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Researching disabled children and young people's views on decision-making: Working reflexively to rethink vulnerability

Introduction

In this article, the aim is to provide a reflexive account of a qualitative research study with children and young people who have Duchenne muscular dystrophy (DMD). The issues that are raised resonate with the main question the research addressed; boys and young men with DMD were interviewed regarding their thoughts on their own participation and decision-making role in medical research. DMD is a muscle-wasting, degenerative condition that mainly affects boys, most need a wheelchair by the age of 12. Respiratory and cardiac problems increase during the late teens, leading to severe disability and an early death (Bushby et al., 2010). Due to the lack of effective treatments for DMD, medical research has mostly focused upon supportive therapies, however, greater knowledge of the causative genetic mutation has led to novel therapeutic research involving clinical trials (Condin, 2014). There may be parents who, understandably, are highly focused on giving their child every chance of taking part in medical research (Woods et al., 2014). Indeed, some parents have taken extreme measures to ensure this by, for example, 'buying in to biotech companies to influence research priorities which will favour their child's condition' (Woods & McCormack, 2013: 249). It is likely there are parents who have focused so intently on the possibility of a 'techno' fix that they have, like some health professionals, overlooked the likelihood that the children and young people have their own priorities and preferences regarding participation in medical research. Bearing these factors in mind, this research study sought to directly explore the

views of boys and young men living with DMD. Pseudonyms are used throughout this article.

Disabled children and young people's participation

The concept of 'vulnerability', whilst contested, may more readily be applied to children, and particularly disabled children who are invited to take part in qualitative research. However, vulnerability, is often regarded as interchangeable with the notion of lacking competence (Carter, 2009), and this has the potential to overlook disabled children's abilities and participatory rights. The systematic positioning of severely disabled children as vulnerable and the discursive tropes the term invokes can impact disabled children's involvement in elements of their health care, decision-making, and their lives more generally. Pertinent to this study, similar concerns are also present regarding children's participation in medical research, wherein research governance can presume children's vulnerability (Nuffield, 2015). Therefore, in this article vulnerability is doubly foregrounded, relating both to disabled children and young people's participation in qualitative research, and medical research. The label of belonging to a 'vulnerable group' can lead to some disabled children being harder to reach and remaining under researched (Carter, 2009; Cocks, 2006). An awareness of the concerns surrounding qualitative research with this 'vulnerable' group meant that the fieldwork was carefully planned and prepared for. Research that relies on accounts provided by parents, doctors and family members is of great value, but it is timely to represent at first hand those with severe disabilities regarding their thoughts on their lives, health care and medical research. Insight provided through qualitative research with disabled children can challenge reductive notions about them, questioning assumptions that they lack competency due to age or disability (Davis & Watson, 2000, Heath et al., 2007). This supports more inclusive

ways of involving them in their care and decision-making, and contributes to broader understandings of disabled children's perspectives.

The Study

Semi-structured interviews were conducted with a total of nine young people aged 10-21 with DMD and one young woman with a less severe but untreatable muscular dystrophy. The young woman's inclusion was prompted by very slow recruitment uptake; however, the focus in this article will be on boys and young men with DMD. Recruitment was implemented via an NHS muscle clinic, two charity organisations and a wheelchair football team. The conceptual framework for the research was informed by the sociology of childhood, which acknowledges that childhood is embedded in social and cultural relationships wherein children are actively involved, rather than passive beings (Prout & James, 1997; Tisdall, 2012). Disability studies also inform the research, challenging the emphasis on disability as an individual, medicalised concern, and focusing on the ways in which disability is socially produced (Hughes & Paterson, 1997). Drawing on work from the sociology of childhood and disability studies, there is a recognition that both children and disabled people can be marginalised by societal attitudes, paternalism, notions of innate vulnerability and dependency (Tisdall, 2012, Connors & Stalker, 2007).

