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SCOPING REVIEW

Community-based respiratory health measures in children and young people with cerebral palsy: A scoping review

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Abstract

Aim: To identify, map, and describe outcome measurement domains and instruments used within a community setting to assess respiratory health in children and young people aged 1 to 18 years, diagnosed with cerebral palsy (CP).

Method: A scoping review methodology informed structured searches in nine databases, grey literature, and registries, conducted in August 2021 (updated in February 2023). Articles were screened for eligibility by two independent researchers. Any outcome measurement instruments used to assess respiratory health or associated impact were extracted, categorized, and mapped to health and health-related domains of the International Classification of Functioning, Disability, and Health.

Results: Seventy-six outcome measurement instruments were identified across 78 articles worldwide between 1970 and 2023. These were categorized into 'Body functions and structures' ($n=20$), 'Activity and performance' ($n=22$), and 'Participation and quality of life' ($n=19$), with a further 15 mapped to 'Health care resources use'.

Interpretation: No consensus of 'what' to measure and 'how' to measure respiratory health in children and young people with CP was found. Moreover, many measures were not replicable in individuals with more severe forms of CP, excluding those at increased risk of respiratory-related morbidity and mortality. Further research is required to agree important outcome domains and associated measures in research and clinical practice.

Cerebral palsy (CP) is the most common physical disability of childhood, affecting 2 to 3 per 1000 live births in resource-rich countries.[1] CP is characterized by changes in tone, movement, and posture, which can affect a child's ability to breathe, cough, and clear their chest effectively.[2] The condition also presents a distinct scope of co-existing impairments and factors that predispose children, aged 1 to 18 years, to respiratory illness, including reduced levels of voluntary activity, sedentary behaviour, learning disability, spinal or chest wall deformity, seizure activity, oropharyngeal motor dysfunction, and gastro-oesophageal reflux.[3–7] Respiratory illness is the leading cause of premature death in children and young people up to 25 years old with CP.[8] It is also the most common reason for this population to attend a primary care consultation, emergency hospital admission, and intensive care, accruing significant health care costs, increasing pressure on hospital resources,[9] and adversely impacting on the quality of life for children, young people, and their families.[10]

Outcome measures are a core component in respiratory health research to determine intervention effectiveness. Current interventions are wide ranging, aimed at tackling multiple, simultaneously occurring risk factors of oral-pharyngeal dysphagia, sialorrhoea, nutritional intake,

Abbreviations: ICF, International Classification of Functioning, Disability, and Health; OMI, outcome measure instrument; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

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dental hygiene, colonization, airway obstruction, and gastrointestinal dysfunction.[6,11] As part of a multidisciplinary approach, physiotherapists deliver conservative management strategies to improve respiratory fitness, breathing mechanics, effective coughing, and posture. This includes therapeutic positioning,[12] airway clearance techniques,[13,14] lung volume recruitment, and exercise.[15–18] These strategies can be delivered flexibly across community and acute care settings, employing preventative and reactive approaches to respiratory health. Evidence to support these strategies has been examined through a Cochrane review,[19] which revealed an overall poor quality of studies, with inconsistent outcome measures, limiting the extent to which results could be compared and generalized. A more recent systematic review of respiratory interventions[20] revealed no significant improvement in respiratory morbidity, observing only 5 of 37 studies were controlled, within which only two were judged to implement strong methodologies. The authors suggested several research avenues in this topic, with numerous interventions yet to be explored. However, attention to quality of study design, adequate sample size, and appropriate outcome measures was deemed vital.

Outcome measures are also widely implemented in clinical practice to assess and monitor respiratory health. Proactive measurement through anticipatory monitoring approaches can detect change, inform early care decisions and timely interventions, minimizing irreversible lung damage and emergency hospital use. This aligns with the UK National Health Service's long-term plan[21] to deliver health care closer to home. Furthermore, it reflects research priorities for paediatric hospital care set by James Lind Alliance Priority Setting Partnerships, questioning how hospital care can be improved, shortened, or even prevented for children who are medically complex.[22]

Respiratory health is typically measured using spirometry, a form of lung function testing used across chronic respiratory conditions[23,24] and neurodisability conditions.[25] However, these measurements are challenging to replicate in children with CP, because of learning, communication, and swallow impairments, skeletal deformities, altered tone, posture, and muscle weakness. In the absence of acceptable outcome measurements to assess, monitor, and evaluate respiratory health, children with CP continue to be vulnerable to respiratory morbidity and mortality.

