

BMJ Open Support programmes for parents of children with intellectual disabilities: a scoping review protocol

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ABSTRACT

Introduction Evidence shows that parents of children with intellectual disabilities experience high levels of stress, emotional challenges and higher levels of depression. Despite these challenges, support for parents has tended to be peripheral in healthcare services of children. There is a lack of research attention given to support programmes for parents of children with intellectual disabilities despite the importance of parenting for the optimal development of children with intellectual disabilities.

Methods and analysis This review will follow the Joanna Briggs Institute scoping review methods manual. The search for relevant studies will be in Medline (EbscoHost), PsychARTICLES, PubMed, CINAHL, Academic Search Complete and SAePublications. Reference mining of full review studies will be conducted. A three-step search strategy will be used, including the use of information management software to manage the search results and to remove duplications. An independent reviewer will extract data using a data extraction tool. Furthermore, another reviewer will screen the included articles in conjunction with the results of the scoping review. The data extracted will be presented in a tabular format with a narrative summary related to the key findings.

Ethics and dissemination This scoping review has received ethical approval (BM20/4/26). A summary of the findings of the study will be published in a peer-reviewed journal. This scoping review will contribute to a better understanding of the support programmes available to parents of children with intellectual disabilities. This could be the first step in highlighting the gaps and future directions for the development and implementation of support programmes for parents of children with intellectual disabilities.

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INTRODUCTION

Intellectual disability (ID) affects 4 in 100 people in South Africa to some degree.¹ IDs are neurodevelopmental disorders that begin in childhood and are characterised by intellectual difficulties as well as difficulties in conceptual, social and practical areas of living. The DSM-5 (The Diagnostic and

STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ This scoping review will provide an evidenced base of available support programmes for parents of children with intellectual disabilities in a wide variety of settings.
- ⇒ A scoping review is practical method to synthesis research in a wide range of methodological approaches, settings and study populations on the available literature of support programmes for parents of children with intellectual disabilities.
- ⇒ This scoping review will highlight the key research gaps and research needs for transformation in the health sector for the support needs of parents who have children with intellectual disabilities.
- ⇒ One of the limitations is that the methodological quality of included studies was not assessed.
- ⇒ The synthesis of data will be limited to English studies that were published in peer-reviewed journals.

Statistical Manual of Mental Disorders, Fifth Edition) diagnosis of IDs requires the satisfaction of three criteria: (1) Deficits in intellectual functioning—‘reasoning, problem solving, planning, abstract thinking, judgement, academic learning and learning from experience’—confirmed by clinical evaluation and individualised standard IQ testing; (2) Deficits in adaptive functioning that significantly hamper conforming to developmental and sociocultural standards for the individual’s independence and ability to meet their social responsibility and (3) The onset of these deficits during the developmental period.²(p33) There is, however, a lack of policy development and implementation for child and adolescent mental health (CAMH) including children and adolescents with IDs, and this is particularly prevalent in low-income and middle-income countries.³ ID places a high burden of disease on low-income and middle-income countries and has become a public health priority because of its continuation throughout the life cycle as well as the impact it has on the family.⁴

Parents of children with IDs experience high levels of stress relating to subjective factors such as feelings of social isolation and life dissatisfaction, and factors relating to societal and institutional marginalisation.⁵ There are a multitude of emotional challenges experienced by these parents, in particular in relation to guilt over possibly being the cause of their child's disability (reasons being both logical and illogical) by parents who have children with IDs.⁵ Parenting a child with IDs is often experienced as being more emotionally demanding and stressful than parenting a child without those challenges.⁵ Depression is a measure that has been used to assess the adverse effects of stress and adaptive capacities of parents who have children with IDs. It has been indicated that mothers of children with IDs experience elevated levels of depression when compared with control group.⁶ However, support for parents in the mental healthcare of children has tended to be a peripheral focus. This indicates that there is a lack of research attention given to support for parents caring for children with IDs, even though parenting is vital for the optimal development and general care of children with this condition.⁷ Studies conducted in South Africa on those caring for individuals with mental disorders and disabilities focused mainly on informal caregiver burden with mainly females being the carers of older persons in the community or the family.⁸ The caregivers' burdens comprise financial strains, privacy issues, social life status and physical and sleep disturbances.⁸ In South Africa, people with mental illness and disabilities experience significant amounts of stigma, which could lead to social isolation, low-self-esteem, depression, poor social skills, marginalisation, unemployment and housing difficulties.⁸