Decision-making

Children's involvement in medical research is considered a vital necessity as the use of medicine for 'children should be guided by the best available evidence of clinical effectiveness... ideally derived from clinical trials conducted with children'

(Department of Health [DH], 2003: 25). This was a starting point for the study as

there is a need to understand the factors influencing children and young people's decisions about taking part in medical research and clinical trials (Broome et al., 2001). Thus far, questions concerning their involvement have mostly been 'debated as ethical or normative principles [but] largely in an empirical vacuum' (Dixon-Woods et al., 2006: 175). Simultaneous to this, children and young people are increasingly encouraged to take an active role in decisions on their health care and treatment (DH, 2003; DH, 2004). For children and young people to be effectively involved during decision-making they should be encouraged to express their views and provided with information that is shared appropriately with them (Alderson, 2007; Lansdown, 2001). Yet their involvement in decisions may be tokenistic (Royal College of Paediatrics & Child Health, 2003; Viper, 2012) and limited by concerns for their protection (Meyer, 2007). Children's involvement can also be constrained if adults fail to explain their illness or treatment needs to them, causing anxiety and fear (Burke, 2010; Drake, 2001). Therefore, a progressive call for greater involvement is circumscribed by worries over safety, doubts about competence, and shaped by the impact of safeguarding failures and scandals in health care (English & Sommerville, 2003; Stalker et al., 2004).

A cautious, protective approach is understandable and it is right that children's best interests are observed; however, precautionary measures can create a culture that is unreflectively restrictive and paternalistic. This does not allow for children's variability and the contextualised nature of individual competence. Competence can be nurtured through the ongoing experience of having an illness or disability (Alderson et al., 2006); when their opinions are taken seriously children acquire the skills to 'develop their thinking and to exercise judgement' (Lansdown, 2001: 12). This skill

may not align with their age because ‘contingencies such as experience and ability can be more salient than age to a child’s competence’ (Alderson, 2007: 2273). The implication is that over-protection and adults’ failure to communicate effectively keeps some children from being fairly represented in the generation of health care research, policy and guidelines. This under-representation can result in ill and disabled children’s marginalisation in their care, in decisions on treatment and their potential involvement in medical research, despite being key stakeholders.

Meaningful involvement

Motivated by the small number of qualitative research studies engaging directly with children and young people with conditions such as DMD, the plan was to hear from participants, and to do so in settings reflecting the relational contexts they live in. It is observed that when children are making decisions these are likely to be made in a family setting (Cave, 2011). Their decisions about treatment and medical research are embedded in social processes not readily apparent to health professionals or regulators overseeing research participation (Schaffer, et al., 2009). Therefore, the intention was to explore the participants’ thoughts on decision-making and to comprehend the contexts that decisions emerge from.

As noted, children’s capabilities may not be age congruent, young children can develop expertise and mature insight on their health needs and the necessities of medication and treatment (Alderson, 2007; Berntsson et al., 2007; Dixon-Woods et al., 1999). Nevertheless, despite a language of inclusion, children with significant support needs are less likely to be meaningfully involved because of assumptions about their ability to participate or make choices (Davis & Watson, 2000; Viper, 2012). Elements of this tension are apparent in a Department of Health document

that describes the training required when communicating with children and:

The need to understand the extent and the limits of children's comprehension at various stages of development (DH, 2003: 16).

If assumptions regarding a child's comprehension are inflexible and are not reassessed in consultation with children, then those considered less competent and/or vulnerable due to age or disability can be marginalised. They may be considered different, with this difference arising 'because the children are judged against supposedly objective criteria' (Davis & Watson, 2000: 214). This limits some disabled children and young people's involvement in decision-making, meaning they have few experiences to draw on when making more significant decisions as they mature (Viper, 2013).

Similar concerns can constrain the way in which health care research is conducted, whereby children perceived to be the 'most vulnerable are most under-represented... even though they are high consumers of services' (Carter, 2009: 859), whilst the least vulnerable (Carter, 2009) are more routinely surveyed. Discourse conflating the need for help with helplessness, physical dependence with overall dependency, and childhood with unmitigated vulnerability is unhelpful.

