frontiers | Frontiers in Education

TYPE Review
PUBLISHED 17 April 2025
DOI 10.3389/feduc.2025.1479668

OPEN ACCESS

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RECEIVED 12 August 2024
ACCEPTED 31 March 2025
PUBLISHED 17 April 2025

CITATION
Tosolini KE, Damen S, Janssen MJ and Minnaert AEMG (2025) A Piagetian lens on cognitive development of children and youths with congenital deafblindness: a scoping review.
Front. Educ. 10:1479668.
doi: 10.3389/feduc.2025.1479668

A Piagetian lens on cognitive development of children and youths with congenital deafblindness: a scoping review

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Children with congenital deafblindness (CDB) experience several developmental delays, but an overview of studies on the cognitive development of children and youths with CDB is lacking. In present review, Jean Piaget's theory of cognitive development was used as a framework to gather and summarize the literature on cognitive development of children and youths with CDB. A scoping review was performed to create a comprehensive overview of studies describing Piagetian cognitive skills in children with CDB. Furthermore, the literature was investigated to get insight into the role of caregivers in the development of Piagetian cognitive skills of children with CDB. Lastly, this scoping review investigated how the Piagetian cognitive skills found in the literature are assessed. Thirteen articles were included in the analysis. Descriptions of cognitive skills were mainly found for the sensorimotor and pre-operational stages. Caregiver support was found to be essential to improve learning opportunities. Dynamic assessment and questionnaires were found to be suitable methods to assess cognition in persons with CDB. More research is needed to evaluate how Piagetian skills develop over time in children and youths with CDB.

KEYWORDS

congenital deafblindness, cognitive development, Piaget, developmental stages, caregiver support, assessment

1 Introduction

Congenital deafblindness (CDB) is a distinct disability that involves a combined vision and hearing impairment of varying degrees. Present review defined CDB in accordance with Dammeyer (2010a) as a combined sensory loss that is either present from birth or has a pre-lingual onset. The vision and hearing loss experienced in CDB is far more impairing than losing a single sense, as one cannot compensate the one sense with another (Dammeyer, 2012). Even a slight impairment of both senses may hinder social interactions and affect cognitive and communicative development (Dammeyer, 2012; Rødbroe and Janssen, 2006; World Federation of the Deafblind, 2018). Children with CDB are more reliant on their tactile sense and therefore have limited and fragmentary access to their environment (Damen et al., 2020). Due to this fragmentary perception, children with CDB have serious problems in three main areas: communication, access to information and mobility (Nelson and Bruce, 2016; World Federation of the Deafblind, 2018). Furthermore, the unique challenges that children with CDB experience can affect their cognitive development (Nicholas, 2020). This may result in several developmental delays in persons with CDB (Damen et al., 2020). The information that

fragmentary perception can provide is often incoherent and disorganized, and children with CDB have difficulties connecting this information to existing knowledge and storing it accordingly. This hinders the child's ability to see relations between events in the surroundings and, accordingly, affects the development of object permanence (Van Dijk and Janssen, 1993). Another consequence is a lesser engagement in exploring their environment resulting in having more difficulty in developing cognitive schemes needed for further development (Van Dijk et al., 1997), since they lack the experience and practice with the world around them.

Yet little research has been conducted on CDB and cognitive functioning (Dammeyer, 2010b). Specifically, there is a lack of studies on cognitive development and factors influencing this development in people with CDB (Ravenscroft and Damen, 2019). A lack of clear information about cognitive development in children with deafblindness can lead to the risk of missing the cognitive potentials of a child with CDB (Rødbroe and Janssen, 2006). Therefore, the authors aimed to contribute to the scientific field of deafblindness by creating the first overview of studies on cognitive development of children with CDB. Since cognition is only accessible through interpretations that we make from observable behavior (Larsen and Damen, 2014), an important first step is to apply a theoretical model of cognition to observable behavior of children with CDB. We chose the perspective of one of the major theories of child development of twentieth-century psychology (Barrouillet, 2015): the developmental theory of Jean Piaget.

The choice of a Piagetian lens for our literature review is based on the impact Piaget's theory of cognitive development had on developmental scientists, educators and others concerned with the course of children's development (Leman et al., 2012). His theory paved the way for most later research on cognition and is still a cornerstone within the field of psychology to this day. The impact and relevance of his theory on later research is emphasized by the extensive amount of secondary literature that was written based on his work—more than 20.000 texts (Ratcliff, 2023). Ratcliff's historical research on Piaget's work shows that Piaget contributed largely not only to the psychological field, but also inspired many other disciplines like pedagogy, philosophy, literature, human sciences and many more. From the early years of the 20th century, he was already considered a classic author, even though his theory was also subject to criticisms and changes. Piaget himself even revised his initial "Standard Theory" to what can be seen as his "New Theory" (Beilin, 1992a), which emphasizes that research on the development of children is always evolving. After the "New Theory," a subsequent rediscovery of his teachings then again growingly inspired new publications (Ratcliff, 2023). To this day, Piaget's work is being used as a foundation for new developments. By applying Piaget's theory to the descriptions of observable behavior in children with CDB in the literature, the authors expected to get new insights in the Piagetian cognitive skills of these children and identify areas that may need support.

An important idea introduced by Piaget is the notion of "schemas" or "cognitive structures," which can be seen as the building blocks of intelligence (Flavell, 1963). These schemas function through the processes of assimilation, by which experiences are understood through existing schemes, and accommodation, the process of modifying a scheme to incorporate a new situation. Piaget believed that through these processes, the structure of a child's schemes undergoes qualitative transformation over time. Piaget ordered these qualitative transformations in several stages. He proposed four stages of development (see Table 1), which occur universally in a fixed order (Gruber and Vonèche, 1977): (1) the sensorimotor stage, (2) the preoperational stage, (3) the concrete operational stage and (4) the formal operational stage (Piaget and Inhelder, 1966).

The sensorimotor schemes that develop in the first stage consist of organized systems of action on objects (Mascolo, 2015). During the sensorimotor stage, infants cannot form representations, the capacity to evoke an absent object or an action not yet carried out (Piaget, 1952), without direct sensory contact. Motor development is also a major task in this phase (Piaget and Inhelder, 1966). At the end of the sensorimotor period, the ability of maintaining a mental image of an object appears: the child now knows that objects exist even if he does not see them (Piaget, 1954). The practical knowledge developed during this first stage will form the basis for the child's ability to form mental representations of the world in later Piaget's stages (Gruber and Vonèche, 1977).

During the preoperational stage, the child develops the ability to represent something by means of a "signifier": an object, event or word that has a symbolic meaning (Piaget and Inhelder, 1966). Children in this stage learn to form representations of events (e.g., words, images) but cannot manipulate them in logical or systematic ways (Mascolo, 2015). In this phase, the child is limited to his own perspective (egocentrism), believing that everyone around him shares his view of the world (e.g., a child closes his eyes and thinks others cannot see him anymore). Further development and refinement of the use of language, based on the knowledge and first signifiers developed in the sensorimotor stage, is one of the most significant developments during the stage. Furthermore, play evolves. Children in this stage do not only perform functional play (i.e., repetitive actions that the child finds enjoyable) and constructive play (i.e., the child manipulates object to construct something); in this stage, children also develop symbolic play. Symbolic play shows the ability to perform make-believe situations and is seen for example when a child is using available objects to represent something else, such as a spoon being waved in the air to represent an airplane, or perform role playing. However, at the preoperational stage, the child is not yet able to perform concrete operations that develop in the subsequent stage (Gruber and Vonèche, 1977).

During the concrete operational stage, with the capacity to think logically using concrete images and representations, children can successfully perform a variety of logical tasks called concrete operations. Concrete operations are operations such as class inclusions, e.g., fathers united with mothers constitute parents, or the reversibility of terms and objects: the opposite of uniting is separating etc. Concrete operations provide a transition between schemes of action of the preoperational stage and general logical structures of the concrete operational stage (Piaget and Inhelder, 1966). Children in this stage learn to order the world by structures and concepts and learn for example to compare lengths and quantities. Children in this stage learn to consider multiple aspects of situations, such as multiple characteristics of objects or the perspectives of other people: they develop out of their egocentric perspective.

Finally, in the formal operational stage, individuals gain the capacity of logical thought using abstract ideas without the need for tangible entities (Mascolo, 2015). Children acquire flexibility in thinking as well as the capacities for abstract thinking and mental

TABLE 1 Piaget's stages of cognitive development.

