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Optimizing school readiness for children with developmental disabilities

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Optimizing school readiness for children with developmental disabilities

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Editorial: Optimizing school readiness for children with developmental disabilities

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Editorial on the Research Topic
Optimizing school readiness for children with developmental disabilities

> *"The only thing worse than being blind is having sight with no vision"*
>
> ~Helen Keller

In September 2015, the 193 Member States of the United Nations (UN) undertook a social contract to advance population health, well-being, and security over the life-course globally under the Sustainable Development Goals (SDGs) (1). This global agenda, for the first time, set out a global vision for early childhood development for children under 5 years with the primary objective of facilitating access to inclusive and equitable quality early childhood education for children with or at risk of developmental disabilities (**Figure 1**). At that time, when the SDGs were launched, limited data was available on the state of the world's children with developmental delays and disabilities. This data gap was swiftly utilized by WHO, UNICEF and the World Bank to promote an early childhood development program in 2018 tagged the Nurturing Care Framework (NCF), based on an estimated 250 million children suspected to be at risk of poor cognitive development due to stunting and extreme poverty in LMICs (2). However, the NCF was neither geared towards promoting school readiness for inclusive education for children with developmental disabilities as envisioned by the SDGs, nor was it endorsed or accredited as a global program under the SDGs (3).

Emerging data since 2018 has now shown that every day, some 145,000 babies are born with or acquire lifelong disability in early childhood (4). The likelihood of a child being disabled is estimated to be ten-fold than of dying before the fifth birthday (5). A landmark report from UNICEF in 2021 further showed that, compared to children without disabilities, children with disabilities are significantly less likely to have foundational reading and numeracy skills, more likely to have never attended school and are more likely to be out of primary school (6). Lack of formal and quality education places children with developmental disabilities at greater risk of not securing gainful employment and at a higher risk of social exclusion and isolation. This trajectory

FIGURE 1
Pathway to early childhood development for inclusive education under the UN Sustainable Development Goals [source: reference (3)].

challenges the moral justice of an exclusive focus on child survival since the era of the Millennium Development Goals (2000–2015) at the expense of a just and equitable society where no one is left behind.

The SDGs global vision to prepare every child, particularly those with developmental disabilities, to receive the best possible education to succeed in life beyond survival inspired the launch of this research topic by the Global Research on Developmental Disabilities Collaborators (GRDDC). GRDDC is a diversified, cross-cultural, and inclusive consortium of professional care providers and parents with and without lived experience of disability dedicated to advancing optimal development for children under 5 years with disabilities. A total of ten papers by 64 authors from all submissions were published drawn from sub-Saharan Africa, South Asia, Latin America, North America, and Europe.

Three papers in this series by Olusanya et al. set out to summarize the available data on children and adolescents with disabilities. The first GRDDC paper (Olusanya et al.) analyzed the latest prevalence estimates of children and adolescents with disabilities reported by UNICEF and the Global Burden of Disease (GBD), two leading publishers of population health metrics for policy makers in global health. The most striking and overarching finding was that the available prevalence estimates of disabilities among children and adolescents generated using either functional approach or statistical modelling can be statistically regarded as comparable and complementary. The choice between these sources is therefore, likely to be guided by the purpose for which the data is required. The second GRDDC paper (Olusanya et al.) addressed a critical gap in the literature on the global and regional estimates of children with cerebral palsy and developmental intellectual disability based on the first-ever WHO-GBD Rehabilitation Need Estimator database. The third GRDDC paper (Olusanya et al.) summarized eligible systematic reviews and meta-analyses of the prevalence of eight prominent developmental disabilities published since the launch of the SDG in 2015. This systematic umbrella review underscored the limitations of traditional systematic reviews and meta-analyses for estimating the global prevalence of developmental disabilities as most of the primary studies were conducted in high-income countries.

Hadders-Algra reviewed the scientific justification for promoting school readiness within the construct of human brain development that emphasizes the role of early detection and intervention for optimal growth and development in LMICs. Nair et al. examined the concept, key dimensions, and evaluation of school readiness for children with disabilities based on an extensive review of the literature and highlighted the critical role of partnership among childcare givers within the health and education sectors in addressing the major challenges in promoting school readiness in LMICs. The paper clarified that school readiness requires targeted interventions for child readiness, school readiness and family and community readiness to facilitate inclusive education. Smythe et al. highlighted the critical role of culturally sensitive parenting interventions and related priorities to support school readiness for children with developmental disabilities in LMICs. Akhbari Ziegler et al. summarized evidence from two pilot studies in Brazil on implementing COPCA (COPing and CAring for infants with special needs), a novel, family-centered early intervention program for infants at high biological risk of neurodevelopmental disability. It can be delivered remotely through tele-coaching, thus overcoming the challenge of access to a physical facility commonly faced by families in LMICs.

Nanyunja et al. demonstrated the feasibility of an affordable, community-based, group, participatory, peer-led program of early care and support for young children (0–3 years) with developmental disabilities and their caregivers through a randomized control trial, as part of a school readiness initiative in Uganda. Breinbauer et al. identified surmountable challenges in serving the needs of children with disabilities under a national early childhood development initiative in Chile, and reported how targeted financial incentives to the education sector has facilitated access to inclusive education for children under 5 years with developmental disabilities. Samia et al. identified obstacles and structural challenges in promoting inclusive education for children and adolescents living with disabilities in Africa.

The articles in this collection complement other publications by GRDDC addressing the need for a disability-focused early childhood development program for LMICs (7–9), visionary global leadership and accountability for early childhood development initiatives by UN agencies (10, 11), and a robust funding and investment mechanism to support school readiness for inclusive

education for children under 5 years with developmental disabilities in LMICs (3, 12). There is an on-going scoping review of the available guidelines by GRDDC to help care givers plan and deliver intervention services from birth against the backdrop of the different models, approaches and scope for early childhood development (13, 14).

In conclusion, the available evidence would suggest that the interest of children with developmental disabilities and their families will be better served by an independent global early childhood development initiative that seeks to optimize school readiness for inclusive education for children under 5 years in line with the commitment under the SDGs. This would require dedicated investment in family-centered early detection and intervention services and for supporting the transition of children with disabilities from home into pre-schools to enable stronger tripartite approach to "ready children", "ready families and communities", and "ready schools". As we mark the mid-point of the SDGs this year, all stakeholders in the disability community must unite and leverage the commitment under the SDGs to make the vision of a purposeful early childhood development by 2030 a reality.

Author contributions

All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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OPINION
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The Role of Parenting Interventions in Optimizing School Readiness for Children With Disabilities in Low and Middle Income Settings

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INTRODUCTION

Children with disabilities include those who have long-term physical, mental, intellectual or sensory impairments that in interaction with various barriers may hinder their full and effective participation in society on an equal basis with their non-disabled peers (1, 2). The protection of children with disabilities is enshrined in the UN Convention on the Rights of the Child (3), Convention on the Rights of Persons with Disabilities (UNCRPD) (2), and the Sustainable Development Goals (SDGs) (4). There is a global commitment to move toward Universal Health Coverage (UHC) as part of efforts to achieve Sustainable Development Goal 3 to "ensure healthy lives and promote wellbeing for all at all ages". The actions of nations to realize the rights of health for all will also impact the probability of achieving SDG target 4.2, which requires, "all girls and boys have access to quality early childhood development, care, and pre-primary education so that they are ready for primary education". The right to education applies to all children, and Article 24 of the UNCRPD (2) ensures that children with disabilities attend their local school and that schools accommodate their specific needs. In signing these pledges, governments have committed to addressing inequity for children with disabilities through the provision of disability-inclusive education and appropriate health services.

Fulfillment of these pledges is important, especially for the 53 million children <5 years of age with developmental disabilities, which include, but are not limited to, epilepsy, intellectual disability, sensory impairments, autism spectrum disorder or attention deficit hyperactivity disorder (5). They face greater challenges accessing quality healthcare services and experience worse health outcomes, especially in low- and middle-income countries (LMICs) (6). Furthermore, a focus on supporting children with disabilities to thrive and transform during their early years is important as this period is critical for maximizing their personal development and achieving their learning outcomes and readiness for school (7). Fulfillment of these pledges requires financial and strategic investment: It is equally essential that the global community recognizes practices

of exclusion. For example, in LMIC the likelihood of a child having an impairment before their fifth birthday was 10 times higher than the likelihood of dying in 2019 (8) and yet integration of inclusive health and education services for children with disabilities remains inequitably deficient (9, 10).

For the purpose of this paper, we use the term "early child development" (ECD) to include health, physical, social, emotional, cognitive and language development in the first 5 years of life (11). Within interventions to promote ECD, we use the term "parenting interventions" to encompass social and behavioral techniques or training that include any primary caregiver of a child with a disability. We refer to school readiness as a child's adequate preparation to engage in a school environment and activities, interconnected with school practices that foster a smooth transition to primary school and parental attitudes toward the school and support for early learning (12). In the context of school readiness, we define parenting interventions as skills training to assist parents in better supporting children with disabilities at home and preparing them for school. We use the term "inclusive education" to mean that different and diverse learners are welcomed and taught side by side with their peers and enjoy safety and participation with informed parental decision-making. It should be delivered in supportive environments in which "all members of the community are welcomed equally, with respect to (the different types of) diversity" (2).

Children with disabilities have limited access to ECD services in resource limited settings; moreover current ECD services are not designed to meet their specific learning, physical, and communication needs (13). It follows that ECD services experience challenges in enabling school readiness for children with disabilities. In this article we propose that culturally sensitive parenting interventions should enable and better inform parents, in partnership with ECD training programs at schools, to improve opportunities for children with disabilities to achieve their optimal potential and thrive in school. Ultimately, strengthening global early child initiatives and health and education systems toward achieving human rights and global development goals for all.

THE IMPORTANCE OF THE EARLY YEARS

Responsive parent-child relationships, and combined parental and community support for learning during the earliest years of life are recognized as being crucial for promoting successful ECD (14). By attending to the child's and the family's needs and strengths, the provision of appropriate and culturally relevant support and services can improve child and family outcomes. This requires the community, teachers and parents to work together with the child toward the successful and inclusive acquisition of individual and desired developmental and learning outcomes (12). These foundations for learning are largely built in the early years of life before a child participates in primary school education. Children who fall behind during these early years seldom catch up with their peers, perpetuating a cycle of underachievement and high dropout rates that continues to harm marginalized children and adolescents (15).

CURRENT STATE OF INCLUSIVE EDUCATION AND CHILD HEALTH

The interrelationship between disability, health and early childhood education is complex. These constructs are overlapping, intertwined and reinforcing, and place children with disabilities in vulnerable situations. In **Figure 1**, we propose a conceptual model based on the current evidence that demonstrates the reinforcing cycle of disability and poverty (16), which in turn limits access to education and health care (1, 17).

Limited access to quality education can have adverse effects on health (e.g., lack of access to school lunch), expose children to violence (e.g., increase of domestic violence during COVID pandemic) (18) and can result in greater adversity. Contextual factors create multiple barriers to successful inclusion as a result of social determinants of health, geographic location, cultural beliefs, discrimination and institutional sexism and racism (19). Thus, a child with disability is vulnerable to poor nutrition (20) and violence (21), at risk of poverty (16) and poor health and wellbeing (22), and has limited access to health care (17) and education (23). These factors are not only linked with disability, but are also interrelated as shown in the diagram.

The relationship between disability, education, health and social protection will not be the same for all; children with disabilities are a highly diverse and heterogeneous group. Nevertheless, they experience limited access to education, including ECD centers, as these centers are not necessarily able to accommodate the needs of children with disabilities through their curriculum programs (24). Disparities also exist between regions of the world in how supports are established for children with disabilities and their families. Health-care systems in high-income countries often support the early diagnosis and intervention with such children, including parenting interventions (25). In LMICs where the majority of children with disabilities live (5), there remains a lack of availability for continuous care and a dearth of information about ECD interventions, including parenting interventions. This paucity of knowledge is perpetuated through research agendas. For example, a recent global systematic review (26) excluded parenting interventions for children with disabilities, and a call for action found that 50% of the registered clinical trials reviewed explicitly excluded young children with disabilities (27, 28). Research agendas are also influenced by global funding for grand challenges and policy makers and there remains a gap in government funding and development assistance for children with disabilities and their families in LMIC, relative to population needs and epidemiology (29, 30).

Optimal support for early child development has lifetime beneficial consequences for educational achievement, adult productivity and population health (27). Despite the wellrecognized importance of investment in children's formative years, only half of the world's preschool-aged children attend pre-primary programs (from age 3 years up to the start of primary

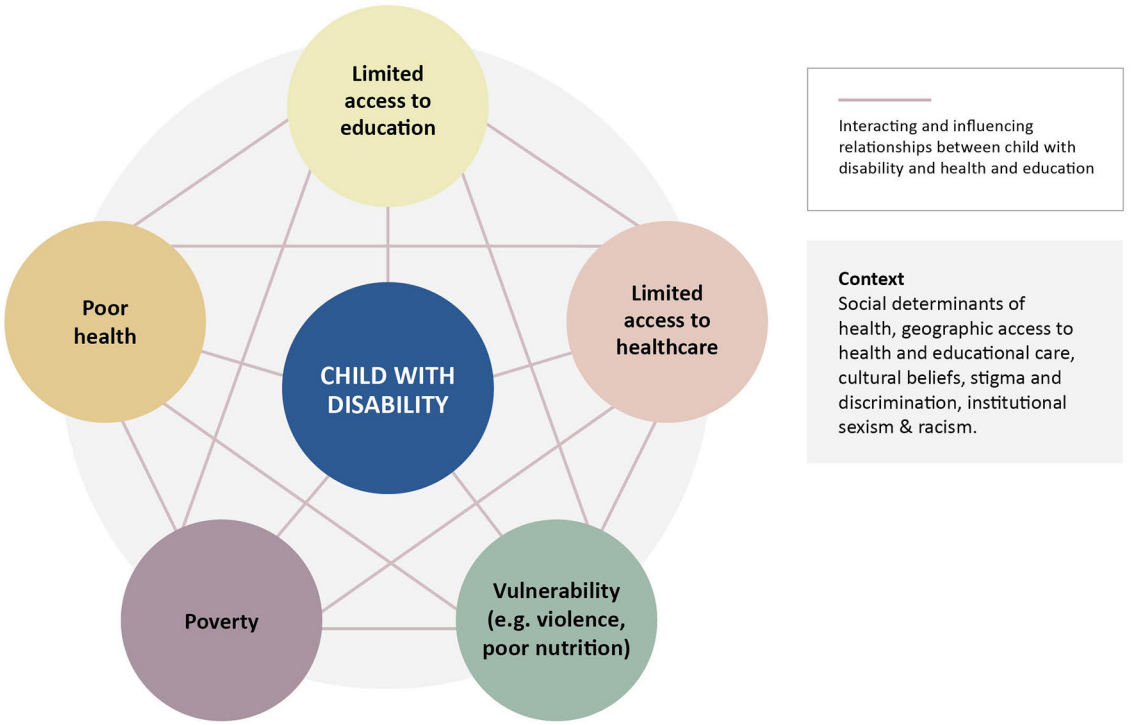

FIGURE 1 | Interrelationship between disability, poor health and limited access to education.

education, often aged 6 years) (31). Whilst formal education usually starts between 5 and 6 years in many LMICs, only 2 in 10 children have the privilege of attending pre-primary programs and barriers are even higher for marginalized groups such as children with disabilities (31). These barriers include a limited accommodation process, physical inaccessibility, negative attitudes, stigma and discrimination, local cultural beliefs, expectations about diverse functionalities and curricula that do not address the needs of children with disabilities (32, 33). It is timely that UNICEF's draft strategic plan (2022–2025) includes a focus on supporting caregivers, communities and schools to provide the environments, care, protection and education that enable children's health, nutrition and development (34). The smooth transition for children to primary education often referred to as "school readiness", requires the school, families/communities and children to work together well before enrolment into school to facilitate communication abilities along with peer social behaviors. The young child's readiness for school focuses on learning and developmental outcomes; the school's readiness for the child usually focuses on school-level outcomes and practices that foster a smooth transition into primary school; and families' readiness for school usually incorporates parent attitudes to the school itself and involvement in the child's early learning as well as development and transition to school (35). Ready schools, parents, children and communities need to work together to achieve this smooth transition. Still, the link between ECD initiatives and school readiness remains contentious as there is a tendency to focus on pre-academic skills (early literacy and numeracy), even at home. Children with disabilities may not be able to achieve pre-academic skills in time or at all, but are more focused on daily living activities (e.g., be toilet trained and able to feed themselves). There remain inequities in the delivery of services for some children with disabilities, and initiatives approach groups of children differently, depending on the functionality and severity of their impairment. (E.g., a child with multiple physical and cognitive disabilities may not be able to participate in many learning and play activities without adequate support.).

Children with disabilities in LMICs can be excluded from school due to parent and caregiver fear of not being able to provide adequate care, teachers being overwhelmed with the presence of children with disabilities and lack of training on inclusion, or classes being simply too large to pay proper attention to the child's needs (24). Families of children with disabilities are often unable to visit the school before the start of the academic year, thus creating significant difficulties for a smooth transition into school for the child. Children with disabilities are further marginalized by the design and structure of school curricula, and by perceptions of their limited abilities (32). Finding ways to meet the individual learning, social, and physical needs of students with disabilities can be challenging in schools and contexts with severely limited resources (32). For example, a child with cerebral palsy may require a modified chair with a flat top with an edge on which to place and move objects. There should be opportunities for ready schools, parents, children and communities to develop the necessary relationships

and information sharing that will support a smoother transition for children with disabilities, from the early child-center into primary school, where all are then ready to engage together (12).

THE ROLE OF PARENTING INTERVENTIONS IN OPTIMIZING SCHOOL READINESS

There are broadly two approaches to providing ECD for children with disabilities, including them in mainstream ECD interventions, and targeting interventions according to their individual needs. There remains a need for inclusive approaches for children with disabilities in mainstream services, as well as within specialist ECD interventions. This means that the role of parents can be particularly crucial to fill existing gaps in service availability. A key factor in optimizing a child's school readiness, is fostering the capabilities of parents and communities to help to scaffold development and early learning. Many interventions that are designed to boost school readiness target parents and parenting skills. Interventions include approaches that increase parent wellbeing, knowledge and confidence, as well as enhance parent sensitivity and responsiveness, enrich parent-child communication and increase parent support for early learning through skills training programs. For example, parenting interventions for children during the first 3 years of life lead to improvements in early cognitive, language, motor, socioemotional development, and reduce child behavior problems across LMICs and high-income countries (26). Parenting interventions also improve parenting knowledge and practices, and parent–child interactions (26, 36, 37).

Accordingly, parenting interventions that include skills training to assist parents in being better able to care for a child with a disability at home and prepare them for school may contribute to optimizing readiness for school. Children with disabilities would benefit from these parenting interventions to improve their child's chances of being school ready and address the challenges that they may face, which are often compounded by community attitudes and beliefs espoused in relation to disability (33). Notably, combining the education of parents and the training of teachers has a greater impact on child outcomes, such as increased language skills (38), and may also benefit social development, improved play and motor skills (39). There is, therefore, a need to deliver parallel training at the school level on the development of pedagogy, teacher skills and positive attitudes to inclusion. Nevertheless, the current policy landscape for ECD has not yet resulted in greater investments and implementation of large-scale national parenting programs and critiques of parenting interventions in LMICs raise ethical challenges and concerns (40, 41).

CRITIQUE OF PARENTING INTERVENTIONS IN LOW RESOURCED SETTINGS

Parenting intervention practices are commonly derived from attachment theory and responsive care, in which the quality of attachment stems from the way that a mother cares for her child, and are presented as the universal standard of good care (40). These practices are typically Euro-American constructs and include little attention to community practices. Such parenting interventions involve encouraging caregivers to change their practices and views, usually with little understanding of how such changes affect child, family, and community (40). Typically, in contexts of limited support and scarce resources, there may be a combined collective input of all caregivers, rather than only the mother or grandmother who assume responsibility for the child's development and welfare. We need to carefully consider existing beliefs, practices, stigma and developmental goals in the targeted communities to ensure that ethical principles are fulfilled (41) all the while preserving the efficacy of parental support for their children within the communities they live in.

There is a clear narrative for inclusive child health, education, and protection (safeguarding) in LMICs; children with disabilities are excluded from recommendations, initiatives and policies. The lack of investment in inclusive education is reflected in low service level inputs and consequently education coverage and outcomes for children with disabilities. Investment in education and scale-up of services for children with disabilities is needed urgently. For instance, the percentage of Gross Domestic Product spent on education in 2019 is 5.3 in North America, but only 3.5 in sub Saharan Africa and 2.5 in South Asia (42). UNESCO's recommendation for government budget for education by 2030 is 4–6% of GDP and/or the allocation of at least 15–20% of public expenditure (43). It is estimated that an additional 50 cents per person annually is the cost for ECD to be incorporated into existing services (44). This level of funding for education, if followed through in LMIC, will translate to significant improvement in public investment in education with measurable outcomes. Partnership between policymakers and educators should support governmental investment in ECD and implementing effective and culturally appropriate parenting interventions.

MULTIDIMENSIONAL ROADMAP FOR INCLUSIVE EDUCATION: PRIORITIES FOR PARENTING INTERVENTIONS AND SCHOOL READINESS

Children with disabilities should be able and have a right, to experience positive wellbeing and full involvement at school rather than merely attending education services. Parents of children with disabilities experience emotional distress, isolation and lack of support, particularly in cultures where unfavorable superstitious beliefs about disability prevail (45), which may trigger profound disappointment, prolonged grief and a sense of hopelessness for a seemingly uncertain future for their children (46). Parenting interventions, for school readiness designed for parents to be able to better care for a child with a disability at home and prepare them for school, may not only instill hope of a better independent and productive living but are also reassuring to the entire family. The role of culturally sensitive parenting interventions in tandem with ready schools

and inclusive communities that are supported by the policy is critical for achieving inclusive and quality education. The intervention needs to be family-centered and ensure that families are more confident to discharge their role effectively. Parenting interventions for parents of children with disabilities should recognize the inherent value of the experience that the parent possesses. They need to be respectful of how children are brought up and educated in the local culture, and build on the local practices, knowledge and strengths that exist in early child education whilst collaborating with local training providers, community and ECD services (24). Active engagement between the health, social welfare and education sectors at all levels is required. Within schools, additional support needs to be provided to support children with disabilities, through assistance to help with toileting, feeding, mobility, communication and safe play. We need to scaffold child development and learning at their level and enrich current parent intervention approaches to work together with training of teachers. This may go some way to preventing the growing gap in provision of early learning and contribute toward achieving the full intent of SDG 4.2.

CALL TO ACTION

As ECD specialists, health professionals and researchers committed to equity and social justice, our task is to reveal patterns of avoidable differences experienced by children with disabilities in accessing inclusive and equitable quality education. We strongly recommend the implementation of system-wide strategies to address the prevailing inequities and barriers, such as the lack of education resources for parents, the lack of training and appropriate resources for preschool professional staff and more importantly the implementation of inclusive education policies by the education sector, which continue to shift the onus to children with disabilities and their families. Close examination of the readiness and capacity of a nation's schools to receive all young children and support their learning and development is needed. Inclusion demands that educators and policymakers consider two key questions when reviewing policies and practices: (1) Who benefits? (47) and (2) Inclusion into what? (48). We call for accelerated political will and action to adapt and deliver parenting interventions for children with disabilities, which are respectful of diverse local contexts, whilst coordinating within existing systems and services. The current evidence suggests that parenting interventions are effective for ECD however, most studies are conducted in high-income settings, which raises questions about generalizability; complementary investment in addressing the needs of the beneficiaries of child survival programs with lifelong impairments in LMICs is required.

CONCLUSION

Children with disabilities face sustained inequities despite the international agenda that supports inclusion. This is perpetuated through exclusive early child initiatives and policies, practice, and research. The global agenda urgently needs to move beyond token recognition of this marginalized group to inclusive early child intervention programs that consider existing practices, cultural beliefs, and developmental goals in the targeted communities. Children with disabilities in LMICs should receive culturally sensitive parenting interventions to improve learning and educational outcomes. These initiatives must be geared toward "school readiness" for educational inclusion of children with disabilities and this necessitates that the community, teachers, and parents work together with children toward successful developmental and learning outcomes. Culturally sensitive parenting interventions, and early child development teaching programs at schools, may thus contribute to strengthening education systems toward achieving the full intent of SDG 4.2 and human rights global development goals for all.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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Cerebral palsy and developmental intellectual disability in children younger than 5 years: Findings from the GBD-WHO Rehabilitation Database 2019

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Objective: Children with developmental disabilities are associated with a high risk of poor school enrollment and educational attainment without timely and appropriate support. Epidemiological data on cerebral palsy and associated comorbidities required for policy intervention in global health are lacking. This paper set out to report the best available evidence on the global and regional prevalence of cerebral palsy (CP) and developmental intellectual disability and the associated "years lived with disability" (YLDs) among children under 5 years of age in 2019.

Methods: We analyzed the collaborative 2019 Rehabilitation Database of the Global Burden of Disease (GBD) Study and World Health Organization for neurological and mental disorders available for 204 countries and territories. Point prevalence and YLDs with 95% uncertainty intervals (UI) are presented.

Results: Globally, 8.1 million (7.1–9.2) or 1.2% of children under 5 years are estimated to have CP with 16.1 million (11.5–21.0) or 2.4% having intellectual disability. Over 98% resided in low-income and middle-income countries (LMICs). CP and intellectual disability accounted for 6.5% and 4.5% of the aggregate YLDs from all causes of adverse health outcomes respectively. African Region recorded the highest prevalence of CP (1.6%) while South-East Asia Region had the highest prevalence of intellectual disability. The top 10 countries accounted for 57.2% of the global prevalence of CP and 62.0% of the global prevalence of intellectual disability.

Conclusion: Based on this Database, CP and intellectual disability are highly prevalent and associated with substantial YLDs among children under 5 years worldwide. Universal early detection and support services are warranted, particularly in LMICs to optimize school readiness for these children toward inclusive education as envisioned by the United Nations' Sustainable Development Goals.

KEYWORDS

cerebral palsy, intellectual disability, rehabilitation, global health, developmental disabilities, global burden of disease, early intervention, SDGs

Introduction

Children under 5 years are widely acknowledged as an important cohort for evaluating the overall health and well-being of any population (1). For several years policymakers have used under-5 mortality as a key indicator of progress in global health and have made targeted reductions in under-5 mortality a central policy objective for global investment in child health (2). The science of human brain development has shown that investments in early childhood, particularly from birth to five years, are the foundation for a prosperous and sustainable society (3, 4). In 2015, the United Nations' Sustainable Development Goals (SDGs) mandated the monitoring of all children under 5 years of age at risk of not realizing their developmental potential to ensure that these children are offered the requisite support services that adequately prepare them for school enrolment (5). However, children with disabilities have a greater risk of poor or sub-optimal development in early childhood compared to children without disabilities (6, 7).

In 2018, the Global Burden of Diseases, Injuries, and Risk Factors Study (GBD) estimated that over 53 million children under 5 years have epilepsy, intellectual disability, hearing loss, vision loss, autism spectrum disorder, or attention deficit hyperactivity disorder (8). Approximately 95% of these children lived in low-income and middle-income countries (LMICs). Although cerebral palsy (CP) is frequently reported as the most common physical disability originating from early childhood (9–11), its exclusion in the GBD 2018 paper was duly acknowledged as a significant limitation (8, 12). To address this omission and provide some indication of the requisite rehabilitation needs, the most recent GBD database produced in collaboration with the World Health Organization (WHO) – the GBD-WHO Rehabilitation Database (labeled as "WHO Rehabilitation Need Estimator") - now

includes data on CP over the life-course (13). This paper sets out to analyze the available global and regional estimates for the prevalence of CP and the associated "years lived with disability" (YLDs). Since developmental intellectual disability (or simply "intellectual disability" hereinafter) is more frequently associated with CP than any other long-term childhood disorder, we also included this condition in this study. The findings will complement our prior reports on the global and regional pattern of developmental disabilities among children under 5 years (8, 14, 15), as well as the recent GBD-related reports on mental and neurological disorders (13, 16).

Methods

A comprehensive description of the methodology for the GBD-WHO Rehabilitation Database including the underlying modeling strategies has been previously reported (13). As with all GBD papers, the substantive data that formed the basis of this analysis adhered to the Guidelines for Accurate and Transparent Health Estimates Reporting (GATHER), which include recommendations on documentation of data sources, estimation methods, statistical analysis, and statistical code (17). In summary, the point prevalence and YLDs are estimated for 25 health conditions selected by a WHO Expert Panel on Rehabilitation (13). The health conditions are grouped into seven GBD aggregate disease and injury categories: musculoskeletal disorders, neurological disorders, sensory impairments, mental disorders, chronic respiratory diseases, cardiovascular diseases, and neoplasms. CP and intellectual disability are included among the neurological disorders and mental disorders categories, respectively. The estimates for each condition are made for 204 countries and territories categorized into the six WHO regions of Africa, Eastern Mediterranean, European, South-East Asia, The Americas, and Western Pacific (see Appendix 1 in Supplementary material). The high-income countries (HICs) from each region were extracted and grouped into a separate category, based on the World Bank criteria.

Cerebral palsy is a group of neurological disorders that appears in infancy or early childhood and permanently affect body movement and muscle coordination (9, 10). The prevalence of CP was determined indirectly by aggregating all sequelae of neonatal disorders and infectious diseases including preterm birth/low birth weight, neonatal encephalopathy due to birth asphyxia and trauma, neonatal sepsis, and other neonatal infections as well as hemolytic disease and other neonatal jaundice with mention of moderate to severe motor impairment (13, 18). These underlying causes of CP were identified from a systematic review of relevant literature using the International Classification of Diseases, Tenth Revision (ICD-10) codes. Children with mild motor impairment, typically those with ambulation who can walk without help, were excluded in the database on the assumption that they were less likely to require rehabilitation (13).

Intellectual disability is typically defined as a condition of below-average intelligence or mental ability originating before the age of 18 years in line with the Diagnostic and Statistical Manual of Mental Disorders by the American Psychiatric Association (19). The prevalence of intellectual disability (IQ score <70) came from a systematic review of publications since 1990 and included studies that estimated the general population prevalence of intellectual disability (13). Intellectual disability was modeled as an impairment and grouped into five bands based on Intelligence Quotient (IQ) scores, ranging from borderline (70–85), mild (50–69), moderate (35–49), severe (20–34), to profound intellectual disability (0–19). In the GBD Study, an impairment is defined as the sequelae of multiple causes for which better data were available to estimate the overall occurrence than for each underlying cause. Borderline intellectual disability was assumed to be less likely to require rehabilitation and was excluded in computing the prevalence of intellectual disability in the database. A child having both CP and intellectual disability was counted separately for each condition.

Years lived with disability are defined as the years of life lived with any short-term or long-term health loss. YLDs are designed to provide a comparable measure of disease burden across diverse health conditions and impairments rather than a measure of functional status as described in the International Classification of Functioning, Disability and Health (ICF) (20). To calculate YLDs for CP and intellectual disability, the estimated prevalence of each condition at the national, regional, and global level was multiplied by an assigned disability weight based on the severity of the disability. Disability weights are population assessments of the magnitude of health loss associated with specific health outcomes, measured on a scale from 0 to 1, where "0" equals a state of "perfect health" and "1" equals death. For example, the assigned weights for CP vary from 0.01 for mild motor impairment, 0.061 for moderate motor impairment and 0.402 for severe motor impairment based on the degree of ambulation. The disability weights for intellectual disability vary from 0.011 for borderline intellectual functioning to 0.2 for profound intellectual disability based on the degree of difficulty in learning to speak, do simple tasks or follow basic instructions (13). The disability weights were estimated from multi-country population-based surveys using pairwise comparison methods between random pairs of health states as described in detail elsewhere (21).

In general, where there are no primary data, estimates rely on predictive covariates and geographical proximity to countries with data. All computations in the GBD Study were conducted 1,000 times to propagate uncertainty around the estimates for prevalence and YLDs. At every step in the modeling process, the distributions were assessed for sampling error of data inputs, the uncertainty of data corrections for measurement errors, the uncertainty in coefficients from model fit, and the uncertainty

of severity distributions and disability weights. Corresponding uncertainty bounds intervals (UIs) for prevalence and YLDs estimates were defined at the 25th and 975th value of 1,000 draws. In this paper, the term "children" refers to children under 5 years of age unless otherwise stated. As this paper is derived from a publicly available database, no ethical approval was required. Estimates are reported along with the 95% uncertainty intervals (UI) in brackets, except stated otherwise.

Results

Globally, of the 662.8 million children younger than 5 years in 2019, 8.1 million (7.1–9.2) or 1.2 % (1.1–1.4) were estimated to have CP and 16.1 million (11.5–21.0) or 2.4% (1.7–3.2%) had intellectual disability (Table 1). About 53% of children with CP and 54% of children with intellectual disability were male. Most children with CP and intellectual disability resided in LMICs. The estimates for HICs were 359,045 children (326,154–397,121) or 0.6% (0.5–0.6) with CP and 886,977 children (727,734–1,088,596) or 1.4% (1.2–1.8) with intellectual disability. Of the total 27.1 million (19.3–36.1) YLDs among children under 5 years from all causes of fatal and non-fatal health outcomes in 2019, CP accounted for 6.5% or 1.8 million (1.2–2.4) YLDs and intellectual disability accounted for 4.5% or 1.2 million (0.8–1.8) YLDs.

Figure 1 shows that the African Region had the highest prevalence of children with CP of 1.6% (1.4–1.8) or 2.7 million (2.4–3.0) and the highest YLDs of 586,762 (408,151–793,947). South-East Asia Region had the highest prevalence of children with intellectual disability of 3.8% (2.4–5.2) or 6.3 million (4.0–8.6) with an associated YLDs 449,331 (270,088–669,598). The geographical distribution of the prevalence of CP and intellectual disability at country level is presented in Figure 2. India recorded the highest population of children with CP and intellectual disability and the associated YLDs (Table 2). The prevalence of CP and the associated YLDs was highest in Bangladesh, while the prevalence of intellectual disability and the associated YLDs was highest in India. The top 10 countries accounted for 57.2% or 4.6 million of all children with CP and 62.0% or ∼10 million of children with intellectual disability globally. These countries also accounted for 57.1% of the global YLDs for CP and 60.4% of the YLDs for intellectual disability. Except for the USA which ranked 10th with the number of children with intellectual disability, the top 10 countries were predominantly LMICs. Among children with CP, 4 (40%) of the top 10 countries with the highest population and 8 (80%) of the top 10 countries with the highest prevalence were from Africa.

Discussion

The dearth of population-based data for specific health conditions from birth across many nations, especially in LMICs, has resulted in a growing reliance on statistical estimation of health outcomes as a surrogate for guiding global health policies and interventions (22). Conceptual and operational challenges in measuring disabilities among children in different cultural contexts at the population-level persist (23, 24). The GBD modeling efforts thus offer an invaluable undertaking in the epidemiology of developmental disorders for global policy intervention. Unlike prior reports, the GBD-WHO collaboration provides an additional layer of quality control for the conventional GBD database through subject expert consultations. Arguably, the estimates reported in this study represent the best available global estimates of children under 5 years with CP and intellectual disability. The findings clearly establish that these conditions are highly prevalent worldwide with LMICs accounting for the greatest burden (i.e., prevalence and YLDs). They also underscore the necessity for primary prevention initiatives and provide independent estimates of the magnitude of the rehabilitation needs for these conditions within the integrated health care systems envisaged by WHO (13).

The global estimate of 1.2% or 12 per 1,000 children under 5 years for CP in this study represents 16.2% of the estimated 50 million of all children and adults with CP (13). This estimate is higher than those in several epidemiological studies which report a global prevalence of between 1 and 4 per 1,000 live births or 1,000 children of all ages (9, 10, 25). However, the global estimates in the literature are almost entirely derived from studies conducted in high-income countries. The GBD-WHO estimate of 0.6% or 5.8 per 1,000 children under 5 years for all high-income countries is higher than the reported estimates for this age group (26, 27). For example, the prevalence of CP among children aged 3–5 years in USA using parental household surveys is estimated at 0.3% or 2.8 per 1,000 (26). However, this estimate is likely to be higher if the younger children below the age of 3 years were included in the estimate. The disproportionately higher prevalence of adverse perinatal and neonatal conditions in LMICs, particularly in sub-Saharan Africa and South Asia, would also suggest higher estimates than reported in the few available population-based studies (9, 28, 29). A detailed comparative analysis with estimates from systematic reviews is difficult principally due to marked variations in methodology, mean age at diagnosis, the age-group of participants, choice of denominator (birth vs. period prevalence) in the underlying studies and the dearth of studies from many regions especially in LMICs (29, 30). In addition, the age range of the children reported across studies varied considerably and the specific prevalence of these conditions among all children under 5 years is seldom reported. In LMICs, children younger than 2 years are commonly excluded because of the view that these conditions are too difficult to detect at this age especially in routine population-based household surveys. Reports also suggest that less than half of children are clinically diagnosed by 24 months of age compared to the

TABLE 1 Global and regional prevalence of cerebral palsy and developmental intellectual disability and the YLDs among children under 5 years in 2019.

Location	Cerebral palsy				Developmental intellectual disability			
	Prevalence	95% uncertainty interval	YLDs	95% uncertainty interval	Prevalence	95% uncertainty interval	YLDs	95% uncertainty interval
African region								
Number	2,684,002.9	2,385,081.1–3,033,950.4	586,762.1	408,150.9–793,946.5	3,310,525.9	2,412,002.9–4,238,791.7	261,499.3	169,643.0–375,684.7
Cases per 100,000	1,616.1	1,436.1–1,826.8	353.3	245.8–478.1	1,993.4	1,452.3–2,552.3	157.5	102.1–226.2
Region of The Americas								
Number	706,407.0	626,193.3–806,022.6	154,178.1	106,230.9–209,656.8	1,189,036.8	978,224.6–1,424,834.2	106,262.7	71,021.0–150,154.6
Cases per 100,000	959.4	850.4–1,094.6	209.4	144.3–284.7	1,614.8	1,328.5–1,935.0	144.3	96.5– 203.9
East Mediterranean Region								
Number	1,053,861.4	933,962.6–1,202,317.7	229,339.6	157,962.2–315,534.9	2,667,911.1	1,779,861.4–3,585,661.5	186,491.8	115,767.2–275,918.6
Cases per 100,000	1,250.5	1,108.2–1,426.6	272.1	187.4–374.4	3,165.6	2,111.9–4,254.6	221.3	137.4–327.4
European Region								
Number	435,109.9	396,450.9–480,023.9	95,126.0	65,963.2–128,310.4	926,164.5	686,743.9–1,178,047.5	70,942.1	46,324.9–101,709.5
Cases per 100,000	805.2	733.6–888.3	176.0	122.1–237.4	1,713.9	1,270.8–2,180.0	131.3	85.7–188.2
South-East Asia Region								
Number	2,357,679.0	2,003,712.7–2,791,591.5	510,862.1	343,125.4–707,366.0	6,317,447.9	4,041,745.9–8,605,559.9	449,331.1	270,088.3–669,597.5
Cases per 100,000	1,427.5	1,213.2–1,690.3	309.3	207.8–428.3	3,825.1	2,447.2–5,210.6	272.1	163.5–405.4
Western Pacific Region								
Number	927,377.8	772,509.7–1,134,088.2	201,611.8	135,266.0–281,416.5	1,766,440.1	1,416,940.5–2,167,897.7	153,070.6	100,938.6–217,222.6
Cases per 100,000	784.8	653.8–959.8	170.6	114.5–238.2	1,494.9	1,199.2–1,834.7	129.5	85.4– 183.8
World Bank High-Income								
Number	359,045.2	326,153.6–397,121.3	78,757.3	54,258.9–105,746.3	886,977.2	727,733.8–1,088,595.9	78,097.0	52,323.3–109,403.1
Cases per 100,000	580.3	527.1–641.8	127.3	87.7–170.9	1,433.6	1,176.2–1,759.4	126.2	84.6– 176.8
Global								
Number	8,071,408.0	7,113,334.0–9,231,577.0	1,757,372.1	1,209,309.2–2,404,752.9	16,057,583.8	11,515,194.1–20,980,652.2	1,222,295.1	782,852.1–1,774,628.7
Cases per 100,000	1,217.7	1,073.2–1,392.7	265.1	182.4–362.8	2,422.5	1,737.2–3,165.3	184.4	118.1–267.7

YLDs, years lived with disability.

