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Exploring the World of Profound Intellectual Disabilities:

Examining the Use of Multi-Sensory Storytelling and an Exploration of the Experience of Mothers to First Born Children With Profound Intellectual Disabilities Making Decisions About Having More Children

by

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Abstract

People with profound intellectual disabilities are often not recognised or included in research. This thesis aims to explore their experiences and those of people who care for them.

A systematic literature review explores the use of multi-sensory storytelling (MSST) with people with profound intellectual disabilities. There is currently limited guidance on interventions for this population and this review aims to explore the possibility of MSST being used therapeutically. A mixed methods review was completed with four databases and three grey literature sources. Data quality was assessed, and an integrated convergent approach was used to synthesis data. Data was extracted verbatim and thematic analysis was conducted. Seven papers were included. Four themes emerged from thematic analysis: MSST is beneficial for people with profound intellectual disabilities, storytellers benefit from MSST, there is mutual enjoyment of MSST and factors to consider when developing, implementing and evaluating MSST. The findings indicate MSST is potentially a feasible and acceptable therapeutic intervention for people with profound intellectual disabilities and should be considered in future research.

The empirical chapter explores the lived experience of mothers with a first-born child, who has profound intellectual disabilities and their decision to have more children. Seven participants were interviewed about their experiences. Transcripts were analysed qualitatively using interpretative phenomenological analysis. Six group experiential themes emerged: 'Grief and loss driving you to have another child', 'Isolation from the world around you', 'The burden of care splits you in two', 'The weight of responsibility of bringing another child into the world when your first has profound intellectual disabilities', 'Finding meaningful support is down to you' and 'Siblings heal and bring joy'. The findings highlight difficulties faced by mothers, demonstrating the need for professional support and the importance of peers for coping strategies and building resilience.

Keywords: Profound Intellectual Disabilities, Multi-Sensory Storytelling, Systematic Review, Mothers, Children, Interpretative Phenomenological Analysis

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Research Thesis: Declaration of Authorship

Print name: Jean Ruth Jevons

Title of thesis: Exploring the World of Profound Intellectual Disabilities:

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I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

1. This work was done wholly or mainly while in candidature for a research degree at this University;
2. Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
3. Where I have consulted the published work of others, this is always clearly attributed;
4. Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
5. I have acknowledged all main sources of help;
6. Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
7. None of this work has been published before submission

Signature: ..... Date: ….August 2025 ..

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Definitions and Abbreviations

MSST……. 'MSST' stands for multi-sensory storytelling. This abbreviation has been used throughout chapter 2 in the systematic review. MSST are a combination of text and stimulus that provide sensory experiences for the person listening. Text and stimulus are presented together. The stimulus will provide opportunities to look, touch, hear, smell, see, and taste for the listener. They have been developed in educational settings to provide inclusive literacy opportunities for people with profound intellectual disabilities.

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1 Bridging Chapter

This chapter will introduce you to the systematic review and empirical chapters of this thesis. It will share the aims and rationale for both chapters within the wider systemic context. The ontological position and epistemological stance will be shared to offer explanations of methodological decisions which were outside the purview of the selected journal guidelines. The axiological positioning of the author and their reflexivity are then discussed to demonstrate how this was managed ethically whilst conducting the research. Finally, plans for dissemination will be shared, including reasoning for the choice of journal, exploring how its values align with the authors.

1.1 Aims and Rationale

The aim of this thesis was to explore the lived experience of people with profound intellectual disabilities, mothers who care for them and those who support them. Given the broad criteria outlined in the DSM5 (American Psychiatric Association [APA], 2013) and the complex variation in phenotypic presentation there is a huge range of diversity, but they all have a profound intellectual disability in common, (British Psychological Society [BPS], 2016), an IQ below 20-25 or a developmental age of two to three years (Maes et al., 2020). Despite these challenges people with profound intellectual disabilities still enjoy relationships with family, carers and those who are familiar to them, initiating and responding to social interactions through gestures and emotional cues (APA, 2013).

Historically researchers have struggled to include people with profound intellectual disabilities meaningfully in studies due to identifying the defining characteristics, recruitment, specific instruments to collect data, data analysis and ethics, including issues of informed consent (Maes et al., 2020). Whilst this body of work hopes to illuminate experiences of people with profound intellectual disabilities and those who care for them, the author recognises the limitations it has in embracing the concept of 'being with' people with profound intellectual disabilities from the inception of the studies, due not only to the time constraints, and wider systemic issues impacting inclusive research (de Haas et al., 2022). However, it strives to explore the experiences of mothers of children with profound intellectual disabilities and how person-centered individualised activities can make meaningful impact in real life.

The covid-19 pandemic shone a light in the UK on the abandonment of those with intellectual disabilities, their vulnerabilities (Shakespeare et al., 2021) and wider societies' perception of them as disposable (Goodley, 2023). This concept of 'otherness' and 'less than' stems from the Western notion of self being autonomous, independent center of thought and agency (Davy 2019). This research hopes to move away from this concept of ideal self and consider relational approaches within the feminist theory of ethics of care, where interpersonal relationships are essential in the

development of sense of self and independence. Davy (2019) proposes that “neither autonomy nor care is privileged, but both are in service of the other: autonomy cannot be enabled without care, and care cannot be enabling without respect for autonomy” (p.102). This not only challenges the way in which we view those who require care, but also those who provide it. Caring roles both professional and unpaid typically fall to women across society (Baldwin & Twigg, 2024). Feminist theory of ethics of care aims to explore the power imbalances associated with the provision of care, and the implications it has for women in the role of carer in wider western society that does not value disability or care (Davy, 2019). Critics of care ethics frequently come from fellow feminists who argue that it will perpetuate the oppression of women in under-valued caring roles, promoting self-sacrifice which is detrimental to the feminist movement (Cawston & Archer, 2018). Hampton (2018), argued that the feminist theory on ethics of care perpetuates gendered roles in society, encouraging women to subjugate themselves to undervalued care roles, to appease and prioritise men within the patriarchal structures of society. However, to view this in a two-dimensional way, only considering gender, is shortsighted. To fully explore the complexities of the ethics of care one must approach from a position of intersectionality, considering how other parts of a person’s identity influence their relation to care, including age, class, education and ethnicity, to name but a few. Therefore, the empirical chapter of this thesis aligns itself with the exploration of the role of care and its relational nature, hoping to explore the experiences of mothers caring for children, who belong to one of the most marginalised groups in society.

The systematic review paper also recognises the importance of understanding the relational experience provided by multi-sensory storytelling for people with profound intellectual disabilities and those who care for them. Research indicates that consistency of care is key to carers developing skilled communication with the people they support, leading to better standards of care for people with intellectual disabilities (Nijhof et al., 2024). This review aims to understand the potential benefits of multi-sensory storytelling and whether, or how it promotes communication, understanding and knowledge. Furthermore, it invites the reader to consider the possible functionality of multi-sensory storytelling as a therapeutic approach for people with profound intellectual disabilities. Given the current lack of guidance by the National Institute for Health Care and Excellence (NICE) (NICE, 2016) and the BPS (BPS, 2016) for psychological interventions for this population, reduction in benefits (Able2UK, 2025) and wider systemic discrimination (Goodley, 2023; Shakespeare et al., 2021), attention from the clinical psychology profession is pertinent to support this marginalised minority.

1.2 Ontology and Epistemology

Both the systematic review and the empirical chapters use qualitative methodologies, thematic synthesis, and interpretative phenomenological analysis (IPA) respectively. The author approached both papers from an individual relativist ontological position. This ontological stance

views reality as an unfixed entity, which is constructed by individual and collective experiences (Pretorius, 2024).

Further, both chapters' methodology followed the epistemology of interpretivism, where knowledge is subjectively constructed in the mind of the individual based on their personal experiences and interpretations of these (William, 2024). From the researcher's perspective we cannot objectively define this knowledge, but we can interpret and make meaning from the understanding the individual has made of their own experiences (Pretorius, 2024). Therefore, it is important to highlight the subjectivity of research positing an interpretivist epistemology (William, 2024), where there is the double hermeneutic of the researcher interpreting the meaning made by the participant of their own lived experience (Pretorius, 2024).

The methodological approaches employed for each paper reflected an individual relativist ontology and interpretivist epistemology (Pretorius, 2024). IPA (Smith & Nizza, 2022) was selected for the empirical as it enabled the use of semi-structured interviews to explore individuals subjective experience and meaning, a methodology aligned with an interpretative epistemology (William, 2024). Whilst thematic synthesis (Thomas & Harden, 2008) was selected for secondary data analysis in the systematic review. Whilst the systematic review was approached from an interpretivist epistemology there is an element of constructivism. To understand a person with profound intellectual disabilities experience of multi-sensory storytelling, data based on the interpretations of the person supporting them was analysed, creating a triple hermeneutic. That is, the interviewees have shared an experience of multi-sensory storytelling with a person with profound intellectual disabilities, and they are verbally reporting their interpretations of the interaction and the meaning made, which included their perspective of what the person with profound intellectual disabilities experienced. So, the researcher's interpretation is based on the interpretation of a constructivist social interaction (William, 2024) between storyteller and listener. Given the scant research on this population, a systematic review including mixed methodological studies was deemed appropriate. Given the complexity of integrating quantitative and qualitative data, findings were extracted verbatim from included papers to promote fidelity to the results during analysis (Lizarondo et al., 2025). Data analysis followed an iterative process, where multiple readings of data and how emerging themes reflected the data were undertaken. Following this, themes were shared and discussed with the wider research team to ensure an accurate representation of the data.

1.3 Reflexivity, Axiology and Ethics

Reflexivity is a key component of qualitative, interpretivist and constructivist research (William, 2024; Pretorius, 2024). It pertains to a level of self-awareness whilst actively participating in the process of research (Palaganas et al., 2017). As a key aspect of qualitative methodology, the author used supervision and a reflective diary to help maintain an awareness of my own thoughts,

beliefs, and feelings, to ensure this was not influencing the outcomes (Smith & Nizza, 2022). An essential aspect of reflexivity is understanding my axiological position, that is, how the values I hold influenced the research and findings (Pretorius, 2024). Part of this is sharing my own personal experiences, beliefs and values to ensure transparency.

My previous experience of working with people with intellectual disabilities was an integral motivation for wanting to complete this research. With over twenty years' experience working across contexts, settings, and lifespan I witnessed firsthand how marginalised people with intellectual disabilities are, especially those with complex, profound and multiple disabilities. Initially I had hoped to conduct primary research on multi-sensory storytelling, that collaborated with people with profound intellectual disabilities, via proxies, if necessary, from inception. This was based on my time spent as a teaching assistant in a special educational needs school where sensory approaches and storytelling was pervasive across the school, curriculum and ages within the school. I saw how exploring sensory experiences through stories supported participation, accessibility, learning, relationships and enjoyment. However, the rigorous ethical scrutiny combined with the time restraints of the course made bringing this project to life very difficult. I fully understand the importance of this thorough ethical assessment, but I had to balance this with completing the academic requirements within the deadline set by the course. I therefore decided to complete a review of the extant literature documenting research into multi-sensory storytelling.

Through my work and social connections, I have seen the challenges faced by families whose children have profound intellectual disabilities at both an individual level and at a family unit level. My own journey of motherhood included one of my children receiving a diagnosis during completion of the research. This made me think more deeply about the impact of diagnosis and disability on mothers and the wider family. Particularly how women decide whether to go on and have further children given the complexity of their experiences of motherhood. To manage this and hold other viewpoints in mind reflexive diaries were kept throughout the process, experts by experience were consulted, the author attended peer supervision with other IPA researchers and regular supervision with the wider research team occurred.

As an intersectional feminist I am aware of the many opportunities I have. I recognised that I could create a space to amplify the voices of other women whose motherhood has been shaped by children with profound intellectual disabilities (López Radrigán et al., 2025). I hope that this research will have real world impact for not only the women who shared their experiences, but also for the children they love and care for.

1.4 Dissemination Plan

This thesis comprises of two further chapters, both of which have been written with the intention of publishing in peer-reviewed journals. The journal I hope to submit to is the 'Journal of Applied Research in Intellectual Disabilities' (JARID). Therefore, the chapters have been written to this journal's authors guidance (see supplementary material 1 for author guidelines). JARID is a peer-reviewed journal based in the UK aiming at an international, multi-disciplinary readership on intellectual disabilities, making it a suitable choice for both the empirical and systematic review paper. It covers topics including quality of life, communication, family issues, mental health, staff training and service provision. All of these are applicable to both chapters in this thesis.

Choosing JARID was also due to their approach to the use of person-first language following consultancy with people who have lived experience of intellectual disabilities. This aligned with my values of promoting all people as valued members of the scientific community and wider society. JARID stipulates that through consultation people with intellectual disabilities do not like to be referred to as abbreviations or acronyms and therefore they should be referred to in full person-first language. Following examples of other published articles in JARID I have referred to people with profound intellectual and multiple disabilities as people with profound intellectual disabilities throughout both chapters, for consistency and readability.

JARID requires a lay summary to be included when submitting a paper for publication. I like the inclusion of this as standard across the publication to increase accessibility of research to all people, including those with intellectual disabilities. The open access option for JARID is important when considering the availability of the findings to those in the wider community of intellectual disabilities and not just those working in research.

Further dissemination will be pursued through an oral presentation of the empirical paper at the British Psychological Faculty for Intellectual Disabilities in May 2026.

1.5 References

Able2UK, (2025, March 26). *Government confirms cuts to disability benefits in spring statement*. https://www.able2uk.com/news/disability-news/benefit-system/government-confirms-cuts-to-disability-benefits-in-spring-statement

American Psychiatric Association [APA]. (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Washington, DC.

Baldwin, S., & Twigg, J. (2024). Women and community care: reflections on a debate. In M. Maclean & D. Groves (Eds.), *Women's issues in social policy* (pp. 117-135). Routledge. https://doi.org/10.4324/9781003522317

British Psychological Society [BPS]. (2016). *Psychological therapies and people who have intellectual disabilities*. https://doi.org/10.53841/bpsrep.2016.rep106

Cawston, A., & Archer, A. (2018). Rehabilitating Self-Sacrifice: Care Ethics and the Politics of Resistance. *International Journal of Philosophical Studies*, *26*(3), 456–477. https://doi.org/10.1080/09672559.2018.1489648

Davy, L. (2019). Between an ethic of care and an ethic of autonomy: Negotiating relational autonomy, disability, and dependency. *Angelaki*, *24*(3), 101-114. https://doi.org/10.1080/0969725X.2019.1620461

de Haas, C., Grace, J., Hope, J., & Nind, M. (2022). Doing research inclusively: Understanding what it means to do research with and alongside people with profound intellectual disabilities. *Social Sciences*, *11*(4), 159. https://doi.org/10.3390/socsci11040159

Goodley, D. (2023). Being human as praxis: for people with learning disabilities. *Subjectivity*, 1. https://doi.org/10.1057/s41286-023-00159-6

Hampton, J. (2018). Feminist contractarianism. In L. M. Antony & C. E. Witt (Eds.), A mind of one's own: Feminist Essays on Reason and Objectivity (2nd ed., pp. 337-368). Routledge

Lizarondo, L., Stern, C., Salmond, S., Carrier, J., Cooper, K., Godfrey, C., Vandyk, M., Pollock, D., Rieger, K., Apostolo, J., Kirkpatrick, P., Borges Dos Santos, K., & Loveday, H. (2025). Methods for data extraction and data transformation in convergent integrated mixed methods systematic reviews. *JBI evidence synthesis*, 10-11124. *DOI:*10.11124/JBIES-24-00331

López Radrigán, C., Aparicio, A., & Tenorio, M. Feminist ethic of care in academic knowledge production: Reflections from disability researchers. *Feminism & Psychology*, 09593535241306536. https://doi.org/10.1177/09593535241306536

Maes, B., Nijs, S., Vandesande, S., Van Keer, I., Arthur-Kelly, M., Dind, J., Goldbart, J., Petitpierre, G., & Van der Putten, A. (2021). Looking back, looking forward: Methodological challenges and future directions in research on persons with profound intellectual and multiple disabilities. *Journal of Applied Research in Intellectual Disabilities*, *34*(1), 250-262. https://doi.org/10.1111/jar.12803

National Institute for Health and Care Excellence [NICE]. (2016). *Mental health problems in people with learning disabilities: prevention, assessment and management*. https://www.nice.org.uk/guidance/ng54

Nijhof, K., Boot, F. H., Naaldenberg, J., Leusink, G. L., & Bevelander, K. E. (2024). Health support of people with intellectual disability and the crucial role of support workers. *BMC Health Services Research*, *24*(1), 4. https://doi.org/10.1186/s12913-023-10206-2

Palaganas, E. C., Sanchez, M. C., Molintas, M. P., & Caricativo, R. D. (2017). Reflexivity in Qualitative Research: A Journey of Learning. *The Qualitative Report, 22*(2), 426-438. https://doi.org/10.46743/2160-3715/2017.2552

Pretorius, L. (2024). Demystifying research paradigms: Navigating ontology, epistemology, and axiology in research. *The Qualitative Report, 29*(10), 2698-2715. https://doi.org/10.46743/2160-3715/2024.7632

Shakespeare, T., Watson, N., Brunner, R., Cullingworth, J., Hameed, S., Scherer, N., Pearson, C., & Reichenberger, V. (2022). Disabled people in Britain and the impact of the COVID-19 pandemic. *Social Policy & Administration*, *56*(1), 103-117. https://doi.org/10.1111/spol.12758

Smith, J. A., & Nizza, I. E. (2022). *Essentials of interpretative phenomenological analysis*. American Psychological Association.

Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC medical research methodology*, *8*, 1-10. https://doi.org/10.1186/1471-2288-8-45

William, F. K. A. (2024). Interpretivism or constructivism: Navigating research paradigms in social science research. *International Journal of Research Publications*, *143*(1), 134-138. https://ijrp.org/paper-detail/6086

2 Multi-Sensory Storytelling for People With Profound Intellectual Disabilities: A Systematic Literature Review

This paper has been prepared in the format required for the *Journal of Applied Research in Intellectual Disabilities*.

2.1 Abstract

Background: There is limited guidance on therapeutic interventions for people with profound intellectual disabilities. This review aimed to explore key outcomes of multi-sensory storytelling (MSST) with this population and those supporting them.

Method: A mixed methods systematic literature review was completed of four databases and three grey literature sources where MSST was used with people with profound intellectual disabilities. Data quality was assessed, and an integrated convergent approach was used to synthesis data. Data was extracted verbatim and thematic synthesis was conducted.

Results: Seven papers were included. Four themes emerged from thematic analysis: MSST is beneficial for people with profound intellectual disabilities; storytellers benefit from MSST; there is mutual enjoyment of MSST; and factors to consider when developing, implementing and evaluating MSST.

Conclusions: Findings indicate the feasibility and acceptability of MSST as a possible therapeutic intervention for people with profound intellectual disabilities. MSST should be considered in future research.

2.2 Accessible Summary

- Professionals have little guidance about how to support people with profound intellectual disabilities and their mental health.
- Multi-sensory storytelling is used in educational settings and could be useful in helping people with profound intellectual disabilities with their mental health.
- Multi-sensory storytelling is useful for the person with profound intellectual disabilities, the person supporting them, and both can find it enjoyable.
- Future research should explore how multi-sensory storytelling can be used to improve mental health services for people with profound intellectual disabilities.

KEYWORDS

Profound Intellectual Disabilities, Multi-Sensory Storytelling, Systematic Review

2.3 Introduction

People with profound intellectual disabilities are a heterogenous group primarily identified by a severe intellectual disability (British Psychological Society [BPS], 2015). As a group they are severely limited in their ability to communicate with, and understand, others (Bellamy, 2010). Their communication is often focused on needs or wants and described as being pre-symbolic, using behaviour, gestures, eye pointing, body language and limited use of symbols or very simple language (Colley, 2020). They have more than one disability, which may include sensory or physical disabilities, complex physical health needs or mental health difficulties (MENCAP, 2024). These physical disabilities and medical conditions often lead to people being wheelchair users (Colley, 2020) and can often present with behaviours that challenge, are harmful or are counterproductive, including self-injury (American Psychiatric Association [APA], 2013). There is a lack of certainty about global prevalence rates. Within the UK there are estimated to be 75,000 people with profound intellectual disabilities (University of Cambridge, 2024), this is approximately 0.11% of the population (based in the Office for National Statistics population estimates at the mid 2021 census) (Office for National Statistics, 2022). The number of people with profound intellectual disabilities in the UK, and globally, is likely to rise due to advances in both neo-natal care and other medical disciplines (Colley, 2020).

Those with profound intellectual disabilities are a minority within a minoritised group of those with intellectual disabilities. They face further marginalisation due to wider systems in western society viewing them as disposable and a lack of appropriate care throughout guidance from governing bodies (Goodley, 2023). There are, however, numerous passionate practitioners working with this population. The work of people like Dave Hewett and Melanie Nind, who developed intensive interaction in the 1980's, has gone on to be a beneficial intervention for people with profound intellectual disabilities (Jefferies, 2009), with potential to reduce the likelihood of trauma and as a restorative intervention (Samuel & Doswell, 2021). However, challenges remain demonstrating the effectiveness of intensive interaction due to conducting ethically sound research which adheres to methodological rigor and transparent reporting (Hutchinson & Bodicoat, 2015). Research with people with profound intellectual disabilities continues through UK-based organisations like Promoting a More Inclusive Society (PAMIS) and the Tizzard Centre. However, historically this has not been translated more broadly into publications in peer reviewed journals. This may be due to difficulties in conducting research inclusively with this population (de Haas et al., 2022). Developments in the application of inclusive methodologies such as photovoice (Chinn & Balota, 2023) within this population gives hope to increased production of inclusive research and therefore a growing body of evidence.

