

Emotional Dysregulation in Children with Deafblindness: A Systematic Literature Review

Master Thesis MSc Pedagogical Sciences/Deafblindness

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Abstract

Children with congenital deafblindness (CDB) encounter significant challenges in their emotional development due to a combination of sensory impairments, limited access to social-emotional cues, and communication barriers. Emotional dysregulation often occurs in this demographic and presents a complex issue that continues to need both basic and applied research. This systematic literature review aimed to identify and synthesize theoretical frameworks, critical insights, and intervention approaches related to emotional dysregulation in children under age 15 with CDB. Following PRISMA guidelines, a structured search was conducted, yielding 16 eligible studies that underwent thematic analysis to uncover patterns in conceptual models, behavioural observations, and intervention strategies.

The findings reveal that emotional dysregulation is commonly linked to arousal instability, difficulties with executive functioning, and sensory processing challenges. The most frequently referenced frameworks include the four-dimensional model of selfregulation, which incorporates cognition, behaviour, emotion, and physiology, as well as the low-arousal approach that guides adult responses to stress and behavioural escalation. Interventions that emphasise co-regulation, environmental predictability, and calming sensory input demonstrate promising effects, although empirical evidence is still limited. This review highlights the necessity for personalised, context-sensitive support and enhanced collaboration between caregivers and professionals. Future research should focus on developing standardised assessment tools and investigating the long-term effectiveness of interventions for children with CDB.

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Table of contents

<u>Abstract</u>	2
Acknowledgements	3
Table of Contents	4
Chapter 1: Introduction	6
1.1 Background and problem definition	6
1.2 Definitions and delimitations	6
1.3 Systematic interventions	9
1.4 Problem Statement	9
1.5 Research Questions	10
1.6 Objectives	10
Chapter 2: Theoretical Framework	11
2.1 Introduction to Emotional Dysregulation in Children	11
2.1.1 Neurobiology and Arousal Regulation	12
2.3 Arousal, Self-Harm and Sensory Integration as Explanatory Models	14
2.4 The Development of Self-Regulation, Emotion Regulation, and Commun	lication in
Children with Congenital Deafblindness	14
Chapter 3: Method	17
3.1 Document Review as a Method	17
3.2 Search Strategy	17
3.3 Study Selection	20
3.4 Data Extraction and Quality Assessment	21
3.5 Analysis Plan	22
Chapter 4: Results	29
4.1 Theories and Models of Emotional Dysregulation	29
4.2 Expressions of Emotional Dysregulation	

4.3 Review interventions and their limitations	
Chapter 5: Discussion	36
5.1 Understanding Emotional Dysregulation in Children with Deafblindness: K	ey
Insights from the Literature	36
5.2 Implications for Practice and Intervention	
5.3 Limitations of the Study	41
5.4 Recommendations for Future Research	42
Chapter 6: Conclusion	44
6.1 Summary of Findings	44
6.2 Intervention Strategies and Limitations	44
6.3 Implications.	44
6.4 Future Directions	45
References	46

Chapter 1: Introduction

1.1 Background and problem definition

I began working with children with diverse abilities in kindergarten and started consulting in deafblindness in 2020. My focus is on self- and emotional regulation, driven by my interest in how children manage strong emotions. I have observed varying staff responses to children's emotional expressions, particularly with deafblindness. This study examines emotional dysregulation in children under 15 with congenital deafblindness, aiming to understand how caregivers can support healthy emotional development.

Emotional development is a central part of a child's well-being, learning and participation. For children with congenital deafblindness (CDB), emotional regulation is often compromised due to limited access to visual and auditory cues, restricted opportunities for incidental learning, and challenges with social reciprocity. These factors may lead to frequent emotional dysregulation. However, their expression of emotional dysregulation is often misinterpreted as behavioural problems rather than regulatory difficulties

1.2 Definitions and delimitations

This section defines and clarifies key concepts relevant to the study, establishing the scope and focus of the review. Specifically, it discusses the terms deafblindness, congenital deafblindness, and emotion regulation.

The review aims to ensure conceptual consistency and delineate the boundaries of the research by clearly defining these concepts. The primary focus is on emotional dysregulation in children under age15 with congenital deafblindness.

Deafblindness is a complex and heterogeneous condition described by a combined vision and hearing impairment that significantly restricts an individual's ability to access information, communicate, and orient in the environment. The literature employs both medical and functional definitions of deafblindness (Ask Larsen & Damen, 2014). Deafblindness can be defined based on medical criteria, including clinical diagnoses of severe hearing and vision impairments, often without explicit reference to its functional impact. However, functionally deafblindness is understood as a combined sensory loss of such severity that the two senses cannot compensate for each other, leading to significant challenges in everyday life.

The Nordic definition, widely used in Norway and other Nordic countries, defines deafblindness as:

"Deafblindness is a combined vision and hearing impairment of such severity that it is hard for the impaired senses to compensate for each other. Thus, deafblindness is a distinct disability (Soelgaard et al., 2024)."

It is crucial to understand that deafblindness does not always mean a total loss of both vision and hearing. Rather, it encompasses a range of sensory impairments that can vary from mild to severe (Dammeyer, 2010). The extent and combination of sensory loss differ greatly among individuals, impacting their communication, access to information, mobility, and social interactions.

Functional deafblindness highlights the combined impact of dual sensory loss on an individual's ability to interact with their surroundings, even when some residual vision or hearing remains. This dual loss creates unique challenges that cannot be fully understood by examining each impairment separately.

Congenital deafblindness refers to combined vision and hearing impairments present from birth or early infancy, before language development. Children with congenital deafblindness often experience profound restrictions in their communication development, access to environmental information, and opportunities for social interaction. These limitations can have a profound impact on cognitive, emotional, and social development (Janssen et al., 2002).

Emotion regulation is a crucial skill that allows individuals to respond and react in ways that are socially acceptable (Gross, 2014). This skill encompasses the ability to modify emotional expressions based on situational needs, foster positive emotions, manage negative emotions, and control how long emotional arousal lasts (Gross, 1998; Lamm & Lewis, 2010).

The ability to regulate emotions develops through a complex interplay between a child's experiences, relationships, and biological and genetic predispositions. Effective emotion regulation typically relies on internal strategies, such as self-awareness, and external supports, including co-regulation from caregivers (Ramirez et al., 2017b).

Emotional dysregulation refers to challenges in appropriately managing emotional responses to different contexts (Gross, 1998). It may present as excessive emotional reactions, difficulty in calming down, or struggling to express feelings constructively.

Children experiencing emotional dysregulation often feel overwhelmed by intense emotions and may engage in behaviours such as self-injury as a coping mechanism. Although these actions might seem maladaptive, they frequently serve a regulatory function for the child (Hejlskov Elvén, 2012).

In children with congenital deafblindness (CDB), several factors may increase the risk of emotional dysregulation. Underlying causes include dual sensory loss, difficulties with executive functioning, and limited opportunities for social interaction and incidental learning (Lamm & Lewis, 2010; von Plessen & Kabicheva, 2010). Due to reduced access to emotional feedback and fewer opportunities for co-regulation, the development of self-regulation skills in children with CDB is particularly challenging. Supporting emotion regulation in this population requires tailored interventions, sensitive caregiver support, and structured opportunities for emotional practice with adults and peers (Hartshorne & Nicholas, 2017).

Chapter 2 provides a more detailed discussion of the manifestations of emotional dysregulation in deafblindness, along with relevant theoretical and neurobiological explanations.

1.3 Systematic interventions

In Scandinavia, two theoretically grounded and developmentally informed approaches are used to support self-regulation in people with congenital deafblindness. These approaches aim to foster emotional development in a structured and proactive manner, rather than relying on reactive or situational strategies.