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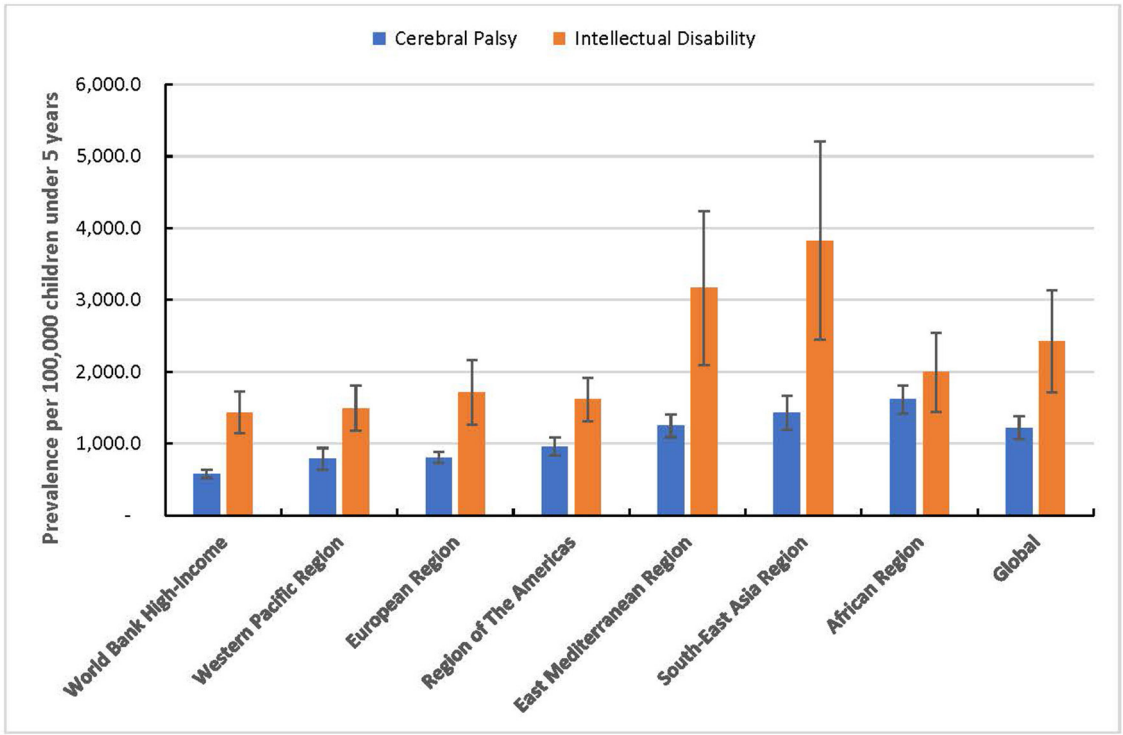

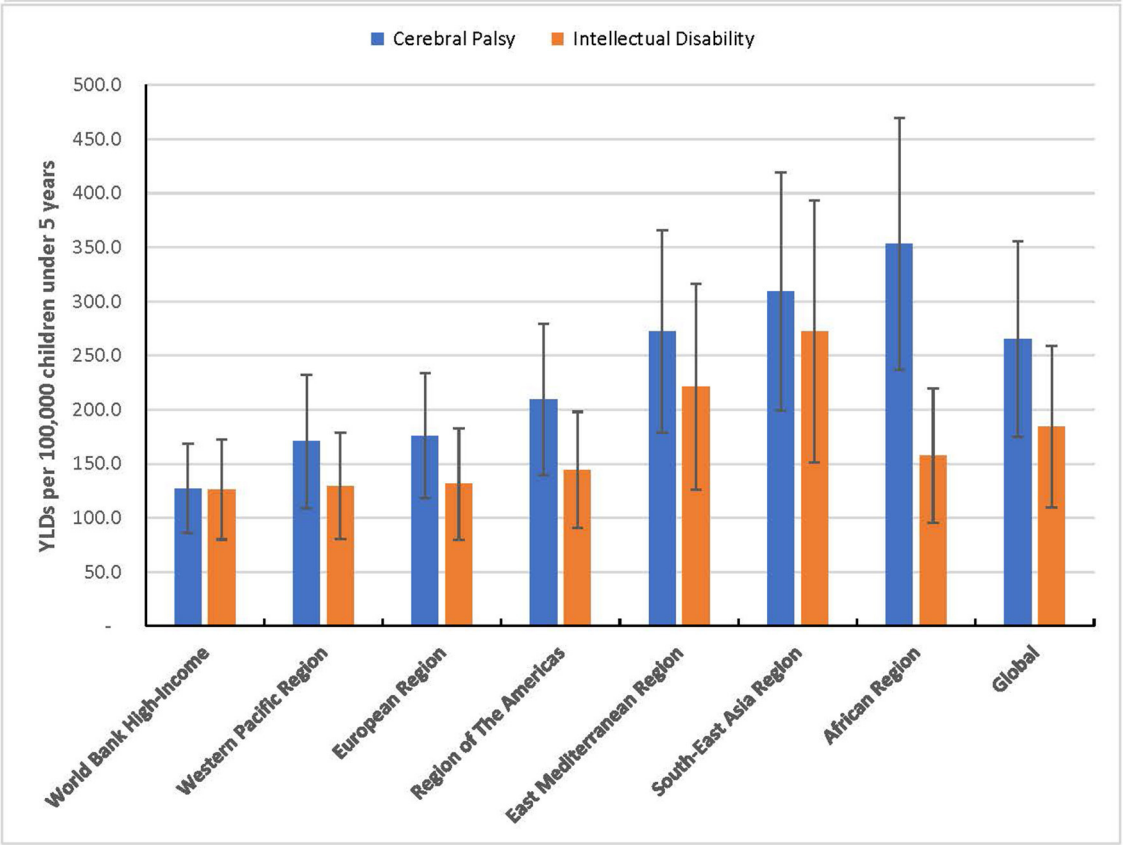

FIGURE 1
Global and regional prevalence of cerebral palsy and developmental intellectual disability and the YLDs among children under 5 years in 2019.

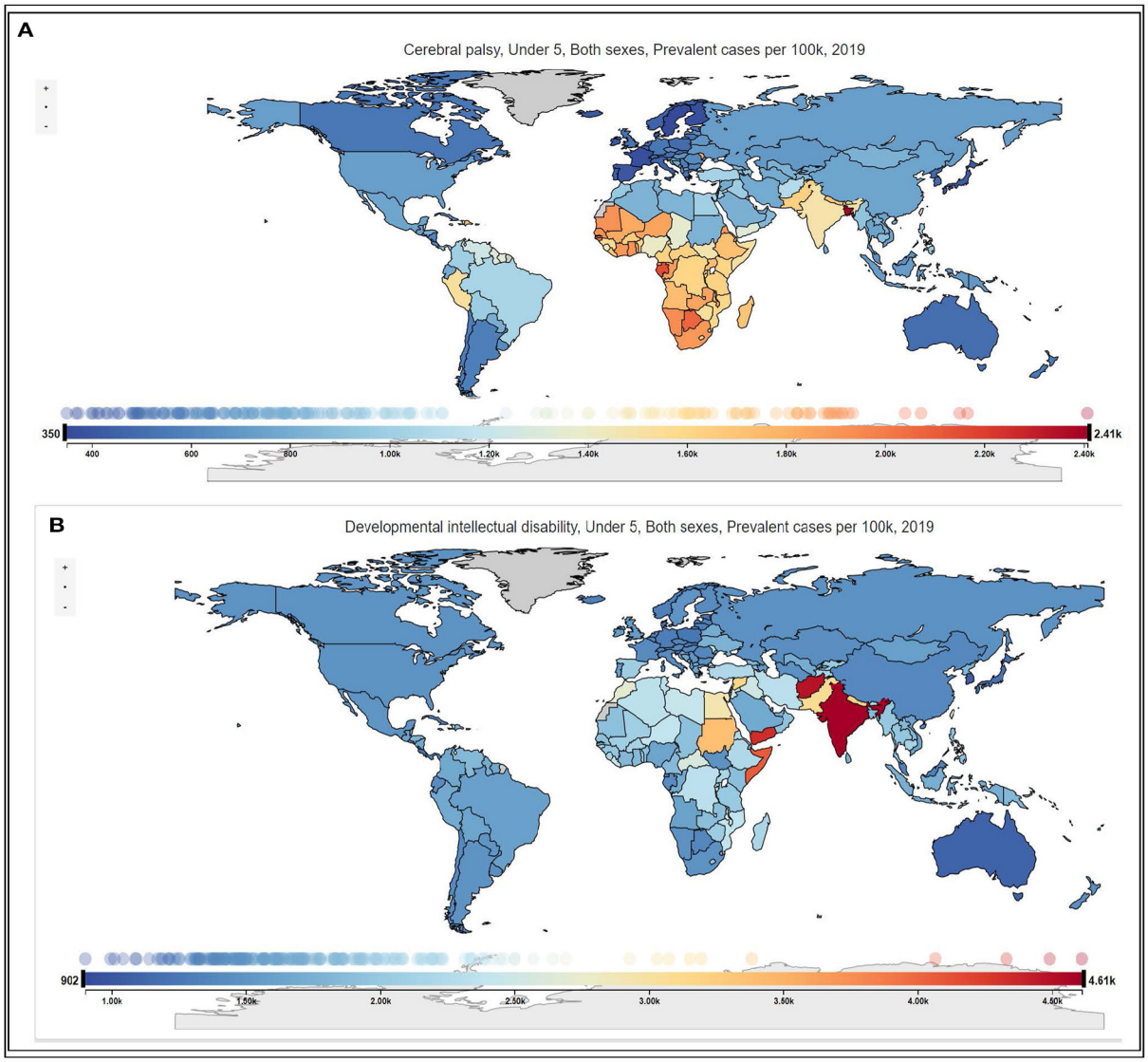

FIGURE 2
(A,B) Global prevalence per 100,000 population of cerebral palsy and developmental intellectual disability among children under 5 years in 2019.

practice in HICs (9). Moreover, many children with CP die before their second birthday, thus remain undiagnosed and many may not be counted at all (31). There is a growing international recognition of the technological advances to make early detection and intervention for CP before the age of 2 years feasible even in countries where clinical diagnosis may typically be delayed until age 4 years (11, 32). Considering the gross under-ascertainment of cases in LMICs, we hold the view that the true global prevalence of CP among children under 5 years possibly lie between the GBD-WHO estimate and reported estimates in the literature from household surveys.

The global estimates for intellectual disability appear to agree with those reported by Maulik et al. in which the global prevalence among children and adolescents was shown as 1.8% (95% CI: 1.5–2.1) (33), considering that the prevalence of intellectual disability is highest in early childhood and declines thereafter among older children. The reported prevalence for HICs also appears plausible compared to the ∼1.6% for children and adolescents in the USA (34). The significantly higher global prevalence of intellectual disability compared to CP in our study is consistent with evidence in the literature principally due to the wider range of comorbidities associated with the former (33–35). Moreover, 1 in 2 children with CP are frequently diagnosed with comorbid intellectual disability (11). The higher estimates of these conditions in LMICs compared with HICs is also in line with previous findings in the literature (9, 10,

33, 36–38), as well as studies among children from low-income households in HICs (35). The substantial YLDs associated with CP and intellectual disability further underscore the need for global initiatives to address these conditions promptly and appropriately when intervention outcomes can be optimized to enhance the opportunities for inclusive formal education as envisaged by the SDGs (5). The higher YLDs associated with CP compared to intellectual disability perhaps reflect the magnitude and scope of the rehabilitation and other support services required by the affected children in early childhood (11, 32).

The disproportionately high prevalence of CP in Africa may be attributable to a constellation of factors which includes the low quality of maternal and child health services and the high proportion of deliveries not attended by skilled health workers. Clinical factors such as birth asphyxia, kernicterus, and neonatal infections have also been implicated as contributors to the high prevalence (36), in contrast to the more common causes like prematurity and low birth weight in HICs (25). Africa's leading contribution to the global burden of CP accords with our earlier report that demonstrated the significantly higher and rising burden of developmental disabilities in Africa among the growing beneficiaries of the highly successful global investments in reducing under-5 mortality since 1990 (14). The leading contribution of Southeast Asia to the global prevalence of intellectual disability appears to be supported by perhaps the most robust nationally representative study on developmental disabilities in India in which the prevalence of CP and intellectual disability in children aged 2 to <6 years was reported as 2.1% (95% CI: 1.3–3.4) and 3.1% (95% CI: 2.2–4.2), respectively (37). The leading risk factors reported for these conditions and other developmental disabilities in India were non-institutionalized delivery, history of perinatal asphyxia, history of neonatal illness, and postnatal neurological (brain) infections. However, the causal factors in about half of the children with intellectual disability are likely to be unknown (33).

It is noteworthy that the countries with the highest population of children with these conditions were not necessarily those with the highest prevalence. Furthermore, the burden of these conditions is highest in the regions of the world that are poorly resourced to provide the requisite support services for these children and their families (9, 38, 39). Consequently, children with these conditions in LMICs are more likely to experience a lower quality of life compared to their peers in HICs (40). These children are also at greater risk of premature mortality (31). While primary prevention should be prioritized, the substantial unmet rehabilitation needs of these children and others with developmental disabilities in LMICs as highlighted by a recent WHO report require urgent and priority attention for these children and their families (39). Whereas the period of interest for early detection and intervention services varies from conception to age 8 years in the literature, it is pertinent to clarify that the focus on the first 5 years of life from birth in this paper is consistent with the most widely recommended clinical framework for the effective management of children with developmental disabilities for school readiness (41). The evidence in this report reinforces our earlier call for a decisive, appropriate, timely and well-coordinated policy intervention to support these children and others with developmental disabilities to place them on the trajectory for school readiness for inclusive education as envisaged under the Sustainable Development Goals (5, 42).

Limitations

Modeling techniques and the use of proxy measures to generate evidence are now common in highlighting the public and global health importance of health conditions that are currently constrained by the lack of adequate and reliable population-based data. This approach is premised on the principle that the absence of ideal data is not evidence of absence of a health condition that truly warrants policy intervention. However, this approach is not without shortcomings. The limitations frequently associated with GBD methodologies have been extensively reported in accordance with the GATHER guidelines (12, 13, 15, 16, 18). Additionally, despite the continuous efforts toward improving the GBD methodology, the current practice of estimating the prevalence of disabilities based on sequelae of underlying health conditions or surrogates is not without drawbacks. For example, on the one hand, the exclusive use of motor deficits for CP is likely to have resulted in over-estimation of its prevalence because not all motor impairments constitute CP. On the other hand, this approach may have under-estimated children with CP who have a wide range of levels of functioning and those with milder difficulties with functioning without moderate or severe motor manifestations (43). It is reported that GBD plans to estimate the prevalence of CP based on a meta-analysis of available data from registries and cohort studies which would provide more insights on any variance attributable to the GBD methodology and the required adjustments in model parameters (13). The GBD estimates for disabilities still do not fully reflect the complex and dynamic relationship between health conditions and contextual personal or environmental factors as envisaged under the ICF, as such they provide a limited picture of disability. In fact, the threshold for rehabilitation which excluded children with mild motor impairments and borderline intellectual disability as well as the sole use of IQ tests may inadvertently exclude children with functional limitations that require intervention. Another limitation is the wide uncertainty around the estimates for YLDs due to the determination of disability weights (8, 13, 15, 18, 21). Disability weights in GBD Study reflect the severity of a disease and are needed to quantify health losses relating to non-fatal outcomes. However, cultural,

TABLE 2 Ten leading countries based on the prevalence of cerebral palsy and developmental intellectual disability and the YLDs among children under 5 years in 2019.

		Prevalence	95% UI	Country	YLDs	95% UI
Cerebral palsy						
Rank based on	**Country**					
No. of cases						
1	India	1,741,232.1	1,464,641.9–2,094,260.6	India	376,934.2	251,818.6–525,062.1
2	China	617,227.6	510,482.7–763,254.5	China	133,982.7	90,676.3–187,606
3	Pakistan	499,455.5	420,258.4–602,818	Pakistan	107,984.6	72,469.9–150,034
4	Nigeria	469,079.2	411,845.5–538,426.6	Nigeria	103,274.0	71,648.7–139,313.1
5	Bangladesh	330,902.7	283,252.2–389,202.8	Bangladesh	72,104.4	49,300.9–99,638.7
6	Ethiopia	286,594.3	248,242.1–333,119.4	Ethiopia	61,847.6	42,509–84,242.6
7	Democratic Republic of the Congo	219,682.7	194,823.1–248,569	Democratic Republic of the Congo	48,524.8	32,983.8–66,965.4
8	Brazil	162,259.4	143,872.2–183,144.3	Brazil	35,227.4	24,238.1–47,840.6
9	United Republic of Tanzania	151,225.0	134,382.6–170,457.2	United Republic of Tanzania	33,121.6	23,190.4–45,329.5
10	Indonesia	141,176.9	118,342–169,567.5	Indonesia	30,529.1	20,508.4–42,394.6
Rank based on						
Rate/100,000						
1	Bangladesh	2,407.3	2,060.6–2,831.4	Bangladesh	524.6	358.7–724.9
2	Comoros	2,166.6	1,921.2–2,445.9	Comoros	469.4	318.9–646.6
3	Gabon	2,150.9	1,944.7–2,387.8	Gabon	467.4	317.8–640.8
4	Botswana	2,072.9	1,866.5–2,321.2	Botswana	449.9	309.5–609.9
5	Guinea-Bissau	2,040.7	1,811.9–2,283.4	Guinea-Bissau	446.4	309.6–599.8
6	Namibia	1,934.8	1,746.1–2,163.4	Namibia	418.2	286.9–569.1
7	Gambia	1,926.5	1,715.7–2,162.5	Gambia	416.4	284.8–564.8
8	Mauritania	1,915.3	1,706.2–2,149	Senegal	415.8	284.9–566.8
9	Senegal	1,912.4	1,710–2,146.6	Mauritania	415.6	288–561.5
10	Ghana	1,905.4	1,671.2–2,152.9	Ghana	415.0	283.6–567.4
Developmental intellectual disability						
Rank based on	**Country**					
No. of cases						
1	India	5,398,051.8	3,375,173.5–7,453,873	India	374,294.2	224,952.6–564,039.1
2	China	1,136,764.1	938,177.4–1,375,414.6	China	101,402.4	66,371.4–142,134.1
3	Pakistan	938,551.6	613,785.6–1,269,948.7	Pakistan	66,873.9	41,700.6–99,348.1
4	Nigeria	548,985.2	435,326.9–685,929	Nigeria	47,227.3	31,529.5–67,219.2
5	Ethiopia	370,801.6	260,947.9–486,018.4	Indonesia	28,322.2	18,743.2–40,323.8
6	Indonesia	342,570.7	257,462.2–435,046.4	Ethiopia	27,446.9	17,640.7–40,004.4
7	Democratic Republic of the Congo	317,774.3	208,151.5–426,136.3	United States of America	24,738.7	16,133–35,132.4
8	Egypt	316,966.1	215,476.6–417,973	Democratic Republic of the Congo	23,174.0	14,565.5–34,256.6
9	Afghanistan	298,448.8	190,228.1–406,802.4	Brazil	22,544.6	15,014.2–31,668.2
10	United States of America	282,629.5	223,324.4–355,956.7	Bangladesh	22,412.2	14,477.1–32,482.1

(Continued)

TABLE 2 Continued

		Prevalence	95% UI	Country	YLDs	95% UI
Rank based on Rate/100,000						
1	India	4,610.9	2,883–6,367	India	319.7	192.2–481.8
2	Afghanistan	4,491.2	2,862.7–6,121.8	Afghanistan	303.6	183.9–462
3	Yemen	4,330.0	2,773.8–5,853.4	Yemen	284.4	169.4–432.2
4	Somalia	4,065.9	2,552.6–5,572.1	Somalia	279.0	170.6–425.4
5	Sudan	3,382.1	2,176.1–4,623.7	Sudan	233.2	142–353.2
6	Nepal	3,194.5	2,100.9–4,296.7	Nepal	230.7	143.5–340.8
7	Syrian Arab Republic	3,151.5	2,087.9–4,262.1	Pakistan	219.1	136.6–325.4
8	Pakistan	3,074.5	2,010.7–4,160.2	Syrian Arab Republic	216.8	133.3–322.5
9	Palestine	3,030.1	2,006.4–4,053.8	Palestine	206.4	125.7–308.3
10	Egypt	2,928.0	1,990.5–3,861.1	Republic of Moldova	205.7	136.4–294.1

YLDs, years lived with disability; UI, uncertainty interval.

educational, environmental, and demographic differences across populations impede the standardization and global comparison of disability weights. Also, disability weights specifically for childhood conditions are still not available. Several ongoing studies on disability weight in different countries are expected to provide further insights on this subject in the future (13). Finally, it was difficult to combine the findings in this study with our earlier reports on developmental disabilities (8, 15), which may be achieved in the future with further improvements in accounting for children with multiple disabilities across multiple developmental domains. Despite these shortcomings, the difficulties in counting and monitoring developmental disabilities routinely through traditional systematic reviews and meta-analyses, household surveys and population-based surveillance programs, particularly in LMICs, means that estimates from statistical modeling remain an invaluable source of data to inform policies and interventions in global health (22).

Conclusion

Evidence from the 2019 GBD-WHO Rehabilitation Database suggests that CP and intellectual disability are highly prevalent and associated with substantial YLDs among children under 5 years globally. The burden of these conditions, as with other previously reported developmental disabilities, is higher in LMICs where very limited support services exist compared to HICs. Early detection and high-quality rehabilitation programs for the affected children must be prioritized globally. The SDGs provide unprecedented opportunity to develop and promote requisite policies and programs to ensure that the affected children are offered the best possible prospects for optimal development and inclusive education. While these estimates represent the best available data for policymakers in global health, evidence from surveillance registries and household surveys suggest that further research is warranted to determine improved estimates for these conditions especially in regions with hardly any primary data sources.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

BO conceived the study. BO and MG wrote the first draft, and finalized the manuscript for submission. AD and JO coordinated the data analysis and interpretation with IHME. SW, MH-A, MG, and N-YB critically reviewed the first draft for important intellectual content and provided overall guidance for the revisions. MN, NA, VK, MS-V, AK-M, TS, CC-H, RH, OC, JA, AE, AS, SG, AW, DW, and CN critically reviewed the first draft for important intellectual content. SW, MH-A, N-YB, and CN provided additional critical review of the final draft. All authors read and approved the final version for submission.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.894546/full#supplementary-material

SUPPLEMENTARY APPENDIX 1
Country grouping by the World Health Organization and the World Bank.

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Education for children and adolescents living with disabilities in sub–Saharan Africa—The gaps and opportunities

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Introduction

The World Health Organization (WHO) defines disability as an umbrella term that covers impairments, activity limitations, and restrictions in participation (1). Disability is not considered a health problem, but rather an interaction between a person's body functions and features of the environments in which they live (1). WHO report a higher prevalence of severe and moderate disabilities in Africa compared to other regions (1). The United Nations Children's Fund (2021) provides a global estimate of 230 million children, ages 0–17 years, living with a disability with 28.9 million children found in Eastern and Southern Africa (2). More than half of these children live in rural settings and only about one third attend a primary school (1). Given the high birth rate of 22.6 births per 1,000 people in East Africa, and successful implementation of interventions that have significantly reduced the under-5 mortality rate in this region, the prevalence of childhood disability can only increase over time (3, 4). This is a pertinent current and future issue given that the estimated likelihood of a child having a disability before their fifth birthday is 10 times higher than the likelihood of dying (377.2 vs. 38.2 per 1,000 live births) (5).

The UN Sustainable Development Goals (6), place early childhood development as an international priority. Specifically, target 4.2 sets out a clear mandate to "ensure that all girls and boys have access to good-quality early childhood development" with specific global indicators measuring the proportion of children under 5 years of age, who are developmentally on track in health, learning and psychosocial wellbeing (7). To achieve optimum early childhood development, the Sustainable Development Goals (SDGs) require regular monitoring of all children's health and wellbeing (7, 8).

Successful models of inclusive education have been implemented in low and middle income countries (LMIC) such as Malaysia which has systematically provided for training of special education teachers from 1990 and created a department for special education in 1995. This was followed a chapter on special education in the education act in 1996 and the education rules that established special schools as well as integrated and inclusive education programs (9). Malaysia implemented the "zero reject" policy in 2019 which aims to ensure that children living with disability can be enrolled in any government or government assisted school of their choice (10).

This exemplary evolution in inclusive education in Malaysia is summarized in Figure 1 below.

In one global data set on children with developmental disorders in Africa, the most common disabilities reported were hearing and visual impairments, intellectual disability and autism spectrum disorder (4). Illiteracy among adults living with disability in Africa compromises potential personal independence, desired social interactions, and exposes this group to exploitation (11).

To adequately meet the needs of CALWD Sub-Saharan Africa needs to refocus its efforts. This redefined focus requires integrated interventions including measures to reduce occurrence of developmental disabilities by targeting preventable biological and environmental contributors, such as sub-optimal perinatal care and economic deprivation; promotion of early diagnosis of disabilities coupled with timely interventions delivered during the time sensitive periods of early brain development; and finally support for wide-ranging, accessible and impactful interventions one of which is inclusive education (3, 11–14). Inclusive education also demands provision of assistive technologies inclusive of hardware and software, and an accommodating environment that allows the best possible attainment for these CALWD (8, 15, 16). This calls for a progressive policy framework driven by governments and relevant partners for realization of these demands (11). Special attention is required for the girl-child living with disability. In East Africa she is much less likely to remain and complete her education compared to her male counterparts and especially so if she hails from an ultra-poor background (10). In addition, cultural norms, biological factors, insecurity, climate change and unprecedented events such as the COVID-19 pandemic have all further contributed to this occurrence (12).

The current state of schools in Africa in accommodating CALWD

Education in young children provides an opportunity to refine developmental abilities that contribute toward highest

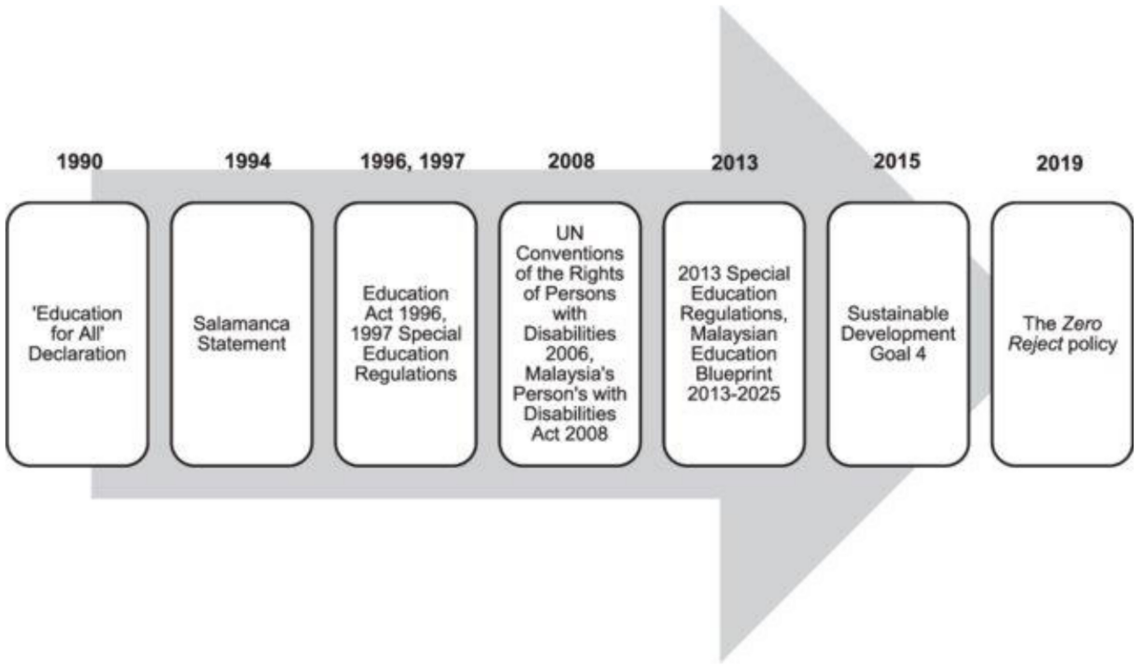

FIGURE 1
The evolution of inclusive education in Malaysia. Ref. Chin (10).

attainable level of personal independence (8). Development of language, cognition, motor abilities and quality of social interaction progresses rapidly throughout the early years of life. It is during this time that tailored education efforts are most likely to be most impactful for all children. Unmet developmental potential in children and young people has social and economic implications for individuals, families and the community at large by negating potential contributions and independence (8, 14, 17).

Children and adolescents who live with physical disability require specific physical accommodations to allow them participate in all-inclusive education settings. Where possible co-location of therapy supports within the school allows CALWD access these services with minimal compromise to school attendance. Such considerations are rarely ever applied within the majority of schools in sub–Saharan Africa and South Asia (8, 12, 15).

A study from South Africa observed that a facility for deaf-blind learners was available but educators and their assistants were ill-equipped to meet the diverse learning needs of these students and had had minimal access to skills upgrade systems which negatively impacted their capacity for optimal skills transfer to learners (18–20). Poor availability of speech language pathologists particularly in East Africa negatively impacts on the possibility for hearing impaired children to receive interventions that prepare them for formal education (21).

In Africa an estimated 350,674 children below 15 years of age are blind and many more are living with undiagnosed low vision (22, 23). Children and adolescents with visual impairment require specifically trained teachers, equipment orientation interventions and ophthalmology services that provide enhancements to make reading possible. The evidence base demonstrates that these support are largely unavailable contrary to the convention on the rights of persons with disability which envisages inclusive education leading to opportunity loss for education for such children who are otherwise capable of learning (24). Gender specific exclusions have also been observed in Africa with school enrolment of visually impaired girls being lower than that of boys (24). Overall transition rates from primary to secondary school for visually impaired children and adolescents is also low in Africa (20).

Instances of bullying and intentional physical violence toward vulnerable children and adolescents with various disabilities have been reported (25, 26). Physical violence from school staff is a particularly common experience among children under 18 years in schools in Kenya and Tanzania (25). Indeed, the frequency of violence toward CALWD is higher than that reported by typically developing children (25, 26). School based interventions such as the "Good-school toolkit" have been effectively utilized to reduce violence toward adolescents living with disability (27, 28).

School readiness and optimizing education for CALWD in Africa

Africa has a predominantly young population and has opportunity to improve economic outputs and quality of life for its communities by empowering CALWD through provision of relevant and contextually appropriate education (29). According to the National Educational Goals Panel, a child's school readiness is dependent on supportive families, communities and schools. Children's school readiness consists of five components; physical health and motor development, social and emotional development, language development, approaches to learning, cognition, and general knowledge (30). Health care providers are in close contact with families prenatally up to young adulthood providing opportunities to optimize school readiness by supporting these five components from the very beginning (30). With the exception of Southern Africa, minimal data exists on efforts to ensure school readiness for CALWD (31, 32).

Early diagnosis of childhood onset disability is a critical first step in improving health related and other outcomes for this population. Studies from south Asia and Africa have demonstrated that assessments lacking adaptation to specific cultural contexts can lead to inaccurate interpretation of performance (33–35). Utilization of locally developed and validated assessment tools as well as inclusion of parents in assessment of CALWD would help identify and place CALWD in appropriate educational settings (33–35). Parents may act as teachers, partners, decision makers and advocates for CALWD and should be continually involved even when their own literacy skills are low (36, 37).

Collaborative models involving parental inputs, training of special education teachers and providing inclusive education that also co-locates therapists operating in the same setting would bring African countries closer to achieving effective education for CALWD (38). To achieve these wide-ranging measures, interventions including policy development and implementation as and changes in social-cultural attitudes toward education for CALDW would be required (9, 10, 27). African countries would need to commit advancements to improve the understanding of the general public regarding education for CALWD in order to realize the vision of an inclusive education (17, 27, 38).

Peer support and social interactions between individuals with disabilities and typically developing children have been shown to have significant positive impacts on the lives of children with disabilities (39). Typically developing children and adolescents better understand the unique needs and strengths CALWD have and can better advocate and accommodate them in their current and future operations when both groups participate in an inclusive education setting (14). This leads to a more cohesive society where

CALWD and their families are "seen," "heard" and have sense of belonging. Africa has the opportunity to educate its communities better on the needs and benefits of inclusive education (27, 38). These understandings would reduce stigma, emotional and physical abuse and eventually improve advocacy for individuals and institutions that support CALWD.

Ratification of Convention on the Rights of Persons with Disabilities by the remaining African states will form a basis for ensuring support for these vulnerable persons (2, 11, 38, 40).

Conclusion

Childhood disability in Africa is currently a significant concern with the numbers of those affected expected to increase over time. This calls for a redefined attention to integrated and multilayered approaches to reduce occurrence and impact of developmental disabilities. Current school environments in Africa largely do not cater to the social, physical, and technological accessibility to education that fosters long term inclusivity of CALWD. In the final analysis this this negates possibility for future independence and positive contribution to society for CALWD. Parents and healthcare workers should be facilitated to participate in nurturing care, assessment and identification of young children with disability which turn fosters school readiness increasing the possibility for CALWD to participate in education. Utilization of contextually appropriate and validated tools to identify CALWD in Africa will contribute to education related advocacy efforts and encourage policy makers fully implement the goals envisioned in SDG 4.

Author contributions

PS and SW provided the concept for the opinion piece. All authors contributed to the submission and approved the final version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Early care and support for young children with developmental disabilities and their caregivers in Uganda: The Baby Ubuntu feasibility trial

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Background: Early care and support provision for young children with developmental disabilities is frequently lacking, yet has potential to improve child and family outcomes, and is crucial for promoting access to healthcare and early education. We evaluated the feasibility, acceptability, early evidence of impact and provider costs of the Baby Ubuntu participatory, peer-facilitated, group program for young children with developmental disabilities and their caregivers in Uganda.

Materials and methods: A feasibility trial, with two parallel groups, compared Baby Ubuntu with standard care. Caregivers and children, aged 6–11 months with moderate-severe neurodevelopmental impairment, were recruited and followed for 12 months. Quantitative and qualitative methods captured information on feasibility (ability to recruit), acceptability (satisfactory attendance), preliminary evidence of impact (family quality of life) and provider costs.

Results: One hundred twenty-six infants (median developmental quotient, 28.7) were recruited and randomized (63 per arm) over 9 months, demonstrating feasibility; 101 (80%) completed the 12-month follow-up assessment (9 died, 12 were lost to follow up, 4 withdrew). Of 63 randomized to the intervention, 59 survived (93%); of these, 51 (86%) attended ≥6 modules

meeting acceptability criteria, and 49 (83%) completed the 12 month follow-up assessment. Qualitatively, Baby Ubuntu was feasible and acceptable to caregivers and facilitators. Enabling factors included community sensitization by local champions, positive and caring attitudes of facilitators toward children with disability, peer support, and the participatory approach to learning. Among 101 (86%) surviving children seen at 12 months, mixed methods evaluation provided qualitative evidence of impact on family knowledge, skills, and attitudes, however impact on a scored family quality of life tool was inconclusive. Barriers included stigma and exclusion, poverty, and the need to manage expectations around the child's progress. Total provider cost for delivering the program per participant was USD 232.

Conclusion: A pilot feasibility trial of the Baby Ubuntu program found it to be feasible and acceptable to children, caregivers and healthcare workers in Uganda. A mixed methods evaluation provided rich programmatic learning including qualitative, but not quantitative, evidence of impact. The cost estimate represents a feasible intervention for this vulnerable group, encouraging financial sustainability at scale.

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KEYWORDS

parenting program, early intervention, developmental disability, feasibility trial, caregiver, young children, Uganda

Introduction

Addressing the needs of the 53 million children under 5 years of age living with developmental disabilities is a global priority (1), with early child development, inclusive of early childhood disability, recognized in the current Sustainable Development Goal (SDG) era. This has been strongly supported by the United Nations Global Strategy for Women's, Children's and Adolescents' Health advocating for all children to not only "survive" but also "thrive" through community and service transformation (2). Supporting young children with disabilities and their caregivers to access inclusive healthcare and early education, remains a crucial component of the SDGs.

Child survivors of common neonatal conditions, such as neonatal encephalopathy (newborn brain injury), preterm birth and neonatal infections, are "at risk" of a wide spectrum of neurodevelopmental difficulties, delays and disabilities (3). These include cerebral palsy and other global developmental disabilities which may limit independent mobility and feeding, and are linked to cognitive delay, epilepsy, visual, hearing and behavioral difficulties. Developmental disabilities have long term physical, emotional, social, and financial consequences for the child and family in any context, but particularly in low-income country (LIC) settings, where availability of, and access to, support services and inclusive early education are often limited and complicated by financial barriers, social stigma and exclusion (4–8). There is also substantial impact on wider society due to the loss of learning potential and economic productivity, perpetuating poverty in the lowest resource settings (9).

Early programs of care and support have the potential to improve neurodevelopmental outcomes for at-risk children (10). Detecting and intervening early is key to taking advantage of the neuroplasticity of the immature developing brain over the first 3 years of life, to maximize the child's functioning and developmental potential (11). Importantly, these programs can also have other positive effects on child and family quality of life, health and wellbeing through family capacity strengthening and enrichment of the care-giving environment (12), and optimizing school readiness through promotion of parenting knowledge, skills, and practices (13).

However, programs of early care and support for caregivers of children with developmental disabilities have been under studied, particularly in LIC settings (14, 15). Such programs are wide-ranging in content and approach but may include physiotherapy, occupational, and speech and language therapy interventions, interactive sessions to improve parent-child interactions, and caregiver mental health and peer support, which can be delivered in child development centers, homes or other community locations (14, 16). Several trials have shown positive effects on child motor and cognitive outcomes and caregiver mental health (17, 18) although the populations included were not assessed to have neurodevelopmental

impairments (NDIs) and may not all have been particularly "at risk" (19–22). Few studies to date, have examined the feasibility, acceptability, impact and cost-effectiveness of such programs in LICs, and how they might be integrated into existing community programs to promote health and access to early education, although studies are underway (23–28). A systematic, sustained and coordinated approach to implementing and monitoring early detection and intervention initiatives is needed, to improve the life chances of millions of affected children and their families.

In Uganda, it is estimated that 3.5% of all children aged 2–4 years and 7.5% of children aged 5–17 years live with a disability (29); however, only 10% have access to rehabilitative services (15). Empowering mothers to access care and promoting inclusion and participation is key to encouraging early development; in Uganda 51% of women reported full participation in household decision-making, an improvement from 38% in 2011 (30). The Ugandan Ministry of Health have highlighted the need for an integrated policy on early child development, which requires a multisectoral approach comprising health, education, sanitation, empowerment, and safeguarding (31, 32).

The Baby Ubuntu program is a community-based, group, participatory, peer-led program of early care and support for young children (0–3 years) with developmental disabilities and their caregivers (15, 33), formerly known as the ABAaNA Early Intervention Program.[1] A conceptual framework of potential pathways to impact of the program at scale is shown in **Figure 1**. Previous non-controlled pre/post mixed-methods evaluations in Uganda and Rwanda have shown a 15–20% increase in family impact quality of life post-intervention (15, 34), however the feasibility, acceptability, impact and scalability of the program have not previously been formally evaluated. We conducted an individually randomized, pilot feasibility trial of the Baby Ubuntu program, inclusive of a mixed-methods evaluation of (i) program feasibility and acceptability for caregivers and healthcare workers (ii) preliminary evidence of impact when compared with standard care (iii) factors important for scale-up and (iv) provider costs of implementation.

Materials and methods

A pilot feasibility single-blind, randomized controlled trial with two parallel groups across two study sites, one urban (Kampala) and one rural (Nakaseke), was undertaken. Neither site had existing formal support services for children with developmental disabilities, although referrals to specialist services including pediatric neurology and physiotherapy were possible (**Supplementary Figure 1**). Full details of the research methodology are described in the published protocol (33).

1 Baby Ubuntu early intervention program online resources: https://www.ubuntu-hub.org/resources/babyubuntu/.

Participants and eligibility

Trial participants were infants aged 6–11 months with moderate-severe NDI [defined as a Griffiths Mental Developmental Scales (35) (quotient <70) and/or Hammersmith Infant Neurological Examination (36) (score <60)], and their primary caregivers (most frequently the mother however may be another relative or carer depending on each family's individual circumstances), and from whom informed written consent was obtained. Exclusion criteria included: age ≥12 months; medical conditions requiring inpatient treatment; unwilling/unable to attend the full program; main residence outside a pre-defined geographic site criterion; non-Luganda or English speakers. Witnessed consent using a thumb print was available to caregivers with low literacy.

