Exploring the perspectives of current and future sibling caregivers to a person with a neurodevelopmental disorder – implications for occupational therapy. A scoping review.
Exploring the perspectives of current and future sibling caregivers to a person with neurodevelopmental disorder – implications for occupational therapy.

A scoping review.

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Abstract

**Introduction:** Numerous studies have examined caregiver burden and stress, yet few represent the perspective from the sibling caregiver of a person with a neurodevelopmental disorder within the context of occupational therapy practice. Therefore, this scoping review provides a review of current evidence.

**Aim:** The aim of the study is to review current literature to find sibling caregiver perspectives on their roles as caregivers to an individual with a neurodevelopmental disorder and whether their perspectives can influence improvements to the occupational therapy practice.

**Method:** A scoping review methodology was chosen to map out the available literature and summarise the findings. The author conducted this review using guidance from the Joanna Briggs Institute.

**Results:** The review resulted in 11 articles and reviewed the perspectives of 1181 sibling participants. The findings resulted in five main themes: (1) Need for education and training opportunities in relation to caregiving; (2) Policy and guideline changes in relation to sibling caregiver involvement; (3) Sibling caregiver inclusion into the care pathway; (4) Need for improvements in service coordination; and (5) Sibling caregivers' lack of knowledge in guardianship matters.
Conclusions: Both positive experiences and negative challenges were reported from sibling experiences when accessing healthcare services. Ideas and strategies were discussed by sibling caregivers in how to improve services.

Significance: This review identified practical strategies which can be used in occupational therapy practice to support and improve services in relation to working with sibling caregivers.

Keywords

Adults; Brothers; Intellectual Disability; Learning Disability; Occupational Therapy; Sisters.
List of abbreviations

Activities of daily living – ADLs
Evidence-based practice – EBP
Neurodevelopmental disorder – NDD
Occupational therapy – OT
Occupational therapists’ – OTs
Exploring the perspectives of current and future sibling caregivers to a person with neurodevelopmental disorder – implications for occupational therapy. A scoping review.

Introduction

The World Health Organization (WHO) estimates that 349 million people worldwide are dependent on caregivers for their care needs (WHO, 2017). Caregivers are defined as an individual who cares for and/or assists a disabled, ill, or frail family member, spouse or friend (Moghimi, 2007). Growing evidence indicates that the number of caregivers is formidably increasing and the Center for Disease Control and Prevention (CDC) states that the effect on caregivers’ quality of life is a public health issue (WHO, 2022; CDC, 2018). Estimating the actual number of caregivers worldwide is difficult, although, the International Alliance of Carer Organizations (IACO) estimates that there are already over 22 million caregivers across Europe alone (IACO, 2021). The Europe Quality of Life Survey estimates that 20% of its population are formal carers and as much as 80% of all long-term care is provided by informal carers - between 10% to 25% of the population (European Commission, 2018).

Totsika, Hastings, and Vagenas (2017) estimate that in future years, social care providers will increasingly rely upon informal caregivers further to provide care to people with intellectual disabilities. As advances in healthcare have increased the life expectancy for adults with neurodevelopmental disabilities (NDD), the possibility of these adults living longer than their parents increases and so does the need for families to plan for long term care (Lindahl et al. 2019) and when parents are no longer able to manage the caregiving role, siblings become the next generation to undertake the role (Heller and Kramer, 2009).
However, the role of siblings as caregivers has not been well documented within existing research (Namkung, Greenberg and Malick, 2017). Arnold, Heller, and Kramer (2012) state how siblings are often overlooked by parents, health professionals and researchers as knowledgeable advocates for their disabled siblings - yet are often the next generation of caregivers. Scorgie (2017) concurs, affirming that within disability research, studies tend to address the effects of caregiving from a parental view, mainly the mother’s perspective, leaving other family members such as siblings, underrepresented. Moghimi (2007) states that a patient’s rehabilitation may be impacted by the burden and stress experienced by a caregiver and should be an area addressed by healthcare providers.

The American Diagnostic and Statistical Manual of Mental Disorders 5th Edition (DSM-5) defines NDD as a group of conditions which typically manifest early with onset in the developmental period. These are a group of conditions which a categorised by developmental deficits which result in impairments in personal, social, academic or occupational functioning. A range of conditions which may include for example, intellectual disabilities, learning disabilities, autism, attention deficit hyperactivity disorder (ADHD), global development delay, communication disorders, tic disorder, motor disorder such as developmental coordination disorders and other unspecified disorders (American Psychiatric Association, 2013).