The first approach is based on the model that is operationalised in the *Assessment Scale for Self-regulation Functions in People with Congenital Deafblindness* (Nicholas et al., 2015). Although not explicitly developed for children, the scale is based on theoretical literature with strong relevance to child development. It outlines how self-regulation evolves from early co-regulation and external support toward independent regulation over time, while also recognising that external regulation may remain important in times of crisis or stress, even in adulthood. Despite its relevance, the scale is not widely used in practice. In my professional work, several practitioners have expressed interest in applying it to children and individuals with deafblindness of various aetiologies, highlighting a broader and recognised need for flexible tools within the deafblindness field.

The second is the *SOS model* – *Side-by-Side on Self-Harm* (Jensen & Selling, 2023), which addresses arousal regulation and self-injurious behaviour in the deafblind population. Akin to Nicholas et al. (2015), the SOS model is not a child-specific model. It draws from literature on intellectual disabilities and the low-arousal approach. It frames self-harm and related behaviours as regulatory attempts in response to overwhelming emotional states.

The low-arousal approach (Hejlskov Elvén, 2011; Hejlskov Elvén & Sjölund, 2018) emphasises emotionally neutral, non-confrontational caregiving in response to escalating behaviour. It recognises that individuals with developmental disabilities are sensitive to emotional contagion from caregivers. As such, the caregiver's ability to remain calm and reduce stimulation is key to preventing further dysregulation.

1.4 Problem statement

Caregivers of children with congenital deafblindness often struggle to understand and support their child's emotional regulation difficulties. At the same time, educators lack appropriate tools to assess emotional dysregulation and are left without effective strategies to guide intervention and support. Despite the complexity of the issue, there is currently no comprehensive overview of what is known about emotional dysregulation in children with

CDB, and few systematic interventions have been empirically evaluated. This gap in the literature makes it difficult for professionals to work proactively and consistently with emotional development in this group. This thesis aims to conduct a systematic literature review to identify theoretical models, key research findings, and existing interventions related to emotional dysregulation in this population.

A structured review clarifies existing knowledge and identifies areas for further research and intervention development.

1.5 Research questions

Main Question:

What are the main theories, models, and findings in the literature concerning emotional dysregulation in children (under the age of 15) with congenital deafblindness? Sub-Questions:

- 1. Which theories and models of emotional dysregulation are used and discussed in studies focusing on children (under 15) with congenital deafblindness?
- 2. Which interventions addressing emotional dysregulation in children with congenital deafblindness are described in the existing literature, and what are their reported limitations?
- 3. What is known about the effectiveness of interventions in supporting emotional regulation in children with congenital deafblindness?

1.6 Objectives

The primary objective of this study is to conduct a systematic literature review to identify theoretical models, key findings, and effective intervention strategies for emotional dysregulation in children with CDB.

Chapter 2: Theoretical framework

This chapter discusses the theoretical perspectives important to understanding emotional dysregulation in children with congenital deafblindness (CDB). The aim is to provide a framework exploring how sensory, cognitive, and neurobiological factors influence emotion regulation in CDB. I also examined one main model developed to support and engage children with CDB.

2.1 Introduction to emotional dysregulation in children

Self-regulation develops through dynamic interaction between biology, genetics, and early relationships with adults (Nicholas et al., 2015). Typical emotional regulation develops through early interactions with caregivers, where infants learn to calm themselves as caregivers respond sensitively to their distress (Janssen et al., 2002; Nordanger & Braarud, 2017).

When children receive assistance from others to regulate their emotions, this process is referred to as *co-regulation*. While internal regulation processes focus on how a child independently manages emotions, co-regulation is a critical foundation for developing selfregulatory skills. These skills emerge through consistent interactions with caregivers, who, by being sensitive to the child's needs, emotional states, and behaviours, can support, intervene, and help re-establish balance when the child becomes dysregulated (Nordanger & Braarud, 2014).

A prerequisite for healthy self-regulation is that caregivers can perceive affective information and establish a secure attachment. The child gradually learns to regulate their emotions through being mirrored by the caregiver. To develop, we depend on interactions with others from birth. At birth, only about 25% of the brain is fully developed, and through these early relationships, the brain continues to grow and mature (Christiansen & Jensen, 2020).

Stern (1998) defines *intersubjectivity* as early affective attunement between caregiver and infant. Intersubjectivity represents the earliest interaction and relational exchange between the child and caregivers. For emotional and cognitive development, the child must have a secure base to explore, established through these early relational experiences.

Individuals with deafblindness must perceive themselves as persons whose expressions and communications are valued and worth attending to by others (Nafstad &

Rødbroe, 2013). Due to sensory barriers, achieving these early experiences of mutual regulation is more complicated in congenital deafblindness. Children with limited access to visual and auditory information may miss incidental cues such as facial expressions, tone of voice, or eye contact, which can delay bonding and the development of trust (Hartshorne & Schmittel, 2016).

2.1.1 Neurobiology and arousal regulation

Neurobiological models explain how the nervous system of a child with deafblindness can be in a constant state of dysregulation. Since their world is often unpredictable and confusing due to limited sensory input, their autonomic nervous system, especially the sympathetic branch that triggers the "fight or flight" response, may be consistently overactive. Meanwhile, the calming influence of the parasympathetic system might be less engaged. Over time, this imbalance can impact physiological regulation and emotional and behavioural control (Christiansen & Jensen, 2020)

Children's developing nervous systems make them particularly vulnerable to stress, even when not deafblind (Nordanger & Braarud, 2017, p. 66). Prolonged stress without effective soothing can alter the brain and increase the risk of somatic illness (Nordanger & Braarud, 2017, p. 89). Arousal regulation is, therefore, fundamental to emotional regulation, requiring a balance between overstimulation and understimulation (Hart, 2016).

2.2 Theoretical model of regulatory dimensions

Hartshorne and Nicholas's (2017) model in Figure 1 describes how literature understands and interconnects various regulatory dimensions.



Figure 1. Self-regulation model adopted from Hartshorne and Nicholas (2017, p. 6)

This model illustrates how self-regulation coordinates activities across four interrelated systems: cognitive, behavioural, emotional, and physiological regulation (Hartshorne & Nicholas, 2017). Each system is deeply interconnected, meaning that challenges in one area will likely affect the others. For instance, difficulties in emotional regulation may also impair behavioural control and cognitive flexibility.

The model adopts a developmental and neurobiological perspective, emphasising that self-regulation emerges through dynamic interactions between internal processes and external environmental demands. In congenital deafblindness, impairments across sensory modalities can significantly affect the maturation of all regulatory domains, underscoring the importance of early, supportive interventions.

While Hartshorne and Nicholas' model provides a comprehensive overview, it is also important to consider specific mechanisms that can impact emotional regulation, such as arousal regulation and sensory integration.

2.3 Arousal, self-harm and sensory integration as explanatory models

Arousal regulation and sensory integration are fundamental to understanding the emotional regulation challenges faced by individuals with deafblindness. Sensory integration refers to the process through which the brain organises and interprets sensory information from the body and the environment. Effective sensory integration supports adaptive responses to stimuli. When this process functions well, sensory input such as touch, sound, movement, and visual information is coordinated efficiently to support regulation, learning, and interaction.

Difficulties with sensory integration can result in challenges with emotional regulation, attention, motor coordination, and behaviour (Lindstrøm, 2022).

Children with deafblindness may experience atypical sensory integration, such as hypersensitivity, hyposensitivity, or difficulties processing and combining sensory inputs, further complicating their ability to modulate emotional states and behavioural responses. Sensory integration occurs in the brainstem and via the spinal cord, while sensory processing occurs in the brain's cerebral cortex (Lindstrøm, 2022).

Jensen and Selling (2023, p. 12) explain how self-injurious behaviour may activate endogenous opioids, providing temporary relief and reinforcing the behaviour. This physiological feedback loop may partly explain why self-harm persists in some individuals who lack other strategies. In addition to behavioural explanations, neurochemical mechanisms may also play a role. Studies in humans and nonhuman primates suggest that the body's release of endogenous opioids, such as endorphins, can help regulate emotional stress in response to pain or distress (Tiefenbacher et al., 2005, p. 4).

With an understanding of these biological and sensory foundations, we now move to how emotional regulation skills develop through communication and interaction, particularly in children with congenital deafblindness.

