



Impact of Research About the Early Development of Children With Intellectual Disability: A Science Mapping Analysis

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The “Warnock Report” (Department for Education and Science, 1978) underlined the importance of early intervention for children with a range of special educational needs and the importance of partnership with families. This paper focuses on young children with intellectual disability to describe the longitudinal research on early development that has emerged since the report, and to describe the scholarly literature that has been impacted by this longitudinal work. First, we conducted a systematic literature search for primary reports of longitudinal studies on the early development of children with intellectual disability. Included studies were those that measured dependent (i.e., developmental outcomes) and independent variables (i.e., risk and resilience factors) on at least two measurement occasions (i.e., truly longitudinal), starting before the end of the 7th year of life, with samples including children with intellectual disability (or related terms). The topics of these studies, and of the publications that have cited these longitudinal studies, were extracted from titles and abstracts using machine reading and subjected to multidimensional clustering (VOSviewer; Van Eck and Waltman, 2016). The resulting body of 101 research studies (about 2.5 studies per year) covered a scattering of topics without a dominant focus. The literature that was impacted by these longitudinal studies consisted of 3,491 scientific publications. Three clusters of topics emerged from mapping the terms used in these publications, which were dominated by (1) syndrome and disorder related terms; (2) autism-related terms; and (3) disability and parent related terms. Topics related to autism and, to a lesser extent, parents showed the strongest increase over time. Topics related to intervention and programmes were mostly linked to the topics disability and parents. Taking into account the science mapping as well as features of the context in which research on intellectual disability takes place, we suggest a collaborative research agenda that systematically links topics relevant for intervention with longitudinal research, in co-creation with families.

Keywords: intellectual disability, longitudinal research, early development, early intervention, systematic review, science mapping

INTRODUCTION

Intellectual Disability is described in ICD-11 as a Disorder of Intellectual Development (Salvador-Carulla et al., 2011). Intellectual disability emerges during the “developmental period” [usually taken to mean before age 18 years; (American Association on Intellectual and Developmental Disabilities (AAIDD), 2010)], and is characterized by low cognitive ability (IQ < 70) and low levels of adaptive functioning (such as communication, and social and independence skills, assessed using standardized tools). Prevalence studies internationally suggest that ~1.5–2.2% of children and adolescents have an intellectual disability (Maulik et al., 2011). Children with an intellectual disability have historically been given a variety of labels including “mental retardation,” “mental handicap,” and “subnormal.” While internationally the term intellectual disability is favored, terminology in the UK education system was heavily influenced by the report of the committee led by baroness Warnock on special educational needs (Department for Education and Science, 1978), which proposed to use “learning difficulties” as the generic term (with some distinction of severity in mild, moderate, or severe). Publications like the Warnock report have not only shifted the social construction of having an intellectual disability but also enhanced awareness of critical needs in a significant group in our society. Charting those needs is one of the tasks that researchers in the field have undertaken.

Current social constructions of intellectual disability emphasize low levels of general intellectual ability and associated low adaptive functioning—relative to levels expected from individuals of the same age. Profiles of abilities and associated needs look very different from child to child, but usually the problems in the domain of mental functioning broadly affect activities and participation. In addition, the putative causes of this cluster of needs are many and varied—ranging from specific genetic conditions (e.g., Down syndrome), to socio-economic circumstances (e.g., extreme poverty or neglect), and environmental toxins (e.g., lead poisoning), with etiology known in a minority of cases (Kaufman et al., 2010). Despite this heterogeneity, the field still categorizes children with intellectual disability, so that needs of this subpopulation of children can be defined on a group basis. Many countries organize specialized educational services with this category of children in mind. Our focus in the current paper, using science mapping methods, is to examine research on the early development of children with intellectual disability, describing themes based on the primary sources on this research as well as the themes within secondary sources that base themselves on the longitudinal research in early development. The focus will be, in terms of the International Classification of Functioning, Disability, and Health (World Health Organization, 2001), on the development of mental functioning, activities, and participation, as these domains are universally affected in people with intellectual disability (in contrast to other body functions and structures). Furthermore, the focus will be on early childhood development, which may be defined as the emergence within the period from conception to age 8 of

sensory-motor, cognitive, communication, and social-emotional skills (World Health Organization (WHO), 2012).

There is broad consensus that intervening early is a good thing in support of any disadvantaged child (UNICEF, 2017). The UK Warnock Report (Department for Education and Science, 1978) devoted a whole chapter to children under five that started with a recognition that the early years are a time of rapid development, and that this time of development is as crucial for children with special educational needs as for all children. However, early intervention and support may be even more important for children with intellectual disability than for all other children. By definition, children with intellectual disability have core developmental delays reflected in their performance on IQ and adaptive behavior assessments. As development in general is rapid in the early years, cognitive and adaptive functioning of children with intellectual disability soon lag behind that of their peers even if their rate of development is only slightly slower than average. To help children with intellectual disability to catch-up developmentally, intervention and support needs to start early in life to shift their trajectory of development and avoid further falling behind.

Beyond dimensions that are a part of the definition of intellectual disability, this group of children face multiple other educational, social, and health inequalities. For example, children with intellectual disability are 4–5 times more likely to have mental health problems compared to other children (Emerson and Hatton, 2007). Families of children with intellectual disability are at increased risk for multiple social/economic risks including poverty and exposure to negative life events (Emerson and Hatton, 2007). In the physical health domain, children with intellectual disability are up to 70% more likely to be obese (Emerson et al., 2016), which in turn increases the long-term risks of obesity-related health problems. These inequalities are apparent early in the lives of children with intellectual disability; by age 5 years at the latest (Totsika et al., 2011; Emerson et al., 2016) and likely even earlier. The early emergence of these inequalities in children’s lives has given rise to entertaining the possibility that early intervention may have large and long-term impact.

In considering any specific intervention, including early interventions, it is important to be able to draw upon evidence that is relevant and that has sufficient coverage of the multiple, complex facets of intellectual disability. Frameworks for complex interventions suggest that the evidence base should start with theory and modeling research (or understanding the “problem”) (e.g., Craig et al., 2008; Thornicroft et al., 2011). This research evidence is then used to design specific interventions that can be tested using robust experimental designs, incorporating mixed methods evaluation, and eventually the testing of the wider scale roll-out of interventions in typical practice. Thus, evidence-based early intervention and early support for children with intellectual disabilities and their families (recognizing that families are the primary context for early development; Department for Education and Science, 1978) would benefit from research on the early development of children with intellectual disability.

