SDS Siblings research – IPA Study

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Qualitative research discussion

“An exploration of the experiences of siblings of people with a diagnosis of Shwachman-Diamond Syndrome.”

- Introduction to researcher and context of research
- Overview of the research
  - Context and existing research
  - Participant demographics
  - Methodology and analysis
  - Main themes
  - Discussion of themes and clinical utility
  - Future research recommendations

Researcher context

- Systemic family psychotherapist – research constituting part of qualifying thesis
  - Working professionally in NHS CAMHS Looked after Children service, prior experience in perinatal services and eating disorder therapy.
  - Background as an occupational therapist in learning disabilities and an MA in English literature – particularly interested in narrative approaches, social constructionist theories, intersectionality and ‘therapy as activism’ (Vikki Reynolds).
- Personal/familial connection with SDS
  - Husband had a brother and a sister who had SDS, who have both sadly passed away.
- Interest in sibling work
  - Close relationship with own sister, eldest of 2 girls, which has been variable throughout life and has taken work!

- I started the research with an expectation about what I might find, based on the experience of me, my husband and his family.
  - Rooted in a particular experience of SDS and recognising that while this is not a generalisable one, and there are many different layers/stages of chronic illnesses, it might provide some insight.
  - Basis for further thought. Research idea and aim went through various iterations.
  - Can't avoid my own belief in the value of therapy, but also recognising that this is often a 'last resort' for families and the best situation would be managing at home, on their own terms.
    - Also realising that there must be a right time, supportive context and motivation for therapy. Particularly challenging in family work where different family members may, and likely will be, at different stages and places.

Context for research/gaps in literature

- No existing studies inside or outside of systemic research literature, on the experiences of siblings of people diagnosed with Shwachman-Diamond Syndrome
- Drawing on literature from other chronic illnesses including sickle cell anaemia, cancer and cystic fibrosis.
  - Sibling relationships are uniquely non-optional, can be engaged with to a chosen degree into adulthood
  - Can have the most longevity in a person's life; formed through 'shared experiences and reminiscences' (Howe et al. 2004, p.444); siblings as 'co-voyagers' through life (Edwards et al. 2006)
  - Sibling relationships as a space to try out roles and identities, consider future relationship dynamics, set boundaries, manage conflict, negotiation, jealousy, sharing (of parental attention, space and possessions) and compromise (Graff et al. 2012).

- Chronic illnesses can disrupt ’typical’ family disorder and expected lifecycle occurrences (moving out, having own family)
- Hanvey et al. (2022) coined term ‘glass children’ to describe relative invisibility that happens to children whose siblings are chronically ill and are necessarily overlooked in terms of parental attention; cystic fibrosis literature favours ‘shadow children’ descriptor (Hodges, 2018).
- Research on younger siblings of people diagnosed with chronic illness shows more propensity towards externalising symptoms like anger and behavioural changes.
  - More likely to seem jealous of increased parental attention, and complicated guilt about their envy (Caspi, 2011)
- Older siblings instead are more likely to show internalising symptoms like anxiety, low mood and low self-esteem (Hollidge, 2001).
  - Potential explanations include attempts to protect family members and not communicating their own needs due to feeling as though parents have enough to contend with.
  - Sense of illegitimacy regarding their own concerns? (Sharpe & Rossiter, 2002)
  - Difference in play styles – increased need for caution
  - Increased need for responsibility in household tasks, potential parentification (Byng-Hall, 2002) whereby they take on the role of a pseudo parent more than a child out of necessity of demands.
- Positives of siblings with chronic illness include increased empathy and compassion, responsibility, independence and life appreciation (Incledon et al., 2013), especially when not defined by sibling.

