum and dad? What are they good at? How do they spend a normal day? Do they work? Do they have any hobbies, interests or skills you can tell me about?
  - Who is at home the most, eg mum or dad? Does anyone come in to help them with running the house and looking after you all?
  - Do you see other family members such as cousins? Grandparents? Do you get to see much of them?
- Do you think of your family as religious? Do you attend a church or mosque etc?
- Tell me about your home, what's good about it? What's not so good about it? Do you have your own room? Do your siblings have their own rooms?
- What about where you live? Do you like it here? Can you describe it a little? E.g. any local shops, transport links, neighbours, schools and colleges close by? Do friends live nearby? What's good about where you live? What could be better about it do you think?
- Transport issues e.g. How do you get out and about? Car? Taxi?

## Appendix C Scenario 1

At the moment, new drugs and treatments are being developed that may help boys with DMD, and these drugs and treatments need to be tested out on the boys so that doctors and scientists can work out if they do make a difference. As part of this it is useful to understand what boys and young men with DMD think about how the research is carried out, how they might find it helpful to have it explained to them and what is important to them when they think about medical research and the possibility of taking part in it.

So today I want to explore your thoughts on medical research and to do that I want to use one or two imaginary situations or scenarios to get your imagination working. Remember, there are no right or wrong answers; I want to hear what you have to say, you are the expert here, and you can say whatever you want to as we are talking.

“I want you to imagine a boy with Duchenne is at the hospital for a routine check-up and a nurse asks the boy and his parents if they would be interested in taking part in some medical research. The nurse explains the research would involve having an injection every week for 24 weeks at the hospital. At the start and then again at the end of their involvement in the research, a very small piece of muscle would be taken so that doctors could look at it under a microscope to see if there is any change or improvement in the muscle fibre. Some of the boys would get the real drug that is being tested out and some boys would get a ‘placebo’, which means that the injection would not have any of the test-drug in it. So, the boys are being asked, with their parents’ help, to decide if they want to take part in some research where they might get the drug that is being tested out, or they might not. The research will help doctors to find out if the drug does make a difference to muscle tissue or not, as doctors are still not sure if the drug really is effective.”

- Imagine the boy in the scenario was a bit younger than you and he came to you for advice, what might you suggest he does as he tries to make up his mind? Prompts: Talks to his parents? or a doctor? Does some research on his own? Other ideas?
- Imagine now that you were that boy/young man; who might you want to ask some questions or discuss it with? Prompts: Parent/ nurse/doctor/family? Why them?
- What sort of questions do you think you might want to ask? Prompts: Is it dangerous, could it harm you, could it help you?
- What do you think your parents' thoughts might be about you taking part? Prompts: Risk, time issues, helping others, hassle?
- Would it bother you that you wouldn't know if you were getting the real drug or the 'placebo', the false drug? Prompts: What if you found out at the end of the 24 weeks of injections that you had been getting the 'placebo', the false drug?
- What would you find helpful as you think it through and make up your mind? Prompts: Leaflets to take home and look at? A discussion with an expert such as a doctor/nurse? A video to watch, useful websites, chat with someone who has taken part in similar research? Mum/dad and family's thoughts on it?
- Do you think it's good to be involved in research even if it brings no benefit to you?
- Any other thoughts or comments?



## Scenario 2



“I want you to imagine that a friend you know who has DMD is being encouraged by his parents to take part in some medical research. In the research, he would be asked to have four scans, a type of X-Ray that takes pictures of the inside of the body, he would have one scan a week for a month, the first two scans would also involve him having dye injected into his arm, which helps to show up his internal organs when doctors look at the images of his body on their computer screen.

The boy’s mum and dad are keen for him to go ahead with the research even though it will not help to improve the boy’s health, they think it is important to help doctors in any way that they can. The boy is not so sure, he wants to help doctors learn all they can about Duchenne and how boys with Duchenne cope with having scans, but he is not so sure about having the dye injected into his arm, he has been told that very occasionally people can be allergic to the dye. He is also not sure about having to spend time hanging around at the hospital, it might be boring; his mum and dad have promised to buy him a new X-Box as a gift if he agrees to take part.”

- What advice might you, as his friend give to him?
- Who else do you think he could talk to for advice? Prompts: His doctor, someone you know who is helpful and knowledgeable, a charity organisation or other group?
- If it was you thinking about having the scans, what might your own thoughts be? Prompts: Risks, helping doctors, helping others, don’t want to spend more time at hospital?
- Do you think it is important for the boy to be able to make up his own mind about taking part? Prompts: Why? Why not?
- In your own opinion, thinking about this imaginary friend, what should be the most important things he thinks about as he makes up his mind? Prompts: His own choices... doing what’s right for him? His parent’s choices? Helping the doctors by agreeing to take part? Doing something useful?

- What might you suggest he does if he is torn between going ahead with the scans as he really wants an X-Box or turning down his parents offer as he just doesn't want to take part? Prompts: What sort of issues could you discuss with him that might help? Why might those issues be important to him? Why are those issues important to you?
- Do you think it's good to be involved in research even if it brings no benefit to you?
- Any other suggestions or ideas you have thought about that we haven't covered here?

#### **Other talking points**

- Some boys are happy for mum and dad to do all the talking at hospital appointments... are you like that? *Prompts: Why are/aren't you happy for them to do the talking? – should boys speak up for themselves more? – Why/why not?*
- If you could tell your doctors how you feel about hospital appointments what would you want them to know? *Prompts: What's good? - What's not so good? - How could they be better?*



## Appendix D



### **Consent Form: Competent minors and over 16's**

#### **Research decisions: Living with Duchenne Muscular Dystrophy**

**Researcher:** Sarah Skyrme.

**Supervisors:** Dr. Janice McLaughlin, Dr. Simon Woods. Newcastle University

**Please circle as appropriate:**

- |  |          |
|--|----------|
| Have you read the information sheet?   | Yes / No |
| Have you been given a copy to keep?  | Yes / No |
| Have you had an opportunity to ask questions and discuss the study?  | Yes / No |
| Have you received answers to all your questions?   | Yes / No |
| Have you received enough information about the study?  | Yes / No |
| Do you understand that you are free to withdraw from the study at any time, without giving a reason for withdrawing .          | Yes / No |
| Are you happy for interviews to be audio-recorded and later transcribed?   | Yes / No |
| Have you been informed that the recordings will be destroyed at the end of the project?  | Yes/No   |
| If you do not wish the interview to be recorded are you happy to have notes taken?   | Yes/No   |
| Would you like to receive copies of your transcripts?  | Yes /No  |
| Do you agree to your personal data being stored on a secure database that only Sarah Skyrme can access?                        | Yes / No |
| Do you agree to take part in this study?   | Yes / No |
| Do you agree that research data which has already been collected can be used if you lose capacity to consent during the study? | Yes/No   |
| Do you understand that if you make a disclosure of risk of harm, the interview may be stopped and appropriate steps taken.     | Yes/No   |

I understand that relevant sections of my medical notes and data collected during the study, may be looked at by individuals from Newcastle University from regulatory



authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.

Yes/No

NAME IN BLOCK CAPITALS .....

Signature..... Date.....