From a developmental perspective on intellectual disability (e.g., Hodapp et al., 1990), developmental pathways for children

generally apply to children with intellectual disability. The main difference is that the pace with which children with intellectual disability develop along these pathways may differ (especially, may be slower). However, this perspective needs to be tested explicitly in studies on the early development of children with intellectual disability. In addition, the developmental perspective on intellectual disability recognizes that there may also be some divergence from typical developmental sequences most notably in the context of specific genetic syndromes. For example, the genetic disorder Rett syndrome is typically associated with severe to profound intellectual disability and early development in this condition is typified by an early period of developmental regression (Cianfaglione et al., 2018).

Very little longitudinal research had been published into the development of children with intellectual disability before the Warnock committee, and no such research was referenced in their report (Department for Education and Science, 1978). The recommendations from the report regarded attending to the origin and course of the special educational needs of children including those with intellectual disability.

The aim of the current paper was to describe the areas that have received most attention in research on early development in the four decades since the Warnock report. To that end, we adopted a bibliometric approach and first asked what peer-reviewed longitudinal research evidence is available on early developmental pathways of children with intellectual disability, what topics of these studies were, and how the topics of this literature are related. Second, we asked what the impact of this longitudinal work has been by mapping the topics of peer-reviewed publications that have cited the longitudinal work, again by describing and depicting the topics and their interrelationships and by examining time trends.

METHODS

The review questions were addressed by performing literature retrieval (Liberati et al., 2009) in **two** steps.

Retrieval of Longitudinal Studies of Early Development Eligibility Criteria

Publications were selected if these: (1) used as inclusion criterion intellectual disability or a clinical condition with intellectual disability as a part of the phenotype (as focal sample, not as comparison sample; per the goals of this review), (2) reported studies where the aim was to quantify non-experimental change in dependent variables or associations between independent and dependent variables on at least two time points (i.e., truly longitudinal), (3) conducted the first measurement wave before end of the 7th year of life for all children (to be flexible enough to incorporate most international perspectives on the focus period for early intervention), (4) appeared as indexed peer reviewed journal articles or chapters (to focus on peer reviewed primary sources of empirical studies), (5) appeared within the domains of psychology and social sciences or in a journal in the field of intellectual disability (to focus on fields that broadly attend to

mental functioning, activities, participation, personal factors, and environmental factors in relation to health conditions and bodily functions; World Health Organization, 2001), and (6) were published before 2018 (to be able to retrieve citing studies in the next step of the research). Publications were considered ineligible if no abstract was available and the full text version could not be retrieved (to enable the investigators to assess eligibility).

Retrieval and Eligibility Assessment

Query strings (**Appendix A**) were entered in the bibliographic databases of Scopus and Web of Science, which provide ongoing coverage of the large majority of international peer reviewed journals in the field of special education and rehabilitation. Records retrieved (Web of Science: number of studies $k = 920$; Scopus: $k = 1,016$) were entered in Endnote to remove duplicates, after which $k = 1,593$ records remained. The authors independently coded the titles and abstracts on eligibility criteria 1–3, turning to the full manuscript if information was missing or unclear. This resulted in 120 candidate publications. Reliability of eligibility assessment varied between kappa = 0.79 to 1.00 ($k = 50$ were double coded). After first screening, candidate publications were fully read to double check compliance on eligibility criteria 1–3, after which 108 publications remained. The final set of $k = 101$ eligible studies was obtained by excluding 7 studies with dependent variables that fell outside the domains of mental functioning, activities, participation, personal factors, and environmental factors in relation to health conditions and bodily functions (eligibility criterion 5). **Figure 1** provides the PRISMA flow diagram for the study selection and results.

Retrieval of Studies Citing Studies of Early Development Eligibility Criteria

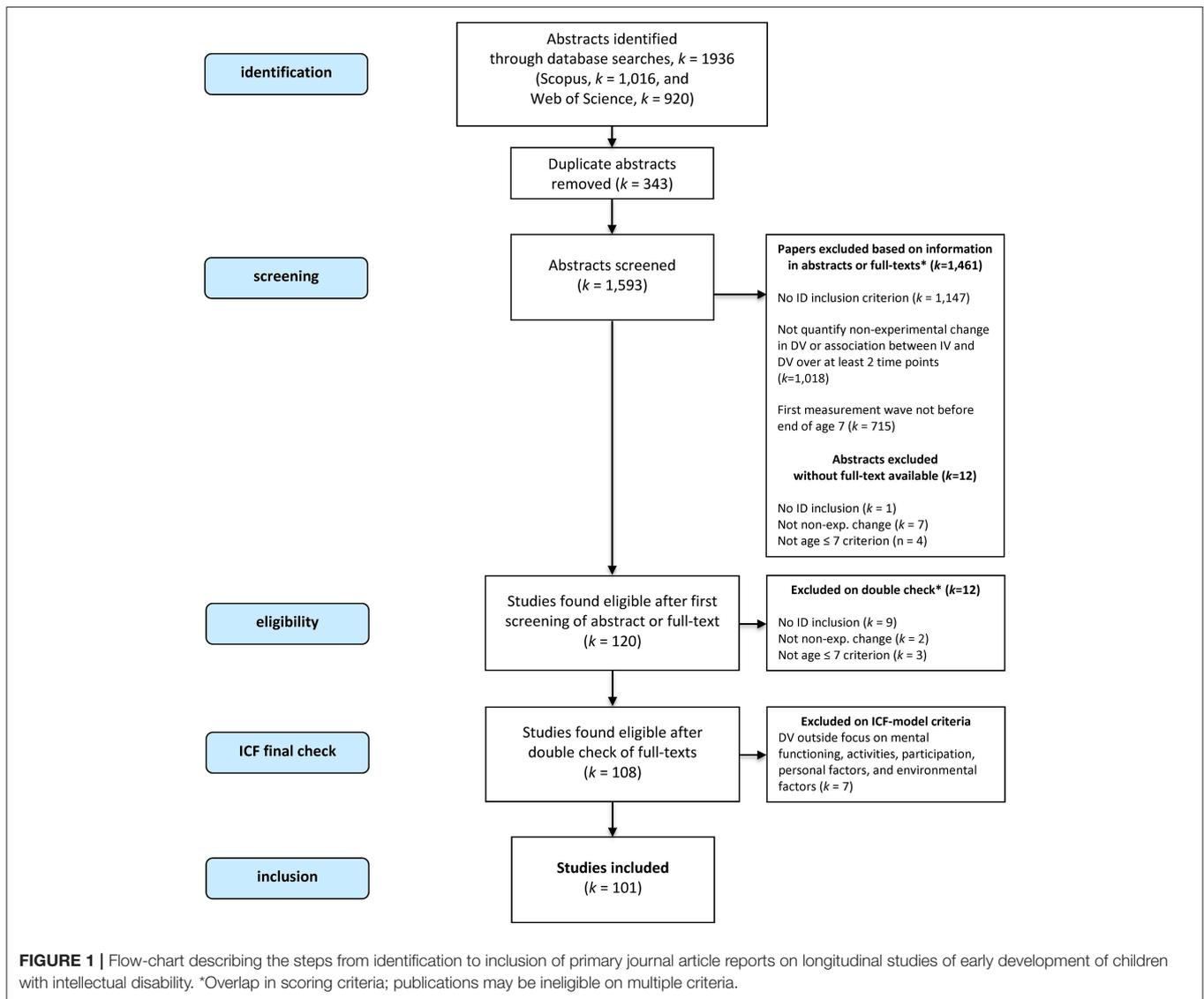
Publications were selected if these: (1) cited one or more of the longitudinal studies identified in step 1 (as per the goal of the study), (2) had full bibliographic records electronically available with title, author list, publication year, abstract, keywords, and reference list (to provide the data necessary for science mapping analysis), which limited the search to journal articles.