There is very limited guidance on therapeutic interventions for people with profound intellectual disabilities. Those that are recommended are typically indirect interventions through proxies, parenting interventions (National Institute for Health and Care Excellence [NICE], 2016) and systemic family therapy (BPS, 2016). Whilst positive behaviour plans may include some direct interventions there are currently no direct psychological interventions recommended by the NICE guidelines specifically for individuals with profound intellectual disabilities (NICE, 2016). The BPS guidance (2016) recommends music therapy as a direct intervention, but this would have limitations for those who have sound sensitivities. The lack of research pertaining to people with profound intellectual disabilitie's psychological wellbeing may be due to professionals prioritising managing their physical health in the context of potentially multiple life limiting conditions, over their emotional and wellbeing needs.

The program Stopping the Over Medication of People with a learning disability and autistic people (STOMP) is a priority within the NHS Long Term Plan for services across NHS England (National Health Service [NHS] England, 2024). STOMP highlights the need for professionals to consider other treatments before prescribing medication. This gives precedence to the identification and development of effective therapeutic interventions for this population. In the Special Educational Needs and Disability code of practice for 0–25-year-olds (Department for Education & Department of Health and Social Care, 2015) those with profound intellectual disabilities are only explicitly mentioned once in relation to needing additional support with their cognition and learning. Whilst guidance states there should be mental health provision from a multi-disciplinary team for those with an intellectual disability, it does not mention the specific needs of those with profound intellectual disabilities or propose how these should be met.

It is estimated 40% of adults and 36% of children with an intellectual disability experience mental health difficulties; this is higher than for the general population (NICE, 2016). The BPS identify profound intellectual disabilities as a factor associated with mental ill-health (BPS, 2016). They highlight that a higher number of GP appointments, living with a paid carer, and urinary incontinence are factors associated with poor mental health, (BPS, 2016), all of which may be more common for this population. However, having a severe physical disability or immobility was not associated with mental health difficulties (BPS, 2016). It is unclear if this is due to an absence of mental health issues, inadequate measures for this population, or this population not meeting the diagnostic criteria for mental health diagnoses, due to diagnostic criteria being written for a typically developing population.

This lack of knowledge and understanding about how to psychologically support people with profound intellectual disabilities is reflected in the literature. There is a scarcity of research exploring

the life, experiences, and worlds of those with profound intellectual disabilities, with them often being excluded entirely (Kellet & Nind, 2014). There is recognition and a movement towards research becoming more inclusive to those with profound intellectual disabilities, not only for participation, but in designing and conducting (de Haas et al., 2022). It is also important to examine the current available literature, to help improve care and better understand how clinical psychology can support people with profound intellectual disabilities.

2.3.1 Multi-Sensory Storytelling

Story telling is an ancient creative art form which allows people of all ages to enjoy new experiences, ideas, learn, understand other perspectives, develop imagination and empathy (Grove, 2022). In typical populations of young people storytelling has been shown to increase psychological resilience (Ramamurthy et al., 2024), reduce anxiety (Abdi et al., 2025), and improve self-regulation (Tillott et al., 2024). Picture books have been shown to be educational and to promote health by supporting adaptive coping strategies for young people who have been newly diagnosed with potentially traumatic conditions (McKeon et al., 2025).

Research has also demonstrated that reading books has a positive impact on the wellbeing, development and social inclusion for people with profound intellectual disabilities (Robinson et al., 2019). Robinson et al., (2019) reported inclusive literature, such as sensory stories, creates meaningful and gratifying exchanges for people participating in reading books.

Senses allow people to develop self-awareness and experience the world around them (Nesayan et al., 2018). For humans to be able to learn and adapt behaviour they need to be able to process and integrate sensations received externally and from their internal world (Bundy & Lane, 2022). There is a lack of research on how a profound intellectual disability affects sensory experiences (Agostine et al., 2022). However, given the types of impairment frequently seen within this population it is likely it has a significant impact on their ability to access sensory experiences and process them. Agostine et al., (2022), provide an example where “limited gross motor movement restricts opportunity to explore the environment, which leads to limited sensorimotor experience needed to make sense of objects. This then delays fine motor skill development and restricts play, which further restricts sensory development” (pp. 3). The inability to access and interact with sensory experiences will not only impact people’s neurological development, but it will shape their ability to engage with life and learn (Grace, 2023). Humpheson (2024) also reported the use of sensory approaches with people who have profound intellectual disabilities supports improved positive cognitive, emotional and social outcomes.

Multi-sensory storytelling (MSST) provides the opportunity for people with profound intellectual disabilities to engage with stories through a combination of narrative and sensory stimuli (Fuller, 2022). In MSST between six and eight sensory stimuli are mounted on a neutral-coloured laminated board for maximum visual acuity and opportunities for exploration (Lambe et al., 2022). Each of these 'pages' are accompanied by one or two sentences which directly relate to the presented stimuli (Lambe et al., 2022) and typically last about three minutes (ten Brug et al., 2016). All the 'pages' and text are stored in a box with a visual and tactile stimulus on the outside, which signals to the person with profound intellectual disabilities the MSST is about to occur (Lambe et al., 2022). The sensory stimuli should be presented with equal value to the words chosen (Grace 2023). Grace (2014) proposes MSST should be shared as a social experience, not just as a form of therapy. Whilst this may be true, exploring the potential outcomes of MSST could be valuable for clinical psychologists in understanding and developing therapeutic interventions for people with profound intellectual disabilities. Whilst MSST has been included in broader reviews, to date there have been no systematic reviews exploring MSST.

2.3.2 Aim

To examine and describe the key outcomes of MSST for people with profound intellectual disabilities and those supporting them.

2.4 Method

This systematic review followed Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Page et al., 2021). It was conducted in line with Hong et al's., (2017), guidance for the convergent integrated synthesis design for reviewing mixed methods studies. The review protocol was registered on the PROSPERO website (CRD42024599801).

2.4.1 Eligibility Criteria

Cooke et al's., (2012) search strategy for mixed methods was employed to structure this thematic synthesis of a mixed methods systematic review. This strategy includes Sample; Phenomenon of Interest; Design; Evaluation; Research type, (SPIDER), see Table 1, (Cooke et al., 2012). The following inclusion criteria were applied to the literature search. The sample included people with profound intellectual disabilities. No age restrictions were applied as it is a lifelong condition. The phenomenon of interest was the experiences and outcomes for those with a profound intellectual disability and those supporting them when participating in MSST. For design qualitative, quantitative and mixed methods were included. The research could have been conducted in any context, including, but not limited to, educational, residential, clinical, hospital, and community settings. Evaluation included any reported qualitative experience reported by participants and any outcomes reported in quantitative results. For research type published and

unpublished studies in the English language were included. Due to a potential paucity in research in this area no cut-off date was employed. Studies from all disciplines would be included due to the small literature base. Papers were excluded if they were a systematic review, conceptual opinion piece or not written in the English language.

Table 1

Application of SPIDER search strategy tool (Cooke et al., 2012) for eligibility criteria

Element of Search Strategy	Inclusion Criteria	Exclusion Criteria
Sample	People with profound intellectual disabilities. No age restriction.	Any papers which do not include people with profound intellectual disabilities.
Phenomenon of Interest	Experience of MSST.	Not exploring the experience of MSST.
Design	Qualitative, quantitative, mixed methods, single case designs.	Not applicable
Evaluation	Any reported qualitative experience reported by participants and any outcomes reported in quantitative results.	Not applicable
Research type	Any context, including, but not limited to, educational, residential, clinical, hospital, and community settings. No cutoff date. Papers from all disciplines.	Systematic review, conceptual opinion piece or not written in the English Language.

Note. MSST = multi-sensory storytelling

2.4.2 Information Sources

A comprehensive search for studies was completed in October 2024 across the following databases: CINAHL; PsycInfo; PsycArticles; and Web of Science. Searches of Pro Quest and PUBMED to incorporate grey literature to reduce publication bias (Paez, 2017). This search was repeated in January 2025 to capture any papers published after October 2024 in the final analysis.

2.4.3 Search Strategy

The development of search terms utilized the Sample and Phenomenon of Interest from the SPIDER acronym (Cooke et al., 2012), and combined using the appropriate Boolean operators, ‘OR’ and ‘AND’. Initial search terms were identified by the primary researcher, followed by a consultation with a university librarian to expand the terms to ensure terminology encompassed cultural and temporal nuances (see Table 2). Due to a paucity in research exploring the experiences of those with

a profound intellectual disability the search terms were kept as broad as possible to capture all relevant research within the search.

Table 2

Search Terms

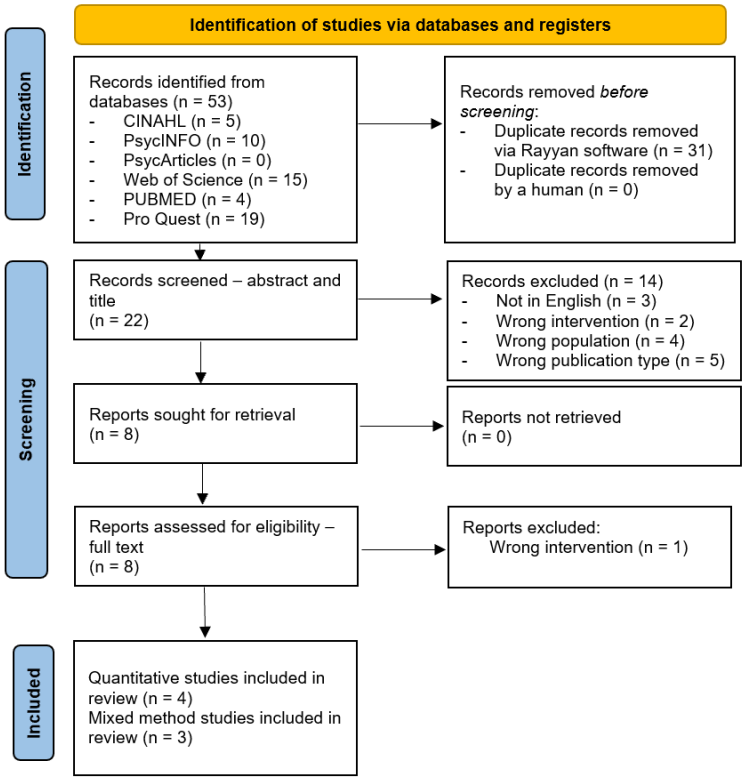
Sample	Phenomenon of Interest
"learning disabilit*" OR "profound and multiple learning disabilit*" OR "PMLD" OR "profound intellectual and multiple learning disabilit*" OR "PIMD" OR "severe learning disabilit*" OR "multiply handicapped" OR "multiply disabled" OR "intellectual retardation" OR "profound disorder of intellectual development" OR "profound intellectual disability*"	"multi-sensory story telling" OR "multi-sensory storytelling" OR "multisensory story telling" OR "multisensory storytelling" OR "sensory stor*" OR "sensory storytelling"

2.4.4 Study Selection

Following searches data was screened and selected in accordance with the PRISMA guidance (Page et al., 2021) and is presented in Figure 1.

Figure 1

PRISMA Diagram for Inclusion Process



An initial search of four electronic databases identified thirty papers: CINAHL (*n*=5), psycINFO (*n* = 10), psycARTICLES (*n* = 0), and Web of Science (*n* = 15). Searches of grey literature sources identified a further twenty-three papers: PUBMED (*n* = 4) and Pro Quest (*n* = 19). The included full text studies were hand searched for additional relevant papers. This did not result in any further papers being identified. Duplicates were removed (*n* = 31) via Rayyan and remaining articles were checked by hand for further duplications (*n* = 0). There were twenty-two papers from electronic databases and grey literature to screen title and abstract. Due to the small numbers of potentially relevant papers at this stage 60% (*n* = 13) were randomly selected to be independently screened by a second reviewer (McDonagh et al., 2013). Following screening for title and abstract eight papers were identified for full text screening. Of these papers one was selected for full text screening due to being unable to establish eligibility based on the title and abstract alone. At full text screening this paper was removed due to the intervention looking at the adherence to delivery structure of MSST, not the outcomes. There were seven papers identified at full text screening to include in the review. 100% papers were screened by a second reviewer (Stoll et al., 2019).

2.4.5 Characteristics of Included Research Papers

Seven papers reported studies including children and adults with profound intellectual disabilities and their care giver. These papers were conducted in the United Kingdom (*n* = 2), Belgium (*n* = 1), Netherlands (*n* = 1) and three recruiting participants from across both Belgium and Netherlands (*n* = 3). Six papers included dyads of a care giver and a person with profound intellectual disabilities (total *n* = 179). The seventh paper's sample included 27 school-based professionals, 18 of whom went on to be observed. This gave an overall total of 385 participants across all seven papers. Four papers observed and coded video recordings of predetermined sessions within the intervention schedule. Two papers combined this with qualitative data from the care giver. One paper completed observations and conducted semi-structured interviews. (See Table 3 for further information on included papers). Preece & Zhao's (2015) study was conducted at an education setting where there was a range of special educational needs, including profound intellectual disabilities, multiple disabilities and visual impairments, severe intellectual disabilities, social, emotional and behavioural difficulties and visual impairment and autism. When scrutinising the samples' descriptive characteristics it appeared it was predominantly comprised of young people with profound intellectual disabilities. It was not possible to extract data specifically related to this group but given the complexity around the clarity of definition of profound intellectual disabilities, and the lack of research in this area, it was decided to include these findings.

Table 3

Reviewed Research Papers

Study	Aims and design	Intervention	Participants	Setting and country
Penne et al., (2012)	To explore possibility of describing staff interactive style during MSST using a global coding tool. Within subjects repeated measures. Sessions 1, 5, and 10 video recorded and coded.	MSST training. MSST developed. MSST delivered.	20 dyads. Professional caregiver and person with profound intellectual disability (young people and adults).	Settings associated with centre of expertise in profound intellectual disabilities. Belgium.
Preece & Zhao, (2015)	To explore how MSST are used within classrooms and schools. What opportunities do MSST provide and what factors affect its use. Exploratory case study. Mixed methods – semi structured Interviews and observations of practice.	MSST delivered.	27 school-based professionals interviewed. 18 teachers observed using MSST with young people, majority with profound intellectual disabilities.	Schools for special educational needs. England.
ten Brug et al., (2013)	Explore if knowledge is gained from MSST and if so, what kind of knowledge this is. How do teachers use and apply this knowledge in the context of MSST. Mixed methods – sessions 1, 10 and 21 video recorded and coded. Qualitative responses recorded from MSST facilitator.	MSST training. MSST developed. MSST delivered.	3 dyads. Teachers and young person with profound intellectual disability.	Centre for special education for students up to the age of 18 years old. Netherlands
ten Brug et al., (2015[a])	To explore the relationship between alertness and active presentation of stimuli in MSST books with people with profound intellectual disabilities. Within subjects repeated measures. Sessions 1, 5, 10 and 20 video recorded and coded.	MSST training. MSST developed. MSST delivered.	27 dyads. 27 Individuals (12 children and 15 adults) with profound intellectual disabilities and 27 support workers.	14 different organisations over 18 different locations. Specific settings unknown. Netherlands.
ten Brug et al., (2015[b])	To understand how guidelines for delivering MSST positively influence the listeners attention. Within subjects repeated measures. Sessions 1, 5 and 10 video recorded and coded.	MSST training. MSST developed. MSST delivered.	45 dyads. Professionals and person with profound intellectual disabilities (66.7% under 18 years old).	Activity centres or group homes. Belgium and Netherlands.

ten Brug et al., (2016)	To understand if MSST gains more attention than regular story books. If there are differences in attention over time between the two types of stories. Non-randomised control study, between participants repeated measures. Sessions 1, 5 and 10 video recorded and coded.	MSST training. MSST developed. MSST delivered. Vs. Regular story telling.	76 dyads. 76 Individuals with profound intellectual disabilities and 76 professionals. 59.2% of people with profound intellectual disabilities were over 18 years old.	Activity centres, schools and residential settings. Belgium and Netherlands.
Young et al., (2011)	Mixed methods – Exploratory case study. Session 1, 4 and 8 video recorded and behavioural observation completed. Parents and professionals delivering MSST interviewed (semi-structured) at the end about the effect of MSST.	MSST developed. MSST delivered.	8 dyads. 5 mothers, 2 teachers and 1 occupational therapist. 8 young people with profound intellectual disabilities.	Schools, nursery's and one home. Scotland.

Note. MSST = multi-sensory storytelling

2.4.6 Summary of results from papers included in the review

The seven papers included in the review all explored MSST but examined different aspects of outcomes, see Table 4 for a summary of their findings. Three of the quantitative papers aimed to explore the cognitive changes associated with MSST. These included aspects of attention towards MSST and the storyteller over regular storytelling (ten Brug et al., 2016), attention to MSST overtime and how stimuli were presented impacted attention towards stimuli (ten Brug et al., 2015[b]). Alertness was also explored, with higher levels of alertness being associated with the presentation style of the storyteller (ten Brug et al., 2015[a]). The findings of these three papers were supported by the qualitative data reported in mixed methodological papers, where storytellers described perceived increases in attention, alertness and overall interaction with MSST (Preece & Zhao, 2015; Young et al., 2011). Qualitative data also highlighted how the presentation style of the storyteller may affect the outcomes for the listener, with storytellers' creativity, experience and teaching style all contributing to outcomes (Preece & Zhao, 2015). However, Penne et al., (2012), found there was no relationship between storyteller's interactive style and their characteristics, nor between that and the characteristics of the listener. Indicating while presentation style is important, there are no indicators people possessing certain characteristics are any better than any other group of people. This was supported qualitatively by the diversity with storytellers across studies; including educational (Preece & Zhao 2015; ten Brug et al., 2013; ten Brug et al, 2015[b]; ten Brug et al., 2016; Young et al, 2011); occupational therapists (Young et al, 2011) professional support staff (Penne et al, 2012; ten Brug et al., 2013; ten Brug et al., 2015[a]; ten Brug et al, 2015[b]; ten Brug et al., 2016) and parents (Young et al, 2011). Preece & Zhao, (2015) and ten Brug et al, (2013), both found that MSST allows the storyteller to learn more about the listener. However, ten Brug et al's., (2013) quantitative findings indicated the storyteller (teacher) did not apply this knowledge actively to adjusting and adapting further sessions. This resonates with the qualitative findings of Preece & Zhao, (2015) that highlight external barriers to delivering effective MSST, including, time, resources, staffing levels and the expectations of eternal inspectorate bodies.

Table 4

Summary of results from papers included in review

Study	Study type	Results
ten Brug et al., (2016)	Quantitative non-randomised	- There was no significant difference in duration for MSST and regular storytelling. - MSST books and stimuli received significantly more attention than regular books. Attention increased in early readings and decreased slightly in later readings. - There was no significant difference in attention to the storyteller between MSST and regular storytelling. - Attention to storyteller did not change over time for either MSST or regular storytelling.
Penne et al., (2012)	Quantitative descriptive	- All staff scored above the mid-point range on the Maternal Behaviour Rating Scale for all categories, indicating they were at least 'moderately sensitive', 'consistently responsive', 'moderately effective', 'acceptant', 'pervasively enjoying', 'moderately overtly expressive' and 'warm' during MSST. Means remained consistent over time points. - A repeated measures multivariate analysis of variance showed no significant main effect of time (session) on the scores for the different staff interactive style dimensions: Wilks' $\lambda = 0.30$, $F_{14,4} = 0.66$, $P = 0.75$. - There was no effect of client characteristics on staff interaction style. - There was no effect of the carers job, experience, experience with the specific client or age of the carer on interactive style.
ten Brug et al., (2015[a])	Quantitative descriptive	- There were higher levels of active alertness when MSST stimuli were actively presented. - Alertness within sessions was not constant and was related to the presentation of the stimuli. - Where stimuli were presented actively there were associations with larger fluctuations in listener alertness.
ten Brug et al., (2015[b])	Quantitative descriptive	- During MSST listeners were attentive for on average 69% of the time. - Attention increased between the first and fifth sessions and then reduced between the fifth and tenth sessions. - Guidelines for storytellers including repetition, actively offering stimuli and duration of MSST were positively to the amount of the listeners attention. - There were no significant correlations found between listeners attention and guidelines for storytellers including adherence to the original test and using a neural background for stimuli.
Preece & Zhao, (2015)	Mixed methods	- Both commercially produced and home-made MSST are used widely within educational settings.

		- MSST were flexible and adaptable in their use for education or pleasurable activities and can be delivered across settings including classrooms of for outdoor activities. They were utilised across the curriculum, in a broad range of subjects. - MSST could be personalised to meet individual student's needs, preferences and abilities ensuring accessibility and meaningfulness. - MSST supported socialisation for people with profound intellectual disabilities, reducing potential isolation and providing opportunities for group interaction and shared experiences. - MSST were used to assess students' progress and to support the development of skills including communication, fine motor skills, memory and cognitive abilities - Internal factors effecting the use of MSST: storytellers' creativity, experience and teaching style. - External factors effecting the use of MSST: time, resources, staffing levels and expectations from inspectors from external bodies. - Other factors effecting the delivery of MSST included training provided, natural skills of the storyteller, opportunities to observe others and access to mentoring.
ten Brug et al., (2013)	Mixed methods	- MSST does provide new knowledge, but teachers are insufficiently aware of it and do not apply it in subsequent practice.
Young et al., (2011)	Mixed methods	- Positive changes in the engagement with the MSST were reported for seven out of eight people with profound intellectual disabilities. Parents or caregivers acting as storytellers reported these positive changes enabled the listener to manage better with the sensitive topic in six of those seven.