Reflections on the study

As introduced, it is increasingly expected that ill and disabled children and young people be involved in decisions on their care and treatment, and that paying attention to their views benefits them and challenges assumptions about their competence. Exploring these concerns can generate insight into the lived experience of DMD and the nuanced ways health care and medical research decisions are

managed between children, parents and doctors. Points are raised that intersect and overlap; this study is sociologically focused research exploring how medical research decisions are made. In doing so, questions are posed about the way in which disabled children's frequent positioning as vulnerable may influence how adults engage with them in decision-making processes. Concerns about their status as 'vulnerable participants' also shaped the way this study was planned for. Hence it is relevant to discuss the experience of preparation for ethics approval, and conducting the fieldwork. These experiences have informed the research process and an ongoing engagement with the findings, resonating with concerns expressed by others regarding ethical review, qualitative research with disabled children, and notions of vulnerability and competence (Carter, 2009; Davis & Watson, 2000; Hagger & Woods, 2005; Heath et al., 2007; Halse & Honey 2005; Stalker et al., 2004).

Ethical Review

The proposed study sought to conduct semi-structured interviews with the participants and to explore their thoughts on decision-making. The topics covered were designed to address matters such as children's perceptions of risk when taking experimental drugs and how they might collaborate with parents and doctors to make decisions. These points of concern are timely as boys with DMD who fit the criteria may be invited to participate in medical research or clinical trials. To improve how they are informed about the research they take part in, they can benefit from appropriate information that helps them reach a decision. Exploring how decisions are reached can contribute to discussions on patients' rights to access experimental drugs (Woods & McCormack, 2013), and the hope expressed by DMD patient

groups for the accelerated development and testing of drugs to modify the condition (Franson & Peay, 2013).

Prior to beginning fieldwork, ethical approval was sought from a Research Ethics Committee (REC). This was necessary as some of the participants were recruited through a National Health Service (NHS) muscle clinic. The recruitment materials, including age-appropriate information sheets and consent/assent forms were devised with support from the National Research Ethics Service (NRES) and from colleagues conducting similar research. An interview schedule was produced and disseminated to the REC along with recruitment materials, a lone worker policy and associated paperwork. However, at the initial ethical review, the proposal was given an unfavourable decision; the REC felt that, for reasons of the researcher's safety, interviews would be better conducted at regional centres such as clinics rather than participants' homes. This concern was unexpected, as it prioritised researcher safety at some cost to the participants, through restricting their choice of venue (Daley, 2015). The boys' health, the limitations on their time, and the reliance this would place on parents to transport them seemed burdensome and, perhaps, unrealistic. It was also surprising as it had been expected that issues raised by the REC would focus on participants' welfare, confidentiality, and the researcher's approach when interviewing severely disabled children. This initial decision was appealed and a favourable opinion given by a second REC who acknowledged the suitability of meeting participants in their own homes. The children and young people spend considerable time in venues such as hospitals and clinics; meeting with them in their own environment was a valuable way of appreciating the life they lead beyond the hospital. It also placed the researcher as a visitor in their homes and, temporarily,

their lives (Dingwall, 2006), in a space where the participants had a real sense of belonging (Daley, 2015).

Seeking ethical approval can seem to be a one-sided conversation (Turner & Webb, 2014) and an obstacle to overcome (Balen et al., 2006); reviewers sometimes limit the parameters of research because they fail to accommodate methodologies involving children in inclusive ways (Balen et al., 2006; Skelton, 2008). The need for protection, both for researcher and those being interviewed, is important but there could be a subtler balance between ensuring high standards are in place and measures imposing 'unnecessary restrictions on potentially worthwhile research' (Stalker et al., 2004: 380). Ethical review is anticipatory and prospective; it says what will happen (Hedgecoe, 2012) rather than what does happen in the field. The precautionary attitude emanating from ethics regulation can mean researchers are conservative in their approach (Clavering & McLaughlin, 2010). Furthermore, and wrapped up in the focus of the study, the risk of harm from a qualitative study is not directly comparable with biomedical research (Dingwall, 2006). Nonetheless, the researcher must be extremely thoughtful in their approach; the subject matter is tentative, potentially touching on matters such as the degenerative nature of DMD and loss of mobility. This demands sensitivity to the contexts of an under-researched group not accustomed to having their views sought, and whose wellbeing is of primary importance.