Retrieval

The longitudinal studies ($k = 101$) retrieved in step 1 were searched in Scopus and Web of Science to identify citing references. Records retrieved (Web of Science: $k = 2,494$; Scopus: $k = 3,448$) were exported to a publication database. Duplicate removal led to $k = 3,491$ unique publications in step 2.

Science Mapping

The citing records retrieved in step 2 were read into the software program VOSviewer 1.6.10 (Van Eck and Waltman, 2016) for the construction and visualization of bibliographic networks. This software projects “nodes,” such as publications, authors, or terms, in a two-dimensional space based on a normalized index for bibliographic similarity (i.e., link strength), such as the number of co-citations of two publications by third publications or the number of times two terms occur together in the same publication (Van Eck and Waltman, 2014). In addition, the



program performs a weighted and parameterized variant of modularity-based clustering on the link strengths to reveal additional distinctions beyond those that can be derived from the two-dimensional scaling (Waltman et al., 2010). To map the topics and themes in the longitudinal studies on early development, a network was created of co-occurrence of terms extracted by natural language processing of titles and abstracts for nouns and adjective-noun combinations. Only terms that occurred 5 times or more were included. The algorithm ranks the terms found based on the extent to which co-occurrence appears systematic or random, keeping only the 60% most relevant terms. Terms were excluded if these referred to longitudinal research, young children, or intellectual disability (because publications were already selected on that basis), if these described study methods (given the interest in substantive focus), or if the terms appeared trivial (such as type of publication, statistical terms, or country of study).

To map the topics and themes in the literature citing longitudinal work on early development, natural language processing was conducted similarly as for the longitudinal studies of titles and abstracts, now limited to terms that occurred at least 50 times. Terms were excluded if these described study methods (given the interest in substantive focus), or if the terms appeared trivial (such as type of publication, statistical terms, or country of study). The full list of deselected terms can be found in **Appendix B**.

RESULTS

Longitudinal Studies of Early Development

References to the longitudinal studies of early development identified in step 1 of the study can be found in **Appendix C**. **Figure 2** maps the machine extracted terms describing the longitudinal studies, indicating the weight of each term (by its

size), and mean publication year indicated by its color (with redness indicating relatively recent use of these terms and blueness indicating relatively early use of these terms). The links connecting terms represent their rate of co-occurrence. **Figure 2** shows that studies referring to “parenting” are of a relatively recent appearance in the literature, this term being present in titles and/or abstracts 14 times since 1999 (9 of which after 2012). “Context” appeared in this literature since 2001 ($k = 10$), while other relatively new topics occurred less frequently. “Syndrome” ($k = 52$), often in combination with “Down” ($k = 41$), continued to be used throughout the period covered by the longitudinal studies. Of the domains of early development (World Health Organization (WHO), 2012), cognitive, communication, and social-emotional skills were represented, with communication (including language) receiving most attention. The sensory-motor domain was not represented. The considerable scatter in the map, with little evidence of dominant topics among this modest set of longitudinal studies, precludes a coherent overall summary of dominant focus and trends.

Bibliographic Impact of Longitudinal Studies

Figure 3 shows the increase in publications citing the longitudinal studies of early development up until 2017 (the last full year with complete bibliographic data). **Figure 4** maps the machine extracted terms describing the publications that cited longitudinal studies, indicating the weight of each term (by its size), and membership (by its color) of one of the three clusters that were extracted from the weights of the links between the terms. The links connecting terms represent their rate of co-occurrence. Only links with a minimum weight of 50 are displayed.

The green cluster consisted of 39 terms, and was dominated by the term “syndrome,” and also, albeit to a lesser extent, by “disorder,” “ability,” and “individual,” reflecting a focus on the diagnosis of individual children. Early development was represented in this cluster with the cognition (“cognitive,” “memory”) and sensory-motor (“motor”) domains. When searching for terms referring to intervention and support, only “patient,” “identification,” and “treatment” were found. Terms referring to education did not occur within this cluster.

The blue cluster consisted of 32 terms, and was dominated by the terms “autism,” “autism spectrum disorder,” “ASD,” and “skill,” reflecting a focus on people with autism spectrum disorders and their skills. Early development was represented in this cluster with the domains of cognition (“joint attention,” “cognitive development,” “learning”), communication (“language,” “language development,” “speech,” “gesture,” “vocabulary,” “word,” “expressive language,” “communication”), and social-emotional skills (“social interaction”). No terms belonged to this cluster that referred to intervention or support, nor to education.

The red cluster consisted of 44 terms, and was dominated by the terms “disability,” “family,” “parent,” and “mother,” reflecting a family focus. In this cluster, early development was represented with the social-emotional skills only (“friendship,” “peer,”