Signature of researcher..... Date.....

## Appendix D



### Parent/Guardian Consent Form

#### Research decisions: Living with Duchenne Muscular Dystrophy

**Researcher:** Sarah Skyrme

**Supervisors:** Janice McLaughlin, Simon Woods. Newcastle University

**Please circle as appropriate:**

- |  |          |
|--|----------|
| Have you and your child read the information sheet?  | Yes / No |
| Have you been given a copy to keep?  | Yes / No |
| Have you had an opportunity to ask questions and discuss the study?  | Yes / No |
| Have you received satisfactory answers to all your questions?  | Yes / No |
| Have you received enough information about the study?  | Yes / No |
| Do you understand that you are free to withdraw your child from the study at any time, without giving a reason for withdrawing ?       | Yes / No |
| Are you happy for interviews to be audio-recorded and later transcribed?   | Yes / No |
| Have you been informed that the recordings will be destroyed at the end of the project   | Yes/No   |
| If you do not wish the interview to be recorded are you happy to have notes taken?   | Yes/No   |
| Do you agree to your child's personal data being stored on a secure database? that only Sarah Skyrme can access?                       | Yes / No |
| On behalf of your child do you agree for them to take part in this study?  | Yes / No |
| Do you agree that research data which has already been collected can be used if your child loses capacity to consent during the study? | Yes/No   |
| Do you understand that if your child makes a disclosure of risk of harm, the interview may be stopped and appropriate steps taken.     | Yes/No   |

I understand that relevant sections of my child's medical notes and data collected

during the study, may be looked at by individuals from Newcastle University from regulatory authorities or from the NHS Trust, where it is relevant to my child's taking part in this research. I give permission for these individuals to have access to my child's records.

Yes/No

NAME IN BLOCK CAPITALS .....

Signature..... Date.....

Signature of researcher.....

**Appendix D**



**Assent Form**

**Research decisions: Living with Duchenne muscular dystrophy**

**Researcher:** Sarah Skyrme.

**Supervisors:** Dr. Janice McLaughlin, Dr. Simon Woods. Newcastle University.

**Please delete as appropriate:**

- Have you read the information sheet? Yes / No
  - Have you been given a copy to keep? Yes / No
  - Have you been able to ask questions about the interview? Yes / No
  - Do you need any more information about the interview? Yes / No
  - Do you understand that you can stop being in the study at any time, without having to say why? Yes / No
  - Are you happy for interviews to be digitally recorded and then typed out? Yes / No
  - Have you been told that the recordings will be destroyed at the end of the project Yes/No
  - If you do not wish the interview to be recorded are you happy to have notes taken? Yes/No
  - Would you like to receive copies of your interview? Yes / No
  - Are you willing to take part in this study? Yes / No
  - Do you understand that if you said something about being hurt or in danger the interview may have to be stopped? Yes/No
- I understand that relevant sections of my medical notes and data collected during the study, may be looked at by individuals from Newcastle University from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.
- Yes/No

NAME IN BLOCK CAPITALS .....

Signature..... Date.....

Signature of researcher..... Date.....

## Appendix E

### 2<sup>nd</sup> Interview Questions

#### 1: Quote from first interview:

S: I know some boys have said that they feel left out (at the hospital), like the doctor's just talking to the parents,

R: I don't really cos I usually talk myself as well,

S: Really? That's really good,

R: Yeah cos they usually talk to me instead of my parents,

S: Right, is that something then that your mum has encouraged, or,

R: No, just as I just grow older I think, probably need to be heard or something,

#### **New question**

You mentioned in the quote above you feel you need to be heard at the hospital, I'd like to know more about why you think that. Eg... Is it important to you that doctors hear your own thoughts?

Why is that important as you get older?

#### 2: Quote from first interview:

S: How would you feel, as you mature, about having appointments on your own without a parent?

R: No I don't think I've done that yet actually,

S: Is that something maybe you could imagine when you're a teenage or?

R: Yeah probably, I think so,

S: And why would you perhaps think that might be a good idea?

R: I don't know, probably saves your parents' time and stuff, and probably you feel more confident about doing it cos you're more independent

#### **New question**

Is it important to you to be independent?

Why is that?

What does independence mean to you?

Would you like to be able to make your own decisions about the drugs and treatments you have when you are older? Why?

Would you still want to discuss decisions with your mum?

#### 3: Quote from first interview:

S: Do you ever find it hard to understand what the doctor or nurse is saying?

R: Not really, no,

S: They kind of speak in a way you

R: Not really, I just know most of the things, my mum's really good at like language and stuff like speaking and reading and stuff, so I read quite a lot myself so I understand quite a few words, it's a bit easier for me to understand them,

#### **New question**

Mum is good at languages & speaking, is it helpful to have such a useful mum?

How do you think you would manage if mum was not so helpful?

4: Quote from first interview:

S: So do you go to any groups specifically for people with Duchenne?

R: Not really no, because I usually like being around able-bodied people,

S: Uhum, yeah I can understand that, can you expand on that a bit more, why would that be?

R: Well I don't know really, some people with a disability you can't really speak to can you... I like it when it's kind of mixed, like at my school there's loads of people with loads of different disabilities and they mix together with everyone else... I know quite a few people at school like XX and XX who's got DMD, yeah loads of people with DMD,

**New question**

I just wanted to check, do you mean that some people with a disability are quieter or have less to say?

Or did you mean perhaps that they have trouble speaking & communicating?

Maybe you could just expand on what you meant for me.

5: Quote from first interview:

S: So... moving on and thinking about DMD, if you wanted to find out something about Duchenne where would you look for information?

R: I don't know, probably off my mum and stuff because she's known quite a lot of it, cos I know all there is to know about it now,

S: So you'd maybe use your, well you know everything there is but you'd use your mum,

R: Yeah I have most of the time, it's only this year I found all of it out so,

**New question**

Tell me how you got interested in finding out more about your condition.

How have you gone about finding out more?

Has this been helpful to you? Why is that?

Do you feel it's good to know as much as possible?

6: Quote from first interview:

S: And who in the family, I think I know this answer, keeps an eye on things to do with DMD?

R: Yeah mainly my mum,

**New question**

What if mum was not so helpful, how would you manage then?

What do you think it would be like for you if you did not have family support?

7: Quote from first interview:

S: So if you had a child with Duchenne how would you manage the whole giving him information?

R: I think I'd do quite well actually, cos if like I've got it as well so I can speak to him about like it's not that bad and everything, and you still have the same quality of life as a person who can walk so,

**New question**

If you had a son with DMD how would you help him make a decision about taking part in

medical research?

What would your thoughts and concerns be? How might you as a parent find out more information?

8: Quote from first interview:

S: Right, so what questions as you're kind of ... thinking this through and making that final decision (about taking part in a clinical trial), what things are coming up into your mind?

R: Well I don't know I just think it might help other people and maybe myself and stuff, to, yeah to try and help,

**New question**

What if the trial was to test a piece of equipment like a new foot splint or a useful gadget, would you be interested in taking part in that?

What do you think research should focus most on between cure and treatments aimed at helping quality of life?

9: Quote from first interview:

S: So thinking about taking this possible new drug would you be concerned about risks with that, would you want to investigate?