Stage	Development of the child	
	Substage/skill	Description
Sensorimotor stage (0–2 years)	Simple reflexes (0–1 month)	Understanding of environment is attained through reflexes such as sucking and crying.
	Primary Circular Reactions (1–4 months)	New schemas and sensations are combined, allowing the child to deliberately engage in actions that are pleasurable, such as sucking his thumb. Movements are mostly centered on the child himself and not destined to achieve and maintain a result in the environment.
	Secondary Circular Reactions (4–8 months)	The child is now aware that his actions influence his environment and purposefully performs actions in order to achieve a desired result. Actions are centered on a result produced and the aim of the action is to maintain that result. Actions can be divided in actions that bring repetition of pleasurable experiences with objects; bodily movements as means of recognition as a response to an event, since language is not yet available; and actions to make an interesting spectacle last by using bodily movements previously used in prolonging other pleasurable experiences.
	Coordination of Reactions (8–12 months)	The child now explores his environment and often imitates the behavior of others. The child is not only interested in repeating or prolonging an interesting effect that he has observed by chance, but pursues an end not immediately attainable by different means. The child is interested in new objects and events but applies only familiar schemata to them, adapted to the new situations. Start of object permanence.
	Tertiary Circular Reactions (12–18 months)	The child begins to experiment and try out new behavior to meet goals. The child seeks novelty, tries different actions and discovers new means to examine all the modalities and aspects of a situation.
	Early Representational Thought (18–24 months)	The child now has sufficient awareness of relationships between events that he can make reasoned previsions. Child does not only apply motor schema but also representative schema. Child begins to recognize and appreciate symbols that represent objects or events. Imitation is characteristic of this stage.
Pre-operational stage (2–7 years)	Symbolic representation	The ability to represent something by means of a signifier.
	Symbolic play	Children perform make-believe play with objects or role-play.
Concrete Operational stage (7–11 years)	Seriation	The ability to (mentally) arrange items based on a quantifiable dimension, such as height or weight.
	Classification	The ability to identify the properties of categories, to relate categories or classes to one another, and use the categorical information to solve problems. E.g., group objects according to some dimension they share.
	Reversibility	The ability to recognize that numbers or objects can be changed and returned to their original condition.
	Conservation	The understanding that something stays the same quantity even though its appearance changes.
	Decentering	The ability to consider multiple aspects of a situation, think about multiple characteristics of objects at the same time and consider perspectives of other people.
	Transitivity	The ability to recognize relationships among various things in a serial order.
Formal Operational stage (12+ years)	Abstract Thought Hypothetic Deductive Reasoning	Thought in this phase does not need to be connected to the concrete objects anymore. Children are able to perform mental experiments and test mental hypotheses.

hypothesis testing; they learn to consider possible alternatives in complex reasoning and problem solving (Piaget and Inhelder, 1966).

According to Piaget, children go through the stages in a universal and unidirectional manner, and later stages build upon earlier ones, making it a hierarchical structure (Mascolo, 2015). In all stages, children go through the process of "equilibration" (Mascolo, 2015; Piaget, 1964): stages have an initial, disorganized period, that lacks stability, where the child is likely to show a mix of earlier structures and incomplete new structures, followed by an achievement period, where the structures function as an integrated whole ("structures d'ensemble"). Some periods in an individual's development are therefore more stable than others, and structures of earlier stages are gradually incorporated into those of later stages, by giving a new meaning to them and integrating them in a new, qualitatively different, and even more complex structure (Flavell, 1963; Morra et al., 2008).

During the years, Piaget's theory has been the target of different criticisms. For instance, Piaget used strict age-thresholds while in practice many age differences are seen between children (Bremner and Wachs, 2014). Furthermore, there has been critique on Piaget's idea that children "actively build" their own development (Piaget, 1964), inferring the impossibility of stimulation of their development. Children from different contexts and countries do in fact receive substantive benefits from interventions that stimulate their cognitive development (Nores and Barnett, 2010). Furthermore, the profound impact caregivers have on the development of children has become clear. In a child's early life, caregivers can fulfill as an external regulatory function when it comes to emotions (Callaghan and Tottenham, 2016) and as mediators between the child and the environment (Hertzman and Boyce, 2010). This way, they support them in developing cognitive, emotional, physiological and behavioral strategies to learn and grow (Singletary, 2019). Thus, criticism suggested that Piaget failed to account for the importance of the social nature of human development (Matusov and Hayes, 2000), even though Piaget actually did not deny the role of what he called social or educational transmission (Piaget, 1964). He acknowledged that adults play a role in children's development, but argued that social transmission in itself is not enough: to receive the information provided by adults, the child must firstly have a structure that enables

him to assimilate that information. Although this idea was later also subject to criticism, these challenges to Piagetian theory introduced core theoretical issues about cognitive development that continue to drive research even today (Miller, 2022). This emphasizes the historical relevance of Piaget's work. Piaget's theory shaped the course of developmental psychology up to the present by not only providing his comprehensive account of development, but also by stimulating criticisms that opened up important new research areas (Miller, 2022).

Since Piaget's ideas are still being used to assess cognitive functioning of both typically developing children and persons with a wide arrange of disabilities, Piagetian principles of development also apply to the development of children with CDB. Research accentuated that Piaget's principles generally apply to people with other disabilities, for example intellectual disability (Morgan, 1986). Children with intellectual disabilities (ID) go through the same sequence of stages of development as children without an ID, but need more time compared to children without ID to develop through the stages (Dockrell and McShane, 1992, cited in Agheana and Folostina, 2015; Weisz and Zigler, 1979). Similarly, children with Down syndrome have been found to follow the same developmental path as typically developing children, although at a slower rate (Dunst, 1990). In other research, Bruce and Muhammad (2009) found that in children with blindness, object permanence develops in a similar sequence though at a slower rate than in children without disabilities, and Peterson (2009) found that Theory of Mind (ToM) is delayed in deaf children born into a hearing community, even though its development progresses for a longer period of time, compared to hearing children. Baron-Cohen (1991) furthermore suggests that autistic children are delayed in their development of ToM due to earlier delays in joint attention and understanding of proto-declarative pointing. This last task shows some similarities with the Piagetian "decentering" task. Finally, Weisz and Zigler (1979) reported findings in which persons with an ID follow the same stages of development in many Piagetian areas such sensorimotor spatial concepts, object permanence, seriation, transitivity, comparison processes, mental imagery, classification, class inclusion and many more.

Similarly, Piagetian principles can be applied to the development of children with CDB. Piaget's observations of the development of infants can provide important information for the assessment of children with CDB. Delays in mastering several cognitive skills are expected in these children, as this is also the case for children with other disabilities. Children with CDB experience only small fragments of the environment compared to the complete sensory information acquired by someone who can see and hear well. Understanding the totality of a situation, based on this fragmented experiences, requires much more effort and processing time (Van Dijk et al., 1997). Since the use of sight and hearing plays a fundamental role in the development of schemata in children (Piaget, 1952), the fragmentary sensory experiences of children with CDB likely have an impact on the information that is assimilated, and, consequently, the content that will be accommodated could differ from that of hearing and sighted children. From the very first circular reaction stages, children progressively accommodate and assimilate what they see and hear. At first, they are only able to perform reproductive assimilation: the assimilation is purely functional, achieved by simply looking and hearing—it revolves around the pleasure of repetition of what they hear and see. Then, by practicing with looking around from different perspectives and listening to or producing increasingly varied sounds, generalizing and recognitory assimilations take place, which brings with it the formation of schemata (Piaget, 1952). Furthermore, from a very young age, coordination exist between vision and hearing. This intersensorial coordination ensures that, e.g., visual schemata are not only coordinated amongst themselves, but also serve vaster assimilations between senses. Piaget states that only by understanding the totality, a meaning can be conveyed to the environment. A possible consequence of CBD could then be that children with this condition, since recognizing, comparing and integrating information within existing knowledge is hard and time-consuming (Damen et al., 2020; Van Dijk et al., 1997), have more difficulty assimilating, accommodating and reaching a state of equilibrium. Since sensorimotor experiences form the building blocks of all later developmental stages, the impact of CDB on these very first experiences possibly greatly impacts the achievement of later developmental stages.

Present scoping review has three aims. Firstly, it aims to provide a comprehensive overview of studies of children with CDB that investigated cognitive skills that are mentioned by Piaget in his Standard Theory. The review did not focus on the processes of acquisition of these skills. Even though we expect that CDB impacts the content of what children with CDB can accommodate and assimilate, since the use of sight and hearing plays a fundamental role in these processes (Piaget, 1952), we wanted to take a first step by focusing on the extent to which and how the various cognitive skills are described in literature. Furthermore, as we expected that most publications would not specifically mention Piagetian skills, we looked for descriptions of observable behavior to be able to link the information to Piagetian skills. Even though we acknowledge that Piaget and others later approached development more as a continuous process rather than a sequentially distinct one, we used his Standard Theory for our analysis of the literature, as it provides a fitting framework for the extraction of data on cognitive development within a population in which, as far as we know, cognitive milestones are not very well described. Thus, the first research question that guided the study is: "How are Piagetian cognitive skills described in literature about children and youths with CDB?"