There are many barriers to caring for children with IDs. This is especially true among families who have complex social situations. For the successful empowerment of families, it is important to address the various obstacles parents of children with IDs consistently face. These obstacles include: the stigma attached to IDs; belief systems surrounding the child and social behaviour (eg, proper disciplining of the child will eradicate the problem), and the attitudes parents have towards mental healthcare services (eg, a lack of trust in service providers based on previous experiences).⁹ Stigma not only affects individuals or groups of individuals who carry the stigmatising attribute (in this case ID) but also spills over into those affiliated with them.¹⁰ Parents of children with IDs often experience affiliated stigma—the process of internalising the public's negative views towards themselves because of their child's disability.¹¹ Affiliated stigma can adversely affect the quality of life of parents because they are undervalued by others and the continuous battles they encounter with service. To cope with the stigma parents may withdraw socially or conceal their child's disability.¹¹ The continuous and long-term use of these coping strategies could potentially lead to lowered self-esteem, and the individual with an ID experiencing increased levels of discrimination.¹¹ It is imperative to understand the belief

systems surrounding the child and social behaviour. Parents' perceptions about the nature of their child's disability is a focal point in many different cultural groups, and disabilities are also viewed differently across different cultures. The perceptions held by parents' and society shape the parents' attitudes towards their children and their disabilities. These perceptions are also instrumental in the resources and treatment that parents will be able to invest in for the training and education of their children with a disability; as well as the views parents have about their children's future.¹² It is also important to understand how the contrasting perspectives of parents and clinicians influences their relations, the care and service provision. This contrast too frequently leads to perpetual blame games between parents and clinicians, where clinicians blame parents for their lack of commitment and parents complain about the lack of understanding from clinicians. This results in children and adolescents not receiving the proper care to address their disability and parents feeling overwhelmed. Therefore, for continuity of care to occur in the treatment of children with IDs through family-focused and patient-centred medical care, collaboration between healthcare professionals and parents should be considered. It is crucial that families and physicians should not view each other as adversaries or untrustworthy, but instead as partners striving to achieve the same goal.⁹

A preliminary search of PROSPERO, MEDLINE, the Cochrane Database of Systematic Reviews and the JBI Evidence Synthesis was conducted. Dew *et al*¹³ reported on the peer support experiences of mothers of children with ID and challenging behaviour. Stuttard *et al*¹⁴ reported on the evaluation of a group-delivered intervention (Riding the Rapids); this programme was developed specifically for parents of children with a disability or autistic spectrum disorder. However, the preliminary search found no current scoping or systematic reviews or systematic and scoping reviews that are currently underway that focused on support programmes for parents of children with IDs. Various studies have been done on support programmes for parents of children with IDs, however, not many have mapped and synthesised the different elements of these programmes to highlight the gaps for future programmes. Therefore, a synthesis of the body of evidence that is currently available in the area is needed to inform evidence-based programme development for parents of children with IDs. A scoping review on the available body of literature on support programmes for parents of children with IDs is proposed. The objective of this scoping review is to describe, map and synthesise the available literature on support programmes for parents of children with IDs.

METHODS AND ANALYSIS

The various systematic approaches available were considered in order to review the published literature on support programmes for parents of children with IDs. We

chose to undertake a scoping review of published literature as this was the best method to describe, map and synthesise the evidence to identify the gaps in literature on parent support programmes. The proposed scoping review will be conducted in accordance with the Joanna Briggs Institute methodology for scoping reviews.¹⁵

Review question(s)

1. What programmes have been carried out and literature published in peer reviewed journals around support programmes for parents of children with intellectual disabilities?
2. What target populations are being addressed in peer reviewed published literature around support programmes for parents of children with intellectual disabilities?
3. What types of interventions are being carried out and evaluated around support for parents who have children with intellectual disabilities?
4. What are the key findings related to the studies carried out around support programmes for parents of children with intellectual disabilities?
5. What are the types of support offered in the support programmes for parents of children with intellectual disabilities?
6. Who offers the support programmes to parents of children with intellectual disabilities?
7. What are the characteristics of support programmes for parents of children with intellectual disabilities?

Inclusion criteria

The inclusion criteria were categorised according to the population, concept and context (PCC) mnemonic recommended by the Joanna Briggs Institute for scoping reviews. This is less restrictive than the population, intervention, comparator, outcome mnemonic which is recommended for systematic reviews.

Participants

- ▶ Studies that include parents of children (0–18 years) with IDs. Parents are defined as men and/or women who perform a parenting role in terms of caring for their child and his/her needs. The term ‘parents’ will include, but is not limited to: biological, adoptive, step, foster parents and guardians.

Concept

- ▶ Studies that explore parent support programmes that are designed to support parents who have children with IDs carried out between 2003 and 2021. This time frame was chosen as in 2003 the CAMH policy framework was developed to be implemented in South Africa.³
- ▶ Included as concepts will be all types of programmes, modes of delivery, setting, leadership and duration. Examples of the types of programmes that could be included are social support, peer support, psychoeducational support, knowledge support, emotional support and all other variants including

multi component interventions. Examples of modes of delivery could include support groups, telephonic interactions and self-delivery (through use of manuals). Examples of settings could include clinical settings, home, community settings. Examples of leadership could include: clinician led, family led and team led.