To address this problem, a scoping review was undertaken to identify, map, and present the breadth of community-based outcome measure instruments (OMIs) implemented in primary research studies for children, aged 1 to 18 years, with CP. The review explored outcome domains measured, according to the International Classification of Functioning, Disability, and Health (ICF) conceptual framework,[26] the community-based context in which they were implemented and by whom, with consideration of limitations or exclusions imposed within research studies. This review excluded invasive measurements or those requiring a hospital stay, aligning with health care and stakeholder priorities aiming to reduce inpatient burden and promote proactive respiratory monitoring closer to home.[21,22]

What this paper adds

- A limited number and size of experimental designs were found.
- Seventy-six measures were identified to assess respiratory health in cerebral palsy.
- No consensus was found in 'what' or 'how' to measure respiratory health.
- Many measures were not replicable in children and young people at risk of poorer respiratory health outcomes.
- Children and young people with comorbidities and learning disability were frequently excluded from studies.

This scoping review aimed to systematically identify, map, and present existing, non-invasive, community-based OMIs used to assess outcome domains of respiratory health in research with children aged 1 to 18 years with CP.

This was achieved through the following objectives: (1) systematically search and identify relevant OMIs implemented in primary research studies, across a cohort of children with CP; (2) map OMIs to domains of the ICF framework;[26] (3) compare participant, measurer, and contextual factors, reported psychometric properties, and limitations or exclusions; (4) inform subsequent research direction in future 'intervention effectiveness' research and community-based clinical practice, within this population of interest.

The Participation, Concept, and Context framework informed the scope and research question for this review. The framework defined: (1) Participants: children aged 1 to 18 years with a clinical diagnosis of CP; (2) Concept: any non-invasive respiratory health measurement conducted using device, clinician, parent, or patient reporting; (3) Context: any OMI feasible to implement in a community-based setting.

The primary question of the scoping review was: 'What community-based OMIs are implemented in primary research to assess domains of respiratory health in children, aged 1 to 18 years, diagnosed with CP?'

The subquestions of the scoping review were: (1) What are the domains of respiratory health measured, according to the ICF?[26] (2) What are the participants' characteristics of those being measured? (3) What environments are used to implement respiratory health measures? (4) What are the measurer characteristics of those implementing respiratory health measures? (5) What psychometric properties are reported for respiratory health measures? (6) What

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are the limitations or exclusions for respiratory health measures?

METHOD

A scoping review methodology was selected to align with the purpose of identifying and mapping existing respiratory health outcome measures in a specific cohort of interest.[27] This review was conducted in accordance with the JBI methodology for scoping reviews for clarity and transparency[28] and registered with JBI in September 2021 which was updated in September 2022 (Appendix S1). As recommended by the JBI, the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) extension for scoping reviews checklist[29] was used to guide high-quality reporting of this review (Appendix S2).

Eligibility criteria

Participants

Authors considered studies including children with a clinical diagnosis of CP. This condition was selected as an exemplar of chronic neurodisability, because of its prevalence, well-defined presentation, and widely established association with respiratory-related morbidity and mortality.[8] Mixed condition cohorts were included if subgroup or individual data were reported for those with CP. The age criteria excluded cohorts under 1 year (mean) because of neonatal differences in respiratory anatomy, physiology, and management, and adults over 18 years (mean) because of differences between adult and paediatric OMIs and health care provision.

Concept

Authors considered any OMI examining respiratory health and/or its impact, encompassing any domain of the ICF framework.[26] OMIs included devices, performance-based measures, and child/proxy/clinician-reported instruments. Authors excluded OMIs involving harmful radiation or ionization, or those requiring implementation in a hospital setting, to reflect the community-based context of interest for this review.

Context

This review considered studies conducting OMIs in community-based settings that did not require a hospital stay, inclusive of outpatient health care clinics, rehabilitation and laboratory facilities, home, and educational settings. These criteria were selected to align with health initiatives aimed to reduce risk of burdensome hospital admissions that impact on quality of life for children and families.[10]

Research designs

Any primary research design, involving quantitative, qualitative, or mixed method, was included to broaden the exploration of respiratory health measures. Authors excluded secondary research and registered protocols. However, reviews and protocols posing a relevant research question were subject to citation screening and hand searching of subsequent published findings respectively.

Types of sources

Authors included unpublished papers and dissertations of primary research if sufficient data were available for extraction.

No date limit was set. Non-English language papers underwent machine translation engine translation and were screened for inclusion.