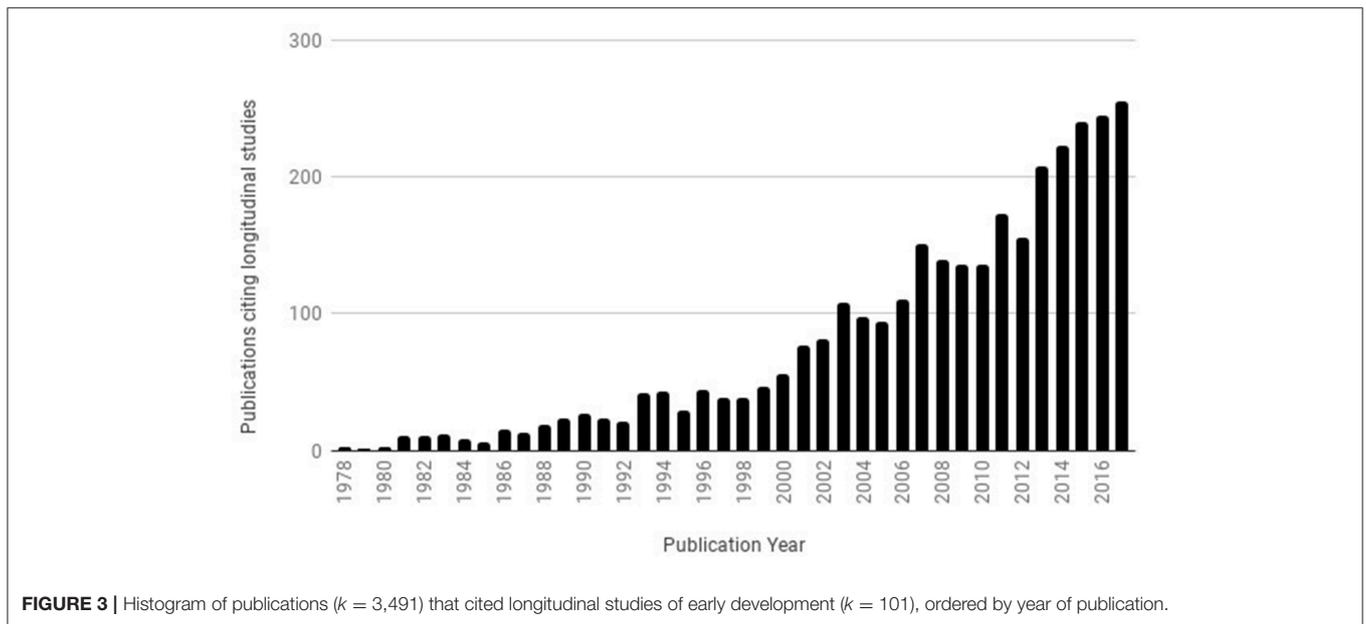
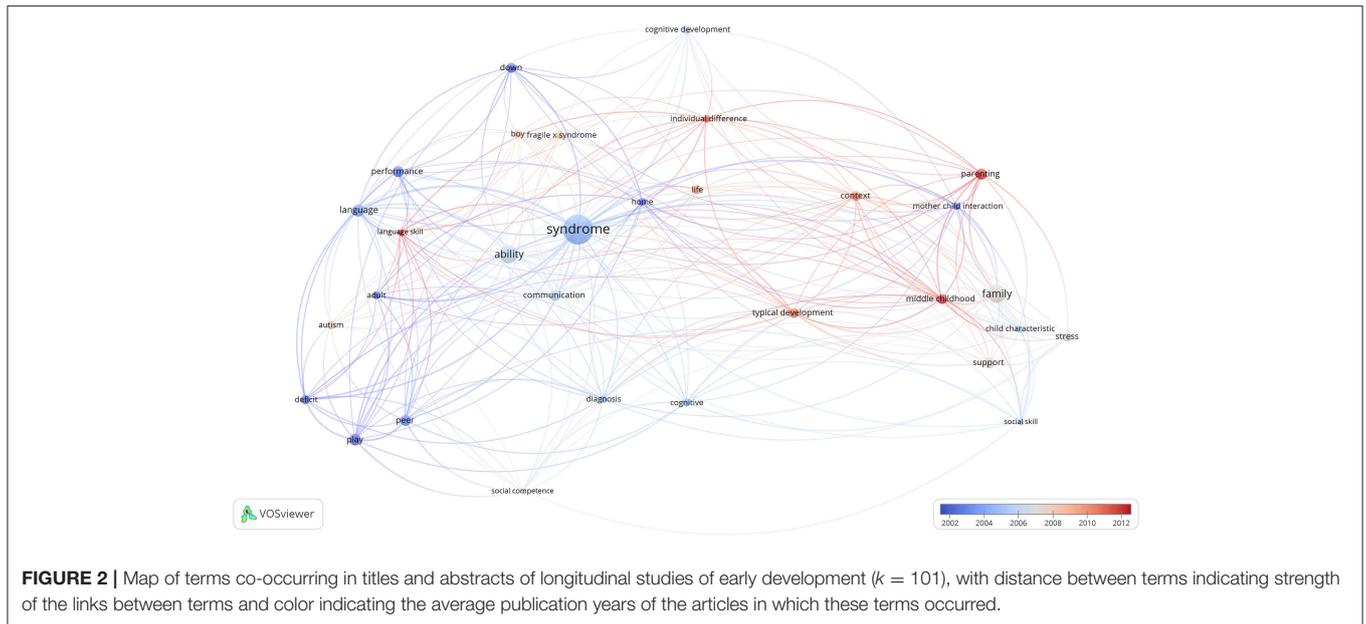
“social competence,” “social skill”). When searching for terms referring to intervention and support, these were also included in this cluster, such as “effectiveness,” “efficacy,” “practice,” “professional,” “program(me),” “service,” and “support.” Terms referring to education were also found in this cluster, such as “education,” “school,” “student,” “special need,” and “teacher.”

To discern the most recent research on which longitudinal studies have had impact, **Figure 5** overlays the clusters from **Figure 4** with mean publication year. There is a clear trend of an increasing number of studies on autism spectrum disorder that cites longitudinal research on young children with intellectual disability. Of the terms referring to intervention and support, “efficacy” ($k = 93$), “effectiveness” ($k = 97$), “practice” ($k = 311$), and “support” ($k = 705$) stand out as being used in relatively more recent literature that cited longitudinal research. Of the terms referring to education, only “school” ($k = 609$) and “student” ($k = 142$) appear in more recent literature citing longitudinal research.

DISCUSSION

Applied scientific research findings may improve understanding of a phenomenon or the processes associated with a problem’s emergence or maintenance. However, the delay in time for such scientific findings feeding into interventions and practice can be substantial—perhaps as much as 20 years (Contopoulos-Ioannidis et al., 2008). Evidence-based interventions are best informed by scientific findings and theory (Craig et al., 2008; Thornicroft et al., 2011), and there is critical need for evidence based early intervention practices for children with intellectual disability (as inequalities affecting them emerge very early in development). Therefore, the research identified in the current study on the early development of children with intellectual disability is of substantial importance both scientifically and in relation to policy and practice.