The second aim of the study is to investigate the role of caregiver support in the development of Piagetian skills of children with CDB. Caregiver support does not play a central role in Piagetian theory, but parental input is considered a source that children use (Maccoby, 1992). Parents in Piagetian theory are seen as an enabling force, and children benefit from the opportunities provided by caregivers to develop cognitive structures (Fuggle et al., 2012). Children use their already developed structures to assimilate information and events provided by adults. Thus, even from a Piagetian perspective, cognitive development cannot be observed separately from their interactions with adults. Furthermore, clear insights into which parental support children with CDB can benefit from, when it comes to the development of Piagetian cognitive skills, are lacking. The second research question therefore is: "How is the role of caregiver support in the development of Piagetian cognitive skills for children and youths with CDB described in the literature?"

The third aim of the study, since assessment of cognitive skills in children with CDB encounters recurring complexities, and suitable assessment instruments within the field are limited, a third aim of the study is to investigate which assessment instruments are used in the literature to assess Piagetian cognitive skills in children with CDB. With the third research question, namely "How are Piagetian

cognitive skills assessed in literature about children and youths with CDB?," The authors wanted to find out if the Piagetian skills described in the literature were assessed with specific instruments and if so, which instruments were used to assess them. Additionally, the authors wanted to find out whether any of the instruments that are described in the literature, are based on Piaget's theory.

2 Method

This study utilized a scoping review to assess what is known about Piagetian developmental stages in relation to the development of children with CDB. A scoping review aims to identify key concepts and gaps in the research literature within a particular research area by systematically searching and synthesizing existing knowledge (Jaiswal et al., 2018). By identifying, evaluating and summarizing the findings of all relevant studies about cognition in deafblind children, a comprehensive overview of studies will be made available. In this scoping review, the methodological framework published by Arksey and O'Malley (2005) was used, which comprises five stages: identifying the research question, identifying relevant studies, selecting studies, charting the data, and then collating, summarizing, and reporting the results.

2.1 Stage 1: identifying the research question

The three central research questions can be found in the introduction.

For the purpose of this study, the population "children and youths with congenital deafblindness" is defined as children and youths aged 0 to 25 with a combination of visual and hearing impairments in various degrees. The term "congenital" had to be mentioned, or a description had to be given mentioning that the impairment was present from birth, had an onset before language development (before the age of two) or had a genetic cause known to be related to CDB, for example CHARGE syndrome. The term "caregivers" is defined as parents, teachers or professional caregivers providing children and youths with CDB with fundamental care.

2.2 Stage 2: identifying relevant studies

Advanced searches were conducted in MEDLINE, ERIC, PsycINFO and Web of Science databases using the search terms that are presented in Table 2. For replicability of the search, "apply equivalent subjects" was removed in the search engines. No date limitation was applied to the search, since the central theme of cognition has been a point of interest for many decades.

As there is no unified definition of the term deafblindness, an extensive array of search terms has been used for the target population to ensure that all possible combinations were used. As for the search terms for cognition: general terms to indicate cognition including cognitive development or cognitive functions have been used, to ensure a broader range of articles.

In addition to the electronic search, further relevant references were explored by means of the snowballing method. This method is considered an important approach to search for primary and relevant studies in addition to database search (Wohlin et al., 2022). It is an effective additional method of information retrieval when an area or question is difficult to search electronically (Horsley et al., 2011).

TABLE 2 Search terms.

Search terms for target population	Search terms for cognition
((deaf AND blind) OR deafblind* OR "deaf-blind*" OR "Dual sensory loss" OR "Dual sensory impair*" OR ("visual impair*" AND "auditory impair*") OR ("visual disab*" AND "auditory disab*") OR ("vision loss" AND "hearing loss"))	(cognition OR "cognitive develop*" OR "cognitive abilit*" OR "cognitive skill*" OR "intellectual develop*" OR develop* OR "cognitive function*)

2.3 Stage 3: study selection

The search resulted in the identification of 3,339 articles in total. After removing duplicates and applying initial search criteria, 1,647 articles remained. A two-stage screening process was used to assess the relevance of these studies, first at the level of title and abstract screening and second at the level of full text review. In the first stage, the titles and abstracts of the 1,647 articles were assessed based on the application of the following criteria: (a) written in English (b) focusing on young people with CDB (0–25 years), (c) focusing on their cognitive development and/or assessment thereof and (d) focusing on aspects of Piagetian theory of cognitive development. In the first stage of title and abstract screening, the authors included not only articles mentioning Piagetian skills but also articles mentioning general cognitive development or general intellectual development to prevent improper exclusions of articles and allow a broader range. This was done in light of our expectation that many authors would not explicitly mention Piagetian skills, but would rather measure or describe behaviors that would be indicative for those skills. Therefore, this screening stage was purposefully kept broad. For example, articles mentioning general terms as "case studies," "characteristics of individuals with CDB," or "the development of children with CDB" in the title or abstract, were included in this stage. To avoid missing information, articles of which the title and abstract did not clearly reveal whether they related to CDB rather than other types of sensory disabilities, were also included.

The selection of articles on specific Piagetian skills was then performed at the second stage, where articles were fully assessed based on the application of the following inclusion criteria: (a) written in English (b) data focused on young people with CDB (0–25 years), (c) data refers to aspects of Piagetian theory of cognitive development. Table 1 provided the fundamental aspects of Piagetian cognitive skills that were used during the search to select relevant information. Of the 1,647 articles, 95 qualified for full-text assessment based on the screening of titles and abstracts. A recurring problem was found during full-text screening: 32 of the articles published before the year 1990 were not available for analysis. An example is that some of the publications were chapters that were published such a long time ago, that they were not accessible through the university library's databases or other libraries. These 32 publications were therefore excluded. During full text analysis of the remaining 63 articles, it became clear

that 14 articles did not focus on CDB. Therefore, they were excluded. Thus, 49 articles remained for the application of the beforementioned inclusion criteria. Since the title and abstract screening was kept purposefully broad, these 49 articles that remained for full text analysis eventually were quite diverse. Of these articles, 20 articles were excluded due to not focusing on cognitive development but rather on for example, language development. Of the remaining articles, 19 were excluded due to the article not containing observable descriptions of cognitive skills that could corresponded to Piagetian theory. Ultimately, 10 articles were included. Finally, a cited reference lists search was conducted using the same criteria. Three publications were added by searching the cited reference lists, increasing the total number of articles to 13.

A flow diagram is presented in Figure 1, following the steps of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA statement) (Moher et al., 2009). EndNote and Rayyan (Ouzzani et al., 2016) were used to manage references, delete duplicates and to screen titles and abstracts of potentially relevant papers. The Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a variety of fields (Kmet et al., 2004) was implemented to screen the quality of included articles. The PRISMA extension for Scoping Reviews (PRISMA-ScR) checklist was used as a guideline for reporting the results (Tricco et al., 2018).

2.3.1 Inter-rater reliability

All the papers were fully assessed by the first author using the criteria mentioned above. A second independent reviewer assessed a random subset of 20% of the papers of the electronic search based on title and abstract. An inter-rater reliability analysis was performed by calculating Cohen's kappa, to determine the consistency between the