Note. MSST = multi-sensory storytelling

2.4.7 Quality Appraisal

The primary researcher conducted a critical appraisal on the seven papers included in the review using the Mixed Methods Appraisal Tool (MMAT; Hong et al., 2018). See Appendix A for the MMAT. Each study was categorized by design using the MMAT algorithm; quantitative non-randomised (*n* = 1), quantitative descriptive (*n* = 3), and mixed methods (*n* = 3). Once categorized each study had two general screening questions and five approach specific questions applied. Overall, the quality of the papers was good, meeting all the appraisal measures set out by Hong et al., (2018). Table 5 summarises the findings of this quality assessment.

Table 5

Quality Appraisal of Review Papers Using the Mixed Methods Appraisal Tool (Hong et al., 2018)

Quantitative non-randomised studies	First Author	S1. Are there clear research questions?	S2. Do the collected data allow to address the research questions?	3.1 Are the participants representative of the population?	3.2 Are measurements appropriate regarding both the outcome and intervention (or exposure)?	3.3 Are there complete outcome data?	3.4 Are the confounders accounted for in the design and analysis?	3.5 During the study period, is the intervention administered (or exposure occurred) as intended?
	ten Brug et al., (2016)	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Quantitative descriptive studies		S1. Are there clear research questions?	S2. Do the collected data allow to address the research questions?	4.1 Is the sampling strategy relevant to address the research question?	4.2 Is the sample representative of the target population?	4.3 Are the measure appropriate?	4.4 Is the risk of nonresponse bias low?	4.5 Is the statistical analysis appropriate to answer the research question?
	Penne et al., (2012)	Yes	Yes	Yes	Yes	Yes	Yes	Yes
	ten Brug et al., (2015[a])	Yes	Yes	Yes	Yes	Yes	Yes	Yes
	ten Brug et al., (2015[b])	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Mixed methods studies		S1. Are there clear research questions?	S2. Do the collected data allow to address the research questions?	5.1 Is there an adequate rationale for using a mixed methods design to address the research question?	5.2 Are the different components of the study effectively integrated to answer the research question?	5.3 Are the outputs of the integration of qualitative and quantitative components adequately addressed?	5.4 Are divergences and inconsistences between quantitative and qualitative results adequately addressed?	5.5 Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?
	Preece & Zhao, (2015)	Yes	Yes	Yes	Yes	Yes	Yes	Yes
	ten Brug et al., (2013)	Yes	Yes	Yes	Yes	Yes	Yes	Yes
	Young et al., (2011)	Yes	Yes	Yes	Yes	Yes	Yes	Yes

2.4.8 Data Extraction Process

Data was extracted and synthesised by the primary researcher. The complete results sections from each paper with tables and figures were extracted. Due to the primary researcher completing extraction, and the numbers of papers identified, data was collated in paper form using word processing software.

2.4.9 Data Synthesis Process

An integrated data-based convergent approach (Hong et al., 2017) was employed as both qualitative and quantitative data could answer the review questions (Lizarondo et al., 2022). This approach, where qualitative and quantitative evidence is extracted prior to analysis, synthesised into one data set and analysed together using the same synthesis method allowing for a more detailed description of the data (Hong et al., 2017). The integration of qualitative and quantitative evidence supports more complete, concrete and nuanced answers to more complex research questions (Heyvaert et al., 2013).

Original data from all papers was extracted verbatim. The quantitative and mixed methods papers included in the review all reported numeric results with comprehensive narrative descriptions. Verbatim extraction and synthesis allowed analysis and findings to remain as close as possible to the data reported in the original studies (Lizarondo et al., 2025). See Table 6 for further details of the data synthesis process.

Table 6

Convergent Integrated Data Extraction Process and Analysis Based on Lizarondo et al., (2025)

Lizarondo et al., (2025) guidance		**Applied Methodology**
Data extraction	All data should be extracted to provide a holistic understanding of all factors involved in MSST participation and the overall understanding of the Phenomenon for participants.	Following identification of papers to be included, all results sections, including numerical statistics, tables, graphs and figures were completely extracted verbatim.
	Data extraction should occur verbatim in mixed methods systematic reviews to ensure data remains as close to data reported in original papers. This minimises misinterpretation by the reviewer at the analysis stage.	All results sections were extracted verbatim. Where there were numeric results, primary authors had provided sufficient narrative text. This meant no qualitisation or contextual information was required. This ensured data remained as close to that reported in original papers.
Checking data	Numerical results and accompanying narratives should be cross checked with each other to ensure data extracted is accurate and reliable.	All results sections were cross checked between narrative and tables, graphs and figures. To ensure data extracted was accurate and reliable. No discrepancies between numeric and narrative data were found.
Data integration	Extracted data (qualitative and quantitative) should be assembled for detailed analysis across data	All data was copied verbatim into one word document in preparation for analysis.
Data analysis	Analysis should identify similarities and create categories. Categories are grouped to generate overall integrated findings.	To formalise this process and give rigour, Thomas & Harden's, (2008) Thematic Synthesis was applied to the data set coding line-by-line, developing descriptive themes, and the generation of analytical themes.

2.4.10Data Analysis

Once data had been integrated the primary researcher followed the principles of Thomas and Harden's (2006) thematic synthesis. The results sections from all seven papers were integrated into one data set (Lizarondo et al., 2025) and a thematic synthesis was carried out (Thomas and Harden 2006). Four papers were purely quantitative and three mixed methods. The three mixed methods (Preece & Zhao, 2015; ten Brug et al., 2013; Young et al., 2011) had substantially longer results sections than the other quantitative papers. Only the three mixed methods papers from the seven included presented quotes as part of their data, therefore it was decided to not use quotes in reporting the thematic synthesis.

2.4.11Researcher Reflexivity

The primary researcher attempted to be aware of their own values and beliefs from previous experiences of working with people with profound intellectual disabilities prior to and during the review process. This was done through checking themes as they emerged ensuring they were grounded in the data being analysed. The themes were then reviewed by the wider research supervisory team to ensure they represented and evidenced by the data from the papers included in the review.

2.5 Results

Four themes and fifteen subthemes were identified during analysis. See table 7 for a summary of themes and subthemes.

Table 7

Themes and Subthemes

Theme	Subtheme
MSST is beneficial for people with profound intellectual disabilities	Cognition
	Interaction
	Social behaviour
	Decrease in maladaptive and stereotypical behaviour
Storytellers benefit from MSST	Learn about and attune to the person with profound intellectual disabilities
	Developing and sharing best practice
Mutual enjoyment of MSST	Person with profound intellectual disabilities enjoys MSST
	Storytellers enjoy MSST
Development, implementation and evaluation of MSST	Individualisation
	Function of MSST
	Environmental factors
	Stimuli
	Presentation
	Barriers
	Evaluation

2.5.1 MSST is Beneficial for People With Profound Intellectual Disabilities

Storytellers and researchers reported multiple observed changes for the people with profound intellectual disabilities participating in MSST. They shared positive changes in the person's

engagement with the MSST in the session and sustained improvements between sessions in other contexts.

2.5.1.1 Cognition. An increase in alertness was explicitly reported across quantitative and qualitative findings in studies (ten Brug et al., 2013; ten Brug et al., 2015[a]; Young et al., 2011). One storyteller in Young et al's., (2011) research reported "He was very attentive and listening more" (William's mother). Quantitative findings also indicated levels of alertness increased over time with repetition of MSST (Young et al., 2011). This was support in other quantitative results where alertness was impacted by the presentation style of the storyteller, with a more active presentation gaining higher levels of active alertness, and passive presentation receiving higher levels of passive alertness (ten Brug et al., 2015[a]). Alertness peaked during the initial presentation of stimuli and decreased over the window of time it was presented (ten Brug et al., 2015[a]).

Listeners paid significantly more attention to MSST books and stimuli than regular books (ten Brug et al., 2016). Furthermore, quantitative and qualitative findings indicated repetition of MSST increased attention towards the MSST within sessions (ten Brug et al., 2016; ten Brug et al., 2013; ten Brug et al., 2015[b]; Young et al., 2011). Quantitative findings demonstrated the length of MSST impacted attention, with longer stories garnering significantly more attention than shorter stories (ten Brug et al., 2015[b]). An active presentation style of the storyteller garnered more attention from the listener (ten Brug et al., 2015[b]). The physical presentation of stimuli impacted attention, with stimuli presented on white backgrounds receiving more attention (ten Brug et al., 2015[b]). This was also supported in qualitative findings where a change in direction of attention was reported, shifting from the storyteller to the stimuli over time: "There was an association between engagement with the story and decreased visual engagement with the storyteller, suggesting a shift in relative attention in the overall context of the storytelling session" (Young et al., 2011).

Additionally, storytellers reported increased listening, anticipation, recognition and decreases in reaction time through repetition of MSST in the listeners with profound intellectual disabilities across quantitative and qualitative findings: "the reaction time went from 10-30 seconds to 5-10 seconds" (author, ten Brug et al, 2013), (ten Brug et al., 2013; Young et al., 2011). A storyteller in Young et al's., (2011) study reported "She is always listening carefully when the story is being read to her….She anticipated the storyline. She was very responsive" (Claire's teacher).

2.5.1.2 Interaction. Increased interaction by the listener was reported quantitatively and qualitatively, seeing them move from passive observer to interacting with stimuli through MSST repetition: “Aden started making sounds along with the triangle and started holding the rainmaker instead of just listening to it” (author, ten Brug et al., 2013) (ten Brug et al., 2013; Young et al., 2011). This was observed and reported qualitatively through storytellers describing increased interest in materials offered through purposeful touching and exploration: “she showed a strong interest in materials and touching these (Rebecca’s mother), (Young et al., 2011). Increased motor interaction led to improved fine motor exploration and manipulation of stimuli (ten Brug et al; 2013). Improved interaction was also reported outside of MSST sessions, where the listeners were increasingly able to tolerate personal care activities, such as hair brushing, with MSST repetition: “During and since the time of the project, Claire has now become more tolerant of having her hair brushed” (Claire’s teacher), (Young et al., 2011). Increased interaction was demonstrated in listeners expressing preferences and with more clarity over time: “At T2, according to Brit, Bob anticipated the toy pig. Before the pig sound was presented, Bob would sit up, laugh, and turn his head towards the pig” (author, ten Brug et al., 2013), (ten Brug et al., 2013; Young et al., 2011).

2.5.1.3 Social Behaviour. Improved social engagement, communication and responsivity was reported qualitatively for those with profound intellectual disabilities (Young et al., 2011). This was identified through increased vocalisations, teamwork with the storyteller, eye gaze, clearer responses and expressing preferences: “Prolonged hand holding and eye contact – very positive response. There has been good team working and Rebecca showed positive interaction with the storyteller” (Rebecca’s occupational therapist), (Young et al., 2011). However, whilst some participants’ eye gaze to storyteller remained consistent over time, some participants decreased towards storyteller and increased in direction to sensory stimuli (Young et al., 2011). MSST were reported qualitatively to create a sense of connection and togetherness: “it gives them a chance to come together as a group” (interviewee) (Preece & Zhao, 2015). One family qualitatively shared reading of MSST was the first time their child with profound learning disabilities had been willing to share an activity with their sibling, enabling them to read as a family: “They do not normally share things but Joseph allowed his brother to ‘share’ hearing his story. For the first time, the three of us read the story as a family” (Joseph’s mother) (Young et al., 2011). Connections between the MSST and real life were shared by storytellers qualitatively reporting increased social engagement and tolerance of social outings outside of MSST sessions: “she is now tolerating social outings with family members which she had not done before” (Claire’s mother), (Young et al., 2011).

2.5.1.4 Decrease in Maladaptive and Stereotypical Behaviour. Quantitative findings and qualitative accounts from storytellers reported a decrease in agitation and anxiety exhibited from the person with profound disabilities during and between sessions of MSST (ten Brug et al., 2015[a]; Young

et al., 2011). Additionally, qualitatively there was a reported decrease in stereotypical behaviour within sessions (Young et al., 2011).

> He has enjoyed the story and everything in it from the word 'go'. This was particularly evident on a couple of occasions when Joseph was extremely anxious and showing physical, challenging behaviour and immediately after this settled very quickly into his story. (Joseph's mother, Young et al., 2011).

Further, people with profound intellectual disabilities were able to participate in activities targeted by the MSST, like dental visits, and spent reduced amounts of time in behaviours that were previously problematic, increasing independence and participation in other enjoyable activities (Young et al., 2011).

> She recognised the symbols/photo in relation to the task…. It has helped Rebecca more than I thought it would and helped her toilet routine which is now more structured, simple and focused…. We do not need to sit her on the toilet for lengthy periods of time any more. Dad does not play the guitar in the toilet now. (Rebecca's mother, Young et al., 2011).

One listener became more accepting over time of the MSST, a favoured activity, ending: "she definitely anticipated the storyline and came to know and accept when the storyline ended" (Sarah's teacher), (Young et al., 2011). Although quantitative results found there were periods of withdrawn behaviour observed during MSST, they typically occurred in earlier MSST sessions and towards the end of individual sessions, possibly related to fatigue (ten Brug et al., 2015[a]).

2.5.2 Storytellers benefit from MSST

Storytellers reported learning and improving their understanding of the person with profound intellectual disabilities. They shared how MSST allowed them to adapt their practice to better meet the needs of the person with profound intellectual disabilities. This knowledge was shared with others, developing wider systemic improvements in practice.

2.5.2.1 Learn About and Attune to the Person With Profound Intellectual Disabilities.

Storytellers qualitatively reported through developing and delivering MSST their knowledge and understanding of the person with profound intellectual disabilities improved: "Carol indicated that she had learned a few things about Catrin; olfactory stimuli now seemed to be a definite interest, presenting opportunities for new activities" (author, ten Brug et al., 2013) (ten Brug et al., 2013; Young et al., 2011). This learning increased attunement between the storyteller and the listener but quantitative results indicated was not always consistently applied (ten Brug et al., 2013).

Storytellers reported quantitatively and qualitatively learning listeners needed more time to focus, attend and react to stimuli: "Instead of a focus time of 0–5 seconds (T1), according to Brit, in T2 Bob needed 5–10 seconds to focus" (author, ten Brug et al., 2013). They shared learning about

environmental preferences when delivering the MSST, sitting position for the person with profound intellectual disabilities, lighting and ambient noise levels: “A little environmental noise changed to complete silence” (author, ten Brug et al., 2013). They qualitatively reported discovering capabilities of the person with profound intellectual disabilities they did not know before, for example learning they could distinguish the size and colour of objects through stimuli offered: “Aden could also use large objects (T3), not just small ones (T2). As for his vision, Amber indicated that Aden could distinguish size as well as colour (T3), not just colour (T2).” (author, ten Brug et al., 2013).

MSST allowed the storyteller to learn about sensory preferences. For example, learning how the people they supported responded favourably to olfactory stimuli, bright colours and soft, calm music: “She paid little attention to the lights, while the shimmering fabrics were more popular. Catrin focused on them making soft, agitated sounds” (author, ten Brug et al., 2013). They discovered stimuli which made the person feel uneasy, noting stimuli such as loud noises did not receive positive responses from the listener: “the honking bus scared him slightly, but he would also laugh at it” (author, ten Brug et al., 2013).

2.5.2.2 Developing and Sharing Best Practice. Quantitative findings indicated repetition of MSST allowed adjustment and adaptation of practice to improve the experience and interaction for the person with profound and intellectual disabilities (ten Brug et al., 2013). Preece & Zhao’s (2015) qualitative findings highlighted the importance of sharing knowledge and experience of good practice between professionals from MSST which improves overall care for the person with profound intellectual disabilities: “I think really here it’s about drawing on other teachers, more experienced storytellers ... drawing on their knowledge and their experience and then sharing really good practice” (Interviewee, Preece & Zhao, 2015).

2.5.3 Mutual Enjoyment of MSST

The storytellers in three of the papers described MSST as an enjoyable activity for the person with profound intellectual disabilities. This was demonstrated through descriptions of the persons response and the use of MSST in leisure activities. Storytellers shared they found the development and implementation of MSST an enjoyable experience.

2.5.3.1 The Person With Profound Intellectual Disabilities Enjoys MSST. The three studies including qualitative data reported people with profound intellectual disabilities finding MSST an enjoyable experience (Preece & Zhao, 2015; ten Brug et al., 2013; Young et al., 2011). This was deduced from observations within MSST sessions of increased relaxation, animation, smiling and laughter: "she was very responsive (e.g. became excited, very animated and lots of smiles" (Claire's teacher, Young et al., 2011), (ten Brug et al., 2013; Young et al., 2011). MSST was an activity offered whilst a less favoured task was completed (PEG feeding), or as a reward for completion of less enjoyable tasks, suggesting MSST is perceived as enjoyable for the person with profound intellectual disabilities by care givers: "Or it might be because the whole class had a really good maths lesson and then it's like a post-lesson reward" (Interviewee, Preece & Zhao, 2015).

2.5.3.2 Storytellers Enjoy MSST. Storytellers explicitly reported qualitatively that the process of MSST is enjoyable for them: "I do quite enjoy the idea of coming up with the story myself" (Interviewee, Preece & Zhao, 2015). This mutual enjoyment through joint attention to MSST supports increased attunement and understanding between the storyteller and the listener, improving overall outcomes for care provision.

2.5.4 Development, Implementation and Evaluation of MSST

Papers included in this review highlighted key areas for consideration when developing and implementing MSST. Firstly, the importance of a person-centred approach, individualising the MSST for the listener in mind. Secondly, the various functions of MSST and how they can be applied. Thirdly within session factors including the environment, stimuli and presentation by the storyteller. There is also consideration of possible barriers to MSST and how one should evaluate MSST.

2.5.4.1 Individualisation. All the studies included in this review identified the importance of MSST being tailored to the individual. Key quantitative and qualitative findings suggest the process should involve current knowledge about the person the MSST is being developed for, what their real-world experiences are and appropriateness of the content to the individual (Preece & Zhao, 2015; ten Brug et al., 2013): "Most of the stimuli were chosen by Amber because she knew Aden would respond well" (ten Brug et al., 2013). Qualitative findings highlighted how in educational settings targets and goals were frequently used to guide the development of MSST (Preece & Zhao, 2015).

> I look at the children's assessments and targets ... I look at fine motor, communication, cognitive, and so on. Each specific to each child in the class ... I set my goals, I develop my story. That's how I do it. (Interviewee, Preece & Zhao, 2015).

Young et al., (2011) reported their utility in addressing personal issues, challenges and supporting the individual to overcome them: "His resistance to touch and explore latex items in earlier readings changed dramatically in the final reading" (Fraser's teacher, Young et al., 2011). Whilst it is the storyteller deciding how, when, why and with whom MSST should occur with (Preece

& Zhao, 2015), the listener should always be at the centre of these decisions. Whilst both quantitative and qualitative studies expounded developing MSST from scratch based on the storyteller's knowledge of the listener (Penne et al., 2012; ten Brug et al., 2013; ten Brug et al., 2015[a]; ten Brug et al., 2015[b]; ten Brug et al., 2016; Young et al., 2016), participants in Preece & Zhao's (2015) mixed methods research used commercially produced scripts and stories, which were adapted with individualised symbols, photos and stimuli:

> For me, the scripts are all about the ideas. I always look at adapting it. So given the specific needs of the students I've got, I might also introduce things like choices with PECS symbols or photographs, embellishing the story and making it even more appropriate to the students' needs, their targets and so on. (Interviewee, Preece & Zhao, 2015).

2.5.4.2 Function of MSST. Quantitative and qualitative findings indicate MSST allows for assessment of educational goals, skills, targets and feedback on adaptations storytellers make (Preece & Zhao, 2015; ten Brug et al., 2013):

> When we devise or adapt a story we integrate their targets in it. So ... somebody's target could be to hold the flashlight for three to five seconds ... and somebody else's would be to move a blanket to look for something underneath it. Throughout the story we try to find as many times ... like three to four times for them to practice that skill. And somebody else's might be to reach for something that is of interest or to track the light. We use the targets and plan around them. (Interviewee, Preece & Zhao, 2015).

Observations and qualitative reports demonstrated MSST is versatile and can be used across a range of individuals, setting and contexts (Preece & Zhao, 2015). Both quantitative and qualitative findings indicated MSST can be used for developing cognitive functions, including attention, (ten Brug et al., 2016; ten Brug et al., 2015[a]; ten Brug et al., 2015[b]) concentration, anticipation and task initiation (Preece & Zhao, 2011). Qualitative data indicated MSST has the capability to support learning (Young et al., 2011), socialisation, entertainment and pleasure (Preece & Zhao, 2015). Preece & Zhao (2015) highlighted MSST provides choices for people with profound intellectual disabilities where they are often not given. This allows the listener to be empowered and develop agency (Preece & Zhao, 2015): "I don't want it ... push it away. That's good because they are telling you that they don't like it. It's all communication. I'll know if they don't like it and that's very important ... because often they won't be given those choices" (Interviewee, Preece & Zhao, 2015). New skills can be introduced and practiced over time in MSST (Preece & Zhao, 2015). Furthermore, storytellers can create opportunities to expose listeners to novel stimuli in a safe environment, to learn more about their responses and preferences (ten Brug et al., 2013).

2.5.4.3 Environmental Factors. Environmental factors depend on the specific listener and will inform how the MSST is individualised. ten Brug et al's., (2013), mixed methodological findings detailed the following factors and the listeners' preferences as having impact on the delivery of MSST: time of day; lighting; and ambient noise: "Lights needed to be dimmed (T2) instead of coming from the right" (author, ten Brug et al., 2013). While MSST are versatile and can be offered indoors, outdoors, across settings and contexts (Preece & Zhao, 2015), qualitative data indicated understanding the individual's preference for space, size of room, positioning within the space and seating is key to them accessing MSST: "Because she preferred brightly lit rooms, ideally she sat near the window" (author, ten Brug et al., 2013).