Search strategy

Authors completed preliminary searches in MEDLINE and CINAHL to examine and index applicable keywords, informing initial search terms. These were cross-referenced with relevant systematic reviews.[19,20] The research team agreed three broad search concepts: 'paediatric', 'neurology', and 'respiratory', with exhaustive use of synonyms to ensure comprehensive searching. These were trialled in MEDLINE before final refinement (see Appendix S3 for an example search strategy).

Full searches were conducted on 3rd August 2021 in the following databases: AMED, CINAHL, EMCare, MEDLINE, PsycInfo, Ethos, Cochrane Central Register of Controlled Trials, Cochrane Database of Systematic Reviews, and PROSPERO. Databases reflected a range of biomedical, allied health, and nursing databases, aligning with multidisciplinary management of respiratory health. Grey literature and registry databases were included to encompass unpublished studies, protocols, and registered trials. To minimize publication bias, final included papers and relevant reviews were subject to citation screening. The searches were repeated on 24th February 2023 to ensure the most up-to-date papers were included in the review. An information specialist was involved in refining the search strategy.

Study/source of evidence selection

All retrieved database citations were uploaded to Endnote 20 reference manager software (Clarivate Analytics, Philadelphia, PA, USA) and duplicates removed. Remaining references were uploaded to a web-based software, Rayyan (Rayyan Systems, Cambridge, MA, USA), for screening. Two research team members undertook a pilot screen of 100 titles and abstracts, informing discussion and refinement of the criteria.

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Two independent reviewers from the research team completed the final five-stage screening process detailed as follows: (1) Title and abstract of all retrieved articles underwent eligibility screening; reviewer discrepancies were provisionally included for full text screen. (2) Eligible titles and abstracts underwent full text screening; reviewer discrepancies were resolved through discussion. All reasons for exclusion were recorded in Rayyan software and in the PRISMA flow diagram. (3) Eligible full texts underwent a second stage of screening to exclude cohorts that did not meet a clinical diagnosis of CP. Reviewers screened cohorts of mixed neurological diagnoses for available individual or subgroup data. All reasons for exclusion were recorded in Rayyan software and in the PRISMA flow diagram (Figure S1). (4) A final refinement stage of screening was performed across all retrieved articles to ensure all relevant papers were identified, including those published in non-English languages. (5) Eligible papers and relevant reviews were subject to citation screening by the first author (RKL) to identify additional primary studies. Authors were contacted from all relevant protocols and trials registrations to confirm published study activity to date.

The results of the search and study inclusion process is presented in a PRISMA flow diagram[30] (Figure S1). Results of the rerun of searches are provided in Figure S2.

Data extraction

The research team codeveloped a data extraction tool in Excel, using the primary and subresearch questions of this review. The tool was piloted with two research team members using the first five eligible papers, prompting amendments and refinement for purpose (Appendix S4). Data extraction included study and participant characteristics, outcome measures of interest and their characteristics (outcome domain, time points, measurer characteristics, and reported psychometric properties), and exclusions or limitations to implementing each outcome measure. The first author (RKL) extracted data from all included papers and a second independent reviewer (SG) extracted every other included paper, amounting to 50% of all papers. Three individual data extraction discrepancies arose between authors, in reference to recording a Gross Motor Function Classification System (GMFCS) level, a sample size, and an outcome measurement for caregiver stress. All three were resolved with discussion. Results are presented in summary tables developed by the research team and adapted from the JBI manual.[28] Where data were missing, authors RKL and SG recorded this as 'not reported'.

Data analysis and presentation

All extracted OMIs were categorized and mapped onto ICF domains and subdomains by two members of the research team, followed by peer discussion and review by the wider research team. A narrative analysis of results was undertaken, presented under headings of subquestions posed in the review, supported by tables and visual graphs where relevant.

RESULTS

Search results

Primary searches retrieved 4805 articles from databases and uploaded to Endnote. After removal of duplicates, 3741 articles were subject to title and abstract screening. A subsequent 583 articles were subject to full text screening, of which 382 were excluded with reasons agreed and recorded in Figure S1. The remaining 201 articles were eligible for a subsequent full text screening of children with CP, excluding a further 163 articles exploring children with neuromuscular conditions ($n = 86$), neurogenetic conditions ($n = 27$), neurodivergent conditions ($n = 15$), other progressive conditions or mixed cohorts without subgroup analysis of CP ($n = 35$). Primary searches retrieved 38 eligible articles. An additional seven articles were found from rerunning and screening of searches (February 2023). These were supplemented by 33 articles sourced through citation screening ($n = 22$) and hand searching relevant reviews ($n = 11$), one of which was a non-English language review which was subject to Google Translation of citations. Results are presented in Figure S1.