People diagnosed with an NDD condition can present with a range of impairments which differ in both type and severity, meaning the levels of support required to manage activities of daily living (ADLs) can also vary (Bathje et al. 2018). Occupational Therapists (OTs) work with people with NDDs and their families by using a person’s daily occupations and activities to develop, recover or maintain skills needed for daily living needed through meeting specific
goals. The American Occupational Therapy Practice Framework: Domain and Process (Framework) states that ADLs are activities that are required for engagement in home, community, financial and health matters (American Occupational Therapy Association, 2020).

Lindahl et al (2019) highlights challenges faced by both parental and sibling caregivers, including that caregivers feel anxiety around the long-term care plans for their adult relatives with an intellectual disability. The authors also highlighted that caregivers are likely to be socially and economically disadvantaged and at a greater risk to develop serious health issues. Moghimi (2007) highlights how occupation is central to a person’s identity and competence and many caregivers usually have a variety of occupations fulfilling their lives – with caregiver role often not being an occupation they have chosen. Also, that caregiver burden is often due to the lack of caregiver training from healthcare professionals, as the primary focus for the healthcare professional is the patient, not the caregiver.

The Royal College of Occupational Therapists (2021) have stated that further research is needed to address how OTs can work more effectively with the family and carers of people who access services. Therefore, it is important to consider the perspective of the sibling caregiver, to understand the challenges faced within healthcare services to be able to improve effective working between healthcare professionals and the sibling caregiver.

**Theoretical background**

As stated, the primary focus for the healthcare professional is foremost, the patient, not the caregiver. Yet, occupational therapy (OT) theory is ideally placed to support both the patient and the caregiver. Whilst traditional models, such as the biomedical model have focused on physical factors to aid medical intervention to cure disease or disorder, OT models have
shifted their focus to occupational engagement as an intervention to aid recovery. Thus, historically causing shifts between the biomedical model and holistic occupation-based approaches in occupational theory (Wong, Fagan and Leland, 2018).

OT conceptual models can be used with all client groups and there are no specific recommendations for certain models to be used with a person with an NDD or their caregiver. Therefore, this paper will explore two occupation-focused models, the Model of Human Occupation (MOHO), the Canadian Model of Occupational Performance and Engagement (CMOP-E) and their applicability with supporting caregivers specifically.

MOHO assesses how people generate and modify their occupations within four components: volition, habituation, occupational performance, and environment in a dynamic open-cycle system of human actions. For example, people are motivated to perform occupations (volition), repeat their performance over time (habituation) and consider their perception of their ability to perform that occupation (occupational performance). This process of events is performed with a social and physical context (environment) (Taylor, 2017).

The CMOP-E includes three main components: the person, occupation, and the environment. The person is the core of the model, and is made up of four aspects, spirituality, affective, cognitive, and physical. Occupation includes the domains of self-care, productivity, and leisure, and these are performed in the context of the environment, which is depicted by the domains of physical, social, cultural, and institutional environment. Within the CMOP-E model, the person and the environment are connected through their occupations. The interaction between these three components results in occupational engagement (Townsend and Polatajko, 2013). When the three components work together, this results in function, dysfunction occurs when there is a disruption to one of the three components (Wong and Fisher, 2015).
There is no current evidence applying the models in the context of directly supporting the sibling caregiver as the primary focus within either model. As therapeutic models focus on the ‘client’ specifically, caregivers generally fall into the category of the ‘environment’ for example, in the CMOP-E - social, and in MOHO – people, which include caregivers as input and feedback from the care recipient’s environment (Demers, 2022). MOHO primarily focuses on the client, yet the CMOP-E provides guidelines for using the model with organisations and communities, which could be explored further in context of the caregiver support with organisations and communities (Wong and Fisher, 2015).

Restall and Egan (2021) state that this is a problem in relation to the terminology used by therapists in relation to ‘client-centredness’. The authors state that ‘client-centredness’ has been defined previously to include individuals, families, groups, communities, and populations. Yet, the authors argue that therapists tend to only utilise models with the perspective of the ‘client’ as the individual, not utilising models with larger groups of people. The authors propose a shift in the therapist’s thinking, away from the single, client-centred approach to occupational participation being the central aspect of the model, thus proposing the caregiver can be both the care-recipient and the caregiver.