2.4 The development of self-regulation, emotion regulation, and communication in children with congenital deafblindness

Deafblindness limits access to social cues and feedback, impacting the development of social and emotional skills. Sensory impairments make it harder to process environmental input and navigate emotional experiences (Nafstad & Rødbroe, 2013). From a developmental perspective, self-regulation emerges gradually through interaction with caregivers. Infants initially depend entirely on external regulation, as adults help soothe distress and maintain emotional balance. Over time, through repeated experiences of sensitive caregiving, children internalise strategies for managing emotions independently (Janssen et al., 2002; Hartshorne & Nicholas, 2017).

This developmental sequence, progressing from external regulation to co-regulation and eventually to self-regulation, is foundational for healthy emotional and cognitive development. However, children with congenital deafblindness often experience disruptions in this path. Limited access to social and emotional cues, such as facial expressions, tone of voice, and mutual gaze, can delay or alter the development of emotion regulation skills (Nafstad & Rødbroe, 2013).

Early affective exchanges, such as reading emotional cues and affective mirroring, may be compromised, which is crucial for establishing secure internal working models of self and others (Stern, 1998). As a result, children with CDB may rely on co-regulation for significantly longer periods than typically developing peers. They may require more precise, structured, and extended adult support to acquire effective emotional regulation strategies.

The building blocks of emotional regulation include recognising internal emotional states, predicting emotional consequences of actions, delaying gratification, using coping strategies, and seeking social support appropriately. In typical development, these skills are supported by rich, multimodal social experiences, which children with deafblindness have limited access to. Therefore, without sufficient and sensitive scaffolding, managing emotional states independently may be difficult, leading to higher vulnerability to dysregulation when faced with stress, unpredictability, or environmental challenges.

Communication development is intrinsically linked to the development of emotion in all children, including children with CDB. Many children with CDB do not have access to conventional language and rely on alternative communication forms. The Diamond Model (Nafstad & Rødbroe, 2013) offers a dialogical framework that fosters emotional resilience through supportive, communicative interactions. This reinforces the importance of emotionally available adults who recognise the child's cues to create safety and predictability (Tetzchner, 2019)

It has been suggested that emotional regulation in children with deafblindness should be viewed as both an individual skill and a relational process. Because of their limited access to conventional cues, they primarily depend on attuned adults who can respond with

sensitivity and predictability. Both positive and negative influences shape emotional development. Positive experiences, such as predictable routines, emotionally safe spaces, tactile communication support, and responsive interactions, lead to the gradual development of self-regulation (Nafstad, 2015). Conversely, unpredictable environments, inconsistent caregiver responses, or overstimulating surroundings can intensify emotional instability and reinforce maladaptive regulation patterns (Nordanger & Braarud, 2017).

Predictable routines reduce uncertainty and support self-regulation (Nafstad & Rødbroe, 2013). Maintaining emotionally safe, attuned, and responsive adult-child interactions helps this gradual developmental process in children with congenital deafblindness (Nafstad, 2015).

Chapter 3: Method

This study employs a systematic literature review to analyse and compare existing research on emotional dysregulation in children with CDB between 2005 and 2025. The PRISMA framework will guide the review process, ensuring transparency and reproducibility.

3.1 Document review as a method

A literature review is used to establish what is already known about a topic, identify existing theoretical frameworks, and uncover gaps in the current knowledge base (Hunziker & Blankenagel, 2024, p. 237). High-quality and relevant literature is essential for credibility. Credibility is often enhanced by reviewing the reference lists of selected studies. (Thagaard, 2018, p. 119).

In a highly specialised field such as congenital deafblindness, the availability of relevant empirical research is limited. This shortage necessitates broadening the scope to include grey literature and theoretical contributions. Many of the included studies were authored or coauthored by the same research teams, which may limit the diversity of perspectives. The relatively small size of the research community also raises the possibility of limited critical engagement between authors and a recurring reliance on shared theoretical foundations. Consequently, this review draws on a combination of peer-reviewed articles, book chapters, and grey literature to capture a comprehensive picture of the current state of knowledge on emotional dysregulation in children with congenital deafblindness.

3.2 Search strategy

The search strategy was designed to identify studies addressing emotional dysregulation in children with congenital deafblindness. Boolean logic was applied to structure effective search strings, using operators such as AND, OR, and NOT, as well as truncation (*) to capture relevant variations of keywords (Hunziker & Blankenagel, 2024, p. 64). The aim was to balance specificity and breadth to capture a comprehensive set of studies across disciplines.

The literature search was conducted using the following databases: SmartCat (University of Groningen), ERIC, Medline, and PsycInfo.

3.2.1 Search terms and combinations

The search included multiple keyword combinations such as:

Search Focus	Keywords/Combinations	
Emotion Regulation	"Emotional dysregulation", "self-regulation"	
Dysregulation	"Self-regulation difficulties", "Behavioural dysregulation"	
Deafblindness-Specific	"Deafblindness", "Dual sensory loss"	
Terms		
Broader Search Queries	"Emotional regulation" OR "emotion regulation" OR "self-	
	control" OR "self-regulation" "Deafblindness" OR "deafblind"	
	OR "deaf-blind" OR "deaf-blindness"	
Boolean Search: Self-	("Self-regulation" OR "self-control" OR "self regulation") AND	
Regulation and	("deafblindness" OR "deafblind" OR "deaf-blind" OR "deaf-	
Deafblindness	blindness")	
Boolean Search:	("Emotion regulation" OR "emotion dysregulation" OR	
Emotion Regulation and	"regulation of emotion" OR "emotional regulation") AND	
Deafblindness	("deafblindness" OR "deafblind" OR "deaf-blind" OR "deaf-	
	blindness")	

3.2.2 Findings and adjustments

The initial database searches using the keyword "emotion regulation" yielded few relevant results. The few hits that did appear were either unrelated to deafblindness or conceptually overlapped with studies using the term "self-regulation." Based on this, the search strategy was adjusted to prioritise "self-regulation" as the primary term, which proved more productive and conceptually appropriate for the literature identified.

The first round of searching with the term "self-regulation" yielded eight potentially relevant studies before applying exclusion criteria. Given the limited volume of research in this field, the strategy was further refined by incorporating synonymous and related terms. This iterative approach also included expanding eligibility criteria to include literature that addressed aspects of emotional dysregulation, even if the specific terminology varied.

The three removed articles by screening:

Article	Screening criteria
Gothelf et al., (2003).	Age of article
	Age of participants
Brown, D. M. (2011).	Relevance
Brown, D. M. (2021).	Relevance

To enhance the comprehensiveness of the review, citation tracking (both forward and backwards snowballing) was used, in addition to searches in grey literature and national library databases such as Bibsys. Expert consultation with researchers in the fields of deafblindness and self-regulation also helped identify studies that may have been missed through conventional database searches, thereby reducing the risk of publication bias.

A key example is the monograph by Nicholas and Hartshorne (2017), which includes several chapters relevant to self-regulation in CHARGE syndrome. One of these, Hartshorne et al. (2007), was retrieved through database searches and is therefore listed accordingly in the PRISMA diagram. Other chapters from the monograph were identified through manual review and are categorised under "other sources."

In addition to planned database searches, a broad exploratory search was conducted using the University of Groningen's SmartCat platform. The keyword "deafblindness" yielded 720 results, which were narrowed down to 639 by applying a 20-year publication limit. All records were rapidly screened by title, and full-text analysis was conducted on potentially relevant entries. This process led to the inclusion of Dammeyer (2014), which, although not using the term "self-regulation," discusses emotional and behavioural challenges in children with congenital deafblindness. The study also highlights the risk of autism misdiagnosis in this population—an issue closely related to emotional and behavioural regulation difficulties (Dammeyer, 2014; T. Hartshorne et al., 2005).

In total, 27 records were initially identified. After removing duplicates and screening titles and abstracts, 16 articles and book chapters were selected for full-text review. These are presented in the PRISMA diagram (Figure 2).