Study characteristics

The scoping review identified 78 eligible studies, originating from 29 countries, with the greatest number of articles conducted in Netherlands ($n = 11$), USA ($n = 10$), Republic of Korea ($n = 9$), and Australia ($n = 8$). Studies spanned five decades, from 1970[31] to 2022 with 60% ($n = 47$) of articles published in the last decade, suggesting increased interest in this area in recent years. Study characteristics are presented in Table S1; OMI characteristics are summarized in Table S2.

Observational designs dominated included studies, with only 19 experimental designs (24%), of which only 11 (14%) reported randomization. Sample size was much lower in experimental designs, the largest cohort recorded as 68,[32] compared to 551 from observational studies.[4] This limited number and size of experimental designs align with findings from topic-related systematic reviews.[19,20]

Subquestion 1: Outcome domains of respiratory health

Outcome domains of measurements were labelled and mapped to the ICF framework[26] core and subdomains, and presented in Figure 1. Core domains included 'Body

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functions and structures', 'Activity and performance', and 'Participation and quality of life'. An additional domain of 'Health care resources use' encompassed measurements of structure and process of care, although this category partially mapped to 'environmental factors' of the ICF and was linked to 'Participation and quality of life'.

Body functions and structures

There were 20 OMIs categorized under 'Body functions and structures', implemented 73 times in total across 51 of 78 studies (65%), and observed to be the most frequent outcome domain measured. Outcome subdomains included

Domain	Sub-domains	Sub-themes
Body functions and structures	Respiration functions	Respiratory rate and rhythm
		Lung capacity, volume, resistance, and flow
		Gas exchange
		Reported respiratory-related symptoms
		Chest wall expansion / movement
	Respiratory muscle functions	Respiratory muscle strength
	Additional respiratory functions	Respiratory function related to sleep
		Respiratory function related to feeding
	Exercise tolerance functions	Respiratory aerobic capacity
		Reported exertion
Activity and performance	General tasks and demands	Functional activity measures
		Handling stress and psychological demands
	Mobility	Walking
		Activity levels / sedentary behaviour
		Changing and maintaining posture
		Aerobic performance / fitness
	Self-care	Activities of daily living
		Sleep activity
		Feeding activity
Participation and quality of life	Community, social, and civic life	Child Self-perception
		Child enjoyment
		Caregiver self-perception
		Caregiver social life and support
	Quality of life for child	Child health
		Child (generic)
		Impact of condition (sleep)
		Impact of condition (respiratory)
		Impact of condition (cerebral palsy)
	Quality of life for caregiver	Caregiver health
		Caregiver (generic)
		Impact of condition (sleep)
		Impact of condition (respiratory)
		Impact of condition (cerebral palsy)
Health care resources use	Healthcare admissions	Emergency care (frequency / duration)
		Secondary care (frequency / duration)
		Intensive care (frequency / duration)
	Healthcare visits	GP primary care (frequency / duration)
	Respiratory-related Illnesses	Frequency / duration
		Impact on non-pharmacological needs
		Impact on pharmacological needs
	Pharmacological care needs	Antibiotic use (frequency / duration)
	Healthcare investigations	Frequency e.g. CXR
	Non pharmacological care needs	Frequency e.g. suction

FIGURE 1 Domains of outcome measurement mapped to the International Classification of Functioning, Disability, and Health. Abbreviation: CXR, chest x-ray.

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respiratory function, respiratory muscle function, exercise tolerance, and additional respiratory-related sleep and swallow function. These OMIs featured in 16 of 19 experimental designs, and were primarily conducted by clinicians in controlled environments such as laboratories. Over half of these OMIs reported participant exclusions, primarily children unable to follow instructions. This was attributed to effort-dependent measures requiring voluntary breath control (e.g. in lung function testing), and compliance wearing of equipment (e.g. facemasks).

Activity and performance

There were 22 OMIs categorized under 'Activity and performance', implemented 51 times in total across 38 of 78 studies (49%). Outcome subdomains included general tasks and demands, mobility, and self-care inclusive of sleep and feeding. Objective activity measures primarily required a controlled clinic or laboratory environment (e.g. for treadmill, shuttle run tests, and cycle ergometry testing), of which all imposed exclusions related to level of mobility and ability to follow instructions.[32–41] In contrast, subjective activity measures were implemented in schools, clinics, and home. OMIs within this domain reported highest levels of psychometric properties, although measurement development studies featuring in this review primarily recruited ambulatory children with CP, questioning application of psychometric properties to a wider cohort of children with CP.