**Aim**

The aim of the study is to review current literature to find current and future caregiver perspectives on their roles as caregivers to an individual with a NDD and whether their perspectives of healthcare services can influence improvements to the OT practice.

**Method**

A scoping review methodology was undertaken. This type of methodology allowed the author to search and summarise themes within the existing literature and highlight potential gaps
across the current research, allowing for recommendations to be made for future practice (Arksey and O’Malley, 2005). This study has utilised the proposed framework from the Joanna Briggs Institute (JBI) scoping review methodology provided by Peters et al. (2020).

**Eligibility Criteria**

Inclusion criteria included (1) adult (aged 18+) current and future sibling caregivers, with an adult brother or sister diagnosed with a neurodevelopmental disability; (2) Literature which focuses primarily on experiences of adult siblings caring for people with neurodevelopmental disorders, living with sibling caregivers or in social service supported homes; (3) OT-based intervention/support or related interventions given by another healthcare professional; (4) Literature published in English language from 2010 to 2021; (5) Literature with abstracts available; and (6) The search included quantitative, qualitative, and mixed method study designs.

Exclusion criteria includes articles that reference (1) child sibling interventions and perspectives; (2) a sibling diagnosis that is not an NDD; (3) literature including interventions with the mother/father or other relatives and their perspectives; (4) literature that focuses on role identity of siblings within a family context; and (5) interventions from an educational or medical perspective.

**Search**

Sources of literature included published and unpublished data available via electronic databases, reference lists and grey literature. A systematic search of the literature was conducted throughout March 2022. The author performed the search via five primary electronic databases: CINAHL, Medline, AMED, PubMed and PsychINFO and two
secondary databases: Cochrane Library and OT Seeker (Table 1). The author employed a variety of search strings which were revised with the assistance of a university librarian. Similar search strings were used across the databases, although were refined dependent on the structure of the database's search parameters. Initially the search term “neurodevelopmental disorders” was utilised to incorporate a broad spectrum of disorders, yet due to the terminology used across literature internationally, the terms “intellectual disability” and “learning disability” were used alongside “neurodevelopmental disorders” to increase the search field and number of hits. Initial searches including the terms “occupational therapy or OT” produced too narrow results and no relevant evidence. Therefore, this search term was removed to broaden the results and articles were selected via their possible relevancy to OT. The search strings also employed a variation of Boolean operators and truncations.

[Insert Table 1]

An electronic search of specific journals was conducted in four journals: The British Journal of Occupational Therapy; Journal of Autism and Developmental Disorders; Research in Developmental Disabilities and Intellectual and Developmental Disabilities. The author limited the search to these journals as the author believed these may give the most relevant information required to answer the research question. A similar search string was used during the journal search, dependent on the search parameters of the journal (Table 2). Eventually, the search string had to be reduced and generalised to “adult sibling or sibling AND caregiving” (using truncations) as the search field was too narrow to produce relevant results.

[Insert Table 2]
A scoping study method aims to identify all existing literature regardless of the study design (Arksey and O’Malley, 2005). Therefore, the author performed a Google search to find potentially relevant authorities, organisations, and key stakeholders relevant to the topic which could provide further evidence related to the study categorised as grey literature. This resulted in the author reviewing information from authorities such as the UK Department of Health and Social care, National Institute for Healthcare and Excellence (NICE) clinical guidelines and charities such as the Sibling and Leadership Network and Sibs, a UK-based charity. The author also conducted searches of 3 search engines, Google Scholar, OAlster and the Beilefeld Academic Search Engine (BASE).

Study selection

The concluding search of the electronic databases resulted in a total of 235 articles, the articles were then exported to EndNote library and some articles were removed as duplicates. Articles were then screened via title and abstract and all literature was screened against both the inclusion and exclusion criteria when selecting literature to be included into the review. The articles retrieved from the journal search were also screened via their title and abstracts and compared against the inclusion and exclusion criteria. When completing the search of the grey literature they author chose to use information from within the past 5 years as the quality and production of grey literature can vary (Paez, 2017). A full-text assessment of 52 potentially relevant combined articles and grey literature pieces were assessed for eligibility by the author. From the full-text assessment, 11 pieces of literature were included in the final selection. The full screening process is depicted in a PRISMA flow diagram (Figure 1).
Data charting

The final 11 studies were reviewed, and key information was extracted into a “Data charting” table (Table 3). As suggested, for a scoping review methodology, a critical analysis of the quality of the selected studies was not performed (Peters et al. 2020). Instead, an iterative approach was used to produce a summary of extracted characteristic and demographic data from the studies. The ‘data charting form’ was conducted independently by the author and was not tested with an additional researcher.