3.3 Study selection

The initial literature search yielded 27 records across peer-reviewed articles, grey literature, and book chapters. After removing duplicates, the remaining sources were screened based on their titles and abstracts. Full-text reviews were then conducted for publications that met the inclusion criteria.

Inclusion criteria:

• The study addressed emotional dysregulation, self-regulation, or related regulatory difficulties in children under 15 with congenital deafblindness.

• Publications included empirical studies, peer-reviewed articles, professional journal contributions, grey literature, or book chapters.

• The text was written in English or a Scandinavian language.

Exclusion criteria:

• The study exclusively focused on individuals over the age of 15.

• The content lacked specific relevance to deafblindness in its discussion of emotional or behavioural regulation.

• Publications older than 20 years were excluded due to significant developments in research and theory since 2005.

These criteria ensured a focused and up-to-date overview of emotional dysregulation in children with congenital deafblindness. After full-text screening, 16 sources were included in the final review. Of these, five were identified through systematic database searches, while eleven were located via hand-searching, citation tracking, or professional consultation. The overall selection process is summarised in the PRISMA flow diagram (Figure 2).

Table 1 provides an overview of all 16 included sources, regardless of study design or participant involvement, while Table 2 presents only the 10 studies that reported empirical data from participants.



PRISMA 2020 flow diagram for new systematic reviews which included searches of databases, registers and other sources

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Figure 2. Literature search process based on PRISMA framework

3.4 Data extraction and quality assessment

Following the final selection of studies, relevant data were systematically extracted using a structured data table. The extracted information included authorship and year, type of publication, methodological approach, participant characteristics, theoretical framework, and key findings related to emotional or behavioural regulation. Special attention was given to whether the study addressed interventions, assessments, or theoretical interpretations of emotional dysregulation in children with congenital deafblindness (CDB).

Given the limited number of empirical studies in this field, the inclusion criteria were intentionally broad. Both peer-reviewed research and grey literature, including professional journal articles, book chapters, and descriptive case reports, were considered, provided they made a meaningful contribution to the research questions. Methodological quality varied significantly across the included sources. While some were peer-reviewed studies with clearly defined designs and data, others were theoretical or practice-oriented texts lacking formal methodological details. Nevertheless, the inclusion of diverse literature was justified due to the limited availability of research and the need to capture a holistic view of emotional dysregulation in this population.

To address this heterogeneity, the review does not use a formal quality grading system. Instead, it considers methodological limitations and contextual factors in interpreting the findings in Chapters 4 and 5. This flexible yet systematic approach allows for the inclusion of underrepresented voices and perspectives while maintaining analytic accuracy (Popay et al., 2006).

3.5 Analysis plan

Given the heterogeneity of study designs, publication types, and theoretical orientations in the literature included, a narrative synthesis approach was chosen. Narrative synthesis is particularly suitable for integrating findings across studies that vary in methodology and cannot be statistically compared (Popay et al., 2006). The synthesis was guided by the overarching research questions and focused on identifying recurring patterns, conceptual frameworks, and implications for practice.

To ensure transparency and traceability, the review process involved multiple steps:

- All included sources were read in full and summarised in structured data extraction tables (see Table 1 and Table 2).
- • Table 1 presents all included sources, and Table 2 presents the subset of studies that included participants
- Information was categorised based on publication type, participant characteristics, theoretical framework, and main findings related to emotional dysregulation.
- The PRISMA flowchart was used to document the selection process from initial identification to final inclusion (see Figure 2).

Following the mapping of the literature, a thematic analysis was conducted. This involved coding the studies based on three focal themes:

- 1. Theoretical models used to conceptualise emotional or self-regulation in children with CDB.
- 2. Intervention strategies aimed at supporting regulation in educational or therapeutic contexts.
- 3. Assessment approaches, including how emotional dysregulation is observed, measured, or described in children with dual sensory loss.

Through this process, key theoretical and empirical trends were identified, along with notable inconsistencies in terminology and methodology. These insights formed the basis for the synthesis presented in Chapter 4 and the critical reflection in Chapter 5.

Table 1

Author(s) & Year	Participants	Main Focus and Findings	Interventions
Type of literature			
1) Bruce et al.	6 children (6–18	Peer interaction and social/self-	Structured sessions
(2016)	y/o)	determination development in	promoting peer
		deafblind students.	interaction.
Peer-reviewed			
2) Dammeyer	71 children	Distinguishing autism from	Suggests specialised
(2014)		sensory-based communication	assessments to prevent
		issues.	misdiagnosis.
Peer-reviewed			
3) Haney et al.	1 case	Behavioural dysregulation in	Behaviour interpreted as
(2017)	Male	CHARGE, including	communication, not just
	age not	impulsivity and meltdowns.	disruption.
Grey literature	specified		
4) T. Hartshorne et	No specific	Overview of CHARGE	No intervention; calls for
al. (2005)	participants	syndrome behaviour and the	tailored support
		need for individualised	strategies.
Peer-reviewed		support.	
5) T. Hartshorne et	100 children	Autistic-like traits in CHARGE	Highlights the need for
al. (2005)		syndrome.	tailored assessments and
			support.
Peer-reviewed			
6) Hartshorne et al.	98 children with	Executive function issues,	Advocates for Executive
(2007)	CHARGE	including impulsivity and	function targeted
		attention shifting, in	interventions.
Peer-reviewed		CHARGE.	
7) Hartshorne and	Theoretical	Social-emotional development	Encourages play,
Schmittel (2016)		delays in deafblind children	inclusion, and

Author(s) & Year	Participants	Main Focus and Findings	Interventions
Type of literature			
		due to reduced social access.	environmental
Peer-reviewed			adaptation.
8) Hartshorne et al.	No specific	behavioural characteristics in	Emphasises the need for
(2017)	participants	CHARGE, emphasising	individualised
		variability, stress sensitivity,	understanding and
Peer-reviewed		and a potential phenotype.	support strategies.
9) Jensen and	Theoretical/Prac	Self-harm in individuals with	Root-cause exploration
Selling (2023)	tical	deafblindness using a low-	and collaborative
		arousal approach.	response.
Grey Literature			
10) Kennert et al.	Descriptive	Cognitive self-regulation:	Suggests scaffolding and
(2016)		planning, monitoring, and	memory supports.
		inhibition challenges.	
Grey Literature			
11) Kennert et al.	Case: 14 y/o	Difficulties in emotional	Focuses on emotion
(2015)	girl. female	regulation in CHARGE arise	recognition and coping
		from sensory and cognitive	strategies.
Grey Literature		issues.	
12) Larsen et al.	Theoretical	Physiological self-regulation in	Promotes adaptive
(2016)		CHARGE and sensory	regulation strategies.
		adaptation deficits.	
Grey Literature			
13) Nelson et al.	1 child,	Self-regulation difficulties due	Interactive tasks,
(2016)	male, 5 v/o	to sensory challenges in the	anticipation strategies,
Peer-reviewed		classroom.	and calming
			interventions.

Author(s) & Year	Participants	Main Focus and Findings	Interventions
Type of literature			
14) Nicholas et al.	Theoretical	Misinterpreted self-regulatory	Focuses on
(2017)		behaviours in CHARGE	understanding the
		children.	behavioural context.
Grey Literature			
15) Dominant of	1	Multi domain calf regulation	Dron o con aven ortivo
15) Ramírez et al.	1 case	Multi-domain self-regulation	Proposes supportive
(2017a)	male, 11 y/o	challenges related to fatigue	routines and awareness of
		and sensory overload.	stamina.
Grey Literature			
16) Skei et al.	35 participants	Executive function deficits in	Stresses need for
(2023)	with CHARGE	CHARGE, especially	Executive function
		inhibition and working	support strategies.
Peer-reviewed		memory.	

Table 2

Theoretical Models and Frameworks used in the included studies. Only studies including participants are presented here.

