

The nature of language impairments in children with cerebral palsy: A scoping review protocol

Michelle McInerney^{a,c,*}, Wendy Pearce^b and Amanda Brignell^{b,d,e}

^a*University College London, London, UK*

^b*Australian Catholic University, Australia*

^c*Murdoch Children's Research Institute, Melbourne, Australia*

^d*Monash University, Melbourne, Australia*

^e*Monash Children's Hospital, Melbourne, Australia*

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Abstract.

BACKGROUND: Children with cerebral palsy (CP) frequently experience language impairments which can significantly affect their communication effectiveness, academic progress and social participation. Research describing language impairments in children with CP is limited, which has direct repercussions for accurate diagnosis; and timely assessment and treatment of communication skills.

OBJECTIVE: Little is known about the profile of language impairments in children with CP. This review seeks to describe language impairments in children with CP. Methods or tools that assess or measure language in children with CP will also be reported.

METHODS: This scoping review will be conducted in accordance with the Joanna Briggs Institute methodology for scoping reviews. The Preferred Reporting Items for Systematic Reviews and Meta-Analysis extension for scoping reviews (PRISMA-ScR) will be used to guide reporting. The International Classification of Functioning, Disability & Health- Child & Youth Version (2007) will be used as the conceptual framework to classify the nature of impairments. Studies that identify language impairments in children with CP (0–18 years) since the 1980s will be included. Seven relevant electronic databases and a targeted sample of the grey literature without any language restrictions will be searched. Two independent reviewers will assess articles for eligibility and three independent reviewers will extract relevant data.

CONCLUSIONS: This scoping review will provide important evidence on, and describe the nature of language impairments in children with CP. It will identify language assessment tools used in diagnosing impairments in children with CP. The findings will help to inform current clinical practice, and guide future directions in research.

Keywords: Children, cerebral palsy, language impairments

1. Introduction

Cerebral Palsy (CP) is the most common cause of childhood disability (Graham et al., 2016) and describes a group of non-progressive developmental

disorders of movement and posture causing activity restriction or disability attributed to disturbances occurring in the foetal or infant brain (Shih et al., 2018, Rosenbaum et al., 2007). CP is a lifelong condition. The birth prevalence in high-income countries is 1.6 per 1000 live births, and this prevalence increases in low- and middle-income countries with a rate of 3.4 per 1000 live births (McIntyre et al.,

*Corresponding authors: Michelle McInerney. E-mail: m.mcinerney@ucl.ac.uk.

2022). Due to the nature of the neurological and motor impairments, and the associated co-morbidities such as sensory impairments and epilepsy, many children with CP will experience communication difficulties which affects their ability to send and receive messages to those around them (Parkes et al., 2010, Hidecker et al., 2011). Breakdowns in communication are therefore common for children with CP and directly affect the child's activity and participation (Watson & Pennington, 2015).

Language impairments frequently underpin the communication challenges that children with CP experience (Hustad et al., 2015). Language is a structured system of communication, encompassing the domains of phonology, morphology, syntax, semantics and pragmatics (Paul et al., 2017). It is the primary means by which humans convey meaning, both in spoken and written forms. Language plays a critical role in facilitating daily activities and social participation (Mei et al., 2016), and impairments in language are also known to frequently co-occur with mental health issues (Hancock et al., 2023). The prevalence of language impairments is high in children with CP. Previously, a single population-based study of children with CP aged 4 years 11 months to 6 years 6 months estimated that 61% of children aged 5–6 years, and 36–74% within this entire age group will present with language impairments (Mei et al., 2015). Cognition and Gross Motor Function Classification System (GMFCS) levels are associated with rates of language impairment (Vaillant et al., 2020, Mei et al., 2016, Hustad et al., 2014). Inferring that children with CP who have greater cognitive and motor impairments are more likely to also present with language impairments.