[Insert Table 3]

Ethical considerations

Scoping reviews do not require ethical approval yet, Suri (2020) suggests critically reflecting upon the position of primary researchers when conducting systematic reviews as the secondary information received may be presented through the subjective lens of the primary researchers. As in systematic reviews, scoping reviews review literature conducted by primary researchers as secondary information. Suri (2020) discusses how researchers of systematic reviews draw upon the philosophical traditions of consequentialism, deontology or virtue ethics when making ethical decisions in relation to secondary research and denotes six steps or guiding principles to ensure the researcher can reduce bias within their systematic or scoping review.

As this review had a sole author, it is important to consider the position of the author (step 1) to understand the author's epistemological perspective, as this demonstrates transparency and limits bias within the review. Duncan (2011) and Suri (2020) both highlight the need for understanding the author's epistemological perspective and how this may impact both
research and practice. In this context, as the author of this paper, I acknowledge that my perspective aligns mostly from a post-positivistic paradigm in deontological ethics, as first and foremost, I, as the author have personal experience as a sibling of a person with an NDD. To ensure my own personal preunderstanding (step 2) or opinion does not transpire into the literature selected for review, it is important as stated by Taylor (2007) that prior to the search, I abide by a clear inclusion/exclusion criterion, to ensure that the internal and external validity of the selected articles are upheld, enhancing the methodological quality. During the search (step 3) it is important as the author to try to scope as much literature as possible, including grey literature, to reduce the possibility of publication and search bias, yet as literature is not peer-review it is important that the author clearly documents each stage of the scoping review study, allowing steps to the study be replicated in future. When evaluating and interpreting results (step 4), it is important that the author consider the quality and relevancy of the evidence in respect to the purpose of the review. Again, abiding by a clear inclusion/exclusion criterion will aid with this and when interpreting the data for thematic analysis, the author can attempt to acknowledge potential bias by using a reflexive approach, reflecting on thoughts and feelings about the selected literature and assumptions that may be generated from the process (step 5). When recommending implications for practice, the author must acknowledge the methodological considerations of the study and how aspects of the study may impact the recommendations (step 6). For example, within this study, acknowledging that the small, limited number of results from this scoping review cannot be generalised to a larger population.

Results

The final 11 studies compromised of 8 qualitative, 2 quantitative and 1 mixed method study. Most of the studies originated from the USA (n = 6) and other countries including, UK (n =
There were a total of 1181 participants in the review, with 1181 (100%) being a sibling of a brother or sister diagnosed with an NDD. 1180 (99.9%) of the participants were over 18 years old, in line with the inclusion criteria, only 1 participant was aged 17 years old, and this study was included due to its relevancy to answering the aim of the study.

To answer the aim accordingly, the author reviewed all 11 articles, read and re-read the articles, kept reflective notes, assessed, and interpreted the data and extracted the key findings from the articles into themes, relevant to answering the aim. The results highlighted that the role of the sibling caregiver was not addressed within OT service in the literature. However, the experiences of how well sibling caregivers are supported within healthcare services were addressed and ideas and opinions of how services can be improved to support sibling caregivers was found from the 11 articles. The data charting process and analysis resulted in key findings in 5 areas: (1) Need for education and training opportunities in relation to caregiving; (2) Policy and guideline changes in relation to sibling caregiver involvement; (3) Sibling caregiver inclusion into the care pathway; (4) Need for improvements in service coordination; and (5) Sibling caregivers' lack of knowledge in guardianship matters.

**Need for education and training opportunities in relation to caregiving.**

The most prominent theme to arise from the literature was the need for sibling caregivers to access opportunities to engage in education and training specific to their caregiving role. Many of the studies chosen for analysis discussed this topic in some context. Service-specific needs included more education and training about the understanding of disability services, roles of staff and more support for the sibling caregiver as they transition into the caring role (Arnold et al. 2012; Rawson 2010). Some sibling caregivers highlighted activity specific
needs, such as time management issues with daily living tasks, knowledge about social activities which can enhance the sibling relationship and advice for supporting their siblings with typical self-care and daily life tasks (Burke et al. 2015; Davys et al. 2016; Mauldin and Saxena 2018; Leane 2019).

