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'At the end of the day, it is more important that he stays happy': an interpretative phenomenological analysis of people who have a sibling with 22q11.2 deletion syndrome

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Abstract

Background 22q11.2 deletion syndrome (22q11.2DS) is the most common microdeletion syndrome. However, there is little research examining the effect of this multisystem disorder on the family, particularly siblings. The current study was a phenomenological exploration of sense-making in siblings of a person with 22q11.2DS.

Method Interpretative phenomenological analysis informed a detailed and open examination of being a sibling of a person with 22q11.2DS. Using in-depth semistructured interviews, five typically developing siblings (two men, three women) of people with 22q11.2DS were individually interviewed, providing the data set for transcription and thematic analysis.

Results The theme '*They are the priority*' overarched two subordinate themes that emerged from participants' descriptions of the struggle with acceptance and finding positive meaning. Participants oscillated between conflicting feelings about their sibling with 22q11.2DS always taking centre stage. For example, they felt anger, guilt and resentment; yet, they also embraced patience, empathy and gratitude.

Conclusions This phenomenological study provides a foundation for future research relating to 22q11.2DS and fostering family wellbeing, particularly around acceptance and psychological growth. The siblings in this study actively withdrew from their family to allow prioritisation of their affected sibling. However, this does not mean that their needs should be overlooked. There are easily accessible resources to support siblings of individuals with disabilities, and it is important for health professionals and parents to consider these options.

Keywords acceptance, disability, interpretative phenomenological analysis, siblings, velocardiofacial syndrome

Introduction

Growing up with a sibling who has a developmental disability can be challenging. It has been suggested that typically developing siblings of children with developmental disabilities are at an increased risk of adjustment problems (Summers *et al.* 1994) including behavioural (Verte *et al.* 2003) and social problems (Constantino *et al.* 2006). On the other hand, there is

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also evidence of good adjustment and resilience amongst many siblings (e.g. Green 2013). Whilst there are many influences on sibling outcomes, there is evidence that siblings' experiences are influenced by the type of disability their sibling has (e.g. Petalas *et al.* 2009). The 22q11.2 deletion syndrome (22q11.2DS, also known as velocardiofacial syndrome) is a disorder encompassing physical, intellectual and behavioural symptoms, yet there is little research regarding the impact the syndrome has on the family system, particularly the siblings. The present study aims to explore the 'lived' experience of being a sibling of a person with 22q11.2DS, with a particular focus on positive and negative subjective interpretations of the unique phenomenon from the siblings' perspective.

22q11.2 deletion syndrome is a complex microdeletion syndrome associated with more than 180 features, such as heart defects and palatal anomalies (McDonald-McGinn *et al.* 1999). The vast majority of people with 22q11.2DS have impairments of intellectual and cognitive functioning. There is also an increased risk of psychiatric disorders such as autism (Fine *et al.* 2005), anxiety (Fung *et al.* 2010), depression (Green *et al.* 2009) and psychosis (Murphy *et al.* 1999). Recent research suggests that the syndrome may occur as often as 1 in 992 births (Grati *et al.* 2015). This, coupled with the low professional and public awareness of the condition, highlights a need to identify the impact, both positive and negative, of 22q11.2DS on the family. The complexity of the phenotype in 22q11.2DS and the chronic nature of many symptoms are likely to pose unique challenges for typically developing siblings.

To date, not much is known about siblings of people with 22q11.2DS. However, a recent study by Okashah and colleagues (Okashah *et al.* 2015) explored how much adolescent siblings knew about the syndrome and also how they perceived the impact of the syndrome on themselves and their affected siblings. The findings indicate clear variability in experiences reported, with some respondents suggesting that 22q11.2DS had changed their life (e.g. less attention from parents), whereas others reported no difference (e.g. their sibling would still be the same without 22q11.2DS). The participants reported that if their sibling did not have 22q11.2DS, they would feel less worried, stressed and guilty; yet, on the other hand, they also reported that they would be less compassionate (Okashah *et al.* 2015). Therefore, although siblings are likely to struggle at times, there is recognition of positive self-change. The experiences of these siblings relate to Folkman's (1997) theory of meaning-based coping, whereby negative psychological states associated with stress can motivate people to create positive psychological states in order to gain relief.