2.5.4.4 Stimuli. Stimuli should be chosen by someone who knows the listener well (ten Brug et al., 2013). Personal preferences should be considered when selecting stimuli and how stimuli might contribute to skill acquisition or goal related behaviour (ten Brug et al., 2013).

ten Brug et al., (2013) detailed considering individual preferences for auditory, olfactory, tactile and visual stimulation based on quantitative and qualitative findings. Qualitative data indicated listeners' abilities should be held in mind, considering any visual or auditory impairment, fine and gross motor skills: "I look at the children's assessments and targets…I look at fine motor, communication, cognitive and so on" (Interviewee, Preece & Zhao, 2015), (Preece & Zhao, 2015; ten Brug et al., 2013). This will inform what is presented and how. For example, understanding visual impairment will allow the storyteller to select and present stimuli in the correct field of vision and distance for the listener to engage: "It was important for Catrin to be offered stimuli in the middle of her visual field that remained over 40 cm away" (author, ten Brug et al., 2013). Everyday items can provide a variety of sensory experiences, including, musical instruments, noisy toys, mirrors, torches, pieces of fabric, music, perfume and egg timers (ten Brug et al., 2013).

2.5.4.5 Presentation. The storytellers participating in most of the studies, quantitative and mixed methods, received training on MSST theory, development and delivery (Penne et al., 2012; ten Brug et al., 2013; ten Brug et al., 2015[a]; ten Brug et al., 2015[b]; ten Brug et al., 2016; Young et al., 2016). The cost of training and the advantages it provides should be assessed. Preece & Zhao (2015) found through qualitive data most of the storytellers in their sample had not received specific training, but had learnt through observation, mentoring from more experienced staff and their own natural style:

> A minority had undertaken training in MSST, while at university, or via their local authority or a charity. Most, however, had not, and based their practice on their natural storyteller style, on their observations of others or on mentoring from more experienced staff (author, Preece & Zhao, 2015).

Individual factors for the storyteller presentation style include tone and volume of speech (ten Brug et al., 2013).

A key component for the storyteller is presentation of stimuli. Quantitative results found active presentation of stimuli was positively correlated with active attention in the person with profound intellectual disabilities, whereas passive presentation was associated with more passive attention to the MSST and the storyteller (ten Brug et al., 2013). Furthermore, quantitative findings highlighted how long stimuli should be presented for, allowing the listener to focus, and respond before moving to the next stimulus object (ten Brug et al., 2013).

Quantitative data showed consistency of MSST between sessions is important to avoid confusion for the listener (ten Brug et al., 2013). However, Penne et al., (2012) reported no significant effect of storyteller's characteristics, including role, age, experience with profound intellectual disabilities or the specific listener. This highlights the accessibility of MSST for all people working with this population.

Quantitative and qualitative findings indicated structure of the MSST should be considered, in terms of number of sentences, length of sentences and corresponding stimuli to be offered (ten Brug et al., 2013; ten Brug et al., 2015[b]). Whilst consistency across MSST sessions is important (ten Brug et al., 2013), quantitatively fidelity to the text appeared to have no effect on attentiveness of the listener (ten Brug et al., 2015[b]), allowing storytellers to respond in the moment to the listener, without being wedded to a script.

2.5.4.6 Barriers. Qualitative data reported individualised MSST can be time intensive for the storyteller and be impacted by wider service staffing levels: "Key external factors were time, resources and staffing levels" (Interviewee, Preece & Zhao, 2015). Therefore, thought should be given to how and where MSST can be developed and implemented. Further, qualitative findings indicated if MSST are to be delivered to groups, due to resource availability, thought should be paid to how MSST can remain interactive and flow, without just passing an object around a circle: "OFSTED requires that your students are doing something all the time. They will no longer accept something going around a group one at a time" (Interviewee, Preece & Zhao, 2015). Cost is also important when developing MSST. ten Brug et al., (2013), highlighted stimuli could be sourced from existing resources mitigating cost.

Another barrier can be the health of the listener. Ill health and hospital admissions for the person with profound intellectual disabilities was qualitatively reported to lead to decreased engagement and interaction with MSST (Young et al., 2011). This was potentially due to being unable to attend sessions, leaving longer periods between sessions reducing consistency and overall impact (Young et al., 2011):

> Both behavioural analysis and interviewee's reports indicated a lack of engagement with the story, and indeed, a decreasing engagement as shown by the behavioural analysis. Throughout the course of the study Martin was ill and off school on several occasions. In addition, his mother, the storyteller, was also ill leading to long delays between sessions. (Author, Young et al, 2011)

2.5.4.7 Evaluation. Quantitative findings highlighted the need for storytellers to attend to the outcome of MSST, adapting and adjusting where necessary (ten Brug et al., 2013). The responses of people with profound intellectual disabilities may not be obvious, so close attention should be paid to their whole body (ten Brug et al., 2013). Six of the seven studies included used video recordings to observe changes in behaviour, with appropriate consent and confidentiality, recordings over time may help identify changes in the person with profound intellectual disabilities. This could contribute evidence of potential positive outcomes, supporting further and more widespread MSST interventions. Adaptation based on feedback will promote ongoing engagement and attunement between the storyteller, MSST and the person with profound intellectual disabilities.

2.6 Discussion

This systematic review used a thematic approach to synthesise data from seven studies (four quantitative and three mixed methodological) to examine and describe the key outcomes of MSST for people with profound intellectual disabilities and those supporting them. Four themes emerged highlighting the benefits of MSST for people with profound intellectual disabilities, those working with people with profound intellectual disabilities, and factors to consider when developing, implementing and evaluating MSST.

This systematic review builds on the evidence found by Humpheson (2024) who found MSST specifically supports positive outcomes for people with profound intellectual disabilities, across social, emotional and cognitive domains. This builds on wider research which indicates providing rich sensory experiences for people with intellectual disabilities promotes engagement, interaction, socialization and enjoyment (Agostine et al., 2022). The findings related to engagement, attention, anticipation and recognition echo findings that MSST improves memory and recall in wider intellectual disability populations (Matos et al., 2015) and gross motor function for people with cerebral palsy (Lee & Kwon, 2022). Further it expands on evidence that MSST promotes engagement, focus and interaction in those with moderate to severe intellectual disabilities (Wolters-Leermakers et al., 2024), Down Syndrome and autism spectrum disorder, (O'Rourke et al., 2021), both of which can present co-morbidly with intellectual disabilities and can be part of a profound presentation.

The theme indicating MSST as beneficial to the person or storyteller working with the person with profound intellectual disabilities is an important finding, demonstrating MSST has the potential

to improve their knowledge, understanding and attunement to the person with profound intellectual disabilities and therefore their own relational experience. This outcome is interesting as it echoes D'Sa et al's., (2024) findings for those working with people with profound intellectual disabilities where the relationship between person and carer is a key factor in being able to holistically meet their needs. If MSST can promote a positive relational experience and attunement between person and carer, this would hopefully improve quality of life and overall psychological wellbeing for the person with an intellectual disability (Nind & Grace, 2024).

The final theme in this review, developing, implementing and evaluating MSST is a call to action for all those working with people with profound intellectual disabilities. It highlights not only the need to develop skills, but an opportunity to recognise people with profound intellectual disabilities as human and psychological beings (Goodley, 2023). It supports other research reporting the need to consider physical space for optimal engagement of people with profound intellectual disabilities (Fitzpatrick & Parker, 2024). ten Brug et al., (2012) published findings on the development, content and application of MSST: guidance, training and ready-made stories are available (Grace, 2023; Grove, 2022). This theme and available literature for those interested in implementing MSST provides confidence and hope for the future of psychological wellbeing for those with profound intellectual disabilities.

2.6.1 Strengths and Limitations of Included Studies

Historically there is a paucity of research including people with intellectual disabilities (de Haas et al., 2022), and this exclusion is even more pervasive for people with profound intellectual disabilities (Kellett & Nind, 2014). Therefore, the inclusion and focus on those people with profound intellectual disabilities is a strength of all the included papers. Additionally, all the papers met the quality assessment markers set out in Hong et al's., (2018) appraisal tool. Each paper explored differing aspects of MSST. This is helpful when considering the broader implications of their utility with people with profound intellectual disabilities, but it means further research is required to explore thoroughly how MSST can be applied as an intervention to support psychological well-being within this population.

A limitation for some of the papers was small sample sizes (ten Brug et al., 2013; Young et al., 2011). Populations of people with profound intellectual disabilities are heterogenous in nature (MENCAP, 2024) and this is compounded by the varying terminology used for people with profound intellectual disabilities (Maes et al., 2019). Therefore, being able to draw conclusions is difficult. However, this is reflective of the population, rather than the specific papers in this review. In addition to this the storytellers had diversity within and between samples. Whilst Penne et al., (2012) found this had no effect on interaction style, individual differences should be considered when drawing conclusions. Young et al., (2011) acknowledged these issues with small diverse samples and

differences in patterns of engagement but suggested this is justification for further multiple single subject designs in further research.

Five of the seven papers included in the review were authored by the same research team and all seven studies were conducted in western Europe, across three countries, Netherlands, Belgium and United Kingdom. Therefore, the impact of culture should be considered when interpreting the data found and the conclusions drawn in this review. It is interesting there were seven papers published between 2011 and 2016 and nothing subsequently. Whilst it is unclear why this is, research into MSST continues with other groups of intellectual disabilities and diagnoses suggesting it could be a transdiagnostic approach.

2.6.2 Strengths and Limitations of the Current Review

A strength of this review is the recognition of a gap in the provision of mental health services for people with profound intellectual disabilities and its aims of considering possible suitable psychological interventions for this population. This review follows the PRISMA guidance (Page et al., 2021) with a protocol published on PROSPERO to ensure transparency and replicability.

A limitation of this review is the small number of papers eligible for inclusion. This highlights the paucity of research done in this area with this population. Due to the small number of papers meeting the inclusion criteria and small sample sizes within these papers, which have heterogenous characteristics, means conclusions should be drawn cautiously. Additionally, thematic synthesis relies on the researcher's interpretation of data and subsequently themes will emerge in the context of their experiences, beliefs and values. To mitigate this the primary researcher undertook regular supervision with the research team and screening of papers for inclusion was undertaken by a second screener. Quantitative data was not qualitised, with results sections lifted verbatim from original papers (Lizarondo et al., 2025). This aided interpretation of the data to be grounded in the original findings and not the researcher's interpretation of them.

Due to the scarcity of research in this area, a mixed methods review supported a more robust approach to answering the research question. Whilst recent guidance was followed to ensure rigor (Lizarondo et al., 2025), it was noted the results sections of papers with mixed methodology were substantially longer than those in purely quantitative papers. Therefore, the proportion of data undergoing thematic synthesis was greater for the mixed methods papers and therefore might skew the conclusions in favour of their findings.

A further limitation is the quality appraisal tool, MMAT (Hong et al., 2018). Whilst this has strengths as a single tool that allows appraisal of methodological quality across designs, it is a relatively new tool that requires further research to explore its discriminatory validity and inter-rater reliability (Hong et al., 2019). This highlights the omission of a second researcher completing the

quality appraisal of the papers included. This was due to time constraints of the wider research project but should be noted when considering the reported quality of the included studies.

2.6.3 Clinical Implications and Further Research

While the conclusions of this review should be considered cautiously given the limitations of both the studies included and the review itself, it suggests MSST could potentially be a positive intervention for people with profound intellectual disabilities. This is an important finding for those working in mental health settings and those working with this population. The findings demonstrate the relational benefits MSST provides to those with profound intellectual disabilities and those supporting them.

In addition, there were positive results observed when MSST was used flexibly. This flexibility included individualised stories, variations in scripts and adapting pre-existing resources for sensory stimuli. This makes MSST both accessible and affordable for people planning to implement them in their everyday practice.

Further, up to date inclusive research is required to understand the potential functions and outcomes of MSST for this population, particularly regarding its acceptability as a psychological intervention. This review focused on MSST which did not include innovative technology, but this is an area which warrants further exploration and has demonstrated improved recall in typically developing populations (Chierichetti & Tombolini, 2023). However, future research should be mindful of the accessibility of MSST with integral technology due to affordability (Grace, 2014). The findings indicate collaborative work and cross discipline training (Devlin, 2024) between educational practitioners implementing MSST as standard practice and those in health care, to channel a broader range of knowledge, skills and experience, may lead to an improved quality of life for those with profound intellectual disabilities (Rossiter et al., 2011).

2.6.4 Conclusion

This review aimed to examine and describe the key outcomes of MSST for people with profound intellectual disabilities and those supporting them. The themes emerging from the data highlight MSST as beneficial to the person with profound intellectual disabilities, the people supporting them and factors to consider when developing, implementing and evaluating MSST. These findings indicate the feasibility and acceptability of MSST as a possible therapeutic psychological approach for people with profound intellectual disabilities should be considered in future research.

2.7 References

Abdi, F., Karamoozian, A., Lotfilou, M., Gholami, F., Shaterian, N., Niasar, A. A., Aghapour, E., & Jandaghian-Bidgoli, M. (2025). Effect of play therapy and storytelling on the anxiety level of

hospitalized children: a randomized controlled trial. *BMC Complementary Medicine and Therapies*, *25*(1), 23. https://doi.org/10.1186/s12906-025-04767-4

Agostine, S., Erickson, K., & D'Ardenne, C. (2022). Sensory experiences and children with severe disabilities: impacts on learning. *Frontiers in Psychology*, *13*, 875085. https://doi.org/10.3389/fpsyg.2022.875085

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Washington, DC.

Bellamy, G., Croot, L., Bush, A., Berry, H., & Smith, A. (2010). A study to define: profound and multiple learning disabilities (PMLD). *Journal of Intellectual Disabilities, 14*(3), 221-235. https://doi.org/10.1177/1744629510386290

British Psychological Society. (2015). *Guidance on the assessment and diagnosis of intellectual disabilities in adulthood*. https://doi.org/10.53841/bpsrep.2015.inf239

British Psychological Society. (2016). *Psychological therapies and people who have intellectual disabilities*. https://doi.org/10.53841/bpsrep.2016.rep106

Bundy, A. C. & Lane, S. J. (2020). *Sensory integration: Theory and practice*. FA Davis.

Chierichetti, C., & Tombolini, E. (2023). Digital multisensory storytelling as educational-didactic methodology. *Italian Journal of Health Education, Sports and Inclusive Didactics*, *7*(2). https://doi.org/10.32043/gsd.v7i2.890

Chinn, D., & Balota, B. (2023). A systematic review of photovoice research methods with people with intellectual disabilities. *Journal of applied research in intellectual disabilities*, *36*(4), 725-738. https://doi.org/10.1111/jar.13106

Colley, A. (2020). The exclusion of learners with PMLD (PIMD) from the policy and practice of inclusive education, in England. *La nouvelle revue-Éducation et société inclusives*, (1), 39-49. https://doi.org/10.3917/nresi.088.0039

Cooke, A., Smith, D., & Booth, A. (2012). Beyond PICO: the SPIDER tool for qualitative evidence synthesis. *Qualitative health research*, *22*(10), 1435-1443. https://doi.org/10.1177/1049732312452938

Department for Education & Department of Health and Social Care. (2015, November 15). *Statutory guidance: SEND code of practice: 0 to 25 years*. https://www.gov.uk/government/publications/send-code-of-practice-0-to-25

de Haas, C., Grace, J., Hope, J., & Nind, M. (2022). Doing research inclusively: understanding what it means to do research with and alongside people with profound intellectual disabilities. *Social Sciences*, *11*(4), 159. https://doi.org/10.3390/socsci11040159

Devlin, E. (2024). Exploring teachers' confidence in addressing mental health issues in learners with Profound and Multiple Learning Difficulties (PMLD) pre and post training. *Wales Journal of Education*, *1*. https://doi.org/10.16922/wje.p5

D'Sa, R., Fletcher, I., & Field, S. (2024). Exploring the experience of working relationships for support workers of adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*, *37*(5), e13285. https://doi.org/10.1111/jar.13285

Fitzpatrick, D., & Parker, R. (2025). What Approaches Described in Research Literature Enhance the Engagement of Children and Young People With Severe or Profound and Multiple Learning Disabilities? A Systematic Literature Review. *British Journal of Learning Disabilities*, *53*(1), 61-73. https://doi.org/10.1111/bld.12619

Fuller, C. (2022). Multi-sensory story-packs. In N. Grove (Ed.), *Storytelling, special needs and disabilities: practical approaches for children and adults* (2nd ed., pp. 112-119). Routledge.

Goodley, D. (2023). Being human as praxis: for people with learning disabilities. *Subjectivity*, 1. https://doi.org/10.1057/s41286-023-00159-6

Grace, J. (2014). Sensory stories: literature for service users. *Learning disability practice, 17*(1), 36-38.

Grace, J. (2023). *Sensory Stories to Support Additional Needs: Making Narratives Accessible Through the Senses* (2nd ed.). Jessica Kingsley Publishers.

Grove, N. (2022). *Storytelling, special needs and disabilities: practical approaches for children and adults* (2nd ed.). Routledge.

Heyvaert, M., Maes, B., & Onghena, P. (2013). Mixed methods research synthesis: definition, framework, and potential. *Quality & Quantity*, *47*, 659-676. https://doi.org/10.1007/s11135-011-9538-6

Hong, Q. N., Fàbregues, S., Bartlett, G., Boardman, F., Cargo, M., Dagenais, P., Gagon, M. P., Griffiths, F., Nicolau, B., O'Cathain, A., Rousseau, M. C., Vedel, I., & Pluye, P. (2018). The Mixed Methods Appraisal Tool (MMAT) version 2018 for information professionals and researchers. *Education for information*, *34*(4), 285-291. https://doi.org/10.3233/EFI-180221

Hong, Q. N., Pluye, P., Bujold, M., & Wassef, M. (2017). Convergent and sequential synthesis designs: implications for conducting and reporting systematic reviews of qualitative and quantitative evidence. *Systematic reviews*, *6*, 1-14. https://doi.org/10.1186/s13643-017-0454-2

Hong, Q. N., Pluye, P., Fàbregues, S., Bartlett, G., Boardman, F., Cargo, M., Dagenais, P., Gagon, M. P., Griffiths, F., Nicolau, B., O'Cathain, A., Rousseau, M. C., & Vedel, I. (2019). Improving the content validity of the mixed methods appraisal tool: a modified e-Delphi study. *Journal of clinical epidemiology*, *111*, 49-59. https://doi.org/10.1016/j.jclinepi.2019.03.008

Humpheson, J. (2024). Sensory approaches for adults with severe or profound and multiple learning disabilities: A systematic literature review. *British Journal of Occupational Therapy*, *87*(3), 129-142. https://doi.org/10.1177/03080226231208717

Hutchinson, N., & Bodicoat, A. (2015). The effectiveness of intensive interaction, a systematic literature review. *Journal of Applied Research in Intellectual Disabilities*, *28*(6), 437-454. https://doi.org/10.1111/jar.12138

Jefferies, L. (2009). Introducing intensive interaction. *Psychologist*, *22*(9), 756-758. https://www.bps.org.uk/psychologist/introducing-intensive-interaction

Kellett, M., & Nind, M. (2014). Ethics in quasi-experimental research on people with severe learning disabilities: Dilemmas and compromises. In *Ethics and Research in Inclusive Education* (pp. 168-175). Routledge.

Lambe, L., Miller, J., & Phillip, M. (2022). Sensitive stories: Tackling challenges for people with profound intellectual disabilities through multi-sensory storytelling. In N. Grove (Ed.), *Storytelling, special needs and disabilities: practical approaches for children and adults* (2nd ed., pp. 143-151). Routledge.