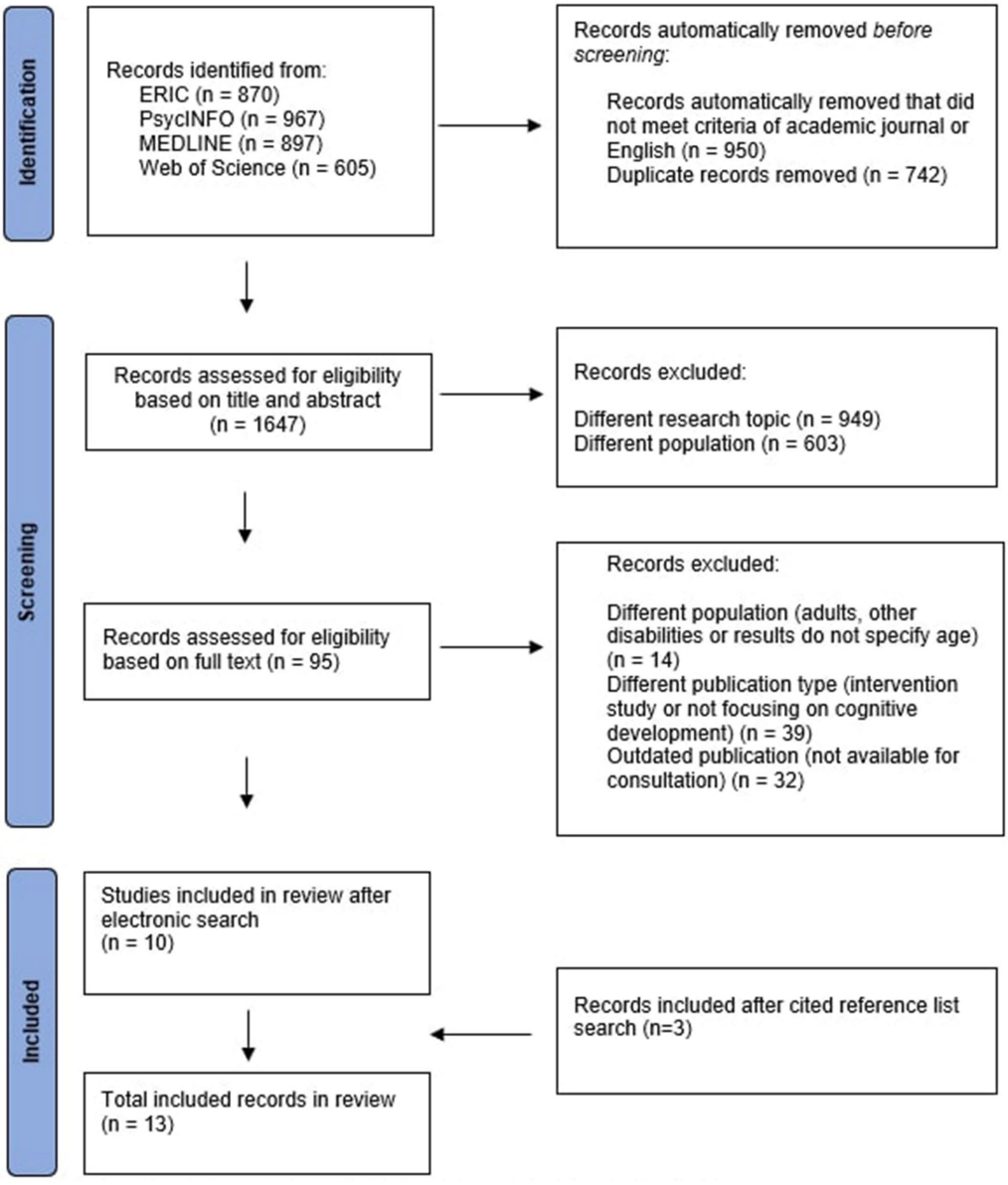

FIGURE 1
Flow of information during the electronic search phase of the scoping review.

two raters. The kappa value was 0.71, indicating a substantial agreement (McHugh, 2012). The percentage of agreement was 97.5%. Discrepancies were mainly caused by different deliberations about "general cognitive development." Studies about language and communication development for example were difficult to judge for their eligibility, since language is intricately connected with cognitive development. Only the development of symbolization as a Piagetian skill was a focus of present review, but reviewers made different deliberations in three cases concerning communication development. Other discrepancies concerned different deliberations about motor development. Another independent reviewer (second author) assessed the analysis of the content of the final inclusions. Only two discrepancies were found within the simple reflexes stage and the symbolic representation stage. The authors discussed the discrepancies until they reached consensus on which information to include in the review.

2.3.2 Quality assessment

Quality assessment of the papers was performed by the first author using the Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a variety of fields (QualSyst) (Kmet et al., 2004). Kmet et al. (2004) describe a checklist to assess the quality of studies, consisting of 14 items to evaluate quantitative studies and 10 items to evaluate qualitative studies. The quality assessment was applied to all empirical studies. Threshold for inclusion was set at a score of 50%. The QualSyst could only be applied to papers presenting empirical studies containing a method section. Other types of papers were still included, to account for a more comprehensive overview of information, given the fact that extensive research on this topic is still lacking.

2.4 Stage 4: charting the data

Extracted data included details regarding name of the authors, year of study, study population, number of participants, central topic relevant to this scoping review and key outcomes.

2.5 Stage 5: summarizing and reporting the results

The results were reported using descriptive summaries. Central topics important to this scoping review were extracted from each article and were summarized. The retrieved data was ordered in a descriptive manner, matching the descriptions to Piagetian stages of development.

3 Results

3.1 Characteristics of included publications (n = 13)

The total search yielded 13 publications. Of the 13 publications, eight were empirical studies, of which one of them a case description (Damen et al., 2020). Three were theoretical contributions that were published in peer-reviewed scientific journals, namely the Journal of Visual Impairment & Blindness and the International Journal of Disability, Development and Education, (Bruce, 2005a, 2005b; Hartmann, 2012) and two were assessment manuals (Nelson et al., 2009; Nicholas, 2020). An overview of the characteristics of the included publications is seen in Table 3. Of the two assessment manuals, Nelson et al. (2009) contained extensive case descriptions, whereas Nicholas (2020) contained short examples and pictures of persons with CDB performing activities with objects without further specifications of, e.g., the age of the person. Therefore, these last ones were not included as case descriptions but were only used to answer the question about assessment methods.

The QualSyst tool was applied to the empirical studies. One study was of adequate quality (Score 50–70%), and six studies were of good to strong quality (score > 70%). Mean quality score of the empirical studies was 87%. The QualSyst Tool was not applied to the theoretical contributions, assessment manuals and case descriptions, not having a methodological design.

The number of participants described in the studies varied from single-cases (n = 5), groups of 17 children (n = 1), groups of parents and teachers (n = 3) to datasets of more than 70 children (n = 1). The age range of children and youths was 0–18 years. The available information about cause of deafblindness and severity of impairments and other disabilities is described in Table 3. Topics of the theoretical contributions included the development and understanding of symbols in children with deafblindness (Bruce, 2005a; Hartmann, 2012) and the importance of the concept of "distancing" during development (Bruce, 2005b). The two assessment manuals introduced the Child Guided Strategies assessment and the Tactile Working Memory Scale (Nelson et al., 2009; Nicholas, 2020). The remaining empirical studies focused on: communication and language profiles of children with CDB (Dammeyer and Larsen, 2016); a case description of using the child guided strategies in the assessment of a girl with CDB (Damen, 2020); video-observation of a case study (Murdoch, 1994); perceptions of teachers and parents on the cognitive functioning of their children with CDB (Narayan and Bruce, 2006; Narayan et al., 2010); the application of the Van Dijk approach to assessment (Nelson et al., 2002); language and play (Pizzo and Bruce, 2010) and analysis of interactions (Vervloed et al., 2006).

Only three publications explicitly mentioned Piaget or Piagetian concepts. In Bruce (2005b), Piagetian theory was mentioned within the theoretical framework or the article. In Vervloed et al. (2006), the participating child was mentioned to be functioning at Piaget's sensorimotor stage 2. And in Nelson et al. (2002), Piagetian theory is mentioned as one of the theories on which the used assessment method is based. All other articles did not explicitly mention Piagetian theory.

Descriptions of cognitive skills in children with congenital deafblindness corresponding to the sensorimotor stage of development were found in eight empirical studies and two theoretical contributions. Four empirical studies and three theoretical contributions were found containing information about skills in the pre-operational stage. For the concrete operational stage, one empirical study and one theoretical contribution was found, and neither studies, nor theoretical contributions were found for the formal operational stage. Table 4 gives an overview of the included studies and contributions for each developmental stage.

TABLE 3 Characteristics of included publications.

S. No.	Reference	Publication type	Participants	Research methodology
1	Bruce (2005a)***	Theoretical article	-	-
2	Bruce (2005b)***	Theoretical article	-	-
3	Damen et al. (2020)***	Case description	One girl with CDB aged 5 born with deafness and blindness as a result of prematurity. Severe motor disability.	-
4	Dammeyer and Larsen (2016)*	Empirical article	71 children with CDB aged 3–18	Questionnaire
5	Hartmann (2012)***	Theoretical article	-	-
6	Murdoch (1994)**	Empirical article	One boy with CDB, 14–25 months, blind with light perception in one eye and moderate to severe sensorineural hearing loss as a result of prematurity.	Observations of video-recordings
7	Narayan and Bruce (2006)*	Empirical article	Eight teachers of children with CDB and eight parents of children with CDB (aged 4–12)	Questionnaire
8	Narayan et al. (2010)*	Empirical article	Eight teachers and eight parents of children with CDB aged 4–12 from the USA, and 19 teachers and 23 parents of children with CDB aged 3–12 from India. Diagnoses of the children with deafblindness from the USA were CHARGE syndrome (3), Retinopathy of Prematurity (RoP) (1), deafblind/developmental disabilities (1), multiple anomalies, including deafblindness (3). Among the deafblind group in India, the details on diagnosis included deafblind (15), Retinopathy of prematurity (3), multiple disabilities with sensory impairment (3).	Questionnaire
9	Nelson et al. (2002)*	Empirical article	One girl with CDB aged 7 with high myopia and mild to moderate hearing loss due to a rare genetic syndrome.	Case study
10	Nelson et al. (2009) ***	Case description	One girl with CDB aged 7 with high myopia, cortical visual impairment and undetermined hearing loss due to Wolf-Hirschhorn syndrome and one boy with CDB aged 2 with blindness, undetermined hearing loss and Zwellweger syndrome.	-
11	Nicholas (2020)***	Assessment manual	-	-
12	Pizzo and Bruce (2010)*	Empirical article	Parents and teachers of 6 students with CDB aged 3–10. Diagnoses varied from CHARGE syndrome (2), 1 demonstrated CHARGE-like characteristics, 2 were born prematurely, one had meningococcal meningitis at age 6 months. Two students had severe visual impairments, 3 were legally blind and 1 had mild visual impairment. The level of hearing loss varied from bilateral severe hearing loss (2), bilateral severe to profound hearing loss (1), bilateral moderate to severe hearing loss (1), moderate to profound hearing loss (1) and unilateral moderate hearing loss (1).	Questionnaire
13	Vervloed et al. (2006)*	Empirical article	One boy with CDB aged 3;4 and his teacher. The boy was born prematurely and was deafblind due to congenital rubella syndrome. He has a profound hearing loss and severe visual impairment.	Observations of video-recordings

*Methodological quality score > 70% (good to strong quality).
**Methodological quality score 50–70% (adequate quality).
***QualSyst not applied.