R: Probably, I'm not really that concerned about too many,

S: Right, that's interesting, if you had any questions about this drug that you're thinking of taking how would you get those questions answered?

R: Probably if I looked deeper into it, contact the person who sent us that first to get more, to get further information on it,

**New question**

Have you made a decision or done some more thinking about taking part?

If so could you tell me a bit more about that, what have you been discussing, what questions have you had?

10: Quote from first interview:

S: So what's mum said about it so far, how does she feel about it?

R: She feels it'd be quite a good opportunity,

**New question**

You mentioned mum thinks it's a good opportunity, what if you decided not to take part, would you feel you could tell mum that?

Why?

11: Quote from first interview:

S: Do you think it's good to be involved in research even if it brings no physical benefit to you?

R: Yeah, I still think it would be important,

S: Right, just expand on that a bit more, even though I know you've said it already,

R: I don't know, just be just important to let them know what it does, that it can have different effects on different people, you just have to...

**New question**

As you said, drugs can work differently on different people, what if the side effects were

unpleasant, what might you decide to do?  
Eg who might you talk to for advice?

**12: Quote from first interview:** (We are discussing a scenario where a boy is being encouraged by his parents to take part in some medical research, the boy in the scenario is not so sure)

S: Do you think it's important for the boy to make up his own mind about taking part?

R: Yeah, quite important,

S: Why?

R: I don't know it's kind of well, up to him, he's not being forced to do it but if he really doesn't want to he doesn't have to,

**New question**

What if the parents were being really pushy, do you think that would be wrong?

Do you think the boy should still have some space to make his own decision?

**13: Quote from first interview:**

S: So, again, I think we've kind of covered this but do you think it's good to be involved in research even if it brings no health benefit to you?

R: Yeah, I think it's really important cos other children's lives could be improved by it,

**New question**

You mentioned that taking part in medical research is important, as it could help other children, are you happy to be involved?

Does it ever seem unfair that you are asked to take part when other young people you know don't have to?

Does it feel good to be doing something?

Why is that, could you tell me a little more?

**14: Quote from first interview:**

R: Oh yeah, and coming back to those wheelchairs, I forgot to mention, also this 4 by 4 one that I've got, it's in the garage,

S: Is that the one where the wheels do that (gestures)

R: Yeah they're like pneumatic tyres, that's quite a good wheelchair it also tilts to allow you to get up and down slopes and stuff so it tilts backwards if it's quite steep going down,

**New question**

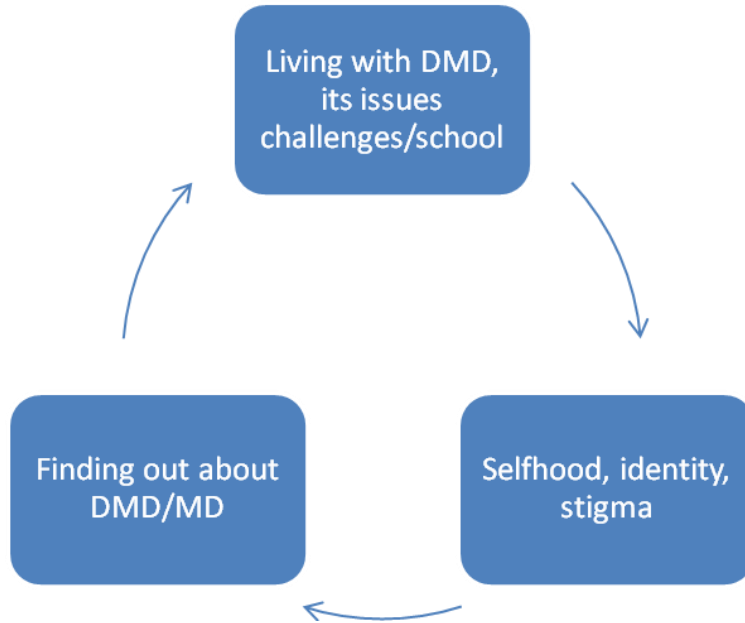
Your wheelchair is important to you, do you think funds from charities should be spent on equipment as well as finding drugs for DMD?

A dad I spoke to recently said that money should only be spent on drugs trials, not equipment, what do you think?