Lee, E. J., & Kwon, H. Y. (2022). Effects of group-activity intervention with multisensory storytelling on gross motor function and activity participation in children with cerebral palsy. *Journal of exercise rehabilitation*, *18*(2), 96. https://doi.org/10.12965/jer.2244028.014

Lizarondo, L., Stern, C., Apostolo, J., Carrier, J., de Borges, K., Godfrey, C., Kirkpatrick, P., Pollock, D., Rieger, K., Salmond, S., Vandyk, A., & Loveday, H. (2022). Five common pitfalls in mixed methods systematic reviews: lessons learned. *Journal of clinical epidemiology*, *148*, 178-183. https://doi.org/10.1016/j.jclinepi.2022.03.014

Lizarondo, L., Stern, C., Salmond, S., Carrier, J., Cooper, K., Godfrey, C., Vandyk, M., Pollock, D., Rieger, K., Apostolo, J., Kirkpatrick, P., Borges Dos Santos, K., & Loveday, H. (2025). Methods

for data extraction and data transformation in convergent integrated mixed methods systematic reviews. *JBI evidence synthesis*, 10-11124. *DOI:*10.11124/JBIES-24-00331

Maes, B., Nijs, S., Vandesande, S., Van Keer, I., Arthur-Kelly, M., Dind, J., Goldbart, J., Petitpierre, G., & Van der Putten, A. (2021). Looking back, looking forward: Methodological challenges and future directions in research on persons with profound intellectual and multiple disabilities. *Journal of Applied Research in Intellectual Disabilities*, *34*(1), 250-262. https://doi.org/10.1111/jar.12803

Matos, A., Rocha, T., Cabral, L., & Bessa, M. (2015). Multi-sensory storytelling to support learning for people with intellectual disability: an exploratory didactic study. *Procedia computer science*, *67*, 12-18. https://doi.org/10.1016/j.procs.2015.09.244

McDonagh, M., Peterson, K., Raina, P., Chang, S., & Shekelle, P. (2013). Avoiding bias in selecting studies. *Methods guide for effectiveness and comparative effectiveness reviews [Internet]*. https://www.ncbi.nlm.nih.gov/books/NBK126701/

McKeon, L., Gildersleeve, J., & Mullens, A. B. (2025). The Strategies of Picture Books as a Mode of Health Communication for Young Children with Coeliac Disease. https://doi.org/10.20944/preprints202503.1626.v1

MENCAP. (2024, November 5). *Profound and multiple learning disabilities (PMLD)*. https://www.mencap.org.uk/learning-disability-explained/learning-disability-and-conditions/profound-and-multiple-learning-disabilities-pmld

National Health Service England. (2024, November 5). *Stopping over mediation of people with a learning disability and autistic people (STOMP) and supporting treatment and appropriate medication in paediatrics (STAMP)*. https://www.england.nhs.uk/learning-disabilities/improving-health/stomp-stamp/

National Institute for Health and Care Excellence. (2016). *Mental health problems in people with learning disabilities: prevention, assessment and management*. https://www.nice.org.uk/guidance/ng54

Nesayan, A., Asadi Gandomani, R., Movallali, G., & Dunn, W. (2018). The relationship between sensory processing patterns and behavioral patterns in children. *Journal of Occupational Therapy, Schools, & Early Intervention*, *11*(2), 124-132. https://doi.org/10.1080/19411243.2018.1432447

Nind, M., & Grace, J. (2024). The emotional wellbeing of students with profound intellectual disabilities and those who work with them: a relational reading. *Disability & Society*, 1-22. https://doi.org/10.1080/09687599.2024.2407819

Office for National Statistics. (2022, November 15). *Population estimates for the UK, England, Wales, Scotland and Northern Ireland: mid-2021.* https://www.ons.gov.uk/peoplepopulationandcommunity/populationandmigration/populationestimates/bulletins/annualmidyearpopulationestimates/mid2021

O'Rourke, J., Main, S., Gray, C., & Lovering, C. (2021). Observations of children with disability during arts-based multisensory story and rhyme activities: Is it all just chimes and perfumes?. *Australasian Journal of Special and Inclusive Education*, *45*(2), 237-251. https://doi.org/10.1017/jsi.2021.8

Paez, A. (2017). Gray literature: An important resource in systematic reviews. Journal of Evidence Based Medicine, 10(3), 233-240. https://doi.org/10.1111/jebm.12266

Page, M. J., McKenzie, J. E., Bossuyt, P. M., Boutron, I., Hoffmann, T. C., Mulrow, C. D., Shamseer, L., Tetzlaff, J. M., Akl, E. A., Brennan, S. E., Chou, R., Glanville, J., Grimshaw, J. M., Hróbjartsson, A., Lalu, M. M., Li, T., Loder, E. W., Mayo-Wilson, E., McDonald, S., (...) & Moher, D. (2021). The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *Bmj 372*. https://doi.org/10.1136/bmj.n71

Penne, A., ten Brug, A., Munde, V., van der Putten, A., Vlaskamp, C., & Maes, B. (2012). Staff interactive style during multisensory storytelling with persons with profound intellectual and

multiple disabilities. *Journal of Intellectual Disability Research*, *56*(2), 167-178. https://doi.org/10.1111/j.1365-2788.2011.01448.x

Preece, D., & Zhao, Y. (2015). Multi-sensory storytelling: a tool for teaching or an intervention technique?. *British Journal of Special Education*, *42*(4), 429-443. https://doi.org/10.1111/1467-8578.12116

Ramamurthy, C., Zuo, P., Armstrong, G., & Andriessen, K. (2024). The impact of storytelling on building resilience in children: A systematic review. *Journal of psychiatric and mental health nursing*, *31*(4), 525-542. https://doi.org/10.1111/jpm.13008

Robinson, D., Moore, N., & Harris, C. (2019). The impact of books on social inclusion and development and well-being among children and young people with severe and profound learning disabilities: Recognising the unrecognised cohort. *British Journal of Learning Disabilities*, *47*(2), 91-104. https://doi.org/10.1111/bld.12262

Rossiter, R., Slaney, K., & Tulloch, L. (2011). Emotion management for young people with severe learning disabilities. *Learning Disability Practice*, *14*(6). doi:10.7748/ldp2011.07.14.6.21.c8617

Samuel, J. & Doswell, S. (2021). The use of intensive interaction in trauma-informed care for people with severe and profound intellectual disabilities. In N. Beail, P. Frankish, & A. Skelly (Eds.), *Trauma and intellectual disability: Acknowledgement, identification and intervention.* (pp. 121-134). Pavilion.

Sheehy, K., & Nind, M. (2005). Emotional well-being for all: mental health and people with profound and multiple learning disabilities. *British Journal of Learning Disabilities*, *33*(1), 34-38. https://doi.org/10.1111/j.1468-3156.2004.00290.x

Stoll, C. R., Izadi, S., Fowler, S., Green, P., Suls, J., & Colditz, G. A. (2019). The value of a second reviewer for study selection in systematic reviews. *Research synthesis methods*, *10*(4), 539-545. https://doi.org/10.1002/jrsm.1369

ten Brug, A., Munde, V. S., van der Putten, A. A., & Vlaskamp, C. (2015[a]). Look closer: the alertness of people with profound intellectual and multiple disabilities during multi-sensory storytelling, a time sequential analysis. *European Journal of Special Needs Education*, *30*(4), 535-550. https://doi.org/10.1080/08856257.2015.1046754

ten Brug, A., van der Putten, A., Penne, A., Maes, B., & Vlaskamp, C. (2012). Multi-sensory storytelling for persons with profound intellectual and multiple disabilities: An analysis of the development, content and application in practice. *Journal of Applied Research in Intellectual Disabilities*, *25*(4), 350-359.https://doi.org/10.1111/j.1468-3148.2011.00671.x

ten Brug, A., van der Putten, A. A., Penne, A., Maes, B., & Vlaskamp, C. (2015[b]). Factors influencing attentiveness of people with profound intellectual and multiple disabilities to multisensory storytelling. *Journal of Policy and Practice in Intellectual Disabilities*, *12*(3), 190-198. https://doi.org/10.1111/jppi.12128

ten Brug, A., van der Putten, A. A. J., Penne, A., Maes, B., & Vlaskamp, C. (2016). Making a difference? A comparison between multi-sensory and regular storytelling for persons with profound intellectual and multiple disabilities. *Journal of Intellectual Disability Research*, *60*(11), 1043-1053. https://doi.org/10.1111/jir.12260

ten Brug, A., van der Putten, A. A., & Vlaskamp, C. (2013). Learn and apply: Using multi-sensory storytelling to gather knowledge about preferences and abilities of children with profound intellectual and multiple disabilities–three case studies. *Journal of intellectual disabilities*, *17*(4), 339-360. https://doi.org/10.1177/1744629513508384

Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC medical research methodology*, *8*, 1-10. https://doi.org/10.1186/1471-2288-8-45

Tillott, S., de Jong, G., & Hurley, D. (2024). Self-regulation through storytelling: A demonstration study detailing the educational book Game On for resilience building in early school children. *Journal of moral education*, 1-20. https://doi.org/10.1080/03057240.2024.2403992

University of Cambridge. (2024, November 15). *Faculty of Education News: "It's almost as if they don't exist": Education policy failing to account for learners with PMLD.* https://news.educ.cam.ac.uk/its-almost-as-if-they-dont-exist

Wolters-Leermakers, N., van Wingerden, E., Gerkema-Nijhof, R., & van Balkom, H. (2024). Sensory Enhanced Interactive Storytelling Technique (SEIS-T): Supporting active communication through short, multimodal narratives. *International Journal of Disability, Development and Education*, 1-20. https://doi.org/10.1080/1034912X.2024.2408673

Young, H., Fenwick, M., Lambe, L., & Hogg, J. (2011). Multi-sensory storytelling as an aid to assisting people with profound intellectual disabilities to cope with sensitive issues: A multiple research methods analysis of engagement and outcomes. *European Journal of Special Needs Education*, *26*(2), 127-142. https://doi.org/10.1080/08856257.2011.563603

3 Chapter 3: "The trauma and the hardship that goes with having a disabled baby is a really heavy load, if that's where your journey ends": Mothers to first born children with profound intellectual disabilities making decisions about further children: An Interpretative Phenomenological Analysis.

This paper has been prepared in the format required for the *Journal of Applied Research in Intellectual Disabilities*.

3.1 Abstract

Background: There is limited knowledge about how caring for a first-born child with profound intellectual disabilities impacts a mother's decision to have subsequent children. This research aims to understand this lived experience.

Method: Seven participants were interviewed about their experiences. Transcripts were analysed qualitatively using interpretative phenomenological analysis.

Results: Six group experiential themes emerged: 'Grief and loss driving you to have another child', 'Isolation from the world around you', 'The burden of care splits you in two', 'The weight of responsibility of bringing another child into the world when your first has profound intellectual disabilities', 'Finding meaningful support is down to you' and 'Siblings heal and bring joy'.

Conclusions: These findings highlight difficulties faced by mothers. It demonstrates the need for professional support and the importance of connection with peers to develop coping strategies and building resilience.

1.2 Accessible summary

- Not much is known about how mothers who have a first-born child with profound intellectual disabilities decide whether to have more children.
- This research explores that experience and what support mothers might need.
- The study found that mothers need support from professionals and from other parents.
- More research is needed to understand how professionals can help.

KEYWORDS

Profound Intellectual Disabilities, Mothers, Children, Interpretative Phenomenological Analysis

3.3 Introduction

People with profound intellectual disabilities are a heterogenous group of individuals (American Psychiatric Association, [APA], 2013), who each have multiple disabilities, with the most

significant being a profound intellectual disability (Doukas et al., 2017). Other complex difficulties include motor, sensory, communication and medical complications (Nakken & Vlaskamp, 2007). The combination of these leads to severely impaired adaptive functioning resulting in pervasive support needs (Schalock et al., 2021).

The burden of care typically falls to parents (Brekke & Alecu, 2023). This often leads to them operating in survival mode and neglecting their own personal needs within the familial system (Geuze et al., 2023). Research shows this burden is felt more acutely for parents of younger children (Tadema & Vlaskamp, 2010) and impacts quality of life, specifically support for their emotional wellbeing (Lahije et al., 2023). The impact of care needs on parents is widespread, negatively impacts physical health, emotional and psychological wellbeing and social health (Shalali et al., 2024).

Research highlights even within countries where gender equality is promoted mothers typically provide more care, which may reduce their working hours and income, when compared to male counterpart's post birth (Wondemu et al., 2022). When comparing experiences of mothers who care for children with disabilities to mothers who do not, there are higher rates of depression, anxiety, musculoskeletal disorders and sleep disorders (Brekke & Alecu, 2023). Higher rates of mental and physical health issues, with an overall increased rate of mortality have been found in mothers caring for children with life limiting conditions (Fraser et al., 2021). Whilst neither study focuses exclusively on caring for a child with a profound intellectual disability, including a broader spectrum of less severe disabilities, these findings could be cautiously recognised as a conservative estimate for those mothers caring for children with the most complex and pervasive needs.

There is limited understanding how caring for a child with a profound intellectual disability impacts a mother's decision making for having subsequent children. In typical populations, for couples who have one child, parental anxiety about their ability to cope, despite support from their partner, decreased the probability of a couple going on to have another child when compared to those without this anxiety (Moilanen et al., 2024). It is therefore possible increased rates of anxiety and other mental health issues may impact parents making the decision to have further children where their first born has profound intellectual disabilities. In a study exploring the wider impacts of the burden of care parents reported having a child with profound intellectual disabilities did influence their decisions to have more children (Geuze et al., 2023). However, Geuze et al., (2023) did not explore the effect of birth order or differentiate between mothers' and partners experience.

To date there is no research exploring the lived experience of mothers whose first-born child has profound intellectual disabilities and how this shapes their decision about having further

children. Aside from the fact they are typically the ones providing care, mothers experience pregnancy, labour and post-natal physical recovery. Therefore, the decision to have another child may impact them disproportionately, whilst they continue to care for their first-born with profound intellectual disabilities.

3.3.1 Research Questions

- What are mother's experiences of parenting a first-born biological child with profound intellectual disabilities?
- How does this influence subsequent decisions about having further children?

1.3 Method

3.4.1 Position Statement

The primary researcher was a trainee clinical psychologist who previously worked across settings supporting individuals with profound intellectual disabilities and their families for over two decades. This experience provided an awareness of the challenges faced and the lack of previous research in this area. They were motivated to explore this further following their own experience of motherhood.

3.4.2 Design

Due to the lack of preexisting knowledge in this area, interpretative phenomenological analysis (IPA) was selected to understand how individuals make sense of their lived experience within their personal and social worlds (Smith & Nizza, 2022). This allowed examination of the phenomenon without predefined theoretical categories (Smith & Nizza, 2022).

Semi-structured interviews were conducted in line with an idiographic approach, collecting detailed first-person accounts, which may get lost in a larger, heterogenous sample, before making between comparisons (Smith & Nizza, 2022). A topic guide was developed for interviews containing open questions, with follow-up questions and prompts (see Appendix B). To ensure relevance and acceptability, the topic guide was developed with experts by experience (EBE), including two mothers of first-born children with profound intellectual disabilities and a Clinical Psychologist working in a community intellectual disabilities service. The EBE's were all reimbursed for their time.

3.4.3 Participants

Purposive sampling was used to recruit homogenous participants (Smith & Nizza, 2022). Recruitment aimed for five to eight participants to allow detailed analysis (Smith & Nizza, 2022; Turpin et al, 1997). Table 8 summarises inclusion criteria.

Table 8

Summary of Inclusion and Exclusion Criteria

Inclusion Criteria	Exclusion Criteria
Biological mothers	Not biological mother
First-born child has a profound intellectual disability	First-born child has no profound intellectual disability
First-born child is the first experience of parenting role	Had previous experience parenting stepchildren
Profound intellectual disability has been present from birth	Profound intellectual disability not attributable to incident post birth
First-born child is still alive	First-born child has passed away [a]
English speaking	Non-English speaking
Resident of the United Kingdom	Not living permanently in the United Kingdom

[a] Profound intellectual disabilities may encompass life-limiting conditions, which can lead to parents out living their children. Parents who had lost their children were not included in this study as bereavement is not the focus of this study, and their experiences, while hugely valuable, may turn the focus away from the research question.

Recruitment was advertised via social media, specialist schools and community settings. Potential participants emailed for further information and were sent a Participant Information Sheet (see Appendix C) and an Informed Consent Form to complete (see Appendix D). If clarity about eligibility was required regarding the definition of profound intellectual disabilities, diagnostic criteria from the Diagnostic and Statistical Manual of Mental Health Disorders V (APA, 2013; see appendix E) was provided.

The sample consisted of seven women with a first-born child with profound intellectual disabilities (see Table 9). All participants received a twenty-pound voucher as a thank you for their time.

Table 9

Participant Demographics

Participant	Age	Number of children	Age of children
Indira	31-40	2	6 years & 8 months
Harriet	51-60	3	17, 15 & 13 years
Gloria	51-60	1	17 years
Frida	51-60	2	16 & 14 years
Cleo	31-40	1	2 years
Etta	31-40	3	11, 8 & 7 years
Elizabeth	-[a]	6	42, 39, 37, 34, 28 & 11 years

[a] Did not share age

Note. Pseudonyms used for participant anonymity. Data on ethnicity was excluded due to the combination of age and number children potentially breaching participants confidentiality.

3.4.4 Ethics

The study was granted ethical approval by the University of Southampton's Ethics Committee (ERGO number 93086, see Appendix F).

3.4.5 Procedure

Interviews between the primary researcher and participant were conducted and recorded via MS Teams video call, lasting between 34-56 minutes (*M* = 43.42 minutes). Due to potentially emotive content participants were told they could take a break, stop the interview or decline to answer any questions. Following the interview participants were given a verbal debrief and a debrief form, including signposting information for support services (see Appendix G). Interviews were transcribed verbatim by the primary researcher and anonymised. All recordings and transcripts were stored on a password protected laptop computer.

3.4.6 Analysis

All participants and individuals mentioned in the data were given a pseudonym. Diagnoses and geographical locations have been redacted to ensure anonymity. The analysis followed the five main IPA phases as outlined by Smith & Nizza (2022). Initially, multiple readings took place, whilst re-listening to recordings for emersion in the data. Exploratory notes on content, use of language, and context were made to the right of the transcript. Next personal experiential statements that captured the participants' perception of their experience were formed and recorded to the left of the transcript. Connections between statements were formed and compiled into a table. These clusters were given a personal experiential theme (PET) supported by direct quotes. This was

repeated for each participant, with 'bracketing' between each transcript (Smith & Nizza, 2022). Cross-case analysis then examined common patterns and idiosyncratic differences between participants, observing how they revealed and clarified aspects of one another (Nizza et al., 2021).

3.4.7 Quality Assurance

Yardley (2000) identified four characteristics of good quality qualitative research. Firstly, sensitivity to context, was met by IPA highlighting individual experiences as unique and prevented comparisons to other populations that would have limited exploration. The second, commitment and rigor, was demonstrated through a supervisory team with expertise in IPA and intellectual disabilities. The primary researcher undertook IPA training, delivered by a contemporary expert. The third, transparency and coherence, was illustrated through a detailed description of methods and analysis, including direct quotes to evidence the theme development and narrative construction across participants (Nizza et al., 2021). The primary researcher engaged in 'bracketing', where they kept a reflective diary (see Appendix H) throughout data collection, recording initial thoughts and feelings following each interview for reflexivity (Smith & Nizza, 2022). Bracketing also included the primary researcher participating in peer supervision with IPA researchers and the supervisory team to promote reflexivity. The primary research also took breaks between interviews to ensure their engagement with subsequent participants remained free of prejudices and judgements occurring based on previous interviews as possible. This involved not only engaging in reflexive practice, but also other non-research related activities.

Once the themes emerged, they were checked with the supervisory team to ensure interpretations were grounded in the data. The fourth, impact and importance, was reflected in narratives shared by the participants and their desire for further support for those embarking on this unique experience of motherhood.

3.5 Results

Analysis resulted in six group experiential themes (GETs) about motherhood with a first-born child with profound intellectual disabilities and making decisions about having further children. These were: 'Grief and loss driving you to have another child', 'Isolation from the world around you', 'The burden of care splits you in two', 'The weight of responsibility of bringing another child into the world when your first has profound intellectual disabilities', 'Finding meaningful support is down to you' and 'Siblings heal and bring joy'. Each theme is described and supported with participant quotes. Each GET and subthemes are listed across participants in Table 7.

Table 10

Group Experiential Themes Across Participants

Group experiential theme	Subtheme	Indira	Harriet	Gloria	Frida	Cleo	Etta	Elizabeth
Grief and loss driving you to have another child	Loss and grief for the motherhood you had hoped for	X	X	X	X	X	X	X
	Craving a 'normal' experience	X	X	X	X	X	X	-
Isolation from the world around you	Loneliness	-	X	X	X	X	-	-
	Others making hurtful comments	-	X	-	-	X	X	X
	You avoid other people	-	X	X	X	X	X	-
The burden of care splits you in two	The care needs of your first are overwhelming and unrelenting	X	X	X	X	X	X	X
	You need to split yourself to meet your children's needs	X	X	X	-	X	X	X
The weight of responsibility of bringing another child into the world when your first has profound intellectual disabilities	Fear and guilt of bringing another child with profound intellectual disabilities into the world	X	X	X	X	X	X	X
	Needing reassurance	X	X	-	X	X	X	X
Finding meaningful support is down to you	Parents need timely and appropriate support	-	X	X	X	X	X	-
	Seeking out support for yourself	-	X	X	X	X	-	X
	Peer support in invaluable	-	X	X	X	X	X	X
Siblings heal and bring joy		X	X	-	X	-	X	X

Note. Pseudonyms used for participant anonymity.

3.5.1 Grief and Loss Driving you to Have Another Child

The first GET highlights the profound sense of loss and grief for the motherhood participants had hoped for, which they felt the disability took from them. They shared how this initiated a desire for a perceived 'typical' experience of motherhood and pushed them on to have another child.

3.5.1.1 Loss and Grief for the Motherhood you had Hoped for. All participants expressed an intense sense of loss for the motherhood they had hoped for. One described the experience of receiving her child's diagnosis being like a physical assault, an expulsion from all she knew and expected from motherhood. The false start in the second sentence emphasises the lack of choice and shock: "you get pushed off the edge of the world. It's like, what you know, everything you thought, isn't going to be" (Frida).

Similarly, another described the moment of finding out her child had a profound intellectual disability as an 'out of body experience', a complete disconnect of consciousness from her body, a phrase used to describe traumatic events, illustrating the pervasive impact this had on her. This was highlighted in the false starts in the second sentence demonstrating how hard it was to find the words to describe the impact of this experience:

> that news, was like an out of body experience, if I'm honest with you. And that, that, it was like a massive grief actually for, for, a loss of a, of a parenthood and a child that you will never have. (Gloria)

3.5.1.2 Craving a 'Normal' Experience. Five participants reported a drive to have another child, which went beyond a typical motherly desire. They described a recognition their parenting experience was so different from others, with typical children, and they craved that experience too: "there was also this desperate need to have an experience which I knew other women had, that I didn't have" (Harriet). Frida explained this was important for her personal fulfillment, to go on and have a different parenting experience, one which was not dictated by complex care needs: "We didn't want our whole experience of parenting to be parenting Sofie with her complex needs" (Frida).