10.3389/feduc.2025.1479668

TABLE 4 Overview of included empirical studies and theoretical contributions for each developmental stage.

Stage	Developmental characteristics	Empirical studies	Theoretical contributions
Sensorimotor stage	Simple Reflexes	-	-
	Primary Circular reactions	Nelson et al. (2009).	-
	Secondary circular reactions	Murdoch (1994); Narayan and Bruce (2006); Narayan et al. (2010); Nelson et al. (2002); Nelson et al. (2009); Damen et al. (2020).	Bruce (2005a).
	Coordination of reactions	Narayan and Bruce (2006); Narayan et al. (2010); Murdoch (1994); Damen et al. (2020); Nelson et al. (2009); Vervloed et al. (2006).	Bruce (2005a); Bruce (2005b).
	Tertiary circular reactions	Narayan et al. (2010); Damen et al. (2020).	
	Early representational thought	Dammeyer and Larsen (2016); Nelson et al. (2002); Nelson et al. (2009).	Bruce (2005b).
Pre-operational stage	Symbolic representation	Dammeyer and Larsen (2016); Narayan et al. (2010).	Bruce (2005a); Hartmann (2012).
	Symbolic play	Narayan and Bruce (2006); Narayan et al. (2010); Pizzo and Bruce (2010).	Bruce (2005b).
Concrete operational stage	Seriation	-	-
	Classification	Narayan and Bruce (2006).	Bruce (2005a).
	Reversibility	-	-
	Conservation	-	-
	Decentering	-	-
	Transitivity	-	-
Formal operational stage	Abstract thought	-	-
	Hypothetic reductive reasoning	-	-

3.2 The sensorimotor stage

3.2.1 Description of cognitive skills

For all substages of the sensorimotor stage except for the simple reflexes stage, a description was found. For the primary circular reactions substage, one description was found in Nelson et al. (2009), where a boy (age 2) was observed intently pulling the ball to his face, which is a sensitive tactile area, to better feel the ball and get more information about it.

Six studies contained descriptions of secondary circular reactions. In these studies, children with deafblindness demonstrated to purposefully perform an action in order to achieve a result. Two studies described examples of children with CDB performing actions to repeat or prolong pleasurable experiences with objects. In the case study of Damen et al. (2020), a five-year-old girl with deafblindness made different efforts to make a toy vibrate again using her hands and face. In the study of Narayan and Bruce (2006), seven teachers and seven parents reported toy activation and switch use as examples. One study described an example of child with CDB expressing recognition of objects or events through bodily expressions: Nelson et al. (2009) described a boy aged 2 signaling that he wanted a vibrator to vibrate on his foot again by lifting his foot, showing that he recognized the object and the previous use of it. He also vocalized to get a toy vibrating again. Four studies described children with CDB performing actions to make an interesting spectacle last by using bodily movements as communicative expressions. Murdoch (1994) described for example how a child (14–25 months of age) clenched his hand on his mother's throat to ask for more vocalizations. In the study of Nelson et al. (2002), a girl with deafblindness aged 7 moved her hands up and down, indicating that she wanted the routine to continue. In the study of Narayan et al. (2010), 75–85% of teachers and parents reported that their children understood that they could influence their surroundings: one child knew for example that when she clapped, her mom would come. In Nelson et al. (2009), a girl aged 7 put her hands together and moved her hands up and down to indicate that she wanted the movement of the tie to continue. She also signalled by bobbing her head and touching her chest that she wanted the singing to continue, showing that she understood her influence on the actions of others.

For the coordination of reactions substage, Vervloed et al. (2006) showed that 18.2% of the interactions between a three-year-old child and his caregiver were initiated by the child to influence the adult's behavior with the goal of overcoming an obstacle. The child touched, manipulated or pushed the adult's hand or arm in order to obtain a desired behavior or object from the adult or to avoid an undesired situation. An example was that the child pointed when he wanted a favorite object. Other examples from this stage mostly described how children with deafblindness explored the environment and mastered object permanence. Murdoch (1994) described how a child (14–25 months of age) explored familiar and unfamiliar faces and compared them to his own, by feeling his father's beard, his uncle's beard and then feeling his own cheek. Narayan and Bruce (2006) reported that all participating teachers in their study saw in their children the ability of exploration. Narayan et al. (2010) describe that at least 75% of participating teachers and parents reported that children with deafblindness explored. Descriptions included discussions of how specific senses were used by the children. Exploring by touch was reported to be common among all children. Other examples were pushing, pulling and pressing objects to know what it does or exploring objects by touching them, feeling around them and

feeling them inside out. All teachers and seven parents in Narayan and Bruce (2006) study reported the understanding of object permanence. An example: two children were reported to come to the rooms of the parents to find them first thing in the morning, showing the knowledge of permanence of people. Narayan et al. (2010) also describe that in their study in India and the USA, 16 of the 19 Indian teachers, 17 of the 23 Indian parents, all teachers in the USA and seven of the eight parents in the USA reported their children to understand object permanence. Examples included students that showed that they knew where their workspace was and where their things were and that they would go to their space to look for familiar objects or by looking in the last place they played with an object, to find it. In the case-study of Damen et al. (2020), a deaf and blind five-year-old girl showed object permanence when she remembered the vibrating toy and the way she could switch it on again after a break, demonstrating object permanence by turning back to the toy and pushing the button with her tongue again. In Nelson et al. (2009), a boy aged 2 reached out and attempted to find the balloon they were playing with when it was no longer within his reach, also demonstrating object permanence.

Narayan et al. (2010) and Damen et al. (2020) gave examples of tertiary circular reactions. In Narayan et al. (2010) examples of children with deafblindness using a chair to climb to something they could not reach or climbing up a window with the support of bars in order to acquire something show how these children were experimenting with ways to get to a desired result. A girl aged 5 in Damen et al. (2020), after trying to make a toy vibrate again with her hands and face, discovered that she could do that by using her tongue. She then proceeded to use her tongue to make the toy vibrate. She repeatedly pushed the button with her tongue after the vibration had stopped, showing a clear understanding of means-end relations.

Finally, for the early representational thought substage, Dammeyer and Larsen (2016) found that 23–58% of children with CDB only mastered pre-intentional communication levels, unconventional pre-symbolic communication and conventional pre-symbolic communication. In Nelson et al. (2002, 2009), a girl aged 7 showed emerging conventional gestures, by telling the assessor she wanted to be lifted by holding out her arms.

3.2.2 Role of caregivers

Two theoretical contributions described the role of caregivers in supporting the development of skills within the sensorimotor stage. Bruce (2005a) explains in her theoretical contribution that children with CDB in the sensorimotor stage of secondary circular reactions need a demonstration by others to understand that they can influence their surroundings, and that they need different examples of cause-effect or tool-use behaviors. For the substage of coordination of reactions, Bruce (2005b) explains that children with congenital deafblindness need support to find, explore and engage with objects. They need environments that are consistent, so that they understand and remember the location of objects. Bruce (2005b) also states that children with deafblindness may not have the opportunity to explore an object in its entirety. They may identify key characteristics that are different from those noted by sighted children, especially when objects are large or dangerous, hindering full exploration. A predictable environment is essential not only to foster exploration, but also for object permanence, by showing the child that objects remain in the same place (Bruce, 2005a, 2005b). To support the early representational thought, Bruce (2005b) describes that children with CDB need more experiences than sighted and hearing children do to develop an understanding that one can use a representation of something to refer to an object, person or event. Bruce explains that representations are based on the child's individual perceptions of objects, and children with CDB base their representations more on their tactile experiences than sighted and hearing children do. A child with CDB might create individual gestures based on motor experiences with the environment. Therefore, motor reenactments could be seen as a form of early representations in children with CDB, just like onomatopoeic expression in hearing children (Bruce, 2005b).

3.3 The pre-operational stage

3.3.1 Descriptions of cognitive skills

Descriptions of symbolic understanding in children with CDB were found in Dammeyer and Larsen (2016). They found that only 1–14% of the participating children with congenital deafblindness in their study used the communicative level "concrete symbols" and 15–19% used the communicative level "abstract symbols." In Narayan et al. (2010), an example was given of a child using 20 word signs expressively on appropriate context. In a theoretical contribution, Bruce (2005a) describes that communication development of most children with CDB is severely delayed and that many of these children will not make the transition from pre-symbolic communication to symbolic communication. Hartmann (2012) also describes that children with deafblindness face challenges in their development of symbolic understanding. She states that they have limited access to the social interactions that facilitate symbolic understanding.