Positive family support may foster this meaning-based coping. Parent and family factors (e.g. socioeconomic status, parental stress and family communication) affect adjustment of typically developing siblings in families who have a child with Down syndrome (Giallo and Gavidia-Payne 2006). In fact, siblings' adjustment was better predicted by these influences than their own coping resources (Giallo and Gavidia-Payne 2006). Longitudinal data from families affected by autism spectrum disorder (ASD) have shown that the psychological adjustment of typically developing siblings also impacts on the outcomes of the child with a disability; for instance, sibling problem behaviours are associated with complex behaviours in the child with ASD up to 3 years later (Hastings *et al.* 2014). These studies emphasise the importance of learning more about siblings' perceptions and reactions, as well as the experiences of the parents and child with the disability.

Adaptation theory may also help to explain the experience of a person who has a sibling with 22q11.2DS. People adapt to their environment through assimilation (i.e. using an existing schema to manage a situation) or accommodation (i.e. changing an existing schema to manage a situation; Piaget 1952). A recent review of the literature regarding siblings of children with ASD highlighted that, although typically developing siblings are vulnerable to behavioural and emotional issues, they can accommodate well to the challenges (Green 2013). Typically developing siblings may not even be at risk of adjustment problems. Parent and teacher reports of mental health symptoms amongst siblings of children with ASD indicated that they did not exhibit a disproportionate prevalence of internalising or externalising symptoms compared with the general population (Dempsey *et al.* 2012). Thus, it is likely that siblings of a person with 22q11.2DS experience challenges but may adapt well.

The voices of siblings are important to consider in the framework of the family system and familial

adaptation. It is within this context that we aimed to explore the subjective interpretations of typically developing young people and adults who have a sibling with 22q11.2DS from a phenomenological epistemological position. We sought siblings' meaning-making (both positive and negative) relating to (1) managing the unique situation of having a sibling with 22q11.2DS, (2) perceptions of change in themselves over time and (3) expectations of their future as influenced by their sibling's disability. Interpretative phenomenological analysis (IPA; Smith 1996), underpinned by phenomenology, double hermeneutics and symbolic interactionism, is a suitable qualitative methodology for this 'lived' experience (Smith 2004) as it seeks idiographic meaning-making.

Methods

Participants

A purposive sample of five typically developing siblings (two males, three females), ranging between 16 and 42 years of a person with 22q11.2DS were recruited from a supporting foundation. Although the foundation's Facebook page has approximately 3000 'likes', it is impossible to know how many people (1) saw the post, (2) engaged with the post (e.g. read the advert properly), (3) fit our inclusion criteria and (4) passed the information onto other potential participants. Therefore, we cannot calculate the response rate. Despite this, the participants were a homogenous group relating to the unique phenomenon under investigation (Smith and Osborn 2008) in that they were all a sibling of a person with 22q11.2DS. The demographic characteristics of participants and their sibling are outlined in Table 1. Pseudonyms are used to protect the participants' confidentiality.

Procedure

Recruitment occurred through an online support group. A brief blurb about the study was posted on the support group's Facebook page. Then, potential participants (or their legal guardian when less than 18 years old) contacted the researchers, at which time they were screened for eligibility and sent participant information and consent forms. Open-ended, semistructured interviews were conducted to allow for deep analysis and sense-making of the participants' rich, personal accounts (Smith and Osborn 2008; Smith 2011). The three topics covered in the interview schedule were (1) managing life as a sibling of a person with 22q11.2DS; (2) expectations of change in their lives; and (3) expectations for the future. Each interview was digitally audio-recorded and lasted approximately an hour, with three conducted face-to-face and two conducted via telephone. The interviews were conducted with the understanding and consent of the participants. Ethical approval was obtained in accordance with the Declaration of Helsinki.