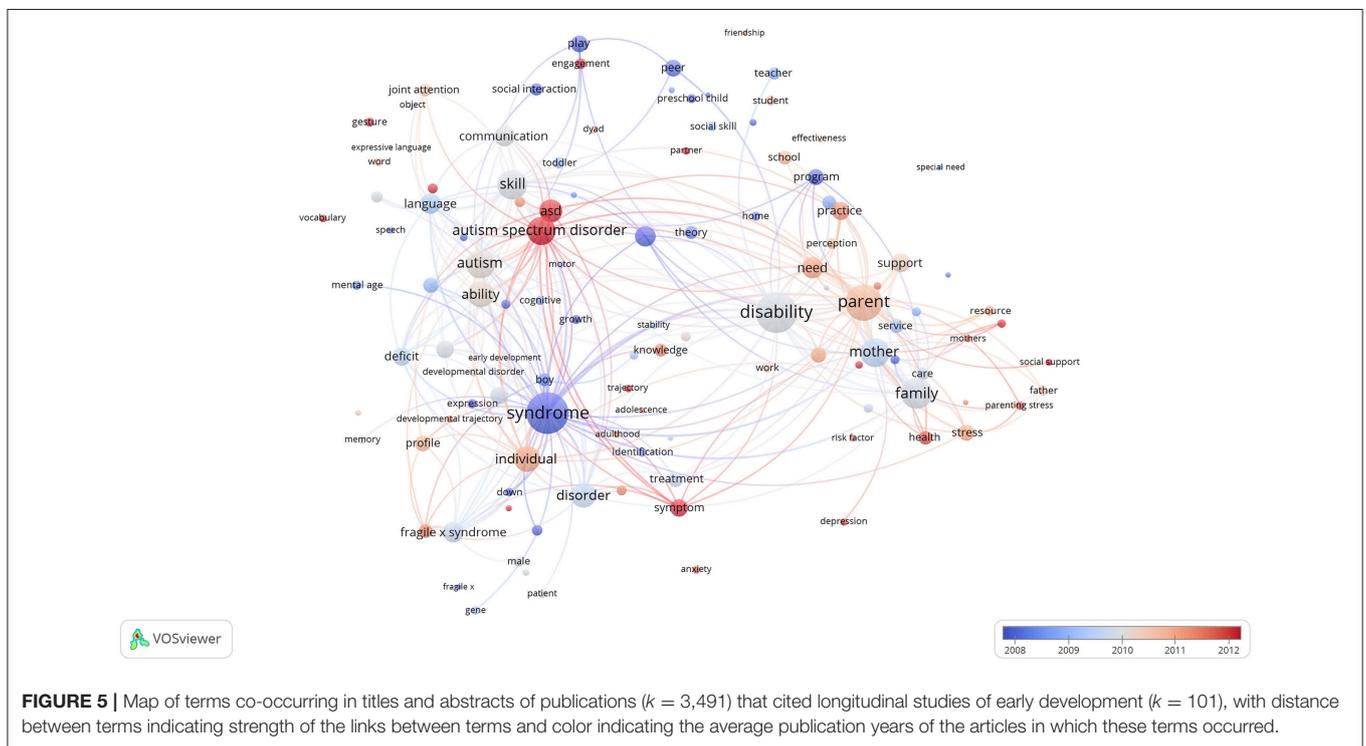
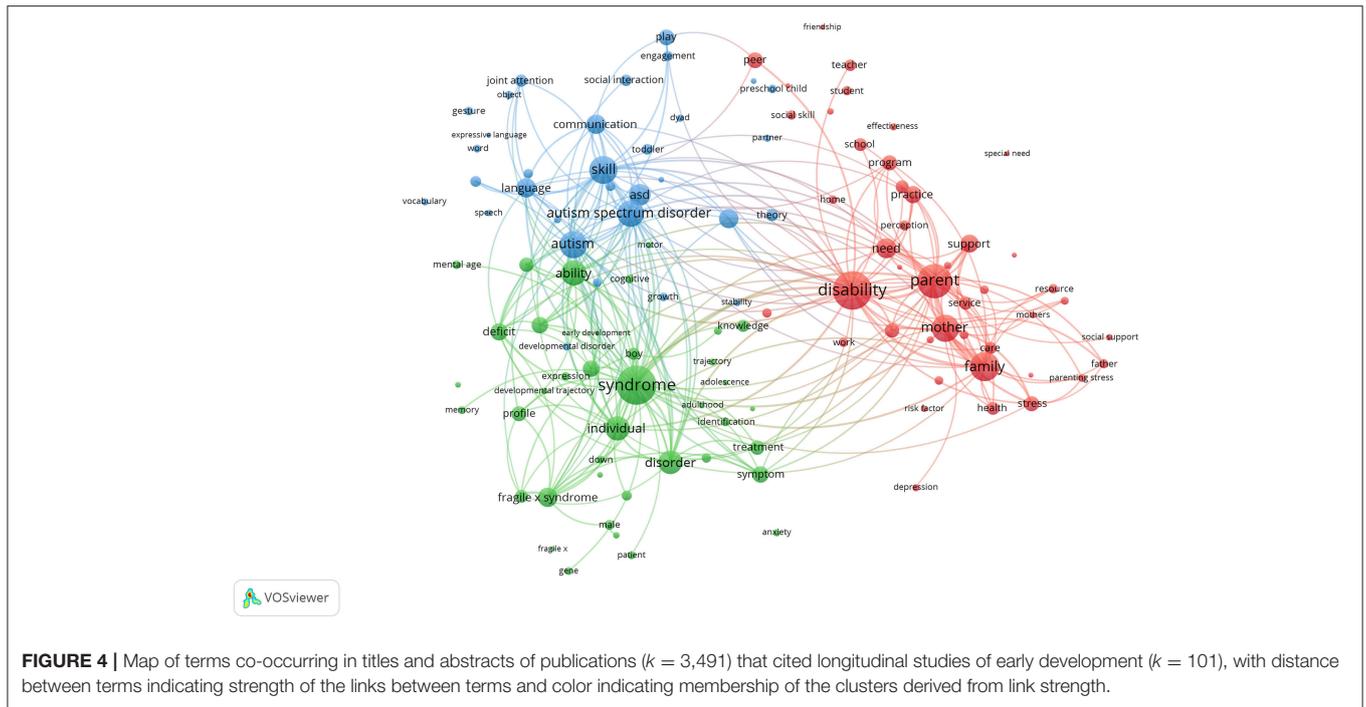
In the systematic review stage of the current study, we identified 101 longitudinal (at least two time points, first data point before children turned 7 years old) research studies addressing educational, psychological and related development of children with intellectual disability. This body of work has been published over more than four decades. Thus, although the total body of relevant work appears at first to be significant, the number of studies published on average per year is a modest 2.5. Running our literature searches without restricting the outputs to studies including terms relating to “intellectual disability” (as a quick search to provide context for what we have found) led to an almost 20-fold larger corpus of potentially relevant papers on early development. This all suggests that the early development of children with intellectual disability has been relatively neglected internationally. Using science mapping approaches to examine the focus of the 101 studies also suggests a lack of coherence or strategic direction for the field of early development in children with intellectual disability. Terms in the records of included studies (**Figure 2**) were varied and showed few trends over time. Although still weak trends, there was some indication of reference to parenting (and to a certain extent to family) in the



more recent research literature. This may signal an increasing focus or recognition of the role of parents and the family in early development of children with intellectual disability, reflecting programmatic and collaborative efforts by people in the field (e.g., Blacher; Baker; Hauser-Cramm). However, given the relatively small number of total studies, this weak trend may have been driven purely by a small number of research groups publishing in the field. Also of note is that 41 of the 101 studies referred to Down syndrome. Although an important sub-group in the population of children with intellectual disability, this relatively large amount of studies referring to the group with Down syndrome seems to represent a relative neglect of other

sub-populations and potential ascertainment bias in the current evidence base.