Cleo, who was still deciding about further children, compared herself to peers expanding their families. This comparison and false start in the second sentence conveyed a sense of longing for the life she and her partner had planned but could not have at this time: "a lot of the friends that we've met from our sort of NCT cohort and what not, that are now having second children. So, it has, it has felt like this is the time that we would have been" (Cleo).

3.5.2 Isolation From the World Around you

In the second GET six participants discussed how they experienced loneliness after their first child was born. They shared feeling unsupported by those around them. This was compounded by

hurtful comments made by friends and family, leading to themselves avoiding others to prevent further pain. This resulted in feeling isolated from the world around them.

3.5.2.1 Loneliness. Frida described the distancing from friends as an inevitable process due to their circumstances: “even our closest friends, you’re always in a, in a different boat than them. And. Yeah. You look at the world from a different perspective. You can’t help it” (Frida). For others the loss of friendship was tinged with hurt: “when he was diagnosed, how people, like I said melted away” (Harriet). The phrase “melted away” implied spending time with Harriet and her child post diagnosis exposed others to an unbearable heat they could not tolerate. Gloria shared this sense of abandonment by those you thought would and should have cared for you “But no one checks in on you, as a parent. Nobody checks in on you. Nobody. Nobody.” (Gloria). The repetition of ‘nobody’ emphasises how alone she felt, when she needed people the most. Harriet shared the importance of social connections and friendships during motherhood, and without them, in the context of a high burden of care, you feel you do not have the resources to have more children: “That affects your decision as well, because you don’t have a support network of friends” (Harriet).

3.5.2.2 Others Making Hurtful Comments. The mothers explicitly spoke about how comments made by others caused pain and the need to develop resilience in the face of this:

> It's, it's, like walking around in the supermarket and in the summer holidays and youngsters running up and down the aisles calling your child a spastic. If you can, if you can find something funny in that, you'll survive. But if you, if you, if everything upsets you, then it will be a struggle. It will be hard. (Elizabeth).

They described how the pain from each comment has a cumulative effect and impacts not only in the moment, but they carry these slights with them, and it exacerbates the burden they already bear:

> people say stuff and it’s like paper cut, paper cut, paper cut. You have to just keep going, but all those paper cuts really wound you in the end. And all of that stuff you carry with you when you have the next child and that impacts. (Harriet)

They shared others’ lack of understanding, and dismissal of their situation, increased the impact it had on them. Leaving them to feel like they had nowhere to turn, when even their closest friends could not offer empathy and compassion: “Like one of my friends when Kiri was first suspected of having microcephaly, she actually said like, oh, what, like a pinhead? And I was like, oh, right. And that was like, one of my closest friends.” (Cleo).

3.5.2.3 You Avoid Other People. The participants spoke about how others' lack of understanding led to social disengagement. It made interactions feel devoid of empathy and meaning, leaving the mothers' needs unmet and so they stopped speaking to others about their children entirely: "so I just don't really even talk about it anymore, to them" (Cleo). The pause before "to them" indicates the sense of feeling 'othered' by the experience. Another shared how avoidance stemmed from self-preservation. If they did not spend time with other people, they could not say things which would cause harm: "I was worried that they would say something that either disappointed me or really stung me" (Harriet). For Etta, avoidance was due to the lack of belonging she felt because of how different her baby was in comparison to other typical babies: "he's completely different from everybody. And you just don't return back to these groups" (Etta). Where Etta refers to herself in the second person in the second sentence, she creates a sense of distance between herself and the experience, indicating a possible need to protect herself from the emotions she felt at that time.

3.5.3 The burden of care splits you in two

For the third GET all participants shared the care needs of their first child were overwhelming and unrelenting. Six described how the continuing care needs impacted their ability to mother their other children. There was a sense this split of caring responsibilities could never be equal and would be at the cost of subsequent children.

3.5.3.1 The Care Needs of Your First are Overwhelming and Unrelenting. All participants described how the level of complex care needs their first child had significantly impacted their life. They shared the need to prioritise the child's needs, and described the sacrifices they had to make, the toll on their mental health and the resilience needed to survive in this world of motherhood. Frida described "we felt that we were up to our eyeballs with, with it" (Frida). This physical metaphor conveyed how overwhelming the burden of care needs were, to the point it was blocking off their nose, mouth and ability to breathe. The participants shared the unrelenting nature of care needs, describing how it was outside the world of typical care a baby needs, and how sustained this was, with no reprieve: "It was not typical, and it did not stop. It went on for years, having to do things like that" (Harriet). Harriet's emphasis on the word "not" emphasised the relentlessness of her experience.

The mothers shared how this burden of care came at significant personal cost: "put my life on hold for that. And that's not easy to do" (Gloria). Where even with good personal resources the care is consuming: "Even I am struggling, like I am well educated" (Indira). Etta shared the deep psychological impact the care had on her, finding fulltime work easier than life at home: "I actually

worked full time. I did three twelve-hour shifts. But I think in hindsight that was because I was quite depressed with Jake, and it was easier to be at work than it was with Jake" (Etta).

3.5.3.2 You Need to Split Yourself to Meet Your Children's Needs. All participants shared having further children led to splitting themselves to meet all their children's needs: "still now, I'm like 70% Avi's mum and 30% Aviraj's mother" (Indira). For one mother the realisation of their child's care needs made it apparent she would not be able to have another child "I could not in my mind, see how I was going to split myself and be able to parent more than him, because of all the needs that, that, he required" (Gloria). They described how this splitting of self leads to guilt and a disparity between the mother you want to be versus the parent you must be because there is no flexibility in the care needs: "I know it's not nice, but it is what it is. Like we don't make a light decision of, to be in XXXXX, away from our other children either" (Etta).

3.5.4 The Weight of Responsibility of Bringing Another Child Into the World When Your First Child has Profound Intellectual Disabilities

A fourth GET across participants was the weight of responsibility linked to deciding to have further children who could have disabilities or choosing to place them into the role of a sibling carer. They shared how they sought reassurance through testing and medical advice. For those who went onto have further children, they felt it was necessary to provide the second child with a sibling with whom they could share this unique experience. They described how other factors contributed to the urgency of the decision. One participant highlighted the pressure of the decision by sharing how a life changing diagnosis, and treatment incompatible with pregnancy felt like a reprieve, as currently the choice has been made for her: "it has taken the pressure of deciding whether to have another child" (Cleo).

3.5.4.1 Fear and Guilt you will Bring Another Child with Disabilities Into the World. All seven participants described fear of future children having disabilities: "Obviously, we were worried about how, going into birth and having another child, and if they would be disabled" (Etta). For some this stemmed from their capacity to cope with additional care needs. For others fear was combined with guilt for choosing to bring another child with profound intellectual disabilities into the world, where they may suffer: "He struggles. He was struggling. And I thought, oh no, I can't do that again to another baby." (Harriet).

Participants shared how fear infiltrated their subsequent pregnancies, and time spent with their newborns. They described how anxiety pushed them to seek reassurance during extra scans and repeatedly asking if the sonographer could see anything concerning "we had extra scans and stuff. And I'd be like, can you see cerebral palsy? Can you see anything?" (Etta). This fear spilled over into the postnatal period, reliving and perpetuating the loss from their first experience: "I missed out on what should be just a really joyous experience. They were, yeah, blighted by the first experience

of just huge trauma and fear and worry" (Harriet). The false start in the second sentence of the quote from Harriet emphasises how difficult it was for Harriet to think about this experience.

Cleo, who was yet to decide, became tearful when sharing the potential guilt about knowingly placing siblings into a caring role and imposing this responsibility on them: "when it's your first, you know, you are, to have any more, you're already considering that they would then have sibling carer roles" (Cleo). Other participants recognised the burden on a single sibling and felt a responsibility to have a third for the second to share the experience with: "she will have a sibling that she can also share this whole family experience together" (Harriet). Another acknowledged due to their eldest's life limiting condition their second would become an only child. So, by having a third, the second would have someone to go through difficult times with: "when we lose him. I keep thinking, well, maybe they'll have each other and can go to the pub together or, you know, talk to each other" (Etta).

3.5.4.2 Needing Reassurance. Participants who had gone on to have further children reported needing reassurance from medics and genetic testing before being able to go ahead: "we wanted to have genetic test, then, we wanted to confirm if it is not genetic" (Indira). Others shared whilst there were tests, they had "quite a lot of risk attached" (Frida). So, choosing to undergo procedures for reassurance was not the easy option. Furthermore, test results took a long time to process, so were not a simple solution to a mother's fears. The time taken for tests was felt more acutely in the context of their 'biological clock': "I was early thirties and it's like, if we are going to do it, we better do it" (Frida). The shift from "I" to "we" indicates Frida feeling the burden of time as an individual experience, but that the consequences stretched beyond her, to her partner and both their futures.

However, tests could be expedited if you are already pregnant. This meant pregnancy without knowing the extent of the risk, potentially leaving them in another unbearable situation if the foetus was found to be disabled: "they, can rush it through in four weeks if you're pregnant, and expecting another one. So, which is a bit late by then, isn't it?" (Cleo). Cleo using third person pronouns to describe being pregnant and expedited testing distances herself from that situation, highlighting how intolerable it would be. For one participant there was no test available, leaving no choice but to take the risk: "It was a leap of faith, a huge one, because you know, you just don't know" (Harriet). Again, the use of third person pronouns enables Harriet to create distance between herself and a period of time that filled with fear and anxiety.

3.5.5 Finding Meaningful Support is Down to you

In the fifth GET all participants spoke to the lack of meaningful support for their mental health and wellbeing from professionals. Support was not always offered, and when it was there was

an absence of skills, knowledge and appropriate treatment options. This led to seeking out private therapists and connecting with other parents of children with profound intellectual disabilities.

3.5.5.1 Parents Need Timely and Appropriate Support. Participants expressed incredulity there was no emotional and psychological support offered during the discovery their child had profound intellectual disabilities, when they desperately needed it: “we couldn’t believe that there was no support, like. And there still isn’t. It’s absolutely bonkers.” (Frida). Others shared when they had the courage to ask for help, they were faced with either long wait lists or no help at all: “The GP was a, I want to say about a years wait for counselling. The local hospital didn’t have anything to offer me. There was no service for this. No service at all” (Gloria). Gloria’s repetition of ‘no service’ emphasises the feeling of abandonment by professionals, leaving them to struggle on in crisis with no support.

For others standard care was offered without consideration of the nuanced support they needed. Whilst professionals acknowledged the intervention was not suitable, there was nothing else. This led to a sense of frustration and resignation things could not improve for them, because there was no help:

> they did say from the outset that this isn’t, you know, not post-natal depression, and we don’t know if CBT would actually be the right kind of therapy for you to do, but they said, you know, if you want to try you can (Cleo).

3.5.5.2 Seeking out Support for Yourself. Due to the lack of appropriate support some sought help independently. For some this meant proactively referring themselves: “I then looked for talking therapy and referred myself” (Harriet). Others, who had no success with NHS services, reached out to third sector services, but found there were still significant waits for treatment. This led others to pursue private therapy, which for some was a positive experience: “I had some sessions privately and I could talk through quite a lot, and it really helped me” (Gloria). Accessing private support comes at a cost to families who are already shouldering the burden of reduced household income due to the level of care needs. Others experience of private support echoed a lack of skills and knowledge needed for these idiosyncratic experiences. This led to interventions feeling unhelpful: “we couldn’t find a counsellor who was particularly qualified like, who knew anything about the situation. We ended up going to like a couple’s counsellor, which wasn’t what we needed” (Frida).

3.5.5.3 Peer Support is Invaluable. Participants expressed the value of peer support from other parents of children with profound intellectual disabilities. They described a sense of belonging and understanding they did not feel with parents of typical children. They spoke about finding a space where they could share advice and tips for navigating caring for a child with complex additional needs: “other parents were the best support I think I ever had” (Elizabeth).

Participants shared how valuable it is meeting other parents in the same circumstances to help understand what is happening to them: “And then I found a few people locally who have undiagnosed children or similar things, and I just find it so much easier to talk to them. Even though everyone's child is so different, because there's so many different ways that they express genetically.” (Cleo). For the participants spending extended periods in hospital compounded their sense of isolation and being able to hear other stories would have helped them process their experiences: “no one explains it to you, or shows you, or gives you people’s experiences. Because even if you read some people’s experiences that are willing to share that, then you understand a bit” (Etta). Etta’s use of “you” as a second-person pronoun creates a sense of distance from this experience, emphasising how difficult the sense of isolation was and a desire to not return there in the present moment.

3.5.6 Siblings Heal and Bring joy

In the final GET participants shared the deep joy more children bought to themselves, their partners and their older siblings. The participants who went on to have further children shared a sense of joy for not only them but their eldest child: “having another kid really made so many positive changes with Avi, what I, what we feel” (Indira). They spoke to siblings giving them purpose and strength to carry on when the burden of care for the eldest felt overwhelming: “I have found the other two a breeze of fresh air and when things are tough with Jake, they keep you going and keep

fighting, instead of collapsing, as they need you too" (Etta). Etta's use of "I" in the first-person pronoun highlights how this experience is one that is easier to share and demonstrates the strength she feels from her subsequent children.

Participants further on in their journey acknowledged if they had not gone on to have more children it would have amplified their difficult experiences and denied themselves the opportunity to lighten the burden they carried: "the trauma and the hardship that goes with having a disabled baby is a really heavy load, if that's where your journey ends" (Harriet). Harriet's use of the third-person pronoun separates her from that experience of trauma and hardship, it reflects her decision to go on and have further children, indicating those feelings are not hers to bear. There was an acute sense of relief and joy they found the courage to go on and have another child despite their previous experience because it enriched all their lives: "I am so glad that we weren't, you know, so heartbroken or so scared that we didn't, you know, risk him" (Frida).

3.6 Discussion

The participants shared when motherhood began with a child with profound intellectual disabilities it was marked with grief, isolation, a sense of burden and responsibility, fear, and strength required to seek support. They also described the deep joy and healing additional children brought to them and their families.

The GETs generated echo other research focused on the lived experience of parents of children with profound intellectual disabilities. The feeling of grief and loss experienced has previously been reported and appears to be an emotional phenomenon linked to a diagnosis of disability, rather than solely associated with first time mothers (Geuze et al., 2023; Geuze & Goossensen, 2021; Sheehan & Geurin, 2018). The desire to have another child for a more typical experience of motherhood was something six participants shared, and this had not been previously reported in mothers where birth order was not detailed.

Feeling isolated is a theme reported in literature by parents of children with a profound intellectual disability (Geuze et al., 2023). This included: others avoiding you, you avoid others due to a lack of understanding and fear of negative responses to your child (Geuze et al., 2023; Geuze & Goossensen, 2021; Sheehan & Guerin, 2018). Prevalence of this experience in the wider population indicates this is not exclusive to mothers of first-born children with profound intellectual disabilities, but the interaction it has with the decision to have more children may be.

The burden of care when parenting a child with profound intellectual disabilities is well documented (Brekke & Alecu, 2023; Geuze et al., 2023; Geuze & Goossensen, 2021; Shalali et al., 2024). Mothers have spoken about existing in parallel worlds (Geuze & Goossensen, 2024) and having to split oneself to balance the care needs of all siblings (Geuze et al., 2023; Willis & Godbold, 2024). Given the varying situations of mothers in this study, it appears existence in dual worlds

persists over time and is not unique to the early days with a child who has profound intellectual disabilities. However, if the burden of care is felt more acutely when the child is younger (Tadema & Vlaskamp, 2010), it may have a greater impact on mothers who are in the process of deciding whether to have further children. This was intensified by the pressure from their biological clock and the time required for genetic screening.

The weight of the decision to have further children following a child with profound intellectual disabilities has been shared previously (Geuze et al., 2023). Parents experiencing guilt about caring responsibilities falling to a sibling is a common narrative. However, Kruithof et al., (2021) found that whilst parents and siblings both considered the responsibility of future care, generally siblings did not perceive it as a burden, but a task they were willing to do. However, siblings did report the lack of transparency in communication about parents' expectations as challenging (Kruithof et al., 2021). The reticence to have open discussions may stem from guilt parents feel for imposing care responsibilities, which may be exacerbated for those with a first-born, who actively make the choice to place siblings in this role. Kruithof et al., (2021) also reported mothers feeling the need to have a third child to share the future burden of care, this was independent of birth order and occurred when it was a second-born child who had profound intellectual disabilities.

The need for meaningful and timely emotional support has been previously highlighted as beneficial for mothers' ongoing psychological wellbeing (Lahije et al., 2023). This is of particular importance during the child's early years (Sheehan & Geurin, 2018). Furthermore, peer support has been repeatedly cited as a key factor in mothers' ability to cope (Geuze et al., 2023; Oakley et al., 2022; Sheehan & Geurin, 2018) as well as building resilience (Willis & Godbold, 2024).

Previous evidence suggests the fear of having another child with profound intellectual disabilities is not unique to first time mothers (Geuze et al., 2023), there is little understanding of how this affects the decision for further children. This study highlighted how the fear of history repeating tainted future pregnancies and parenting experiences, despite going on to have a typically developing child.

Although the burden of care is undeniable there is support in wider literature for parenting a child with profound intellectual disabilities being a positive experience (Geuze et al., 2023). In other studies parents have similarly reported personal growth, sharing a sense of happiness and fulfilment (Beighton & Wills, 2019; Geuze & Goossensen, 2024). Like the mothers in this study, others have reported gratitude for the experience, for all they had learnt and how it shaped them as individuals (Sheehan & Geurin, 2018). Furthermore, research supports that siblings' experiences are positive for all involved (Dorsman et al 2024; Geuze et al., 2023).

3.6.1 Strengths and Limitations

A strength was using IPA to explore the experiences of mothers through a rich and detailed narrative, in their own words (Smith, 2011). This is important due to the minimal research conducted with this population and the implications caring has for this group (Brekke & Alecu, 2023; Fraser et al., 2021). IPA promotes accessibility whilst permitting complexity, through a balance of prescription and flexibility to explore the individual experience (Todorova, 2011). Through following step-by-step guidance (Smith & Nizza, 2022) this study demonstrated fidelity to IPA theoretical underpinnings by holding phenomenology, hermeneutics and the idiographic nature at the centre of the methodology (Smith, 2011). This was achieved through close adherence to guidance in the extant literature of standards for IPA and qualitative research (Levitt et al., 2018; Smith, 2011; Yardley, 2000). However, a limitation of this research, within an IPA framework, was conducting the interviews virtually. This may have limited the researcher's ability to fully observe and interpret the participants body language in the context of their spoken word, tone and intonation. However, on balance conducting the interviews virtually at a time and date that was convenient to the participant increased the accessibility for participation around other demands on their time, including the complex and high care needs of their first-born child.

A key aspect of IPA is a homogenous sample (Smith & Nizza, 2022). However, people with profound intellectual disabilities present with great heterogeneity (Doukas et al., 2017). Therefore, while their mothers met the inclusion criteria, their individual experiences, given their child's presentation, may have varied greatly. Another potential difference was the position of participants in their journey of motherhood. Six had completed their families during participation, whereas only one was still deciding about further children. This could be attributable to the early years of parenting a child with profound intellectual disabilities being more emotionally fraught than later (Sheehan & Guerin 2018). It may have been easier for those further from the point of decision to speak about the process and therefore were more likely to volunteer to participate. It may be helpful for further research to focus on those currently making decisions about whether to have further children to understand the experiences within the current socio-economic and political context given the changes that may occur over time in the provision of care and support offered.

Context is key in IPA, and this includes cultural situatedness (Todorova, 2011). Although ethnicity is not reported to protect anonymity, there was diversity within the sample, and this may explain some of the divergences between individual participants. It would be interesting for future research to explore the intersectionality of motherhood and culture, for example within individualistic and collectivist societies to understand how this might impact decisions about further children. It would also be important to explore the fathers and partners of the mothers' perspectives on having further children in the context of their first born having profound intellectual disabilities.

3.6.2 Recommendations

The findings indicate the need for meaningful, appropriate and timely support for mothers of children with profound intellectual disabilities. Furthermore, that this is implemented and delivered by professionals who understand the nuances of adjustment and impact this caring role has on the individual (Sheehan & Guerin 2018). It is key professionals including GP's, midwives, health visitors and other allied health professionals are trained how to support mothers and where to signpost for support. Clinical psychologists can play a key role in the consumption and dissemination of evidence in meaningful and pragmatic ways to support other professionals adopting psychologically informed thinking to deliver meaningful, appropriate and timely support.

This research highlights the value of peer support. Signposting to local third sector services, especially in the early years, should be standard care for mothers of first-born children with profound intellectual disabilities. Services may consider peer specialists within teams offering support during the period of adjustment to their child's diagnosis. Clinical Psychologists could support other professionals to develop co-produced pathways within services to bolster opportunities for peer support to be offered in a meaningful way. They may also use consultation skills to support third sector services to improve support offered to mothers across contexts and settings.

Exploration of adapted therapy for this population may prove valuable. Wider research into psychological flexibility and acceptance and commitment therapy, has proved to be beneficial for parents of children with disabilities (Gur & Reich, 2023). Therefore, future research could assess the feasibility and acceptability of this approach with this population. This is a key role for Clinical Psychologist's to undertake. Clinical Psychologists are trained to not only deliver therapy but think holistically about the person and how therapy can be adapted to improve not only accessibility, but also effectiveness from an evidence-based perspective.

3.6.3 Conclusion

These findings highlight the difficulties faced by mothers to first born children with profound intellectual disabilities when thinking about having further children. It demonstrates the need for nuanced and timely professional support throughout their journeys of motherhood. It emphasises the importance of connection with peers in similar circumstances as means to develop coping strategies and building resilience.

3.7 References

American Psychiatric Association, (2013). *Diagnostic and statistical manual of mental disorders: DSM-5*(Vol. 5, No. 5). Washington, DC: American psychiatric association.