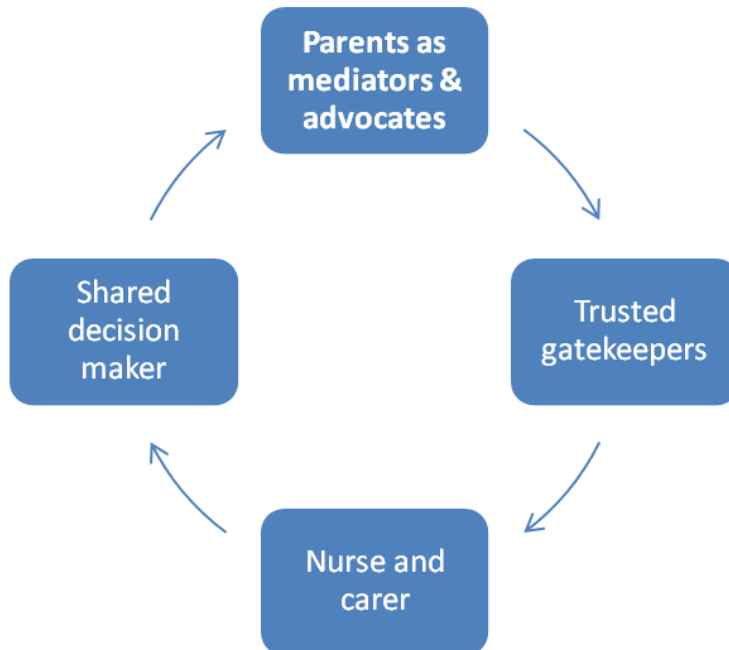


**Appendix F**  
**PowerPoint Themes**

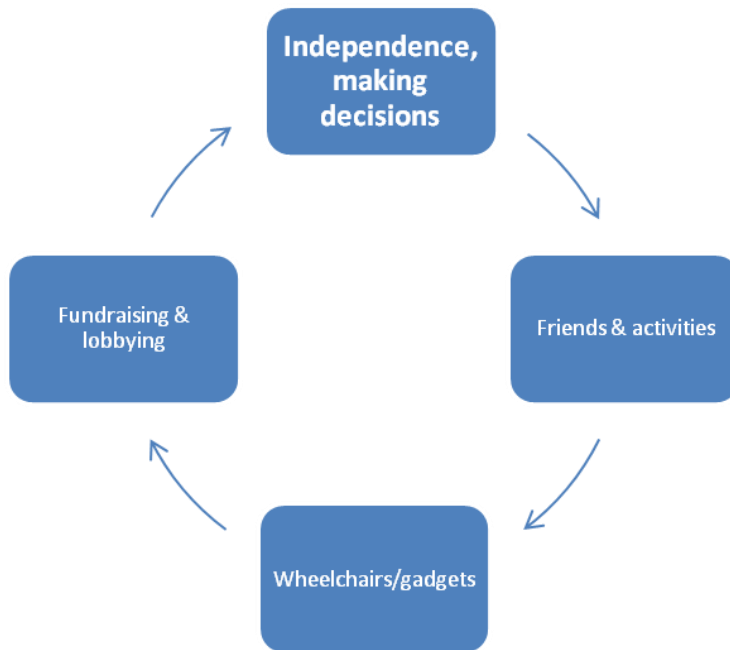
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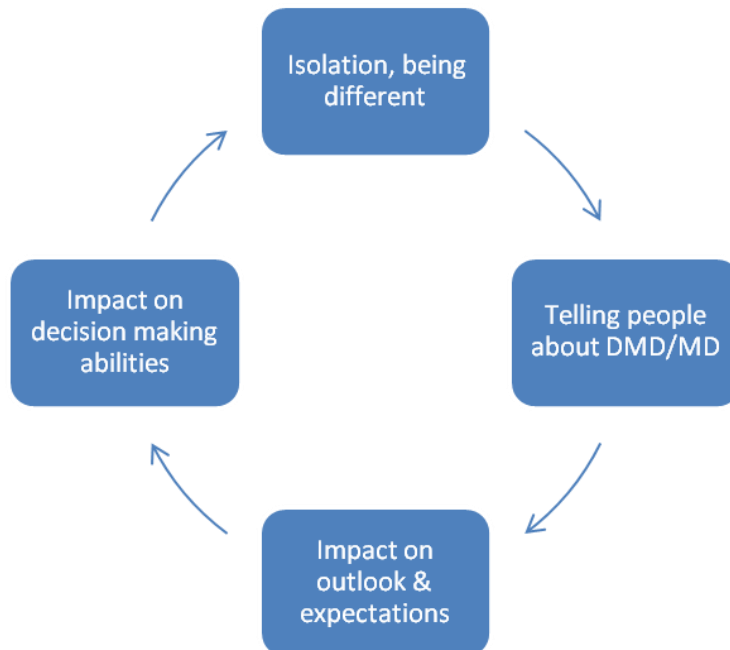
**Parents**