Examples of symbolic play in children with deafblindness were found amongst others in the study of Narayan and Bruce (2006), where one parent discussed how their child reenacted scenes from stories and films. In the study of Narayan et al. (2010) one teacher reported that a student "pretends to answer the phone." In the study of Pizzo and Bruce (2010) students with deafblindness were rated as being capable of performing "pretend self-play" at a higher rate than the students with multiple disabilities. For other categories of play, like pretend other-play or combinational symbolic play, no differences between groups was found. None of the students showed symbolic play involving dolls or play involving symbolic substitution of one item for another.

3.3.2 Role of caregivers

The role of caregivers in supporting symbolic understanding was described by Bruce (2005a) and Hartmann (2012). Bruce (2005a) explains the importance of offering emotionally charged, shared experiences, since gestures that emerge in these experiences are more likely to hold meaning for the child. Hartmann (2012) also states that to understand how symbols are used to refer to objects, people, concepts and environments children with CDB benefit from adults that provide them with meaningful opportunities to develop a basic understanding of the world around them. Hartmann emphasizes that symbolic understanding occurs within interactions that build in complexity and she too describes the importance of shared activities. Shared activities will help the child with CDB in creating concepts and experiences the child will recall later, therefore helping the child to refine symbolic understanding.

TABLE 5 Overview of assessment methods.

Assessment method	Literature	Cognitive skills measured in literature
Child Guided Strategies/The Van Dijk Method of Assessment	Nelson et al. (2002); Nelson et al. (2009); Damen et al. (2020).	Cause-effect, means-ends, object permanence, pre-symbolic understanding and symbolic understanding.
Rowland Communication Matrix	Dammeyer and Larsen (2016); Pizzo and Bruce (2010).	Pre-symbolic and symbolic understanding.
Tactile Working Memory Scale	Nicholas (2020)	Object manipulation, exploration and classification.
Self-developed questionnaires	Dammeyer and Larsen (2016); Narayan and Bruce (2006); Narayan et al. (2010); Pizzo and Bruce (2010).	Cause-effect, exploration, object permanence, pre-symbolic understanding, symbolic understanding, symbolic play and classification.

To support symbolic play, Bruce (2005b) describes that adults can use objects and gestures during pretend play to facilitate the acquisition of symbolic skills in children with CDB. Pizzo and Bruce (2010) add that children with CDB may need additional opportunities to engage in symbolic play, as developing symbolic relationships in play can be challenging.

3.4 The concrete operational stage

3.4.1 Description of cognitive skills

One study (i.e., Narayan and Bruce, 2006) described an example of children with CDB showing aspects of concrete operations. In their study, one teacher reported that the child "generalized knowledge about old toys to new toys," (Narayan and Bruce, 2006, p. 14). We interpreted this as that the child showed that he was aware of different properties of the old toys and of the fact that these properties may apply to other toys.

3.4.2 Role of caregivers

Bruce (2005a) states that children with CDB need support in identifying key physical features, functional uses and similar characteristics of objects. Extensive (joint) exploration of objects and the provision of multiple exemplars to identify similar key features in new objects helps the child's development of categorization. Bruce describes that the first encounter with an object will determine the perception of it, therefore repetitive experiences with objects help the child with CDB better understand the features of the object, thus supporting categorization. Furthermore, multiple sensory input allows the child to have more points of comparisons when considering if objects belong to the same category.

3.5 Assessment methods

In eight of the publications, at least one instrument to assess cognitive skills that correspond to skills from Piagetian theory was mentioned. Only one instrument, the Child Guided Strategies, mentioned Piaget in the theoretical framework on which it was based. None of the other described instruments were based on Piagetian theory. Table 5 gives an overview of the different assessment methods and the cognitive skills that were assessed in the included publications.

In four publications, cognitive skills were measured by means of questionnaires for parents, teachers and professionals. In Dammeyer and Larsen (2016), professionals filled in questionnaires about the included children to provide information about the communication modes of the children, including pre-verbal and symbolic understanding. In Narayan and Bruce (2006) and Narayan et al. (2010), parents and teachers of children with CDB filled in a questionnaire on cognitive functioning including exploration, cause-effect, means-end relations, object permanence and classification. In Pizzo and Bruce (2010), the Play Assessment Questionnaire was used with parents and teachers to assess several forms of object manipulation and forms of symbolic play such as pretend self-play, pretend other-play to other people, pretend other-play to dolls and substitutional play.

In three publications, the Child Guided Strategies (CGS) was used to assess the development of children with CDB (Nelson et al., 2002; Nelson et al., 2009; Damen, 2020). The CGS is a dynamic assessment specifically developed for children with severe multiple disabilities such as CDB. It aims to assess the underlying processes of learning by observing eight areas: biobehavioral state, orienting response, learning channels, approach-withdrawal, memory, interactions, communication and problem solving. Piagetian skills assessed with this method in the three publications were: cause-effect, means-ends relations, object permanence, pre-symbolic understanding and symbolic understanding.

Dammeyer and Larsen (2016) and Pizzo and Bruce (2010) used the Rowland Communication Matrix (Rowland, 2009) to assess symbolic understanding. The Rowland Communication Matrix is an instrument designed to evaluate the communicative functions and forms of expressive communication skills in children with severe and multiple disabilities.

One publication described The Tactile Working Memory Scale (TWMS), another dynamic assessment (Nicholas, 2020), was specifically developed for persons with CDB to assess tactile working memory and includes items that assess object manipulation, exploration and classification.

4 Discussion

Using Piaget's theory of cognitive development, we aimed to provide a comprehensive overview of literature describing Piagetian cognitive skills and the assessment thereof in children and youths with congenital deafblindness. With the first research question, the authors wanted to investigate how Piagetian cognitive skills are described in literature about children and youths with CDB. For the sensorimotor stage, descriptions mostly involved children performing physical actions to achieve specific results, by engaging with their environment or influencing the behaviors of adults around them, or descriptions of children exploring and manipulating objects in their environment, showing cause-effect understanding, means-end knowledge and object permanence. Some descriptions of children with CDB mastering pre-symbolic communication were found.

In some cases of which the age was given, behaviors within the sensorimotor stage that can be expected at that age were found present. For example, the boy of 14–25 months of age in Murdoch (1994) showed behaviors corresponding with the secondary circular reactions stage, usually developing between 4 and 8 months. Furthermore, he showed behaviors corresponding with the coordination of reactions stage, usually developing between 8 and 12 months. Similarly, the three-year-old girl in Vervloed et al. (2006), the seven-year-old girl in Nelson et al. (2002) the girl (age 7) and boy (age 2) in Nelson et al. (2009) and the five-year-old girl in Damen (2020) showed behaviors ranging from the primary circular reactions to the tertiary circular reactions stages that children that age are expected to master (e.g., object permanence, means-ends, cause-effect). At the same time, these descriptions do not give information about when the children acquired the skills. Therefore, no statements can be made on whether delays are present. Based on the descriptions found, only some inferences can be made. For example, the seven-year-old girl in Nelson et al. (2002, 2009) showed only emerging conventional gestures, thus showing a delay since one might expect a seven-year-old girl to master skills beyond the sensorimotor stage. Furthermore, about the boy of 14–25 months from Murdoch (1994), no descriptions were given beyond the coordination of reactions stage. Since behaviors within last two sub-stages of the sensorimotor stage usually develop until 24 months of age, one might wonder if the child described by Murdoch (1994) is thus showing delays in those stages. The boy who participated in the study by Vervloed et al. (2006), 3 years and 4 months of age, was described as functioning only at the second sensorimotor stage. Even though our analyses showed that he performed actions corresponded with the fourth sensorimotor stage, still a developmental delay can be inferred. Unfortunately, since describing Piagetian skills was not the goal of any of the reviewed publications, it could also be that skills were still present but not described. Furthermore, none of the publications provided information about age of onset of the skills, making it impossible to make a statement about the developmental trajectories of those skills. This finding highlights the lack of knowledge that exists within the field of deafblindness when it comes to cognitive development of children with CDB and underlines the great need for longitudinal research within this group. Yet, the descriptions we found in the literature, show that Piagetian skills at the sensorimotor stage are observable in children with CDB, which is relevant for both assessment and intervention purposes.

For the pre-operational stage, it was found that symbolic communication is severely delayed in most children with CDB and that most of them seemingly do not transition to the symbolic phase of communication. Only less than a fifth of 71 children aged 3–18 from Dammeyer and Larsen (2016) mastered abstract symbols, inferring a delay in symbolic communication in most children with CDB. Regarding symbolic play, examples of reenactments and pretend self-play were described. No examples were found of symbolic play involving dolls or other items. None of the descriptions within the pre-operational stage could be related to specific ages of the children, since only group descriptions and age range of the group was given.

For the concrete operational stage, one example of aspects of concrete operations regarding the generalization of knowledge was mentioned, but no specific skill from this stage was clearly described. Besides, no descriptions were found of cognitive skills at the formal operational stage.