- Genetic elements of chronic illness and ‘survivor’s guilt’ for not being impacted in unaffected siblings (Packman et al., 2005) – shame and attempts to compensate.
- Family therapy theory in chronic illness posits that having a space to process ideas and emotions can improve adjustment, communication and wellbeing in a scale called ‘family coherence’ (Kissane et al., 2016)
- Chronic illness as disruptive to established family hierarchies, implicating boundaries, discipline, communication, connection and openness.
- Communication around different stages of chronic illness enables family’s ability to develop a narrative, which helps with conflict resolution and ability to meet one another’s needs (Kissane et al., 2016).
- Majority of research has been on 1:1 psychological support, growing understanding of how illnesses impact entire family system and providing a platform to discuss these concerns, instead of shying away from them, can be beneficial.
  - Growing research on whole family work to increase confidence in management of diagnosis and attainment of coherence. Platform to discuss emotions that are potentially otherwise avoided, contributing to wellbeing and healing (Shields et al., 2012).

Research question and aims

- Aim to explore the childhood and adulthood experiences of siblings of people with a diagnosis of Shwachman-Diamond Syndrome, in their own words.
- Particular interest in perception of self and resources accessed.
- Questions around diagnosis process, experiences of being a child and an adult, and what in their opinion has helped them navigate their experiences.

Interpretative Phenomenological Analysis (IPA) methodology

- Qualitative research, focusing on subjective and emotional experiences instead of the empirical or the quantitative (Dingwall, 2008).
  - Specific psychological experiences and how they are understood.
- IPA recognises the role of the researcher as part of the process, which seemed important reflecting on my own experiences with SDS and how this informs both the questions I'm interested in, and how I'll interpret the data I find.
  - How do people in a particular group make sense of their experiences? How does the researcher understand their account of this?
    - If I as researcher do not consider and name this, I risk biasing the research. I am not neutral.

- IPA gives people a chance to explore their experiences in their own words, often through semi-structured interviews about an hour in length, on video conferencing software.
- Rooted in ’social constructionism’ (Burr, 2015), the idea that reality is understood and developed through conversation with other people, and the context of culture cannot be ignored.
- Methodology where researcher recognises their contributions as epistemologically central, felt vital, hence IPA.

Ethics

- Ethical approval for this study was granted by University of Exeter's CEDAR (Clinical Education, Development and Research) department.
- Ethical approval granted by the University of Exeter on the basis that due to the potential psychological distress from discussing emotive subjects, the participants were over 18.
- Research supervisor, Kate Campbell
- CEDAR is an Applied Psychological Practice Centre of Excellence.
- Each research interview included a psychological debrief and signposting for support as required. Please get in touch if you would like any more information about the ethics process involved with this study.