Publications citing the 101 core studies have been increasing over time, especially over the most recent 5 years to 2018 (Figure 3). These data may indicate a growing and recent attention to the early development of children with intellectual disability, or at least in research addressing this topic. The related aspect of the science mapping (focus and trends of the 3,491 studies citing the original 101 studies) showed three clusters of research that have been citing studies about early development of children with intellectual disability (Figures 4, 5). The first cluster shows that research referring to autism spectrum disorder



has been citing studies of the early development of children with intellectual disability, and that this is a recent trend in particular. It is important to note that we did not search for research studies focused on autism (in the absence of intellectual disability) as our interest was in research on the early development of children

with intellectual disability (also including those who may have an additional diagnosis or label of autism). However, drawing on research on the early development of children with intellectual disability in the autism literature may reflect a number of scientific trends. For example, this may reflect a simple increase

in the volume of autism research as it has become a funding priority internationally. Furthermore, children with intellectual disability and Down Syndrome in particular are often used as a comparison group in studies focusing on children with autism (e.g., Baranek et al., 2019). The autism research citing studies of early development of children with intellectual disability was not associated with frequent use of intervention-related terms. Clarifying linkages and trends regarding autism and intervention will require more comprehensive mapping of the research on early development in children with autism.

A second cluster of terms from citing studies in **Figure 4** derive from medical terminology including “syndrome,” “disorder,” and “ability.” Intervention-related terms did also occur in this cluster but again they can also be seen to have a more medical flavor (patient, identification, treatment). There was also a clear time trend for this cluster of more medical terms from citing studies (**Figure 5**) appearing in older literature. Our searches sought out research of a primarily psychological, social or educational nature on the development of children with intellectual disability. Therefore, it is of interest that a more medically-focused cluster of citing studies was found. However, the fact that this cluster of terms was found in older citing research may reflect a general move away from medical models of disorder to an increasing functional, activity, and participation focused understanding of disability (World Health Organization, 2001; Bertelli et al., 2016).

The third cluster of terms from citing studies represents studies focusing on environmental factors. This third cluster was dominated by terms relating to family [including parent(s)], but also included multiple terms relating to social dimensions of intervention and to education. Thus, there is a body of research citing studies of the early development of children with intellectual disability that has a dual focus on families, and on intervention and supports. This body of work has been present in the field for some time, but does not have such a strong recent trend (in the same way that autism is showing) (**Figure 5**). Given the key role of the family in early child development, one might have expected increasing interest in the developmental environment, as well as in the implications for family life. However, the science mapping of citing studies did show a clear time trend for increasing occurrence of terms in citing studies that referred to intervention and support (efficacy, effectiveness, practice, and support) (**Figure 5**), showing that intervention as a component of the environment appears to integrate insights on development and considerations of the family context.

In summary, we found a disappointingly small body of international research literature on the early development of children with intellectual disability but on a broad range of subjects (suggesting lack of focus). Science mapping analysis revealed some encouraging trends in the use of research on early development of children with intellectual disability. Most significantly, more recent research citing studies of early development in intellectual disability were more likely to also make reference to intervention and support. Perhaps of some concern was that, despite dominant terms relating to the family and child within the family in the 101 early development studies themselves, recent citing studies were more likely to be referring

to terms relating to autism than to the family. Our analysis suggests a priority for more research on the early development of children with intellectual disability.

Toward a Road-Map for Early Developmental Research in Intellectual Disability

Funding, organizing, and maintaining longitudinal studies in all fields is a considerable challenge. In intellectual disability, there are at least three additional challenges. First, the prevalence rate of intellectual disability even in childhood is low overall (Maulik et al., 2011), and these numbers drop dramatically when breaking down this population in subpopulations with distinct known etiologies (Kaufman et al., 2010) let alone phenotypes. Finding sufficient numbers of young children and their families to achieve reliable estimates of developmental pathways and test hypotheses about developmental mechanisms with sufficient statistical power will, therefore, require additional resources and/or collaboration across research groups and countries. Second, ascertainment of intellectual disability when children are very young is fraught with problems. Intellectual and adaptive functioning of young children show variation across individuals and time, and time is needed to be able to conclude that functioning remains in the range for intellectual disability. Existing studies have typically focused on populations (and “diagnoses”) that may be more typically applied to young children and are likely strongly associated with identified intellectual disability as a child ages. In particular, the constructs of Global Developmental Delay or developmental delays in key domains (e.g., language and/or social behavior) may be easier to identify in young children. Some etiologies (e.g., Down syndrome) are also easily identified without even the need for complex biological testing. Third, children who have lower levels of cognitive and adaptive functioning (especially those with severe to profound intellectual disability) present researchers with challenges given the paucity of measures of development and opportunities to test children to establish their developmental level.

These three challenges, and likely others, may explain the relatively small amount of research on the early development of children with intellectual disability. Despite increasingly clear data on the developmental inequalities affecting children with intellectual disability, it may also be the case that international policy has yet to fully recognize the developmental needs and priority for early intervention for this population of children. Without policy prioritization, funding for research on the early development of children with intellectual disability is also unlikely to be easily available, resulting in fewer incentives for researchers to focus their energy and careers in the field of intellectual disability. We propose two inter-linked strategic developments to increase the volume and relevance of research on the early development of children with intellectual disability: Partnership and co-creation; and innovative and creative research designs and methods.

Partnership and co-creation is needed at a policy level, across countries and cultures, and most importantly between families of young children with intellectual disability and researchers. In terms of the latter, our science mapping did not identify a core of research referring to co-creation, co-production, or co-design with families of children with intellectual disabilities. Thus, families may not as a matter of course be involved in contributing to research questions about early development or partnering with researchers in longitudinal research processes. An alternative explanation is that co-production and co-design may have been happening in the research literature but that the way research is reported by scientists does not emphasize these features. Either way, a stronger partnership between families and researchers is required. Closer connection with families will not only ensure directly relevant questions about early development of children with intellectual disability are asked, but will mean that the findings of early development research might be more rapidly applied (at least by families, who have a considerable interest in ensuring the best possible developmental environment for their child). Examples of co-production and co-creation by parents, professionals, and researchers are emerging in neighboring fields, leveraging the opportunities of digital platforms (e.g., a digital platform for asking questions, finding information, and preparing for consultation with professionals for parents of children with physical disabilities; Alsem et al., 2017).