Participation and quality of life

There were 19 OMIs categorized under 'Participation and quality of life', implemented 22 times across 12 of 78 studies (15%). These OMIs featured in only 5 of the 19 experimental designs. Despite being the least frequent outcome domain measured, the number of OMIs was comparable to other domains, noting high variability. Outcome subdomains included community, social and civic life, and quality of life measures related to the child and/or the caregiver. OMIs in this domain were implemented across a variety of research designs, context, and cohorts, imposing the least number of exclusions. This domain encompassed 4 out of 5 child-reported OMIs.[32,39,42] All measures contributed ordinal data sourced from Likert scales.

Health care resources use

There were 15 OMIs of 'Health care resources use', implemented 45 times, across 18 studies, of which only three were experimental designs.[14,43,44] These OMIs described structures or processes of respiratory-related care, contributing discrete levels of data, calculated using frequency or length of care (in days). Most frequently implemented measures included number ($n=9$) and duration ($n=8$) of respiratory-related hospitalizations. Although categorized under 'Health care resources use', repeated hospitalizations also overlapped with subthemes of 'Participation and quality of life'.

Subquestion 2: Participant characteristics

Eligible studies included participants aged 1 to 18 years old, inclusive of sex. Several studies ($n=13$) enrolled mixed cohorts over 18 years, encompassing children and young people up to 26 years, although the mean age remained within eligibility criteria. There were no significant differences in the outcome domains or OMIs used in paediatric compared to mixed age cohorts. However, selected OMIs such as the Self-Perception Profile for Children and the TNO-AZL Questionnaire for Children's Health-Related Quality of Life[32] were used across the mixed cohort despite reported validity in paediatric populations only.

Of 78 included studies, 16 (21%) did not report GMFCS level, limiting full analysis of this subquestion. Eighteen studies included children of 'any' GMFCS classification, representing GMFCS levels proportionately, notably 32.5% of participants were classified in GMFCS levels IV to V, aligning with recent registry literature.[45] These studies implemented OMIs across all ICF domains,[26] most commonly within 'Activity and performance'. However, these measures did not examine the 'mobility' subdomain, but rather parent-reported sleep[46–55] or feeding activity.[3–5,56] Figure 2 provides a visual comparison of measurement domains across identified GMFCS cohorts.

Of the studies reporting GMFCS levels, 22 (35%) recruited only ambulatory children classified in GMFCS levels I to III, representing the most prevalent group of participants. These studies overwhelmingly favoured OMIs of 'Body functions and structures', specifically lung function, respiratory muscle strength, and aerobic capacity, or 'Activity and performance' domains, specifically aerobic exercise testing. Notably, none of these studies implemented measures of 'Health care resources use', likely attributed to lower risk of respiratory morbidity amongst ambulant children with CP compared to their non-ambulant counterparts.[3,4]

Thirteen studies selected children classified in GMFCS levels IV to V, reflecting those most at risk of respiratory illness. Within this domain, no 'Activity and performance' measures were implemented. 'Body functions and structures' measures primarily examined parent-reported respiratory symptoms[57,58] and non-effort-dependent measures; this included home oximetry, capnography, respiratory observations,[59–63] and a portable interrupter technique used to measure airway resistance,[60,64] noted to be validated in young children without a neurological condition.[65]

Four outlier studies[60,63,66,67] attempted to implement effort-dependent measures in children classified in GMFCS levels IV to V, including lung function tests and maximal

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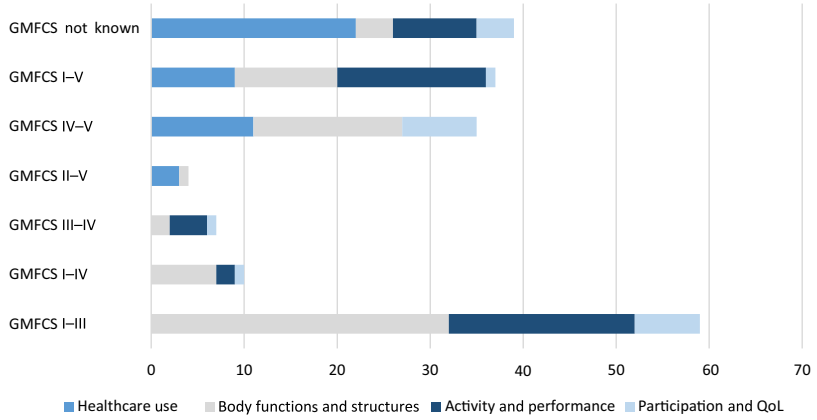

FIGURE 2 Visual presentation of measurement domains across reported Gross Motor Function Classification Scale (GMFCS) cohort groups. Abbreviation: QoL, quality of life.