Author(s) & Year	Model / Framework	Application and Relevance
Dammeyer (2014)	Autism Behaviour Checklist	Used to distinguish sensory-based social
	(ABC) – Differential	issues from autism traits in children with
	Diagnostic Tool	deafblindness.
Hartshorne and Schmittel	Attachment and Social-	Applied to describe how limited incidental
(2016)	Emotional Development	learning affects social development in
	Theories	deafblind children.
Hartshorne et al. (2017)	Describes CHARGE	Describes behaviour changes as responses
	syndrome behaviour shaped	to pain, anxiety, or overstimulation,
	by a unique pattern and low	highlighting that these behaviours aim to
	stress tolerance.	regain control or express distress.
Jensen and Selling (2023)	Low-arousal Approach	Used to explore and address the underlying
		causes of self-harm behaviour.
Kennert et al. (2016)	Metacognitive and Executive	Explains challenges in planning, attention
	Function Models	regulation, and cognitive flexibility
Larsen et al. (2016)	Sensory Integration Theory	Explains physiological self-regulation
		challenges as responses to sensory
		overload.
Nelson et al. (2016)	Vygotsky's Social Learning	Basis for scaffolded classroom intervention
	Theory & Emotional	supporting internalisation of self-
	Regulation	regulation.
Nicholas et al. (2017)	Functional Behaviour	Behaviour is viewed as adaptive and
	Perspective	meaningful rather than disruptive or
		inappropriate.
Ramirez et al. (2017a)	Self-Regulatory Strength and	Highlights the impact of stamina loss on
	Depletion Theory	emotional and behavioural control.

Skei et al. (2023)	Miyake's Executive Function	Used to interpret executive function
	Model	deficits (e.g., inhibition, shifting, working
		memory) in CHARGE.

Chapter 4: Results

The studies included in this literature review involve empirical investigations of children with congenital deafblindness and theoretical summaries. Together, the analysed studies comprise approximately 715 participants. The review encompassed a total of ten empirical studies, covering a range of methodological designs. One peer-reviewed study employed a single-case multiple-baseline design involving a 5-year-old child with deafblindness (Nelson et al., 2016). Additionally, several other case studies with single participants, although not all peer-reviewed, were included to capture rich, detailed information (Haney et al., 2017; Kennert et al., 2015; Ramirez et al., 2017a). Other studies used survey-based methods, cross-sectional population data, and grounded theory. For example, Bruce et al. (2016) applied grounded theory to examine peer interaction and self-determination in six children aged 6–18.

Several studies were published as part of the *Dbi Self-Regulation in Individuals with CHARGE Syndrome* monograph (Ramirez et al., 2017b), initially developed as master's thesis projects. These studies include structured case analyses of children with CHARGE syndrome and provide rich, observation-based empirical data on different domains of self-regulation, for example, Kennert et al. (2015, 2016). Dammeyer (2014) presented quantitative data on 71 children with congenital deafblindness (mean age 11.3 years), and Skei et al. (2023) conducted a population-based study of 35 individuals with CHARGE syndrome, including a subgroup under the age of 15. Hartshorne et al. (2005) contributed large-scale survey data on autistic-like traits and executive dysfunction in children with CHARGE syndrome. From individual case studies to population-level analyses, this methodological variety offers depth and breadth to understanding emotional dysregulation in deafblind children.

However, it is impossible to determine whether certain individuals are included in multiple studies, making the number of unique children represented in the material likely lower. This consideration is important when interpreting the findings. Table 1, located in Chapter 3, provides an overview of all included studies, detailing authors, methodology, participant characteristics, and main themes.

The following section presents the results from all included studies (N = 16), organised by main themes related to emotional dysregulation and self-regulation in children with congenital deafblindness. First, the theories and models of emotional dysregulation used in the studies are discussed. This is followed by an overview of the most central findings

concerning the prevalence, expression, and understanding of emotional dysregulation, as well as factors that promote or hinder the development of self-regulation in children under the age of 15 with CDB.

4.1 Theories and models of emotional dysregulation

Several studies, primarily among the 10 included participant studies (see Table 2), utilised theoretical frameworks to understand emotional dysregulation in children with congenital deafblindness, particularly in CHARGE syndrome. Although "emotional dysregulation" is not always explicitly mentioned, related constructs such as behavioural outbursts, self-harming behaviour, or difficulties with self-regulation are often discussed and interpreted within broader theoretical models.

The most frequently referenced model across the included studies is the fourdimensional model of self-regulation developed by Ramirez et al. (2017b). This model conceptualises self-regulation as consisting of four interrelated domains: cognitive, behavioural, emotional, and physiological regulation. Although the model itself was introduced in the theoretical monograph (Ramirez et al., 2017b), it was applied or referenced in three of the included participant studies: Ramirez et al. (2017a), Nicholas et al. (2017), and Kennert et al. (2016), all published as part of the *Dbi Self-Regulation in Individuals with CHARGE Syndrome* monograph. These studies draw on the model to structure observations, interpret regulatory challenges, and formulate intervention strategies for children and youth with CHARGE syndrome. In line with these multidimensional perspectives, Ramirez et al. (2017b) define emotional dysregulation as the inability to properly exhibit, inhibit, and alter thoughts, emotions, physiology, and behaviour. This definition supports the notion that dysregulation spans across domains, reinforcing the use of comprehensive models such as the four-dimensional framework.

For example, Ramirez et al. (2017a) describe the case of an 11-year-old boy facing aggressive outbursts and self-injury. They apply the four-dimensional model to explore how low stamina, emotional sensitivity, rigid thinking, and impulsivity interact to create a cycle of dysregulation. Interventions included sensory breaks, emotional labelling strategies, and visual schedules to support behavioural and cognitive regulation. Similarly, Kennert et al. (2016) present a case study focusing on cognitive self-regulation. They highlight how difficulties with executive functions, such as planning, monitoring, and inhibition, can affect

motivation and task completion. Their interventions focused on scaffolding techniques to strengthen attention control and support goal-directed behaviour.

In the article of Nicholas et al. (2017; "The Importance of Self-Regulation"), the significance of self-regulation for individuals with CHARGE syndrome across four interconnected areas is discussed: cognitive, behavioural, emotional, and physiological. They highlight challenging behaviours, such as withdrawing from social situations, acting impulsively, or even self-injury, can play essential adaptive roles or function as forms of communication. By employing this four-dimensional model, the authors suggest that we should view these behaviours not just as symptoms of dysregulation but as responses to deeper issues such as stress, cognitive overload, or unmet needs for self-regulation.

Similarly, Haney et al. (2017) illustrate how behavioural dysregulation in individuals with CHARGE syndrome is often linked to interacting factors such as sensory overload, executive dysfunction, and emotional stress. Behaviours such as impulsivity, fixation, tantrums, and aggression are described not as purely disruptive, but as coping mechanisms in response to internal states or environmental demands. These must be interpreted within the context of the individual's sensory and cognitive profile.

In line with this, Hartshorne et al. (2017) describe a behavioural phenotype in CHARGE syndrome, characterised by a low threshold for sensory overload and rapid escalation of stress responses. They argue that many challenging behaviours should be understood as expressions of stress, sensory overwhelm, or executive dysfunction rather than deliberate misbehaviour. This perspective further supports the theoretical framing that emotional dysregulation must be understood considering the persons neurodevelopmental and sensory context.

In addition to these cognitive and behavioural aspects, Larsen et al. (2016) underscore the role of physiological self-regulation in CHARGE syndrome. Children with CHARGE often experience difficulties maintaining homeostasis due to vestibular dysfunction, low muscle tone, and atypical sensory processing. These challenges can compromise the ability to regulate arousal levels and bodily states, making children with CHARGE syndrome more vulnerable to stress and emotional overload. The authors describe how disruptions in physiological self-regulation, such as over- or under-reactivity to sensory input, can cascade into emotional and behavioural dysregulation. By highlighting these physiological

foundations, the study deepens our understanding of how bodily regulation interacts with emotional control in children with CHARGE syndrome.