Hinek and Milakovic (2019) reported sibling caregivers requiring other types of support, such as challenges faced with communication, interaction and challenging behaviours of their brother and sisters. The sibling caregivers suggested healthcare services could support further by linking them to other typically developing sibling caregivers to share experiences. Rawson (2010) also noted that sibling caregivers discussed concerns that their sibling’s cognitive development changes as they age and similarly, Mauldin and Saxena (2018) highlight the difference in sibling caregiver needs at different life stages, noting that task-based activities increase as the sibling's age increases. One study had implemented a training program with future sibling caregivers, in which future sibling caregivers attended four 2-hr training sessions about sibling perspectives, identifying support needs, learning problem solving skills, legal and financial planning and finally writing a letter of intent and connecting to other sibling organisations. Outcomes from this study demonstrated that participants felt more empowered, connected to others, felt improved communication with family, and had more knowledge of adult disability services (Lee and Burke, 2021). Another study reported that the sibling participants felt that their preferred way to access information is from online website sources (Redquest et al. 2020).

**Policy and guideline changes in relation to sibling caregiver involvement.**

The second most prominent topic refers to how policies can be changed to express the ‘voice’ of sibling caregivers and how these caregivers are powerful advocates for their
brothers/sisters. Some of the articles referred to advocacy. Arnold et al. (2012) states that the disability advocacy movement would benefit from involving sibling caregivers. Many sibling caregivers stated their preference for learning more about advocacy, supporting their siblings in self-advocacy and stated that having a greater involvement in advocacy matters may lead to a positive outcome in current caregiving (Brady et al. 2019; Lee et al. 2019). In the sibling training program conducted by Lee and Burke (2021) sibling participants of this study reported feeling more empowered when handling advocacy matters.

**Sibling caregiver inclusion into the care pathway.**

Inclusion of sibling caregivers into the care pathway was a recurring theme in the data. Some of the articles discussed how sibling caregivers should be included in their brother/sisters’ care planning from the start of service intervention or stated issues within current service delivery. Several participants discussed how they felt as the sibling caregiver that they had important information that related to the care needs of their brother/sister, or they were already heavily involved with their brother/sister’s care planning (Arnold et al. 2012; Burke et al. 2015; Rawson 2010). Yet, these sibling caregivers did not feel that they were either included in the care planning of their brother/sister or were not included early enough (Arnold et al. 2012). Also, some sibling participants discussed their concern with transitioning into the caregiver role (Burke et al. 2015). Many sibling caregivers stated that services had poor coordination and reported a distrust in services or a lack of confidence in service providers (Arnold et al. 2012; Davys et al. 2016).

**Need for improvements in service coordination.**

Many sibling caregivers shared frustrations related to the need for improvements within the system and how access to other services could support their needs. Two of the articles
revealed their frustrations with the ‘system’, needing more services to support their own health and wellbeing and for information to be shared more freely, with support in understanding bureaucracy and ultimately coordination of services (Arnold et al. 2012; Burke et al. 2015).

**Sibling caregivers' lack of knowledge in guardianship matters.**

Three of the articles referred to sibling caregivers expressing their ‘lack of knowledge’, in relation – predominately, to legalities about their role as a caregiver. Nearly all participants in the study completed by Rawson (2010) reported a lack of knowledge about guardianship and their rights. Brady et al. (2019) stated that 40% of participants reported they did not understand guardianship, yet all participants reported that guardianship was necessary for their brother/sister. Participants suggested that siblings could be involved in care planning as early as 16 years of age (Rawson 2010).

**Discussion**

The aim of this study is to review current literature to find current and future sibling caregiver perspectives on their roles as caregivers to an individual with a neurodevelopmental disorder and whether their perspectives of healthcare services can influence improvements to the OT practice.

Implications for research

In relation to research, the most prominent need arose around sibling caregivers feeling a need for their role as a current or prospective sibling caregiver to be included specifically at both a local and national policy level, suggesting that this may lead to their inclusion to their brother or sister’s care pathway right from the start. Many national policies do not reflect the
role of the sibling caregiver. For example, in the UK, the Royal College of Occupational Therapists (2022) highlights laws and policies which guide OT services and there is little reflection on the role of the sibling caregiver. Concurrent with this, ‘The Care Act 2014’ UK law states rights specifically for both young carers and parents, yet not siblings. Formalising documentation to include siblings as caregivers, would ensure the sibling caregiver is included in the planning process.