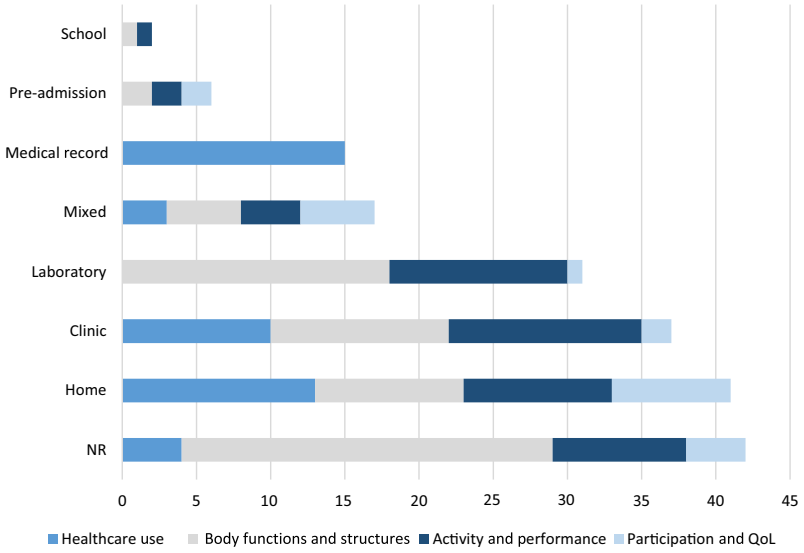

FIGURE 3 Visual presentation of measurement domains across reported contexts. Abbreviation: QoL, quality of life.

oxygen consumption during exercise. These studies all excluded participants who were unable to follow instructions. Once enrolled, participants were further excluded because of intolerance or agitation when wearing a face mask, postural instability, primitive reflex activity, and inability to cooperate with procedures, with one study excluding 50% of their recruited cohort.[63]

Subquestion 3: Measurement context

Of 78 included studies, 18 (23%) did not specify the environment in which OMIs had been implemented, although authors considered them to be feasible to implement in the community. Three studies sourced 'Health care resources use' from medical records.[9,68,69] Six studies reported flexible or mixed contexts in which measurements could be undertaken in school, home, clinic, online, or telephone. The remaining studies varied in context, most commonly including outpatient facilities or clinics ($n = 20$), followed by home ($n = 15$), laboratories ($n = 13$), school ($n = 1$), and pre-admission environments ($n = 2$). Figure 3 provides a visual comparison of measurement domains across identified contexts.

Objective measures were implemented in laboratories or outpatient facilities, primarily measuring 'Body functions and structures' (e.g. lung function) or 'Activity and performance' (e.g. aerobic exercise testing). Only three objective measures were implemented in the home: an activity monitoring device,[70] home oximetry,[61,62] and airway mucus encumbrance clinical assessment.[62] Subjective measures represented all ICF domains, although dominated by 'Health care resources use' and 'Participation and quality of life'. These were implemented flexibly across all contexts. Only two subjective measures were implemented in a laboratory environment: the Borg scale and the Children's OMNI Scale of Perceived Exertion, both of which were linked to aerobic exercise testing.[33,39]

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Subquestion 4: Measurer characteristics

Measurer characteristics informed a subquestion for this review, to explore key stakeholders in the assessment of respiratory health, whilst identifying measurers who may be at risk of research burden. Of 78 included studies, only 10 measurements across three studies[14,71,72] did not report measurer characteristics. Clinicians or researchers implemented 104 measures across 50 studies, largely mapping to 'Body functions and structures' and 'Activity and performance' domains. These were primarily objective, with few subjective measures reported by clinicians, limited to 'Health care resources use'.