Participant recruitment and demographics

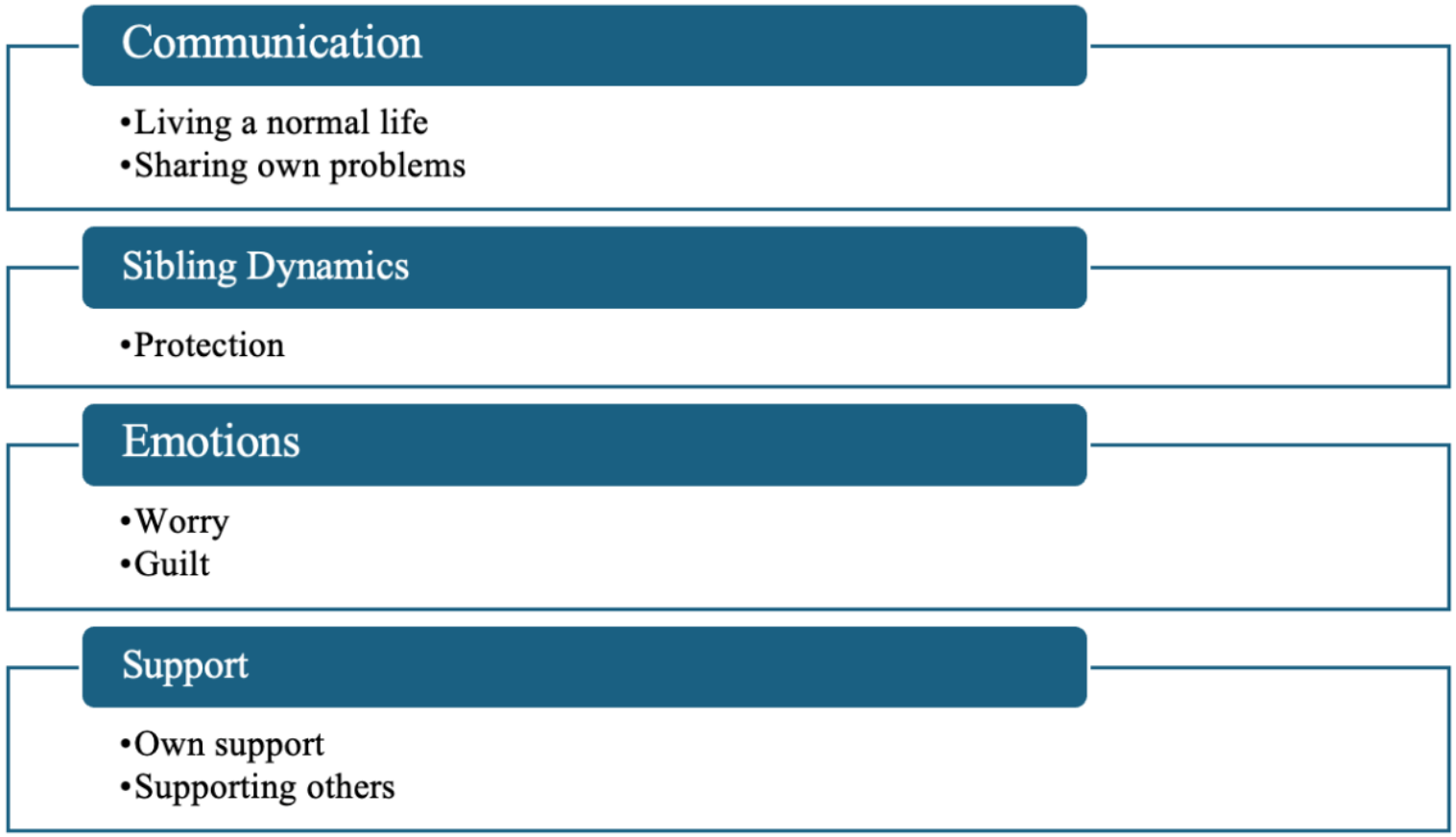
- Participants were recruited purposively through SDS-UK and SDS Alliance (USA based) charities, through their mailing lists.
- Prospective participants then contacted me.
- 5 participants, an ideal sample size for small-scale IPA study.

Gender	Age	Education	Ethnicity	Geography
Male	21	High school	White Caucasian	USA
Male	55	Postgraduate degree	White Caucasian	UK
Female	39	Undergraduate degree	Mixed European-Asian	UK
Male	18	Apprenticeship	White Caucasian	UK
Female	27	Postgraduate degree	White Caucasian	UK

Participants

- Participants were three cisgender men and two cisgender women.
- Mean age was 32.
- Pseudonyms were randomly assigned: Dominic, Eve, Jon, Stef and Matt.
- No participant had accessed SDS-related therapeutic intervention.
- Two participants had siblings die of SDS/related complications.
- One had a sibling undergoing cancer treatment, and two described their sibling as well.