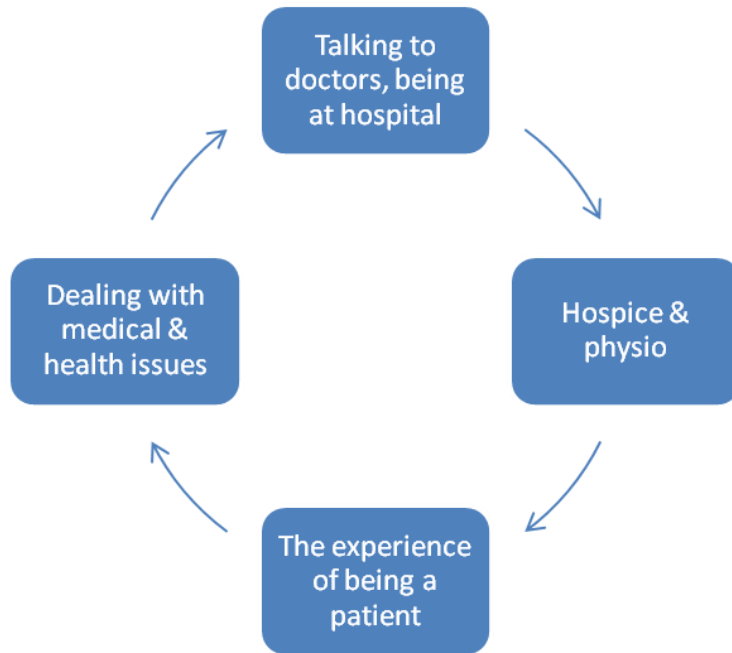
**Independence**



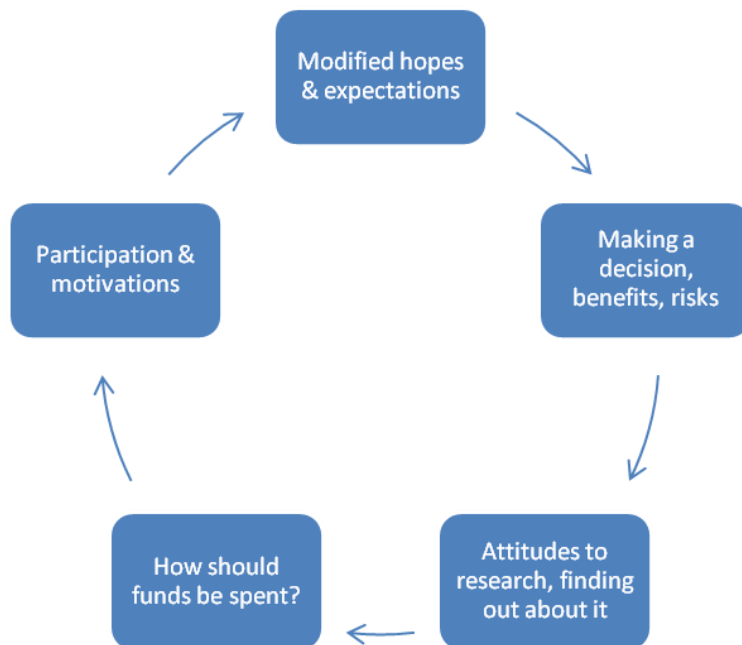
**Isolation**



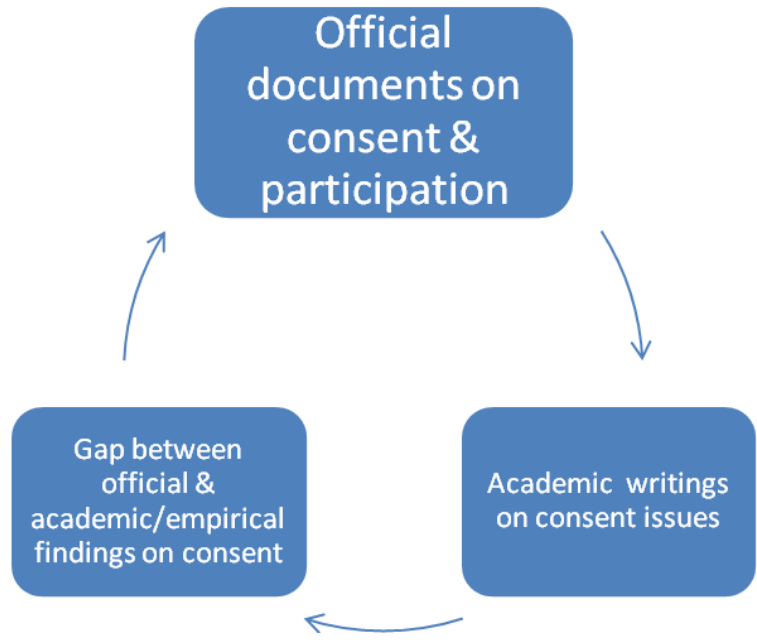
## Doctors and Hospitals



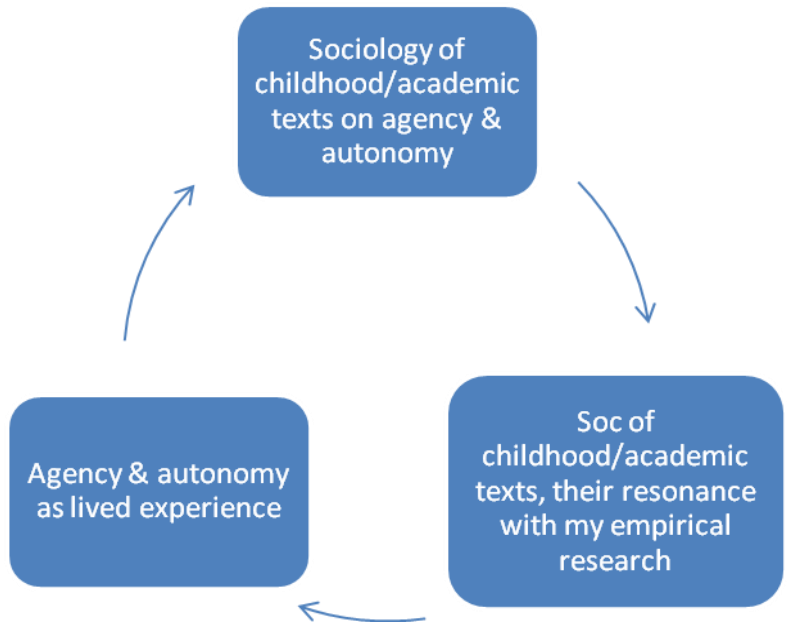
## Research Decisions



**Discourse Analysis**



**Sociology of Childhood**



## Appendix G

### Lone worker protocol

#### Visits to research participant's home

##### **Planning the visit**

- Participant should be informed in advance of the purpose of the visit
- Telephone confirmation of the meeting time and date should occur before travel
- Plan your route especially if the area is not well known to you. Consider parking options in the area.
- Avoid evening visits or late afternoon in winter months even to areas that you normally consider safe.
- Affluent areas attract opportunist thieves

##### **Making the visit**

- Ensure colleagues are aware of your timetable
- Login with Supervisor before commencing the visit
- Keep your mobile phone switched on and within reach at all times
- Only logout with Supervisor when your visit is complete
- If during your visit you encountered problems ensure this is reported to Supervisor
- If you do not log in with the Supervisor, the database of participant's personal details will be accessed and they will be contacted to check whether you have arrived
- If you still cannot be contacted the police will be contacted

**Car safety:** The following guidelines are relevant for any car journey you make during the project

##### **General advice**

- Plan your route and parking options carefully. Always carry a current A-Z area map
- Ensure your car is well maintained and you have sufficient fuel

- Carry a torch for emergencies
- Valuables should be locked in the boot and not kept on you personally. It is preferable not to carry large sums of money, chequebooks and/or credit cards with you unless absolutely necessary
- Lock your car doors and windows whilst driving but especially when you are stationary at a junction or traffic lights. Stay alert when stationary, particularly in areas of notoriety
- During summer months it is preferable to use a sunroof to aid ventilation. Should you have to open your car window remain vigilant – particularly when stationary
- Do not, under any circumstances, pick up anyone in your car even if they are vaguely familiar to you or their cause seems genuine
- Never leave your car inadvertently unlocked or unattended whilst removing items of equipment. It takes seconds for an opportunist thief to remove items from your car – do not make life easier for them!
- Have your keys ready when returning to the car. Avoid the need to search your bag for them

### **Personal safety advice**

- Should you believe you are being followed whilst driving it is recommended that you drive to the nearest Police Station (should you know its location) or alternatively pull into a garage, shop or public house
- Call the Police immediately if you feel unable to drive away safely
- Should a person or persons enter your car whilst you are stationary try to stay calm, sound your horn continuously and switch on your hazard warning lights – hopefully this will attract attention. Shout for assistance as loudly as you can. Never, under any circumstances attempt to chase after the assailant. Remember that despite your anger no items are more valuable than your life! Should the assailant attempt to remove valuable items or goods from your person do not compromise your personal safety by putting up a struggle.

### **In the event of a breakdown**

- Pull off the road and switch on your hazard warning lights
- Use your mobile telephone to contact your breakdown company or car hire

company or Police as appropriate to circumstances

- Stay in your car with all doors and windows locked if safe to do so. Await assistance. Staying in the car on the hard shoulder of a motorway is not considered safe. Walk up the embankment a safe distance and wait
- It is preferable not to agree to assistance from any well-meaning passers-by unless you are totally confident of their intentions

### **In the event of car crime**

- Report this to the Police and ask to be issued with a crime number for insurance purposes
- Report the incident via the NHS Trust Incident Reporting system

### **Nuisance calls/Harassment**

- Under no circumstances, not even those you may deem exceptional are you to issue any participant with your personal mobile or home telephone number. All participants should be advised to contact your work number or research mobile number.
- Keep individual copies of telephone numbers safe to maximise security for yourself and colleagues.
- If contacting a participant from home or on your personal mobile telephone, always prefix the number with the digits 141 thereby ensuring they cannot trace your individual telephone number.

# Appendix H

## Recruitment Materials for Action Duchenne/NHS Clinic/Wheelchair Football



### Participant information sheet for 12-15 year olds

Research project:

#### **Research decisions: Living with Duchenne Muscular Dystrophy**

- Would you be interested in taking part in a research project about Duchenne and medical research?

#### **1. What is the project for?**

My name is Sarah Skyrme and I am a student at Newcastle University. I would really like to hear what you have to say about taking part in medical research and what would help you decide whether you want to take part in it or not. Medical research means tests to try and find out what new drugs or treatments work best for Duchenne.

I have contacted one of your parents or a guardian and have asked them to pass this information on to you.

#### **2. Do I have to take part?**

No you don't! You should think carefully about it and decide if you want to take part or not. Talk about it with your parent or guardian. Even if you agree to take part, and sign a form agreeing, you can change your mind at any time.

#### **3. What will happen if I take part?**

We can decide where to have the interview, we will sit together and I will ask you questions. You can decide if you want your parent or guardian to be there too. The interview will take about 1-2 hours or less if you want and you can choose if it is



recorded. If you prefer I can just take notes during the interview and not use the recorder. A full copy of the notes will be offered to you after I have typed them out. I may want to speak to you a second time but we can decide that later.

#### **4. Why do my thoughts matter?**

This is a chance for boys and young men like you to share their thoughts with other people. It would be good if you could spare some time. The findings will help us to understand what you think about medical research.

#### **5. Will people know who I am if I take part?**

I will not use your real name or let anyone know who you are when I write about the project. The recordings/notes will be destroyed at the end of the research.

#### **6. Is there any time when someone might know what I said?**

If you said something about being hurt or in danger, or that someone else is being hurt then I may need to stop the interview and discuss the issue with other people.

#### **7. What is the next step?**

If you want to take part in the project or ask me for more information then you or your parent or guardian can contact me on:

Email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)

Mobile: 07xxxxxx Thank you for reading this sheet and for thinking about taking part in my project, I hopefully look forward to meeting you, *Sarah Skyrme*.

## Appendix H



### **Participant information sheet 16-21 year olds**

Research project:

#### **Research decisions: Living with Duchenne Muscular Dystrophy**

**Researcher:** Sarah Skyrme.

**Supervisors:** Dr. Janice McLaughlin, Phone:0191 222 7511.

Dr. Simon Woods, Phone: 0191 2083254.