- ▶ Full text.
- ▶ Peer reviewed.
- ▶ Literature published in the English medium.

Context

- ▶ Clinical settings.
- ▶ Community settings.
- ▶ Virtual settings or any other settings where support programmes for parents of children with IDs are delivered.
- ▶ All countries and economic settings will be included in the study.
- ▶ Quantitative, qualitative and mixed-methods study approaches will be considered for inclusion. Quantitative study designs to be considered for the review will include experimental and quasi-experimental including randomised control trials, non-randomised control trials, pretest post-test studies and interrupted time series. In addition, cohort studies, case–control studies and cross-sectional studies will be considered for inclusion. All qualitative research including explanatory, exploratory, descriptive and applied types of research using different application techniques will be considered for inclusion.

Exclusion criteria

Excluded from the study will be systematic reviews, scoping reviews and studies that were not interventions and interventions supporting parents with IDs.

Search strategy

The search strategy will follow a three-phase approach with an aim to find published studies. A literature search will be conducted to identify parent support programmes aimed at parents who have children with IDs. In the first phase, an initial limited search of Medline (EbscoHost) and PubMed will be undertaken to identify articles on the topic. The text words contained in the titles and abstracts of relevant articles, and the index terms used to describe the articles related to the proposed scoping review will inform the second phase where a full search strategy for relevant database platforms; these include Medline (Ebscohost), PsychARTICLES, CINAHL, PubMed, Academic Search Complete and SA ePublications. PubMed is detailed in online supplemental appendix 1. In the third phase of the search, the reference lists of articles selected for full-text review will be screened for additional papers.

Following the search, all identified records will be collated and uploaded into Mendeley (Mendeley, Elsevier, Netherlands) and duplicates removed. Following a pilot test, titles and abstracts will then be screened by

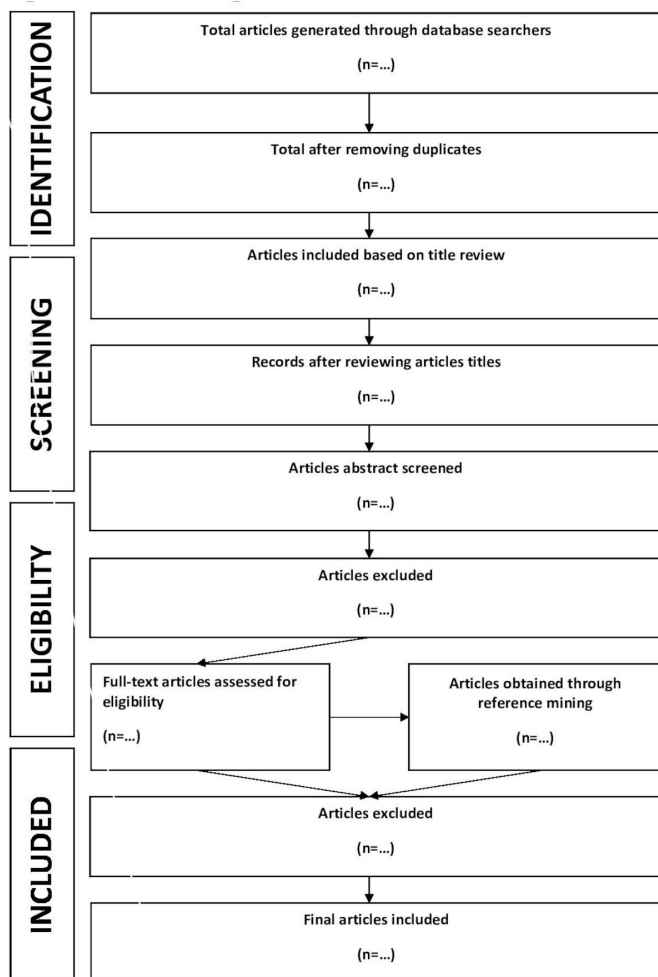


Figure 1 PRISMA flow diagram. PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-analyses.

one independent reviewer (JEM) for assessment against the inclusion criteria for the review. Potentially relevant papers will be retrieved and their citations and details will be recorded in Microsoft Excel. The full text of the selected studies will go through a detailed assessment against the inclusion criteria by two independent reviewers (JEM and LW). Disagreements will be resolved via discussion and where no agreement can be reached a third independent reviewer (TM) will be consulted for further discussion. Reasons for exclusion of full-text papers that do not meet the inclusion criteria will be recorded and reported on in the final scoping review. The results of the search will be reported in full in the final scoping review and presented in a Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) flow diagram (figure 1).¹⁶