Braun, V., & Clarke, V. (2021). Can I use TA? Should I use TA? Should I not use TA? Comparing reflexive thematic analysis and other pattern-based qualitative analytic approaches. *Counselling and psychotherapy research*, *21*(1), 37-47. https://doi.org/10.1002/capr.12360

Brekke, I., & Alecu, A. (2023). The health of mothers caring for a child with a disability: a longitudinal study. *BMC Women's Health*, *23*(1), 639. https://doi.org/10.1186/s12905-023-02798-y

Dorsman, N. I., Luijkx, J., Van der Schans, C. P., Van der Putten, A. A. J., & Waninge, A. (2024). Experiences of Adult Siblings of Individuals with an Intellectual Disability and Pervasive Support Needs. *Research and Practice for Persons with Severe Disabilities*, *49*(3), 155-173. https://doi.org/10.1177/15407969241245611

Doukas, T., Fergusson, A., Fullerton, M., & Grace, J. (2017). Supporting people with profound and multiple learning disabilities: Core & essential service standards 1st edition. https://www.pmldlink.org.uk/wp-content/uploads/2017/11/Standards-PMLD-h-web.pdf

Fraser, L. K., Murtagh, F. E., Aldridge, J., Sheldon, T., Gilbody, S., & Hewitt, C. (2021). Health of mothers of children with a life-limiting condition: a comparative cohort study. *Archives of Disease in Childhood*, *106*(10), 987-993. https://doi.org/10.1136/archdischild-2020-320655

Geuze, L., Goossensen, A., & Schrevel, S. (2023). "Continuously struggling for balance": The lived experiences of Dutch parents caring for children with profound intellectual and multiple disabilities. *Journal of Intellectual & Developmental Disability*, *48*(2), 161-171. https://doi.org/10.3109/13668250.2022.2073707

Geuze, L., & Goossensen, A. (2021). Exploring the experiences of Dutch parents caring for children with profound intellectual and multiple disabilities: A thematic analysis of their blogs. *Global Qualitative Nursing Research*, *8*, 23333936211028170. https://doi.org/10.1177/23333936211028170

Geuze, L., & Goossensen, A. (2024). Caring for children with profound intellectual and multiple disabilities: images and metaphors expressed by Dutch parents. *Disability & society*, *39*(7), 1840-1858. https://doi.org/10.1080/09687599.2023.2164846

Geuze, L., Schrevel, S., & Goossensen, A. (2023). "It is important that we also remain a person ourselves": A qualitative study about the role of healthcare and social welfare services by Dutch parents caring for a child with profound intellectual and multiple disabilities at home. *SSM-Qualitative Research in Health*, *4*, 100326. https://doi.org/10.1016/j.ssmqr.2023.100326

Gur, A., & Reich, A. (2023). Psychological flexibility of parents of children with disabilities: A systematic literature review. *Research in Developmental Disabilities*, *136*, 104490. https://doi.org/10.1016/j.ridd.2023.104490

Kruithof, K., IJzerman, L., Nieuwenhuijse, A., Huisman, S., Schippers, A., Willems, D., & Olsman, E. (2021). Siblings' and parents' perspectives on the future care for their family member with profound intellectual and multiple disabilities: A qualitative study. *Journal of Intellectual & Developmental Disability*, *46*(4), 351-361. https://doi.org/10.3109/13668250.2021.1892261

Lahaije, S. T., Luijkx, J., Waninge, A., & Van der Putten, A. A. (2023). Well-being of families with a child with profound intellectual and multiple disabilities. *Research and Practice for Persons with Severe Disabilities*, *48*(2), 63-78. https://doi.org/10.1177/15407969231173916

Levitt, H. M., Bamberg, M., Creswell, J. W., Frost, D. M., Josselson, R., & Suárez-Orozco, C. (2018). Journal article reporting standards for qualitative primary, qualitative meta-analytic, and mixed methods research in psychology: The APA Publications and Communications Board task force report. *American Psychologist*, *73*(1), 26. http://dx.doi.org/10.1037/amp0000151

Moilanen, S., Räikkönen, E., Lammi-Taskula, J., Duvander, A. Z., & Alasuutari, M. (2024). Do parenthood worries impede the birth of a second child? Differences according to the parent's gender and spousal support in Finland. *Journal of Family Research*, *36*, 103-125. https://doi.org/10.20377/jfr-968

Nakken, H., & Vlaskamp, C. (2007). A need for a taxonomy for profound intellectual and multiple disabilities. *Journal of Policy and Practice in intellectual Disabilities*, *4*(2), 83-87. https://doi.org/10.1111/j.1741-1130.2007.00104.x

Nizza, I. E., Farr, J., & Smith, J. A. (2021). Achieving excellence in interpretative phenomenological analysis (IPA): Four markers of high quality. *Qualitative Research in Psychology*, *18*(3), 369-386. https://doi.org/10.1080/14780887.2020.1854404

Oakley, S., Dunbar, H., & de Vries, K. (2022). Parent-led strategies supporting personal well-being when caring for a child with a life-limiting condition: A scoping review. *Journal of Child Health Care*, *26*(4), 648-667. https://doi.org/10.1177/13674935211026122

Pietkiewicz, I., & Smith, J. A. (2014). A practical guide to using interpretative phenomenological analysis in qualitative research psychology. *Psychological journal*, *20*(1), 7-14. DOI:10.14691/CPPJ.20.1.7

Schalock, R. L., Luckasson, R., & Tassé, M. J. (2021). An overview of intellectual disability: Definition, diagnosis, classification, and systems of supports. *American journal on intellectual and developmental disabilities*, *126*(6), 439-442. https://doi.org/10.1352/1944-7558-126.6.439

Shahali, S., Tavousi, M., Sadighi, J., Kermani, R. M., & Rostami, R. (2024). Health challenges faced by parents of children with disabilities: a scoping review. *BMC pediatrics*, *24*(1), 619. https://doi.org/10.1186/s12887-024-05104-3

Sheehan, P., & Guerin, S. (2018). Exploring the range of emotional response experienced when parenting a child with an intellectual disability: The role of dual process. *British Journal of Learning Disabilities*, *46*(2), 109-117. https://doi.org/10.1111/bld.12221

Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health psychology review*, *5*(1), 9-27. https://doi.org/10.1080/17437199.2010.510659

Smith, J. A., & Nizza, I. E. (2022). *Essentials of interpretative phenomenological analysis*. American Psychological Association.

Tadema, A. C., & Vlaskamp, C. (2010). The time and effort in taking care for children with profound intellectual and multiple disabilities: A study on care load and support. *British journal of learning disabilities*, *38*(1), 41-48. https://doi.org/10.1111/j.1468-3156.2009.00561.x

Todorova, I. (2011). Explorations with interpretative phenomenological analysis in different socio-cultural contexts: Commentary on J. Smith: 'Evaluating the contribution of interpretative phenomenological analysis'. *Health Psychology Review*, *5*(1), 34-38. https://doi.org/10.1080/17437199.2010.520115

Turpin, G., Barley, V., Beail, N., Scaife, J., Slade, P., Smith, J. A., & Walsh, S. (1997). Standards for research projects and theses involving qualitative methods: suggested guidelines for trainees and courses. In *Clinical Psychology Forum* (Vol. 108, pp. 3-7). The British Psychological Society.

Willis, E., & Godbold, R. (2024). Children's complex health: maternal experiences of care and decision making. *Journal of Child Health Care*, *28*(4), 786-803. https://doi.org/10.1177/13674935231158456

Wondemu, M. Y., Joranger, P., Hermansen, Å., & Brekke, I. (2022). Impact of child disability on parental employment and labour income: a quasi-experimental study of parents of children with disabilities in Norway. *BMC Public Health*, *22*(1), 1813. https://doi.org/10.1186/s12889-022-14195-5

Yardley, L. (2000). Dilemmas in qualitative health research. *Psychology and health*, *15*(2), 215-228. https://doi.org/10.1080/08870440008400302

Appendix A Mixed Methods Appraisal Tool

Part I: Mixed Methods Appraisal Tool (MMAT), version 2018

Category of study designs	Methodological quality criteria	Responses			
		Yes	No	Can't tell	Comments
Screening questions (for all types)	S1. Are there clear research questions?				
	S2. Do the collected data allow to address the research questions?				
	Further appraisal may not be feasible or appropriate when the answer is 'No' or 'Can't tell' to one or both screening questions.				
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?				
	1.2. Are the qualitative data collection methods adequate to address the research question?				
	1.3. Are the findings adequately derived from the data?				
	1.4. Is the interpretation of results sufficiently substantiated by data?				
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?				
2. Quantitative randomized controlled trials	2.1. Is randomization appropriately performed?				
	2.2. Are the groups comparable at baseline?				
	2.3. Are there complete outcome data?				
	2.4. Are outcome assessors blinded to the intervention provided?				
	2.5 Did the participants adhere to the assigned intervention?				
3. Quantitative non-randomized	3.1. Are the participants representative of the target population?				
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?				
	3.3. Are there complete outcome data?				
	3.4. Are the confounders accounted for in the design and analysis?				
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?				
4. Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?				
	4.2. Is the sample representative of the target population?				
	4.3. Are the measurements appropriate?				
	4.4. Is the risk of nonresponse bias low?				
	4.5. Is the statistical analysis appropriate to answer the research question?				
5. Mixed methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question?				
	5.2. Are the different components of the study effectively integrated to answer the research question?				
	5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?				
	5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?				
	5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?				

For further information on the MMAT please see Microsoft Word - MMAT_2018_criteria-manual_2018-08-08.docx

Appendix B Topic Guide

Topic guide

Title: An Interpretative Phenomenological Analysis: What are mothers experiences of making family planning decisions when their first-born child has profound intellectual and multiple disabilities?

ERGO Number: 93086

Topics	Prompt	Interview Question
Introduction and background	Nature of study, aim and confidentiality. Explain if want to stop at any point or take a break. Demographic questions.	
Wellbeing	Wellbeing check-in	1. I hope you are well and are happy to go ahead with the interview? 2. Are you in a confidential place and feel able to talk freely without being overheard?
Family	Current family configuration	3. Can you tell me a bit about your family? - *Is there anyone else important to your family in your life?*
Family planning decisions	Decision to become mother Subsequent children	4. Can you tell me about your decision to become a mother? 5. Issues? 6. After having _____ what was you experience of thinking about having further children? 7. In what ways do you think, if any, did your experience of parenting _____ shape your decision to have further children?
Wider systems	External influences Support	8. Outside of you and your partner, was there anything else that influenced your decision? *Prompts for these areas if not mentioned:* - *Family* - *Friends* - *Professionals* - *Life circumstances* 9. In what ways, if any did health professionals support you with making decisions? 10. Is there any support you think would have been helpful? - *From health professionals, if not mentioned*

Effects of decision making	How the decision about having further children has impacted the person as individual and within wider systems Similarities/differences of impact to person and partner	11. How do you think the process of making family planning decisions has shaped your life? *Prompts for theses areas if not mentioned:* - *How do you think this decision has affected your family?* - *Your relationships?* - *You as a person?* 12. How do you think you experience of this decision making has compared to your partner/biological father/current partner?
Advice	Knowledge they would want to share with others	13. What would be your advice to other parents of children with PIMD be for family planning? 14. Is there anything else you think is important to tell me?
Ending	Additional information and debriefing form distribution. Plan for research outcome – thesis and publication. Contact details and information if they decide they would like to withdraw from the study. Information regarding voucher.	

Appendix C Participant Information Sheet

Participant Information Sheet

Study Title: An Interpretative Phenomenological Analysis: What are mothers experiences of making family planning decisions when their first-born child has profound intellectual and multiple disabilities?

Researcher: Jean Jevons

ERGO number: 93086

You are being invited to take part in the above research study. To help you decide whether you would like to take part or not, it is important that you understand why the research is being done and what it will involve.

Please read the information below carefully and ask questions if anything is not clear or you would like more information before you decide to take part in this research. You may like to discuss it with others, but it is up to you to decide whether or not to take part. If you are happy to participate you will be asked to sign a consent form.

What is the research about?

I am a Trainee Clinical Psychologist studying at the University of Southampton and this research forms part of the submission for my doctoral thesis. This research hopes to further understand about how a mother's experience of parenting a first-born child with profound intellectual and multiple disabilities (PIMD) informs subsequent decisions about family planning. Although research has explored parents experience of caring for a child with PIMD, and peoples experience of being a sibling to those with PIMD, there is little understanding about the process mothers go through when deciding to have further children. Understanding this experience will help identify if psychological support to mothers and their families is needed when making these decisions. It may also highlight any ongoing support needed following any decisions made.

Why have I been asked to participate?

You have been asked to participate because you are the biological mother to a first-born child who has had PIMD since birth. They were also your first experience of parenting; you can speak English and are living in the United Kingdom.

What will happen to me if I take part?

If you decide you would like to take part you will be asked some demographic data, including age, relationship status, number and age(s) of children, eldest child's diagnosis, ethnicity, and employment status. Gathering this information will help us understand a bit more about your circumstances and contextualise any information you choose to share in the interview. You will then be asked to take part in an interview, lasting no longer than an hour where you will be asked about your experiences of being a mother to a child with PMLD and how it shaped your future family planning decisions. The interview will take place over MS Teams. Hopefully this will cause the least disruption to you schedule and will mean it can take place in the comfort of your own home, in a confidential space you feel able to speak freely. This will be a relatively informal conversation where you will have space to talk about your experiences. If there are any questions you do not feel

comfortable answering, we can move on, and this will not affect your participation in the research. If at any point during the interview you decide you no longer want to take part, you can withdraw, and your data will be removed from the study.

The interviews will be recorded on MS Teams. This will therefore include audio and visual. This is to support the interview process and enable the interview to be transcribed for analysis. You will be asked to sign a consent form to indicate you agree to having the interview recorded. The recordings will be kept securely on a password protected laptop in a password protected file. The recordings will only be listened to those directly involved in the research.

Once the interviews have been transcribed, they will be analysed to explore common themes and issues that appear to be significant to the participants.

The results of the research will be written in the form of a doctoral thesis and submitted to the University of Southampton as part of the researchers' award of Doctorate in Clinical Psychology. The thesis may also be submitted for publication in a scientific journal to help share the knowledge found. All contributions from participants will be anonymised prior to submission to the University or scientific journal.

As a thank you for full participation in this research you will receive a £20.00 voucher.

Are there any benefits in my taking part?

Many people find participating in research interesting, or an opportunity to reflect on their experiences and how they have responded to life experiences. It is hoped that this research will also benefit future mothers going through these experiences by understanding what support and help would be helpful. Whilst there is no direct benefit for participants, you will receive a £20.00 pound voucher as a thank you for your participation.

Are there any risks involved?

There is a chance you may find discussing your previous experiences upsetting. If you become distressed during or following the interview, information about support services will be provided.

What data will be collected?

The data collected will be the content of the interview. This will be recorded via MS Teams. The laptop being used is password protected. All demographic information shared will be viewed by the researcher, Jean Jevons and stored securely on an encrypted computer system provided by the university. If there are ant paper copies these will be scanned onto the same system and hard copies will be destroyed.

All participant contact details will be kept on a password protected laptop for the duration of the research to maintain contact with participants for as long as necessary, and no longer. These will be the contact details offered by the participants in line with their preferred method of communication, i.e., phone or email.

Identifiable information will not be used in the write up of the research project. Quotes will be included, but remain anonymous, for example, "participant 2, in her thirties said…."

Will my participation be confidential?

Your participation and the information we collect about you during the course of the research will be kept strictly confidential.

Only members of the research team and responsible members of the University of Southampton may be given access to data about you for monitoring purposes and/or to carry out an audit of the study to ensure that the research is complying with applicable regulations. Individuals from regulatory authorities (people who check that we are carrying out the study correctly) may require access to your data. All these people have a duty to keep your information, as a research participant, strictly confidential.

Interview recordings will be made, so they can be transcribed later. Once transcripts have been made the recording will be destroyed. The transcripts will be kept on a password protected laptop. Contact information and demographic information will also be stored securely on the password protected laptop. If there are any hard copies of information provided by the participant they will be scanned, saved with the other information electronically and the paper copies will be destroyed. Electronic copies will be destroyed once the research has been completed.

Do I have to take part?

No, it is entirely up to you to decide whether or not to take part. If you decide you want to take part, you will need to sign a consent form to show you have agreed to take part.

If you would like to participate, you can contact the researcher (Jean Jevons) via email at the following address: J.R.Jevons@soton.ac.uk.

What happens if I change my mind?

You have the right to change your mind and withdraw at any time without giving a reason and without your participant rights (or routine care if a patient) being affected. To withdraw from the research, you have two weeks following the interview to inform the researcher of your decision. You can do this by emailing them at J.R.Jevons@soton.ac.uk. Requests to withdraw after two weeks cannot be honoured, as the data will have been transcribed and anonymised.

What will happen to the results of the research?

Your personal details will remain strictly confidential. Research findings made available in any reports or publications will not include information that can directly identify you without your specific consent.

The findings of this research will be submitted to the University of Southampton as part of the researchers work towards completing a Doctorate in Clinical Psychology and will be submitted to a scientific journal for publication.

If you would like a summary of the findings following the completion of the research, you can indicate this on the consent form. You will receive this via email.

Where can I get more information?

For further information, please contact Jean Jevons at J.R.Jevons@soton.ac.uk.

What happens if there is a problem?

If you have a concern about any aspect of this study, you should speak to the researchers who will do their best to answer your questions.

Jean Jevons: J.R.Jevons@soton.ac.uk

Dr Melanie Hodgkinson: M.J.Hodgkinson@soton.ac.uk

Dr Cheryl Jones: cj1a09@soton.ac.uk

If you remain unhappy or have a complaint about any aspect of this study, please contact the University of Southampton Research Integrity and Governance Manager (023 8059 5058, rgoinfo@soton.ac.uk).

Data Protection Privacy Notice

The University of Southampton conducts research to the highest standards of research integrity. As a publicly funded organisation, the University has to ensure that it is in the public interest when we use personally identifiable information about people who have agreed to take part in research. This means that when you agree to take part in a research study, we will use information about you in the ways needed, and for the purposes specified, to conduct and complete the research project. Under data protection law, 'Personal data' means any information that relates to and is capable of identifying a living individual. The University's data protection policy governing the use of personal data by the University can be found on its website (https://www.southampton.ac.uk/legalservices/what-we-do/data-protection-and-foi.page).

This Participant Information Sheet tells you what data will be collected for this project and whether this includes any personal data. Please ask the research team if you have any questions or are unclear what data is being collected about you.

Our privacy notice for research participants provides more information on how the University of Southampton collects and uses your personal data when you take part in one of our research projects and can be found at http://www.southampton.ac.uk/assets/sharepoint/intranet/ls/Public/Research%20and%20Integrity%20Privacy%20Notice/Privacy%20Notice%20for%20Research%20Participants.pdf

Any personal data we collect in this study will be used only for the purposes of carrying out our research and will be handled according to the University's policies in line with data protection law. If any personal data is used from which you can be identified directly, it will not be disclosed to anyone else without your consent unless the University of Southampton is required by law to disclose it.

Data protection law requires us to have a valid legal reason ('lawful basis') to process and use your Personal data. The lawful basis for processing personal information in this research study is for the performance of a task carried out in the public interest. Personal data collected for research will not be used for any other purpose.

For the purposes of data protection law, the University of Southampton is the 'Data Controller' for this study, which means that we are responsible for looking after your information and using it properly. The University of Southampton will keep identifiable information about you for 10 years after the study has finished after which time any link between you and your information will be removed.

To safeguard your rights, we will use the minimum personal data necessary to achieve our research study objectives. Your data protection rights – such as to access, change, or transfer such information - may be limited, however, in order for the research output to be reliable and accurate. The University will not do anything with your personal data that you would not reasonably expect.

If you have any questions about how your personal data is used, or wish to exercise any of your rights, please consult the University's data protection webpage (https://www.southampton.ac.uk/legalservices/what-we-do/data-protection-and-foi.page) where you can make a request using our online form. If you need further assistance, please contact the University's Data Protection Officer (data.protection@soton.ac.uk).

Thank you. For taking the time to read this information and considering if you would like to participate in this research project.

Appendix D Consent Form

CONSENT FORM

University of Southampton

Study title: An Interpretative Phenomenological Analysis: What are mothers experiences of making family planning decisions when their first-born child has profound intellectual and multiple disabilities?

Researcher name: Jean Jevons
ERGO number: 93086

Participant Identification Number (if applicable):

Please initial the box(es) if you agree with the statement(s):

I have read and understood the information sheet (16 August Version 2 of participant information sheet) and have had the opportunity to ask questions about the study.	
I agree to take part in this research project and agree for my data to be used for the purpose of this study.	
I understand my participation is voluntary and I may withdraw for up to two weeks after the interview for any reason without my participation rights being affected.	
I understand that I may be quoted directly in reports of the research but that I will not be directly identified (e.g. that my name will not be used).	
I understand that taking part in the study involves audio and video recording which will be transcribed and then destroyed for the purposes set out in the participant information sheet.	
I agree to take part in the interview for the purposes set out in the participation information sheet and understand that these will be recorded using audio and video.	
I understand that my personal information collected about me such as my name or where I live will not be shared beyond the study team.	
I understand that special category information will be collected about me to achieve the objectives of the study. This will include age, gender, and ethnicity.	
I would like to receive a summary of the research findings via email.	

Name of participant (print name)…….

Signature of participant…….

Date…….. …….

Name of researcher (print name)……..

Signature of researcher ……..

Date…….

Appendix E Participant Screening Information

Screening email to be sent to clarify PIMD diagnosis with mother interested in participating.