My name is Sarah Skyrme and I am a PhD student at Newcastle University. You are being invited to take part in this research study and it is important that you understand the research project and your part in it. Initially I contacted one of your parents or a guardian and have asked them to pass this information on to you.

The information below explains what the project is about and why you are being asked to take part. Do ask if there is anything that is unclear or about which you want more information, you can phone or email me and I will be happy to help.

You may also want to talk with your parent or guardian about whether to take part or not (to help with this your parent or guardian also has been provided with an information sheet).

1. The aims of the project are:

- To discuss your thoughts and opinions on any research decisions you have had to make, or your thoughts on how you might make a decision to take part in medical research. Medical research means tests and trials to try out new drugs or treatments for Duchenne.
- To understand how making a decision is influenced by people, such as your parents, and by information from the internet and patient support groups.

- There is no 'right' or 'wrong' answer, I would really value any thoughts and opinions you have.

2. Do I have to take part?

You should think carefully about if you want to take part or not, and even if you agree to take part, you can change your mind at any time. I will discuss with you any questions you have and then, before we proceed I will ask you to sign a consent form where you give your agreement to take part in the project, but you can withdraw from the project at any time you want.

3. What will happen if I take part?

I will interview you at a time and place suitable to you and if you wish a parent/guardian can be present. There is no time limit on the interview and if you agree, I will be using a digital audio-recorder, or if you prefer I can just take notes and not use the recorder. A copy of the interview will be offered to you after I have typed it out. I may wish to speak to you a second time but we can decide that later.

4. Will people know who I am if I take part?

All your details and information will be kept private; I will not use your real name when I write out the project. The recordings will be destroyed at the end of the research and I will not discuss your participation with anyone you know.

5. Is there any time when disclosure might be needed?

If you told me something that suggested you or someone else may be at risk of harm, then I would point this out and we would discuss together if the interview should continue and the next steps to take.

6. What will happen to the information from the interview?

The information will be used in my PhD project and will be shared with interested groups and people at conferences and presentations and will be published on the TREAT-NMD website.

7. Who is organising and funding the research?

The research is organised through Newcastle University in partnership with PEALS (Policy, Ethics and Life Sciences) and is funded by the Economic and Social Research Council and TREAT-NMD.

8. What is the next step?

This information should help you decide if you want to take part in the project, you and/or your parent or guardian can contact me for a chat or any more information you need on:

Email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)

Mobile: 07xxxxx

Thank you for reading this sheet and for thinking about taking part in my project, I hopefully look forward to meeting you,

*Sarah Skyrme.*

## Appendix H



### **Parent/Guardian information sheet**

Research project:

#### **Research decisions: Living with Duchenne Muscular Dystrophy**

**Researcher:** Sarah Skyrme.

**Supervisors:** Dr. Janice McLaughlin, Phone: 0191 222 7511

Dr. Simon Woods, Phone: 0191 2083254

My name is Sarah Skyrme and I am a PhD student at Newcastle University. I would like your help in inviting your son to take part in this research project, therefore it is important that both you and your son understand his part in it. The information below explains what the project is about and why I would like your son to take part. Do ask if there is anything that is unclear or about which you want more information, you can phone or email me and I will be happy to help. You will also want to talk with your son about his thoughts on whether or not to take part as it is important he makes his own choice about participating.

1. The aims of the project are:

- To discuss your son's thoughts and opinions on any research decisions he has had to make or, his thoughts on how he might make a decision to take part in medical research. Medical research means tests and trials to try out new drugs or treatments for Duchenne.
- To understand how making a decision is influenced by people, such as you and other family members, and by information from the internet and patient support groups.
- There is no 'right' or 'wrong' answer, I would really value any thoughts and opinions your son has.

2. Does my son have to take part?

It is important that you discuss with your son whether he wants to take part or not, and even if your son agrees to take part, he can change his mind at any time. I will discuss with you any questions you have and then, before we proceed I will ask your son to sign a consent form if he is over 16, or an assent form if under 16. If your son signs an assent form you will also sign a consent form. Your son can withdraw from the project at any time and without giving a reason.

3. What will happen if my son takes part?

I will interview your son at a time and place suitable to you both, we can discuss beforehand if your son would like you to be present or not. There is no time limit on the interview and if your son agrees, I will be using a digital audio-recorder, but if he prefers I can take notes and not use the recorder. A copy of the interview will be offered to your son after I type it out. I may wish to speak to your son a second time but we can decide that later.

4. Will people know my son's identity if he takes part?

All your son's details and information will be kept private; I will not use his real name or identify him when I write out the project. The recordings will be destroyed at the end of the research and I will not discuss your son's participation with anyone he knows.

5. Is there any time when disclosure might be needed?

If your son told me something that suggested he or someone else may be at risk of harm, then I would point this out and we would discuss together if the interview should continue and the appropriate steps to take.

6. What will happen to the information from the interview?

The information will be used in my PhD project and will be shared with interested groups and people at conferences and presentations and will be published on the TREAT-NMD website.

7. Who is organising and funding the research?

The research is organised through Newcastle University in partnership with PEALS (Policy, Ethics and Life Sciences) and is funded by the Economic and Social Research Council and TREAT-NMD.

8. What is the next step?

This information should help you support your son as he decides if he wants to take part in the project, you and your son can contact me for a chat or any more information you need on:

Email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)

Mobile: 07xxxx

Thank you for reading this sheet and for thinking about supporting your son in taking part in my project, I hopefully look forward to meeting you,

*Sarah Skyrme.*

## **Appendix H**

**Letter to parents, sent as hard copy and/or as email (adapted for each recruitment route by the insertion of 'Action Duchenne' 'NHS Clinic' or 'Wheelchair Football')**



### **'Research Decisions: Living with Duchenne Muscular Dystrophy' A request for participants for a PhD research project**

Dear Parent/Guardian,

As the parent of a son with Duchenne Muscular Dystrophy, this brief request has been sent to you by (Action Duchenne/NHS Clinic/Wheelchair Football) on my behalf. My name is Sarah Skyrme and I am a PhD student at Newcastle University, I am currently recruiting participants aged between 12-21 with DMD who might be interested in being interviewed about their thoughts and experiences regarding DMD and medical research. I am writing to ask you if you could discuss the project with your son and give him the opportunity to consider participating in the research.

The project is funded by Treat-NMD and the ESRC and is an exciting opportunity for boys and young men with DMD to speak up and have their voices heard. The findings will help us to understand more about the boys' own part in approaching or making a decision to participate in medical research. The boys do not have to have taken part in any medical research to participate in this project; their thoughts and opinions will still be of great value.

Please find enclosed some materials you can give your son to help him (and you) decide whether I can speak to you both further about his possible participation: these include a poster, leaflet, an information sheet for you and a choice of two age specific information sheets - please would you give your son either the '12-15' or '16-21' Participant Information sheet according to his age. There is also a reply slip with a stamped addressed envelope where you can indicate that you or your son want to know more and how best to contact you (or your son). Alternatively you can contact me on my mobile phone number or by email. If your son is interested in participating



I am most happy to discuss further any questions you or he has, we can do this in person or by phone.