Despite the prevalence of language impairments in children with CP, significant knowledge gaps exist. We know little about the nature of the presenting language impairment(s) in CP in childhood and whether there are differences in profiles of language impairments across the different sub-classifications of CP, that is, pathological movement pattern sub-type (e.g., spastic vs dystonic), topography (e.g., hemiplegia vs quadriplegia) etc. Even less is known about the impacts of language impairments on health and other areas of functioning, for example, how impairments in specific domains of language such as receptive language affects communication skills and abilities for participation. A lack of knowledge on this topic has direct implications for clinical assessment and intervention practice, and affects the supports provided to children with CP and language impairments. For

example, there are currently no international clinical practice guidelines, and no agreed protocol amongst therapists for paediatric language assessment in children with CP. Further knowledge about the nature of language impairments and their impacts across childhood in children with CP will help to inform assessment, intervention planning and implementation.

1.1. Review questions

This scoping review aims to identify and describe the nature of receptive and expressive language impairments in children with cerebral palsy (CP). Specific research questions include:

1. What is the nature of language impairments, inclusive of form (morphosyntax), content (semantics) and use (pragmatics), and varied modalities (e.g., spoken, written literacy and AAC), in children with CP from 0–18 years?
2. Are language impairments in children with CP described as an impairment at a body functions and structure level, activity, or participation level of health?
3. Are specific types of language impairments associated with different sub-classifications of CP e.g., spastic vs dyskinetic, hemiplegic vs. Quadriplegic, GMFCS level etc.?
4. What assessment tools or methods have been used to measure language in children with CP?

2. Methods

The proposed scoping review will be conducted in accordance with the Joanna Briggs Institute (JBI) methodology for scoping reviews (Peters et al., 2020). The Preferred Reporting Items for Systematic Reviews and Meta-analysis extension for Scoping Reviews (PRISMA-ScR) will be used to guide the reporting of the review (Tricco et al., 2018). A preliminary search of two databases was completed initially, and no current or underway systematic review or scoping reviews on the topic were identified. The International Classification of Functioning Disability and Health (ICF CY: World Health Organization, 2007) will be used as the conceptual framework for the study to classify impairments across the different health levels. As best practice, the protocol will also be registered with the Open Science Framework (OSF).

2.1. Eligibility criteria

Children with a diagnosis of CP. Specific inclusion and exclusion criteria are outlined below:

Inclusion criteria:

- (1) All children aged 0–18 years with a clearly stated diagnosis of CP will be included. The diagnosis of bulbar CP (Worster-Drought Syndrome) will also be included.
- (2) Participants diagnosed with CP i.e., congenital or acquired before the age of two years attributed to impairment of the brain and/or neuromuscular system.
- (3) Participants included in the study must have an identified language difficulty or impairment i.e., any reference to a language difficulty. Terms may include but are not limited to receptive (comprehension) or expressive deficits, vocabulary delays, social communication deficits and difficulties using language in a social context. Literacy difficulties are also of interest under the umbrella term of language.
- (4) Language (not speech) must be an identified impairment or primary focus or outcome of the study. Speech sounds disorders, stuttering or voice impairments were not the focus of this study.

Exclusion criteria:

- (1) Neurodisability sub-populations who have degenerative or progressive disorders e.g., muscular dystrophy, mitochondrial disorders etc. will be excluded.
- (2) A population of children who acquire a brain injury after the age of two years.
- (3) People aged 19 years and greater.
- (4) It is not clear from the population description that the sample/ person has CP.
- (5) The data for the CP sample cannot be extracted as it is reported.
- (6) The distinction between speech and language impairment is not made clear.

2.2. Types of sources

Research studies in all languages published since 1980s to May 2023 will be included. Studies from all clinical health settings or environments, and educational settings such as hospital, school, and research settings are of interest and will be included. Any study that outlines an impairment related to all domains

of language will be included for example, receptive, expressive, syntax, grammar, semantics etc. Experimental and quasi-experimental studies will be considered including randomized controlled trials (RCTs), non-RCTs, n-of-1 trials or single-case experimental design studies (SCEDs) such as multiple baseline, alternating treatment and changing criterion treatment designs; pre-post A-B designs and case studies. Qualitative research that describes language impairments will also be considered and may include grounded theory, phenomenology, ethnography, and ‘action research’ methodologies.