In the field

Those working to develop life-saving drugs for children with untreatable conditions like DMD can benefit from studies that map out some of the factors influencing decisions (Broome et al., 2001). Therefore, the research explored how the participants might approach taking part in medical research and the role familial and sociocultural influences have in that approach. Qualitative research is contingent and however much researchers prepare for a study, what happens on arrival at a busy home may not entirely match the plans (Abbott, 2012). Indeed, there was a disparity between the preparation for ethical review and the experience in the field, which was organic, responsive and dialogic. After the necessities of extensive paperwork (Stalker et al., 2004) and a sense of powerlessness (Turner & Webb, 2014) during preparation for ethical review, connecting with participants and their families shifted the research to an immersive experience. Preparation was replaced with the challenge of gleaning perceptions and nascent data, whilst acting ethically and empathically as children and parents shared highly personal accounts.

Many of the insights parents shared were impromptu, occurring during informal chat over a cup of tea, or in the initial phone call when an interview date was being arranged. Parents readily disclosed concerns for their child's health, commenting on the lack of medical research applicable to their stage of disease progression, and sharing information that was sometimes painful to hear. Most of these comments could not be reported because of their confidential nature, and due to the fact it was the boys who were the focus of the interviews, not the parents. However, their words have proven significant, making a major contribution to the way the research was conducted and is reported on. The parents' words depict the close, supportive

relationships boys and parents have (Skyrme, 2016), these insights were instrumental in informing the analysis. Whilst it was plausible to imagine such encounters might happen, their candour and spontaneity indicate how parents live in the moment, but also how for some there is an ongoing sense of grief at their child's diagnosis and shortened life span.

Qualitative research; negotiated and dynamic

As the fieldwork phase began, the interview schedule was implemented; this had taken many revisions to get 'right' and it contained a range of open-ended questions. Notwithstanding this, it became apparent during initial fieldwork that revision was needed. The questions concerning medical research, such as what the participants thought of it and how they might decide to take part in this research, lacked context; the questions were too remote from their daily lives and experiences. Medical research, whilst making progress, is likely to be too late for the current generation of maturing boys with DMD. Several parents spoke of their endeavours to keep their son as well as possible, whilst accepting that a breakthrough in medicine was a distant hope. The current lack of drugs and therapies for DMD and the related loss of function mean that discussions on a topic which could cause despondency (Condin, 2002; Young et al., 2003) are likely to be limited amongst family members. It was not apparent that the participants themselves were despondent; however, most did not expect a medical discovery that would be relevant to them. When asked if medical research was a subject he spent time thinking about Adam (16) was representative of the general attitude:

There's no point... when there's nothing major happening... Just got to wait for it to happen, wait for them to make another stage [of drug development].

Ollie (14) described how, in the absence of therapies to halt muscle-wasting, he has gradually lost mobility:

I first started going in a wheelchair when I'd probably be about 9 I think... I needed to have loads of equipment at primary [school], I needed to have a chair, I used to have a manual wheelchair and then I went in this [electric chair] so yeah, I might have walked for a bit more but I injured my leg... I did my Achilles so that went and I couldn't walk anymore so that was it.

Meeting with the families face to face provided vital insight, some at a non-verbal level that enabled an emotional and intellectual understanding of life with DMD. Their thoughts on medical research are tempered by the realities of muscle wasting and, in the absence of a scientific miracle, families must cope with daily life and their child's needs (Samson et al., 2009). This early finding called for the development of an approach that was appropriate to the participants. Researchers and ethical review committees should be amenable to the ways in which qualitative research is a negotiated and dynamic dialogue, as well as a co-production of meaning (Abbott, 2012), shaped by the context and uniqueness of each encounter.