The Royal College of Occupational Therapists (2022) holds regular consultations to discuss aspects of policy and changes needed. OTs may find it beneficial to involve themselves with local meetings, such as these to ensure they can influence policies relevant to sibling caregivers. However, OT services are also instructed to meet certain expectations set out by healthcare authorities. Often, OTs report problems in meeting recommendations relayed from research and using evidence-based practice (EBP). Practical barriers such as lack of time, little support from their employment and a need for more training prevented using EBP in practice (Reep, 2018). Therefore, the author recommends that from this review, OTs consider the most significant strategies which could be implemented into service in their country, whilst acknowledging the barriers in their practice.

Implications for practice

In relation to practice, the results have highlighted the most prominent views of both current and future sibling caregivers in relation to both their personal situations and sibling perspectives of healthcare services in general. Therefore, this review finds some practical strategies that can be taken into consideration within OT, to improve services.
Relevant strategies from participants include prominently, providing access to educational opportunities in relation to caregiving for their sibling. Many strategies were suggested by participants of the studies, these included sibling-specific training days, group work scenarios, sibling-specific information books or websites and more information sessions relating to their sibling’s condition and how this may change over time. One study included a sibling training program, tested with a small sample of 18 sibling participants and resulted in positive feedback from the sibling participants (Lee and Burke, 2021). Further research is needed to demonstrate whether this type of sibling training program could be supported by OTs as the study by Request et al. (2020) reported that sibling participants would prefer to receive information via online sources, Therefore, how to disperse this information most effectively would need to be considered.

Some participants discussed daily living specific issues of their own, for example, time-management as a barrier to their own performance in supporting their sibling with an NDD. Some of the current sibling participants discussed requiring more assistance when supporting the care of their sibling in their ADLs. This is possibly where OTs can work more effectively in practice – by providing further support to sibling caregivers by arranging support days in the form of training.

Including sibling caregivers into the care pathway was discussed as an important problem by the sibling participants of the studies. Concerns arose around the feasibility of being able to share information with healthcare professionals about their brother or sister, being included to the care planning stages early enough and challenges around transitioning into the caregiver role. Ensuring involvement in the OT process of siblings at a younger age in the care
planning process may provide more opportunities for siblings to learn the key elements involved in care planning and their rights as caregivers. As the sibling participants reported distrust and a lack of confidence in service providers, it may be one strategy to ensure that future or prospective sibling caregivers of a person with an NDD, are invited to join care planning meetings within a multi-disciplinary setting. Prospective sibling caregivers would be able to discuss pertinent information regarding both their brother or sister, and themselves in their future transition to the caregiver role. This may start to or continue to build trust between the families and the healthcare services.

Transitioning into roles and role identity is one aspect which falls into the OT domain and is one area where OTs are more able to support sibling caregivers. Termed ‘occupational identity’, this refers to a person’s sense of self, from how they engage in their own occupations (Hansson, et al. 2022). This includes work, social and leisure occupations. Howes and Ellison (2022) describe how threats to participation in leisure and social activities impacted and adapted some caregiver’s occupational participation to engage and meet the demands of the caregiving role. The authors also state that some caregivers are required to change their role at work or give up their role entirely to meet the demands of being a caregiver, yet also that OTs have the knowledge and skills to support caregivers with their occupational participation, which can have a positive impact on their occupational identity.

There are some aspects from the results, that sibling participants discussed - that don’t fall into directly into the OT domain, these were identified as finding more information related to guardianship matters, knowledge about advocacy and sibling participants concerns over their own health and wellbeing. However, OTs being able to or continuing to signpost/refer to
relevant services would support service coordination further in this instance. In the UK the Royal College of Occupational Therapists (2019) actively promotes OTs to signpost to local community support services to enhance social participation.

Conclusions

Although all the participants in the studies were siblings who had brother/sister with an NDD, there was no link found specifically between the role of siblings within OT service. As The Royal College of Occupational Therapists (2021) stated that further research is needed to address how OT’s can work more effectively with family and carers of people who access OT services, this review demonstrates that more research is required in this area, both in areas of research and practice. This review demonstrates some practical strategies which OTs can take, which may help to improve OT services, from the sibling caregiver perspective. Although, further empirical research is required to truly reflect how the sibling caregiver role is experienced within the domain of OT.