2.3. Search strategy

The search strategy aims to include both published and unpublished sources. A population intervention comparison outcome (PICO) framework will be applied to develop the research question, wherein key concepts relating to the population of CP, and language impairment will be generated. An initial preliminary search of the CINAHL databases will be completed to further identify and refine keywords to develop a comprehensive search strategy (See Appendix 1). The primary search will be through seven electronic published literature databases including the following: APA Psych Info, CINAHL, Cochrane Registry of Controlled Trials (CENTRAL), EMBASE, MEDLINE, Scopus, and Web of Science. The search strategy including the identified key words, subject heading, and index terms will be adapted for each of the included seven databases and/or source. A focused sample of the grey literature will also be sourced and restricted to the following: 1) Accessing relevant position papers from Professional Speech and Language Therapy Organizations e.g., Speech Pathology Association of Australia Ltd. (SPA) etc.; 2) Health/ governmental reports on language difficulties in children with CP; ProQuest Dissertation and Theses Global. The reference lists of all included sources of evidence will be screened for additional studies.

2.4. Study/source of evidence selection

After the search is completed, all identified citations will be exported to the reference management software Endnote (version 20) (www.endnote.com). An.xml file will then be imported to Covidence (www.covidence.org) directly from Endnote20. The Covidence setting whereby two researchers are required to screen each title and abstract will be

enabled. Initial screening by title and abstract against the inclusion and exclusion criteria will therefore be completed by two independent reviewers. Relevant resources will be retrieved in full and will be assessed against the inclusion criteria by two independent reviewers. Any full texts that do not meet the inclusion criteria will be recorded and reported. At each stage of the selection process, any disagreements will be resolved through discussion, and/or involve a third party (AB, MMcI, WP) until a consensus is reached.

2.5. *Data extraction*

A data extraction form will be developed, piloted, and adapted, as appropriate to extract relevant information from the studies included. Data to be extracted will include the research design/ type of source, country of origin, age of participants, type and severity of CP, language impairment type (receptive vs expressive), domains of language affected (syntax, semantics, phonology, morphology, pragmatics), level of impairment-health focus i.e., body structures and function, activity, and participation etc., and type of assessment, diagnostic method or language measure used. Two researchers will independently extract data from all included studies with any disagreement resolved through a consensus discussion with a third research member of the team. If necessary, authors of papers will be contacted to request missing or additional data for studies completed.

2.6. *Data analysis and presentation*

The study characteristics and basic data will be presented in written form and/or visually e.g., using tabulation, graphical and other visual methods. Depending on the quantity of papers included, where possible, data relating to different sub-classifications of CP will be included e.g., topography (diplegia, quadriplegia etc.), movement or motor-impairment type (spastic, dyskinetic etc.), GMFCS level etc. From a language impairment perspective, depending on the nature of the available data uncovered in the included sources, information on language domains (form, content, use), and modalities (oral and written language, AAC) etc. together with any impacts of language impairment for children with CP will also be reported. Differences in language presentations according to subclassification of CP will be analyzed.

The results of this review will be published and made available in a relevant scientific journal in addi-

tion to being presented at scientific, research and clinically-related conferences where appropriate.

3. Discussion

Specifically, this scoping review aims to systematically identify and map the profile of language impairments in children with CP and the assessment or outcome measurement tools used. It is anticipated that this review will increase our knowledge of the breadth of language impairments in children with CP, across the different sub-classifications of CP, and provide evidence on the impact to their functioning or health. The findings from this review will have implications for research and clinical practice. A better understanding of the profile of language impairments and how language is assessed in this population, will help to determine gaps in our knowledge and help to highlight future research directions e.g., best practice in assessment of language in children with different sub-classifications of CP. The knowledge gained will therefore contribute to optimising the care that children with CP and language impairments receive.

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Conflict of interest

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Supplementary material

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