Vignettes

The questions in the initial schedule presumed boys with DMD are reasonably well informed about medical research; as explained, this was not the case amongst all the participants. Therefore, vignettes were devised to situate the issues and stimulate constructive discussions. These were mailed out prior to the interview, offering an opportunity for participants to consider their responses; it is suggested this can help reduce some of the power imbalance inherent in the interview setting (Jepson et al., 2015). In the vignettes, participants were asked to imagine a fictional friend with DMD comes to them for advice because he is thinking of taking part in medical research, but is concerned about the risks in taking an experimental drug. In

another vignette, the friend feels he is being coerced into taking part by his parents; in both cases participants were asked how they would advise this 'friend'. This approach invited an imaginary context for expressing thoughts; it also offered some critical distance as a fictional person was being discussed rather than the participant. This enabled a way of exploring medical research decisions, but also, unexpectedly, created a space for the participants to speak about other issues they regarded as important.

Whilst the vignettes worked with most participants, two young men found them less constructive, one 17-year-old commented that it was hard to speculate on an imaginary situation as 'it's a scenario isn't it?'. Likewise, an 18-year-old found the idea of an imaginary friend seeking their advice 'daft' [silly], mainly because he thought the boy should not ask his advice but make his own decision. With these two older boys, direct questions proved to be more appropriate and logical to them, matching their style of communicating. However, a 21-year-old responded well to the imagined settings; hence, researchers must be prepared to act intuitively, working alongside participants to establish effective, shared communication. Overall, vignettes provided a framework within which to explore ideas and follow thoughts through rather than gathering definitive answers. The flexible narrative space created opportunities for participants to discuss key experiences.

Narrative space

With little prompting the participants explained their concerns with some health care experiences and described the vital role parents have in supporting them. Comments included reflections on their experiences of discomfort, surgery, and the limits of

health and social care. Drawing on the participants' imaginative skills (Alderson, 1992) set up a flow of conversation that helped them express their thoughts (Royal College of Nursing, 2011) as they moved between fictional settings and real-life examples. They spoke eloquently about disablist, condescending attitudes among the public and their peers, and explained how they wanted health professionals to speak to them. Watson (2012) suggests that we may need to develop new approaches that allow disabled children to contribute to our research agendas, ensuring 'that we ask children for their perspectives and allow them to identify what it is that give their lives quality' (199). While the study sought to address decision-making, broader matters were spontaneously raised by the participants. The topics they discussed help to contextualise their experiences, addressing the primary question whilst also contributing to understandings that are more complete and nuanced (Watson, 2012).

Challenging vulnerability

The participants' insights construct a subtle understanding of life with DMD, challenging a discourse of disabled children's dependency and vulnerability (Davis & Watson, 2000). Their observations depict how they negotiate a path through their lives, revealing competence to be a way of relating to others and not just an individualised skill (Alderson, 1992). What emerged was how, in the absence of tangible scientific progress, the boys and their parents live and cope to the best of their abilities. For the participants this includes maintaining some independence, giving their lives meaning, and achieving realisable goals (Abbott & Carpenter, 2014; Gibson et al., 2009; Skyrme, 2017). There was not a direct or measurable correlation between age and level of competence and understanding amongst the participants. Rick, aged 12, expressed a preference for making his own decisions and speaking

directly with doctors to gather information on medical research. He understood that a clinical trial was unlikely to be of direct benefit to him, commenting that 'it's just to experiment with what would happen'. Meanwhile an 18-year-old was less clear about the experimental nature of trials, commenting that taking part would be worth it as, 'it might be getting rid of your disease'. Another 18-year-old had not heard of the use of placebo in medical research and commented:

It's a bit cruel making a bunch of kids have injections that's not going to do anything and people know it.

This indicates that children and young people who may be invited to take part in medical research have understandings about research protocol and terminology that are variable and not strictly related to age and stage. Therefore, age-related information must be supplemented with interactive dialogue and activities that ensure children and young people's misapprehensions are clarified. In the process of thinking about taking part in medical research, careful explanation and good quality communication can support decision-making (Nuffield, 2015). Concerns about vulnerability should be responded to by working in partnership with children and engaging with them in the way research is designed and conducted (Nuffield, 2015). The findings, some of which are reported here, contribute to improved knowledge about disabled children and young people's thoughts on involvement in medical research decision-making. They challenge presumptions of disabled children's innate vulnerability, indicating that children, including those facing adversity, can be active in interpreting their worlds and in making choices (Boyden, 2003).