Parent/caregivers implemented 64 measures across 32 studies, mapping to all levels of the ICF. These were subjective, reported through ordinal scale questionnaires or discrete 'Health care resources use'. Only seven recordings of child-reported measures were found across four (5%) studies; this included two 'Body functions and structures' measures, the OMNI Scale of Perceived Exertion[33] and the Borg scale,[39] and five 'Participation and quality of life' measures, including the Self-Perception Profile for Children, the Children's Assessment of Participation and Enjoyment, the TNO-AZL Questionnaire for Children's Health-Related Quality of Life,[32] patient-reported KINDL,[39] and the Children's Quality of Life Scale.[42] Figure 4 provides a visual comparison of measurement domains across identified measurer characteristics.

Subquestion 5: Psychometric properties

Forty OMIs reported psychometric properties, which were extracted and categorized into reliability, validity, responsiveness, or clinical utility, aligning with established health measurement taxonomy.[73,74] Forty-six (54%) studies reported at least one psychometric property for one of their implemented OMIs. Of these, 31 studies reported at least one property for *all* their implemented measures. There were no trends noted between level of reporting and research method.

Within this review, six studies directly explored measurement development: Brehm et al.[33] and van den Berg-Emons et al.[38] examined reliability of cycle ergometry to assess peak aerobic power; Gorter et al. measured reliability[75] and feasibility[70] of a treadmill test; Verschuren et al. examined reliability, reproducibility, and validity of shuttle run tests[34] and shuttle ride tests.[35] Four of the six studies recruited children with CP classified in GMFCS levels I to III.

Validity was most reported in studies ($n = 37$), followed by reliability ($n = 29$) and clinical utility ($n = 6$). Reliability and clinical utility featured primarily in OMIs of 'Body functions and structures' and 'Activity and performance', whilst validity was reported across all measurement domains. Four outlier studies reported validity, but acknowledged using the OMI outside its intended population or method.[58,62,70,76] One study reported responsiveness in a validated sleep questionnaire.[77]

Completeness of reported measurement properties differed across outcome domains. 'Activity and performance' OMI properties were most widely reported, representing 17 (77%) of the 22 identified OMIs. 'Participation and quality of life' OMI properties were reported for 13 (68%) of the 19 identified OMIs in this domain. 'Body functions and structures' OMI properties were reported for 11 (55%) of the 20 identified OMIs. Table S2 presents these findings.

Subquestion 5: Exclusions or limitations

A key rationale for this review, and what sets children with CP apart from other chronic respiratory populations, is the difficulty replicating 'criterion standard' respiratory measures such as spirometry.[23,78] Reported eligibility criteria of

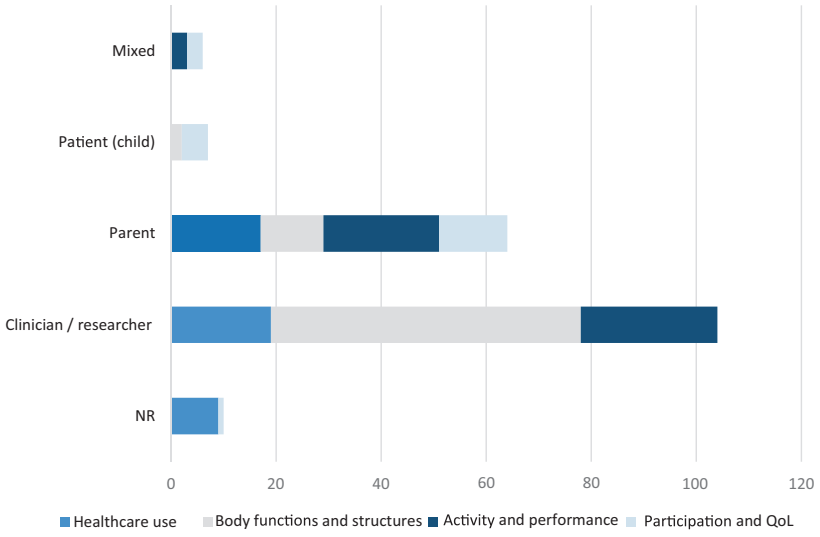

FIGURE 4 Visual presentation of measurement domains across reported measurer characteristics. Abbreviation: QoL, quality of life.

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cohorts warranted examination to determine application of existing OMIs to a wider population of children with CP and respiratory health impairment.

Of 78 included studies, 23 (29%) did not report exclusion criteria for their study, limiting a full analysis for this review. Of the remaining 55 studies, the most common exclusion criterion was an 'inability to follow instructions' or a cognitive impairment, reported in 28 studies. This criterion was most prevalent in studies recruiting GMFCS levels I to III cohorts and justified through need to comply with effort-dependent measures of lung function, aerobic performance, and exercise testing. However, this exclusion was also noted in 10 studies enrolling children in GMFCS levels IV to V, such as Park et al.,[66] who recruited 112 children in GMFCS levels IV to V for chest x-ray examination, but only 10 were able to cooperate in lung function testing.