Screening, recruitment and randomization

At-risk infants in the community were screened for eligibility using the Malawi Developmental Assessment Tool (MDAT) (37). Comprehensive neurodevelopmental assessment using the Griffiths Mental Developmental Scales (GMDS) and HINE, was performed for those screening positive for developmental delay. Infants and their caregivers were randomized in a 1:1 ratio to the intervention or standard care (SC) arm using a random number generator prior to the commencement of the study, as previously described (33).

The intervention

The Baby Ubuntu program manual is freely available to download (footnote 1). The program is divided into ten modules, each lasting 2–3 h, covering understanding disability, positioning and carrying, feeding, mobilizing, communication, play, everyday activities, and the child within the community (34). Modules are delivered over 4–6 months, incorporating at least one home visit. The Baby Ubuntu groups are facilitated by an "Expert Parent," themselves a caregiver of a child with developmental disability, with or without a healthcare professional. Facilitators receive 5 days of structured training with ongoing supervision and mentorship by a Baby Ubuntu "Master Trainer." The program manual is freely available to download. Program groups of 6–10 participating families were selected based on locality.

Standard care (SC) referred to existing local services which includes some limited access to physiotherapy and assistive devices, seizure management, audiology, ophthalmology, and nutritional support (**Supplementary Figure 1**), and

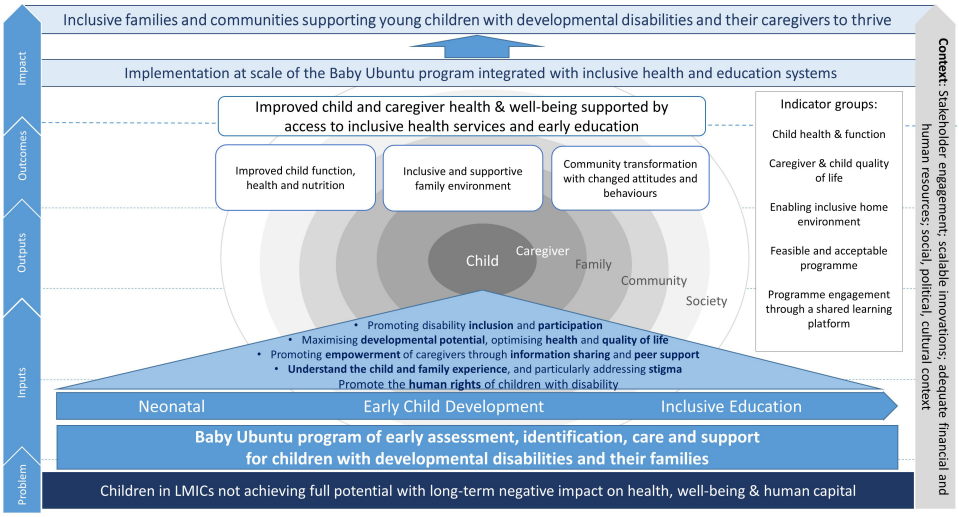

FIGURE 1
Conceptual framework for the Baby Ubuntu program.

this group were offered entry to the program following completion of the study.

Outcome measurement

Quantitative data

Feasibility was evaluated quantitatively as the total number recruited and randomized to each arm over a pre-specified time period (9 months). Acceptability amongst caregivers and healthcare workers was assessed by the protocol violation rate, e.g., participants in the intervention arm being treated as if in the control arm or vice versa, and by pre-specified criteria for "satisfactory attendance" (≥6 modules).

A number of outcome measure tools were piloted and used to examine for early evidence of impact on child and caregiver outcomes (33). These included; *Family quality of life* (QoL) assessed using the scored Pediatric Quality of Life Family Impact module (PedsQL) (38); *Child motor functioning* assessed by the mobility score of the Pediatric Evaluation Disability Inventory (PEDI-CAT) (39); *Child cognitive function* as assessed by the Griffiths Mental Developmental Scales (GMDS) (35); *Child growth and nutritional status* assessed by weight, height, occipito-frontal head circumference and estimation of hemoglobin (HemoCue AB, Angelholm, Sweden); *Caregiver mental health* as assessed using the Self-Referral Questionnaire (SRQ) and the Parenting Stress index (PSI) (40); *Caregiver-child attachment* using the Maternal Infant Responsiveness Instrument (MIRI) (41); and *Quality of the home environment* assessed using the Infant Toddler-Home Observation for the Measurement of the Environment (IT-HOME) (42).

For quantitative data, participants in both arms were assessed by study staff blind to trial allocation, at three time points; pre-intervention (age 6–11 months), at program completion (age 12–17 months), and at 12 months post-completion (age 18–23 months). Outcome measure tools were administered by trained study staff including nurses, doctors, physiotherapist, occupational therapist, medical clinical officer, and a clinical psychologist.

Qualitative data

A social scientist conducted in-depth interviews (IDIs) with 20 randomly selected primary female caregivers in the intervention and SC arms at baseline and endline, and nine intervention arm male caregivers at endline. Participants selected for IDI were contacted and interviewed in the local language. Interviews were conducted at the study site, and later transcribed into English by the social scientist for analysis.

To further develop our understanding of program feasibility, acceptability, impact and scale-up, focus group discussions (FGDs) with female caregivers (two FGDs per site) and healthcare workers (one FGD with healthcare workers from both sites) were conducted. In addition, a stakeholder workshop for investigators, study staff, program facilitators and caregivers was held.

Provider costs

A cost analysis was conducted to examine program costs including set-up (training, equipment and furniture, pre-program expenditure) and running costs (staff, building,

supplies, transport refunds, home visits, outreach) over a 1-year time period. Costs relating to the trial, as opposed to implementation of the intervention, were not included in provider costs. Information was gathered from financial data recorded by the project implementation team and facility and program staff interviews. Costs were allocated according to the implementation activities of the program: recruitment, education sessions, and home visits. Costs were inputted into an Excel-based costing tool, in prices in the currency that the cost was incurred; British pounds (GBP) and Ugandan shillings (UGX), and annualized to obtain the economic costs. Costs were incurred in 2018 then inflated to May 2022 prices based on the consumer price index of the currency of the initial recorded cost.[2] Finally, all costs were converted to 2022 US dollars (USD).

Sample size

The trial aimed to recruit 126 children and their caregivers; 63 per arm. Allowing for a 20% dropout rate, this sample size gives 90% power to detect a minimal relative difference of 20% on PedsQL Family Impact score between the intervention and control arms, at 5% significance level, assuming a mean PedsQL score of 65 in the SC arm and SD of 20 in both arms (based on data from a pilot pre- and post-evaluation study). The provider cost analysis was performed using all participants completing the program in the intervention arm ($n = 56$).

Data analysis

Feasibility of participant recruitment and randomization was assessed quantitatively by the total number recruited and randomized to each arm, with feasibility demonstrated if the target sample size of 126 was achieved within the 9-month recruitment period. Acceptability was assessed quantitatively by (i) calculating the protocol violation rate and (ii) summarizing the number of program sessions attended with 6 or more defined as acceptable. Analyses compared outcomes between intervention and control arms at the end of the program, and again 6 months later. On the advice of the DSMB and following CONSORT reporting guidelines for pilot and feasibility trials, we did not plan any formal statistical tests due to the preliminary nature of the trial; instead confidence intervals provide inference around the possible range of effect sizes. Regression models were used to adjust comparisons for baseline measures of the outcomes. Analysis was done on an intention-to-treat basis and missing data were not imputed. The DSMB did not instigate any interim analyses or stopping guidance.

Qualitative data were analyzed using a thematic framework approach around the topics of feasibility, acceptability, impact and scale-up. Two social scientists reviewed the interview transcripts to identify the codes and themes based on the study objectives and other interesting themes that emerged from the data. We described the experiences of children and caregivers relating to the intervention received including the impact of the disability, parental confidence level, inclusion in community life and experiences of stigma and discrimination. We examined changes in these domains over the follow-up period and explored attributions of change. In addition, we performed social mapping of parent networks and group discussions with staff on their perspectives and experiences of using the program.

Results

In total, 126 infants were recruited between 25th January and 16th October 2018, with 101 (80.2%) participants completing the final follow-up assessment at 18–23 months, by 2nd October 2019. Twelve (9.5%) were lost to follow up, 4 (3.2%) withdrew, and 9 (7.1%) died. Of 63 randomized to the intervention, 59 survived (93%); of these, 51 (86%) attended ≥6 modules, and 49 (83%) completed the follow-up assessment at 18–23 months. The flow of trial participants is outlined in **Figure 2**.

Baseline characteristics and descriptive analyses

Baseline characteristics of recruited children are presented in **Table 1**. Median age at recruitment was 9.5 months. Most recruited infants had severe NDI (GMDS DQ < 55).

Descriptive analyses of qualitative research participants

Of the 20 randomly selected female caregivers, all participated in IDIs at baseline and 16 (80%) at endline; the four who were not interviewed had a child that died during the study period. Nine of the 20 caregivers (45%) were <25 years old (mean age 25.7 years), and for 55% (11/20) the recruited child was their first born. All but 2 were biological parents; a grandmother and a maternal aunt were caring for two children due to one mother being away and another having a disability. All participants had some primary education and most lived separately from the child's father (either fully or partially separated) at the time of recruitment. At the rural study site (total 10), the majority (8) were small-scale farmers; at the urban site (total 10) six were engaged in small-scale business.

Male caregivers [total, 9 (5 rural, 4 urban)] interviewed were older (mean 36 years, range 27–52), and all were considered married; 2 in their second marriage, and 3 had more than one

2 Uganda consumer price index 2016/17: https://www.ubos.org/wp-content/uploads/publications/12_2021CPI_PUBLICATION_NOVEMBER_2021.pdf.

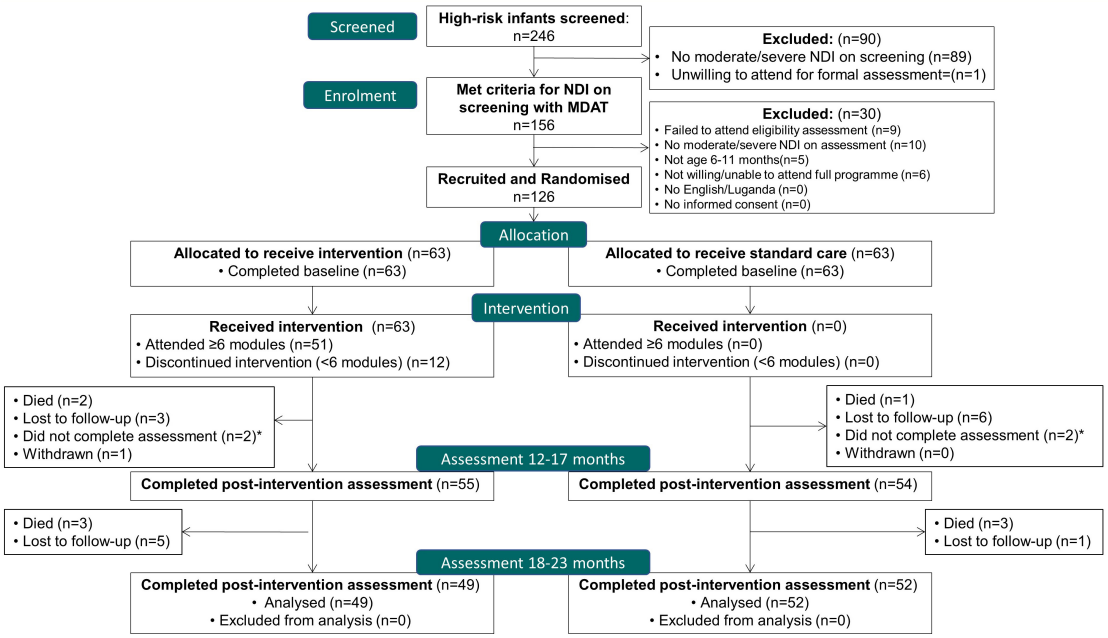

FIGURE 2
Flow of participants; CONSORT flow diagram. *Two participants in each arm did not complete the 12–17 month assessment but returned for the 18–23 month visit.

wife. All had some primary school education, and 5 had some secondary school education; all were employed, the majority in manual labor.

Focus group discussions at baseline and endline included, 4 with a total of 32 female caregivers (8 from each arm at each site), and 2 FGDs with 10 healthcare workers (4 urban, 6 rural). The stakeholder workshop held in November 2019 was attended by 6 research investigators, 14 study staff or healthcare workers (HCWs), 3 program facilitators, 4 caregivers, and 5 other stakeholders.

Program feasibility and acceptability

In total, the target number (n = 126) of infants were recruited in less than 9 months meeting the primary feasibility outcome. Acceptability of the program was indicated by all children receiving either the intervention or standard care according to allocation (no protocol violations). A total of 51 intervention arm families attended six or more sessions (84%, allowing for the two children who died during the intervention).

Qualitative analysis of the findings provided evidence that the intervention was both feasible and acceptable to most participants, facilitators and HCWs. Major enabling factors cited were peer support from other caregivers; local community members acting as "champions" to support mobilization families to participate in the program; the positive and caring attitudes of HCWs and facilitators toward children with disability creating a conducive enabling social environment; good accessibility of training and materials; and incentives including transport reimbursement. Program barriers included lack of engagement of male caregivers, lack of community awareness around child disability, superstition around etiology of disability including discrimination, and challenges relating to poverty including traveling to sessions. Proposed solutions included active early engagement of male caregivers, earlier and greater emphasis on community sensitization to promote wider engagement and geographically locating groups at more local community clinics rather than central referral hospitals, and the provision of social protection and other livelihood support. Themes and sub-themes emerging from the qualitative analysis on feasibility and acceptability are presented in **Supplementary Table 1**.

Prior to the program, caregivers and HCWs expressed concerns that particularly in the rural setting, there was very limited provision of care services available for children with disabilities. Whilst specialty services were available at both sites (**Supplementary Figure 1**), access to these services was complicated by HCWs often lacking knowledge and skills in managing children with disability which hindered communication with caregivers and timely referral. Exclusion of children with developmental disability from services was commonly described by caregivers. One female caregiver reported being sent away from the facility to see a herbalist because she had attended the same facility several times and the

TABLE 1 Baseline characteristics of study participants by trial arm.

Characteristic	Intervention ($n = 63$)	Standard care ($n = 63$)	Overall ($n = 126$)
Age in months, median (IQR) [range]	9.4 (7.2–10.2) [6.0–11.9]	9.5 (7.8–10.2) [6.0–12.6]	9.5 (7.5–10.2) [6.0–12.6]
Sex (Male)	32 (51%)	32 (51%)	64 (51%)
Mother's education			
None/Primary	21 (34%)	26 (41%)	47 (38%)
Secondary	29 (48%)	29 (46%)	58 (47%)
Tertiary	11 (18%)	8 (13%)	19 (15%)
Father's education			
None/Primary	20 (34%)	16 (27%)	36 (30%)
Secondary	22 (37%)	29 (48%)	51 (43%)
Tertiary	17 (29%)	15 (25%)	32 (27%)
DQ median (IQR) [range]	33.4 (16.3–45.5) [0.6–75.5]	27.4 (12.2–36.1) [3.0–80.0]	28.7 (14.2–42.6) [0.6–80.0]
HINE score median (IQR) [range]	33 (23.5–44) [13 –68]	30 (21–47) [10–69]	32.5 (22–46) [10–69]
Weight-for-age z-score, median (IQR) [range]	−2.4 (−3.8, −1.3) [−6.1, 0.8]	−2.4 (−3.3, −1.2) [−5.8, 1.3]	−2.4 (−3.5, −1.2) [−6.1, 1.3]
Head circumference-for-age z-score, median (IQR) [range]	−2.6 (−4.1, −1.1) [−6.0, 6.0]	−2.2 (−3.5, −0.9) [−6.0, 6.0]	−2.4 (−3.8, −1.0) [−6.0, 6.0]

DQ, developmental quotient on griffiths mental development scales II; HINE, hammersmith infant neurological examination.

child was not improving. This was a theme that came through particularly strongly at the stakeholders meeting.

One female caregiver from the intervention arm shared how she felt when she first joined the program "*I used to ask myself so many questions why my child was different… I felt that I was alone, but when (the program coordinator) invited me, she explained to me that we were going to meet in groups and learn how to take care of our children. I got excited.*"

Feasibility of the program

Participant recruitment was greatly facilitated through community health outreach by program coordinators and facilitators. HCWs described the importance of community champions in both community sensitization, identification and mobilization of families of children with developmental disability.

Pre-intervention, female caregivers described high levels of stigma and emotional, social and financial burden associated with caring for a child with developmental disability, and the substantial barriers that this represented to program enrollment. Several female caregivers reported that the program helped reduce their own self-stigma and blame, which helped them to understand and accept the condition and situation of their children. An 18-year old female caregiver shared the importance of the program helping her to understand that her child's disability was not her fault, or caused by "witchcraft," and that she did not need to segregate him from other children. Community sensitization to issues around child disability were highlighted by both female and male caregivers. Fathers particularly emphasized the importance of sensitizing everyone in the community to the learnings from the program as child disability could affect others; in his words "*Today it's me, tomorrow it's you*" (IDI P8).

Amongst barriers to feasibility and acceptability, mentioned by caregivers and HCWs, was the limited engagement of fathers. Whilst none of the interviewed fathers had attended the program directly, most appreciated the support provided to their children and were keen to implement what had been learnt. As one father said: "*as long as you have chosen to be a parent, you have to be involved at all stages, because that is your child too.*" (IDI, P1). Some fathers were exasperated by the number of different programs which come and go at the hospital, especially if they failed to deliver on perceived promises.

> "*…I sent in my child with the mother… to help out with treating the blisters [on] her head but she was told that they couldn't help…because that was not part of (the program) and she didn't get any help. So how is the program going to help me?*" (IDI, P4).

At the outset of the study, most caregivers were struggling financially with caring for their child with developmental disability due to increased care needs and reduced ability to work. Some caregivers reported that as a result of participating in the intervention they were able to return to work, having gained skills on how to care for the child. One of the mother's said, "*I had given up on work since no one could accept to stay with my child, but because I learnt to feed him and how to make him calm, I have returned to work leaving the boy with one of my relatives.*" However, poverty was a clear theme throughout the IDIs. More than half of the female caregivers continued to struggle with transport costs and the cost of specialized care when referred. Caregivers reported poverty as the most important contributor to continued poor quality of life, despite the other positive impacts identified for themselves and their child.

Poverty was commonly identified as a barrier to attendance and was exacerbated by the costs of care, but also loss of income due to caring responsibilities. Prospectively meeting the cost of transport, despite the program reimbursing travel costs, was particularly challenging for some, especially those traveling significant distances due to wide geographical spread of some group members. One female caregiver observed that "*the place was good though it is too far from our homes.*"

Poverty was also cited as a key factor undermining access to care and impact on quality of life. One HCW said "*Most of the families are poor and stay deep in villages so, raising*

money to take their children for treatment on a regular basis may be difficult." Although most caregivers found the training modules especially the practical sessions, easy to implement, some caregivers particularly from rural communities reported that the nutrition module, whilst very useful, was expensive because of the different foodstuffs recommended.

Female caregivers identified the program coordinator as key to success of the groups. They particularly valued the coordinator and facilitators being easy to contact, which enhanced attendance and adherence. Phone call reminders to caregivers about their appointments was valued and reported to positively influence attendance.

Acceptability of the program

Facilitators and HCWs were reported to be accommodating, respectful and friendly which made the caregivers feel confident in participating. This strengthened the rapport between HCWs and caregivers, and promoted emotional wellbeing and a sense of acceptance. One female caregiver said, "*I remember on the first day, I came very early in the morning and had not carried tea for my child. She was all crying with mucus dripping from her nose. As soon as I stepped on the door the (healthcare worker) got the baby from me and carried him before I even greeted her. . . it is so rare (and) encouraged me to keep coming. . .*" The provision of simple refreshments during the group sessions and transport reimbursements were also identified by female caregivers and HCWs as contributing positively to attendance and acceptability.

Most participants within the intervention arm strongly attributed program acceptability, and impact, to the psychosocial support from other caregivers in the group and Expert Parent facilitators. For example, during a post FGD, one of the participants said, "*they could teach us something they have been through which is good. . . and I always wanted to come to meet with them because we would interact freely*." Group composition of participants from the same or neighboring communities motivated caregivers to attend sessions and promoted peer support from within the immediate community. Peer support through the facilitated group, partnered with individualized support from group facilitators, was reported as particularly powerful in meeting the variable needs of different caregiver-child dyads. Participants reported that community (home) visits supported sharing of individual challenges and barriers in more depth, and provided opportunities for one-to-one discussion of issues.

The use of health facilities as the site for group meetings was well received by participants and many reported that this promoted access to other facility-based health services from which they had frequently felt excluded. In addition, participants reported that group meetings facilitated provision of medication and other adjunct medical and therapeutic consultations, and this further incentivized regular attendance. However, HCWs felt that clearer referral pathways to specialist services were also required, particularly for speech and hearing problems.

Facilitators reported that session debriefs led by Master Trainers were highly valued, enabling reflection on learning experiences, delivery of the program, and enabling them to track progress. The participatory approach of the program promoted caregivers to share their experiences, and encouraged hands-on practice of practical skills such as feeding, improving their confidence in caring for their child. One female caregiver said "*We were taught to communicate with (our children) more often so that their brain may grow and learn how to speak. Putting the child nearby you when you are doing work so that the child can also learn how to do what you doing for example when you are washing utensils*."

The caregivers and HCWs were positive about the training materials, and the utilization of everyday items. However, some felt financially challenged in needing to procure materials themselves, e.g., foodstuffs for the feeding module. Caregivers were positive about the sessions being delivered in the local language. However, low literacy levels was highlighted as a barrier to feasibility and acceptability by both caregivers and HCWs. They felt that additional visual materials including pictures and videos would be valuable, as well as more translation from English to local languages. Expert parent facilitators reported that the provision of appropriate training materials ["information, education and communication" (IEC) materials] supported effective content delivery; they also strongly valued the ongoing supervision and mentorship offered by the Master Trainers.

Whilst seeing improvement in their child's functioning encouraged caregivers to return to the program sessions, facilitators and HCWs talked extensively of the need to balance realistic expectations around progress with maintaining hope. This was particularly relevant for those who had children with more severe impairment. Unrealistic caregiver expectations were felt to be a key cause of caregiver disengagement from the program: a female caregiver said, "*. . .if I do not see any change in my child's health I stop coming. Because that will be wastage of energy and money for nothing*."

Impact of the program

A number of tools for measuring child and caregiver outcomes were piloted to examine for early quantitative evidence of impact. **Table 2** reports the crude and adjusted differences seen between the SC and intervention arm immediately and at 6 months post program completion (12 months post-enrollment). Wide confidence intervals consistent with either a beneficial effect, no effect, or a detrimental effect of the intervention were seen (**Table 2**).

Whilst quantitative tools did not provide clear evidence of program impact, qualitative findings provided supportive

TABLE 2 Outcomes at baseline, 6 and 12 months post-enrollment to the baby Ubuntu program, by arm.

Outcome	Baseline measures		Outcomes at 6 months post-enrollment				Outcomes at 12 months post-enrollment			
	Standard care ($n = 63$) mean (SD)	Baby Ubuntu ($n = 63$) mean (SD)	Standard care ($n = 54$) mean (SD)	Baby Ubuntu ($n = 55$) mean (SD)	Crude difference (95% CI)[1]	Adjusted difference (95% CI)[2]	Standard care ($n = 52$) mean (SD)	Baby Ubuntu ($n = 49$) mean (SD)	Crude difference (95% CI)[1]	Adjusted difference (95% CI)[2]
PedsQL										
Total score	61.2 (19.1)	64.8 (18.7)	60.1 (18.1)	58.2 (18.1)	−1.9 (−8.8, 5.0)	−5.4 (−11.4, 0.6)	58.2 (21.8)	61.5 (22.6)	3.4 (−5.4, 12.1)	−0.7 (−8.9, 7.5)
Physical functioning	65.2 (20.8)	64.8 (21.5)	64.8 (22.0)	61.4 (25.0)	−3.3 (−12.3, 5.7)	−3.9 (−12.1, 4.3)	64.0 (23.2)	64.1 (27.5)	0.1 (−9.9, 10.1)	−1.0 (−10.6, 8.6)
Emotional functioning	53.8 (29.6)	59.4 (27.9)	53.8 (28.8)	55.5 (28.1)	1.7 (−9.2, 12.6)	−0.8 (−10.9, 9.3)	51.4 (30.3)	56.5 (31.4)	5.1 (−7.1, 17.2)	1.1 (−10.3, 12.5)
Social functioning	59.9 (30.2)	63/3 (28.6)	56.0 (31.6)	48.3 (30.3)	−7.8 (−19.6, 4.0)	−10.8 (−22.1, 0.5)	52.2 (34.3)	53.2 (32.7)	1.0 (−12.2, 14.3)	−1.6 (−14.6, 11.5)
Cognitive functioning	77.9 (23.3)	79.0 (24.3)	75.9 (23.9)	71.9 (24.1)	−4.1 (−13.3, 5.1)	−5.7 (−14.0, 2.5)	71.8 (25.1)	78.8 (25.9)	6.9 (−3.1, 17.0)	4.8 (−5.2, 14.9)
Communication	56.7 (27.4)	55.8 (25.5)	48.0 (19.6)	51.2 (23.5)	3.3 (−5.0, 11.5)	2.3 (−5.7, 10.3)	55.1 (22.1)	59.9 (31.8)	4.8 (−6.0, 15.6)	3.0 (−7.6, 13.6)
Worry	44.9 (26.5)	51.9 (26.3)	46.9 (26.0)	44.4 (25.2)	−2.5 (−12.3, 7.3)	−7.0 (−16.0, 2.0)	44.3 (27.9)	51.9 (30.4)	7.6 (−3.9, 19.1)	2.1 (−8.9, 13.1)
Daily activities	54.4 (35.1)	62.8 (33.4)	52.2 (31.6)	51.4 (38.1)	−0.8 (−14.2,12.5)	−7.8 (−20.2, 4.6)	50.5 (35.8)	49.1 (38.0)	−1.3 (−15.8, 13.2)	−4.1 (−18.7, 10.5)
Family relationships	70.7 (28.0)	76.9 (29.3)	74.0 (27.6)	73.7 (29.5)	−0.3 (−11.2, 10.6)	−3.9 (−14.4, 6.6)	69.5 (31.3)	70.9 (32.0)	1.4 (−11.0, 13.9)	−2.4 (−14.2, 9.5)
PEDI mobility score[3]	36.3 (5.9)	35.3 (6.7)	35.7 (7.7)	37.2 (7.5)	1.6 (−1.5, 4.6)	1.5 (−1.3, 4.3)	39.3 (6.8)	39.7 (8.0)	0.5 (−2.6, 3.5)	0.4 (−2.3, 3.2)
Developmental quotients										
Global DQ[4]	28.9 (18.5)	31.3 (19.7)	27.6 (19.7)	31.7 (22.3)	4.0 (−4.0, 12.0)	3.0 (−4.5, 10.6)	16.4 (13.9)	20.4 (16.7)	4.0 (−2.1, 10.1)	2.4 (−2.3, 7.0)
Locomotor	28.6 (23.0)	29.8 (23.5)	18.4 (20.4)	21.6 (21.1)	3.2 (−5.1, 11.5)	3.4 (−3.4, 10.3)	16.8 (15.1)	18.6 (18.2)	1.9 (−4.7, 8.5)	1.8 (−3.8, 7.3)
Personal social	33.2 (23.6)	40.2 (27.5)	25.9 (19.2)	30.4 (24.7)	4.5 (−4.2, 13.2)	1.0 (−5.5, 7.5)	20.4 (17.0)	25.1 (19.9)	4.7 (−2.6, 12.1)	2.2 (−3.6, 8.1)
Speech and hearing	36.7 (22.1)	38.3 (23.7)	23.2 (14.6)	29.6 (17.0)	6.4 (0.2, 12.7)	6.1 (0.8, 11.4)	18.9 (12.1)	22.6 (14.0)	3.8 (−1.4, 8.9)	3.1 (−1.3, 7.4)
Eye and hand	21.7 (20.2)	23.9 (21.1)	15.8 (18.8)	21.4 (22.3)	5.6 (−2.6, 13.7)	5.2 (0.2, 10.2)	12.6 (15.4)	17.7 (18.8)	5.2 (−1.6, 12.0)	4.7 (−0.3, 9.7)
Performance	21.4 (17.4)	21.3 (16.0)	15.4 (15.2)	20.5 (20.0)	5.1 (−1.9, 12.1)	6.6 (0.9, 12.3)	13.2 (14.6)	16.6 (16.3)	3.4 (−2.8, 9.5)	4.8 (−0.1, 9.8)
Anthropometry[5]										
Weight-for-age z-score	−2.3 (1.5)	−2.5 (1.7)	−2.5 (1.8)	−2.6 (1.7)	−0.1 (−0.8, 0.6)	0.0 (−0.5, 0.5)	−2.8 (1.5)	−3.4 (1.7)	−0.6 (−1.3, 0.0)	−0.5 (−1.0, 0.0)
Height-for-age z-score	−2.0 (1.6)	−2.4 (2.2)	−2.5 (1.8)	−2.9 (1.9)	−0.4 (−1.1, 0.3)	−0.3 (−0.9, 0.3)	−2.8 (1.5)	−3.2 (1.7)	−0.4 (−1.1, 0.2)	−0.3 (−0.9, 0.2)
HC-for-age z-score	−2.2 (2.0)	−2.2 (2.8)	−2.7 (2.5)	−2.5 (2.6)	0.2 (−0.7, 1.2)	0.3 (−0.4, 0.9)	−3.2 (2.3)	−3.0 (2.7)	0.2 (−0.8, 1.2)	0.1 (−0.5, 0.7)
MUAC-for-age z-score	−1.0 (1.4)	−1.1 (1.5)	−1.1 (1.4)	−1.0 (1.9)	0.1 (−0.6, 0.8)	0.3 (−0.2, 0.9)	−1.1 (1.2)	−1.6 (1.8)	−0.5 (−1.1, 0.1)	−0.4 (−0.9, 0.2)
Caregiver wellbeing										
SRQ score[6]	7.0 (4.7)	6.9 (4.7)	7.3 (4.9)	7.9 (5.2)	0.6 (−1.4, 2.6)	1.1 (−0.7, 3.0)	8.5 (5.2)	7.5 (4.9)	−1.0 (−3.0, 1.0)	−0.5 (−2.3, 1.3)
MIRI score[7]	77.3 (12.7)	78.8 (13.6)	80.5 (12.7)	81.6 (12.0)	1.1 (−4.1, 6.3)	−0.3 (−5.4, 4.7)	82.2 (14.3)	84.4 (12.7)	2.2 (−3.3, 7.7)	0.4 (−4.7, 5.5)
PSI score[8]	90.2 (28.0)	90.9 (26.1)	91.2 (26.4)	87.6 (24.8)	−3.6 (−13.7, 6.4)	−6.8 (−15.9, 1.4)	84.6 (29.2)	91.7 (27.6)	7.2 (−4.8, 19.1)	2.8 (−7.3, 12.8)
HOME[9]										
Total score	21.8 (4.5)	20.0 (5.0)					24.7 (4.8)	22.2 (5.1)	−2.5 (−5.4, 0.5)	−1.2 (−3.7, 1.3)
Responsivity	5.8 (2.1)	5.3 (2.6)					7.0 (2.1)	6.8 (2.2)	−0.1 (−1.4, 1.1)	−0.0 (−1.3, 1.2)
Acceptance	5.3 (1.1)	5.5 (0.8)					5.5 (0.9)	5.5 (0.7)	0.0 (−0.5, 0.5)	0.0 (−0.5, 0.5)
Organization	3.6 (1.1)	3.9 (1.1)					4.6 (1.0)	3.7 (1.3)	−0.9 (−1.6, −0.2)	−1.0 (−1.6, −0.4)
Learning material	2.8 (2.2)	1.7 (1.6)					2.6 (2.2)	1.7 (1.3)	−0.9 (−1.9, 0.2)	−0.3 (−1.3, 0.7)
Involvement	2.9 (1.1)	2.7 (1.2)					3.3 (1.3)	3.2 (1.2)	−0.1 (−0.9, 0.6)	0.0 (−0.7, 0.7)
Variety	1.3 (1.4)	0.9 (0.9)					1.7 (1.0)	1.3 (1.0)	−0.4 (−1.0, 0.2)	−0.3 (−0.9, 0.3)

PEDI, pediatric evaluation of disability inventory; HC, head circumference; MUAC, mid-upper arm circumference; SRQ, self-report questionnaire; MIRI, maternal-infant responsiveness index; PSI, parent stress index; HOME, home observation for the measurement of the environment. [1]Baby Ubuntu vs. standard care (reference group). [2]Adjusted for corresponding outcome assessed at baseline (before randomization). [3]PEDI missing for 5 in standard care arm, 6 in Baby Ubuntu arm at 6 months and missing for 4 in standard care arm, 2 in Baby Ubuntu arm at 12 months. [4]Griffiths sub-quotient scores missing for 3 in standard care arm, 2 in Baby Ubuntu arm at baseline and missing for 4 in standard care arm and 4 in Baby Ubuntu arm at 6 months. [5]Anthropometry missing for 1 in Baby Ubuntu arm at 12 months, z-scores below −6 were imputed to have value −6. [6]SRQ missing for 5 in standard care arm, 7 in Baby Ubuntu arm at 6 months and missing for 1 in standard care arm at 12 months. [7]MIRI missing for 9 in standard care, 11 in Baby Ubuntu arm at 6 months and missing for 3 in standard care arm, 3 in Baby Ubuntu arm at 12 months. [8]PSI missing for 5 in standard care arm, 2 in Baby Ubuntu arm at 6 months and missing for 4 in standard care arm, 7 in Baby Ubuntu arm at 12 months. [9]HOME done for 27 at baseline and 24 at 12 months in standard care, done for 23 at baseline and 22 at 12 months in Baby Ubuntu arm, not done at 6 months.

evidence of impact for children, caregivers and healthcare workers. Key reported impacts included improved perceptions and attitudes toward the ability of children with disabilities, caregiver psychosocial and emotional wellbeing, child function and wellbeing, confidence in child care, peer-support and information sharing. Themes and sub-themes from the qualitative analysis on impact are presented in Supplementary Table 1.

Impact for children

The most reported positive impact for children, particularly in the urban site, was greater inclusion in everyday life at both

family and community level. Prior to the program, caregivers reported societal exclusion, for example, being cast out by their families, children denied clan names, and HCWs refusing to treat children. Following the program, caregivers reported being encouraged by their family to take their child to hospital, HCWs explaining their treatment plan, and being visited by and invited to eat with members of the community. One of the mothers explained that when she learnt the importance of play and peers during a group session, she approached her neighbors, explained her son's condition and invited their children to play with her son. This improved her son's social skills and facilitated his acceptance by community members. Another mother said: "*When my child goes to play at the neighbors, they no longer chase him [after they] explained to them the causes of the condition*."

Most female caregivers receiving the program reported improvements in their child's health, development and function. One mother of a child with hydrocephalus said, "*Whenever you are trying to play with him, he extends the hand to you, he is able to turn himself and lie on his stomach, when you try to cover him from direct sunlight, he pulls off the cloth, he eats very well, he is able to sit when I help him lift his head. . . he is ever happy*."

Similarly, 7 of the 9 fathers interviewed reported seeing a positive impact of the program on their child, including their child's growth, energy levels, and motor and language development: "*Now you can see that what she was unable to do in the past, she can now do*." However, two of the fathers reported seeing no impact; one said "*I have not seen any difference or anything she has gotten from the program maybe apart from the transport reimbursement and the questions she is asked whenever she comes to the hospital for a review*."

In the intervention group, caregivers were supported in accessing routine child health services, and were referred to relevant specialist services. Due to the care and knowledge shared, HCWs reported that children avoided recurring health issues that may have led to secondary disabilities. Caregivers reported that they were better able to identify when their child had health issues and when to seek care appropriately: "*My child does not talk but from what I was taught, I know when he is hungry, sick or about to get sick and I don't wait. . . I either call (the Baby Ubuntu coordinator) or go to our clinic*."

Impact on caregivers

Most caregivers reported positive impacts on their psychosocial and physical wellbeing, peer support and advocacy. Many reported feeling "love" and "hope" for the first time since their child's disability had become apparent. For many, however, there was a diminishing of perceived positive effects over time, particularly in emotional wellbeing.

Prior to the program, caregivers reported negative emotions and physical symptoms such as anger, fatigue, and headaches. For example, one of the rural site female caregivers said, "*My child cries day and night. I find it difficult to concentrate and do some work because every time I am carrying the child. . . I feel like abandoning it to his family because even his father stopped supporting us. . .*(she broke down into tears and as she cleaned her face, she added), *I don't know what to do. . .*" Caregiver attitudes toward disability in the intervention arm changed and they became more resilient and hopeful with reported reductions in stress, isolation, and self-stigma. After the program, each of the caregivers reported a positive change in their own psychosocial wellbeing. They reported that the program increased their understanding of their child's condition and facilitated acceptance of their child, restoring hope and happiness.

Benefits identified by caregivers included a clear understanding of their child's condition. Witnessing an improvement in their child's development gave them hope and encouraged caregivers to return to the groups; though conversely a lack of developmental progression left some caregivers disheartened.

Caregivers reported becoming increasingly confident in caring for their children and becoming "child disability champions." They particularly valued the peer support aspects of the program including sharing information and supporting one another, as well as other families with affected children not participating in the study. One of the mothers said:

> "*I preach the gospel everywhere I go, taxis, markets, shops, etc. because I was taught. I tell them that 'omwana yakoowa' (child born with birth asphyxia), I tell people a lot about the condition if they ask. I also normally tell people with such children to take them for physiotherapy. I have got many friends through this and I am no longer despised because of my child's condition. People keep coming to me, especially young girls who are pregnant, and I also advise them to go to hospital early. I no longer feel ashamed*."

Female caregivers from the control arm mentioned that although they had a place to take their children for treatment, they did not receive much help, and this was evident from their emotional states during interviews. However, through social networks within communities, a few of the control arm mothers had been linked to rehabilitation centers and thereafter had noticed some positive changes in their children.

Impact on healthcare workers

Healthcare workers reported positive changes in their perception of child disability and ability to support affected children and families; this increased their hope and motivation. They reported improvement in the quality of their service delivery due to enhanced skills and knowledge, including a better understanding of referral pathways and resources. This contrasted with prior to the program, particularly in the rural setting, a lack of understanding about child disability reported by HCWs which negatively affected their clinical management, communication with caregivers, and timely referral to specialist

services. One HCW said, "*Children at risk are now given timely and proper care unlike before. . .*" Another stated, "*Our attitude, and that of the caregivers toward these babies, has changed. We no longer view them as useless babies because we have seen most of them achieve. . .*"

Awareness of the etiology also increased; a midwife said in a FGD, "*Being a midwife, I am very keen now and supportive to mothers during labor. I knew the effects of asphyxia even before this intervention, but knowing did not call for any action. Being part of this program has provoked me to take action and I have made initiatives to talk to my fellow midwives about the dangers of asphyxia and how it can be prevented.*" HCWs also appreciated that treating children with disabilities requires collaboration within the multi-disciplinary team, promoting teamwork and a sense of working together to achieve common goals. However, some of the HCWs reported that the increased referrals from the community led to a marked increase in their workload. Whilst this in part related directly to clinical care, they also found that in the absence of social workers it became their responsibility to provide psychosocial support to caregivers which substantially increased their burden.

Impact on wider family and community

Prior to the program, all caregiver participants reported that they received little or no support, and they attributed this to stigma around child disability and high levels of discrimination. Post program, female caregivers reported receiving increased social support from medical teams, community members and families. They frequently attributed this to increased community awareness around child disability facilitating support systems at community level. Caregivers described the program as demystifying community superstitions that their child's condition was a curse or contagious; one female caregiver said "*Ever since my husband interacted with the (program team) my relationship with him improved. He even asks me whether I have done certain roles like feeding him. . .it seems he knows what to do now.*"

However, there were still reports of low levels of support from some extended family members, and continued stigma particularly amongst the caregivers whose child's progress was slower. Whilst female caregivers were generally positive regarding the impact on the wider family and community, none of the fathers interviewed mentioned this.

Scalability

Themes and sub-themes from qualitative data analysis on scale-up are shown in **Supplementary Table 1**.