Families of young children with intellectual disability, and researchers in the field of intellectual disability, also have a direct, current and future shared interest in influencing early intervention/early education policy and also research funding policy around the world. Thus, we call for families and researchers to work together strategically to bring early years development and the need for early intervention to the attention of policy makers who can ensure that the early development of children with intellectual disability becomes a policy priority. Research is also needed on developing and evaluating models of co-production between families and researchers, and the putative impact of different approaches to this partnership on policy. With attention to children with intellectual disability in early years/early education policy, families and researchers might then also be able to work together to approach and influence research funders cross-nationally. In addition, families as partners will play a key role in the training and development of researchers working with young children with intellectual disability. To keep the research questions relevant, and to ensure that research methods are inclusive (especially considering the challenges associated with research with children with severe to profound intellectual disability), close connection and partnership with families are crucial.

One strategy to address the challenges outlined earlier (of a rare condition like intellectual disability, methods to ascertain likely intellectual disability early in development, and creative methods to include children with more severe intellectual disability) is to foster scientific partnership. This requires not only that researchers join forces, but research funding agencies, organizations representing and working with

families, and regulators also participate in such teams (cf. Webster, 2019, for the neighboring field of special education). International co-operation could increase available sample sizes, increase the overall size and relevance of research through collaborative funding arrangements, and enhance agreement to use similar measures and tools. For example, in the Netherlands, a minimal data set was developed to facilitate interoperability and reusability of data to answer questions, for example, affecting smaller numbers of children such as those with rare genetic conditions associated with intellectual disability (Kunselser et al., 2016). Research teams working together could also share and plan to address key questions in the field strategically (e.g., one team in one country seeks funding to work on one problem, and a research team in another country works on another problem thus creating synergy; Salas et al., 2018). While co-production needs to be carefully considered on a case by case basis (Oliver et al., 2019), in intellectual disability research many instances can be found where the benefits of co-production will outweigh these costs.

The second area of strategy to change the trajectory of research on the early development of children with intellectual disability is to take advantage of new and emerging research methods and designs. We suggest four general approaches that would benefit early development research in intellectual disability. First, countries around the world have been investing more in administrative data across multiple domains of public policy, but including children and families. Early development research on children with intellectual disability would benefit from international standards for data to gather about young children that would allow those with an intellectual disability to be identified with some confidence. Second, many countries also invest in large population-based studies of children and families. It is possible in some of these to identify children who are likely to have an intellectual disability and related developmental conditions and thus to uniquely consider matters of child development at a population level (cf. Totsika and Hastings, 2012). When making these national investments, it would not take much additional effort for the designers of population surveys to include methods that would allow children with intellectual disability to be more easily identified. Exclusion from population level surveys is in any case likely inconsistent with the rights of people with ID to be included in matters related to their health and well-being (United Nations, 2006). Third, although the consent and data sharing issues will need to be considered very carefully, families of young children with intellectual disability across collaborating international research groups could be approached to join national or international research registers (cf. the Netherlands Autism Register: www.nar.nl; Grove et al., 2018). Fourth, and related also to the previous point, a repository of protocols, measures and methods in longitudinal studies of young children with intellectual disability is needed to make sure that creative and excellent research approaches are more widely used, researchers do not have to “re-invent the wheel,” and that data are more easily combined to consider new research questions without the expense of commissioning new research studies.

CONCLUSION

In the current paper, and science mapping study, we have argued for the importance of basing early interventions for children with intellectual disability on the foundations of high quality developmental research. Mapping the use of early development research in intellectual disability, we found only moderate evidence of links to research on intervention and arguably a concerning disconnection. However, our analysis gives only a partial picture of the state of early developmental science in intellectual disability. A related systematic review and synthesis of early intervention research in intellectual disability would be informative in this regard. It is possible, for example, that early interventions in intellectual disability have been directly informed by mainstream developmental theory and/or research studies of typical development.

Given that we have argued that developmental processes may be relatively universal although likely at a slower pace in intellectual disability, does it matter if there is both a lack of early development research in intellectual disability and a potential disconnection with early intervention science? We contend that the answer to this question is: Yes, it does matter. In particular, it is clear that the social (and especially the family) environment both partially determines and also interacts with children's development. This means that the development of children with intellectual disability is likely shaped by different environmental influences than for other children at the same developmental age. For example, puberty and the social/family response to developing sexuality in a physical sense may occur for adolescents with intellectual disability when their social and communication functioning lags behind their peers. In addition, families' experiences are different: their child with intellectual disability may engage in challenging behaviors that are not only uniquely stressful, but lead to considerable public stigma for families. Modeling the effects of any intervention is thus not as simple as applying a one-size-fits-all "developmental delay" approach. Lacking direct research evidence about the development of children with intellectual disability, any intervention not informed by such research may have unexpected, and even damaging, outcomes.

REFERENCES

- Alsem, M. W., van Meeteren, K. M., Verhoef, M., Schmitz, M. J. W. M., Jongmans, M. J., Meily-Visser, J. M. A., et al. (2017). Co-creation of a digital tool for the empowerment of parents of children with physical disabilities. *Res. Involv. Engagem.* 3:26. doi: 10.1186/s40900-017-0079-6
- American Association on Intellectual and Developmental Disabilities (AAIDD) (2010). *Intellectual Disability: Definition, Classification, and Systems Of Supports*. Washington, DC: American Association on Intellectual and Developmental Disabilities (AAIDD).
- Baranek, G. T., Carlson, M., Sideris, J., Kirby, A. V., Watson, L. R., Williams, K. L., et al. (2019). Longitudinal assessment of stability of sensory features in children with autism spectrum disorder or other developmental disabilities. *Autism Res.* 12, 100–111. doi: 10.1002/aur.2008

Without underpinning developmental research, it is also difficult to understand how or why early interventions are working successfully. This will be especially limiting at the stage where evidence-based early intervention strategies need to be rolled out widely in practice. Successful implementation will, in part, be determined by strategies to maintain the changes in developmental and family processes that are targeted by the intervention. While the Warnock report in 1978 did not focus on the critical need to understand developmental mechanisms in children with intellectual disability, the findings of the current study suggest continuing relevance of high quality longitudinal work.

DATA AVAILABILITY

The bibliographic datasets generated and analyzed for this study can be fully obtained using the search commands in the **Appendix A**, using proprietary databases of Thomson Reuters (Web of Science) and Elsevier (Scopus). The bibliometric map data with which interactive versions of the maps (**Figures 2, 4, 5**) can be created are open and can be accessed at <https://osf.io/s7hde/> (doi: 10.17605/OSF.IO/S7HDE).