Reflective Practice

Adjustments and revision can be a necessary part of exploratory research, although the additional time and extra funding to support this 'breathing space' may not be built into the research budget. Funded studies can also place an expectation on specific and actionable findings (Abbott, 2012) that, not unreasonably, respond to the research question and minimise accounts of the researcher's non-linear route toward findings. Nevertheless, it is productive to reflect on experiences in the field such as non-verbal interactions, researcher 'hunches' and the balance of remaining a polite guest in homes (Yee & Andrew, 2006) whilst being there for a specific reason. It is observed that children's research benefits from researchers questioning their 'research methods and the academic and personal assumptions that they carry... in to the field' (Davis, 1998: 327). Although these issues may not fit into final reports they are an integral part of many research encounters. Discussion of these issues helps build contextualised understandings on the intersection of disability and childhood in the lives of those who are young and severely disabled.

Understanding childhood disability

Empirical work based on disabled children's own experiences has been less apparent in the sociology of childhood (Moran-Ellis, 2010). Likewise, disabled children have been underrepresented in disability studies (Connors & Stalker, 2007; Watson, 2012). Researching the lived experience and perspectives of disabled children helps avoid the homogenising and decontextualising of children (Brady et al., 2015) and childhood. Issues arising from this study indicate that the way we communicate with those we are researching matters, and that competence is not

static but is situated in sociocultural and infrastructural contexts that together impact lives (Watson et al., 1999). Alderson (1993) discusses how 'children become competent by first being treated as if they are competent' (Alderson 1993: 173).

Therefore, researchers must reflect and practice this through highlighting more empathic and democratising approaches to research participation (Aldridge, 2014).

As Davis and Watson (2000) contend:

Children's rights are intertwined with relationships and anything which enables the establishment and maintenance of empowering relationships, will also act to support the rights of children. (Davis & Watson, 2000)

The constraints in reaching and listening to severely disabled children are contrasted with the necessity of doing so if their perspectives are to be incorporated into social research, health care policy, and medical research guidelines. Their contributions counter the tendency to minimise children's potential for demonstrating judgement and decision-making capabilities. Regarding medical research participation, these capabilities should be recognised and encouraged; they can help researchers ensure children's willing involvement in research that may lead to life-saving therapies (Woods & McCormack, 2013).

Concluding points

Attempts to future-proof children's health and social care, concerns about risk, and scandals in health care all impact the shaping of policy and practice, contributing to categories influencing the embodied lives of children and young people (Brady et al., 2015). Inflexible age and stage based restrictions can miss contextualised abilities;

these abilities can be encouraged, in part, through qualitative research processes that accommodate difference. How we speak about disabled children and young people in our research practice and their representation in policy is important; uncontested notions of vulnerability can contribute to a circular discursive dynamic (Meyer, 2007). As Fisher (2012) observes, vulnerability may be

Regarded as a label that is embedded within discourses of recognition and misrecognition that influence how power is embedded in society. (7)

Unchallenged reductive beliefs and practices marginalise disabled children and young people, misrepresenting their capacity for involvement; yet these individuals have insightful perspectives to share regarding their lived experiences.

To better inform how disabled children and young people are cared for we need to hear from them, developing in-depth understandings of their experiences and values. To hear from them and represent them as faithfully as possible we need to overcome concerns about their lack of competence and the harm we may do them (Carter, 2009; Heath et al., 2007). Concerns about children's vulnerability should not limit their involvement in health research (Carter, 2009), and research that is ethically and socially productive must seek out those who are poorly represented (Fisher, 2012). This representation could, amongst others, include severely disabled children, children living in areas of armed conflict, those experiencing domestic violence, and children in hospital (Boyden, 2003; Overlien, 2017; Stalker et al., 2004). This is likely to involve complex negotiations around access, consent, and researcher and participant safety. Nevertheless, if social research is to be ethically robust and a

morally sound, enabling force that fairly represents those deemed vulnerable, then these are the issues that researchers and their associated institutes must deal with.

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