Facilitators to scale-up of the program were identified during interviews with caregivers and during the stakeholders' workshop. The key facilitator mentioned by most of the participants was the relevance of the program to potential users. They reported that the program was suitable for their needs and the demand for similar services in communities was high. However, several mentioned the importance of increasing awareness in the community through community meeting and radio/television programs, in addition to peer-to-peer communication. One female caregiver said"*. . .teaching about this condition on television and radio (is important) because there are people still hiding their children away in the house not wanting others to see them. . .and it is difficult to sensitize her about the child's condition and the available solution.*" Community engagement was seen as key and a strong driver to successful scale-up. Caregivers mentioned that engaging fathers, religious leaders and traditional healers as advocates influenced acceptability and therefore scale-up. They gave examples of the strong existing beliefs and authority of these community members, and the importance of collaboration to enable access.

> "*When I had just got this child, I was advised to go to different powerful people. I went to priests, pastors, witch doctors and old women and they had their own explanations. The pastors were telling me it was a curse, the witch doctors and elders were telling me it was something to do with clan spirits and though all of them gave me what to use, none of it worked until I came here.*"–Female Caregiver

On the same note, HCWs gave examples of existing service providers and disabled people's groups to collaborate with for capacity building and service delivery, to improve cost-effectiveness and sustainability. Bringing services closer to people was felt to increase access, use and involvement. Most families were unable to access intervention services due to distance, and HCWs' limited understanding of the needs of families at the community level. They felt that integrated community services would increase fathers' engagement, acceptance, peer-to-peer support and timely intervention seeking.

> "*. . .. if the program can be taken out of the hospitals to the communities just like it is for HIV. This is because most of the people in communities don't know that our children are in this condition because they were born tired, they think it is due to supernatural powers like witchcrafts, sacred oracles and so keep demoralizing us and our husbands from giving us support.*"–Female caregiver

Poverty of participants and lack of finances for transport also limited attendance. One female caregiver said, "*Most of the families are poor and stay deep in villages so, raising money to take their children for treatment on a regular basis may be difficult.*" Limited resources and capacity of healthcare services, with rigid systems/teams and lack of political were also considered important barriers to scale.

Government financial and political investment, at local, regional and national levels, was identified as important for scalability and sustainability at the stakeholder workshop.

TABLE 3 Program provider economic costs in 2022 US Dollars (USD) (n = 56).

Cost category	Type of cost in 2022 USD		
	Cost	% of Total cost	Cost per participant
Set-up costs			
Equipment and furniture	511.15	29.38%	9.13
Training and pre-trial expenses[1]	1,228.48	70.62%	21.94
Total set-up costs	**1,739.62**	**11.81%**	**31.06**
Running costs			
Staff[2]	3,971.67	30.57%	70.92
Building costs[3]	2,426.80	18.68%	43.34
Refreshments	416.24	2.83%	7.43
Office supplies	346.46	2.35%	6.19
Airtime	740.23	5.02%	13.22
Transport refund for participants	3,982.02	30.65%	71.11
Transport costs for facilitators	79.67	0.61%	1.42
Home visits[4]	1,029.55	7.92%	18.38
Total running costs	**12,992.64**	**88.19%**	**232.01**
Total cost	**14,732.26**		**263.07**

[1]Includes costs of formatting and printing the education guides, toys and mats for children, and training of implementors. Costs of development and piloting of education guide were excluded. [2]Includes proportion of gross salary (inclusive of net pay + National Social Security Fund + Pay as You Earn) multiplied by time allocated to project implementation. [3]The cost of the square footage of hospital spaces used for study activities as a fraction of the total facility cost multiplied by the number of hours of use for education sessions. [4]Inclusive of both staff time and transport costs, as these were compensated at a fixed rate per participant.

Furthermore, stakeholders felt that involvement of non-governmental organizations operating in the region could help roll-out and scale-up of the program. Stakeholders also mentioned development of a "train the trainers" program as key in facilitating scale-up of Baby Ubuntu master trainers.

Costs of the program

The total costs of setting up and running the program from a provider perspective were USD 14,732.26. The running costs of the program per participant were $232.01 if transport reimbursement for participants was included, and $160.90 if excluded. The total setup cost for the program was $1,739.62, equivalent to a per participant setup cost of $31.06 (Table 3).

Discussion

Baby Ubuntu is a community-based, participatory group rehabilitation program, co-facilitated by healthcare workers and expert parents, that aims to provide an affordable solution to providing early care and support for young children with developmental disability and their caregivers and promote access to child health services and early education. In this pilot feasibility trial in Uganda, with high levels of identified need, Baby Ubuntu was found to be feasible and acceptable in both urban and rural settings. Whilst our mixed methods evaluation provided qualitative evidence of impact on family knowledge, skills, and attitudes, quantitative evaluation of family impact quality of life was inconclusive amongst this population of children with severe developmental impairments. Important programmatic barriers included stigma and exclusion, poverty, and the need to manage expectations around the child's progress. Facilitating factors for scale included community level engagement and sensitization around child disability, and the need to embed the program within existing community health systems. Limited capacity of already overstretched existing healthcare systems was also identified as a challenge. The provider cost estimate represents a feasible intervention for this vulnerable group, encouraging financial sustainability at scale.

Quantitative and qualitative findings supported the program's feasibility and acceptability to most participants, facilitators and healthcare workers. Important enablers identified were peer-support from fellow caregivers, and respectful care toward children and their caregivers by program facilitators, which strongly facilitated attendance. Participatory content was reported to support caregivers in understanding their child's lived experience and needs, relevant not only to meeting immediate care needs, but also accessing routine child health services and early education from which they had been frequently turned away (13, 43, 44). In our study, high levels of self- and community-level stigma were experienced by caregivers, in accordance with our previous research (4, 45, 46), presenting a substantial barrier to accessing services, including healthcare, rehabilitative services and early education. This highlights the need for interventions to consider community-level enablers and barriers and to broaden the programmatic focus from the child to include the wider family and community. Specifically community awareness around childhood disability was seen as crucial in combatting superstition, discrimination and exclusion (47). Early identification of eligible families was strongly facilitated by local community champions, with stakeholders clearly identifying the need for integration with current established community healthcare systems to support early identification of those most in need.

Barriers to attendance included poverty, geographical challenges (distance traveled) and lack of engagement of male caregivers. The effects of poverty on a child's development and education readiness has been found to be highest in the earliest years, mediated through several factors including the home environment (13). Given the strong influence of the home on young children's learning and development, the lack of ability of low-income families to modify the effects of poverty may inhibit access to both healthcare and early education. Traveling long-distances to meet with their group was stated as a barrier by many, despite provided transport reimbursements. Increasing program reach and coverage would likely mitigate

this by reducing the geographical coverage of individual groups. Whilst there is a tendency for health and education programs to focus on supporting female caregivers, who frequently shoulder the greatest caregiving responsibility, paternal engagement is clearly key and has been associated with a number of positive development and education outcomes (13). In our study, fathers were frequently considered gatekeepers of access to groups, health services and, by extension, early education.

Qualitative findings reported positive impacts on a range of child and caregiver outcomes, including perception of the child's health and abilities, however this did not clearly translate to quantitative measures of neurocognitive functioning and quality of life. Whilst intensive therapy interventions for infants with disabilities have been shown to have positive impact on early child functioning (22, 48), it is recognized that this is more challenging in those with the severest of NDIs as seen in the Baby Ubuntu trial cohort. Tools for measuring outcomes lack standardization and validation in LMICs (14), and may not have been sensitive or specific enough to detect a change in our cohort. Positive qualitative experiences reported by caregivers may reflect psychological bias from the belief in the intervention, peer support and motivation. It is possible that the early identification of children at a young age, when the extent of their developmental disability is just becoming apparent, may also undermine the ability for impact on quality of life as families come to terms with their child's condition. Poverty was also a commonly identified barrier to both feasibility and impact, meaning that whilst improvements in knowledge, skills and attitudes were valued, they did not always translate to an improvement in quality of life at family level when poverty still remained. It is also possible, that a longer period of administration (dose dependence) is required to have a positive and holistic impact on the child, caregiver and family. These may explain the significant improvement in family quality of life seen in pre- and post-evaluations, in previous non-controlled studies (15, 34).

The socio-emotional and psychological impact of caring for a child with early child developmental disability on caregivers was clearly reported. Many female caregivers showed physical signs of psychosocial distress during baseline interviews however described the program as transformational in their attitude and behaviors, leading to a more accepting and loving parenting approach. The peer support of others, themselves caregivers of children with disability, was described as key in reducing feelings of isolation, suggesting the important contribution of this to both acceptability and impact. The environment provided in the home, indicated by caregiver engagement in learning activities such as play, is considered to be a strong characteristic of developmental and educational readiness of families (13).

With regards to factors important for scale-up, stakeholders reported a high level of need and demand in the community for early care and support parenting interventions. Community sensitization and engagements were identified as crucial in successful implementation and impact of the program. Stakeholders reported community acceptability attributed to the relevance of the intervention, and of the participatory approach of delivery. In particular, this enabled acceptability and buy-in that can be leveraged to enable both scale-up and sustainability. Participatory approaches have been demonstrated elsewhere to promote smooth integration and scale up of interventions in public health and education (49). Community engagement promoted ownership, support and advocacy for affected families, and enabled smooth implementation, but also highlighted the need to embed the program within existing community health systems. The main barrier to implementation at scale identified by stakeholders, was the limited capacity of already overstretched existing healthcare systems. This has been identified previously in an evaluation of human resources across several early child development programs (50).

The reported provider costs associated with delivering the program add to the evidence of feasibility. The majority of costs were running costs, largely related to workforce and transportation reimbursement for participants. In this research study, transportation costs were largely provider allocated, however this would be less likely if integrated with community health services at scale. Increased program reach and coverage should however reduce this cost through closer geographically located groups. In addition, there may be additional economies at scale, when implementation is streamlined and potentially more efficient (51), particularly when integrated into routine community health services (52). Overall, there is a relative paucity of costing data on parenting interventions for children with disabilities in LIC settings. One cost-effectiveness analysis of a community-level intervention for children and adults with disabilities in Nepal reported a cost per participant of 630 Euros (53). However, this study included both adults and children with a wide range of disabilities and followed a community-based rehabilitation model including health, education, livelihoods, social context and empowerment, and thus may not be directly comparable.

Strengths and limitations

Our Baby Ubuntu feasibility trial reports some of the first evidence from sub-Saharan Africa on feasibility and acceptability of parenting interventions for young children with developmental disabilities. Whilst a comprehensive systematic review of parenting interventions for children during the first 3 years of life supported impact on early child development outcomes across all income settings, programs targeting children with developmental disabilities were not included (54). Given that the trial evaluated a participatory, facilitated group intervention, it was not possible for program facilitators and all research staff to be blind to allocation. However, to

mitigate any bias in reporting outcome measures, study staff performing endline assessments were blind to allocation arm and all other clinical data, with staff from the urban site conducting the rural site assessments and vice versa. It was challenging to protect against the community-level component of the intervention contaminating the control arm in this individually randomized trial, and there were known incidences that intervention arm caregivers shared program content with those from the control arm. It is possible that this undermined the ability to observe differences in outcomes between the arms. A cluster-randomized designed trial is planned for more rigorous evaluation of process, impact, and cost-effectiveness of this complex intervention. It is also possible that the outcome measure tools themselves were limited in their ability to detect change through lack of validation in the LIC setting or through difficulties with accurate translation and interpretation. All attempts were made to mitigate this risk, including choosing validated tools where possible, and those that have been used previously in LIC studies, ideally studies relating to child disability.

Conclusion

The Baby Ubuntu program aims to provide an affordable solution to early care and support for children with developmental disability and their families. Our feasibility trial found the Baby Ubuntu program to be feasible and acceptable to families in both urban and rural settings in Uganda. This mixed methods evaluation provided strong qualitative evidence of impact on family knowledge, skills, attitudes and quality of life, however this was less clear on quantitative evaluation. Facilitating factors for scale included community level engagement and sensitization around child disability and the need to embed the program within existing community health systems. Important barriers included stigma, poverty, limited capacity of existing healthcare systems and highlighted the need to manage expectations around the child's progress. The cost estimate represents a feasible intervention for this vulnerable group, encouraging financial sustainability at scale. Planned work includes integration of Baby Ubuntu within government community health systems in Uganda and Rwanda, development of program modules targeting pre-school readiness and livelihood support, and a cluster-randomized trial and mixed-methods evaluation of process, impact and cost-effectiveness at scale.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by London School of Hygiene & Tropical Medicine; MRC/UVRI and LSHTM Uganda Research Unit; Mulago Hospital; Kiwoko Hospital; Uganda National Council for Science and Technology; Uganda President's Office. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

CT conceived and designed the feasibility trial with substantial contribution from MN, DL, JN, EW, CM, JS, KK, and FC. MN, CN, MZ, JN, BM, DK, SS, RN, FC, AM, and CT developed trial methodology. CN, MK-L, SS, RN, JN, AM, KK, CO, and MN conducted the data collection. EW, RN, and CT conducted the data analysis. KK and GG designed the cost analysis. KK conducted data collection. ET and KK conducted data analysis under the guidance of GG. CT, MN, SS, MK-L, CN and ET were written the first version of the manuscript. All authors contributed to the final version of the manuscript.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2022.981976/full#supplementary-material

SUPPLEMENTARY FIGURE 1
Standard care referral pathways for children with developmental concerns/disability at Mulago Hospital (urban site) and Kiwoko Hospital (rural site).

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Global and regional prevalence of disabilities among children and adolescents: Analysis of findings from global health databases

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Objective: The United Nations' Sustainable Development Goals (SDGs) require population-based data on children with disabilities to inform global policies and intervention programs. We set out to compare the prevalence estimates of disabilities among children and adolescents younger than 20 years as reported by the world's leading organizations for global health statistics.

Methods: We purposively searched the disability reports and databases of the United Nations Children's Fund (UNICEF), the World Health Organization (WHO), the World Bank and the Global Burden of Diseases (GBD) Study. We analyzed the latest disability data reported by these organizations since 2015. We examined the methodologies adopted in generating the reported prevalence estimates and evaluated the degree of agreement among the data sources using Welch's test of statistical difference, and the two one-sided *t*-test (TOST) for statistical equivalence.

Results: Only UNICEF and GBD provided the most comprehensive prevalence estimates of disabilities in children and adolescents. Globally, UNICEF estimated that 28.9 million (4.3%) children aged 0–4 years, 207.4 million (12.5%) children aged 5–17 years and 236.4 million (10.1%) children aged 0–17 years have moderate-to-severe disabilities based on household surveys of child functional status. Using the UNICEF estimated prevalence of 10.1%, approximately 266 million children aged 0–19 years are expected to have moderate-to-severe disabilities. In contrast, GBD 2019 estimated that 49.8 million (7.5%) children aged under 5 years, 241.5 million (12.6%) children aged 5–19 years and 291.3 million (11.3%) children younger than 20 years have mild-to-severe disabilities. In both databases, Sub-Saharan Africa and South Asia accounted for more than half of children with disabilities. A comparison of the UNICEF and GBD estimates showed that the overall mean prevalence estimates for children under 5 years were statistically different and not statistically equivalent

based on ±3 percentage-point margin. However, the prevalence estimates for children 5–19 years and <20 years were not statistically different and were statistically equivalent.

Conclusion: Prevalence estimates of disabilities among children and adolescents generated using either functional approach or statistical modeling appear to be comparable and complementary. Improved alignment of the age-groups, thresholds of disability and the estimation process across databases, particularly among children under 5 years should be considered. Children and adolescents with disabilities will be well-served by a variety of complementary data sources to optimize their health and well-being as envisioned in the SDGs.

KEYWORDS

developmental disabilities, functional impairments, global health, Global Burden of Disease, statistical modeling, low-income and middle-income countries, SDGs, ICF

Introduction

The disability-inclusive provisions in the Sustainable Development Goals (SDGs) require policy interventions to address the needs of children with disabilities and bridge the inequalities that exist between children with and without disabilities (1). However, unlike child mortality which has improved substantially since 2000 (2), reliable global estimates of children with disabilities have been lacking, a situation that has often been misconstrued as evidence that disability is not an important or a serious enough public or global health issue (3). For many years, the absence of consensus on the definition and measurement of disability to facilitate comparable data cross-nationally has been a major challenge in generating the necessary estimates (4). Estimates generated traditionally from systematic reviews and meta-analyses of specific disabilities are often unusable to justify global initiatives because of substantial variations in the quality and methodologies of the underlying studies including the poor representation of high-burden populations from low- and middle-income countries (5, 6). These reservations have accounted for the growing reliance by policymakers on alternative approaches and sources of global estimation of population health metrics including household surveys and statistical modeling (6).

In 2001, the World Health Organization (WHO) launched the International Classification of Functioning, Disability and Health (ICF) to standardize the evaluation of disabilities over the life course (7). In the same year, the Washington Group on Disability Statistics (Washington Group) was commissioned under the auspices of the United Nations (UN) to develop suitable disability measures that will facilitate comparable disability data within the ICF framework (8). In 2006, the Convention on the Rights of Persons with Disabilities (CRPD) provided an operational definition for children with disabilities as "children 18 years or younger who have 'long-term physical, mental, intellectual, or sensory impairments which in interaction with various barriers may hinder their full and effective participation in society on an equal basis with others." (9). In the same year, some 150 million children were estimated by the United Nations Children's Fund (UNICEF) to have disabilities (10). However, no details were provided on how this estimate was generated, and the age range of children included.

In the first World Health Report on Disability published by WHO in 2011, 93 million children (0–15 years) were estimated to have a moderate-to-severe disability, and 13 million had a severe disability (11). These estimates were based on statistical modeling of limited data sources by the Global Burden of Diseases, Injuries, and Risk Factors Study (GBD) 2004, and they excluded children with mild but functionally disabling impairments which was inconsistent with the ICF framework (7). Additionally, the proportion of children under 5 years with disabilities, who were likely to benefit most from early childhood intervention services, was not reported. While these WHO estimates were reported with reservation by UNICEF (12), they were widely cited in the literature and by several UN agencies until an update was published in 2020 based on the GBD 2017 data (13).

Several provisions of the SDGs for disability issues, especially for inclusive education (SDG 4), now make it imperative to generate estimates of children with disabilities (1). These provisions are reinforced by the urgent need to address the disturbing disparities between the global trends in mortality and morbidity among children and adolescents since 2000 (14, 15). This article, therefore, set out to analyze the global and regional estimates of children and adolescents younger than 20 years with disabilities published in global health databases since the launch of the SDGs in 2015.

Methods

Data sources and approaches to disability estimation

For this study, we purposively searched disability reports and databases of UNICEF, WHO, the World Bank and the GBD produced by the Institute for Health Metrics and Evaluation (IHME), as these are presently the leading sources of population-based data for research and policy decisions in global health. The methodological approaches used by these databases were examined to provide context for the reported prevalence estimates of disabilities in children and adolescents. For the remainder of this study, the term "children with disabilities" refers to "children and adolescents with disabilities" below the age of 20 years, except otherwise stated. We relied entirely on the data available to the public and did not contact the organizations for any additional information. A summary of the key features of the available data sources is presented in Table 1.

UNICEF Disability Report 2022

In 2016, UNICEF in partnership with the Washington Group developed a Child Functioning Module (CFM) for inclusion in its routine Multiple Indicator Cluster Survey (MICS) implemented worldwide (16, 17). The CFM appears to conform largely with the biopsychosocial model of disability, by focusing on the presence and extent of functional difficulties rather than on body structure or conditions. It consists of two questionnaires, one with 16 questions for children aged 2–4 years and the other with 24 questions for children aged 5–7 years. The questionnaires are designed to assess functional difficulties in 8 developmental domains of hearing, vision, mobility, fine motor, communication/comprehension, emotions, learning, and playing; and are administered to mothers and primary care givers of eligible children. Responses reflect different levels of severity measured on a 4-level Likert rating scale (0 = no difficulty, 1 = some difficulty, 2 = a lot of difficulty and 3 = cannot do at all). This scale allows the proportion of children with mild difficulties (those who respond "at least some difficulty"), or moderate difficulties (those who respond "a lot of difficulty") or those with severe difficulties (those who respond "cannot do at all") on one or more domain of functioning to be estimated. For reporting purposes, a child with a disability is considered as one with a score level of 3 or 4 in one or more of the 8 functional domains, which meant that children with the mildest degrees of difficulty are excluded. In the UNICEF report first published in 2021, data were collected from 103 data sources (across 43 countries and areas) representing 84 per cent of the world's population of children and at least 50 per cent of population of children in each world region (https://data.unicef.org/resources/children-with-disabilities-report-2021/). Data were first-of-all collected using three different instruments: UNICEF/Washington Group Child Functioning Module, Washington Group Short Set on Functioning and Global Activity Limitation Indicator; and later harmonized (17). After data harmonization, and due to significant variability across countries and regions, a meta-analytical technique was used to estimate the prevalence rates of children with disabilities for each country, 95% confidence intervals (CI) and the child population for all age groups. The estimates for children under 2 years were extrapolated from the estimates computed for children aged 2 to 4 years. It is important to clarify that the results do not provide epidemiological characteristics of any disease or impairment; rather, they provide an indication of the prevalence of moderate-to-severe functional difficulties that, in interaction with various barriers, can place children at increased risk for non-participation and exclusion.

The World Bank and WHO

The WHO and the World Bank Group developed the Model Disability Survey (MDS) tool in 2011 for collecting data on functioning and disability based on ICF framework (18). It is primarily designed as a standalone household survey for adults, with a shorter version to be integrated in health and other population surveys to readily facilitate the continuous monitoring of functioning and disability in a region or a country. There is an optional module for children which uniquely makes additional provision for eliciting information on health conditions, diagnosis, and treatment from the respondents. However, no global or regional estimates of children with disabilities have been published yet from the MDS. In 2020, WHO collaborated with IHME to produce the first-ever estimates of persons who experience a health condition over the course of their life that would benefit from rehabilitation based on the substantive GBD 2019 database (19, 20). The customized database was titled WHO Rehabilitation Need Estimator. A group of experts in the field of rehabilitation was convened by WHO to select specific health conditions in all age groups for which rehabilitation is a key intervention as part of an overall management plan. A total of 25 health conditions were selected for inclusion into this database. The selection for the first time included cerebral palsy as a distinct entity in the GBD database but excluded epilepsy and attention-deficit/hyperactivity disorder which were included in prior reports of children with developmental disabilities and the substantive GBD 2019 database (21, 22). For consistency, we opted to consider the six developmental disabilities reported in the substantive GBD 2019 database (https://vizhub.healthdata.org/gbd-results/). Moreover, this decision allowed the inclusion of all children with developmental disabilities regardless of expert opinion on the need for rehabilitation. The disabilities included are hearing loss, vision loss, developmental intellectual disability, epilepsy, autism spectrum disorders and attention-deficit/hyperactivity disorder. As previously reported, GBD

TABLE 1 Summary of data sources for global estimates of disabilities in children and adolescents.

	UNICEF	WHO-World Bank	IHME	WHO-IHME
Title	Multiple Indicator Cluster Survey (MICS)	Model Disability Survey (MDS)	Global Burden of Disease (GBD)	WHO Rehabilitation Need Estimator
Disability model	Biopsychosocial/ICF	Biopsychosocial/ICF	Medical	Medical
Disability measurement	Parent (or household member)-reported functional difficulties	Parent (or household member)-reported functional difficulties and known impairments	Diagnosis of impairments based on the International Classification of Diseases (ICD) codes	Diagnosis of impairments based on the International Classification of Diseases (ICD) codes
Sources of data input	Household surveys	Household surveys	Systematic reviews of the literature, hospital and claims databases, health surveys, case notification systems, cohort studies, and multinational survey data	Systematic reviews of the literature, hospital and claims databases, health surveys, case notification systems, cohort studies, and multinational survey data Rehabilitation experts
Measurement tool(s)	UNICEF/Washington Group Child Functioning Module, Washington Group Short Set on Functioning, and Global Activity Limitation Indicator	Children version of the Model Disability Survey (MDS) questionnaire	Statistical modelling of sequelae of health conditions	Statistical modelling of sequelae of health conditions
Age group(s)	2–4 years 5–17 years	<5 years 5–12 years 13–17 years	0–19 years	0–19 years
Countries covered	43	Not available	193	193
Included in analysis	Yes	No, data collection on-going	Yes	No. fewer impairments in children reported

UNICEF, United Nations Children's Fund; IHME, Institute for Health Metrics and Evaluation; WHO: World Health Organization.
ICF, International Classification of Functioning, Disability and Health.

estimates of children with developmental intellectual disability include a high proportion of children with cerebral palsy (21). The rehabilitation needs of children younger than 5 years with cerebral palsy and intellectual disability have also been reported previously (23).

GBD 2019 by IHME

The details of the methodologies for the six developmental disabilities selected have been extensively reported (13, 21, 22). In summary, the case definitions and diagnostic criteria were based on the WHO's global standard for diagnostic health information - International Classification of Diseases (ICD) codes (ICD-9 and ICD-10) - complemented with relevant guidelines, such as the Diagnostic and Statistical Manual of Mental Disorders (DSM)-IV-TR and the Guidelines for Epidemiologic Studies on Epilepsy (22). Hearing loss was defined as the quietest sound an individual can hear in their better ear, based on the pure-tone average (PTA) of audiometric thresholds of 0·5, 1, 2 and 4 kHz. Severity levels were classified from mild (PTA from 20 dB) to complete hearing loss (PTA > 95 dB). Vision loss was defined as an impairment resulting from all causes of moderate and worse distance vision loss, visual acuity of <6/18 according to the Snellen chart, and uncorrected presbyopia, or near vision worse than N6 or N8 at 40 cm when best-corrected distance visual acuity was better than 6/12.

Developmental intellectual disability (or "intellectual disability" hereinafter) was defined as a condition of below-average intelligence or mental ability, with multiple severity levels. Severities were defined according to intelligence quotient (IQ) scores, ranging from borderline intellectual disability (IQ 70–85) to profound intellectual disability (IQ 0–19). Epilepsy was defined as an impairment due to idiopathic epilepsy and epilepsy secondary to known infectious and neonatal causes. This definition included cases of active epilepsy with at least one seizure in the previous 5 years, regardless of treatment. Autism spectrum disorders referred to a group of neurodevelopmental disorders with early childhood onset, incorporating disability from pervasive impairment in several areas of development, including social interaction and communication skills, plus restricted and repetitive patterns of

behaviors or interests. Attention-deficit/hyperactivity disorder was defined as an externalizing disorder, incorporating disability from persistent inattention and/or hyperactivity/impulsivity using the DSM-IVTR (314.0, 314.01) and ICD-10 (F90) criteria.

In summary, the prevalence estimation for each condition started with the compilation of all available data inputs from systematic reviews of the literature, hospital and claims databases, health surveys, case notification systems, cohort studies, and multinational survey data. A comprehensive list of the sources of input data for each condition is publicly available at the Global Health Data Exchange (https://ghdx.healthdata.org/gbd-2019/data-input-sources). In the data preparation, efforts were made to i) optimize the comparability of data derived from various sources using different methods; ii) find a consistent set of estimates across prevalence data; and iii) generate estimates for locations with sparse or no data by using available information from other locations combined with covariates.

Prevalence estimates were generated using DisMod-MR 2.1, a statistical modeling technique developed specifically for the GBD project (13, 22). This is a Bayesian meta-regression tool that synthesizes epidemiological data for fatal and non-fatal health outcomes from disparate settings and sources, adjusting for different case definitions/diagnostic criteria or sampling methods, to generate internally consistent estimates by geographical location, year, age group, and sex. An overview of the analytical framework is provided in the Supplementary Figure S1 (13). Sophisticated and validated statistical modeling techniques were used to address sparse and often inconsistent data, especially for diseases, injuries, risk factors and countries for which data were insufficient (22). At every step in the modeling process, the distributions were assessed for sampling error of data inputs, the uncertainty of data corrections for measurement errors, the uncertainty in coefficients from model fit, and the uncertainty of severity distributions. Corresponding uncertainty bounds intervals (UI) for prevalence estimates were defined at the 25th and 975th value of 1,000 draws. The entire GBD process adhered to the Guidelines for Accurate and Transparent Health Estimates Reporting (GATHER), which include recommendations on documentation of data sources, estimation methods, statistical analysis, and statistical code (24).

Statistical analysis

For our analysis, the most recent global and regional prevalence estimates of disabilities were extracted using the World Bank classification: Europe and Central Asia (ECA), East Asia and the Pacific (EAP), Eastern and Southern Africa (ESA), Latin America and the Caribbean (LAC), Middle East and North Africa (MENA), North America (NA), South Asia (SA), and West and Central Africa (WCA). A complete list of countries and areas in the regions and subregions is available at: https://data.unicef.org/regionalclassifications/. We assumed that the age groups of 0–4 years and 5–17 years used by UNICEF are comparable to the GBD age groups of under 5 years and 5–19 years, respectively. The population of children in each group that was used by UNICEF and GBD to estimate the total number of children with disabilities was compared to the official population data provided by the United Nations Population Division for each age group (25). We assessed the degree of agreement between prevalence estimates based on four criteria: statistical difference, statistical equivalence, absolute prevalence difference and prevalence ratio (26–28). Statistical difference was assessed using the Welch's *t*-test to determine the probability that the estimates from both sources are different. Statistical equivalence, which determines whether two estimates are equivalent, was explored using the two one-sided *t*-test of equivalence (TOST) based on a priori ±3 percentage-point margin typically used for comparing prevalence estimates around 10% (27). We sought to determine if the estimates for each age group were (i) statistically different and statistically equivalent, (ii) statistically different and not statistically equivalent, (iii) not statistically different and statistically equivalent, or (iv) not statistically different and not statistically equivalent (28). The absolute and relative differences were also assessed to determine whether the differences were meaningful based on *a priori* goodness-of-fit criteria of 15%, (or 0.85 to 1.15) for prevalence ratio and ≤5 percentage point for the absolute difference (26, 27). All tests of statistical significance were based on critical level of $p < 0.05$. The JAMOVI program for Windows version 2.2.5.0 with TOSTER module were used for analyses, as well as IBM SPSS Statistics for Windows Version 22 where possible for verification.

Results

The disability prevalence estimates reported by UNICEF are presented in Table 2. A total of 28.9 million or 4.3% (95% CI: 4.1–4.6) of children aged 0–4 years, 207.4 million or 12.5% (95% CI: 11.7–13.3) of children aged 5–17 years, and 236.4 million or 10.1% (95% CI: 9.6–10.6) of all children aged 0–17 years were estimated to have moderate-to-severe disabilities globally. Sub-Saharan Africa (29.6% or 69.9 million) and South Asia (27.3% or 64.4 million) accounted for more than half of these children. West and Central Africa accounted for 58.7% (28.9 million) of children with disabilities in Sub-Saharan Africa. Middle East and North Africa recorded the highest prevalence (13.1%) while Europe and Central Asia had the least prevalence (5.5%) of children with disabilities. Children aged 0–4 years accounted for 12.2% of all children with disabilities.

In contrast, the GBD estimated that at least 49.8 million (7.5%) of children under 5 years (Table 3), 241.5 million (12.6%) of children aged 5–19 years (Table 4), and 291.3 million (11.3%) of all children younger than 20 years (Table 5)

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TABLE 2 Global and regional prevalence estimates of disabilities among children younger than 18 years from UNICEF 2022.

Region	Children under aged 0 to 4 years			Children aged 5 to 17 years			Children aged 0 to 17 years		
	%	95% CI	Number of children ('000)	%	95% CI	Number of children ('000)	%	95% CI	Number of children ('000)
North America	4.4	3.9–4.9	943	12.0	11.3–12.7	7,073	9.9	9.5–10.4	8,016
Europe and Central Asia	2.7	2.4–3.1	1,515	6.5	5.6–7.4	9,299	5.5	4.9–6.0	10,814
East Asia and the Pacific	3.5	3.3–3.8	5,333	9.5	7.5–11.6	37,788	7.8	6.7–9.1	43,121
Latin America and the Caribbean	3.8	3.3–4.5	1,978	12.6	11.5–13.7	17,102	10.2	9.6–10.8	19,080
South Asia	3.7	2.9–4.7	6,254	13.0	10.2–16.1	58,177	10.5	9.0–12.2	64,431
Middle East and North Africa	4.5	3.3–6.0	2,246	16.9	13.5–20.5	18,694	13.1	11.3–15.1	20,940
Sub-Saharan Africa	6.0	5.2–7.0	10,648	15.9	13.3–18.6	59,300	12.7	11.2–14.3	69,948
Eastern and Southern Africa	5.2	4.5–6.0	4,509	12.8	11.2–14.4	24,356	10.4	9.5–11.3	28,865
West and Central Africa	6.8	5.8–7.9	6,139	18.9	15.3–22.7	34,944	14.9	12.8–17.2	41,083
Global	4.3	4.1–4.6	28,917	12.5	11.7–13.3	207,433	10.1	9.6–10.6	236,350

UNICEF 2022 (Reference 17).

have mild-to-severe disabilities globally. South Asia (33.8% or 98.5 million) and Sub-Saharan Africa (20.5% or 59.8 million) accounted for more than half of these children. West and Central Africa accounted for 53.2 % (31.7 million) of children with disabilities in Sub-Saharan Africa. The highest prevalence of children with disabilities (13.6%) occurred in South Asia and the least prevalence (8.9%) in Europe and Central Asia. Children under 5 years accounted for 17.1% of all children with disabilities. Among children under 5 years, developmental intellectual disability was most prevalent (3.2%) while attention-deficit/hyperactivity disorder was least prevalent (0.2%). In contrast, among all children, hearing loss was most prevalent (4.0%), while autism spectrum disorders were the least prevalent disabilities (0.4%).

The statistical comparison of the estimates from both UNICEF and GBD is summarized in Table 6. The *t*-tests for the overall mean for each age group only showed statistically significant difference among children under 5 years ($p = 0.003$). At a ±3 percentage point margin, the TOST showed that the estimates from both sources were statistically equivalent, except for children under 5 years ($p = 0.375$). None of the global and regional prevalence ratios for children under 5 years fell within the goodness-of-fit criteria while all absolute differences for the combined category of children under 20 years fell within goodness-of-fit criteria. The goodness-of-fit criteria were met in North America, Latin America and the Caribbean, Sub-Saharan Africa and globally for all age categories except for children under 5 years. The largest absolute difference in estimates globally was recorded among children under 5 years. The regional pattern of the global estimates of children under 5 years with disabilities is also presented in Figure 1. The largest contributor to the difference between both data sources was South Asia where a 6.7 percentage point difference was recorded, and the GBD estimate was almost 3-fold of the estimate by UNICEF. Similar data for the other age groups are presented in Figures 2, 3. The populations of children in each group used by UNICEF and GBD for estimating the total number of children with disabilities globally are summarized in Figure 4.

Discussion

It is important to clarify the significance of the findings on the prevalence estimates reported from different databases against the backdrop of the adverse consequences confronting children with disabilities over the life course (10–12, 17, 29, 30). Globally, the likelihood of a surviving child having a disability is estimated to be at least 10 times higher than that of dying before their fifth birthday (29). When compared to children without disabilities, children with disabilities are 42% less likely to have foundational reading and numeracy skills, 49% more likely to have never attended school, 47% more likely to drop out of primary school and 20% less likely to have expectations of a

TABLE 3 Global and regional prevalence estimates (95% uncertainty intervals) of disabilities among children younger than 5 years from GBD 2019.

Region	Metric	Hearing loss	Vision loss	Epilepsy	Developmental intellectual disability	Autism spectrum disorders	Attention-deficit/hyperactivity disorders	Total*
North America	Number	216680 (178560–253606)	144266 (111242–184772)	116174 (86582–147558)	365935 (288933–444460)	163530 (137322–191935)	88839 (57190–129131)	1,095,424
	Cases per 100,000	1033 (852–1209)	688 (531–881)	554 (413–704)	1745 (1378–2119)	780 (655–915)	424 (273–616)	5,224
Europe and Central Asia	Number	734399 (613799–844935)	442598 (344672–561172)	325486 (254438–405246)	968321 (759685–1177813)	288885 (241844–341950)	129066 (85207–183064)	2,888,755
	Cases per 100,000	1387 (1159–1596)	836 (651–1060)	615 (481–766)	1829 (1435–2224)	546 (457–646)	244 (161–346)	5,457
East Asia and the Pacific	Number	3438113 (2956219–3913264)	1297733 (1038429–1626274)	823451 (609592–1057118)	2727757 (2193412–3288485)	655238 (536229–777688)	494302 (328841–691713)	9,436,594
	Cases per 100,000	2321 (1996–2642)	876 (701–1098)	556 (412–714)	1842 (1481–2220)	443 (362–525)	334 (222–467)	6,372
Latin America and the Caribbean	Number	955072 (815257–1083241)	537396 (425765–677756)	407933 (314902–517147)	920348 (751406–1093649)	235268 (195519–280124)	164888 (109929–235240)	3,220,905
	Cases per 100,000	1810 (1545–2052)	1018 (807–1284)	773 (597–980)	1744 (1424–2072)	446 (371–531)	313 (209–446)	6,104
South Asia	Number	3874622 (3255133–4475660)	1957304 (1557653–2441206)	1125281 (800287–1471321)	10126841 (7607751–12675553)	618664 (507916–741150)	203205 (131076–295087)	17,905,917
	Cases per 100,000	2245 (1886–2593)	1134 (903–1415)	652 (464–853)	5866 (4407–7343)	359 (295–430)	118 (76–171)	10,374
Middle East and North Africa	Number	488204 (402462–571691)	484094 (386408–607876)	349893 (273786–431567)	1495407 (1130592–1865688)	163087 (134699–194489)	93415 (62260–134040)	3,074,100
	Cases per 100,000	1117 (921–1308)	1107 (884–1390)	801 (627–987)	3420 (2586–4266)	373 (308–445)	214 (143–307)	7,032
Sub-Saharan Africa	Number	4430374 (3751757–5105139)	1060410 (858727–1318551)	1281867 (971635–1601732)	4380762 (3367541–5401625)	785503 (649093–939682)	192373 (125070–284836)	12,131,289
	Cases per 100,000	2591 (2194–2985)	620 (503–771)	750 (569–937)	2562 (1969–3159)	460 (380–550)	113 (74–167)	7,096

(Continued)

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TABLE 3 (Continued)

Region	Metric	Hearing loss	Vision loss	Epilepsy	Developmental intellectual disability	Autism spectrum disorders	Attention-deficit/hyperactivity disorders	Total*
Western Sub-Saharan Africa	Number	2056777 (1733992–2379728)	486542 (396448–600924)	535773 (412020–676979)	1569783 (1214410–1929542)	334776 (276395–400912)	83100 (53414–123565)	5,066,751
	Cases per 100,000	2828 (2385–3272)	669 (546–827)	737 (567–931)	2159 (1670–2653)	461 (381–552)	115 (74–170)	6,969
Central Sub-Saharan Africa	Number	476231 (403059–544899)	105598 (83203–135057)	159777 (112730–212478)	569843 (436073–705495)	94749 (77403–113592)	22322 (14378–32171)	1,428,520
	Cases per 100,000	2301 (1948–2633)	511 (402–653)	772 (545–1027)	2753 (2107–3409)	458 (374–549)	108 (70–156)	6,903
Eastern Sub-Saharan Africa	Number	1622747 (1369967–1865589)	358640 (289879–446935)	489671 (362217–625240)	1824540 (1391859–2248468)	299975 (247740–358721)	68924 (44638–101370)	4,664,497
	Cases per 100,000	2531 (2136–2909)	560 (452–697)	764 (565–975)	2845 (2171–3506)	468 (387–560)	108 (70–159)	7,276
Southern Sub-Saharan Africa	Number	214764 (182842–247327)	55604 (44030–69664)	63914 (44593–86373)	154615 (122342–186165)	37204 (30649–44371)	8776 (5687–13128)	534,877
	Cases per 100,000	2653 (2259–3055)	687 (544–861)	790 (551–1067)	1910 (1512–2300)	460 (379–549)	109 (71–163)	6,609
Global	Number	14148322 (12036835–16216298)	5928288 (4749336–7364009)	4433545 (3376788–5567220)	20998409 (16142819–25947466)	2912437 (2418074–3461585)	1367582 (898677–1947054)	49,788,583
	Cases per 100,000	2135 (1816–2447)	895 (717–1111)	669 (510–840)	3168 (2436–3915)	440 (365–523)	207 (136–294)	7,514

*95% uncertainty intervals not available for all disabilities.

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TABLE 4 Global and regional prevalence estimates (95% uncertainty intervals) of disabilities among children aged 5 to 19 years from GBD 2019.