Regards, *Sarah Skyrme*. Sarah's Email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)  
Sarah's mobile: [077XXXX](tel:077XXXX)

**Appendix H**  
**Recruitment Poster**

**Research Decisions:  
Living with Duchenne Muscular Dystrophy**



Are you 12-21 and would you be interested in talking about your experiences of having Duchenne Muscular Dystrophy? Would you like to help us understand what boys and young men with Duchenne think about medical research for Duchenne? By medical research we mean tests and trials to try out new drugs or treatments for Duchenne.

- **It's important that you make up your own mind about if you want to take part in this project, you are free to choose.**
  - **If you do choose to get involved you will be asked to take part in an interview.**
  - **The researcher, Sarah, will sit and talk with you about your thoughts on Duchenne and medical research.**
  - **The interview can be at your house if you like and how long it lasts is up to you.**
  - **Your name and other personal details will not be written down.**
  - **You can choose if the interview is audio recorded or if you just want Sarah to take some written notes.**
  - **You will get a copy of the final report when it is finished.**
- This is your opportunity to have a say and share your thoughts and experiences.**

**If you want to hear more about taking part then please contact me using the enclosed reply slip.**

**Or, you can email, text or phone me using the numbers below. I look forward to hearing from you.**

**Sarah's telephone number: 077XXXX**

**Email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)**

**Web address: <http://research.ncl.ac.uk/researchduchenne>**

**Appendix J**  
**Recruitment information for MDC website**

**Muscular Dystrophy Campaign**

**Research decisions: Living with Muscular Dystrophy**

My name is Sarah Skyrme and I am a Sociology PhD student at Newcastle University, I am interested in interviewing young people aged 12 to 21 who have Muscular Dystrophy. The aims of the project are:

- To discuss your thoughts and opinions on different kinds of medical research - whether you have participated in any or not. By medical research I mean tests and trials to try out new drugs, treatments or therapies for Muscular Dystrophy.
- To understand what might influence your decision to participate in activities such as medical research. For example, other people, such as your parents, or information from the internet, or patient support groups.

There is no 'right' or 'wrong' answer, I would really value any thoughts and opinions you have.

The information will be used in my PhD and will be shared with interested groups at conferences, on websites and in academic papers.

The interview can be at your house if you like and how long it lasts is up to you.

Your name and personal details will not be written down in the finished report. This is your opportunity to have a say and share your thoughts and experiences.

If you want to hear more about taking part then please contact me on my mobile or by email.

- Sarah's mobile number: 077XXXXXX (please leave a message if I don't pick up and I will get back to you)
- Email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)

**Appendix J**  
**Recruitment article for MDC website**

**Newcastle University postgraduate student studies attitudes to medical research amongst young people with muscular dystrophy**

Recruitment is underway for a PhD project being carried out by Sarah Skyrme, she is interviewing young people aged 12-21 who have muscular dystrophy in order to better understand how they think about medical research and how they might approach taking part in it.

The project will contribute to a better understanding of young people's perspectives on taking part in medical research and the findings will be disseminated amongst interested parties such as clinical researchers, patient organisations and young people themselves.

The project is funded by the Economic and Social Research Council, in partnership with Treat-NMD and is based in the Policy, Ethics and Life Sciences Research Centre at Newcastle University.

If you are interested in being interviewed by Sarah please phone: 077XXXX or email: Duchenne.Research@ncl.ac.uk

## **Appendix J**

### **Recruitment material for the MDC website**

#### **Calling young people with muscular dystrophy to participate in a research interview about clinical trials**

*A study is asking people aged 12 to 21 who have muscular dystrophy about their attitudes towards medical research and taking part in it. The results of the project will help to influence how young people who wish to take part in clinical trials are involved in the consent process.*

In this PhD project, researcher Sarah Skyrme aims to interview young people with muscular dystrophy in order to better understand how they would approach making a decision to take part in medical research, and who might influence their decision – for example parents, other family members, peers and charities.

Before taking part in a trial, a doctor, nurse or other researcher has to ask potential participants for their permission. This is called ‘obtaining informed consent’. The process of consent follows strict guidelines to make sure that participants are not pressured into making a decision and they fully understand what they are signing up to.

This research gives young people an opportunity to speak up and express their own thoughts and priorities independent from their parents. The results of the project will *help to influence how young people who wish to take part in clinical trials are involved in the consent process.*

If you decide to take part in this study, Sarah will sit and talk with you about your thoughts on muscular dystrophy and medical research. The interview can be at your house if you like and how long it lasts is up to you. Your name and other personal details will not be written down and you can choose whether the interview is recorded or not. You will get a copy of the final report when it is finished.

The project is funded by the Economic and Social Research Council, in partnership with Treat-NMD and is based in the Policy, Ethics and Life Sciences Research Centre at Newcastle University.

If you are interested in being interviewed by Sarah please phone: 077XXXX or email: [Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)

## Appendix J

### Recruitment material for the MDC 'Trailblazers' website

#### **Have your say about medical research**

A study is asking people aged 12 to 21 who have muscular dystrophy about their attitudes towards medical research and taking part in it. The results of the project will help to influence how young people who wish to take part in clinical trials are involved in the consent process.



In this PhD project, researcher Sarah Skyrme aims to interview young people with muscular dystrophy in order to better understand how they would approach making a decision to take part in medical research, and who might influence their decision - for example parents, other family members, peers and charities.

Before taking part in a trial, a doctor, nurse or other researcher has to ask potential participants for their permission. This is called obtaining 'informed consent'. The process of consent follows strict guidelines to make sure that participants are not pressured into making a decision and they fully understand what they are signing up to.

This research gives young people an opportunity to speak up and express their own thoughts and priorities independent from their parents.



If you decide to take part in this study, Sarah will sit and talk with you about your thoughts on muscular dystrophy and medical research.