Methodological considerations

There are methodological limitations within this review. The review had one sole author. As suggested by Taylor (2007) to enhance the rigour and reduce bias within this study, the author should have engaged at least one more reviewer of the data. To increase the reliability of the results as recommended by Arksey and O’Malley (2005), the author has clearly detailed the process and steps taken to complete this review, to ensure the process can be repeated.
Other limitations include that only a partial number of databases, journals and grey literature sources were searched, in only English language, limiting the number of relevant articles found. The search was limited to articles which included an abstract only, further hindering the scope of the search and enhancing the possibility that some literature which may have fitted the inclusion criteria was excluded.

Finally, most of the studies included in the review used convenience, snowball, or deliberate sampling method types, the highest method of sampling used was convenience sampling and then snowball sampling. This demonstrates that the primary researchers may have been unable to attain a purposive sample which can lead to a biased and unrepresentative sample within the scoping review. reducing the transferability of the review and the ability to generalise the findings to a larger population (Taylor, 2007). Also, majority of the selected studies reported recruiting their participants through Autism/sibling or disability networks and organisations. Therefore, it is likely a high percentage of the siblings may already have some form of support network in place and may not show a typical representation of the sample. It is clear within the selected literature that there are a small group of researchers who regularly engage in research about sibling caregiving and these authors crossover and co-author other selected literature. Whilst this may impact the quality and content of the data available, it also demonstrates the current limited available evidence and supports the conclusion of the need for further research.
References

Articles included into the review are marked with an *


https://doi.org/10.1016/j.socscimed.2019.04.022


https://doi.org/10.1097/01.TGR.0000284770.39958.79


Royal College of Occupational Therapists. (2021). *Identifying research priorities for occupational therapy in the UK: What matters most to the people accessing and delivering services?* https://www.rcot.co.uk/identifying-research-priorities-occupational-therapy-uk


Table 1. Electronic database search

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<td>neurodevelopmental disorders AND sibling</td>
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<tr>
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<td>learning disabilit* AND sibling</td>
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<tr>
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<td>autis* AND sibling</td>
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Table 2. Journal search

<table>
<thead>
<tr>
<th>Journal</th>
<th>Limits</th>
<th>Search terms</th>
<th>Number of hits</th>
<th>Chosen by abstract</th>
<th>Final number of articles included</th>
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</thead>
<tbody>
<tr>
<td>1. British Journal of Occupational Therapy</td>
<td>2010-2021</td>
<td>sibling AND caregiv*</td>
<td>29</td>
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<tr>
<td>2. Journal of Autism and Developmental Disorders</td>
<td>2010-2021, English language</td>
<td>adult sibling* AND caregiv*</td>
<td>286</td>
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<tr>
<td>3. Research in Developmental Disabilities</td>
<td>2010-2021</td>
<td>adult sibling AND caregiving</td>
<td>39</td>
<td>6</td>
<td>0</td>
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<tr>
<td>4. Intellectual and Developmental Disabilities</td>
<td>2010-01-01 to 2021-12-31, English language</td>
<td>adult siblings AND caregiv*</td>
<td>51</td>
<td>8</td>
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</tbody>
</table>
Table 3. Data charting