Together, these perspectives are backed by neurobiological explanations and models of executive function. Sensory stress and executive dysfunction are often identified as factors contributing to low frustration tolerance, difficulties with impulse control, and emotional vulnerability. In several case descriptions (Hartshorne et al., 2007; Ramirez et al., 2017a; Nicholas et al., 2017), these mechanisms are directly linked to issues with emotion regulation and self-injurious behaviour.

4.2 Expressions of emotional dysregulation

Children experiencing emotional dysregulation often feel intensely overwhelmed by their emotions. To cope with this discomfort, they may resort to behaviours such as selfinjury. Such behaviours are described in several case studies based on the four-dimensional model of self-regulation (Ramirez et al., 2017a) and theories of executive dysfunction (Haney et al., 2017). Jensen and Selling (2023) also discuss this, emphasising that we should address self-harm in children with CDB, even if it is not currently an issue for their well-being. While these behaviours may appear maladaptive, they often serve an important regulatory function for the child. Children with CDB do not have the same opportunities to observe how other children or adults react to and regulate themselves in response to environmental stimuli. From early infancy, children typically moderate their arousal levels, orient themselves, and respond to stimuli from their surroundings (Nelson et al., 2016). However, children with CDB do not have the same access to these experiences. They are often exposed to environments that are either overwhelming or lacking in stimulation, and unpredictable sensory information can lead to persistent hyperarousal. These arousal-related difficulties contribute to stress and reduced emotional flexibility, particularly in unfamiliar or overstimulating environments (Haney et al., 2017; Larsen et al., 2016). Regulating arousal is therefore fundamental to developing emotional stability and adaptive behaviour. As Skei et al. (2023) emphasise, executive functions are closely tied to physiological activation and emotional control, even though "arousal" is not used explicitly. Jensen and Selling (2023) further argue that a child's ability to regulate arousal is essential for preventing self-harm and enabling participation, learning, and social development. As Nafstad (2015) points out, the emotional regulation required to maintain communicative agency in such contexts is substantial, as children with deafblindness often must negotiate meaning without relying on conventional social cues and language.

Several of the reviewed studies focus specifically on CHARGE syndrome and the distinct characteristics related to self-regulation in children with this condition (n=11). CHARGE syndrome is one of the leading causes of deafblindness, and this population is one of the largest to study (Bruce et al., 2016, p. 150). Children with CHARGE syndrome often exhibit emotional dysregulation through heightened reactivity to sensory stimuli, frequent emotional outbursts, difficulty transitioning between activities, and increased impulsivity. Furthermore, many children with CHARGE syndrome struggle with managing frustration and exhibit perseverative behaviours, where they become fixated on specific routines or interests. Additionally, their ability to self-soothe or seek external regulation from caregivers may be diminished due to executive dysfunction and sensory integration challenges (Kennert et al., 2015)

Key differences emerge between children with deafblindness without CHARGE syndrome and children with CHARGE syndrome. While both groups experience difficulties with emotional regulation due to sensory limitations, children with CHARGE syndrome tend to display more severe executive dysfunction, leading to more significant difficulties in adapting to new situations and maintaining behavioural flexibility (Skei et al., 2023). Moreover, children with CHARGE syndrome are likely to exhibit autism-like traits, such as rigid thought patterns and repetitive behaviours, which can further complicate their ability to regulate emotions effectively. Although Dammeyer (2014, p. 1096) describes a higher risk of autism-like symptoms in children with deafblindness, impaired social interaction and repetitive behaviour are also observed in children with CBD without CHARGE syndrome.

4.3 Review interventions and their limitations

Internal and external factors influence emotional dysregulation in children with CDB. Studies highlight the roles of executive dysfunction and heightened arousal in CHARGE syndrome. (Ramirez et al., 2017a; Skei et al., 2023), Limited access to consistent and meaningful social interaction further complicates emotional development (Hartshorne & Schmittel, 2016). These underlying mechanisms inform how intervention strategies are selected and implemented across studies.

Six of the ten included empirical studies present targeted interventions, most of which are grounded in the four-dimensional model of self-regulation or draw on theories of executive dysfunction and arousal regulation. In addition, the theoretical and practice-based

framework presented by Jensen and Selling (2023) offers guidance on how to apply a lowarousal approach in emotionally challenging situations.

A recurring theme across the included studies is the importance of predictability and environmental structure in supporting emotional regulation. Interventions often rely on visual timetables, tactile calendars, and consistent routines to reduce anxiety and promote stability (Hartshorne & Schmittel, 2016; Nelson et al., 2016; Ramirez et al., 2017a). Such strategies are particularly effective for children who struggle with transitions, uncertainty, or sensory overload, standard features in children with CHARGE syndrome. From a theoretical perspective, these methods address both the behavioural and physiological dimensions of self-regulation by lowering arousal and enhancing the child's sense of control.

In line with these principles, Nelson et al. (2016) identified three promising strategies for reducing challenging behaviour. The first involves providing meaningful and interactive activities. The second strategy utilises visual support tools, such as calendar systems, to increase predictability and structure in daily routines. The third strategy incorporates various methods of stress regulation, including physical contact, emotional reflection, and sensory aids like weighted blankets and similar tools.

Hartshorne and Schmittel (2016) highlight how essential it is to create environments and activities that naturally encourage social interactions among children with deafblindness. They recommend using thoughtfully chosen "social toys" that foster joint attention and engagement. Additionally, designing open and uncluttered physical spaces can help minimise sensory overload and enhance mobility, making things more accessible for these children. This kind of adaptation is vital as it not only eases environmental stress but also plays an essential role in supporting emotional regulation. Furthermore, they suggest that children interacting with those who are deafblind should have typical developmental milestones, as this allows for valuable opportunities to model behaviours and learn through interactions, key components in these children's social and emotional growth.

Managing stress is important for effective interventions, as lower stress levels significantly improve a child's ability to learn, connect with others, and develop selfregulation skills. To help reduce stress, we can switch between familiar activities and new experiences, adjust our surroundings, like controlling noise and lighting, and encourage calming routines. These methods are especially beneficial for children with sensory sensitivities or those who experience heightened arousal responses, which are common traits

in individuals with CHARGE syndrome. Although co-regulation is not explicitly defined in Kennert et al. (2015), their case description of a teenage girl with CHARGE syndrome illustrates key co-regulatory practices. Through consistent adult support, emotional scaffolding, and environmental adjustments, the child gradually developed greater self-awareness and improved emotion regulation.

One of the first steps in supporting a child's emotional self-regulation is to help them identify, label, and understand their feelings. This skill becomes particularly crucial for children with CHARGE syndrome, who might face challenges recognising their emotions and linking physical sensations to specific feelings. Research by Kennert et al. (2015) highlights the importance of emotional scaffolding, such as naming emotions for the child, utilising visual aids, and demonstrating appropriate responses. In their study, these techniques were instrumental in helping a 14-year-old girl with intense emotional outbursts reflect on her feelings and develop coping strategies. This approach aligns with the emotional component of a four-dimensional model and supports the transition from dependence on external support to enhanced internal self-regulation. Moreover, giving children strategies to avoid or prepare for situations that may provoke strong emotions can help reduce both the frequency and intensity of emotional dysregulation.

in addition to emotion recognition and expression, several studies highlight the importance of preparing children to cope with intense negative affect (Nelson et al., 2016; Ramirez et al., 2017a; Kennert et al., 2015). This may include teaching specific calming strategies, as well as helping the child recognise and avoid emotionally triggering situations when appropriate. As illustrated in Kennert et al. (2015), proactive avoidance, such as preparing for transitions or minimising exposure to overwhelming environments, can reduce stress and support more effective self-regulation. These strategies are not about withdrawal but about improving the child's ability to predict and manage difficult emotions within their developmental capacity.

Chapter 5: Discussion

This literature review aimed to identify and synthesise the main theories, models, and findings concerning emotional dysregulation in children under the age of 15 with congenital deafblindness. The findings demonstrate that emotional dysregulation is a significant and recurring challenge in this population, often characterised by difficulties regulating arousal, behaviour, and affective responses. These difficulties are frequently attributed to a combination of executive dysfunction, sensory impairments, communication challenges, and stress-related vulnerability.