Analysis

Each interview was transcribed verbatim. Following the analysis procedures for IPA as outlined by Smith *et al.* (2009), transcriptions were read and reread, with preliminary themes or ideas noted in the margins. This descriptive analysis then led to more

Table 1 Participant and child characteristics

	Participant			**Affected sibling of participant**			
	Gender	**Age**	**Birth order**	**Gender**	**Age**	**Birth order**	**Developmental ability***
'Laura'	F	16	Eldest	F	10	3/4	4
'Matt'	M	17	Eldest	F	4	4/4	5
'Kate'	F	31	Eldest	M	29	2/2	6
'Peta'	F	42	Eldest	M	35	2/2	5
'Tom'	M	26	Youngest	F	29	1/2	5

*The child's developmental ability as rated by the participant on a scale of 1–7, where 1 = severely delayed and 7 = not delayed at all.

interpretative explanations (Smith *et al.* 1999). Each transcript was analysed completely before proceeding to the next. A table was created with all themes apparent in the transcripts, grouped into clusters under appropriate headings (Table 2). Conclusions were drawn in terms of similarities and differences between participants.

Rigour check

The first stage of analysis (i.e. noting of ideas and potential emergent themes in the margin) was conducted independently by each author for each transcript. This acted as a rigour check, where the first author's interpretations were compared with the other authors'. Each author had similar interpretations, and any small differences were clarified through robust discussion and referral to the interview transcripts for evidence. We used Yardley's (2000) guidelines for assessing the quality of qualitative research to ensure rigour and quality.

Sensitivity to context was initially addressed by reviewing relevant literature. The methodological approach of IPA was led by the philosophy of phenomenology, symbolic interactionism and critical realism. IPA is committed to exploring, describing and interpreting unique phenomena. In this case, we sought to understand the 'lived' experience of being a sibling of a person with 22q11.2DS (Smith *et al.* 2009). By using a hermeneutic exploration and taking a critical realism stance, a researcher is able to capture both the objective and relative truths of the participants. However, as the researchers' access to the participants' personal world is affected by their own conceptions, a double hermeneutic is involved, that is, the researcher making sense of the participant making sense of their experience (Smith and Osborn 2008). The authors were sensitive to the context of life with 22q11.2DS because all are current researchers in 22q11.2DS and family functioning. This research focuses on siblings because families have identified its importance when the researchers engage with them through research, clinical practice, online support groups and family conferences. It is unrealistic to assume that biases such as these can be fully bracketed. However, reflexive practices (e.g. discussion and independent audits) were utilised throughout to ensure that the researchers were not forcing the data into preconceived interpretations based on their knowledge of the literature, contact with families affected by 22q11.2DS and personal experiences.

Commitment, rigour, transparency and coherence are apparent through the researchers' engagement and experience of studying the impact of disability on the family unit, with parallel training in IPA. Throughout the analysis, the authors consistently referred to the transcripts and recordings to ensure that they were staying true to the data. Quotes from participants are provided in this manuscript as supporting evidence for the themes and the double hermeneutic process. The audit trail accounted for the systematic examination at each level of analysis (e.g. transcripts, independent audits, meetings, notes and tracking between authors). This allowed for transparency of the findings and enhanced the quality and transferability.

Results

One superordinate theme: They are the priority overarches two subordinate themes: (1) Jekyll and

Table 2 Stages of interpretative phenomenological analysis

Stage	Description
1	Relistening, transcription, reading and rereading
2	Developing emergent themes through independent interpretation (e.g. noting)
3	Credibility established through robust author discussion
4	Repeating stages 1–3 for the other four cases
5	Searching for connections across emergent themes, identifying convergence and divergence
6	Clustering of subordinate themes 'Jekyll and Hyde of acceptance' and 'Made me a better person' under the superordinate theme 'They are the priority'
7	Reviewing transcripts to validate interpretations in the results

Hyde of acceptance and (2) Made me a better person (Table 3).

They are the priority

The overarching theme describes the participants' acceptance of their sibling taking precedence in their family's life. This is not necessarily emotionally loaded. For example, Matt understands that his affected sibling needs more attention and time from his mother than he does. He actively strives to become an independent and responsible person to reduce the caregiving burden on his mother. Although he longs for more time with his mother, he does not place blame on anyone and willingly resigns from his childhood:

> I am 18 and I do the stuff I need to do for myself which I don't mind… I sometimes do want to hang around with my mum, but we just don't get that time together. [Matt, 17 years]

The same lack of resentment is also demonstrated in the siblings' response to the pressure to 'achieve for two'. They are grateful for their own abilities and skills, recognising that their affected sibling does not have these natural advantages. In light of this, the participants welcome the opportunity to give the spotlight to their sibling:

> This puts pressure on me because now my kids are the only grandkids my parents have got. I need to make sure that I visit my parents often... But at the end of the day I think it is more important that he stays happy. [Kate, 31 years]

However, the required selflessness can be stifling too. At times, the participants feel that they must repress their feelings, especially regarding their frustrations with their sibling. They worry about placing a greater burden on their parents; however, not having the chance to process their experiences or debrief leaves them frustrated with their affected sibling:

> I don't always make sense of everything that is happening, I just try to keep the peace. [Laura, 16 years]

Jekyll and Hyde of acceptance

In many forms, the participants have had to adapt to the situation that they have been thrust into by chance. Acceptance for these participants is a constant juggling act of conflicting feelings. That is, they have creeping thoughts of anger, yet also appreciate the good that has come out of their experiences.

Pragmatic acceptance. This theme describes the rational acceptance exhibited by the participants in relation to their affected sibling. Instead of feeling guilt or placing blame, they understand that they cannot change the 22q11.2DS and thus adjust accordingly:

Table 3 Summary of themes

Superordinate theme: They are the priority	
Subordinate themes	**Description**
Jekyll and Hyde of acceptance	Acceptance is a juggling act of conflicting feelings. Positive aspects of the experience are not as constructive as they appear. For example, participants recognise positive change in themselves (e.g. empathy) but have difficulty applying this to their affected sibling. Also, seemingly negative reactions (e.g. avoidance) are actually adaptive and self-protective for the participants in terms of the constant challenges they face.
Made me a better person	The strain of having a sibling with 22q11.2 deletion syndrome (DS) provides a foundation for positive change. Participants feel pride regarding their siblings' achievements, recognising that they must overcome extra challenges because of the syndrome. Participants understand that they will need to make sacrifices for their sibling in the future (e.g. taking on a caring role) but happily prepare for this.

> I do feel sad but one part of me says that this is life. … I would never blame my sister… it's not mum's fault either... [Matt, 17 years]

The motto of 'this is life' is echoed by all the participants. Sometimes, it is expressed as resignation, but generally, the participants appreciate that life can simply be difficult at times. Having a sibling with 22q11.2DS is just one example of a challenge one may face:

> It could have been different but I don't want it different. I love her. There would still be hard things in life… that's life. [Laura, 16 years]

Grudgingly growthful. Psychological growth (i.e. positive change in psychological functioning, such as empathy; Joseph and Linley 2005) is emerging in the participants; however, they are not all willing to embrace this aspect of their experience. Although they recognise the positive impact having a sibling with a disability can bring, some do so grudgingly. The participants' resentment over the strain and stress their sibling has brought into their lives is evident, despite acknowledgement of the positive changes they have personally undergone. Laura, in particular, recognises that she has become a more empathic person, but she does not display this understanding in relation to her affected sibling:

> My sister does not even want to 'give it a go', she does not even try. … But I can now empathise and understand other people's problems. I feel more empathetic for disabled kids at school. [Laura, 16 years]

Tom touches on the normalcy of his affected sibling and the associated experiences for his family. It is simply the way life has been for them, with little reflection. Although he is a compassionate brother, he does not recognise this in himself, nor does he appreciate that his empathy is a unique gift:

> I have learnt to be more patient, more understanding of disability and more open to disability… I have never given it any special thought. There is nothing special about all that. [Tom, 26 years]

Adaptive avoidance. The mixture of acceptance reactions continues through avoidance. The participants remove themselves from stressful situations in self-protective manner. Peta, in particular, finds the family dynamics overwhelming and must separate herself in order to avoid a spiral of jealousy and guilt about the attention her affected brother receives:

> It's always about him. … I still talk to my family but I have isolated myself… [it's] the best thing that I have done. [Peta, 42 years]

The other participants do not feel the need to take responsibility for their family situation and their sibling's idiosyncrasies or behaviour: a potentially positive attribute, as many of these issues are features of the syndrome that cannot be changed. Guilt or shame is considered unnecessary. Despite helping out on occasion, the participants recognise that they are a sibling rather than a caregiver and happily leave the parents to diffuse testing situations:

> I am not embarrassed about her… I can look past all the superficial crap and just don't worry about it. … Usually I leave it for mum to sort it out, I just walk away. [Matt, 17 years]