Concerning the second research question focusing on the role of caregivers in supporting cognitive development of children with CDB, it was found that children with CDB need consistence in their environments and support in finding and exploring objects, and that they need more support in experiencing their environment than sighted and hearing children do, to help them develop skills in the sensorimotor and pre-operational stage. The importance of enabling shared experiences was also highlighted. Within the concrete operational stage, it was mostly found that children with CDB need support in identifying, exploring and comparing objects to be able to experience the different characteristics of object, before being able to categorize, decenter or understand reversibility, conservation and transitivity. No descriptions were found of support for the development of the formal operational stage, but it could be inferred from the described support at previous stages that additional support would be needed in all stages.

With the third research question, the authors wanted to investigate if and how Piagetian cognitive skills are assessed in literature about children and youths with CDB. Furthermore, we wanted to investigate if the instruments described in the literature were based on Piagetian theory. One instrument was described that used Piagetian principles as a theoretical framework, concomitantly with principles from other developmental theories such as Vygotsky, namely the Child Guided Strategies (Nelson et al., 2002, 2009). This dynamic assessment instrument seems to be appropriate to observe several Piagetian cognitive skills, since one of the core principles of the CGS is that learning processes are based on sensorimotor experiences. Dynamic assessments such as the Child Guided Strategies (CGS) and the Tactile Working memory Scale (TWMS)(Nicholas et al., 2019) are specifically developed for this target group and were used in multiple publications. These methods specifically focus on measuring the learning potential of children with CDB and the use of mediating strategies of the assessor is pivotal during the assessment procedure.

Questionnaires also seem to be appropriate instruments to gather data on Piagetian cognitive skills of children with CDB, as in several publications parents, teachers or professionals provided specific information about such skills in children with CDB. Finally, the Rowland Communication Matrix can be used to measure symbolic understanding.

Even though the scare amount of literature on cognitive development in combination with insufficient information about the children in the literature impeded a thorough analysis of data on cognitive development, some aspects within the results have to be mentioned. For example, engaging in independent acts with objects seems to be occurring rarely in the descriptions found. If we look at our findings within the secondary circular reactions stage, it is noticeable that only two examples were found of children manipulating objects to recreate a pleasurable event. All other examples in that stage describe children with CDB trying to influence the adult's behavior. Similarly, looking at symbolic play, some examples of children with CDB were found performing reenactments or pretend self-play, but the authors specifically mentioned that no examples of pretend play with objects was found. Yet, when looking at the descriptions of caregiver support, no descriptions were found of caregivers supporting independent acts with objects, whereas mostly descriptions of caregivers supporting joint exploration were found. This finding is in line with the literature review of Rorije et al. (2023), which revealed that caregivers prefer supporting autonomy of children with CDB by

remaining present and nearby, instead of supporting the children's will to act independently. Furthermore, this finding could be seen as a reflection of transactional models of development, which emphasize the bidirectional, recursive nature of relationships already introduced by Bell (1968): the environment has an influence on the children, but children also have an influence on their environment (Sameroff, 2010). Possibly, children with CDB engage less in objects because caregivers support them in other ways, while at the same time caregivers do not support them in engaging with objects because they see that the children do not initiate acts with objects.

Most publications described cognitive skills in the sensorimotor stage ($n = 10$) or pre-operational stage ($n = 8$). Only two publications described cognitive skills within the concrete operational stage and no publications were found describing cognitive skills of the formal operational stage. The scarce quantity of publications found describing skills at the concrete and formal operational stages could indicate that children with CDB rarely achieve these stages. Different causes for this can be inferred. Even though we found that most children showed the mastery of cognitive skills within the sensorimotor stage, it became clear that they experienced substantial delays and needed additional support as early as in this first developmental stage. Earlier research supports our findings that children with CDB lack experience and practice with their environments (Damen et al., 2020) and that the experiences that they do have are usually not only limited, but also different than those of sighted and hearing children. Furthermore, children with CDB experience difficulties in the stage of developing symbolic understanding. Other research adds to this finding, by stating that children with CDB have fewer possibilities to understand the functions of eye contact, facial expressions or speech (Damen et al., 2015). Consequently, children with CDB not only have less experiences on which to build on during the sensorimotor stage, but are also hindered in their learning from the opportunities that adults can provide them.

On the other hand, the authors ponder whether the scarce quantity of articles could also relate to a lack of suitable ways to assess cognitive skills in children with CDB. There is an ongoing debate on how much a lack of proper adaptations of assessment instruments contributes to the frequently reported severe developmental delays in people with congenital deafblindness (Dalby et al., 2009, in Ravenscroft and Damen, 2019). The question arises whether it is indeed these children who do not progress to higher stages of development, or whether it is a lack of accurate ways to map and observe cognitive skills in this target group. Our results suggest that meaningful observations can be made to some extent, for example, within the sensorimotor stage. Yet in other stages, for example when looking at forms of symbolic play, one might question if we are able to interpret symbolic play as such. Possibly, a child with CDB does show symbolic play with objects but the behavior could easily be interpreted as sensorimotor exploration, since a child with CDB is in both cases more reliant on the sense of touch. As can be seen in our results, dynamic assessments and combinations of formal and informal assessment are used within the field of CDB to account for the complexities of assessing cognition in children and youths with CDB. Specific instruments such as the Callier-Azusa Scale, which was specifically developed for children with CDB, are only mentioned as being used in combination with other tools to gather information about the child with CDB (Narayan and Bruce, 2006; Narayan et al., 2010). Other instruments that are used to assess cognitive development in typical children, for example the Uzgiris-Hunt ordinal Scales of Psychological Development (Uzgiris and Hunt, 1975), which is based on Piaget's theory, were not mentioned at all, possibly because of the inapplicability of these instruments for the assessment of children with congenital deafblindness. Nelson et al. (2002) emphasize that such instruments fail to take into account the impact of sensory disabilities on all areas of development. Furthermore, they fail to take into account the fact that children with sensory impairments are disadvantaged by unfamiliar settings and may not understand what they are asked to do.

Present scoping review provided a first look into what is known about Piagetian cognitive skills in children with CDB. Based on our findings, the authors now want to propose a dual shift forward, to take the next step in research on this population. Firstly, it would be interesting to research how children with CDB develop cognition by looking at models of cognitive development that integrate concepts from Piaget's Standard Theory with newer concepts. Piaget himself, for example, shifted his view on development to a more continuous course of development in his New Theory. Other neo-Piagetians further built on his theories and introduced concepts such as attentional capacity and information-processing theory to explain both the development from one Piagetian stage to the next and individual differences in developmental rate (Morra et al., 2008; Pascual-Leone, 1987). Others proposed that executive control structures are the building blocks of the developmental stages, arguing that variations in processing efficiency may explain differences in the rate at which individuals progress through cognitive stages (Case et al., 1996; Mascolo, 2015). Furthermore, researchers emphasized the contextual and social factors in these processes much more than Piaget did (Fischer and Bidell, 2006; Morra et al., 2008) or discerned core capacities that include processing speed, control of processing, and storage and systems of higher order such as metacognitive structures that govern self-understanding and self-monitoring (Demetriou, 2022; Mascolo, 2015).

In general, rather than primarily focusing on describing cognitive development in terms of the acquisition of skills like Piaget, a common thread between neo-Piagetian theories can be found in their interest in higher-order functions of cognition. With their theories on the capacity of retaining and processing units of information and on executive structures, neo-Piagetian research has been contributing in a relevant way to the discussion on higher-order cognitive functions such as working memory, inhibition and executive control, also known as executive functions (Morra et al., 2008). The authors of present scoping review, aiming to provide a primer on an understudied topic, decided to focus on clearly defined, though more static, cognitive skills, and therefore did not include neither the processes of acquisition of skills nor later developmental theories. Nevertheless, we are aware of the importance of these processes and the contribution of later developmental theories, and a suggestion for research is therefore to include these aspects in future studies, gradually working towards a more comprehensive overview.

Secondly, since children with CDB are even more reliant on social interactions to learn about their environments than sighted and hearing children, they may have a greater need for processes of assisted learning such as scaffolding or mediated learning. Therefore, the authors propose a shift toward the more dynamic look on cognition proposed by Vygotsky. The need for caregiver support in children with CDB is in line with his social-constructivist theory,

which states that a child's cognitive development is inseparable from the socio-cultural structures that surround the child (Vygotsky, 1978). While Piaget focused on the interaction of infants with the physical world, Vygotsky studied children's development at a preschool age and beyond, within social interactions. Both scaffolding (Wood et al., 1976) and mediating learning (Feuerstein et al., 1986) build upon Piaget and Vygotsky, and focus on the idea that an individual's skills are related to the amount of and quality of tutoring strategies received from caregivers.