Overview of main themes



1) Communication

- **‘Living a normal life’**
  - Enabled by reducing communication about SDS? ‘*a priority*’ for one participant; conversations limited to the practical and pragmatic, instead of the emotional.
  - Different stages with SDS – earlier in the process, participants spoke about not *‘needing*’ to discuss SDS but recognising this may change over the illness’ duration.
  - Enabled by SDS’ relative invisibility. ‘*Existing and persisting in the background*’
  - Participants talked about their mistaken understanding that medical intervention removed SDS *‘I thought the tablets the fixed SDS.’* Frequency of medical appointments, nonetheless.
  - Avoiding negative conversation made instances of death more jarring – one participant noted not understanding that the hospitalisation was different to any other time.
  - Also extended to not talking about the future, and instead ‘*caring in the moment.’* Wondered if this removed opportunities to discuss it for sibling with SDS as they may not initiate.

1) Communication

- **Communication of own problems**
  - All participants compared any of their difficulties with that of their sibling. Recognised the problems of SDS/comorbidities as '*worse*' and felt '*bad*' talking about their own.
  - Extended to some participants not sharing concerns with their parents, e.g., school bullying, learning difficulties, because of recognising their parents being '*overloaded*' and '*having enough to deal with*.'
  - Widened support network of school and grandparents to share problems with.
  - Some participants also recognised the context of their gender, time of childhood and family patterns of communication as contributing to them not sharing their own difficulties – '*it wasn't done in my family*.' This was seemingly irrespective of SDS.

2) Sibling dynamics

- Differences in participants – some oldest and some younger or youngest.
- Recognition of parental expectation on chores as being the same.
- Role of siblings at healthcare appointments as providing '*distraction*' and '*light-hearted comic relief*' for both parties.
- In keeping with literature, younger siblings spoke about their anger and '*misbehaviour',* externalising their emotions and regretting this as an adult.
- Older siblings spoke about their desire to '*be good*' in recognising their parents' load.
- Increased reciprocity, equal relationships and respect identified with increasing age in both siblings.

2) Sibling dynamics

- **Protection**
- Understanding of social hierarchies and the influence of co-morbidities with SDS including social/communication challenges; one participant discussed ensuring school peers were '*aware of [him] as older brother.'*
  - Recognised this was *'taken for granted*' by parents and not explicitly requested/communicated
- Carefulness and '*caution*' in play styles initiated by parents; fear of '*roughing up*' but no significant differences in closeness.
- One participant discussed being a *'second mum' to* her brother, but this was not requested by parents and they tried to discourage.
  - Linked to her awareness of how much her parents were managing, as well as her perception of her brother's relative '*weakness*.'

3) Emotions

- **Worry**
  - Linked to protection and anxiety/uncertainty about the prognosis of SDS in participants. No clear solutions, lots of healthcare appointments, especially when more unwell.
  - Experience of worry feeling '*inherited*' and learned from observing parental worry.
  - Understanding of why parents shielded their worry but sense that as children, they noticed the worry regardless, and this amplified their own worry.
    - '*Kids notice stuff. Even if you pretend that you're not worried, they know you are*.'
  - Worry as self-directed in the genetic testing/diagnostic process, which gives insight into the experiences of their unwell siblings.
  - Relationship with worry as changing following bereavement and being redirected to other family members in one participant: '*I'm never not worrying about people I love*.'

3) Emotions

- **Luck and guilt**
- Guilt regarding genetic carrier status and not testing positive, recognising that this is linked purely to chance.
  - Conflicting '*relief*' for testing negative but not being able to discuss this due to siblings' experiences. Resulting dilemma and avoidance of discussions.
- Rareness of SDS and confusion about its occurrence/presentation.
- Increased parental attention and guilt about raising awareness of this. Recognition of the context of the attention as not being favourable – '*I wouldn't swap places for the extra attention*' – but not always remembering this as a child.
  - Not universally experienced amongst participants as there being increased parental attention for unwell sibling.

4) Support and resources

- **Own support**
- All participants lauded the importance of social networks and friendships – intimate connections, validation and understanding.
- '*Distraction'* provided by hobbies, sport, religion and therapy.
- Different stages and needs for support correlated with different stages of SDS illness – post-bereavement, identified more need for therapeutic intervention in participants; earlier in illness, participants noted they *'haven't needed*' additional support.