Region	Metric	Hearing loss	Vision loss	Epilepsy	Developmental intellectual disability	Autism spectrum disorders	Attention-deficit/hyperactivity disorders	Total*
North America	Number	1226942 (1069237–1389908)	829331 (673427–1015300)	464116 (377040–566934)	1127243 (881538–1386467)	509875 (428720–599971)	3365797 (2261084–4803878)	7,523,304
	Cases per 100,000	1780 (1551–2016)	1203 (977–1473)	674 (547–823)	1635 (1279–2011)	740 (622–871)	4882 (3280–6967)	10,914
Europe and Central Asia	Number	4583297 (4026175–5148943)	2222942 (1818252–2714517)	1158295 (896858–1486995)	2744266 (2120532–3364352)	838512 (701345–993733)	4550322 (3110708–6365862)	16,097,634
	Cases per 100,000	2843 (2498–3194)	1379 (1128–1684)	719 (557–923)	1703 (1316–2087)	521 (436–617)	2823 (1930–3949)	9,988
East Asia and the Pacific	Number	22128237 (19553458–24940194)	5991479 (4904617–7257251)	2552101 (1987366–3300801)	7356504 (5854718–8964365)	1770957 (1461017–2120794)	15649369 (10849799–21465647)	55,448,647
	Cases per 100,000	5181 (4578–5840)	1403 (1149–1700)	598 (466–773)	1723 (1371–2099)	415 (343–497)	3664 (2541–5026)	12,984
Latin America and the Caribbean	Number	6648994 (5902702–7477938)	2930755 (2386395–3569863)	1438411 (1127718–1831419)	2550241 (2058769–3060952)	670370 (555365–798010)	6064849 (4188888–8551760)	20,303,620
	Cases per 100,000	4153 (3687–4671)	1831 (1491–2230)	899 (705–1144)	1593 (1286–1912)	419 (347–499)	3788 (2617–5342)	12,683
South Asia	Number	28211185 (24273897–32232474)	8138965 (6764205–9851146)	4151288 (3099984–5359019)	30468226 (22914705–38180927)	1834585 (1512061–2209420)	7824749 (5139601–11183238)	80,628,998
	Cases per 100,000	5126 (4410–5856)	1479 (1229–1790)	755 (564–974)	5536 (4163–6937)	334 (275–402)	1422 (934–2032)	14,652
Middle East and North Africa	Number	2813081 (2427661–3208346)	2477027 (2036894–2992981)	957295 (764983–1203938)	4016620 (3022254–5024694)	435499 (359818–520101)	3169552 (2163344–4468032)	13,869,074
	Cases per 100,000	2258 (1949–2575)	1988 (1635–2403)	769 (614–967)	3224 (2426–4033)	350 (289–418)	2544 (1737–3586)	11,133

(Continued)

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TABLE 4 (Continued)

Region	Metric	Hearing loss	Vision loss	Epilepsy	Developmental intellectual disability	Autism spectrum disorders	Attention-deficit/hyperactivity disorders	Total*
Sub-Saharan Africa	Number	22442961 (19306417–25523614)	4073575 (3435394–4846169)	3459890 (2673539–4485332)	9863944 (7536483–12220930)	1807381 (1487757–2159942)	5801510 (3840848–8271417)	47,449,261
	Cases per 100,000	5314 (4571–6043)	965 (814–1148)	820 (633–1062)	2336 (1785–2894)	428 (353–512)	1374 (910–1959)	11,237
Western Sub-Saharan Africa	Number	10039827 (8585704–11437723)	1772251 (1501258–2105376)	1358388 (1023033–1787672)	3372979 (2584303–4197495)	750778 (618519–898133)	2446774 (1607405–3493043)	19,740,997
	Cases per 100,000	5720 (4891–6516)	1010 (856–1200)	774 (583–1019)	1922 (1473–2392)	428 (353–512)	1394 (916–1990)	11,248
Central Sub-Saharan Africa	Number	2440218 (2112922–2752972)	412296 (342047–495657)	448016 (286390–650824)	1279151 (971797–1596102)	215881 (176255–257883)	655630 (431619–945625)	5,451,192
	Cases per 100,000	4836 (4188–5456)	818 (678–983)	888 (568–1290)	2535 (1926–3164)	428 (350–512)	1300 (856–1874)	10,805
Eastern Sub-Saharan Africa	Number	8396926 (7232002–9572424)	1380137 (1157587–1644255)	1370261 (1044622–1785873)	4146836 (3166775–5147128)	696321 (574120–831487)	2096146 (1388361–3004361)	18,086,627
	Cases per 100,000	5269 (4538–6006)	866 (727–1032)	860 (656–1121)	2602 (1987–3230)	437 (361–522)	1316 (872–1885)	11,350
Southern Sub-Saharan Africa	Number	1233551 (1062885–1402959)	241357 (200575–289619)	202537 (153864–262472)	400906 (317513–486844)	96865 (79345–115927)	297468 (195820–427506)	2,472,684
	Cases per 100,000	5479 (4721–6231)	1072 (891–1287)	900 (684–1166)	1781 (1411–2163)	431 (353–515)	1322 (870–1899)	10,985
Global	Number	88121532 (76891578–99618793)	26684718 (21991143–32187072)	14192633 (11172414–18071433)	58160929 (44335927–72217829)	7873281 (6532083–9413240)	46477791 (31750591–64830750)	241,510,884
	Cases per 100,000	4599 (4013–5199)	1393 (1148–1680)	741 (583–943)	3035 (2314–3769)	411 (341–492)	2426 (1657–3383)	12,605

*95% uncertainty intervals not available for all disabilities.

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TABLE 5 Global and regional prevalence estimates (95% uncertainty intervals) of disabilities among children younger than 20 years from GBD 2019.

Region	Metric	Hearing loss	Vision loss	Epilepsy	Developmental intellectual disability	Autism spectrum disorders	Attention-deficit/hyperactivity disorders	Total*
North America	Number	1443622 (1260095–1623672)	973597 (801351–1171707)	580289 (474814–694717)	1493177 (1172810–1827421)	673405 (566292–791760)	3454636 (2317514–4930022)	8,618,726
	Cases per 100,000	1606 (1402–1806)	1083 (892–1303)	646 (528–773)	1661 (1305–2032)	749 (630–881)	3842 (2577–5482)	9,587
Europe and Central Asia	Number	5317696 (4661136–5957133)	2665540 (2220754–3201168)	1483781 (1161312–1851246)	3712587 (2885054–4541304)	1127397 (943188–1336345)	4679388 (3206381–6539742)	18,986,389
	Cases per 100,000	2483 (2177–2782)	1245 (1037–1495)	693 (543–865)	1734 (1347–2121)	527 (441–624)	2185 (1497–3054)	8,867
East Asia and the Pacific	Number	25566349 (22666249–28615270)	7289211 (6079418–8726594)	3375552 (2681974–4248571)	10084260 (8052049–12264190)	2426195 (1995116–2897387)	16143670 (11192831–22148095)	64,885,237
	Cases per 100,000	4445 (3941–4975)	1268 (1057–1517)	587 (467–739)	1753 (1400–2132)	422 (347–504)	2807 (1946–3850)	11,282
Latin America and the Caribbean	Number	7604066 (6768579–8453553)	3468151 (2896484–4159632)	1846344 (1478702–2287072)	3470589 (2818649–4153597)	905637 (753110–1077016)	6229737 (4299528–8776093)	23,524,524
	Cases per 100,000	3572 (3180–3971)	1629 (1361–1954)	868 (695–1075)	1631 (1324–1951)	426 (354–506)	2927 (2020–4123)	11,053
South Asia	Number	32085806 (27728413–36385664)	10096269 (8439115–11984658)	5276568 (4094348–6599348)	40595067 (30539944–50842037)	2453248 (2019630–2962215)	8027954 (5266209–11471660)	98,534,912
	Cases per 100,000	4438 (3835–5032)	1397 (1168–1658)	730 (567–913)	5615 (4224–7032)	340 (280–410)	1111 (729–1587)	13,631
Middle East and North Africa	Number	3301284 (2862269–3746353)	2961120 (2494463–3525529)	1307187 (1063318–1619453)	5512026 (4148436–6892949)	598586 (495243–713797)	3262966 (2224812–4598774)	16,943,169
	Cases per 100,000	1962 (1701–2226)	1760 (1482–2095)	777 (632–963)	3275 (2465–4095)	356 (295–425)	1939 (1322–2732)	10,069

(Continued)

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TABLE 5 (Continued)

Region	Metric	Hearing loss	Vision loss	Epilepsy	Developmental intellectual disability	Autism spectrum disorders	Attention-deficit/hyperactivity disorders	Total*
Sub-Saharan Africa	Number	26873334 (23225530–30370158)	5133984 (4380184–6041603)	4741756 (3782633–5953097)	14244706 (10926006–17570979)	2592883 (2136090–3099711)	5993883 (3966944–8542723)	59,580,546
	Cases per 100,000	4529 (3914–5118)	866 (739–1019)	800 (638–1004)	2401 (1842–2961)	437 (360–523)	1010 (669–1440)	10,043
Western Sub-Saharan Africa	Number	12096603 (10409381–13721117)	2258793 (1943559–2638917)	1894161 (1469697–2416851)	4942762 (3802172–6119971)	1085553 (896738–1299438)	2529873 (1663706–3611220)	24,807,745
	Cases per 100,000	4873 (4193–5527)	910 (783–1063)	763 (592–974)	1991 (1532–2466)	438 (362–524)	1019 (671–1455)	9,994
Central Sub-Saharan Africa	Number	2916448 (2512575–3258254)	517894 (433288–616952)	607792 (401690–844674)	1848993 (1407885–2300062)	310629 (253551–371262)	677951 (445679–977406)	6,879,707
	Cases per 100,000	4099 (3531–4579)	728 (609–867)	855 (565–1187)	2599 (1979–3233)	437 (357–522)	953 (627–1374)	9,671
Eastern Sub-Saharan Africa	Number	10019673 (8666559–11379839)	1738777 (1488881–2044281)	1859931 (1438907–2348494)	5971376 (4562094–7412340)	996295 (821527–1190208)	2165070 (1432587–3105380)	22,751,122
	Cases per 100,000	4483 (3878–5092)	778 (667–915)	833 (644–1051)	2672 (2041–3317)	446 (368–533)	969 (641–1390)	10,181
Southern Sub-Saharan Africa	Number	1448315 (1250897–1636349)	296961 (249538–352908)	266451 (207123–340369)	555520 (438187–673890)	134069 (109930–160120)	306243 (201417–440151)	3,007,559
	Cases per 100,000	4731 (4087–5346)	971 (816–1153)	871 (677–1112)	1815 (1432–2202)	438 (360–524)	1001 (658–1438)	9,827
Global	Number	102269853 (89657165–115064557)	32613006 (27412553–38676284)	18626177 (15136201–23044362)	79159337 (60490508–98168458)	10785718 (8953061–12859912)	47845372 (32634830–66892474)	291,299,463
	Cases per 100,000	3966 (3477–4462)	1265 (1063–1500)	723 (587–894)	3070 (2346–3807)	419 (348–499)	1855 (1266–2594)	11,298

*95% uncertainty intervals not available for all disabilities.

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TABLE 6 Comparison of global and regional prevalence estimates (%) between UNICEF 2022 and GBD 2019.

Region	UNICEF			GBD 2019			Prevalence ratio: Criterion 0.85 to 1.15			Absolute difference: Criterion ±5		
	<5 years	5 to 17 years	<18 years	<5 years	5 to 19 years	<20 years	<5 years	5 to 19 years	<20 years	<5 years	5 to 19 years	<20 years
North America	4.4	12.0	9.9	5.2	10.9	9.6	1.2	**0.9**	**1.0**	**0.8**	**−1.1**	**−0.3**
Europe and Central Asia	2.7	6.5	5.5	5.5	10.0	8.9	2.0	1.5	1.6	**2.8**	**3.5**	**3.4**
East Asia and the Pacific	3.5	9.5	7.8	6.4	13.0	11.3	1.8	1.4	1.4	**2.9**	**3.5**	**3.5**
Latin America and the Caribbean	3.8	12.6	10.2	6.1	12.7	11.1	1.6	**1.0**	**1.1**	**2.3**	**0.1**	**0.9**
South Asia	3.7	13.0	10.5	10.4	14.7	13.6	2.8	**1.1**	1.3	6.7	**1.7**	**3.1**
Middle East and North Africa	4.5	16.9	13.1	7.0	11.1	10.1	1.6	**0.7**	**0.8**	**2.5**	−5.8	**−3.0**
Sub-Saharan Africa	6.0	15.9	12.7	7.1	11.2	10.0	1.2	**0.7**	**0.8**	**1.1**	**−4.7**	**−2.7**
Global	4.3	12.5	10.1	7.5	12.6	11.3	1.7	**1.0**	**1.1**	**3.2**	**0.1**	**1.2**

t-Test for statistical difference in overall mean in each age group: <5 years (t = 4.185, df = 11.4, p = 0.001); 5 to 19 years (t = 0.264, df = 9.71, p = 0.797); < 20 years (t = 0.746, df = 11.37, p = 0.471).
Two one-sided test (TOST) for statistical equivalence of overall mean in each age group, GBD vs UNICEF: <5 years (6.9% vs 4.1%, p = 0.375); 5 to 19 years (12.0 vs. 12.4%, p = 0.0.032); <20 years (10.7 vs. 10.0%, p = 0.024).

better life (17). Available reports also suggest that between 80 and 90% of people with disabilities of working age are likely to be unemployed in low- and middle-income countries compared to between 50 and 70% in high-income countries (30). Given the peculiar challenges often associated with measuring disability across various functional domains (4, 8), our primary goal was to examine the degree of alignment between the reported estimates from data sources that rely on different methodologies with a view to highlighting areas for further consideration.

A key finding in this study is that available prevalence estimates of children with disabilities from UNICEF and GBD appear complementary and emphasize the need for appropriate policy interventions from early childhood. The comparability of the prevalence estimates of disabilities among all children and adolescents as a group, despite the differences in the approaches to estimation is noteworthy. The GBD estimate of all children with mild-to-moderate disabilities exceeded the estimate of moderate-to-severe disabilities from UNICEF by 55 million or 23.2%. This variance can be attributed to several factors. Firstly, the UNICEF estimates excluded children aged 18 and 19 years. The inclusion of these children by GBD is consistent with the adolescent age group used by the UN Population Division (25) and the United Nations Inter-Agency Group for Child Mortality Estimation that comprises the UN, UNICEF, WHO and the World Bank (2). It is unclear why this group of children was excluded in the substantive survey tool designed by the Washington Group that was adopted by UNICEF. Secondly, the population of all children in each group that served as denominator for computing the estimated prevalence differed. For example, the world population of children 0–19 years in 2019 by the UN Population Division was ∼2.6 billion (25), same as the GBD denominator for estimating the prevalence for this age group. In contrast, the population of children aged 0–17 years and 5–17 years used as denominator by UNICEF was 2.3 billion and 1.7 billion, respectively. If the prevalence of 12.5% for children aged 5–17 years reported by UNICEF were applied to the 1.9 billion children aged 5–19 years by UN, the prevalence of disabilities among all children (0–19 years) would have increased to ∼266 million compared to 291 million by GBD 2019. Thirdly, the reported estimates by UNICEF excluded mild disabilities in all age groups. However, mild disabilities are always significantly more prevalent than moderate-to-severe disabilities regardless of the approach to measurement (4). It is understandable that the child functioning module is likely to produce spurious findings as it relies entirely on subjective assessment by respondents. It is, therefore, not unlikely that children who have mild activity limitations might not be reported as having a disability while some children without disability may also be erroneously reported as disabled (31). However, the decision to exclude mild disabilities is inconsistent with the ICF provisions which recognize that the affected children may encounter functional difficulties under different environmental conditions (7). For example, children with

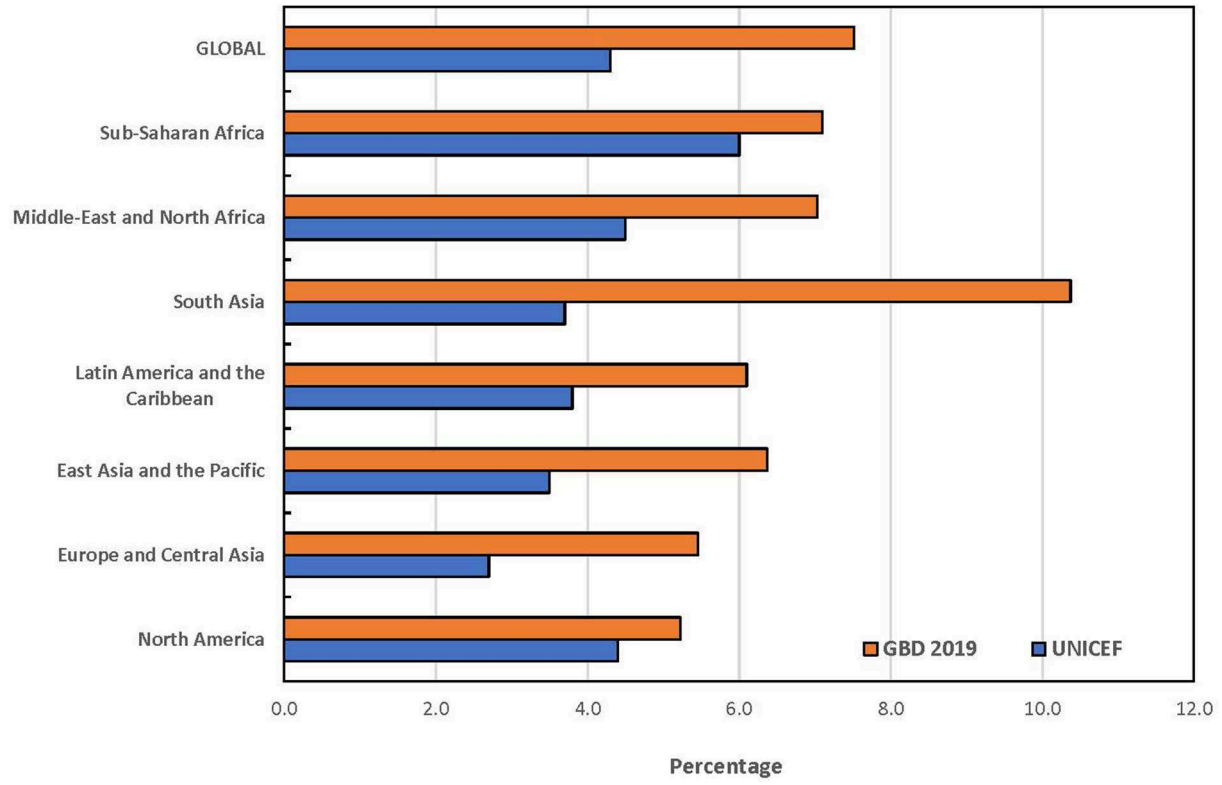

FIGURE 1
Prevalence estimates of disabilities among children under 5 years by UNICEF and GBD 2019.

minimal hearing loss (comprising slight or mild bilateral and unilateral hearing impairments), are frequently associated with adverse effects across different functional domains including speech and language development, academic performance, and social interactions (32, 33).

The significant disparities in the prevalence estimates among children under 5 years also merit clarification because of their special relevance to the subsisting global commitments for early childhood development under the SDGs (4.2.1) for this age group (1, 21, 29). The child functioning module used in country surveys excluded children under 2 years because of the challenges in eliciting functional limitations reliably through parental response. Usually, the effects of some impairments in infants may not be apparent to the parents because they are too young to have developed the ability to carry out activities that are normal for older children. However, UNICEF recognizes that data for this age group is vital and opted to assume that the estimate for children under 2 years could be informed by the estimate for children aged 2 to 4 years in each country (17). However, this imputation does not adequately reflect the evidence on the magnitude of the incidence of neurodevelopmental impairments associated with the perinatal disorders, especially in low- and middle-income countries where perinatal care is poor (34). For example, both UNICEF and GBD agree that Sub-Saharan Africa and South Asia are associated with the poorest maternal and child health complications and remain the largest contributors to disabilities among children globally. Moreover, very limited evidence exists on the validation of the child functioning module among a large sample of children 2–4 years compared to older children in these high burden regions (35, 36). The true global prevalence of children under 5 years with disabilities is therefore likely to be closer to the GBD 2019 estimate of 50 million approximately.

Considering the peculiar challenges in disability measurement, estimates of disabilities using different approaches must necessarily be evaluated within the context of the intended purpose. UNICEF data is aimed at identifying children with functional limitations over a pre-specified range of domains as part of national population censuses and surveys. The UNICEF data also uniquely provide insights into the performance of these children across key indicators of early child development compared to children without disabilities. However, the estimates are not intended to provide information on the diagnostic entities underlying the survey responses based on the available ICD codes. Attempts to use survey responses, for example, as a first stage screening to identify people with clinical impairments, service and assistive product referral needs in four functional domains (vision, hearing, mobility,

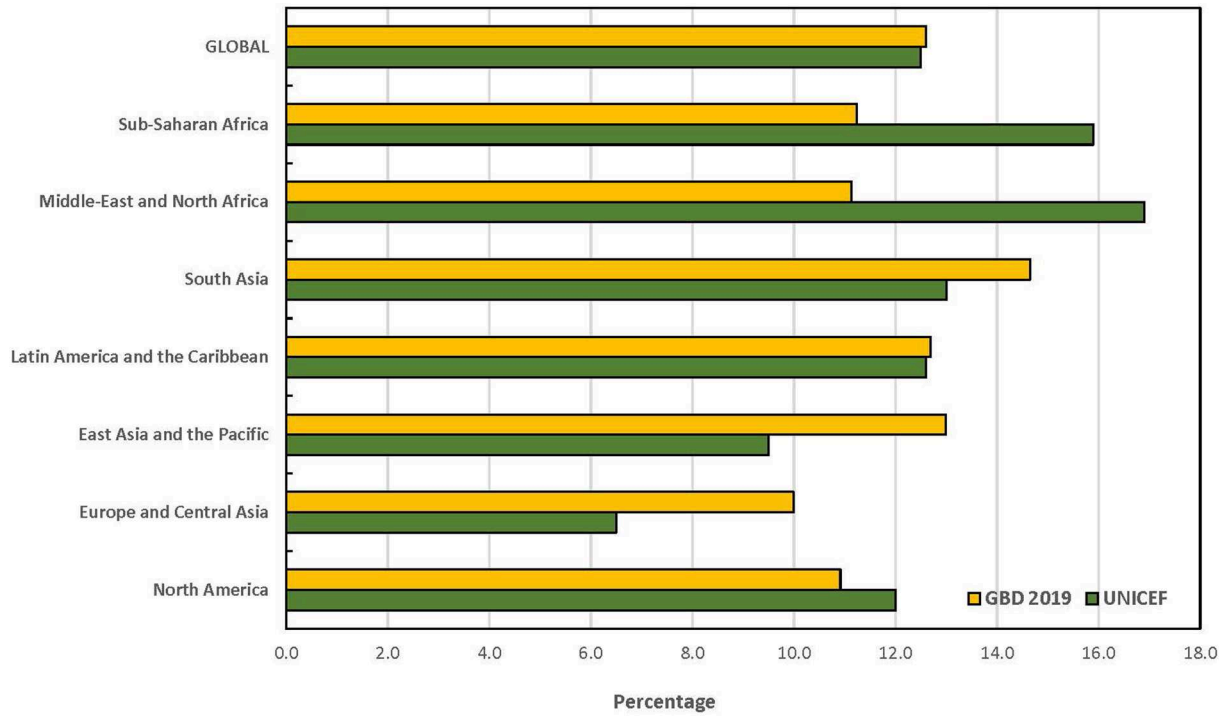

FIGURE 2
Prevalence estimates of disabilities among children aged 5 to 19 years by UNICEF and GBD 2019.

and cognition) have been shown to be associated with less-than-optimal sensitivity and specificity (37). In fact, UNICEF specifically stated that the results should not be used to assess the epidemiological characteristics of any disease or impairment but an indication of the prevalence of moderate to severe functional difficulties that, in interaction with various barriers, can place children at increased risk for non-participation and exclusion (17). In contrast, the GBD primarily sets out to quantify the long-term sequelae associated with diverse health conditions based on ICD codes to inform appropriate interventions (primary, secondary and tertiary prevention) within the healthcare systems. The estimates provide information on the scope, nature and magnitude of the rehabilitation services that are required to support children with specific disabilities. While the GBD estimates do not cover all known disabilities, they are notably consistent with the recognition of specific diagnostic disability entities under the US' Individuals with Disabilities Education Act (IDEA) 2004 (38) and the UK Equality Act 2010 (39). Additionally, the ICF views disability as an umbrella term for impairments, activity limitations, and participation restrictions and denotes the negative aspects of the interaction between an individual (with a health condition) and that individual's contextual factors (environmental and personal factors) (7). The ICF also underscores its complementarity with the ICD diagnostic entities.

Disability measurement is frequently linked with models for conceptualizing disability (17, 40–43). The predominant and oldest model - the medical or biomedical model - defines disability primarily as a medical condition resulting from some physiological impairment that can either be prevented or managed to optimize individual functioning (17, 40, 41). The social model emerged in the 1970's to present disability as not due to an individual pathology but as a failure of the policy, cultural and physical environments to accommodate differences in function (42). Unfortunately, the social model evolved from a narrow and restricted conceptualization of disability beyond physical impairment (41, 42). The biopsychosocial model was later introduced to address the limitations of the medical model in recognizing the psychological, social, and behavioral dimensions of a medical condition (43, 44), and became the focus of the ICF. However, the ICF was never intended to replace the medical model but to enhance it (45). While it may be easier to elicit functional difficulties through household surveys, such responses do not provide a pathway for the effective care of children with disabilities within the health systems (37). In fact, it is difficult to identify children with self-limiting constitutional developmental delays based on survey responses. Any suggestion that these models of disability are mutually exclusive is therefore erroneous, counter-productive, and inconsistent with the ICF principles

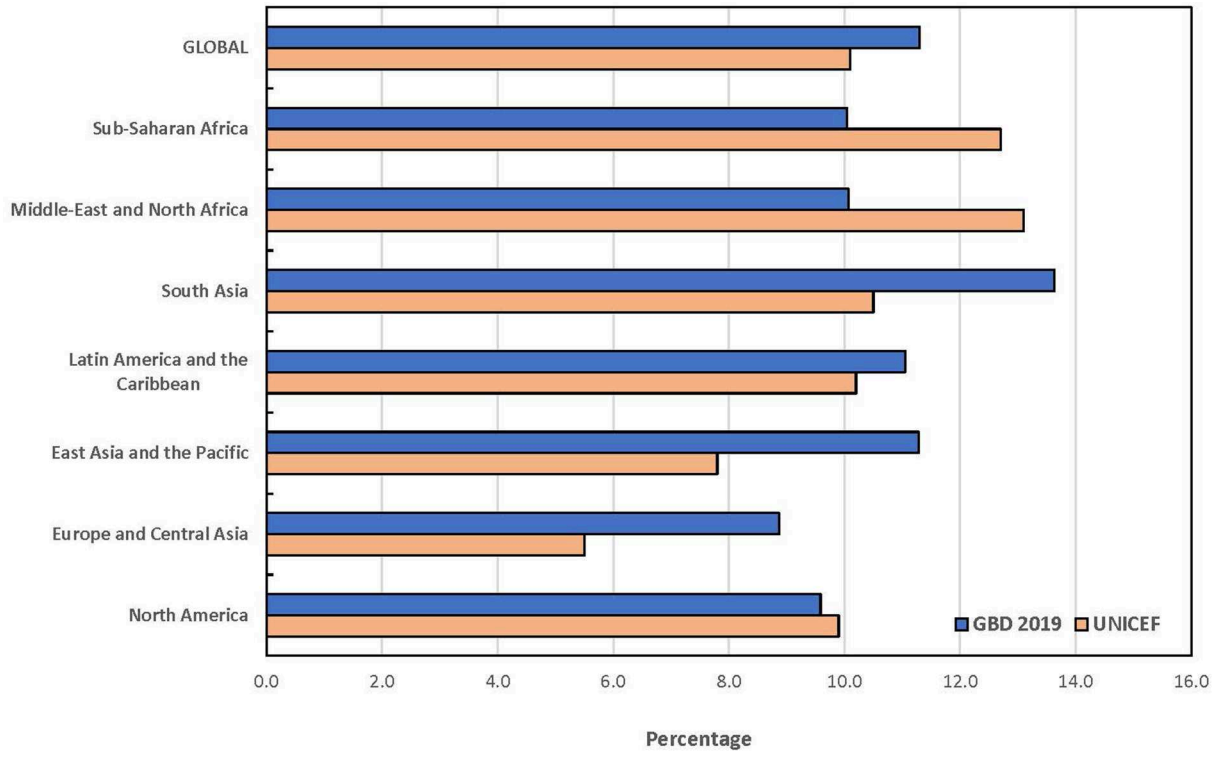

FIGURE 3
Prevalence estimates of disabilities among children under 20 years by UNICEF and GBD 2019.

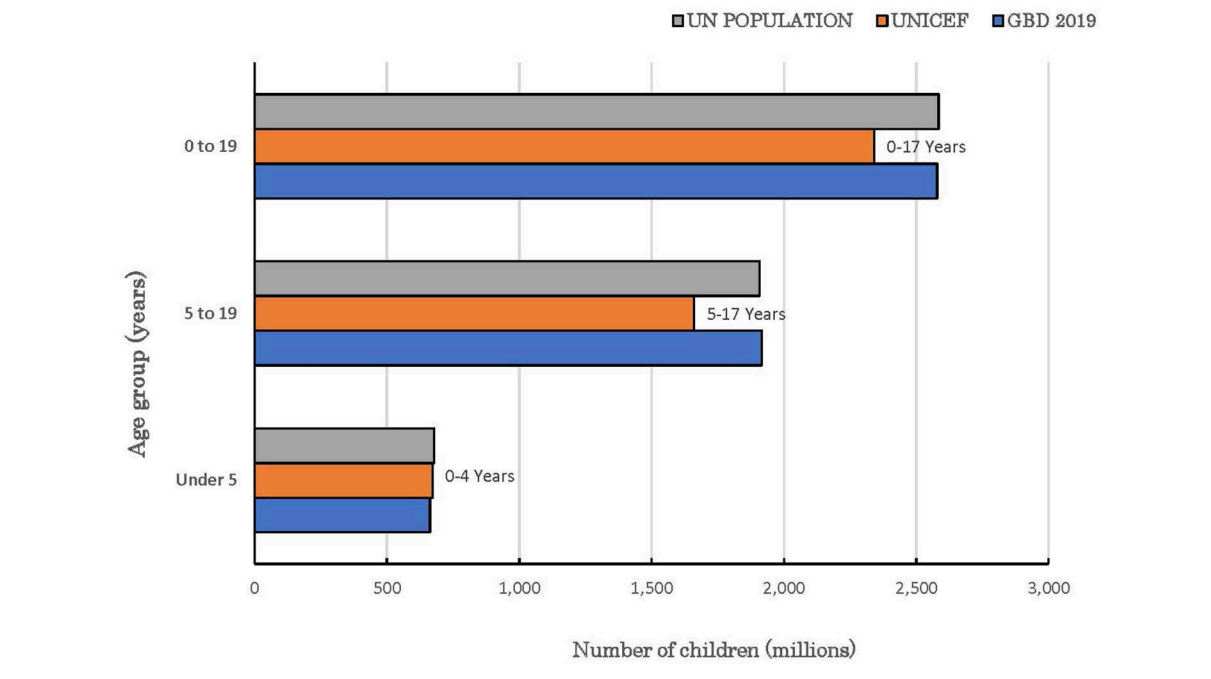

FIGURE 4
Global population of children with and without disabilities in the reported age groups.

embraced by UNICEF (40–42). For example, routine screening and confirmation of babies for congenital hearing impairment is legally mandatory within the first 3 months of life in many high-income countries well-before the functional difficulties associated with hearing impairment become apparent usually after 18–24 months (46). Functional approach to prevalence estimate will miss such infants. No single approach to prevalence estimation is flawless, better, or sufficient by itself to serve the multidimensional interests of children with disabilities. This fact is duly acknowledged by UNICEF and GBD (13, 17, 21, 22). The ongoing implementation of the MDS tool designed to elicit information on functional limitations and associated health conditions by WHO and the World Bank (18) is likely to offer a more robust comparative analysis of prevalence estimates in future.

Additionally, neither UNICEF nor GBD cover the full spectrum of known disabilities in children. Thus, the reported prevalence should appropriately be regarded as the minimum estimates among children with disabilities. All estimation approaches require some degree of imputation and statistical adjustments, and concerns have been raised on modeling approaches in general and particularly for those used by GBD (6, 47). While efforts to improve the reliability of such estimates are needed, the COVID-19 pandemic has further underscored the need for different approaches to prevalence estimation outside the traditional in-person house-to-house surveys.

The focus of this paper was to examine how the available global and regional estimates of disabilities among children can be optimized to facilitate the implementation of policies and action plans for achieving inclusive education as envisioned in the SDGs and reinforced by CRPD (2, 9). In our view, the estimates from both sources, using functional approach and the identification of specific impairments associated with various health conditions should be regarded as complementary and in line with the ICF framework. While an effort by UNICEF to include children younger than 2 years through data imputation based on findings among children 2–4 years is commendable, we wish to reiterate earlier calls on the need to expand the CFM to include children younger than 2 years in line with the principles and concept of early childhood development globally (48). This is not only consistent with the spirit and letter of the SDG of leaving no child behind, but also allows for improved age-specific comparison across all databases. Additionally, there is need to highlight the inequalities among children and adolescents with disabilities in low- and middle-income countries compared to high-income countries across all data sources and indicators of functioning status which are required for any effective rights-based advocacy.

Some limitations of this study are worthy of emphasis. For example, the age range covered by both data sources differed and the lack of adequate validation studies for child functioning module for children under 5 years would have compromised the estimates by UNICEF as reference standards for assessing data from other sources. Our inability to obtain 95% uncertainty intervals for the combined estimates of the six disabilities included in the GBD 2019 as at the time of this study is a limitation that can be resolved in future with additional inputs from the organization. Notwithstanding, the overarching evidence from the available data sources demonstrate the magnitude of disabilities among children and adolescents that need to be addressed within the SDGs framework to ensure improved developmental trajectory for the affected children from early childhood for optimal educational opportunities.

Conclusion

The global and regional prevalence estimates of children and adolescents younger than 20 years with disabilities relevant to the monitoring requirements of the SDGs are now provided by UNICEF and GBD. The latest prevalence estimates of disabilities reported from these two sources are generally comparable but would require improved alignment of the age groups and the selected severity thresholds, especially for children under 5 years. The ICF conceptually encapsulates the medical and social models of disability, and no single data source presently fully satisfies the biophysiological paradigm of this framework. While the UNICEF data provides unique and valuable insights on the functional challenges faced by children with disabilities compared to children without disabilities, the GBD data offer equally valuable insights on the nature of the medical services that will assist these children optimize their functional performance. We conclude that the interests of children with disabilities and their families will continue to be well-served by data from a variety of complementary sources to inform global policy interventions. Future analysis is likely to be boosted by the inclusion of findings from the ongoing MDS implementation by WHO and the World Bank.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author/s.

Author contributions

BO drafted the manuscript. VK, AS, FO, and AD critically reviewed the draft, contributed to the statistical analysis, and suggested essential edits. BO and AD are guarantors. All authors contributed to revising the manuscript, and approved the final version as submitted.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2022.977453/full#supplementary-material

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Early Childhood Development policy in Chile: Progress and pitfalls supporting children with developmental disabilities toward school readiness

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Introduction

There is now evidence indicating that first 5 years of life are of major importance for learning and health across life course (1, 2). This period is key in providing detection and timely supports for children with developmental disabilities (3, 4). Because of this, many investments have been done around the world (5). Specifically for this article, we illustrate the case of Chile. This paper provides an overview of some investments in public health, social protection, and education that Chile has done in supporting children with developmental disabilities during early childhood. The authors also provide their opinion about progress as well as barriers affecting school readiness for children with developmental disabilities during the last decade.

Background and Early Childhood Development (ECD) programs currently funded by Chile for children with disabilities

In 2000, UNESCO considered that Chilean educational results were poor compared to countries with similar economic development and that the large gaps between rich and poor groups were alarming (6). However, the absence of national data on developmental delays in children made it impossible to identify the fraction of the population in need of preventive or supportive services. In 2005, the National Survey of Health and Quality of Life integrated a parent report on the developmental milestones of children from 3 months to 5 years. The objective was to estimate the magnitude of developmental delays so that the health, education, and social sectors could plan their budgets and programs (7).

Since then, Chile has made significant investments in policies aimed at the early detection, prevention, and services for developmental disabilities in children under 5 years of age (8).

Since 2005, a new policy guarantying by law to the entire population, opportunity to access health services and financial protection—through Explicit Guaranties from a list of health conditions, as well as extra support for high-cost eligible medical treatments, increased access to medical treatments that might support some children with disabilities (9, 10). For example, any child born before 32 weeks of gestation or born with <1,500 gr has guaranteed access to hearing screening. If the screening reveals significant hearing deficit (more than 35 decibels), children are eligible to receive headphones, cochlear implants, and speech and language therapy. Since 2013, children under 4 years old with moderate, severe, or profound deafness (more than 40 decibels hearing deficit) have also access to headphones, cochlear implants, and therapy (11).

Another action taken in the health sector is the integrated health guideline for primary health care providers, recommended by the Ministry of Health (12). It includes a chapter for children with special needs, with special attention to Down Syndrome and Autism. This includes 24 services guaranteed for children with Down Syndrome during their first 5 years of life. The section for autism has a guideline for autism screening between 16 and 30 months of age, using the M-CHAT-R/F, validated for Chile, with high sensitivity and specificity (13). Chile started collecting M-CHAT-R/F data from all public health services in 2019. Unfortunately, COVID-19 restrictions reduced significantly screening services since 2020, making it hard to assess its impact.

Regarding education, Chile has been gradually implementing inclusive education, starting at the pre-kinder level since 2009 (14). Public schools receive financial support to include children with developmental disabilities (known as "permanent special needs") in regular schools. The Ministry of Education provides financial supplement for every child with developmental disabilities integrated into regular education, with a cap of 2 students per class. Schools who integrate deaf or blind children in small class sizes (maximum 8 children) receive an additional financial supplement (15). Access to inclusive education for children with developmental disabilities under 5 years old has increased (16).

In 2007, a cross-sectoral system of integrated services through the Social Protection sector was implemented. The national Early Childhood Development policy "Chile Crece Contigo" (ChCC, Chile grows with you) coordinates activities offered across nine ministries, from the prenatal period up to 9 years old (17, 18). In 2019, ChCC identified lack of timely services for eligible children with developmental disabilities (from 60% most socially vulnerable households), including autism, and developed pilot programs for supporting children with developmental disabilities called now Inclusive Rooms in 21 communities ("comunas") across the country (19). This program finances the training and services of interdisciplinary teams, including speech and language pathologists, occupational, and/or physical therapists, who educate the parents and provide direct developmental services for children under 4 years of age. This pilot program also coordinates benefits that children with the national disability credential (issued by the National Service of Disability, SENADIS) can access. Example of benefits that can support children's readiness for school include assistive communication devices, such as tablets with speech generating devices for non-speaking autistic children over 4 years old, Braille typewriters, as well as other assistive technology (20).

Discussion: Pitfalls and suggestions

Despite the large progress implementing ECD policies, Chile has currently not reported a national indicator on proportion of children under 5 years of age who are developmentally on track in health, learning, and psychosocial well-being. The governmental related website indicates that data is being developed, studied, or analyzed since 2019 (21) but indicator still is not available. Moreover, lack of randomized evaluations of several interventions scaled up in Chile makes difficult to recognize the cost-effectiveness of this large investments on child outcomes.

Following on a comparison of two health surveys separated by 10 years, Chilean government reported a massive reduction from 25% in 2006 to 11% in 2017 on developmental delay rates after implementing ChCC. However, only children at age 3 years old group exhibit these differences and no significant changes are observed at other age groups (22). On the other hand, experimental evidence from a large, randomized trial on a parenting intervention showed robust effects on reducing language developmental delays and lower rates of socioemotional developmental delays on families who were offered parenting classes in primary health care using the Canadian well-known Nobody's Perfect Parenting Program that was adapted to the Chilean culture (23). However, take up of the program was still small and generalizability to children with developmental delays was not possible because they were listed among the exclusion criteria of the target population.

Furthermore, we could not find a rigorous study following up children from lower socioeconomic background with cochlear implants and their school readiness, placement, and educational outcomes. We know that access to language for deaf people with developmental disabilities in a linguistically accessible environment, adapted to their communicative needs—including the use of their national sign language, is both a basic need and a fundamental human right (24). This aspect should be strengthened in the available programs.