Never-ending struggle. At times, the participants feel like the challenges they face as a family are overwhelming. There is a sense of selfishness if one is to discuss their frustration, as they all know that 22q11.2DS is no one's fault, least of all their affected sibling. As the quote from Laura demonstrates, the participants have difficulty adequately expressing themselves because they must always inhibit their feelings to keep the peace:

> It's hard living with her because she is very badly behaved... She can be a little bit cute sometimes but... It's hard to put it in words. [Laura, 16 years]

The participants' reactions to a stressful living situation are not validated by those around them, who see it as less important and more transient than those facing the sibling. Sympathy is always directed to the child with 22q11.2DS. Sometimes, it is impossible not to lash out at their sibling in anger. This is followed by shame because they are so used to keeping quiet:

> She will not understand why we told her off, and then we feel bad about that. [Matt, 17 years]

What about me?. When thinking about the never-ending struggle, the participants begin to reflect on what their life could have been like without a sibling affected by 22q11.2DS. They feel jealousy towards their imagined self, free of the daily stresses they encounter. They ruminate about different scenarios that could have improved their life:

> I got no attention after he was born… my mind often thinks how it would have been if it was reversed [Peta, 42 years]

The reality the participants face is one of constant concession. It is 'her way or the highway'. There is no space for compromise or fairness; the affected sibling seems to always come out on top:

> I come home once a month but if she has already planned something, then she can't change her day, even if it's a five-minute job. [Tom, 26 years]

Living under their affected sibling's rule, the participants find themselves asking 'what about me?' Their needs and desires become lost (by themselves and their parents) in the pursuit of their sibling's happiness:

> If it's not done according to her way, then she is not happy and does not behave well and she gives us so much stress. If she wants something and can't get it she makes us feel bad and finally gets her way. [Laura, 16 years]

Made me a better person

This subordinate theme refers to the psychological growth the participants have undergone. The strain of having a sibling with 22q11.2DS is wearing, leaving the participants susceptible to burnout. However, it also provides a foundation for them to become a better person by embracing patience, empathy and gratitude. Kate embodies these attributes, positively reappraising all experiences surrounding her brother with 22q11.2DS:

> I am a patient person… I am pretty sure some of it is from the experience of growing up with him, it all needed patience… It has given me greater awareness of a lot of difficulties that the other people have. I really appreciate what a lot of people go through around me and in life as well… I think it has made me a better mother… I am really grateful of what he has taught us. [Kate, 31 years]

What could their life have been without 22q?. Grief for the affected sibling is captured in this theme. The participants imagine their sibling's potential without 22q11.2DS and feel sorrow for the life they could have had. Even small milestones or achievements bring about a fierce pride, with the often concealed mixture of sadness and pride bubbling to the surface:

> I love him, he is absolutely beautiful… I am sure he can achieve the best, he can (teary)… Having letdowns is hard and it's harder to get back and continue on… I just want to appreciate his little wins. [Kate, 31 years]

The love that the participants feel for their affected sibling outweighs the negative effect of 22q11.2DS. They feel close as a family when they momentarily forget the impact of the syndrome. There is a wistful longing for the imitative normalcy to last:

> Her face will just light up, she will start playing with you… you kind of forget that moment that she has got something. [Matt, 17 years]

Responsibility for future. For the younger participants, there is uncertainty about the trajectory of the syndrome. They are concerned for what might happen because they do not have a thorough knowledge of what 22q11.2DS entails. Helpless to find answers and unwilling to cause more problems for their already burdened parents by asking, they carry this anxiety alone:

> I think there might be something that develops over time, I don't know but that's something that's in my head, one thing that I am worried about. [Matt, 17 years]

Although, at times, the participants avoid thoughts of their role in the future, the burden of responsibility also weighs on them. For the older participants, there is recognition that one day, their parents will no longer be able to care for the affected sibling. Kate is biding her time, cramming a lifetime of experience and achievement in before she 'resigns' from her own life. She spoke about the urgency she felt to achieve

her goals (e.g. living overseas, studying and providing her parents with grandchildren) before she steps into her carer's role. Although she feels sorrow for this sacrifice, she is glad to take on this position with the knowledge that she can care for her brother appropriately and keep him happy. She has taken active steps to prepare and considers it a personal (rather than forced) choice, which makes the change seem less of an obligation:

> I worry for his future. I discussed this with my partner - he said that we can move to my brother, when time comes… Other than that I always did what I wanted to do. I have always liked to strive for a lot and I have also always been conscious of the fact that my parents have only got me. [Kate, 31 years]

Discussion

Although a number of studies have explored the experiences of individuals who have siblings with disabilities, little is known about how people are affected when their sibling has a complex multisystem disorder such as 22q11.2DS. The current study undertook a phenomenological exploration of sense-making in siblings of a person with 22q11.2DS and identified both positive and negative influences of having a sibling with the syndrome. Naturally, each person had their own emotional journey; however, there were aspects of their journey that the participants in the current study shared. Conflicting feelings were particularly evident, with the overarching theme of '*They are the priority*' describing reactions to the attention the affected sibling receives. All participants reported sacrifice, such as frequently making compromises that were more favourable for their affected sibling. Although the participants mourned what could have been without the condition, they were realistic and accepting. At times, they disengaged from potentially distressing events for self-preservation and did not want to embrace the positive meaning-making they recognised in themselves (e.g. empathy) due to resentment. Yet, they knew it was there and focused their efforts into positively reappraising their circumstances. This is consistent with the theory of meaning-based coping (Folkman 1997), whereby the siblings were motivated to create positive psychological states. Overall, the participants were accepting despite their struggles and felt proud, compassionate and grateful. These results are similar to siblings of people with other disabilities (discussed in the succeeding texts). Further research is needed to determine whether there are challenges and joys that are specific to people who have a sibling with 22q11.2DS compared with other disabilities.

As per Piaget's (1952) adaptation theory, the participants assimilated and accommodated their schemas to manage the impact of 22q11.2DS on their sibling and the family unit. Despite the increased risk of mental health problems associated with the challenges of having a sibling with a disability may bring, the participants were largely managing well. This is not necessarily unusual: Siblings of individuals affected by disability are often resilient (e.g. Dempsey *et al.* 2012; Green 2013). For example, even though children with siblings who have a disability generally report higher scores on depression and anxiety than children with typically developing siblings, many still fall within the normal range, indicating positive adjustment (McHale and Gamble 1989). These results are encouraging; however, it does not mean that siblings' needs (or risk of poor outcomes) should be neglected. Rather, it provides an opportunity to explore what promotes this hardiness and how the family system contributes both positively and negatively to their outcomes.

Feelings of shame and guilt regarding the disability have recently been reported amongst siblings of a child with 22q11.2DS (Okashah *et al.* 2015). However, this was not particularly present in our sample. Although Peta battled with these feelings, the rest of the participants expressed a pragmatic approach, viewing their sibling (and their potentially problematic behaviours) as separate to their own identity. They accepted that guilt would not change neither the presence nor the impact of the syndrome and thus actively avoided such thoughts. As the current participants were all in their late teens or adulthood, this could be an age effect with the current findings better reflecting an increased ability of rationalising their experiences in a way that younger children struggle with.

It has been suggested that complex behaviours, rather than the disability in itself, have negative impacts on siblings (e.g. not being able to participate in activities due to the affected sibling; Neece *et al.* 2010). In keeping with this, Laura commented that

she often had to put her own wants and needs aside because of her sibling's behaviour issues. Although the participants in this study coped well with the additional demands that having a sibling with 22q11.2DS placed on them, some siblings may suffer as a result of not having the opportunity to express themselves or feel validated. In terms of family system theory, this could indicate that the current participants may be protected from poor outcomes by explicit support from the rest of their family. For example, the participants generally reported reassurance from parents that they did not need to care for the affected child. For siblings who do not have those resources, health professionals are well-placed to advise families on appropriate resources to ensure that the family system stays intact.

Finally, the participants reported a sense of responsibility for their sibling's future. Kate especially was actively preparing for the time when she would need to take over the care of her brother. This is consistent with research on sibling relationships for older adults with ID, many of whom had a sibling who advocated for them, looked after their wellbeing and supervised their care (or acted as a primary carer; Bigby 1997). Further, several participants described uncertainty regarding what is likely to happen to their sibling in the future but did not want to question their already overburdened parents. Interestingly, similar themes were reported in Okashah *et al.*'s (2015) article. Parents reported that they shared information about 22q11.2DS with the typically developing sibling as issues arose or when asked, with only 41% of parents having discussed future needs. It is important for health professionals to consider siblings in the care of a child with a developmental disability and to provide information appropriate to their developmental stage (Kisler and McConachie 2010). Siblings should also be considered in genetic counselling sessions. This can help reduce the anxiety some of the participants in the current study reported regarding 'not knowing' and help facilitate an open discussion between parents and children around future needs for the person with 22q11.2DS.