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Population</th>
<th>Sample</th>
<th>Recruitment Method</th>
<th>Study design</th>
<th>Ethical considerations</th>
<th>Purpose</th>
</tr>
</thead>
<tbody>
<tr>
<td>Arnold, C, Heller, T and</td>
<td>2012</td>
<td>139 sibling</td>
<td>Convenience</td>
<td>Recruited through an online sibling group and state-wide sibling conference.</td>
<td>Grounded theory</td>
<td>Consent and ethics for study not reported.</td>
<td>Examines support needs of adult siblings of people with developmental disabilities</td>
</tr>
<tr>
<td>Kramer (2012)</td>
<td></td>
<td>participants</td>
<td></td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>US</td>
<td></td>
<td></td>
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<tr>
<td>Brady, A., Burke, M.,</td>
<td>2019</td>
<td>10 sibling</td>
<td>Combination of convenience and theoretical sampling</td>
<td>Recruited through disability organisations and social media.</td>
<td>Grounded theory</td>
<td>Board ethics gained.</td>
<td>Explore the knowledge and understanding of guardianship for siblings</td>
</tr>
<tr>
<td>Landon, T., and Oertle, K.</td>
<td></td>
<td>participants (3 current caregivers, 1 past, 6 possible future caregivers)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(2019)</td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>US</td>
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<td></td>
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<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Burke, M., Fish, T., and</td>
<td>2015</td>
<td>42 sibling</td>
<td>Not reported</td>
<td>Personal contacts and social media used to recruit from three USA states through sibling organisations.</td>
<td>Qualitative study</td>
<td>Board ethics gained. Participant consent gained.</td>
<td>Compare the perceptions of caregiving between adult siblings who currently are caregivers and those who are anticipated caregivers in the future</td>
</tr>
<tr>
<td>Lawton, K. (2015)</td>
<td></td>
<td>participants (25 current caregivers and 17 anticipated caregivers.)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Authors</td>
<td>Country</td>
<td>Participants</td>
<td>Recruitment Method</td>
<td>Study Design</td>
<td>Consent</td>
<td>Ethics</td>
<td>Research Objectives</td>
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<tr>
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</tr>
<tr>
<td>Hinek, S., and Milakovic (2019)</td>
<td>Croatia</td>
<td>6 sibling participants</td>
<td>Deliberate sampling</td>
<td>Qualitative study</td>
<td>Oral consent gained, ethics for study not reported</td>
<td>To explore the experiences of siblings living with a brother with ASD and the perceptions of received and needed support.</td>
<td></td>
</tr>
<tr>
<td>Leane, M (2019)</td>
<td>Ireland</td>
<td>25 sibling participants</td>
<td>Not reported</td>
<td>Qualitative study</td>
<td>Board ethics gained. Written consent gained. Anonymity reported.</td>
<td>To explore perceptions of sibling relationships and expectations and aspirations of their caregiving roles in the future.</td>
<td></td>
</tr>
<tr>
<td>Lee, C., Burke, M., Arnold, C., and Owen, A. (2019)</td>
<td>US</td>
<td>429 sibling participants</td>
<td>Convenience sampling</td>
<td>Quantitative study</td>
<td>Board ethics gained. Consent not reported.</td>
<td>To explore the bioecological correlates in current sibling caregiving to identify who is likely to become a caregiver and identified for support.</td>
<td></td>
</tr>
<tr>
<td>Lee., C., and Burke, M. (2021)</td>
<td>US</td>
<td>18 sibling participants (all future planned caregivers)</td>
<td>Selective snowball sampling</td>
<td>Mixed method study</td>
<td>Board ethics gained. Written consent gained.</td>
<td>To evaluate the outcomes and feasibility of the Sibling Training for Early Future Planning (STEP) program.</td>
<td></td>
</tr>
<tr>
<td>Author(s)</td>
<td>Year</td>
<td>Country</td>
<td>Sample Size</td>
<td>Sampling Method</td>
<td>Methodology</td>
<td>Ethics</td>
<td>Focus</td>
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<td>----------------------------------------------------------------------</td>
</tr>
<tr>
<td>Mauldin, L., and Saxena, M</td>
<td>2018</td>
<td>US</td>
<td>224 siblings</td>
<td>Snowball sampling</td>
<td>Link to web survey sent to 245 organisations and agencies across USA.</td>
<td>Grounded theory</td>
<td>Board ethics gained. Consent not reported.</td>
</tr>
<tr>
<td>Rawson, H</td>
<td>2010</td>
<td>UK</td>
<td>13 siblings</td>
<td>Not reported</td>
<td>Siblings invited via letter to their home via their brother/sisters who attend a special needs residential school.</td>
<td>Qualitative study</td>
<td>Board ethics gained. Consent gained. Anonymity reported.</td>
</tr>
<tr>
<td>Redquest, B., Tint, A., Ries, H., Goll, E., Rossi, B., and Lunsky, Y</td>
<td>2020</td>
<td>Canada</td>
<td>260 siblings</td>
<td>Not reported</td>
<td>Secondary analysis of data.</td>
<td>Quantitative study</td>
<td>Board ethics gained.</td>
</tr>
</tbody>
</table>
Figure 1: PRISMA 2020 flow diagram (Page, et al. 2020)

*Consider, if feasible to do so, reporting the number of records identified from each database or register searched (rather than the total number across all databases/registers).

**If automation tools were used, indicate how many records were excluded by a human and how many were excluded by automation tools.