Lissi et al. (25) included among their recommendations to the Ministry of Health, that early detected deaf children

and their parents would be supported also by trained deaf psychologist and deaf educator during the early years, ensuring access to Chilean sign language and culture. The absence of this early support contributes to language deprivation with a profound impact on the quality of their educational inclusion and overall future development (24, 25). Currently, eligible deaf children under 4 years old are being supported by the new Inclusive Rooms, coordinated by ChCC. We encourage ChCC representatives of the Inclusive rooms to take Lissi et al.'s recommendations and to develop plans that exist in place to conduct a rigorous study to evaluate the impact of the pilot programs for children with developmental disabilities. Such plan should assess not only children's school readiness but also the readiness of schools to receive and support the children transitioning from this program to formal education (26).

Another barrier is the fragmentation of data collection. Chile's system to obtain a disability credential, through SENADIS, does not seem to have a publicly available database with the number of unidentified children who obtained the disability credential, disaggregated by diagnosis, age, gender, socio-economic status, and benefits provided, that can be analyzed for planning. In other words, no data, no problem, no action. Moreover, a recent study evaluating access of children with disabilities to services, identified that families are reluctant to obtain the national disability credential for their children. They described the process as cumbersome, slow and feel afraid of stigmatization (27). In addition, the information regarding benefits for children under 5 years of age in the SENADIS website is difficult to understand because is not presented in a friendly and accessible way.

In addition to the impact evaluation, and fragmentation, another pitfall of Chilean investments for school readiness of children with developmental disabilities is the lack of involvement of the strong disability community in Chile. It will be highly desirable that the Chilean disability community could help shaping the design of services, including an evaluation component, for young children with disabilities. Chile has a strong deaf community and a growing autism community. Current best practices recommend Community-Based Participatory Research involving scientific professionals and experts by experience working together in developing, implementing, and disseminating research (28). "Nothing about us without us"[1] has become an expression that communicates that "no decision that influences people with disabilities should be made without their participation." This movement provided the ethical basis, within international human rights, in calling State members to guarantee the participation of people with disabilities in all aspects of public policy (29). Neurodivergent communities point out the need to generate spaces for literacy, awareness, and development of knowledge about neurodivergences through instances of dialogue and co-creation of projects that include representative actors of existing neurodivergencies (30).

Lastly, the current new government administration has expressed interest in increasing funding for mental health. We hope that this very much needed attention to this important area, "no health without mental health" (31), includes also young children with disabilities. There is evidence that states that deaf persons show higher levels of anxiety and depression compared to the general population (32, 33). Also, the recent Lancet commission on Autism identified that anxiety among autistic children starts in infancy (Figure 2, page 275) (34). Pukki et al. (35) replied to the Lancet commission on autism, sharing the autistic perspectives on the future of clinical autism research. Among their recommendations, the autistic authors urge to focus more resources on mental health support, among other (35, 36). It is not clear to us how Chile's ECD program will support the transition of children with disabilities to schools, where bullying starts early and children with disabilities are often targets, affecting their learning and well-being. Currently, there is new evidence that neurodivergent children are at more risk of co-associated anxiety, depression, and suicide. School exclusion and bullying can be a modifiable factor (37). In addition, there are reports of high stress levels within the family environments of children with disabilities (38). It is a priority to include mental health support and services for parents and caregivers within national early childhood programs.

To have more inclusive communities, and therefore, better mental health of children with neurodevelopmental disabilities, it is important to focus on acceptance, significantly changing current practices, shifting from deficits to strengths-based approaches. We propose that a good way to start is to work on awareness and changes of pathologist and ableist[2] views of disabilities (39). Chile has done significant progress in Early Childhood Development policies and programs. Children with disabilities are being included with more targeted services in the last few years. However, we still have a very ableist view of approach. Their lives are worthy as they are, and this needs to be highlighted. It is necessary to address disability as part of human diversity, where the design of health and education programs incorporate dignified and respectful perspectives of their identity, from those who live the experience. We hope that our views shared in this opinion article help to strengthen Chile's programs and be a model for the world also in this area.

1 "Nothing about us without us" is a phrase used by South African disability rights activists Michael Masutha and William Rowland in the 1990s; and then in 1998, J. Charlton begins to use and spread it.

2 According to Nario-Redmond, ableism is the discrimination of and social prejudice against people with disabilities based on the belief that typical abilities are superior (39).

Author contributions

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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The developing brain: Challenges and opportunities to promote school readiness in young children at risk of neurodevelopmental disorders in low- and middle-income countries

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This paper discusses possibilities for early detection and early intervention in infants with or at increased risk of neurodevelopmental disorders in low- and middle-income countries (LMICs). The brain's high rate of developmental activity in the early years post-term challenges early detection. It also offers opportunities for early intervention and facilitation of school readiness. The paper proposes that in the first year post-term two early detection options are feasible for LMICs: (a) caregiver screening questionnaires that carry little costs but predict neurodevelopmental disorders only moderately well; (b) the Hammersmith Infant Neurological Examination and Standardized Infant NeuroDevelopmental Assessment (SINDA) which are easy tools that predict neurodisability well but require assessment by health professionals. The young brain's neuroplasticity offers great opportunities for early intervention. Ample evidence indicates that families play a critical role in early intervention of infants at increased risk of neurodevelopmental disorders. Other interventional key elements are responsive parenting and stimulation of infant development. The intervention's composition and delivery mode depend on the infant's risk profile. For instance, in infants with moderately increased risk (e.g., preterm infants) lay community health workers may provide major parts of intervention, whereas in children with neurodisability (e.g., cerebral palsy) health professionals play a larger role.

Abbreviations
ASD, Autism spectrum disorders; ASQ, Ages and Stages Questionnaire; CHW, Community health worker; CIMT, Constraint-induced movement therapy; COPCA, COPing with and CAring for infants with special needs; CP, Cerebral palsy; EGL, External granular layer; GAME, Goals Activity Motor Enrichment; GMA, General movement assessment; HICs, High-income countries; HINE, Hammersmith Infant Neurological Examination; LMICs, Low- and middle-income countries; M-CHAT, Modified Checklist for Autism in Toddlers; MRI, Magnetic resonance imaging; R-ND, increased biological Risk of or with a Neurodevelopmental Disorder; PEDS, Parents' Evaluations of Developmental Status; PMA, Postmenstrual age; SINDA, Standardized Infant Neurodevelopmental Assessment

KEYWORDS

brain development, cortical subplate, infant, early detection, early intervention, neurodevelopmental disorders, cerebral palsy, low and middle income countries

Introduction

Global mortality in children aged under 5 years decreased by 60% between 1990 and 2020 due to the impact of the United Nations' Millennium Development Goals (1). Unfortunately, this accomplishment was not paralleled by a similar decrease in childhood disability (2). The combination of an increase in surviving children particularly in low- and middle-income countries (LMICs), a rapid population growth in LMICS, and often fragile health care systems in these countries, contributed to a high prevalence of children with neurodevelopmental disabilities (1, 2). It has been estimated that over 53 million children under 5 years had neurodevelopmental disabilities globally in 2016 (3). Over 90% of these children lived in LMICs (1, 4).

The United Nations Convention on the Rights of Persons with Disabilities (2006) and the United Nations Sustainable Development Goal 4 (2015) declared that children with disabilities have the right of inclusive education (5, 6). Nonetheless, UNICEF statistics revealed that many children with disabilities do not receive proper support and adequate education (7). UNICEF's data indicate that children with disabilities are 25% less likely to receive early stimulation and responsive care, 25% less likely to attend early childhood education and 49% more likely to have never attended primary school than children without disabilities (7). In order to improve this situation, it is mandatory that children with neurodevelopmental disorders, such as cerebral palsy (CP), intellectual disability and autism spectrum disorders (ASD), are detected at early age and receive early intervention (2, 8). Early detection and early intervention will result in improved school readiness, as they allow for optimal preparation of family and child so that the child may fully engage in learning experiences at school.

This perspective paper aims to discuss methods available for early detection and early intervention in infants with an increased biological risk of or with a neurodevelopmental disorder (hereafter: infants with R-ND). It pays special attention to those methods that are mostly geared to the health care situation in LMICs. Early detection and early intervention occur in a developmental timeframe that is characterized by abundant brain development. Therefore, the paper first summarizes the developmental changes in the young human brain and its implications for early detection and early intervention. It focuses on the first two postnatal years. The following two sections briefly review knowledge on early detection of and early intervention in infants with R-ND. The last section discusses how early detection and early intervention in infants with R-ND may be achieved best in LMICs. It stresses the importance of family involvement and the need of adaptation to local situations, including cultural habits and beliefs.

Early human brain development: Opportunities and challenges

Early human brain development

The development of the human nervous system is a long-lasting and intricate process based on ingenious interactions between genes, environmental information and experience (9). **Figure 1** provides an overview of the elementary components of brain development. The majority of neurons and glial cells are generated during prenatal life. Many neurons do not stay at their origin's site but migrate during gestation to their final destination. Neuronal differentiation, synapse production and myelination start early in fetal life to become very active in gestation's last trimester and the first year post-term. Thereafter, these processes continue at a slower pace.

Brain development is not only a matter of production of elements; it also involves massive elimination. About half of generated neurons die through programmed cell death, particularly during gestation's third trimester. Also, axons are initially produced in excess and later partially removed, especially during the end of gestation and the first 3 months post-term. Throughout life, synapses are formed and eliminated, with synapse elimination peaking between the onset of puberty and early adulthood (9).

The combination of production and regression gives rise to temporary structures and connections. Major transient structures are the cortical subplate and the cerebellar external granular layer (EGL; **Table 1**). The cortical subplate is a temporary structure between the developing white matter and cortical plate. It hosts the first generations of cortical neurons and plays a critical role in cortical development being the major site of neuronal differentiation, synaptogenesis and synaptic activity in the fetal cortex. It receives the first cortical afferents (10). The cortical subplate, which is most prominently present between 28- and 34-weeks postmenstrual age (PMA), mediates fetal behavior. From mid-gestation neurons in the subplate start to die and next generations of migrating cortical neurons begin to populate the cortical plate, i.e., the site of the permanent cortical networks. Around 3 months post-term, the subplate has largely disappeared in the

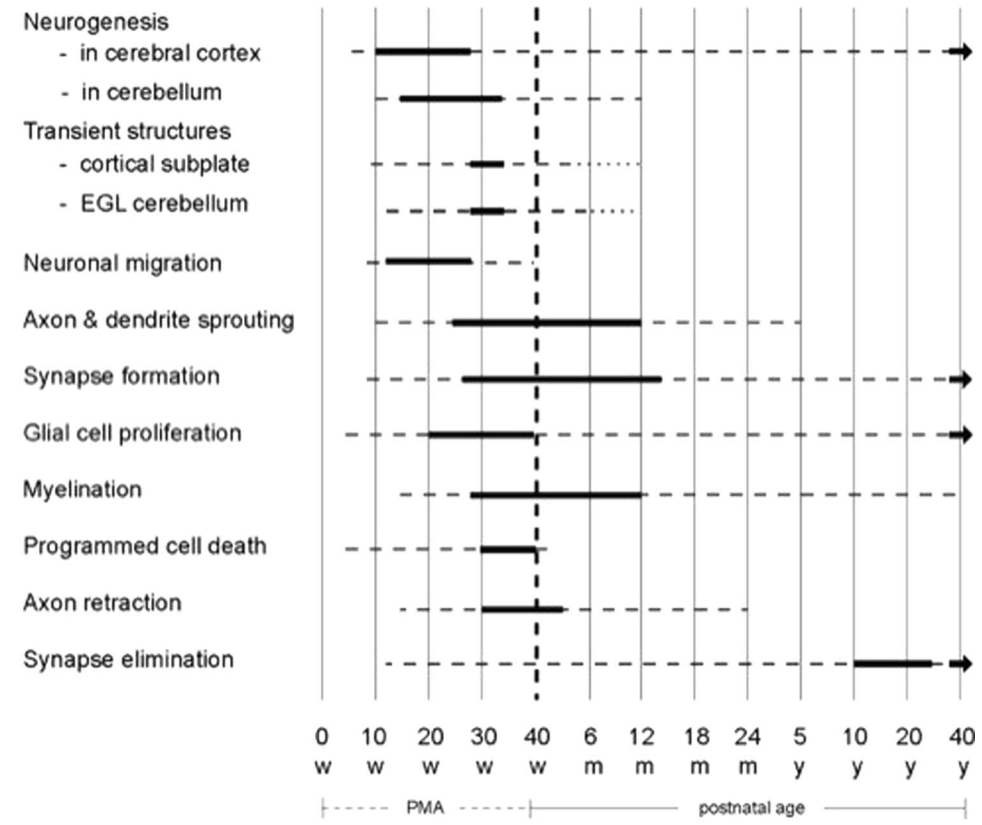

FIGURE 1
Schematic overview of the developmental processes occurring in the human brain. The bold lines indicate that the processes mentioned on the left side are very active, the broken lines denote that the processes still continue but less abundantly. The diagram is based on reference (9). EGL = external granular layer; m = months; PMA = postmenstrual age; w = weeks; y = years. Figure reproduced with permission from "Early Detection and Early Intervention in Developmental Motor Disorders—from neuroscience to participation" by Mijna Hadders-Algra (ed.) published by Mac Keith Press in its Clinics in Developmental Medicine Series, ISBN number 978-1-911612-43-8 (11).

TABLE 1 Transient structures in the developing human brain.

Structure	Function	Period of presence
Cortical subplate in primary motor, sensory and visual cortex	- Pivotal role in shaping of permanent circuitries in cortical plate - Mediation of sensorimotor behavior in early life	- Most prominently present at 28–34 week PMA - Largely dissolved around 3 months post-term
Cortical subplate in frontal, temporal and parietal association cortex	- Pivotal role in shaping of permanent circuitries in cortical plate - Mediation of social and motor behavior in early life	- Most prominently present at 28–34 week PMA - Largely dissolved around 12 months post-term
Cerebellar external granular layer	- Production of the granule cells, the most numerous cells of the cerebellum and brain	- Most prominently present at 28–34 week PMA - Dissolving between 6–12 months post-term

For details see references (9) and (10).

primary motor, sensory and visual cortex, but it takes until the age of 12 months before the subplate has largely dissolved in the frontal, temporal and parietal association areas (9, 10). This means that infant behavior before subplate dissolution is based on activity in the networks in the "fetal" subplate and the cortical plate. First, after the disappearance of major parts

of the cortical subplate, infant behavior is mainly mediated by the permanent cortical networks (9, 10). The other significant temporary structure is the cerebellar EGL. The EGL produces the granule cells, the most numerous cells of the brain. The EGL emerges around 15 weeks PMA and is most prominently present between 28- and 34-weeks PMA. Thereafter, it shrinks and disappears completely between 6- and 12-months post-term (9).

As mentioned above, axon development is also characterized by a combination of growth and regression. A well-known example is the axon retraction in the corticospinal tract (11). This tract begins with bilateral projections. Retraction of the ipsilateral projection starts in gestation's last trimester and is largely completed around the age of 2 years (11). This implies that, first at 2 years, the corticospinal tract has achieved its adult configuration with predominantly contralateral projections.

Implications of early brain development for early detection and early intervention

The brain's developmental activity in the first two years post-term results in specific windows of vulnerability for adverse events, such as inadequate nutrition, preterm birth, or hypoxic-ischemic events (12). The events' unfavorable effect often impacts development in multiple domains, including motor, cognitive, communication and socio-emotional abilities (12). The brain's high developmental activity also has important implications for early detection and early intervention in neurodevelopmental disorders. It offers opportunities and challenges. The brain's great developmental activity generates the opportunity of high neuroplasticity. Neuroplasticity may result in "growing out of dysfunction". This means that signs of neurological dysfunction that may be present at early age in infants with prenatal, perinatal, or neonatal complications (with or without a brain lesion) may disappear with increasing age (13, 14). Moreover, the high neuroplasticity offers opportunities for early intervention. For instance, it is well known that developmental stimulation in preterm infants results in improved cognitive and motor outcome (15).

The brain's high rate of developmental activity also induces challenges, particularly for early detection of neurodevelopmental disorders. The developmental changes may not only result in resolution of neurological signs, but they may also be associated with the emergence of signs, i.e., "growing into a deficit". The developing brain usually needs time to express signs of specific neurodevelopmental disorders. The early signs of CP manifest especially from 3 months post-term onwards, i.e., from the time that the cortical subplate in the primary motor and sensory cortex has dissolved (16). Ample evidence has demonstrated that abnormal general movements at 3 months post-term are a powerful predictor of CP (16, 17). The asymmetries of unilateral spastic CP are subtly expressed from 3 to 5 months onwards and become increasingly clear during the rest of the first year when the corticospinal tract reorganizes (18, 19). The early signs of ASD such as impaired social communication, atypical sensory responsivity and repetitive behavior, become clinically predictive from 12 months onwards, i.e., at the age that the cortical subplate has largely disappeared in the cortical association areas and the EGL has vanished (20).

The above described and other early signs of increased risk of disability generally do not allow for the diagnosis of a specific neurodevelopmental disorder. Currently the average age at the diagnosis of CP is 12 months (21), and of ASD, 43 months (22). Nonetheless, it is important to realize that a diagnosis is not needed to start early intervention. Knowing that an infant is at increased risk of neurodevelopmental disorders invokes the need of early intervention (17).

Early detection of neurodevelopmental disorders

World-wide developmental screening tools are most often used to detect infants with R-ND. Commonly applied methods are caregiver questionnaires [e.g., Parents' Evaluations of Developmental Status (PEDS) (23), Ages and Stages Questionnaire (ASQ) (24)], and the Denver Developmental Screening Test (25). These methods are largely based on attainment of developmental milestones. From the age of 2 years these methods are relatively good in detecting children with developmental delay (26–28). However, their ability to detect children with neurodevelopmental disorders during the first two years is less satisfactory, with sensitivities of 40%–60% and specificities of 59%–77% (29, 30). The most frequently used caregiver questionnaire to detect ASD is the Modified Checklist for Autism in Toddlers [M-CHAT (31)]. In children aged at least 12 months M-CHAT has moderate predictive power in children at increased familial risk of ASD (32).

Five years ago, a systematic review on early prediction of CP indicated that the best methods available for young infants were magnetic resonance imaging (MRI) at term age, and the general movement assessment (GMA) around 3-month post-term (17). In term infants with hypoxic-ischemic encephalopathy, MRI-scans predict CP with sensitivities and specificities of 70%–90% (32). In preterm infants, term-MRI predicts CP with a sensitivity and specificity of 77%–79% (33). GMA is based on the evaluation of the quality of 3 min of general movements in supine. The presence of general movements with seriously reduced movement variation and lacking the age-specific fidgety movements around 3 months post-term predicts CP with a sensitivity and specificity of 91%–98% (16, 34).

The review of Novak et al. (17) also indicated that throughout infancy the Hammersmith Infant Neurological Examination (HINE) is a good instrument to detect CP. It does not only

TABLE 2 Properties of HINE and SINDA's neurological scale.

Property	HINE	SINDA's neurological scale[a]
Age range (corrected age)	2–3 months - 24 months	6 weeks – 12 months
Domains	- cranial nerve function - posture - movements - muscle tone - reflexes	- spontaneous movement (special attention quality) - cranial nerve function - motor reactions - muscle tone - reflexes
Number of items	26	28
Scoring of items	- ranging from 1 to 4 - criteria for atypical age-dependent	- dichotomous - criteria for atypical not dependent on age
Cut-off for at risk score	varies for different ages and different studies; cut-offs only reported for 3, 6, 9, 12 and 18 months	identical for entire age range: ≤21
Time needed, including administration	<10 min	<10 min
Normative data	not available	present in manual
Reliability	good	good
Prediction of CP		
Sensitivity	- 90%–100%	- 91%–100%
Specificity	- 85%–100%	- 81%–85%
Prediction of CP and/or intellectual disability	intellectual disability	CP and/or intellectual disability
Sensitivity	- 51%–82%	- 83%–89%
Specificity	- 71%–90%	- 94%–96%
Performed by	health professionals	health professionals
Training	*via* website with instructional videos; no manual available	*via* manual and accompanying >160 video clips

[a]SINDA has two additional scales: a developmental and a socio-emotional scale. The developmental scale has 15 items per months covering cognition, communication, gross and fine motor development. An "at risk" developmental score predicts intellectual disability with a sensitivity of 77% and a specificity of 92%. The socio-emotional scale addresses interaction, emotionality, self-regulation and reactivity. Emotionality and self-regulation predict with sensitivities of 32%–40% and specificities of 85%–98% behavioral and emotional problems at ≥2 years (37).
For details see (35–40).

predict CP, but also intellectual disability [**Table 2** (39, 40)]. More recently, the Standardized Infant Neurodevelopmental Assessment (SINDA) has been developed. SINDA consists of a neurological, developmental, and socio-emotional scale (36–38). SINDA's neurological scale predicts CP and intellectual disability well; its developmental scale also predicts intellectual disability (**Table 2**; 15, 32, 45).

Early intervention in infants with or at increased risk of neurodevelopmental disorders

This section focusses on early intervention in infants with R-ND during the first two years. Families play a pivotal role in early intervention (41–43). They form the infants' major environment. Also, family members are the key persons impacting child development through daily interaction during caregiving and play. Details of the intervention approach depend in part on the nature of the infant's risk profile. To this end three groups of infants may be distinguished: (a) infants with prenatal, perinatal, or neonatal complications without a significant brain lesion; (b) infants with a significant brain lesion or neurological signs suggestive of such a lesion; and (c) infants at increased familial risk of ASD.

For the first group of infants, many intervention programs are available (44). Ample evidence exists that sensitive and responsive parent-infant interaction and stimulation of infant development are associated with better family well-being and favorable infant development (11, 32, 45).

Less evidence exists on the effective elements of early intervention in infants with a significant brain lesion (32, 45, 46). Nonetheless, available information suggests that the following key elements are beneficial (32, 45, 46): (a) family involvement; (b) focus on the child's activity domain, i.e., on the child's mobility, learning and knowledge, and

communication, and not on impairments such as deviant muscle tone or atypical reflexes; (c) early introduction of assistive devices to promote activities and participation and to prevent contractures and deformities; (d) emphasis on activities and participation of family and child (45). Programs that include these elements are Goals Activity Motor Enrichment (GAME) (47, 48), the Small Step Program (49), COPing with and CAring for infants with special needs (COPCA) (50–52), and - for infants at increased risk of unilateral CP - baby constraint-induced movement therapy (baby-CIMT) (53), and intensive bimanual activities (54). These programs aim to challenge children to explore by self-generated movements with trial and error their own body and the physical and social world.

Knowledge on effective intervention in infants at increased risk of ASD is limited as most intervention studies have been performed in children diagnosed with ASD, implying an age of at least 2.5 years (32). Recent systematic reviews (55–59) suggested but did not prove that in children with ASD, a developmental approach with or without behavioral components is associated with a positive effect on social communication. The evidence on the effect of intervention in infants at increased risk of ASD is very limited (55). The data available suggest that a caregiver-mediated social communication intervention may be associated with improved child attention and social communication and better caregiver responsiveness (55, 60, 61).

Discussion and conclusion

The rapidly developing brain during infancy imposes challenges for early detection and offers opportunities for early intervention. This is true for high income countries (HICs), but the situation in LMICs is significantly more challenging due to the large number of infants with R-ND in combination with limited resources for early detection and early intervention (62, 63).

Early detection by means of caregiver questionnaires is more cost-effective than that based on testing by professionals. This makes questionnaires (especially PEDS and ASQ) attractive for LMICs despite their less favorable detection properties than assessments by professionals. Nonetheless, barriers such as low caregiver education, illiteracy, and linguistic and cultural diversity may impede general implementation of screening questionnaires (64–67). Assistance by paraprofessional community health workers (CHWs) (68) may reduce these barriers (69) but will increase costs.

The best tools for detection of infants at high risk of neurodevelopmental disorders in the first year post-term are MRI at term, GMA, HINE and SINDA. MRI requires expensive equipment making it less feasible for LMICs. Videorecording of spontaneous movements in GMA is easy and may be performed by caregivers using mobile phones, although educational and linguistic barriers may limit successful recording (70, 71). The latter problem may be solved by videorecording by lay CHWs (68). However, the evaluation of general movement quality requires ample experience, which hampers the implementation of GMA, particularly in LMICs (72, 73). In the future, this situation may change through the application of automated GMA (74–76). Of the best detection tools, HINE and SINDA's neurological scale are the most cost-effective options. HINE and SINDA require the skills of health professionals working in infant health care. Both methods take relatively little time, they do not require an expensive toolkit and they have good predictive properties. HINE covers a larger age range than SINDA. Yet, SINDA's neurological scale has the practical advantages of having a detailed manual and being easier than HINE, as its items and cut-off for "at risk" are independent of infant age (**Table 2**).

Most early childhood development programs in LMICs focus on health and nutrition in children living in poverty (77). Of course, attention to health and nutrition is quintessential, as health and growth are basic requirements for children to reach their developmental potential. However, the LMIC-literature pays little attention to early intervention in infants at increased risk of neurodevelopmental disorders due to prenatal, perinatal, or neonatal complications, e.g., preterm infants. But it is conceivable that the early intervention strategies that are effective in preterm infants in HICs are also beneficial for preterm infants in LMICs. Actually, the effective strategies to promote development in socially disadvantaged infants in LMICs have large similarities to those applied in preterm infants in HICs (45, 78, 79). Key-elements of both approaches are family involvement, support of caregivers in provision of responsive caregiving, and stimulation of infant development (15, 45, 78, 80). These interventions may be provided by trained lay CHWs to groups of caregivers in the local community with or without home visits by the CHW (81). The home visits may also be replaced by tele-coaching (82). It is conceivable that similar family-community approaches may also work in young children at increased risk of or with ASD. Yet, as described above, evidence on the best intervention approaches in these children is still lacking.

Gradually it is becoming clear which early intervention strategies are beneficial for infants with R-ND due to a significant brain lesion. Essential elements are family involvement, focus on activities and participation of child and family, and prevention of contractures and deformities. Guidance of families with a child with neurodisability is more complex than guidance of families with a preterm infant. It requires more professional effort. Studies performed in LMICs indicate that a combination of caregiver group sessions ran by health professionals in combination with (a) tele-coaching by health professionals and/or (b) home visits by trained lay

CHWs may be feasible means to deliver intervention services in infants at increased likelihood of or with neurodevelopmental disorders (82, 83). In the implementation of these early intervention services, it is important to recognize cultural diversity in understanding neurodisability (84). Accordingly, the first steps in early intervention consist of discussing with the family the child's condition, its significance for child, family and community, and the goals of early intervention.

In conclusion, the young brain's neuroplasticity imposes challenges and offers opportunities. It is challenging to detect in the first year infants with R-ND, as the brain needs time to get rid of its temporary structures and to express specific dysfunction. Nonetheless, our hands are not empty: the PEDS, ASQ, HINE and SINDA offer feasible early detection tools for LMICs. Early intervention needs to be geared to the characteristics of child and family. In early intervention for infants with R-ND, the family plays a critical role. In LMICs, families generally are firmly imbedded in the local community, as LMIC-societies function more collectivistic than societies in the individualistic HICs (85). The interdependent societal organization in LMICs may offer specific opportunities for early intervention (84), e.g., through the help of lay CHWs. Cultural integration is a prerequisite for successful early intervention in LMICs (86–88). Adequate early intervention in infants with R-ND will pave the way for school readiness by enhancing attitudes, awareness, knowledge and skills of families and communities, early implementation of assistive devices, and optimizing children's motor, cognitive, communication and socio-emotional skills (1, 8, 45).

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material, further inquiries can be directed to the corresponding author/s.

Author contributions

MH-A is the only author of the paper.

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Conflict of interest

MH-A is one of the authors of the manual of the SINDA.

Publisher's note

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Global prevalence of developmental disabilities in children and adolescents: A systematic umbrella review

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Aim: The provisions of the United Nation's Sustainable Development Goals (SDGs) for disability-inclusive education have stimulated a growing interest in ascertaining the prevalence of children with developmental disabilities globally. We aimed to systematically summarize the prevalence estimates of developmental disabilities in children and adolescents reported in systematic reviews and meta-analyses.

Methods: For this umbrella review we searched PubMed, Scopus, Embase, PsycINFO, and Cochrane Library for systematic reviews published in English between September 2015 and August 2022. Two reviewers independently assessed study eligibility, extracted the data, and assessed risk of bias. We reported the proportion of the global prevalence estimates attributed to country income levels for specific developmental disabilities. Prevalence estimates for the selected disabilities were compared with those reported in the Global Burden of Disease (GBD) Study 2019.

Results: Based on our inclusion criteria, 10 systematic reviews reporting prevalence estimates for attention-deficit/hyperactivity disorder, autism spectrum disorder, cerebral palsy, developmental intellectual disability, epilepsy, hearing loss, vision loss and developmental dyslexia were selected from 3,456 identified articles. Global prevalence estimates were derived from cohorts in high-income countries in all cases except epilepsy and were calculated from nine to 56 countries. Sensory impairments were the most prevalent disabilities (approximately 13%) and cerebral palsy was the least prevalent disability (approximately 0.2–0.3%) based on the eligible reviews. Pooled estimates for geographical regions were available for vision loss and developmental dyslexia. All studies had a moderate to high risk of bias. GBD prevalence estimates were lower for all disabilities except cerebral palsy and intellectual disability.

Conclusion: Available estimates from systematic reviews and meta-analyses do not provide representative evidence on the global and regional prevalence

of developmental disabilities among children and adolescents due to limited geographical coverage and substantial heterogeneity in methodology across studies. Population-based data for all regions using other approaches such as reported in the GBD Study are warranted to inform global health policy and intervention.

KEYWORDS

developmental disabilities, global health, Global Burden of Disease, developmental epidemiology, early childhood development, inclusive education, SDG 4.2

Introduction

The United Nations' Sustainable Development Goals (SDGs) are widely embraced, especially in low- and middle-income countries (LMICs), as the priority global agenda for improving population health and well-being by 2030 (1). The disability-inclusive provisions of the SDGs have stimulated a growing interest in children and adolescents (hereinafter reported as "children") with developmental disabilities globally (2, 3). The Convention on the Rights of Persons with Disabilities (CRPD) defines persons with disabilities to include "those who have long-term physical, mental, intellectual or sensory impairments which in interaction with various barriers may hinder their full and effective participation in society on an equal basis with others" (4). Developmental disabilities are frequently defined as chronic physical, cognitive, speech or language, psychological, or self-care conditions that typically originate during childhood before the age of 22 years; are likely to continue indefinitely; and require additional coordinated services, support, or other assistance for an extended duration or during a lifetime; and represent a subset of conditions that affect children with special health care needs (5, 6). Right from birth, children with developmental disabilities, especially in LMICs experience stigma along with negative attitudes and beliefs that place them at increased risk of neglect, exploitation, and violence, as well as premature death including infanticide (2). These children also perform significantly poorer than children without disabilities across virtually all indicators of health and educational wellbeing in early childhood (2).

Up-to-date prevalence estimates are essential to raise awareness and inform policy initiatives, service planning, resource allocation, and research priorities (2). Evidence from global health databases suggests that about 240 million children globally have developmental disabilities based on parent-reported functional difficulties compared to 290 million children using statistical modeling techniques (3). Although systematic reviews and meta-analyses are more suited for evaluating the effectiveness of health interventions and accuracy of diagnostic tests from clinical trials (7–9), it is not uncommon to use pooled prevalence estimates from individual primary studies as proxies for the global and regional prevalence of children with developmental disabilities (10–13). However, it is unclear how such prevalence estimates compare with those reported in global health databases from the World Health Organization (WHO), United Nations Children's Fund (UNICEF), the World Bank or the Global Burden of Disease (GBD) Study published by the Institute for Health Metrics and Evaluation (IHME), USA. Umbrella reviews are increasingly being used to summarize evidence from systematic reviews and meta-analyses, especially for health care interventions (14, 15). We, therefore, set out to conduct an umbrella review of systematic reviews and meta-analyses of the prevalence estimates of developmental disabilities for comparison with estimates from other sources of population data in global health. The primary goal of this umbrella review was to provide a narrative synthesis of the selected reviews due to well-documented differences in the methodological approaches to disability measurement (3).

Methods

The protocol for this systematic umbrella review was registered in the International Prospective Register of Systematic Reviews (PROSPERO), reference number #CRD42022373552 (https://www.crd.york.ac.uk/prospero/#searchadvanced). We adopted the Preferred Reporting Items for Overviews of Reviews (PRIOR) statement for conducting umbrella reviews (16). This statement was considered more up-to-date and better suited for an umbrella review than the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). The term "reviews" in this paper is used for published articles that are systematic reviews and meta-analyses of primary studies. The term "primary studies" refers to any original research or investigation conducted to determine the prevalence of specific developmental disabilities in a defined population.

Search strategy and selection criteria

We searched PubMed, Scopus, EMBASE, PsycINFO, and Cochrane Library in October 2022 using the terms ("prevalence" OR "incidence") AND ("disability" OR "impairment" OR "disorder"), filtered for systematic reviews and meta-analyses, English Language, and children under 20 years published between September 2015 (when the SDGs were launched) and August 2022. Eligible systematic reviews were those that were peer-reviewed with a clearly stated research question, systematic search of at least two databases and systematic data synthesis. No supplementary search for primary studies was conducted (16). The GBD Study from IHME (https://vizhub.healthdata.org/gbd-results/) is presently the only global health database that provides global, regional, and national prevalence estimates of specific disabilities among children and adolescents according to the American Psychiatric Association's (APA's) Diagnostic and Statistical Manual of Mental Disorders (DSM) (17), or WHO's International Classification of Diseases (ICD) codes (18). The selection of specific disabilities for our umbrella review was therefore guided by those typically reported by GBD database to facilitate appropriate comparability (3). These disabilities include attention-deficit/hyperactivity disorder (ADHD), autism spectrum disorder (or simply "autism" hereinafter), cerebral palsy, developmental intellectual disability, epilepsy, hearing loss

and vision loss. We also included developmental dyslexia because of its relevance to the disability-inclusive education provision in the SDGs (1). Developmental dyslexia is a specific impairment characterized by severe and persistent problems in the acquisition of reading skills and it is not typically reported by GBD. Two independent reviewers/authors (BOO and TS) searched titles and abstracts for eligibility and evaluated the full texts of the eligible articles for inclusion. Any unresolved conflict was to be referred to a third reviewer/author (FAO) for adjudication. Reviews that provided pooled estimates with confidence intervals of the selected disabilities were included. In general, these reviews assessed the heterogeneity of the eligible primary studies and performed random effects meta-analysis to estimate the pooled prevalence of a disability. No distinction was made between reviews that evaluated population-based primary studies and those based on a random sample of participants. We excluded reviews that focused on a specific population group such as children who are born preterm, those with different birth weights, refugees, children exposed to HIV or malnourished children. We also excluded reviews that reported a subset of children with a specific disability such as children with refractive errors among those with vision loss as well as reviews that were published before September 2015, that focused on specific countries, one geographical region, or had less than 10 primary studies as such reviews were unlikely to accurately reflect the overall prevalence of disability among all children and adolescents. In order to minimize the risk of missing other relevant systematic reviews, a further manual search of PubMed and selected child health journals was conducted specifically for each of the eight selected disabilities. The reference lists of included reviews were also searched for the identification of additional eligible references.

Data extraction

The citations for the retrieved reviews were first migrated to separate spreadsheets based on the standard fields in each database. A combined spreadsheet was then created for the selected articles with the following fields: source database, year of publication, authors, title, journal, abstract and journal link to the full text. From the full text of the selected articles, the following information were extracted by two authors (BOO and TS): name of disability, citation, year of publication, databases searched, number of primary studies, number of countries covered, proportion of countries from LMICs, overall study size, age group of the reported prevalence estimate, global prevalence estimate, prevalence estimate for high-income countries (HICs), prevalence estimate for LMICs, and remarks. The composition of HICs and LMICs is based on the 2022 World Bank classification (https://data.worldbank.org/country/XO).

Evaluation of the methodological quality

The risk of bias (quality) of included reviews was assessed independently by two reviewers (BOO and TS). The Assessment of Multiple Systematic Reviews (AMSTAR2) tool (available at https://amstar.ca/Amstar-2.php) (19) and the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for umbrella reviews (20) were used as neither tool covered all relevant sources of bias in reviews on the prevalence estimates of developmental disabilities. For instance, AMSTAR2 was specifically designed for health intervention research but it is more comprehensive than JBI checklist and accounts for the quality of the primary studies included in the meta-analysis, without limiting the quality assessment to the technical aspects of the meta-analysis itself. The AMSTAR2 questionnaire has 16 criteria and requires reviewers to respond with a "Yes" or "Partial Yes" or "No" or "No Meta-analysis" option. Overall quality was classified as "critically low," "low," "moderate," and "high" (17). JBI consists of 10 criteria scored as being "met" (1), "not met" (0), or "unclear" (UC), resulting in an overall quality score of 0 to 10. The scores were categorized as low (0–4), medium (5–7), and high-quality (8–10) reviews. Disagreements on risk of bias ratings were resolved through discussion.

Global Burden of Disease estimates

The latest GBD estimates of developmental disabilities in children and adolescents in 2019 were obtained from two publications (3, 21), which were extracted from the substantive GBD 2019 Database (https://vizhub.healthdata.org/gbd-results/) and the GBD-WHO Rehabilitation Database or "WHO Rehabilitation Need Estimator" (https://vizhub.healthdata.org/rehabilitation/). These are the only sources of global and regional prevalence estimates of specific developmental disabilities covering 204 countries and territories, including the 193 UN Member States. The GBD methodology has been extensively reported (3, 21, 22). In summary, the prevalence estimation for each condition begins with the compilation of all available data inputs from systematic reviews of the literature, hospital and claims databases, health surveys, case notification systems, cohort studies, and multinational survey data. A comprehensive list of the sources of input data for each condition is publicly available at the Global Health Data Exchange (https://ghdx.healthdata.org/gbd-2019/data-input-sources). In the data preparation, efforts were made to (i) optimize the comparability of data derived from various sources using different methods; (ii) find a consistent set of estimates across prevalence data; and (iii) generate estimates for locations with sparse or no data by using available information from other locations combined with covariates. Prevalence estimates are then generated using DisMod-MR 2.1, a statistical modeling technique developed specifically for the GBD project. This is a Bayesian meta-regression tool that synthesizes epidemiological data for fatal and non-fatal health outcomes from disparate settings and sources, adjusting for different case definitions/diagnostic criteria or sampling methods, to generate internally consistent estimates by geographical location, year, age group, and sex. The GBD database contains estimates from 1990 to 2019 and are accompanied by the corresponding 95% uncertainty bounds intervals (UI). Prevalence estimates are available for seven of the eight selected disabilities. Developmental dyslexia is presently not included in the GBD databases. We did a narrative synthesis of included studies in comparison to the GBD (2019) study and compared prevalence estimates for the eight selected disabilities.

Results

The initial search of the five bibliographic databases yielded 3453 articles composed as follows: Scopus ($n = 1,788$), PubMed ($n = 681$), EMBASE ($n = 755$), PsycINFO ($n = 87$) and Cochrane Library ($n =$

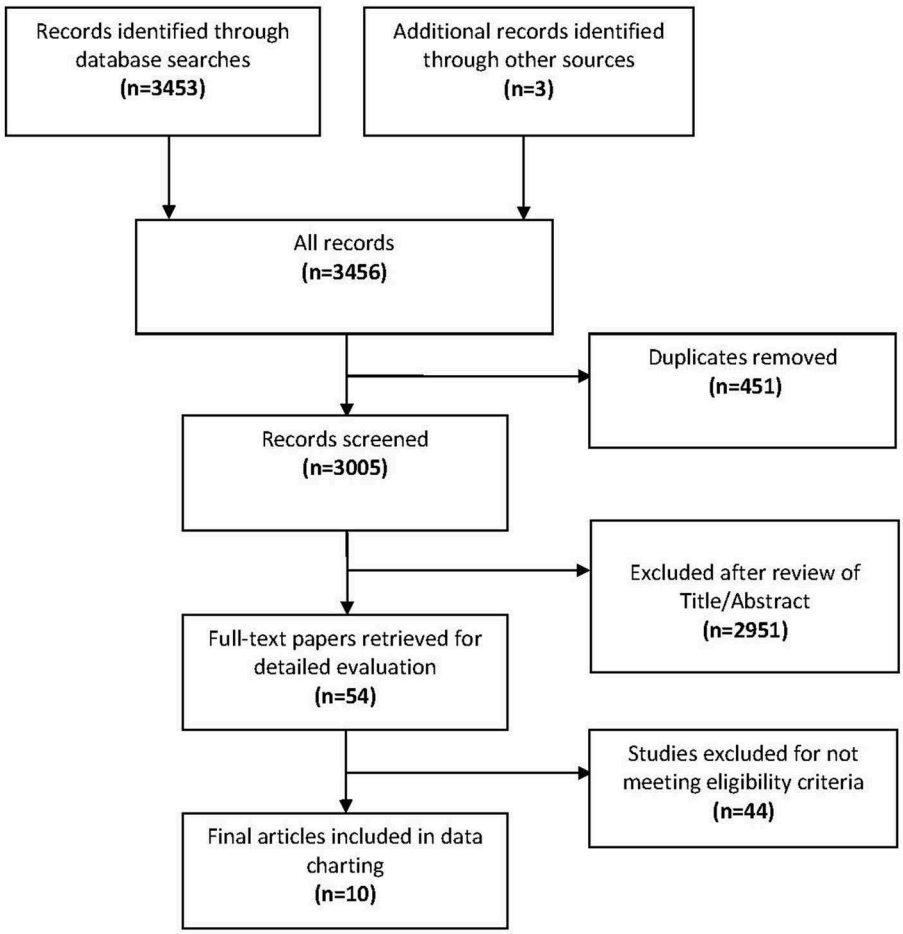

FIGURE 1
Flow diagram of the study selection process and results.