Limitations

The variance in participants' ages presents a challenge for interpretation. For example, a teenager who lives with their affected sibling (e.g. Laura, 16 years) is likely to have different joys and challenges compared with an adult who lives a fairly separate life from their family (e.g. Peta, 42 years). However, the similarities between the participants' experiences in the current study suggest that there are issues common to siblings of a person with 22q11.2DS, regardless of their age. Further, the qualitative nature of the current study meant that generalisation and causality were not sought. Instead, an understanding of the 'lived' experience of this particular group of participants was pursued through an in-depth qualitative analysis. Another limitation is that, although 22q11.2DS is a genetic disorder, none of the siblings expressed concern about the genetic implications of the condition. This is in contrast to previous literature (e.g. McAllister *et al.* 2007), which has shown that genetic conditions can be associated with guilt and worry about genetic risk. This may be because the participants are not at an increased risk of having a child with 22q11.2DS or because they are at a stage in their sense-making where reproduction is less significant than other issues they face (e.g. Laura and Matt are 16 and 17 years old respectively, and thus, the social impact of their siblings' behaviours may be more salient than future genetic implications). However, we did not specifically ask the participants to ruminate on their experience as related to the genetics of 22q11.2DS.

The double hermeneutics of the analytic process may have impacted both positively and negatively on the researchers' interpretations. The researchers' experience in family functioning as related to 22q11.2DS provided rich insight into these siblings' accounts. However, the researchers also brought personal experiences to the analysis as parents and relatives of people affected by disabilities. It was important to be mindful in order to remain focused on the participants' stories instead of pursuing the researchers' own experiences and agendas. For example, one researcher feared resentment towards her parents in her own life and believed that the participants were experiencing these feelings. Robust discussion between authors and referral to the transcripts revealed that this was often not the case (e.g. Matt stated, 'it's not mum's fault'). Even though the researchers' biases may have affected the interviews and analyses, the knowledge and experiences were also valuable. According to Maslow (1966: p.45), 'there is no substitute for experience,

none at all', which, in this context, means that the researchers' own experiences gave the insight and ability to see the humanness in the participants' accounts, rather than simply a psychologically theoretical understanding. In an effort to enhance the study's quality, Yardley's (2000) recommendations guided the research and acted as a rigour check. Therefore, despite the limitations, this study contributes to the 22q11.2DS literature by highlighting both positive and negative interpreted impacts on siblings.

Future research

This study provides a platform for future research regarding 22q11.2DS and family wellbeing, particularly as related to siblings' adjustment. More research is needed to determine whether these participants' experiences are typical for siblings of people with 22q11.2DS or indeed whether 22q11.2DS affects siblings in a way that is different or similar to other disabilities. For example, none of the participants in this study reported concerns regarding the genetic implications of the syndrome, which has been reported in other genetic conditions (McAllister *et al.* 2007). Further, this study did not highlight a sibling experience that was unique to 22q11.2DS, as comparable results have been found in siblings of people with other developmental disabilities. This is important for future research aiming to identify siblings' support needs, which has implications for support services and whether they need to be tailored specifically to 22q11.2DS.

Conclusions

Although the participants in this study accepted that their affected sibling is the priority in their family, it is still important that siblings of a person with 22q11.2DS feel included, loved and appreciated, as it is clear that they experience challenges and thus may be at risk of negative outcomes. However, positive outcomes such as gratitude and psychological growth were present in our sample and are realistic and achievable for other siblings in similar situations. Clinicians should refer to Kisler and McConachie's (2010) guidelines for managing disability diagnoses, which acknowledge the importance of information provision for relatives of the affected child, including siblings. Peer support for siblings can be accessed through Sibshops, a forum for typically developing siblings of a child with a disability.

Conflict of interest

No conflicts of interest.

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Ethics

Ethical approval was obtained from the research University of Newcastle Human Ethics Committee.

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