Thank you for expressing interest in participating in this research. Given the nature of this research, specifically exploring mothers experience with children who Profound Intellectual and Multiple Disabilities (PIMD), it is important to clarify what we mean by this for this piece of research.

What is Profound Intellectual and Multiple Disability?

The child must have a profound learning disability.

This means they do not communicate using symbols i.e. words, PECS etc, but rely on physical objects of reference. They may understand some verbal prompts, but their understanding of speech and gestures will be limited. In turn they will express their emotions and needs through non-verbal and non-symbolic (no writing, PECS etc) communication. They may also enjoy relationships with family, carers and those who are familiar. With these people they may initiate and respond to social interactions through gestures and emotional cues.

Typically, they will also experience physical disabilities, which may often mean they are a wheelchair user. They will also be dependent for all activities of daily living, including toileting, personal care needs and other health and safety tasks. They may be able to participate in some of these tasks but are not able to complete any of them independently.

They may also have sensory impairments. This could include vision or hearing, or other wider sensory issues linked to how their body experiences the world. These sensory and physical difficulties may also prevent them from engaging in social, recreational, and vocational activities.

People with PIMD also tend to have complex physical health needs. This may include, but is not limited epilepsy and being fed via feeding tubes directly into their stomachs.

For some people with PIMD they may also have behaviour that us counterproductive, harmful and interfere with day-to-day activities and functioning.

What PIMD is not and how this can be confused

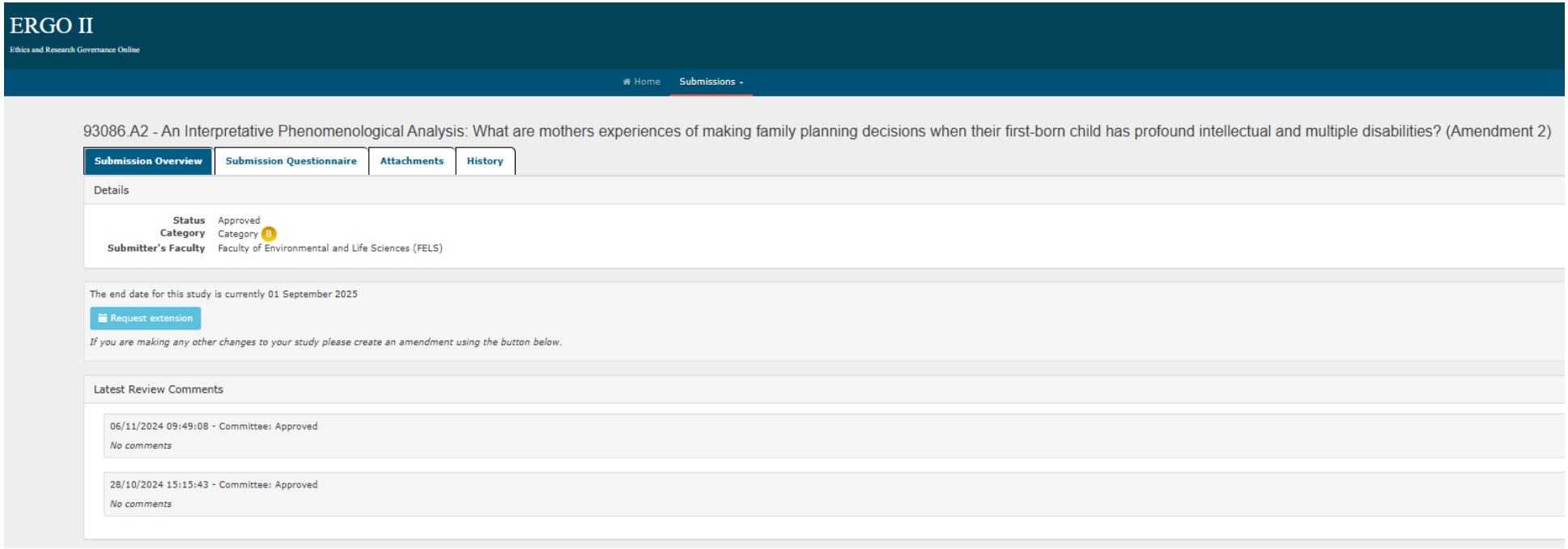
There are no specific diagnoses that fall under the term PIMD. This makes it hard to explicitly say what PIMD is and is not. For example, whilst someone may have Autism and a Severe Learning Disability, they would not have PIMD if they could perform some tasks independently, or used PECS, to communicate. A person may be able to move independently from aids like a wheelchair, but they must also meet the criteria of a profound learning disability and difficulties with communication.

Prior to participating in this research, it is important to understand if your child meets the criteria for PIMD. This is because this study is focusing very specifically on the mothers of these people and their

experiences. It is not to say the experience of mothers of children with other disabilities is not important or useful to research. There may be other research more suitable to you and your child that will be available to participate in in the future.

If you are unsure whether your child meets these criteria, then I am more than happy to discuss it with you prior to proceeding with the study.

Appendix F Ethics



Appendix G Debrief Form

Debriefing Form

Study Title: An Interpretative Phenomenological Analysis: What are mothers experiences of making family planning decisions when their first-born child has profound intellectual and multiple disabilities?

Ethics/ERGO number: 93086

Researcher(s): Jean Jevons, Dr Cheryl Jones & Dr Melanie Hodgkinson

University email(s): J.R.Jevons@soton.ac.uk
cj1a09@soton.ac.uk
M.J.Hodgkinson@soton.ac.uk

Version and date: 26 July 2024 Version 1

Thank you for taking part in our research project. Your contribution is very valued and greatly appreciated.

Purpose of the study

The aim of this research is to gain an understanding of how the experience of parenting for a mother whose, first born child has profound intellectual and multiple disabilities (PIMD), impacts their decisions about future family planning. It is known that parenting a child with PIMD can be experienced as stressful and a burden, but people have also reported lots of positive aspects. There has also been research conducted into siblings of people with PIMD, providing understanding of their experience. However there has not been any research into the decision-making process around family planning for families whose first born child has PIMD.

It is hoped that this research will help understand the experience of mothers and inform support for their wellbeing they might need during these times of decision making. Your data will help to further understanding about ways in which parents caring for a child with PMLD and thinking about further child can be supported to make the right decision for them and their families.

Confidentiality

Results of this study will not include your name or any other identifying characteristics.

Study results

If you would like to receive a copy of the dissertation when it is completed, please let us know by using the contact details provided on this form.

Further support

If taking part in this study has caused you discomfort or distress, you can contact the following organisations for support:

Your GP is the first person you should seek help from. If they are not available or you feel unable to keep yourself or those around you safe, please call NHS 111, 999 or attend A&E.

Other national services that are available are:

Samaritans: The Samaritans are a service that you can call in times of crisis. Their help line is open 24 hours a day, 7 days a week. You can also email, but they may take several days to respond.
Website: www.samaritans.org
Phone: 116 123 (free to call from UK and Ireland)
Email: jo@samaritans.org

Shout: Shout is a free, confidential, 24/7 text support service for anyone in the UK who is struggling to cope. Start a conversation by texting the word 'Shout' to 85258.

Further reading

If you would like to learn more about this area of research, you can refer to the following resources:

Geuze, L., Goossensen, A., & Schrevel, S. (2023). "Continuously struggling for balance": The lived experiences of Dutch parents caring for children with profound intellectual and multiple disabilities. *Journal of Intellectual & Developmental Disability*, *48*(2), 161-171. https://doi.org/10.3109/13668250.2022.2073707

Further information

If you have any concerns or questions about this study, please contact Jean Jevons at J.R.Jevons@soton.ac.uk who will do their best to help.

If you remain unhappy or would like to make a formal complaint, please contact the Head of Research Integrity and Governance, University of Southampton, by emailing: rgoinfo@soton.ac.uk, or calling: + 44 2380 595058. Please quote the Ethics/ERGO number which can be found at the top of this form. Please note that if you participated in an anonymous survey, by making a complaint, you might be no longer anonymous.

Thank you again for your participation in this research.

Appendix H Reflective Diary Excerpt

Interview with "Indira"

I was in admiration of Indira's strength individually and with her husband, especially being so far from friends and family. During the interview she had to take a phone call from her son's school. I felt empathy for her, relating to the gut wrench you feel when you get a call from people caring for your child. I was in awe of the way she candidly spoke about how difficult life is, even though she is so capable. Also, the sacrifices she has made for her family in terms of her career, but how much joy her children bought her. I was also in admiration of how she shared her experience of accessing an abortion. There is so much shame surrounding it, but she spoke with courage and bravery.

Interview with "Harriet"

Hearing about Harriet's experience of having two children so close together really resonated with me. Also, how she described having to split herself, being in two places as a mother, and lacking connection with other mothers. I felt inspired by her tenacity and courage to speak about her experiences. She struck me as being brave, resilient and strong! I also appreciated her drive and motivation to share her story to help others.

Supplementary Material 1 Journal Guidelines

JARID Author Guidelines

Sections

1. Submission

Authors should kindly note that submission implies that the content has not been published or submitted for publication elsewhere except as a brief abstract in the proceedings of a scientific meeting or symposium.

Once the submission materials have been prepared in accordance with the Author Guidelines, new submissions should be made via the Research Exchange submission portal: **https://wiley.atyponrex.com/journal/JAR**. Should your manuscript proceed to the revision stage, you will be directed to make your revisions via the same submission portal. You may check the status of your submission at anytime by logging in to submission.wiley.com and clicking the "My Submissions" button. For technical help with the submission system, please review our **FAQs** or contact **submissionhelp@wiley.com**.

Wiley Publishing Networks

This journal participates in the Wiley Special Education publishing network and the **Wiley Developmental Science Publishing Network**. This exciting collaboration amongst our Special Education and Developmental journals simplifies and speeds up the publication process, helping authors find the right home for their research. At the Editors' judgement, suitable papers not accepted by one journal may be recommended for referral to another journal(s) in the network. Authors decide whether to accept the referral, with the option to transfer their paper with or without revisions. Once the referral is accepted, submission happens automatically, along with any previous reviewer reports, thereby relieving pressure on the peer review process. While a transfer does not guarantee acceptance, it is more likely to lead to a successful outcome for authors by helping them to find a route to publication quickly and easily.

2. AIMS AND SCOPE

JARID is an international, peer-reviewed journal which draws together findings derived from original applied research in intellectual disabilities. The journal is an important forum for the dissemination of ideas to promote valued lifestyles for people with intellectual disabilities. It reports on research from the UK and overseas by authors from all relevant professional disciplines. It is aimed at an international, multi-disciplinary readership.

In order for a paper to be considered for publication, it must be about people with intellectual disabilities. Manuscripts which focus upon autism will be considered only when the focus is also upon intellectual disabilities. Papers which focus upon autism and exclude people with intellectual disabilities will not be considered.

The topics it covers include community living, quality of life, challenging behaviour, communication, sexuality, medication, ageing, supported employment, family issues, mental health, physical health, autism, economic issues, social networks, staff stress, staff training, epidemiology and service provision.

Theoretical papers are also considered provided the implications for therapeutic action or enhancing quality of life are clear. Both quantitative and qualitative methodologies are welcomed. All original and review articles continue to undergo a rigorous, peer-refereeing process.

3. MANUSCRIPT CATEGORIES AND REQUIREMENTS

Original Articles**,* including Clinical Trials (see guidance within section 5), ***Review Articles and ***Brief Reports*** are accepted by the Journal. ***Theoretical Papers*** are also considered, provided the implications for therapeutic action or enhancing quality of life are clear. Both quantitative and qualitative methodologies are welcomed. Articles are accepted for publication only at the discretion of the Editor. Authors who are submitting original articles where qualitative methods have been used must ensure that their choice of method is well justified and issues relating to methodological rigor are effectively addressed.

Articles and ***Theoretical Papers*** should not exceed 6000 words;

Review Articles should not exceed 7000 words;

Brief Reports should not exceed 2000 words.

All word limits are inclusive of the abstract. References, Words in Tables, Captions/Legends, Figure and Figure captions/legends are excluded from the word limits.

Please note that papers submitted for Special Issues should also not exceed 6000 words.

4. PREPARING THE SUBMISSION

Use of Language

The language used to describe disability differs across countries, cultures and disciplinary fields, and continues to evolve. All manuscripts submitted to JARID must use language that promotes the value of all people as full members of our shared society. Pejorative language inclusive of euphemisms must not be used. For JARID this includes the use of older language that has been used to describe people with intellectual disabilities such as “retarded”, "special needs", "disease", “handicapped”, or “mentally handicapped”. Using any terms which are offensive, or patronising may lead to rejection of your submitted manuscript.

JARID recommends using person-first and/or identity-first language thoughtfully and appropriately. For example, the language used to describe both people with intellectual disabilities and autistic people has evolved based on recent advocacy efforts. When referring to people with autism, it is acceptable to use either identity-first language (e.g., “autistic people”) or person-first language (e.g., people with autism”), while identity-first language is not used to describe people with intellectual disabilities, where person-first language is preferred. Thus, people with intellectual disabilities should be referred to as people with intellectual disabilities.

We have consulted with over 40 self-advocates through Learning Disability England which included the North West Self-Advocacy Group, as well as Self-Advocacy Together and asked them what language we should use when writing about people with intellectual disabilities.

People with intellectual disabilities said that they do not like to be referred to by acronyms or abbreviations. Authors must therefore not use an abbreviation to describe intellectual disabilities such as “ID” or “LD”. Instead, use person-first language such as children, teenagers, adults, or people with intellectual disabilities, avoiding acronyms or abbreviations.

The terms “learning disabilities” and “learning difficulties”, though used in some countries to refer to people with intellectual disabilities, can cause confusion among readers. These terms are not used by the journal to refer to people with intellectual disabilities. Authors must only use the term “learning disabilities or difficulties” where this refers to a specific learning disability/disorder– such as a specific learning difficulty in reading, written expression or mathematics. **If “learning disabilities” or “learning difficulties” are used, authors must not use an abbreviation.**

Free Format Submission

JARID now offers **Free Format submission** for a simplified and streamlined submission process. Before you submit, you will need:

- A Cover Letter
- Your manuscript: this should be an editable file including text, figures, and tables, or separate files – whichever you prefer. All required sections should be contained in your manuscript, including titles, keywords, abstract, lay summary, introduction, methods, results, discussion/conclusions and acknowledgements.
- Figures and tables should have legends. Figures should be uploaded in the highest resolution possible.
- References may be submitted in any style or format, as long as it is consistent throughout the manuscript.
- Supporting information should be submitted in separate files. Click here for Wiley's FAQs on supporting/supplemental information.

- Note: if data, scripts, or other artefacts used to generate the analyses presented in the paper are available via a publicly available data repository, authors should include a reference to the location of the material within their paper
- An ORCID ID, freely available at https://orcid.org (Why is this important? Your article, if accepted and published, will be attached to your ORCID profile. Institutions and funders are increasingly requiring authors to have ORCID IDs.)
- The title page of the manuscript, including:

- Your co-author details, including affiliation and email address. (Why is this important? We need to keep all co-authors informed of the outcome of the peer review process.)
- Statements relating to our ethics and integrity policies, which may include any of the following (Why are these important? We need to uphold rigorous ethical standards for the research we consider for publication):

- data availability statement
- funding statement
- conflict of interest disclosure
- ethics approval statement
- patient consent statement
- permission to reproduce material from other sources
- clinical trial registration

JARID has a double-anonymized peer review process so please ensure that all identifying information such as author names and affiliations, acknowledgements or explicit mentions of author institution in the text are on a separate page.

Abstract
All papers should have a structured abstract (maximum 150 words) as follows: Background, Method, Results, and Conclusions. The abstract should provide an outline of the research questions, the design, essential findings and main conclusions of the study. We kindly request that authors place the abstract and title at the beginning of the main manuscript document.

Lay Summary
Please provide 3 or 4 bullet points summarizing the main finding of your work, the impact of it for people with intellectual disabilities and for the research community.

Authorship
On initial submission, the submitting author will be prompted to provide the email address and country for all contributing authors.

The Research Exchange submission system will extract listed affiliations from the manuscript and then ask the submitting author to verify each author's affiliation institution(s). Authors are encouraged to include the complete affiliation addresses in the manuscript (Institution Name, Country, Department Name, Institution City, and Post Code). When verifying their institution, authors will also be asked to locate their base institution only (not necessarily the department or school).

Please refer to the journal's authorship policy in the Editorial Policies and Ethical Considerations section for details on eligibility for author listing.

Acknowledgments
Contributions from anyone who does not meet the criteria for authorship should be listed, with permission from the contributor, in an Acknowledgments section. Financial and material support should also be mentioned. Thanks to anonymous reviewers are not appropriate.

Conflict of Interest Statement
Authors will be asked to provide a conflict of interest statement during the submission process. For details on what to include in this section, see the section 'Conflict of Interest' in the Editorial Policies and Ethical Considerations section below. Submitting authors should ensure they liaise with all co-authors to confirm agreement with the final statement.

Wiley Author Resources

Manuscript Preparation Tips: Wiley has a range of resources for authors preparing manuscripts for submission available **here**. In particular, authors may benefit from referring to Wiley's best practice tips on **Writing for Search Engine Optimization.**

Article Preparation Support
Wiley Editing Services offers expert help with English Language Editing, as well as translation, manuscript formatting, figure illustration, figure formatting, and graphical abstract design â€“ so you can submit your manuscript with confidence.

Also, check out our resources for **Preparing Your Article** for general guidance about writing and preparing your manuscript.

Video Abstracts
A video abstract can be a quick way to make the message of your research accessible to a much larger audience. Wiley and its partner Research Square offer a service of professionally produced video abstracts, available to authors of articles accepted in this journal. You can learn more about it by **clicking here**. If you have any questions, please direct them to **videoabstracts@wiley.com**.

5. EDITORIAL POLICIES AND ETHICAL CONSIDERATIONS

Peer Review and Acceptance
Manuscripts are judged on the significance of the contribution to the literature, the quality of analysis and the clarity of presentation. Papers are expected to demonstrate originality and meaningful engagement with the global literature.

Except where otherwise stated, manuscripts are double-anonymized peer reviewed by anonymous reviewers in addition to the Editor. Ensure that all personally identifiable information is removed from your manuscript before you upload it to help protect your identity through the peer review process. Authors are asked not to post information about their submitted manuscripts to social media or websites until a final decision about the paper has been made; again, the reason for this is to help protect the double- anonymized

peer review process. Authors who do not work in such a way as to help maintain the double-anonymized peer review process may have their manuscript rejected.

Final acceptance or rejection rests with the Editor-in-Chief, who reserves the right to refuse any material for publication or to edit any contribution to ensure that it conforms with the requirements of the journal

In-house submissions, i.e. papers authored by Editors or Editorial Board members of the title, will be sent to Editors unaffiliated with the author or institution and monitored carefully to ensure there is no peer review bias.

Wiley's policy on the confidentiality of the review process is **available here.**

Refer and Transfer Program

Wiley believes that no valuable research should go unshared. This journal participates in Wiley's **Refer & Transfer program**. If your manuscript is not accepted, you may receive a recommendation to transfer your manuscript to another suitable Wiley journal, either through a referral from the journal's editor or through our Transfer Desk Assistant.

Human Studies and Subjects
For manuscripts reporting studies that involve human participants, including but extending beyond medical research, a statement identifying the ethics committee that approved the study and confirmation that the study conforms to recognized standards is required, for example: **Declaration of Helsinki**; **US Federal Policy for the Protection of Human Subjects**; or **European Medicines Agency Guidelines for Good Clinical Practice**. It should also state clearly in the text that all persons gave their informed consent prior to their inclusion in the study.

Patient anonymity should be preserved. Photographs need to be cropped sufficiently to prevent human subjects being recognized (or an eye bar should be used). Images and information from individual participants will only be published where the authors have obtained the individual's free prior informed consent. Authors do not need to provide a copy of the consent form to the publisher; however, in signing the author license to publish, authors are required to confirm that consent has been obtained. Wiley has a **standard patient consent form** available for use.

Clinical Trial Registration
The journal requires that clinical trials are prospectively registered in a publicly accessible database and clinical trial registration numbers should be included in all papers that report their results.

Clinical trials should be reported using the CONSORT guidelines available at **www.consort-statement.org**. A CONSORT checklist should also be included in the submission material (**www.consort-statement.org**).

The Journal encourages authors submitting manuscripts reporting from a clinical trial to register the trials in any of the following free, public trials registries: **www.clinicaltrials.org**, **www.isrctn.org**.

Authors are asked to include the name of the trial register and the clinical trial registration number at the end of the abstract. If the trial is not registered, or was registered retrospectively, the reasons for this should be explained.

Conflict of Interest

The journal requires that all authors disclose any potential sources of conflict of interest. Any interest or relationship, financial or otherwise that might be perceived as influencing an author's objectivity is considered a potential source of conflict of interest. These must be disclosed when directly relevant or directly related to the work that the authors describe in their manuscript. Potential sources of conflict of interest include, but are not limited to: patent or stock ownership, membership of a company board of directors, membership of an advisory board or committee for a company, and consultancy for or receipt of speaker's fees from a company. The existence of a conflict of interest does not preclude publication. If the authors have no conflict of interest to declare, they must also state this at submission. It is the responsibility of the corresponding author to review this policy with all authors and collectively to disclose with the submission ALL pertinent commercial and other relationships.

Authorship

The list of authors should accurately illustrate who contributed to the work and how. All those listed as authors should qualify for authorship according to the following criteria:

1. Have made substantial contributions to conception and design, or acquisition of , or analysis and interpretation of data; and
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3. Given final approval of the version to be published. Each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content; and
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