Regarding the first sub-question, the review found that the most applied theoretical model was the four-dimensional model of self-regulation developed by Ramirez et al. (2017b), including cognitive, behavioural, emotional, and physiological regulation. This model was used to frame the complex interplay between sensory loss and regulatory difficulties. In addition, elements from executive function theory and early developmental frameworks, such as attachment theory and co-regulation, were referenced in several studies.

A limited number of intervention studies were identified regarding the second subquestion. The described interventions primarily involved structured routines, tactile-visual anticipation systems, co-regulation strategies, and calming sensory input. These were often embedded in individual educational or therapeutic contexts. However, most interventions were case-based, and their generalizability remains limited.

As for the third sub-question, while the reported outcomes of interventions were generally positive, such as reductions in dysregulated behaviours and increased participation, the evidence for long-term effectiveness is limited. The studies rarely employed standardised measures, and most lacked longitudinal follow-up, making it difficult to draw firm conclusions about efficacy.

The following sections will interpret these findings with existing theory and research, and discuss implications for practice, limitations, and directions for future work.

5.1 Understanding emotional dysregulation in children with deafblindness: key insights from the literature

This section addresses the first research question by synthesising the key models and recurring empirical findings regarding emotional dysregulation in children with congenital deafblindness (CDB). Across the reviewed literature, emotional dysregulation was rarely

described as an isolated issue but was instead linked to a broader pattern of difficulties involving cognitive, emotional, behavioural, and physiological mechanisms (Hartshorne & Nicholas, 2017). A combination of sensory loss, neurobiological vulnerability, and environmental stressors shaped these challenges. In response to the research question, three theoretical frameworks and four cross-study themes were identified, each helping to explain different aspects of how emotional dysregulation manifests and is understood in this population.

The first theoretical approach identified across the reviewed studies was the fourdimensional model of self-regulation (Ramirez et al., 2017b), which conceptualises regulation as a dynamic interplay between cognitive, emotional, behavioural, and physiological processes. This model has been used to explain how sensory impairments, executive dysfunction, and emotional instability frequently co-occur and interact in children with CDB (Ramirez et al., 2017a; Ramirez et al., 2017b).

The second approach draws on executive function theory, which plays a central role in understanding emotional dysregulation, especially among children with CHARGE syndrome. Several studies have demonstrated that difficulties with impulse control, emotional flexibility, and attentional shifting can significantly influence how children regulate their emotional responses in demanding or unpredictable environments (Hartshorne et al., 2007; Skei et al., 2023). This theory provides a neuropsychological lens for interpreting challenges to regulation in daily life.

Third, several practice-oriented texts emphasise the use of the low-arousal approach (Jensen & Selling, 2023). This perspective emphasises the role of arousal regulation and the risk of escalation when support persons respond with high affect. It highlights the importance of calming strategies, predictable environments, and attuned adult responses (Jensen & Selling, 2023). Unlike the cognitive frameworks, this approach offers concrete guidance for adult responses and support strategies.

In terms of empirical findings, four recurring themes were identified across the reviewed literature.

First, emotional dysregulation in children with CDB is often expressed through behaviours such as self-injury, aggression, or social withdrawal. Importantly, these behaviours do not always indicate deliberate opposition or behavioural disorders. Instead, several authors emphasise that such actions may serve adaptive purposes, acting as regulatory

or communicative responses to overwhelming internal states or external demands (Haney et al., 2017; Jensen & Selling, 2023; Ramirez et al., 2017b)

Second, the sensory impairments characteristic of CDB have a substantial impact on the child's access to emotional feedback and co-regulation. Research shows that reduced or absent visual and auditory input limits the child's ability to interpret social-emotional cues, making it more difficult to develop emotional understanding and apply appropriate strategies (Bruce et al., 2016; Hartshorne & Schmittel, 2016; Nelson et al., 2016) This restricted access increases the likelihood of regulatory breakdowns in situations of stress or change.

Third, co-regulation and structured, predictable environments are crucial external supports for emotional regulation in children with CDB. Studies show that interventions involving consistent routines, tactile or visual schedules, and emotionally attuned adult responses are associated with reduced dysregulated behaviour and increased classroom participation (Hartshorne & Schmittel, 2016; Nelson et al., 2016). These findings are consistent with broader theories of self-regulation, which emphasise the importance of adult scaffolding in helping children with complex needs to develop regulation skills (Bodrova & Leong, 2005; Nelson et al., 2016; Ramirez et al., 2017a)

Fourth, the reviewed studies revealed considerable variation in how emotional dysregulation was defined and described. While some explicitly used the term "emotional dysregulation," others referred more broadly to concepts such as self-regulation or behavioural difficulties. This conceptual inconsistency reflects a growing understanding that dysregulation in children with congenital deafblindness is not a single emotional problem. However, rather a complex outcome of interactions between internal vulnerabilities—such as executive dysfunction—and external factors like sensory overload, unpredictability, and limited access to social cues (Dammeyer, 2014; Larsen et al., 2016; Ramirez et al., 2017b) These variations make cross-study comparisons more difficult and underline the need for more precise and consistent terminology in future research.

Taken together, the findings in this review support a holistic and contextualised understanding of emotional dysregulation in children with congenital deafblindness. Rather than viewing dysregulation as an isolated issue, it should be understood as part of a broader regulatory profile shaped by both neurodevelopmental and environmental factors.

5.2 Implications for practice and intervention

The review highlights the need for personalised, context-sensitive strategies to support emotional regulation in CDB. Given that emotional dysregulation may arise from a range of factors, including executive function difficulties and atypical sensory processing, interventions must be varied and tailored to address each child's unique developmental and communicative profile.

A developmental and relational approach should be adopted. Emotional regulation in children with CDB must be seen as part of a developmental process that evolves through interaction with caregivers and the environment. Professionals should emphasise co-regulation and responsive communication strategies as essential to the child's capacity to develop self-regulation over time (Nicholas et al., 2017; Ramirez et al., 2017b).

A key finding in the literature is the significance of supportive and emotionally attuned communication. Children with CDB often depend on others to help them recognise and manage their emotions. This dependence should not be seen as a weakness, but rather as a natural aspect of development, particularly given their limited access to social cues. Caregivers and educators, therefore, play a critical role by establishing predictable routines, labelling emotional states, and responding consistently to signs of stress. (Hartshorne & Schmittel, 2016; Jensen & Selling, 2023; Nelson et al., 2016)

Another key implication concerns the value of structure and environmental predictability. Multiple studies have shown that tools such as tactile calendars, visual timetables, and routine-based interventions can substantially reduce anxiety and dysregulation in children. These supports help provide a sense of control and security, enabling children to anticipate transitions better and regulate emotional responses (Hartshorne & Schmittel, 2016; Nelson et al., 2016; Ramirez et al., 2017a)

The review also underscores the importance of implementing strategies that directly support the development of self-regulatory skills. These include helping children identify physical sensations associated with emotional states and introducing calming techniques that can be internalised over time (Jensen & Selling, 2023; Larsen et al., 2016; Nelson et al., 2016). Such strategies must be adapted to the child's preferred modes of communication, cognitive profile, and sensory sensitivities (Ramirez et al., 2017a).

The use of low-arousal strategies is also strongly recommended. Practitioners should be trained to recognise early stress indicators and respond with calm, non-confrontational approaches. Environments should be structured to reduce overstimulation, and calming input, such as deep pressure, safe physical contact, or movement-based activities, may support regulation (Jensen & Selling, 2023).

Importantly, not all dysregulated behaviours should be interpreted as disruptive or oppositional. These behaviours may represent adaptive responses to stress, pain, or frustration, particularly when the child is unable to express needs or discomfort in more conventional ways. They are often best understood as signs of unmet needs or as efforts to cope with internal or external demands (Haney et al., 2017; Jensen & Selling, 2023). In such situations, how the adult regulates their affect and interprets the child's signals becomes crucial for de-escalating and providing practical support (Jensen & Selling, 2023).