142). Three articles were identified from outside the databases giving a total of 3,456 articles (Figure 1). A total of 54 articles were selected for full-text review based on the inclusion and exclusion criteria. After the review of the full-texts, 44 articles were excluded and the reasons for their exclusion are summarized in Supplementary Table S1. The most common reason for exclusion was the absence of global and regional prevalence estimates for children and adolescents. Of the 10 articles selected for inclusion that reported pooled global prevalence estimates of disabilities (10–12, 23–29), three articles focused on ASD and the remaining seven articles were each focused on one disability. A summary of the selected reviews is presented in Table 1. The primary studies covered by the selected systematic reviews and meta-analyses ranged from 14 to 88 articles and the vast majority were from HICs. The reported age groups varied across most reviews except for cerebral palsy, hearing loss and vision loss. Prevalence estimates of developmental disabilities in LMICs were only reported for ASD, cerebral palsy, and developmental dyslexia. Prevalence estimates for the WHO or World Bank world regions were reported for developmental dyslexia and vision loss. Since the prevalence estimates from most of the systematic reviews were derived from primary studies conducted in HICs, the GBD global estimates were reported along with the estimates for HICs as prevalence estimates for LMICs as a group are not reported separately by GBD (Figure 2).

Attention-deficit/hyperactivity disorder

Barican et al. reported a pooled prevalence of 3.7% (95% CI: 2.3–5.7) in children aged 4–18 years from 14 primary studies in 11 countries (23). The primary studies covered the period January 1990 to February 2021, and specifically excluded studies from LMICs. The GBD estimated the prevalence of ADHD among children 0–19 years as 1.9% (95% UI: 1.3–2.6) in 2019 (Table 1). The GBD prevalence estimate of ADHD for HICs is approximately 3.0% (95% UI: 2.0–4.2), suggesting a far lower estimate for LMICs (Figure 2).

Autism spectrum disorder

Three reviews all published in 2022 reported prevalence estimates of ASD DHD that ranged from 0.6% (95% CI: 0.4–1.0) to a median of 1.0% (Interquartile range: 1.1–4.4) (11, 24, 25). One study by Barican et al. reported estimates for ADHD and ASD, but the estimate for ASD was not considered as it was derived from only four primary studies (23). None of the reviews provided pooled estimates specifically for children and adolescents. The primary studies covered ranged from 51 to 71 articles derived from 25 to 41 countries, less than half of which were LMICs in all three reviews. One of the reviews by Wang et al. aimed to determine the prevalence of gastrointestinal

TABLE 1 Prevalence estimates of developmental disabilities in children and adolescents reported in systematic reviews (2015–2022) compared to GBD 2019 estimates.

Condition	N	References	Year of publication	Databases used	Number studies	Countries [LMICs]	Overall study size	Age group	Prevalence_Global (95% confidence interval)*	Prevalence [HICs]	Prevalence [LMICs]	Remarks	GBD 2019 [0–19 years]
Attention-deficit/ hyperactivity disorder (ADHD)	1	Barican et al. (23)	2022	MEDLINE, EMBASE	14	11 [0]	61,545	4–18 years	3.7% (2.3–5.7)	3.7% (2.3–5.7)	Not reported	Regional population of children with condition.	1.9% (1.3–2.6)
Autism spectrum disorder (ASD)	2	Zeidan et al. (11)	2022	MEDLINE	71	34 [16]	Not reported	0–89 years, predominantly below 18 years	100/10,000 (IQR: 1.09/10,000 to 436.0/10,000)	Not reported	Not reported	Regional population of children with condition not reported.	0.4% (0.3–0.5)
	3	Salari et al. (25)	2022	Science Direct, PubMed, Scopus, SID, Magiran, Web of Science, Google Scholar	74	41 [15]	30,212,757	0–27 years	0.6% (0.4–1)	Not reported	Not reported	Limited regional population of children with condition reported.	
	4	Wang et al. (26)	2022	PubMed, EMBASE, Web of Science	51	25 [6]	548,413,748	All ages, predominantly school children	98/10,000 (81/10,000–118/10,000)	85/10,000 (67/10,000–105/10,000)	155/10,000 (111/10,000–204/10,000)	Limited regional population of children with condition reported.	
Cerebral palsy	5	McIntyre et al. (10)	2022	MEDLINE, EMBASE	41	27 [6]	Not reported	0–18 years	Not reported	1.6/1,000 (1.5–1.7) live births	3.4/1,000 (3.0–3.9) live births	Global and regional population of children with condition not reported.	0.9% (0.8–1.0)#
Developmental intellectual disability (DID)	6	McKenzie et al. (27)	2016	PubMed, MEDLINE, EMBASE, PsycINFO, Cochrane	18	9 [2]	Not reported	Child & adolescent	0.22–1.55%	Not reported	Not reported	Pooled estimate not reported. Highest reported estimate came from USA in 1996.	3.1% (2.3–3.8)
Epilepsy	7	Fiest et al. (28)	2017	MEDLINE, EMBASE	24	42 [34]	Not reported	0–9 years 10–19 years	5.19/1,000 (3.54–7.62) [0–9 years]; 8.86/1,000 (6.58–11.92) [10–19 years]	Not reported	Not reported	Regional population of children with condition not reported.	0.7% (0.6–0.9)

(Continued)

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TABLE 1 (Continued)

Condition	N	References	Year of publication	Databases used	Number studies	Countries [LMICs]	Overall study size	Age group	Prevalence_Global (95% confidence interval)*	Prevalence [HICs]	Prevalence [LMICs]	Remarks	GBD 2019 [0–19 years]
Hearing loss	8	Wang et al. (29)	2019	MEDLINE, EMBASE	88	39 [23]	3,360,850	0–18 years	13.1% (10.0–17.0) [>15 dBHL]; 8.1% (1.3–19.8) [>20 dBHL]	Not reported	Not reported	Global and regional population of children with condition not reported.	4.0% (3.5–4.5)
Vision loss	9	Yekta et al. (12)	2022	PubMed, Scopus, and Web of Science	80	28 [19]	769,720	0–19 years	12.72% (9.26–16.19) [UCVA of 20/40 or worse in better eye]; 7.26% (4.34–10.19) [UCVA of 20/60 or worse in better eye]	Not reported	Not reported	Global and regional population of children with condition reported.	1.3% (1.1–1.5)
Developmental dyslexia	10	Yang et al. (30)	2022	PubMed, EMBASE, Web of Science, Cochrane, EBSCO host, ProQuest, Springerlink, & 5 Others^	58	16 [6]	Not reported	6–13 years	7.10% (6.27–7.97)	7.10% (5.54–8.82)	7.10% (6.10–8.20) [MICs]	Global and regional population of children with condition reported.	Not Available

GBD, Global Burden of Disease (GBD); LMICs, Low- and middle-income countries; MICs, Middle-income countries; HICs, High-income countries; UCVA, Uncorrected visual acuity. *Except stated otherwise, #Derived from GBD-WHO Rehabilitation Need Estimator Database, ^China National Knowledge Infrastructure, Wanfang, CQ-VIP, China Hospital Knowledge Database, OATD database.

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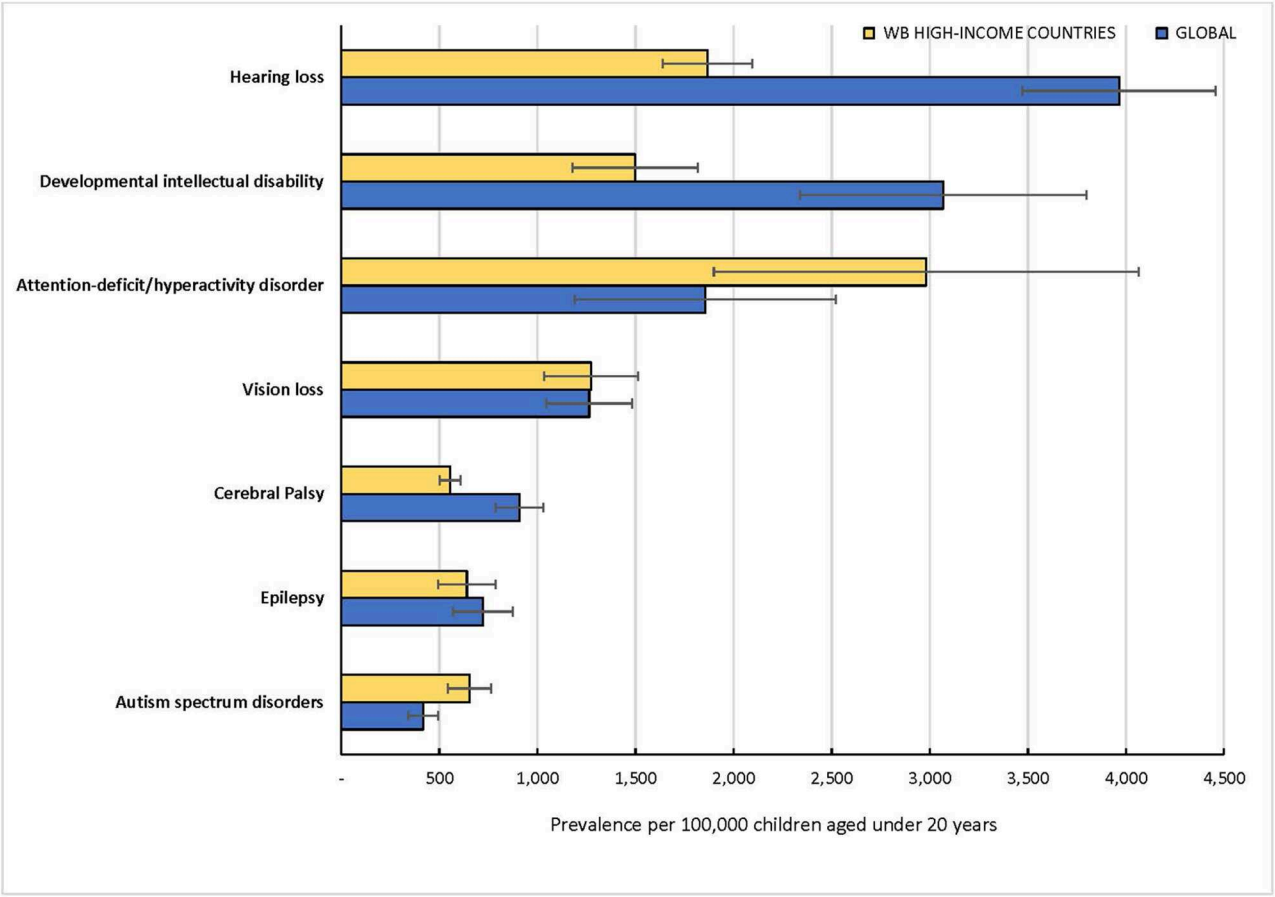

FIGURE 2
Prevalence estimates of selected developmental disabilities in children under 20 years in 2019 by the Global Burden of Disease (GBD) Study Group.

symptoms in individuals with ASD and reported pooled estimates of ASD for HICs (0.9%, 95% CI: 0.8–1.2) and LMICs (1.6%, 95% CI: 1.1–2.0) (25). Four of the 51 primary studies in this review involved individuals older 20 years or older and the selected studies were published between 2001 and 2022. Regional estimates were reported for Africa (3.0%, 95% CI: 2.5–3.4), Oceania (2.6%, 95% CI: 1.6–3.8), the Americas (1.3%, 95% CI: 1.1–1.6), Asia (0.3%, 95% CI: 0.3–0.4) and Europe (0.7%, 95% CI: 0.6–0.8). The GBD global estimate for ASD was 0.4% (95% UI: 0.3–0.5) with a higher prevalence of 0.7% (95% UI: 0.6–0.8) estimated for HICs (Figure 2), suggesting a lower prevalence for LMICs compared to HICs.

Cerebral palsy

The included systematic review by McIntyre et al. reported prevalence estimates for HICs and LMICs separately (10). A total of 41 primary studies were included in the review derived predominantly from surveillance registries in 27 countries, six of which were LMICs. The review covered studies published between January 2011 and November 2020 and the sample included children with birth year of 1995 and beyond. The estimated birth prevalence of cerebral palsy was approximately 0.2% (95% CI: 0.1–0.2) for HICs and 0.3% (95% CI: 0.3–0.4) for LMICs among children 0–18 years. A pooled global estimate was not reported nor estimates by geographical world regions. The meta-analysis was based on children with birth year from 2010. The GBD estimate for cerebral palsy was 0.9% (95% UI: 0.8–2.0) globally based on children with moderate to severe motor impairment (21). The prevalence estimate for HICs was 0.6% (95% UI: 0.5–0.6) which would suggest a higher prevalence for LMICs than the reported global estimate.

Developmental intellectual disability

Only one systematic review by McKenzie et al. published in 2016 was identified for this study (26). The review included primary studies published between 2010 to 2015 and no meta-analysis was conducted. There were 18 primary studies covering all age groups from 9 countries, and all but 2 countries were HICs. Prevalence was highly variable across studies and ranged from 0.22 % in 2007–2008 (USA) to 1.55 % in 1996 (USA) among children and adolescents. The GBD global estimate was 3.1% (95% UI: 2.3–3.8) and the estimate for HICs was 1.5% (95% UI: 1.2–1.8), suggesting a significantly higher prevalence for LMICs than the global estimate (Figure 2).

Epilepsy

One systematic review by Fiest et al. published in 2017 was eligible for inclusion (27). The review covered the period from 1985 to October 2013 and included 63 primary studies in all age groups (0–60+ years) from 42 countries, only 8 of which were HICs.

Prevalence estimate was reported separately for children aged 0–9 years (0.5%, 95% CI: 0.4–0.8) and children/adolescents aged 10–19 years (0.9%, 95% CI: 0.7–1.2). Overall pooled estimates for all age groups were reported separately for HICs and LMICs but not for children and adolescents. The GBD global estimate was 0.7% (95% UI: 0.6–0.9) and the estimate for HICs was 0.6% (95% UI: 0.5–0.8), suggesting a significantly higher prevalence for LMICs than the global estimate (Figure 2).

Hearing loss

The systematic review by Wang et al. published in 2019 was the only eligible study (28). The review was specifically conducted for children aged 0–18 years and included 88 articles published between January 1996 and August 2017 from 39 countries, 23 (or roughly 60%) of which were LMICs. The review computed pooled estimates at different hearing threshold levels, and the prevalence decreased as the severity of hearing loss (the threshold cutoff) increased. Prevalence estimates ranged widely from as low as 0.1% (95% CI, 0.1–0.2) when hearing loss was defined using a lower frequency average (0.5, 1, and 2 kHz) with a hearing threshold/level of 40 decibel (40-dBHL) in both ears to as high as 17.9% (95% CI: 15.9–20.0) when using a full frequency average (0.5 to 8 kHz) with a 15 dBHL threshold in 1 or both ears. Two global prevalence estimates using the most reported thresholds for hearing loss were presented: 13.1% (95% CI: 10.0–17.0) based on >15 dBHL and 8.1% (95% CI: 1.3–19.8) based on >20 dBHL. As recommended by the WHO, the GBD uses 20 dBHL threshold for all its computations. The global prevalence was estimated as 4.0% (95% UI: 3.5–4.5) while the estimate for HICs was 1.9% (95% UI: 1.6–2.1), which suggests a higher prevalence for LMICs than the global estimate (Figure 2).

Vision loss

One systematic review by Yekta et al. published in 2022 met our inclusion criteria (12). The review included 80 studies published between 1971 and 2018 from 28 countries, 19 of which are LMICs. It is the only systematic review that was specifically conducted among children and adolescents below 20 years. It was also the only review that reported estimates for all WHO regions. The global prevalence of vision loss was 12.7% (95% CI: 9.3–16.2) based on uncorrected visual acuity (UCVA) of 20/40 or worse in the better eye, and 7.3% (95% CI: 4.3–10.2%) based on UCVA of 20/60 or worse in the better eye. The GBD global prevalence was estimated as 1.3% (95% UI: 1.1–1.5) using visual acuity of less than 6/18 according to the Snellen chart, while the estimate for HICs was 1.3% (95% UI: 1.1–1.5) (Figure 2).

Developmental dyslexia

One systematic review by Yang et al. published in 2022 provided the most comprehensive and up-to-date status of children with developmental dyslexia globally (29). The review covered 58 primary studies published as far back as the 1950s until June 2021 and involved school children aged 6–13 years. A total of 58 studies were selected for the review drawn from 16 countries, 6 of which were LMICs. The pooled global prevalence was 7.1% (95% CI: 6.3–8.0%). The prevalence estimates for HICs (7.1%, 95% CI: 5.5–8.8%) and middle-income countries (7.1%, 95% CI: 6.1–8.2%) were similar. Pooled estimates based on WHO regions were also reported. However, developmental dyslexia is not included in the GBD database.

Risk of bias

The quality of the selected reviews is summarized in Supplementary Tables S2, S3. The inter-rater reliability after the first round of independent evaluation was 94.8% for the AMSTAR2 and 98.3% for JBI Checklist. Differences were resolved by consensus. For example, the AMSTAR2 required authors to provide a list of excluded reviews and justify the exclusions. This accounted for most of the discrepancies between the two raters. It was therefore agreed that reviews that reported the excluded primary studies in the PRISMA flow diagram with explanations for the exclusion should be considered as satisfying this criterion. Based on AMSTAR2, none of the reviews met the criteria for high quality and the most were either of low or critically low quality. In contrast, based on JBI checklist, none of the reviews were of low quality. In fact, 9 of the reviews were of high quality and 3 of medium quality.

Discussion

We set out to provide an overview of the pooled prevalence estimates of commonly reported disabilities in children and adolescents derived from systematic reviews and meta-analyses, published approximately midway into the SDGs and to compare the findings with estimates from alternative data sources in global health. To our best knowledge, this is the first systematic umbrella review on the global prevalence of the selected disabilities in children and adolescents. The principal finding was that sensory impairments were the most prevalent disabilities (13.1% for hearing loss and 12.7% for vision loss) while cerebral palsy was the least prevalent disability (approximately 0.2%) globally.

Another important finding was that most of the global prevalence estimates were derived from primary studies conducted in HICs and estimates for LMICs were reported for only three disabilities: autism spectrum disorder, cerebral palsy, and developmental dyslexia. The highest number of countries providing primary data for any disability was 56, which is 29% of all UN Member States that signed the SDGs. Regional prevalence estimates were only available for autism spectrum disorder, vision loss and developmental dyslexia. In contrast, the GBD estimates were available for high-income countries which gave indications on the contribution of LMICs to the global prevalence for the selected disabilities except developmental dyslexia. For example, the contributions of LMICs to the global prevalence of hearing loss and intellectual disability were substantially higher than those from high-income countries, in contrast to findings on autism spectrum disorder and ADHD.

Another notable finding was that the age groups of children reported in the reviews varied which makes direct comparison of estimates challenging. Furthermore, the global prevalence estimates reported for ADHD, autism, epilepsy, hearing loss and vision loss in systematic reviews were higher than those reported by the GBD. In contrast, prevalence estimates for cerebral palsy and intellectual

disability from systematic reviews were lower than those reported by GBD. Based on GBD data, hearing loss was the most prevalent disability (4.0%) and autism was the least prevalent (0.4%) disability in children and adolescents. The modeling techniques used by GBD for each of the disabilities and the number of countries covered would have accounted for the differences in the global prevalence estimates between the GBD and the systematic reviews. However, the pooled prevalence estimate for cerebral palsy for LMICs of approximately 0.3% does not appear to reflect the well documented disproportionately high burden of the risk factors for cerebral palsy and the reported prevalence estimates in young children in LMICs, especially in South Asia and sub-Saharan Africa (21, 30, 31). For example, in one robust population-based study in India, the prevalence estimate of up to 2.1% for neuromotor impairments including cerebral palsy was reported (31).

Another major finding was the sharp contrast in the quality rating of the included reviews from two different assessment tools. The major reason for the poor quality rating based on AMSTAR2 were that most of the reviews (8 out of 12) did not provide an explicit statement that the review methods were established prior to the conduct of the review which constitutes a major risk of bias in all included reviews (19). In addition, none of the reviews reported the sources of funding for the primary studies that were selected. For these and other reasons we concluded that the available reviews are generally not of a high quality to inform policy interventions in global health.

These findings would suggest that prevalence estimates derived from systematic review and meta-analyses are unlikely to provide comparable data for different disabilities to satisfy the requirements for policy and investment decisions in global health, especially in relation to population-level information for service planning. Prevalence estimates for geographical world regions were not available for most disabilities. More crucially, it was difficult to combine the estimates from the various reviews to determine an overall global estimate of disabilities in children and adolescents due to marked variability of study designs, methodological approaches, sampling strategies, and the diagnostic criteria used in case ascertainment (32). These limitations have accounted for the growing reliance by policymakers on alternative approaches and sources of global estimation of population health metrics including household surveys and statistical modeling (3, 22, 33). In order to address these limitations, the GBD for example, utilizes sophisticated statistical techniques to (i) optimize the comparability of data derived from various sources using different methods; (ii) find a consistent set of estimates across prevalence data; and (iii) generate estimates for locations with sparse or no data by using available information from other locations combined with covariates (22). However, it is important to clarify that GBD estimates are equally associated with several limitations which have been reported extensively in the literature (3, 21, 22). For example, The GBD methodology of estimating the prevalence of disabilities based on sequelae of the underlying health conditions or surrogates may result in over-estimation or under-estimation due to the difficulty in accurately accounting for idiopathic impairments. Behavioral conditions such as ASD and ADHD, continue to rely on sparse data in many regions, particularly LMICs. In addition, The GBD estimates for disabilities still do not fully reflect the complex and dynamic relationship between health conditions and contextual personal or environmental factors under the ICF, as such they provide a limited picture of disability. It is also important to mention that while cerebral palsy is least prevalent among the selected developmental disabilities, it is the leading cause of early-onset physical disability. Considering that cerebral palsy is lifelong and very disabling for some people, the impact in terms of disability-adjusted life years makes cerebral palsy a more significant condition from a public health perspective than its low prevalence might suggest (34). The use of live births as denominator in computing the prevalence, is also unlikely to reflect the extent of the disability in the population optimally.

A major strength of this study is that the findings from the systematic reviews were compared with the latest prevalence estimates in the GBD database, which is novel. We had previously demonstrated that the prevalence estimate of disabilities in children and adolescents (<20 years) by GBD and UNICEF were not statistically different and were statistically equivalent (3). The study also complied with the key quality measures recommended by AMSTAR2, including prior registration with PROSPERO and the provision of a separate list of excluded reviews and reasons for exclusion. Another unique feature was the quality evaluation of the included reviews using two separate risk-of-bias tools. We also included developmental dyslexia which is the most common type of learning disability, accounting for approximately 80% of all learning disabilities but rarely reported in the global health literature (29, 35).

A few limitations of this umbrella review are worth restating. First, the electronic databases searched were not exhaustive which would have resulted in a potential selection bias. For example, we excluded non-English articles, and we did not search Web of Science, Google Scholar, and regional databases such as the WHO Library (WHOLIS), LILACS (formerly Latin America Index Medicus) and African Index Medicus for additional eligible articles from LMICs, which could have biased the findings. Second, no meta-analysis of the reported estimates was undertaken, primarily due to heterogeneity in the methods, age groups and the sample sizes of the included reviews. However, umbrella reviews in general are aimed at summarizing the evidence rather than to re-synthesize primary studies (20). Third, there was wide variation in the period covered by selected reviews which would have made comparison of reported estimates across disabilities biased and inconsistent. Fourth, prevalence estimates reported for HICs frequently mask the health and social inequalities in rural and isolated areas designated as medical deserts due to inadequate access to medical care.

Conclusion

Up-to-date prevalence estimates of disabilities in children and adolescents are essential to raise awareness and inform policy initiatives, service planning, resource allocation, and research priorities. However, available estimates from systematic reviews and meta-analyses do not provide representative evidence on the global and regional prevalence of developmental disabilities due to limited geographical coverage and substantial heterogeneity in methodology across the primary studies. Population-based data for all regions that reflect and adjust for these limitations such as those reported by GBD Study periodically are warranted to inform global health policy and intervention.

Author contributions

BO drafted the manuscript. TS, FO, MN, MS, and AD critically reviewed the draft and suggested essential edits. All authors contributed to revising the manuscript and have approved of the final version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

The reviewer CS declared a past co-authorship with the author BO to the handling editor.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1122009/full#supplementary-material

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Promoting school readiness in children with developmental disabilities in LMICs

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The United Nations' Sustainable Development Goals (SDGs) explicitly acknowledge inclusive and equitable quality education as the primary goal of any global initiatives for early childhood development for children under 5 years with developmental delays and disabilities. Primary education provides the foundation for lifelong learning, vocational attainment, and economically independent living. Globally, the majority (over 90%) of children with developmental disabilities reside in low- and middle-income countries (LMICs). These children are significantly less likely to have foundational reading and numeracy skills, more likely to have never attended school and more likely to be out of primary school, compared to children without disabilities. Concerted and well-coordinated efforts to prepare these children in early childhood for inclusive education constitute a moral and ethical priority for all countries. This paper sets out to examine the concept and dimensions of school readiness for children under 5 years from an extensive narrative review of the literature. It identifies the barriers and challenges for school readiness for children with disabilities and the limitations of the available tools for evaluating school readiness. It concludes by emphasizing the critical role of inter-disciplinary engagement among pediatric caregivers in promoting school readiness in partnership with the families and community where the children reside. Overall, the paper highlights the need for appropriate policy initiatives at the global and national levels to promote school readiness specifically for children under 5 years with developmental disabilities in LMICs, if the aspirational goal of inclusive education by 2030 under the SDGs is to be realized.

KEYWORDS

school readiness, inclusive education, developmental disabilities, early detection, early intervention, SDG 4, developing countries

Introduction

Developmental disabilities (or simply "disabilities" hereinafter) are chronic physical, cognitive, speech or language, psychological, or self-care conditions that typically originate during childhood; are likely to continue indefinitely; and require additional coordinated services, support, or other assistance for an extended duration or during a lifetime (1, 2). These conditions include but not limited to hearing impairment, vision loss, cerebral palsy, epilepsy, intellectual disability, autism spectrum disorder, attention-deficit/hyperactivity disorder, speech and language disorders, and specific learning disabilities. Globally, more than 50 million children aged under-5 years are estimated to have disabilities (3). A recent report from UNICEF suggests that, compared to children without disabilities, children with disabilities are 42% less likely to have foundational reading and numeracy skills, 49% more likely to have never attended school, 47% more likely to be out of primary school, 33% more

likely to be out of lower-secondary school, 27% more likely to be out of upper-secondary school, and 20% less likely to have expectations of a better life (4). The United Nation's Sustainable Development Goals (SDGs) have provided the political and policy framework for ensuring that children under-5 years with disabilities are promptly identified and supported to benefit from inclusive and equitable quality education (5). SDG 4.2 specifically calls for actions to ensure that all girls and boys have access to quality early childhood development (ECD), care and pre-primary education so that they are ready for primary education by 2030. Thus, school readiness is a critical component of the global health agenda for children under 5 years with disabilities. This has been reinforced by the 2015 Incheon Declaration and Framework for Action for the implementation of Sustainable Development Goal 4 (Education 2030) led by UNESCO (6). It is also consistent with the United Nations Convention on the Rights of the Child (7), and the United Nations Convention of the Rights of Persons with Disabilities (8).

In this mini-review, we set out to: (i) examine the concept and dimensions of school readiness with respect to inclusive education among children under 5 years with disabilities; (ii) identify the barriers and challenges for school readiness for children with disabilities from the perspective of child, school and family/community; (iii) examine the limitations of the available tools for the evaluation of school readiness; and (iv) highlight the role of pediatric caregivers in facilitating school readiness for children with disabilities in low- and middle-income countries (LMICs). The articles and reports used in this narrative review were identified through targeted searches of the PubMed, Scopus and Google using the terms "school readiness" and "childhood disability." Additional articles were identified from the references of selected publications and reports.

The concept and dimensions of school readiness

School readiness is a measure of the preparedness of a child, with age-appropriate physical and emotional wellbeing as well as social, language and cognitive or intellectual competencies to succeed in school. The concept of preparedness and competencies for school readiness has evolved with time from a maturational construct (wherein the maturity level of the child was solely responsible for the attainment of appropriate skills helpful for success in school) (9), to a social construct (wherein the child has an active role in becoming ready for school through a wide range of interactions between the child and his environment) (10).

School readiness comprises three interconnected dimensions: the readiness of the individual child for primary school enrolment and participation; the school's readiness to provide optimal learning environment for the child; and family and community supports that contribute to child readiness for school, as depicted in Figure 1 (11, 12). "Ready children" have skills, abilities and attitudes that are required for a smooth and successful transition to school, such as, self-regulation, early literacy, early numeracy, motor, social-emotional, and executive function skills. "Ready schools" have appropriately trained teachers and high quality of support services to provide smooth transitions for children irrespective of their abilities and at their own pace. Family and community readiness involves parenting beliefs, attitudes, and practices, which are quite varied across cultures and socio-economic groups, as well as community support. These dimensions are applicable to all children. However, children with disabilities have peculiar challenges that require special attention over and above those without disabilities in order to foster school readiness for inclusive education.

School readiness for children with disabilities

In line with SDG 4.2, school readiness for children with disabilities must be geared toward inclusive education that allows full and effective participation, accessibility, attendance, and achievement along with children without disabilities (6–8). An overview of the three dimensions of school readiness for children with disabilities is presented below.

Child's readiness for school

The domains of school readiness for any child typically include (i) Health and Physical Development, ii) Emotional WellBeing and Social Competence, (iii) Approaches to Learning, iv) Communicative Skills, and v) Cognition and General Knowledge (13). When children's physical health forms the basis for the development of school readiness skills and successful transition to school (14), the school readiness skills of children with disabilities assume greater importance as they are less likely to engage in the process of education itself. Compared to other children, those with disabilities are less likely to start school, have lower levels of attendance, have lesser chance for higher education, and have lower school retention rates (15–17).

Lack of access to timely detection and intervention services is perhaps the greatest barrier to school readiness faced by children with disabilities (18). Routine newborn screening and developmental monitoring are generally not offered in many LMICs. Where services exist, poverty, discrimination, stigma, and abuse may constitute additional barriers (19). As a result, these children falter in all the essential domains of child development for school readiness (13). Specific disabilities are also associated with unique challenges. For example, children with Autism Spectrum Disorders may experience less emotional readiness as they have more externalizing behaviors and difficulties with self-regulation which adversely affects their engagement in the classroom activities as well as social interactions with teachers and peers (20).

Studies also show considerable impairment in cognition and general knowledge, lower academic scores, increased grade retention and dropout rates among children with ADHD (21). This is because hyperactivity and impulsivity affect social interactions and the so-called normal classroom behaviors like paying attention to the teacher or activities, being able to sit still in the class etc., and interpersonal issues due to poor emotional control.

Preschool age children with Cerebral Palsy have been found to perform well below their peers in areas of mobility, self-care, social interactions, and communication skills. Hence, the need for timely screening and intervention for these children so as to prepare them

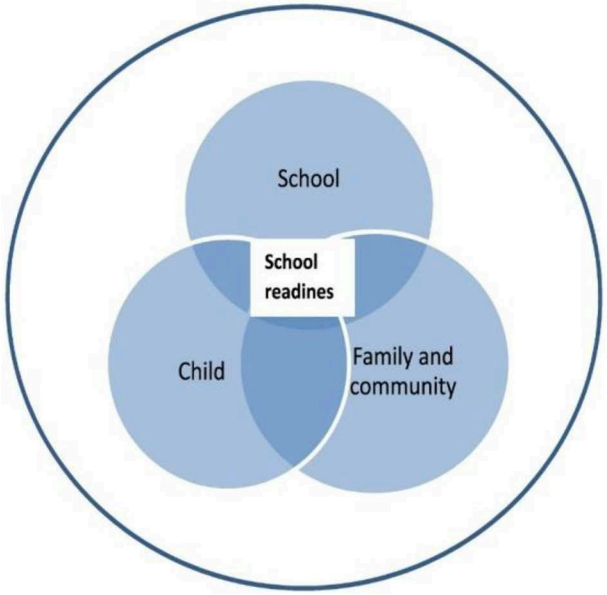

FIGURE 1
UNICEF's model for school readiness (12).

for school entry (22). Similarly, studies show that enrolment in early intervention services for deaf or hard-of-hearing children well before 6 months of age help establish healthy trajectories of early childhood development, thereby reducing later academic challenges (23).

School's readiness for the child

For schools to be ready to provide developmentally appropriate education for children with disabilities in an inclusive educational setting, they need to satisfy diverse learning needs and preferences in the present-day classrooms. As one of the goals of inclusive education is not only to accept children with disabilities, but also to welcome them, schools need to bring about systematic changes not only in the way schools' function, but also in the attitudes, beliefs, and value systems of all stakeholders of the school including families and community at large. Studies have shown that although children with disabilities liked attending school most of the time they are discouraged by discrimination, prejudice and non-acceptance from peers (24). Those in school are all too often excluded within the school setting and are not placed with peers in their own age group and receive poor-quality learning opportunities. Hence, the need for proper guidelines for implementing inclusive education in schools (25).

Studies conducted in LMICs have shown that teachers do not have adequate knowledge about disabilities and inclusive education and only few teachers receive requisite hands-on training beyond lectures. As a result, many teachers lack confidence in teaching children with disabilities resulting in the belief that children with disabilities should be taught out of mainstream education system (19). Many schools also lack infrastructural facilities to meet the needs of children with different forms of disability. Schools, therefore, need to be adequately funded and equipped to receive children with disabilities. UNICEF's Child Friendly Schools (CFS) can be considered as a model of school's readiness for children with disabilities (26). The characteristics of child friendly schools are: (i) child-centered approach in teaching and learning; (ii) hygienic; (iii) healthy; (iv) safe-adherence to safety regulations in construction of buildings and playgrounds; (v) protective; (vi) gender sensitive and (vii) inclusive. CFS also links the three dimensions of school readiness by involving the family and community in children's learning and development (26).

Family/community's readiness for school

Parents play a crucial and indispensable role in fostering school readiness of children with disabilities. Parents act as decision makers on behalf of their children and assist others in making decisions about their children in school related matters. They act as teachers not only at home but also as partners in the classroom. And their role as an advocate for their child also makes them the most important group in the school community setup (25).

However, parents in LMICs must overcome several personal and societal challenges in getting their children with disabilities ready for school. Studies have shown that the main obstacles to transition to primary school for children with disabilities in sub-Saharan Africa are related to stigma, financial limitations including costs to the family, resources in school and travel (19, 24, 27). Problems associated with accessing health care and education facilities also affects children's health, development, and education as these programs and services may often be costly, not inclusive and situated in urban areas. Although some countries have a good network of community-based services for children, there is a dearth of knowledgeable and skilled service providers for disability. Challenges in physically reaching the schools is also a factor affecting schooling for children with disabilities in some communities. Children with disabilities have also been found to miss out on essential vaccinations and basic treatment for common childhood illnesses which compromises their school readiness and smooth transition to formal education (28). Parental empowerment and community enlightenment are needed to foster school readiness for children with disabilities. Parental perception on disabilities, their concerns about school, their perception of benefit from schooling to their child with a disability must also be considered and addressed as appropriate.

Evaluation of school readiness

Even after almost 50 years of research, the concept of School Readiness and its assessment is an area wherein a consensus among the stakeholders is still emerging (29). Evidence in the literature shows varied approaches to the dimensions of school readiness assessment such as the age at which school readiness should be assessed, which is dependent on the transition age to primary school and varies with the education policies of each

country (27, 30–33). The types and dimensions of assessment, as well as the reliability and validity of the assessment tools, especially when test scores form the basis for denial of entry or admission to special education are important. Additional considerations include who the assessor should be, the settings and frequency of assessments, cultural sensitivity of assessment tools, communicating school readiness status of children with their parents and using readiness data for other purposes of curriculum planning. However, the appropriateness of school readiness tools for children with disability remain largely untested in LMICs (27). A summary of available tools is presented in Table 1.

Evaluating child's readiness

Tools for assessing school readiness in children in general are varied and consists of screening tests, diagnostic tests, and generic school readiness tests. It was observed that only few instruments considered the contextual aspects of children's learning, the quality of environment (34, 35), the individual and group differences in the patterns of child development as well as impairment or disability (31). However, most of the tools conserved the biological-maturational aspect linked to the achievement levels in various domains of development suitable for each age.

For young children (0–6 years), there are five conditions for which routine screening programs have been recommended and implemented in several countries: (i) congenital metabolic conditions, (ii) hearing, (iii) vision, (iv) developmental and behavioral disorders, and (v) autism spectrum disorder (ASD) (36). School Readiness module and scale to assess the outcome of the intervention in pre-schoolers with autism spectrum disorder has been developed and validated in a developing country but is yet to be widely used (37). Some 32.6% of 4-year-olds assessed using The Jamaica School Readiness Assessment (JSRA) in Jamaica in 2017 and 2018 were identified as having at least one developmental problem (36). Early Development Instrument (EDI), a teacher administered tool for assessing the development of children in the age group of 3.5–6.5 years, has been widely used in Canada and is in use in Ethiopia, Malawi, and Mozambique (38). The International Development and Early Learning Assessment (IDELA) is a global tool administered by trained enumerators to assess early learning and development of children in the 3.5–6-year age group (39); but school readiness threshold is not available and certain IDELA score range is not indicative of developmental delay. IDELA has been used in 45 countries and has been adapted for use in Bhutan (31). Malawi Development Assessment tool is another tool with good specificity in identifying developmental delay in children from low-income settings. This has been used in Zimbabwe, Pakistan, Kenya, Uganda, Bangladesh, Tanzania, and Nepal (39).

Lastly, the Nursery Evaluation Scale Trivandrum (Abridged Version) is a simple, cost-effective screening tool to assess the development of children from 48 months to 72 months to be used in the community settings by community health workers (40). The 3rd, 50th, and 97th percentile age placement in months have been provided.

Evaluating school's readiness

School Assessment Tool (Reflection Matrix) has been designed to assist the stakeholders of the school community to assess the current family and community engagement practices and thereby implementing strategies to strengthen them (41). This assessment tool helps schools understand their position on the continuum of engagement and where further development is required. This tool aligns with the seven dimensions of Family-School Partnerships Framework: (i) communicating; (ii) connecting learning at home and at school; (iii) building community and identity; (iv) recognizing the role of the family; (v) consultative decision-making; (vi) collaborating beyond the school; and (vii) participating (41, 42). This tool can be culturally adapted for LMICs because of its simplicity.

Government of India launched Accessible India Campaign (Sugamya Bharat Abhiyan) in 2015 to achieve universal accessibility for persons with disabilities. A checklist was developed to assess the accessibility of schools in India for children with disabilities as part of the guidebook titled: "Making Schools Accessible to Children with Disabilities" (43).

Evaluating family's readiness

Specific tools aimed at assessing family's readiness for school are rare, even in high-income countries. A tool currently used in Australia under the Albuquerque Public Schools Family and Community Engagement Policy, addresses issues that may be considered in evaluating parent engagement in school readiness (44).

Intervention programs for school readiness

Evidence shows that disadvantaged students with or at risk of disabilities are those making the most dramatic gains from ECD programs and in turn from school readiness programs (45). Examples of intervention programs to facilitate school readiness in children with disabilities include the "Head Start Program" in the USA (46), and the Integrated Child Development Services (ICDS) in India (47).