Data extraction

Data will be extracted from papers included in the scoping review by one independent reviewer (JEM) and assessed by two independent reviewers (LW and TM) using a data extraction tool developed by the reviewers. The draft data extraction instrument (online supplemental appendix 2) was adapted from the Methodology for Joanna Biggs Institute scoping reviews¹³ and Hoagwood *et al*⁷ to answer

the review question. The data extracted will include specific details about the population, concept, context, methods leadership, programme characteristics and key findings relevant to the research question. Modifications of the JBI data extraction tool will consist of the following items: intervention name, type of support provided, leadership (eg, clinician led), and programme characteristics. If necessary modifications to the data extraction tool will be made and revised during the data extraction process of each included study. All modifications made will be detailed in the full scoping review. If any disagreements arise between reviewers (JEM and LW), these will be resolved through discussion with a third reviewer (TM). Authors of papers will be contacted to request missing or additional data, where required. If authors do not respond to the request a decision to include or exclude the article based the information available.

Synthesis and reporting of results

The data extracted will be presented in a tabular format with a narrative summary related to the key findings. The results will be presented following the PCC format as well as the main conceptual categories used in the data extraction tool. A narrative description of the findings will be guided by the thematic analysis of the qualitative findings. The tabulated results using descriptive analysis, accompanied by the narrative results will be presented and framed in relation to the review objective and research question of this scoping review. A checklist for protocol submission guidelines PRISMA-Protocols has been completed (online supplemental appendix 3).¹⁷

Public and patient involvement

No patient involved

ETHICS AND DISSEMINATION

This scoping review has received ethical approval from the University of the Western Cape Biomedical Research Ethics Committee (BM20/4/26). A summary of the findings of the study will be published in a peer-reviewed journal. This scoping review will contribute to a better understanding of the support programs available to parents of children with intellectual disabilities. This could be the first step in highlighting the gaps and future directions for the development and implementation of support programs for parents of children with intellectual disabilities.

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REFERENCES

- Western Cape Forum for Intellectual Disability [Internet]. Western Cape forum for intellectual disability, 2020. Available: <https://wcfid.co.za/frequently-asked/> [Accessed 15 Jan 2019].
- American Psychiatric Association. *Diagnostic and statistical manual of mental disorders*. 5th ed, 2013.
- Mokitimi S, Schneider M, de Vries PJ. Child and adolescent mental health policy in South Africa: history, current policy development and implementation, and policy analysis. *Int J Ment Health Syst* 2018;12:36.
- Durkin MS, Schneider H, Pathania VS. Chapter 49: Learning and developmental disabilities. In: Jamison DT, Measham AR, eds. *Disease control priorities in developing countries [Internet]*. 2nd ed. Washington DC: The International Bank for Reconstruction and Development / The World Bank, 2006.
- Azeem MW, Dogar IA, Shah S, et al. Anxiety and depression among parents of children with intellectual disability in Pakistan. *J Can Acad Child Adolesc Psychiatry* 2013;22:290–5.
- Norlin D, Broberg M. Parents of children with and without intellectual disability: couple relationship and individual well-being. *J Intellect Disabil Res* 2013;57:552–66.
- Hoagwood KE, Cavaleri MA, Serene Olin S, et al. Family support in children's mental health: a review and synthesis. *Clin Child Fam Psychol Rev* 2010;13:1–45.
- Yakubu YA, Schutte DW. Caregiver attributes and socio-demographic determinants of caregiving burden in selected low-income communities in Cape town, South Africa. *J Compassionate Health Care* 2018;5.
- Kelleher KJ, Hoagwood K. Beyond blame: parents as partners. *J Pediatr* 2015;167:795–6.
- Stigma GE. *Notes on the management of spoiled identity*. New York: Simon and Shuster, 1963.
- Mitter N, Ali A, Scior K. Stigma experienced by families of individuals with intellectual disabilities and autism: a systematic review. *Res Dev Disabil* 2019;89:10–21.
- Mohamed Madi S, Mandy A, Aranda K. The perception of disability among mothers living with a child with cerebral palsy in Saudi Arabia. *Glob Qual Nurs Res* 2019;6:1–11.
- Dew A, Collings S, Dowse L, et al. 'I don't feel like I'm in this on my own': Peer support for mothers of children with intellectual disability and challenging behaviour. *J Intellect Disabil* 2019;23:344–58.
- Stuttard L, Beresford B, Clarke S, et al. Riding the rapids: living with autism or disability--an evaluation of a parenting support intervention for parents of disabled children. *Res Dev Disabil* 2014;35:2371–83.
- Peters MDJ, Godfrey C, McInerney P. Chapter 11: Scoping Reviews (2020 version). In: Aromataris E, Munn Z, eds. *JBI manual for evidence synthesis [Internet]*. Adelaide: JBI, 2020. <https://synthesismanual.jbi.global>
- Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med* 2009;6:e1000097.
- Moher D, Shamseer L, Clarke M, et al. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* 2015;4:1–9.