AUTHOR CONTRIBUTIONS

CS and RH contributed conception and plan for the review. CS collected bibliographic data and performed the bibliometric analyses. CS, MvR, and CES scored abstracts for eligibility and prepared the flow chart and supplementary tables. CS and RH wrote the first draft of the manuscript. All authors contributed to manuscript revision, read and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/feduc.2019.00041/full#supplementary-material>

- Bertelli, M. O., Munir, K., Harris, J., and Salvador-Carulla, L. (2016). "Intellectual developmental disorders": reflections on the international consensus document for redefining "mental retardation-intellectual disability" in ICD-11. *Adv. Ment. Health Intellect. Disabil.* 10, 36–58. doi: 10.1108/AMHID-10-2015-0050
- Cianfaglione, R., Felce, D., Hastings, R., Kerr, M., and Clarke, A. (2018). "Rett syndrome," in *The SAGE Encyclopedia of Intellectual and Developmental Disorders*, ed E. B. Braaten (Thousand Oaks, CA: Sage), 1384–1387.
- Contopoulos-Ioannidis, D. G., Alexiou, G. A., Gouvas, T. C., and Ioannidis, J. P. A. (2008). Life cycle of translational research for medical interventions. *Science* 321, 1298–1299. doi: 10.1126/science.1160622
- Craig, P., Dieppe, P., Macintyre, S., Mitchie, S., Nazareth, I., and Petticrew, M. (2008). Developing and evaluating complex interventions: the new Medical Research Council guidance. *Br. Med. J.* 337, 979–983. doi: 10.1136/bmj.a1655

- Department for Education and Science (1978). *Enquiry Into the Education of Handicapped Children and Young People* (The Warnock Report). London: HMSO.
- Emerson, E., and Hatton, C. (2007). Mental health of children and adolescents with intellectual disabilities in Britain. *Br. J. Psychiatry*. 191, 493–499. doi: 10.1192/bjp.bp.107.038729
- Emerson, E., Robertson, J., Baines, S., and Hatton, C. (2016). Obesity in British children with and without intellectual disability: cohort study. *BMC Public Health*. 16:644. doi: 10.1186/s12889-016-3309-1
- Grove, R., Hoekstra, R. A., Wierda, M., and Begeer, S. (2018). Special interests and subjective wellbeing in autistic adults. *Autism Res.* 11, 766–775. doi: 10.1002/aur.1931
- Hodapp, R. M., Burack, J. A., and Zigler, E., (Eds.). (1990). *Issues in the Developmental Approach To Mental Retardation*. Cambridge: CUP.
- Kaufman, L., Ayub, M., and Vincent, J. B. (2010). The genetic basis of non-syndromic intellectual disability: a review. *J. Neurodev. Disord.* 2, 182–209. doi: 10.1007/s11689-010-9055-2
- Kunseler, F. C., Schuengel, C., Embregts, P. J. C. M., and Mergler, S. (2016). *Basis Minimale Dataset (MDS) Verstandelijke Beperking [Basic Minimal Dataset (MDS) intellectual disability]*. Vrije Universiteit Amsterdam. Retrieved from: <https://www.zonmw.nl/nl/onderzoek-resultaten/gehandicapten-chronisch-zieken/programmas/programma-detail/gewoon-bijzonder-nationaal-programma-gehandicapten/t/werken-met-de-minimale-data-set/> (accessed March 07, 2019).
- Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P. A., et al. (2009). The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. *PLoS Med.* 6:e1000100. doi: 10.1371/journal.pmed.1000100
- Maulik, P. K., Mascarenhas, M. N., Mathers, C. D., Dua, T., and Saxena, S. (2011). Prevalence of intellectual disability: a meta-analysis of population-based studies. *Res. Dev. Disabil.* 32, 419–436. doi: 10.1016/j.ridd.2010.12.018
- Oliver, K., Kothari, A., and Mays, N. (2019). The dark side of coproduction: do the costs outweigh the benefits for health research? *Health Res. Policy Syst.* 17:33. doi: 10.1186/s12961-019-0432-3
- Salas, E., Reyes, D. L., and McDaniel, S. H. (2018). The science of teamwork: progress, reflections, and the road ahead. *Am. Psychol.* 73, 593–600. doi: 10.1037/amp0000334
- Salvador-Carulla, L., Reed, G. M., Vaez-Azizi, L. M., Cooper, S. A., Martinez-Leal, R., Bertelli, M., et al. (2011). Intellectual developmental disorders: towards a new name, definition and framework for “mental retardation/intellectual disability” in ICD-11. *World Psychiatry* 10, 175–180. doi: 10.1002/j.2051-5545.2011.tb00045.x
- Thornicroft, G., Lempp, H., and Tansella, M. (2011). The place of implementation science in the translational medicine continuum. *Psychol. Med.* 41, 2015–2021. doi: 10.1017/S0033291711000109
- Totsika, V., and Hastings, R. P. (2012). How can population cohort studies contribute to our understanding of low prevalence clinical disorders? The case of autism spectrum disorders. *Neuropsychiatry* 2, 87–91. doi: 10.2217/np.12.12
- Totsika, V., Hastings, R. P., Emerson, E., Berridge, D. M., and Lancaster, G. A. (2011). Behavior problems at 5 years of age and maternal mental health in autism and intellectual disability. *J. Abnorm. Child Psychol.* 39:1137. doi: 10.1007/s10802-011-9534-2
- UNICEF (2017). *Early Moments Matter for Every Child*. New York, NY: UNICEF.
- United Nations (2006). *Convention on the Rights of Persons With Disabilities (CRPD)*. Retrieved from: <https://www.un.org/development/desa/disabilities/convention-on-the-rights-of-persons-with-disabilities.html> (accessed March 07, 2019).
- Van Eck, N. J., and Waltman, L. (2014). *VosViewer Manual*. Leiden: Leiden University. Retrieved from: <http://www.vosviewer.com/> (accessed August 26, 2018).
- Van Eck, N. J., and Waltman, L. (2016). *VosViewer* (Version 1.6.10). Retrieved from: <http://www.vosviewer.com/> (accessed February 19, 2019).
- Waltman, L., Van Eck, N. J., and Noyons, E. C. M. (2010). A unified approach to mapping and clustering of bibliometric networks. *J. Informetr.* 4, 629–635. doi: 10.1016/j.joi.2010.07.002
- Webster, R. (2019). A blueprint for evidence-based practice? Assessing the Warnock Inquiry’s proposals for research and development in special education 40 years on. *Front. Educ.* 4. doi: 10.3389/educ.2019.00017
- World Health Organization (WHO) (2001). *International Classification of Functioning, Disability and Health* (ICF). Geneva: WHO.
- World Health Organization (WHO) (2012). *Early Childhood Development and Disability*: Discussion paper. Geneva: WHO. Retrieved from: <https://apps.who.int/iris/handle/10665/75355> (accessed April 21, 2019).

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