4) Support and resources

- **Supporting others**
- Based on understanding of challenges faced by their siblings, all participants noted how in various ways they aim to support others – fundraising and awareness raising, employment, politically, morally.
- '*Try working hard but also playing with a different deck of cards*' – recognition of societal barriers.
- Some participants located their decision to work in health and social care as being linked to their sibling – *'I want to try to help people like him.'*
- Helping others as a way of feeling closer to sibling after they have passed away..
- Also enhanced by travel and extracurriculars – noting that SDS is not the only defining experience, of siblings or affected individuals.

Discussion of themes/clinical recommendations

- Identified themes as informed by intersectional elements of my identity as researcher, as well as those of the specific participants, instead of being generalisable – offering some preliminary insights to build upon in future research.
- SDS not universally experienced, uneven trajectory in symptoms, diagnosis, co-morbidities (Tan et al. 2019).
- Fear of causing upset in other family members leads to less open communication, leading to a 'reciprocal silence' (Deavin et al. 2018) that siblings feel unable to break. Potentially problematic feedback loops; participants remained aware of the worry of their parents, but internalised it, blamed themselves and felt confused.
  - Wherever possible, communication about SDS is key. **Conversations from diagnosis onwards may help understanding and strengthen relationships** – helping with family coherence. But should only be done when more understanding has been achieved, **or practical suggestions about roles** to take can be made, otherwise may increase fear (Lummer-Aikey & Goldstein, 2020).

- Importance of siblings feeling as though they have roles in the process – clear tasks to help with purpose and less alienation/helplessness.
  - Siblings appreciated being able to provide distraction around hospital appointments, helping with involvement, productivity and value.
  - **Participants indicated that siblings are already aware and worried; both parties find involvement helpful. Important for healthcare professionals to be aware of.**
  - **If communication precedent is established early on, conversations will continue.**
  - **Preference is for families to manage their challenges internally and need therapeutic work as last resort.**
- Considering what **a 'normal' life is in the context of culture, individual/specific family values** about health and wellbeing, expectations about life course trajectory, timing of achievements… – healthcare professionals should ensure that these are asked and kept central to treatment.
- If SDS is held central, this influences relationships with individuation, roles etc. For future research studies, considerations about whether siblings are alive or have passed away and considering them **separately in different projects.**

- Confusion amongst siblings about prognosis and knowing who to talk to – **signposting and information for family members as being available throughout illness process.**
- Helping physically and offering protection feels more accessible as a role for siblings than talking about emotions.
- Participants recognised the context of increased parental attention and despite it being challenging at times, did not begrudge it.
  - **Importance of having wider networks of support for siblings** including school and extended family, especially grandparents. This buffers potential lack of attention.
  - Social networks able to be stronger at bolstering support when there is **acceptance of the illness** and its prognosis.
- Participants all experienced guilt and wanted a forum to discuss this but recognised that they would be grappling this on a life-long basis.
- Consensus about supporting others amongst all participants, but also **recognising the importance of other factors impacting worldview** including travel, reading, media and hobbies.

Future research suggestions

- Childhood accounts where ethically permissible as current research is based on adult accounts
- Comparison between parental and sibling accounts, especially as research indicates parents tend to rate sibling relationships more negatively (Sharpe & Rossiter, 2002).
- Wider geographical and global majority participant recruitment
  - Comparison to research on conditions that disproportionately impact minoritised communities, like sickle cell anaemia.
- Future mixed methods research to increase validity and reliability through triangulation (Flick, 2018)
- Separate research based on phase of illness with SDS, especially focusing on bereavement lens.

Future direction for this study

- Aim to publish in Chronic Illness journal in 2025
- Contribution to Association of Family Therapists' monthly practice magazine, Context, with the publication date of August 2025.

- Please contact me with any questions, suggestions or comments – I'd really love to hear from you ☺
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