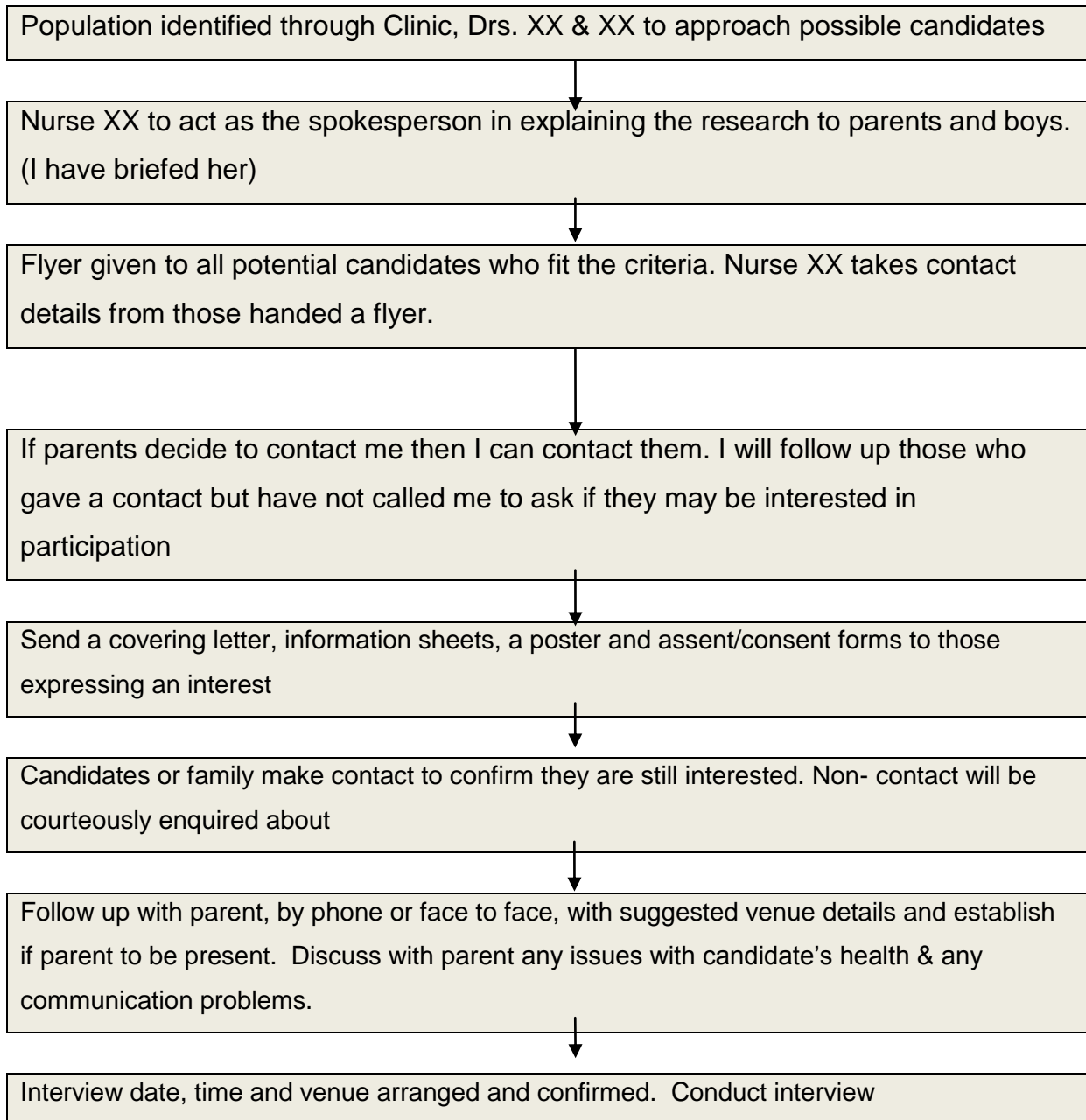
The interview can be at your house if you like and how long it lasts is up to you. Your name and other personal details will not be written down and you can choose whether the interview is recorded or not. You will get a copy of the final report when it is finished.

The project is funded by the Economic and Social Research Council, in partnership with Treat-NMD and is based in the Policy, Ethics and Life Sciences Research Centre at Newcastle University.

**If you are interested in being interviewed by Sarah please phone: 077XXXX or email:**

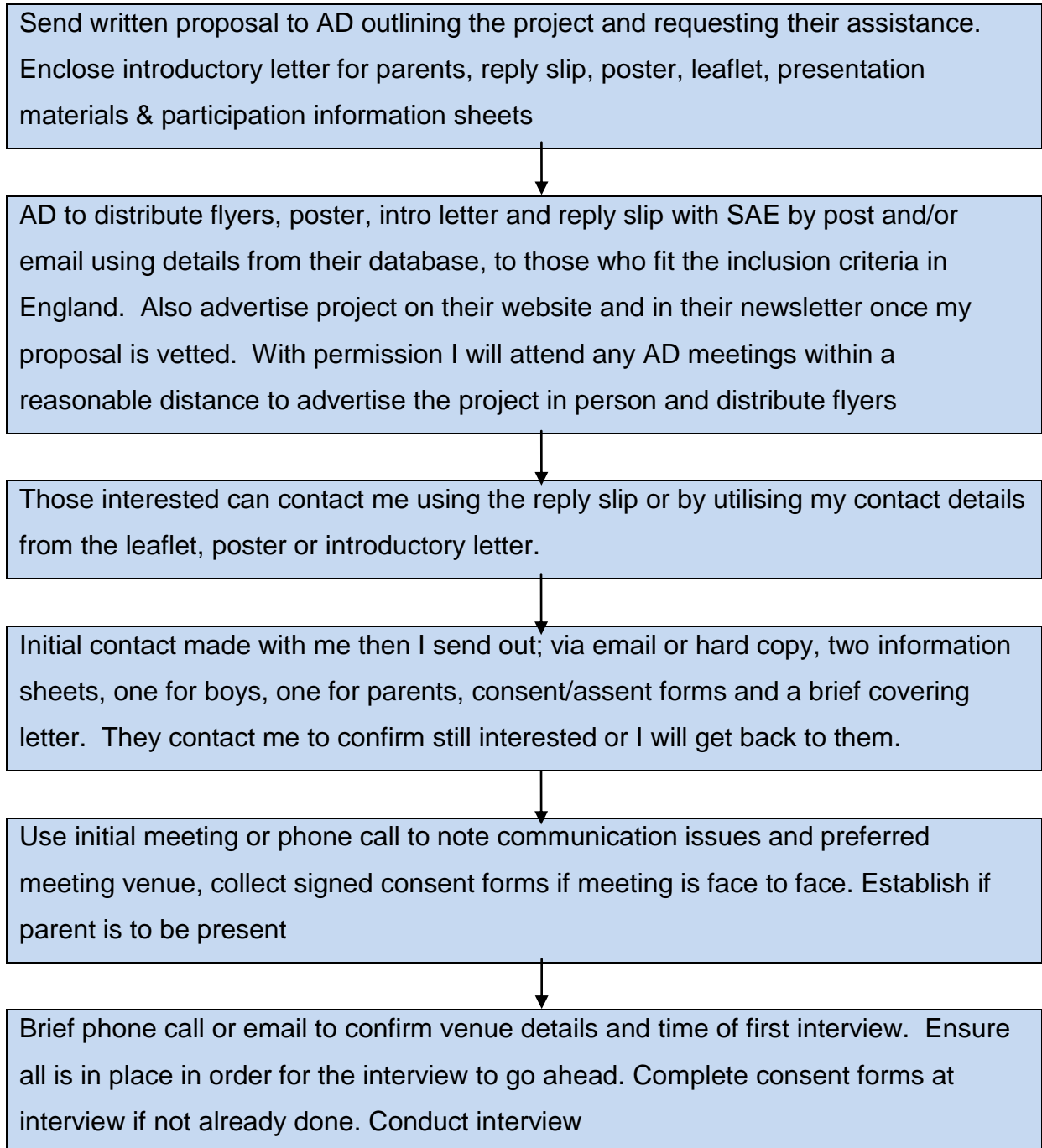
[Duchenne.Research@ncl.ac.uk](mailto:Duchenne.Research@ncl.ac.uk)

**Appendix K**  
**NHS Clinic flow chart (original chart, prior to amendments)**





**Appendix L**  
**Flow chart: Action Duchenne (AD)**



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