Within interventions focused on improving interaction and communication abilities, this shift has already been made. The importance of caregivers has been addressed by researchers when looking at interactions and intersubjective communication. Janssen et al. (2006) revealed that caregivers can be effectively supported in improving the quality of interactions with children with CDB and Damen et al. (2014) also found that an intervention for caregivers had a positive effect on intersubjective communication. These interventions are based on the more social aspect of learning of Vygotsky's (1978) and theories of Trevarthen and Aitken's (2001), and focus on improving interpersonal communication such as turn-taking, imitation and other attunement strategies. Within the dynamic assessments that are used within the field of CDB, this shift also seems to have been made, as can be seen by the CGS and the TWMS that are specifically developed for this target group and are based on several developmental theories combined. Focusing on the dynamic aspect of cognitive development of children with CDB seems therefore pivotal to do justice to the learning potential of these children. Yet, there are no interventions specifically developed for this aim, and the majority of interventions for children with CDB focus on social interaction and communication (Sundqvist et al., 2022). Developing an intervention focused on mediating strategies to support cognitive development of children with CDB seems thus necessary. In light of the aforementioned, it can be concluded that the shift to a more dynamic view on cognitive development still has to be made when it comes to interventions targeting cognitive development. Since children with CDB are so reliant on their caregivers, the cognitive development of children with CDB could benefit from an intervention specifically targeting caregivers, where the use of mediating strategies should play a central role. As Manford et al. (2024) also concluded in their scoping review, children with CDB need individualized support from professionals that understand deafblindness to ensure that children with CDB reach their full potential. Piagetian cognitive skills could form a starting point for measuring several cognitive milestones of children with CDB. Furthermore, socio-cognitive perspectives of development, such as Nelson (1996) or Tomasello (1999), who take into account the child and their social experiences, could provide relevant insights for the development of an intervention aimed at the complex interplay between child-characteristics and caregiver support in the cognitive development of children with CDB.

4.1 Limitations and recommendations for future research and practice

In present scoping review, we chose to use Piaget's "Standard Theory" as a lens to analyze the found literature. We acknowledge that choosing one theory when analyzing the data may restrict the bandwidth of the analyses. Furthermore, we acknowledge that Piaget himself at a later stage moved in a different direction with his "New Theory." In the latter, he saw development more as a continuous process instead of clearly sequentially distinct. At the same time, even though Piaget's theory changed through time, it still has maintained continuity in most of its core assumptions (Beilin, 1992b). In present scoping review, the choice for analyzing the data through one specific theory allowed for a more in depth approach. Since the scope of present review was to provide only a first step in analyzing the available data on cognitive development of children with CDB, Piaget's Standard Theory provided us with a definite set of skills that could be extracted from the literature. Naturally, some limitations accompany this choice. Firstly, it must be noted that even in Piagetian theory, not all skills are clearly sequentially distinct, and many skills develop alongside each other and are interdependent. Thus, one limitation of the study is that choices had to be made when placing skills within a specific stage or substage. Exploration and imitation for example were placed in the coordination of reactions stage, but some forms of exploration and imitation do develop earlier. Similarly, the finding of children "generalizing knowledge" was placed within the concrete operational stage as classification, but one could argue that some information gets lost with this choice, since processes underlying classification skills are more complex and span across multiple stages of development. A recommendation for future research is to take into account the processes that underly the acquisition of skills, and the impact of sensory impairments on these processes to create a more comprehensive overview. Piaget's "New Theory" or other developments based on his theory such as Neo-Piagetian theories could provide other perspectives to describe these processes. A systematic review could then be recommended.

A second limitation can be found in the number of included publications. Only nine empirical studies were found, of which none were longitudinal studies. Therefore, no descriptions could be given about the course of development of the cognitive skills of the participants. Apparently, data on cognitive skills in children with CDB is even more scarce than we anticipated, and longitudinal data on how cognitive skills develop in children with CDB is lacking. Present scoping review provided a first look on how cognitive skills are described in the literature. A recommendation for research would be to gather more longitudinal data on how cognition of children with CDB develops by carrying out longitudinal studies or looking into retrospective data from case descriptions in the past. Possibly, an archival research could yield relevant information, since research on cognition has widely been documented in books and case descriptions that are not always accessible within modern databases.

A third limitation of this study is that only in five publications the ages of each participating child were specified. All other publications either did not have any participants, the participants were parents and teachers, or only gave a mean age and characteristics of the group. Therefore, the descriptions of the cognitive skills could be matched to the age of the children only in the five publications describing case studies. Furthermore, no conclusions could be drawn about the ages at which children with CDB achieve certain milestones, since no descriptions were found about age of onset of the observed behaviors. Therefore, no information could be given on developmental trajectories of the children. The lack of information on the participants within the already scarce amount of publications, in combination with the absence of information on the age of onset of skills in children with CDB, impeded us to make a critical analysis of the specific

cognitive delays of individuals with CDB. This lack of information emphasizes the need for longitudinal research on this population the gain more insights on the developmental trajectories of these children and the impact of child characteristics on these trajectories.

The lack of clear information on the participants is further emphasized by the lack of a clear definition of CDB in literature and practice. In present research, only articles describing "congenital deafblindness" or describing the onset of deafblindness as "from birth" or "before language acquisition" were taken into account. Yet, a specific age of onset when speaking of "onset before language acquisition" is not always given. Furthermore, different publications use different definitions of CDB. In Dammeyer and Larsen (2016) for example, the Nordic Definition was used, but in Narayan et al. (2010), the authors explain that the diagnosis as given by the parents was accepted for the study, as this is presumed to be based on medical records. Thus, even though all articles focus on congenital deafblindness, some unclarity still exists. This further highlights that the field of CDB is a relatively young one and that fundamental aspects are still being researched.

A fourth limitation lies in the screening process. Two researchers analyzed the articles for eligibility to calculate Cohen's Kappa in the first round of screening. Cohen's Kappa indicated a substantial agreement—thus not a perfect agreement. The authors see this limitations as a reflection of the challenge of researching cognition, being complex mental processes that are sometimes hard to grasp objectively. Even though the percentage of agreement was high, inferring that overall the two researchers did agree on the inclusions and exclusions, in some cases they made different deliberation about which cognitive skills should be included in the first round of selection, which was purposefully kept broad since we expected scarce data on this topic. Due to the broad search, more articles containing unclear titles and descriptions were included for full text analysis. A consequence was that in the second screening phase, the Piagetian lens could only be applied later in the screening process, since articles had firstly to be screened based on more general aspect such as the relevance of the topic. For example, an article mentioning cognitive development in a school context was firstly included, since cognition was mentioned, but was later excluded since the topic eventually was not relevant for our review. For future reviews, it could therefore be recommended to start with a more focused approach.

Furthermore, since none of the included literature specifically focused on Piagetian theory, all descriptions of behaviors were extracted by the authors from articles that had other aims than looking at Piagetian theory. To select and describe the Piagetian skills in children with CDB the researchers had to deduce the cognitive skills from the observable behavior described in the literature. Therefore, some interpretation bias is conceivable, even though efforts were made to minimize this by adhering to a clear description of the cognitive skills made beforehand and by discussing the included results with the second author until consensus was reached. On top of that, we were reliant on second-hand data both during title and abstract screening and during data extraction based on full-text. For example, Nelson et al. (2009) describe that a girl put her hands together and moved her hands up and down to indicate that she wanted the movement of the tie to continue. This is an interpretation that the authors of the original article have made. We did not perform the observation ourselves. Thus, we had to rely on the descriptions of original authors. We tried to address this problem by comparing the descriptions found with the descriptions of observations by Piaget himself.

To conclude, the authors want to end with some recommendations for practice. Delays of different cognitive milestones of children with CDB and the reliance on their caregivers became apparent in present review. To support cognitive development of children with CDB, the authors suggest the development of an intervention specifically focusing on the support of caregivers in stimulating the development of cognitive milestones of their children with CDB. Intervention should deliberately focus on coaching caregivers in applying those strategies that stimulate cognitive skills in children with CDB. The Tactile Working Memory Scale (Nicholas et al., 2019), for example, does provide suggestions about which caregiver strategies can be used to support tactile working memory, based on the outcomes of the assessment. Yet, caregivers are not trained in applying those strategies. To our knowledge, no interventions have been previously developed that focus on improving both the cognitive skills of children with CDB and the strategies that caregivers can use to do so. Furthermore, in assessing cognition, we recommend the use of multi-method, multi-informant assessments, in line with Nicholas (2020), and specifically we recommend the use of dynamic assessment to ensure that during assessment and intervention individual strengths and potentials are taken into account, in line with Boers et al. (2013).

Author contributions

KT: Conceptualization, Data curation, Formal analysis, Writing – original draft, Writing – review & editing. SD: Conceptualization, Formal analysis, Supervision, Writing – review & editing. MJ: Conceptualization, Supervision, Writing – review & editing. AM: Conceptualization, Supervision, Writing – review & editing.

Funding

The author(s) declare that financial support was received for the research and/or publication of this article. This work was supported by Fonds Instituut voor Doven (Sint-Michielsgestel, The Netherlands) and H.D. Guyotfonds (Haren/Groningen, The Netherlands).

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 author(s) declared that they were an editorial board member of Frontiers, at the time of submission. This had no impact on the peer review process and the final decision.

Publisher's note

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