Caregivers and staff require accessible and ongoing training in emotional development, sensory processing, and self-regulation. Strengthening their competence and confidence can enhance their ability to respond effectively to emotional challenges in everyday situations. Given the complexity of regulatory difficulties often observed in this population, interdisciplinary collaboration, particularly among professionals in special education, psychology, communication, and occupational therapy, is strongly recommended.

Finally, this review highlights a clear gap between theoretical understanding and the implementation of assessment and intervention practices. Although the literature describes several promising approaches, these are rarely embedded in standardised systems or evaluated through longitudinal research. There is a particular need for assessment tools tailored to children with congenital deafblindness. Such tools should capture the complexity of emotional regulation by incorporating behavioural, physiological, and communicative indicators, moving beyond reliance on traditional verbal feedback (Dammeyer, 2014; Ramirez et al., 2017b).

In summary, a shift is needed, from simply managing behaviour to understanding the relational, developmental, and sensory contexts in which behaviours occur. When caregivers and professionals are supported to reflect on the underlying meaning of behaviour and provided with concrete strategies for response, children with CBD are more likely to develop emotional regulation skills that foster participation, learning, and well-being.

5.3 Limitations of the study

This study is subject to several limitations that reflect the challenges inherent in conducting a literature review on a highly specialised and under-researched topic. One central limitation encountered in this review was the lack of consistency in how core concepts such as *emotional dysregulation*, *self-regulation*, and *behavioural regulation* were defined and applied across studies. These terms were often used interchangeably or without clear definitions, which made comparison and synthesis more challenging. For instance, Dammeyer (2014) referred broadly to behavioural and emotional difficulties without explicitly labelling them as emotional dysregulation. Similarly, Bruce et al. (2016) and Nelson et al. (2016) framed their findings in terms of self-regulation model that delineated emotional, cognitive, behavioural, and physiological domains. This model also served as a conceptual foundation in studies by Nicholas et al. (2017), Kennert et al. (2015), and Ramirez et al. (2017a). Such conceptual variation required careful interpretation to determine relevance to emotional dysregulation and may have affected which studies were included or excluded.

Another limitation lies in the limited availability of studies in this highly specialised field, which restricts the scope of the review. Despite a broad and systematic search strategy, only a small number of articles met the inclusion criteria. Many included publications were found in grey literature or internal monographs, and only a few were peer-reviewed empirical studies. This reflects both the scarcity of research and the fragmentation of knowledge about emotional regulation in children with congenital deafblindness. The inclusion of non-peer-reviewed literature was a deliberate strategy to address this scarcity, but it may affect the generalisability and perceived rigour of the findings.

A third limitation concerns the frequent focus on CHARGE syndrome across the included literature. Although CHARGE is one of the most common causes of CBD, this overrepresentation may limit the applicability of the findings to children with other aetiologies, such as congenital rubella, prematurity, or rare genetic syndromes. As a result, the review may not capture the full variability of emotional regulation challenges across the broader population of children with CDB.

A further limitation concerns methodological constraints within the reviewed studies. Many relied on small sample sizes, single-subject designs, or descriptive case studies, which limit the generalizability of the findings. Moreover, recurring authorship across multiple studies raises concerns about sample overlap, which was not always clearly disclosed. This makes it difficult to determine whether findings represent a broad evidence base or repeated analysis of the same individuals.

Finally, publication bias should be considered. Studies reporting positive intervention outcomes are more likely to be published, potentially skewing the available evidence base. In addition, the geographic concentration of studies, mainly from Nordic and Anglo-American contexts, may limit the cultural transferability of the findings. This is especially important in a field where cultural practices, service structures, and educational systems can shape how emotional dysregulation is perceived and supported.

Despite these limitations, the review offers a valuable insight into current knowledge and highlights critical areas for future research. The inclusion of diverse types of literature from peer-reviewed studies to master student research and book chapters, though potentially a limitation, also enhances the review by capturing a broader range of perspectives and practices relevant to children with congenital deafblindness.

5.4 Recommendations for future research

Although the reviewed studies provide valuable insights into emotional dysregulation and self-regulation in children with congenital deafblindness (CDB), several important knowledge gaps remain that limit the field's development.

First, many of the studies were small in scale, often involving single-case designs (Nelson et al., 2016) or descriptive analyses based on limited participant groups (Kennert et al., 2016; Kennert et al., 2015). This restricts the generalisability of findings and highlights the need for larger empirical studies that include more diverse participants across age groups, diagnoses, and cultural contexts.

Second, although some intervention studies have shown positive effects on reducing dysregulation and increasing participation (Nelson et al., 2016), few have been replicated or tested in larger samples. Longitudinal studies are lacking, and interventions are rarely implemented or evaluated in naturalistic settings such as homes or mainstream schools. This limitation restricts our understanding of how effective strategies can be sustained over time and across diverse environments.

Third, several studies point to the absence of standardised tools for assessing emotional dysregulation in this population (Dammeyer, 2014; T. Hartshorne et al., 2005). Due to the complexity of dual sensory loss, often in combination with intellectual disability, traditional assessment tools may be unsuitable or misleading, increasing the risk of misdiagnosis or under-identification of regulatory challenges. (Gross, 2014).

Moreover, the overrepresentation of CHARGE syndrome in the reviewed literature means that children with other causes of deafblindness are underrepresented. This limitation restricts generalizability and hinders the conclusion that aetiology may influence emotional regulation (Bruce et al., 2016; Skei et al., 2023). Future research should aim to include children from a broader range of aetiological backgrounds and investigate how differences in cognitive, communicative, and sensory profiles shape emotional development (Hartshorne et al., 2007).

Hartshorne and Schmittel (2016) underscore the complexity of social-emotional development in this population, highlighting barriers in developing empathy, secure attachments, and social competence. These areas are closely related to emotional regulation but remain underexplored in empirical research.

Based on these gaps, future research should prioritise: (1) the development of standardised, ecologically valid assessment tools that reflect the communication and sensory profiles of children with CDB; and (2) longitudinal and multisite studies that examine the development of emotional regulation across different support contexts, aetiologies, and educational environments. Such research is necessary to create more nuanced, effective, and inclusive support systems for children with congenital deafblindness.

Chapter 6: Conclusion

This chapter provides a concise summary of the main findings and recommendations. This systematic literature review aims to explore emotional dysregulation in children with CDB, a topic that is often underrepresented. It addresses three research questions:

- 1. What are the main findings and theories focused on in emotional dysregulation in children under 15 with deafblindness?
- 2. What are the main theories/models used?
- 3. How do existing interventions address emotional dysregulation, and what are their limitations?

6.1 Summary of findings

The findings reveal that emotional dysregulation in CDB is a complex issue arising from sensory impairments, executive dysfunction, and limited communication and social interaction. These behaviours, rather than disruptive, serve as adaptive responses to internal stress and external unpredictability. The most used model was the four-dimensional self-regulation framework (Ramirez et al., 2017b), describing regulation as dynamic interaction between cognitive, emotional, behavioural, and physiological processes.

6.2 Intervention strategies and limitations

Identified strategies include co-regulation, predictable routines, tactile-visual supports, and low-arousal environments. While promising, these approaches rely mainly on descriptive or syndrome-specific studies, particularly for CHARGE syndrome. The lack of standardised assessment tools for emotional dysregulation in children with dual sensory loss limits accurate diagnosis and tailored interventions.

6.3 Implications

This review emphasises the significance of emotional dysregulation in CDB as a relational, context-sensitive process. Interventions must focus on empathetic relationships, sensory-informed practices, and tailored supports. By viewing behaviours as communicative and regulatory strategies rather than problems, practitioners can respond more effectively and compassionately.

6.4 Future directions

Future research should prioritise validated assessment tools, evaluate intervention outcomes with more diverse samples, and explore longitudinal impacts of early regulation support. Expanding the empirical base will help professionals and families better support the emotional well-being, participation, and learning of children with congenital deafblindness.

These insights establish a foundation for improving emotional well-being and participation for these children.

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* = articles included in the systematic review

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