Head Start Programs (USA)

The "Head Start" and "Early Head Start" Programs were launched in 1965 targeted at children from birth to 5 years of age hailing from low-income families, and foster care systems. The services are offered at no charge to parents. Children with disabilities and special needs are also catered for in the Head Start Programs. The Early Head Start component caters to the needs of expectant mothers, infants, and toddlers and are mostly provided in the child's own home through weekly home visits, while the Head Start Program is aimed at promoting school readiness for all children 3–5 years of age through center-based activities (46). The

TABLE 1 Instruments for assessing child/school and family readiness.

(a). Instruments for assessing child's readiness								
Name of instrument	Assessor	Functional domains	Age group	Feasibility	Reliability	Validity	Scoring	Experience with total population implementation
1. The Jamaica school readiness assessment (JSRA)	Teacher	JSRA has three components: The Eleven Question Screen (EQS) an adapted version of ten question screening, the child behavior rating scale and the early learning scales. The functional domains assessed are development, behavior, early literacy skills, and early numeracy skills, approaches to learning	4 years −4 years 11 months	Feasible for classroom settings where teacher completes the questionnaires based on observation. Based on the normative cutoff points decisions about further evaluations are made	The standardized alpha for the approaches to learning (0.81), early literacy (0.89), and early numeracy (0.87) areas indicated strong internal consistency for all three areas. Internal consistency was also examined for the CBRS, and the standardized alpha was 0.86, also indicating strong internal consistency	Original study showed high sensitivity and specificity for original TQS The CBRS has demonstrated strong predictive validity with reading and math achievement in elementary grades and validated in different cultural contexts	Cut off scores for each of the component instruments has been identified for comparison against normative sample	In Jamaica, Bangladesh and Pakistan TQS had relatively poor sensitivity for serious vision and hearing disorders that had not been previously identified and a low positive predictive value of less than 25% for serious disability. Hence positive screen result therefore needs to be followed by a clinical diagnostic evaluation to confirm the presence or absence of disability
2. Early development instrument (EDI)	Teacher/educator	Physical health and wellbeing, social competence, emotional maturity, language and cognitive development, communication skills and general knowledge	4 to 7 years	An easy to administer paper pencil/digital three-point Likert type scale which can be administered with minimal training, requires only 15–20 minutes for Individual child. This instrument is intended to collect individual child's data but results are not interpreted for individual child and not for diagnostic purposes	Internal consistency (alpha) ranged from 0.84 to 0.96. Test-retest reliability coefficients ranged from 0.82 to 0.94. Inter-rater reliability (as measured by correlation of school-teacher and daycare teacher scores, as well as parent-teacher scores) ranged from 0.36–0.80	Validity studies based on Content validity, response processes, internal structure as well as in relation with other variables like social competence, physical health, emotional maturity, language development three years after initial EDI administration as well as academic outcome at the end of first grade demonstrated good validity	Percentile cut-points, and norm-referenced scores (based on national results from Canada) are available for comparison. Children who score in the lowest 10th percentile on one or more domains are categorized as vulnerable	EDI was finalized in 2000 in Ontario. Most provinces continue to implement the EDI on a regular basis. Many countries have implemented the EDI with suitable adaptations to local settings to ensure validity and relevance across settings
3. The international development and early learning assessment (IDELA)	Trained enumerator/ community member	Early numeracy, early literacy, social-emotional development, and motor skills	3.5–6 years	Direct individual skill assessments of children are done for all the 22 items on the instrument through direct child interview and observation, which takes ∼30 min for each child. Requires minimal set of materials for administering the test	High inter rater reliability was observed in different settings	All domains of development measured by IDELA are predictive of later academic performance in Early primary school, and the domains of Emergent Literacy and Emergent Numeracy are the strongest predictors of Early Grade Reading Assessment and Early Grade Maths Assessment. Internal consistency calculations were performed for both the overall IDELA instrument and four of the subscales for the countries where IDELA has been administered	75% correct scoring is considered as fine mastery and 25% correct scoring is considered as struggling for overall assessment s and for a particular functional domain	IDELA has been used in 45 countries to assess the ECE interventions aimed at achieving SDG 4.2 goals. Further predictive validity studies that investigate whether there are IDELA score ranges associated with better primary school outcomes are needed before performance benchmarks can be established as per the original study

(Continued)

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TABLE 1 (Continued)

(a). Instruments for assessing child's readiness								
Name of instrument	Assessor	Functional domains	Age group	Feasibility	Reliability	Validity	Scoring	Experience with total population implementation
4. Malawi development assessment tool**	Trained health worker	Gross motor, fine motor, language, and social skills	0–6 years	Technically sound and suitable for African rural settings. Could be used by with little training and the items are easy to understand as pictorial representations of many items are provided in the tool.	Overall, reliability was excellent ($k > 0.75$) for 99% (134/136) of interobserver immediate reliability this table, for 89% (121/136) interobserver delayed reliability, and 71% (96/136) of intra-observer–delayed 2-wk assessments	Very high sensitivity (97%), and specificity 82%	Age norms for 25, 50, 75, and 90% percent of the children passing each item was determined which acts as normal reference values for each functional domain milestones	Authors have mentioned that limited resource settings can use this scale for initial assessment of children's development as well as outcome measurement tool for interventions
5. Nursery evaluation scale Trivandrum (Abridged version)	Community health worker	Gross motor development, fine motor development, cognitive development, receptive language development and personal social and expressive language development	48 months-72 months	Brief, simple, cost effective and easy to administer screening tool which requires minimal training and less time for administering in community setting. It provides scope for continuous evaluation of children to monitor their progress after offering inbuilt intervention programs for each item.	NEST abridged is a shorter version of NEST Full version. Psychometric studies of NEST full version have been published in the Indian Academy of Pediatrics Textbook Vth Edition*	Psychometric studies of NEST full version have been published in the Indian academy of pediatrics textbook	3rd, 50th, and 97th normative Percentile age placements for each item is available for comparison	Large population experiences are yet to be documented for NEST abridged version although it is available for NEST Full version
(b). Instruments for assessing school's readiness								
1. School assessment tool (reflection matrix)	Members of school community	It includes seven dimensions of family-school partnerships framework: (i) communicating; (ii) connecting learning at home and at school; (iii) building community and identity; (iv) recognizing the role of the family; (v) consultative decision-making; (vi) collaborating beyond the school; and (vii) participating	Not applicable	Contains individual, school and group assessment proformas. easy to administer, the results of individual assessments are collated onto group assessment proforma. These results after discussion with the members about the school's current stage on each dimension is entered into the school profile overview proforma and the differences in rating between groups discussed and action plans formulated	Not available	Not available	Not available	Not available

(Continued)

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TABLE 1 (Continued)

Name of instrument	Assessor	Functional domains	Age group	Feasibility	Reliability	Validity	Scoring	Experience with total population implementation
2. Checklist to assess the accessibility of schools for children with disabilities	Parents, school administrators, school management committee, civil works personnel	Entry/exit, ramps, stairs, corridors, signage, doors, boards, windows, flooring, drinking water units, toilets, playgrounds and emergency preparedness	Not applicable	The checklist outlines access requirements to comply with the diverse needs of all children, including children with disabilities and to use the guidebook to understand as to improve the accessibility by working on areas identified as requiring improvement. This can be used in planning, designing and implementation of school related construction works or for self-assessment, monitoring and maintenance purpose, third party audits, advocacies for improving accessibility to schools etc.	Not available	Not available	Yes or NO response with a remarks column for noting observations and reference column indicating the required section in the guidebook for improving particular design element	Not available
(c). Instruments for assessing family's readiness								
1. Family engagement best practices rubric and assessment	Individuals, teacher groups, family groups, student groups or by the whole school community	Communication, strengthening relationships and capacity, connecting learning at home and at school, recognizing the role of the family, shared decision making, collaborating with community and participating	Not applicable	Based on the individual assessment family engagement action plan to be prepared	Not available	Not available	Three stages of, Developing, Building, Sustaining, within each element to represent a continuum of engagement based on YES/NO/ DON'T KNOW responses for each statement	Not available

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Head Start Program is highly successful and exemplifies a useful framework for developing culturally appropriate intervention programs in LMICs.

Integrated child development services (ICDS)—India

Integrated Child Development Services launched in 1975, is one of the world's largest and unique ECD programs (47). The objectives of this program are: (i) to improve the health and nutritional status of children under 6 years; (ii) to lay the foundation for the physical, psychological, and social development of the child; (iii) to reduce malnutrition, mortality, morbidity as well as school dropout rates; (iv) to promote inter department coordination at the policy as well as implementation level so as to promote child development; and (v) to enhance mother's capability to meet the health and nutritional requirements of their children through proper health and nutrition education. ICDS focusses on an integrated and life cycle approach in delivering services to its beneficiaries: children under 6 years of age, pregnant women, and lactating mothers. All the services of ICDS are provided through its grassroot level center called the Anganwadi center, manned by Anganwadi worker and an assistant. The services provided to children under 6 years of age, adolescent girls, and pregnant and lactating mothers through Anganwadi are: supplementary nutrition (to bridge the gap between the Recommended Dietary Allowances (RDA) and the Average Daily Intake (ADI) of the target group), health check-up, referral services and immunization. ICDS also aims at breaking the vicious cycles of malnutrition, mortality and morbidity and reduced learning capacity as well as provide non formal education to children between 3 to 6 years of age (47).

Anganwadi workers have been trained in identifying developmental delay in children from birth to 2 years of age using Trivandrum Developmental Scale developed at Child Development Center, Trivandrum and to assess school readiness as a continuous assessment program using Nursery Evaluation Scale Trivandrum in 2- to 6-year-old children. Anganwadi workers are also trained in providing family Life education sessions to adolescents belonging to their Anganwadi area. In the financial year 2021, more than 89 million mothers and children had benefited from ICDS (48). One evaluation study conducted in three states in India demonstrated that ICDS also has a significant benefit for the mental development of the children (49).

Role of pediatric caregivers in promoting school readiness

The scientific, ethical, and political framework for optimizing school readiness for inclusive education for children with disabilities as envisaged by the SDGs has been reported in the literature (18, 50, 51). Pediatric caregivers, including nurses, physicians and other primary care professionals, community health workers and rehabilitation specialists have a significant role in promoting school readiness for all children, right from birth through pediatric consultations as well as advocacy (52, 53). Available evidence from both pediatrics and education shows that children with disabilities start school farther behind than their peers without disabilities (4). Inter-disciplinary work between pediatrics and education to drive the implementation of evidence-based solutions will ultimately improve the developmental trajectory for better outcomes for these children. For instance, the healthcare system is the only sector that enjoys highest contacts with children before school entry, particularly, through routine immunization programs in communities with high rates of births outside hospitals. National guidelines similar to the policy document from the American Academy of Pediatrics (AAP) on early detection and intervention provide caregivers with opportunities for improving physical, socio-emotional and educational health of young children with other advocacy groups (53). Ensuring children's regular and timely visits to the well-child clinics is a way of ensuring healthy child development and school readiness. These visits, apart from screening for risks factors and the early identification and intervention for disabilities, provide opportunities for pediatric caregivers to monitor and ensure parental education on children's growth, development, and nutrition, handling behavioral issues, as well as the importance of quality parent-child interaction within a positive home environment. The importance of family-centered services cannot be over-emphasized (54–56).

Community support systems through home visits can be used for promoting school readiness, family support programs and early intervention services (57). Kindergarten screening, rather than a gatekeeping test for age-eligible children to enter school should be a tool to guide planning, curriculum, and instruction to support developmental and academic achievement for diverse groups of children. A school readiness curriculum for increasing the pediatric resident's knowledge and confidence in addressing school readiness in clinics has also been developed and evaluated for pediatric residents (58). The International Pediatric Association has also issued a position statement that addresses the training needs of the pediatric service providers (59). These recommendations can be adapted for use in LMICs within the pediatric community of caregivers to ensure that efforts to facilitate early detection and intervention for children with disabilities are appropriately geared toward school readiness.

Conclusion

Inclusive education has been acknowledged as a global priority for children with disabilities under the SDGs. However, there is limited evidence of progress toward systematic promotion of school readiness in LMICs across the dimensions of child readiness, school readiness and family/community readiness. Intervention programs in early childhood for children with disabilities are still not explicitly structured and evaluated to facilitate school readiness for inclusive education. Policy interventions to address barriers to school readiness for inclusive education among families, the community, and

schools at the country level in LMICs should be considered. Additionally, there is an urgent need to train and empower all pediatric health caregivers to recognize and embrace school readiness for children with disabilities as an early childhood development priority as envisioned by the SDGs framework for global child health, inclusive education, and development.

Author contributions

MN and RR conceptualized and drafted the manuscript. BO critically reviewed the draft and suggested essential edits. All authors contributed to revising the manuscript, and approved the final version as submitted.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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The potential of COPCA's coaching for families with infants with special needs in low- and middle-income countries

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Infants at high biological risk of or with a neurodevelopmental disorder run a high risk of delayed school readiness. This is especially true for infants in low- and middle-income countries (LMICs). This perspective paper first summarizes evidence on intervention elements that are effective in promoting family well-being and child development in infants at high biological risk in high income countries. Crucial elements are family centeredness, goal orientation, a home setting, focus on activity and participation, and challenging the infant to explore the world and the own body by means of self-produced movements. The studies revealed that coaching as applied in COPCA (COPing and CAring for infants with special needs) is a pivotal element determining the success of intervention.The paper continues by describing COPCA and its coaching. Next, we report on two pilot studies addressing COPCA's implementation in Brazil. Finally, we discuss why COPCA is a promising early intervention program for infants at high biological risk of neurodisability in LMICs: COPCA is adapted to the families' strengths and needs, it empowers families and promotes child development therewith facilitating school readiness. Moreover, it may be delivered by tele-coaching therewith eliminating families' burden to travel to distant intervention clinics.

KEYWORDS

family-centered, early intervention, low-and middle-income countries, high risk infants, COPCA, coaching

Introduction

Infants at high biologically high risk of neurodevelopmental disability are, for example, infants born preterm or infants with a neonatal hypoxic-ischemic encephalopathy. Neurodevelopmental disability consists of a heterogeneous group of disorders, including cerebral palsy (CP), intellectual disability and autism spectrum disorders (1). The disorders affect multiple domains of activities and participation, such as mobility, learning and applying knowledge and communication (1). The presence of neurodevelopmental

Abbreviations
AIMS, albert infant motor scale (AIMS); APAE, associação de pais e amigos dos excepcionais; COPCA, COPing with and CAring for infants with special needs; CP, cerebral palsy; GMFM, gross motor function measure; HICs, high-income countries; LMICs, low- and middle-income countries; PNAISC, brazilian national policy on integral attention to the health of the child; SUS, sistema unico de saúde.

disability puts children at risk of limited school readiness. This is true for children world-wide, but the problem of limited school readiness is particularly pressing in low-and middle-income countries (LIMCs), as a high proportion of children with neurodevelopmental disability live in these countries (2) where early and appropriate intervention support to children and families is less available due to financial issues or limited accessibility (3).

It is generally agreed that infants and children at high risk of or diagnosed with neurodevelopmental disability should receive early intervention (4, 5). Literature suggests that effective early intervention programs are family-centered, goal-oriented, occur in the home setting in an enriched environment, focus on activity and participation, and challenge the infant to explore the world and the own body by means of self-produced motor behavior with trial and error. In addition, constrained-induced movement therapy or bimanual training are recommended for infants with clear asymmetries or unilateral CP, and early provision of assistive devices is recommended in infants who in early life already show substantial mobility limitations, e.g., due to a brain lesion. It is gradually acknowledged that children benefit more from the implementation of development-enhancing strategies during daily activities than from intervention activities more or less restricted to the intervention sessions themselves, as the child has more opportunities to practice in the former than in the latter situation. Coaching of the family members is a successful and modern means to let families appreciate how they in their own way can promote their child's development (1, 4).

Coaching is increasingly applied in early intervention and pediatric rehabilitation to foster family empowerment and child development. However, the application of coaching approaches confronts health professionals with challenges involving changes in professional role and associated behavior, and acquisition of coaching skills (6). Examples of coaching approaches designed for this field, with growing evidence for the effectiveness of coaching are "Coping with and caring for infants with special needs" (COPCA) (7, 8), Occupational Performance Coaching (OPC) (9), and Solution-Focused Coaching in Pediatric Rehabilitation (SFC-peds) (10). In COPCA positive associations between coaching of family members and (a) infant mobility and (b) empowerment and quality of life of the family have been demonstrated (11–14). OPC has been associated with positive effects on parents' self-efficacy and self-competences and on participation and occupational performance of children with neurodisability (15). Other studies suggest that SFC-peds is beneficial for the attainment of participation and friendship goals and increased sense of empowerment of children and youth with disabilities, and for the enhancement of skills and knowledge of their parents. All these approaches are family-centered and use reflection and feedback as intervention strategies (16–18). COPCA does not use video-feedback to coach families. Video-feedback is, for instance, used in situations in which families have established already problematic interactions with their children (19). In these situations, video-feedback helps family members to discover and correct maladaptive behavior. In COPCA, i.e., in the situation of intervention in early childhood the situation is different, maladaptive interactions did not have time to develop. COPCA's goal is to enhance the families' own capacities to solve problems. To this end, it uses dialogue with the family, shared observation of daily care giving activities (without video), and provision of hints and suggestions.

Coaching is a major ingredient of the early intervention program COPCA. In the following sections of this perspective paper, we first describe COPCA and the characteristics of COPCA's coaching. In the next section we report on two pilot studies addressing COPCA's implementation in Brazil. In the last section we discuss why COPCA, and its coaching strategy turns COPCA into a promising early intervention program for infants at high biological risk of neurodisability in LMICs: COPCA is adapted to the family's strengths, needs and culture, it empowers the family, it promotes child development and—ultimately—this will result in increased school readiness. Moreover, COPCA may be delivered by tele-coaching therewith eliminating the family's burden to travel to distant intervention clinics.

COPCA and coaching in COPCA

COPCA is a family-centered early intervention program, which includes all above mentioned components (7, 8). Becoming a COPCA coach requires a professional education course of 3×2 days and two individual coaching sessions of one hour (8). COPCA has been designed for infants at high biological risk of neurodisability. COPCA has two aims: 1) to enhance empowerment of individual families in the process of decision-making regarding activities and participation of child and family; and 2) to promote infant development in general and especially the child's mobility allowing for optimal participation in daily life and to prevent contractures and deformities.

Coaching is COPCA's major strategy. The goal of coaching is to empower family members to discover their own strategies, capacities, and competences to challenge the infant with special needs in naturally occurring parenting situations. COPCA's coaching approach is goal-oriented and complies with the three criteria of Ives (20): it is non-directive, solution-focused and performance driven. Being non-directive implies that the coach is a facilitator and stimulator of ideas and actions and not a trainer or instructor. Solution-focused implies a focus on finding solutions to achieve specific aims. Being performance driven emphasizes the focus on changing actions to improve performance through understanding of circumstances.

In COPCA family members are equal and active partners in the intervention. They are actively involved goal setters, decision makers and supporters of the child with special needs. They are engaged in daily care activities in naturally occurring parenting situations. In COPCA health professionals act as a coach. In this role health professionals observe, listen, ask, and provide information. The coach honors families as experts of their lives and believes that every family member is creative and resourceful. In coaching, relationships between family members and health professionals are of critical importance.

TABLE 1 Coaching strategies in COPCA.

Coaching Strategies	Definition
Information exchange	Information exchange means all communication tuned to the guidance of the infant and the family as an entity. This includes exchange of knowledge, and exchange of information related to the development of the infant or the actual situation of the infant and family.
Active listening	Active listening implies to listen attentively and concentrated. Paying attention to nonverbal signs of the partner, and—when needed—respond to the non-verbal signs.
Shared observation	Shared observation means that the caregiver and health professional jointly observe the infant's motor activities, or that the health professional observes caregiver-infant interactions during daily activities, and that caregiver and health professional share their observations with each other.
Provision of hints and suggestions	Hints and suggestions invite caregivers to implement their own strategies aiming to promote child development or to evolve own ideas during the implementation.
Asking reflective questions	Reflection means scrutinizing and comparative mediation about different aspects of knowledge, skills, desires, aims, actions, or observations. It includes the evaluation of behaviors and/or results of the current intervention. Reflection enables realization, analyses and/or generation of alternative behavior strategies to better reach the own aims. Questions which may inspire reflection, are called reflective feedback.
Provision of feedback	Two different kinds of feedback may be provided, informative feedback and affirmative feedback. Informative feedback means to share information directly related to an action of the caregiver or to an observation. Affirmative feedback means to affirm an action or information of the caregiver.
Illustration with example	The health professional explicitly models an intervention strategy (with the infant or a doll) and the caregiver observes the action of the health professional or acts together with the health professional. The example serves as a hint or suggestion, it does not aim at instruction. Typically, the illustration is accompanied by verbal information and followed by actions of the caregiver with the infant.
Joint planning	Joint planning means that at the end of the session, the parents and the coach together plan which activities the family will try out during daily routine activities during the interval between the sessions.

COPCA coaches focus on the whole family as a unit, implement equal partnership and recommend families to find their own solution. Therefore, COPCA coaches respect families' autonomy and acknowledge families' own criteria for quality of life. The coach has confidence in families' competences and capacities: family members are the key persons in the intervention. The family's values, routines and rituals are respected. COPCA takes place in an enriched real-life environment, during daily care giving activities like playing, dressing, feeding, or bathing. Enriched implies that caregivers receive hints and suggestions how they can use material available in the home environment to play with the child. No expensive material is needed. In COPCA sessions, family members receive coaching on how to promote infant development, for instance how to challenge the infant to self-produced motor behavior. This involves discussions of coach and family members on how to offer the infant opportunities to explore the environment, and how to let the infant experience trial and error. To this end the coaching strategies specified in **Table 1** are used. COPCA coaches appreciate the unique situation of each family, including the family's cultural background. They recognize the families' coping strategies and offer tailored interventions that are adapted to the strengths, resources, decisions, goals and needs of the family members and the child with special needs. The coaching strategies are adapted to the individual needs of the family in a non-directive way. Typically, the COPCA intervention starts with the COPCA coach visiting the family once a week for 45 to 60 min. After a few weeks, the frequency can usually be reduced to every two weeks and later once a month. Since the intervention is adapted to the individual needs of the family, the procedure is flexible.

The effectiveness of COPCA's coaching strategies in infants at high biological risk of neurodevelopmental disorders has been demonstrated in three randomized controlled trials performed in high-income countries (HICs). Coaching of family members was not only associated with improved cognition and better mobility of the infant in daily life, but also with better family empowerment and well-being (11–14, 21). Interestingly, the positive effect of COPCA intervention continued after the end of the intervention (measured about one year after the randomized interventions had stopped), suggesting that families had learned the principles of COPCA on how to stimulate their child's development during daily life activities (11, 14).

Benefit of COPCA'S coaching in LMICs: The example of Brazil

Pediatric health and developmental care in Brazil

Brazil is a country with a large territorial extension and cultural diversity. Although access to remote areas and wealth inequalities challenge universal coverage by public health services, child health and nutrition indicators improved considerably over the last three decades (22). Nonetheless, problems ranging from absence of universal coverage of basic sanitation to limited access to health care services persist. Brazil belongs to the ten countries with the highest rate of preterm births worldwide (11.2% of live births) (23). As in other LMICs, it is common that children pair biological risk with psychosocial risk, resulting in risk accumulation (24). Psychosocial risk factors include food restriction, low parental education, and poor social and environmental stimulation.

Only a few studies addressed the prevalence of developmental delay in Brazil. A population-based study, performed in the Northeast, revealed that 9.2% of children 0–6 years were delayed in at least one developmental domain (25). Another study from the Northeast in infants aged 0–28 months reported that 23% of infants were suspected of a delay in personal-social skills and

20% of a delay in language skills (26). A third study, carried out in Brazil's south, indicated that 32% of infants aged 0–36 months were suspected of developmental delay (27). We also know that the prevalence of disabilities among children aged under 5 years in Latin America and the Caribbean is higher than that in Europe, Central Asia and North America, but not as high as in South Asia and Africa (2). These data—varied as they may be—underline that a considerable proportion of young Brazilian children need early intervention services.

Access to early intervention in Brazil is, however, not easy. It is linked to access to health care in general, which occurs either through the private system—for the minority of the population that can afford it—or through public health care, i.e., through the Unified Health System (Sistema Único de Saúde (SUS)). The SUS, which is used by most people, offers full, universal, and free access to health services. It has front doors at the community level, with basic healthcare units for primary care. Implemented in 2015, the Brazilian National Policy on Integral Attention to the Health of the Child (28) prioritizes health care actions for young children, especially for those in a vulnerable context. The actions start with humanized and qualified care during pregnancy, childbirth, and care for the newborn, including actions directed to preterm and low birth weight newborns offered by SUS. The latter care ranges from kangaroo care and specialized hospital care to shared infant follow-up by hospitals, university centers and primary care teams. Infants, who during follow-up are diagnosed with a delayed or atypical development, are referred for early intervention to rehabilitation or specialized centers, such as the Associação de Pais e Amigos dos Excepcionais (29).

COPCA in Brazil

A recent review on early intervention (30), indicated that in Brazil a rehabilitative model of care is used, i.e., a model applying clinical approaches and child-centered care. This means that early intervention in Brazil in general differs from the internationally recommended good practices (5). Therefore, we recently embarked on the implementation of COPCA in Brazil, as COPCA has several advantages to the typical early intervention approaches in Brazil. First, COPCA fully complies with the international guidelines. Second, COPCA is family centered and home based. This means that family members are coached to find their own ways in rearing the child with special needs in their own home environment. This also implies that families do not need to travel to a center that provides early intervention, and that, ultimately, less health professionals are needed. The latter is an advantage in a country with overall shortage of health professionals (31), including those specialized in early intervention.

A recent Brazilian law, that provides guidelines to formulate and implement public policies for young children (32), and the national healthcare policy (20) recommend early intervention at home. Nonetheless, only a few of such programs are currently available, for example "Primeira Infância Melhor" (22) and "Programa Criança Feliz" (33). These programs are, however, aimed at children at psychosocial risk, not at children at high biological risk. A study from Ghana (Fonzi et al., 2021) indicated that also caregivers of children with cerebral palsy preferred to receive home care, as home care was associated with a reduction of treatment costs, caregiver burden and social stigma (34). Intervention at home with possible cost reduction may be strategic, as some families have difficulties to attend follow-up sessions in clinics due to economic challenges (35).

The use of COPCA's coaching techniques ensures that families are supported in advancing problem-solving strategies to promote development of their child with or at high risk of neurodevelopmental disabilities. COPCA also provides families with opportunities to learn about child development in the context that makes sense to the family. Last, but not least, COPCA's coaching results in family empowerment and may help families to be more assertive throughout a lifetime of care. Current recommendations for improving health and social systems for children in LMICs indeed include redesigning health service delivery models to maximize outcomes, not only to empower families to better care for children, but also to demand better services (36).

We recently reported about our experience with COPCA in Brazil (37) in a case series study with five Brazilian children. Four of the five families had a low income. Three children had been diagnosed with cerebral palsy (Gross Motor Function Classification System levels III, IV and V), one was an infant at high biological risk due to perinatal hypoxia/ischemia, and another child had psychosocial risk due to adverse childhood experiences. The children's families were coached by physical therapy students, who were supervised by a certified COPCA® coach. The families received seven weekly one-hour home visits, with COPCA coaching. After the seven weeks of intervention, the three children with cerebral palsy showed an increase of more than 5% in the target areas of the Gross Motor Function Measure (GMFM-88) (38), a gain that is considered clinically important (**Figure 1**). In addition, the percentile ranking score on the Albert Infant Motor Scale (AIMS) (39) increased in the infant at biological risk. The AIMS percentile scores of the infant at psychosocial risk did not change during the intervention (**Table 2**). The latter may have been due to the student's limited experience to cope with the challenging psychosocial needs of the family. The study also showed that all families were very satisfied with the results obtained during the short intervention—also the family of the infant at psychosocial risk—and their responses indicated that they felt empowered.

In another study, performed during the COVID pandemic, physical therapy students provided intervention supervised by a certified COPCA® coach in seven preterm infants (gestational age at birth 29–36 weeks; correct age at start intervention 5–14 months corrected ag) *via* telemonitoring. This means that we implemented COPCA's coaching *via* the video-call option of WhatsApp. After eight weeks with a weekly tele-COPCA-coaching session all infants had reached the goal that had been determined in partnership with the families at the beginning of the intervention. In addition, all infants showed a substantial

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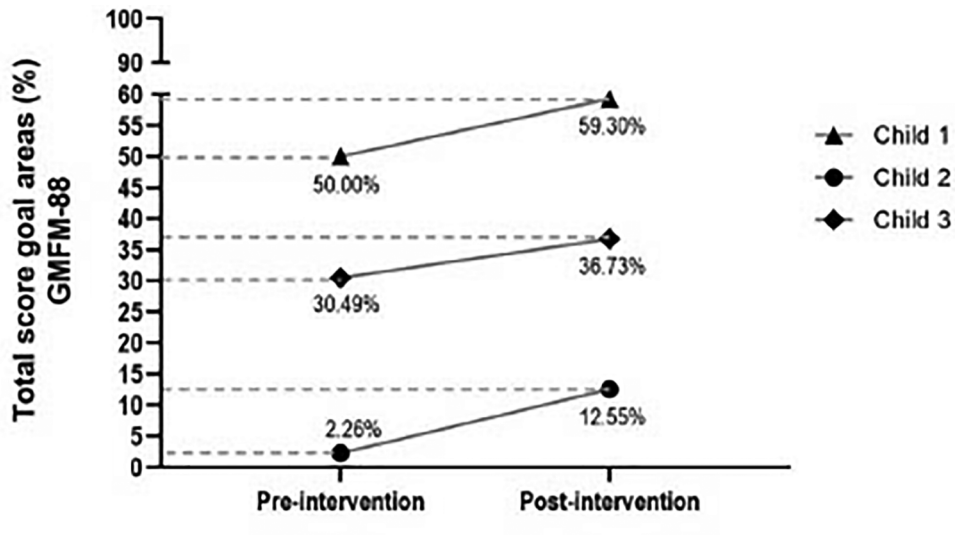

FIGURE 1
Changes in GMFM-88 percentage scores in the goal areas of the three children with CP of the Brazilian case series study during the 7 weeks of Family-Centered Care intervention (led by a COPCA coach). Figure reprinted from reference 38 with permission of Revista Fisioterapia em Movimento (Curibita, PR, Brazil: DOAJ).

increase in the AIMS percentile scores (**Figure 2**). Moreover, the caregivers were satisfied with the results and felt supported and empowered by the approach.

The second case series showed that it is feasible to implement COPCA by means of tele-intervention. Tele-coaching of COPCA may be an attractive early intervention strategy for Brazil, as it eliminates the family's burden to travel to a clinic. The travel burden is a well-known factor reducing adherence to early intervention (40, 41). Tele-coaching of COPCA also enables a virtual visit to the infant's home. It therewith allows for the visualization of the natural home environment, and it facilitates exchange of information and discussion of activities that fit within the infant´s reality. In addition, as tele-guidance makes it impossible for the health professional to touch and handle the child, the transition to a really family-centered approach is more easily achieved. In other words, tele-guidance facilitates the implementation of COPCA's coaching strategies. Tele-guidance can also be used in combination with face-to-face care; for instance, in families who live in distant communities or in rural areas, which is very common in Brazil.

Conceivably, one barrier to the implementation of COPCA, not investigated in these two case studies, maybe parental objection to the novel approach of coaching, as it so different from the most used approaches to developmental physical therapy in Brazil. These interventions consist of hands-on approaches such as neurodevelopmental treatment and suit therapy (42). In these traditional approaches, the therapist acts as an expert, who handles the child and instructs parents what to do. COPCA's coaching implies a different role of the family members, which might meet resistance. However, it should be realized that in the countries in which COPCA was first applied, i.e., in the Netherlands and Switzerland, similar primary worries on COPCA's implementation existed. Nonetheless, COPCA's implementation in daily practice revealed that the families gladly accepted the new approach, after having received information of the approach's background. In the Brazilian case studies the parents in the study seemed to appreciate COPCA, but it must be noted that parents might have been motivated to try COPCA as some children had not been making gains with the traditional approaches, and the other children were either on a waiting list

TABLE 2 Changes in the Alberta infant motor scales (AIMS) scores in the two children without CP of the Brazilian case-series study.

	Child 4		Child 5	
	Pre-intervention	Post-intervention	Pre-intervention	Post-intervention
Percentile	**<25**	**50**	**10**	**10**
Total score	**15**	**29**	**49**	**52**
Prone	4	8	21	21
Supine	6	8	9	9
Sitting	3	10	12	12
Standing	2	3	7	10

Percentile scores based on Piper & Darrah, 1994 (39). Table adapted from Cunha et al. (37). Child 4 was at high biological risk due to perinatal hypoxia/ischemia, child 5 was at psychosocial risk due to adverse childhood experiences.

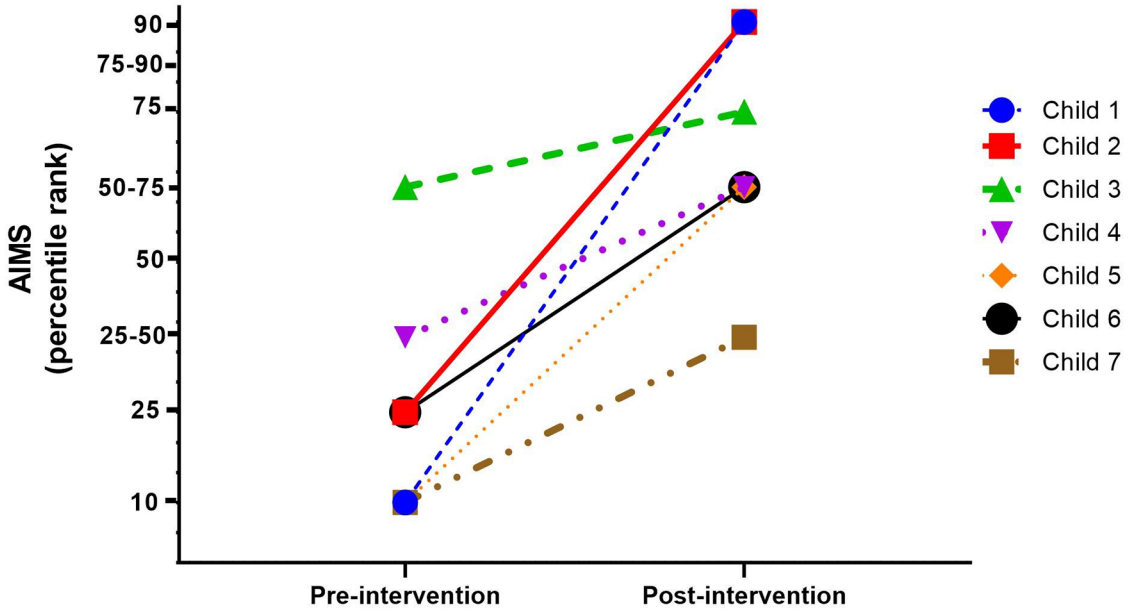

FIGURE 2
Developmental trajectories in percentile rank scores of the AIMS of the seven preterm infants before and after 8 weeks of COPCA intervention (results of the second Brazilian pilot study).

or did not have access to other forms of intervention due to the COVID-19 pandemic. This means that future studies need to address in the LMICs the perception of family members of COPCA, including its advantages and disadvantages compared to traditional approaches.

Based on the theoretical reflections, the overall child health care situation in Brazil, and the promising results of the pilot studies, we believe that COPCA is an early intervention program that may contribute to overcoming the challenges encountered in Brazil's early intervention services for infants at high biological risk. Even though more data are needed to support this assertion, there were no barriers to the application of COPCA coaching with low-income families. COPCA fits to Brazil's primary care because its coaching strategy works with the resources that are available in the home environment, and the family does not need to go to a rehabilitation center or pay for expensive equipment. COPCA's coaching may be delivered face-to-face or *via* tele-guidance or by a combination of both approaches.

Discussion and conclusion

Between 1990 and 2020 mortality in children aged under 5 years decreased by 60% due to the impact of the United Nations' Millennium Development Goals (3). Fortunate as this may be, this gave—in combination with the rapid population growth in LMICs—also rise to an increase of infants at high biological risk of neurodevelopmental disorders. As a result, more than 90% of children with disabilities live in LMICs (3, 43). This implies that the need of adequate early intervention in LMICs is high.

Most early intervention programs in LMICs focus on families in challenging social conditions, for instance families dealing with poverty (44). Early intervention in these situations is most effective when it consists of parenting interventions, i.e., of intervention that aim to improve caregivers' knowledge, attitudes, practices, and skills, including responsive caregiving. Such interventions allow caregivers to promote in their own situation optimal early child development (18, 45).

Little is known on early intervention in the infants with highest needs, i.e., the infants at high biological risk of neurodisability in LMICs (46, 47). It may be assumed that they will benefit from the same intervention strategies that are profitable for infants at high biological risk in HICs. But knowing which interventional elements are effective in promoting developmental outcome is one thing, implementing early intervention in challenging social situations, as frequently met in LMICs, is quite another thing. The intervention needs to reach the families (48). The latter implies that the intervention has to take into account the families' culture, perceptions, finances and levels of stress (48).

Our preliminary data with COPCA in Brazil suggest that COPCA is an early intervention program that may serve early intervention in infants at high biological risk in LMICs. COPCA's coaching strategies are tailored to the needs of individual families, as family autonomy is a crucial element in COPCA. Family members learn through the empowering dialogue with the COPCA coach in which way they can promote their child's development, in their own situation according to their own cultural norms. In addition, COPCA's coaching may be delivered by tele-coaching therewith eliminating the family's burden for travelling to distant early intervention clinics. Larger scale studies are needed to support COPCÁs effectiveness for

early intervention in LMICs as well as to identify possible barriers to its implementation and how to overcome them. Intervention by health professionals is associated with substantial costs, which may hamper the implementation of the intervention. We also recommend studies that evaluate which part of the COPCA intervention may be delivered by lay or paraprofessional community health workers and which part needs to be taken care of by fully educated COPCA coaches.

In conclusion, LMICs face the challenge of implementation of effective early intervention services for a high number of infants at high biological risk of neurodisability. Increasing evidence in HICs indicates that interventions in which families are empowered to find their own solutions, on how they can promote their child's development during daily care giving activities, are associated with better child development and favorable family outcome. A major strategy to reach these goals is coaching, which is COPCA's fundamental intervention strategy. Two pilot studies in Brazil indicated that COPCA's coaching technique, including its tele-coaching option, turns COPCA in a promising early intervention for infants at high biological risk in LMICs. COPCA's positive effect on family empowerment and child development suggest that COPCA may be associated also with improved school readiness.

Data availability statement

Publicly available datasets were analyzed in this study. This data can be found here: Cunha RFM da, Costa KB, Morais RL de S. Family-centered care on a physiotherapy course: case reports. Fisioter mov (2022) 35: doi: 10.1590/fm.2022.35301.

Author contributions

SAZ and MHA conceptualized and designed the study. All authors drafted the initial manuscript and reviewed and revised the manuscript before the submission. SAZ wrote particularly the first two sections, RLSM and LM wrote the third section in the first instance and MHA wrote primarily the discussion and conclusion section. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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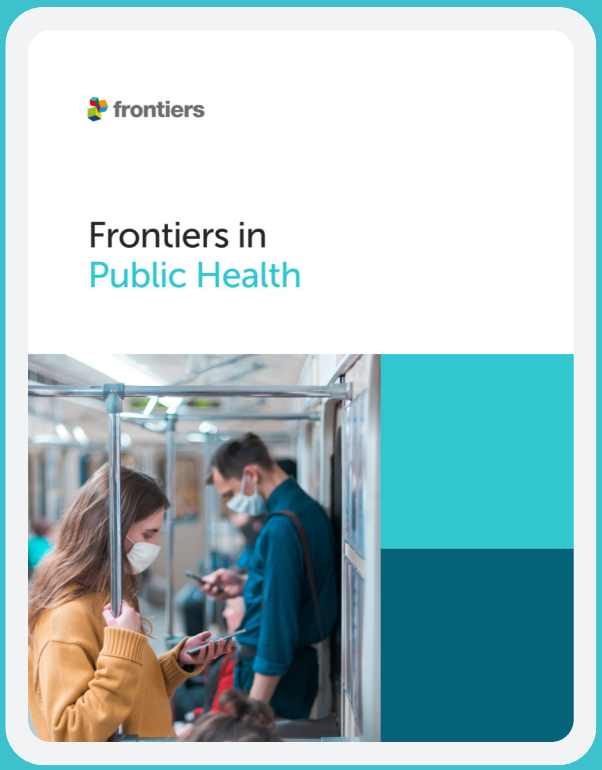

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