The second most common exclusion was children with a known acute or chronic respiratory impairment such as asthma ($n = 18$ studies), frequently justified by risk of exacerbation during exercise testing or resisted breathing techniques. However, 13 studies purposely recruited children with respiratory morbidity or related symptoms, such as drooling, and aspiration. All but one of these studies[79] included children in GMFCS levels IV to V, recruiting cohorts at highest risk of respiratory-related mortality.[5] These studies were dominated by subjective measures of 'Health care resources use', 'Body functions and structures', and 'Participation and quality of life'.

Further exclusions included other comorbidities such as seizures ($n = 13$), medication ($n = 9$), posture or level of mobility ($n = 10$), orthopaedic or spinal deformity ($n = 7$), recent surgery or botulinum neurotoxin A injection ($n = 7$), pain ($n = 1$), and behaviour ($n = 1$). Exclusions related to treatment contraindications were justified in five studies, related specifically to aerobic fitness testing. Many of these exclusions reflect factors associated with more severe forms of CP and poorer respiratory health outcomes.[5,7]

Figure 5 provides a visual comparison of exclusions across GMFCS cohorts.

DISCUSSION

The purpose of this scoping review was to identify, map, and present respiratory health outcome domains and measures implemented in primary research studies for children with CP. The review revealed a limited number and size of experimental designs, lacking consensus of 'what' and 'how' respiratory health is measured in research. This limits comparison and synthesis of findings across studies, aligning with concerns raised by Blackmore et al.[20] and Winfield et al.[19]

The review identified 76 OMIs across 78 studies, which were mapped to core ICF domains: 'Body functions and structures', 'Activity and performance', and 'Participation and quality of life'.[26] 'Health care resources use' was adopted as an additional domain as it only partially mapped to ICF 'environmental factors'.[26] On reflection, authors may consider alternative conceptual frameworks in future research with increased suitability towards outcomes, such as the COMET-informed taxonomy,[80] reflecting wider 'Health care resources use', associated with respiratory-related morbidity and mortality. This particular domain provides valuable information to inform sustainability in health care and aligns with UK health initiatives[21] and the James Lind Alliance.[22] However, these constructs do not provide anticipatory monitoring opportunities to detect early change, inform timely interventions, and minimize lung damage, such as those observed in best practices for other chronic respiratory illnesses.[23] Furthermore, the discrete data do not capture meaningful impact of these constructs on children and their families, suggesting the need to combine such measures with meaningful 'Participation and quality of life' outcomes.[10]

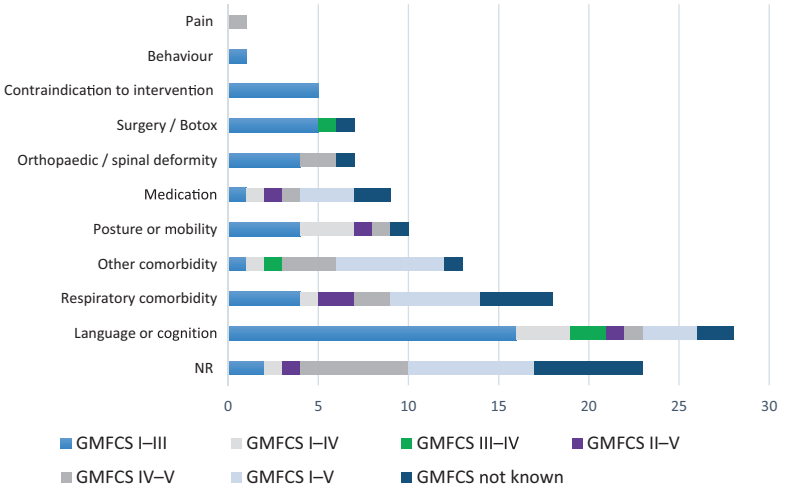

FIGURE 5 Visual presentation of exclusion criteria across reported across reported Gross Motor Function Classification Scale (GMFCS) cohort groups. Abbreviation: QoL, quality of life.

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Across ICF domains, 'Body functions and structures' featured greatest consensus of OMIs. However, findings suggest increased consensus was achieved through recruiting homogenous cohorts with milder forms of CP, excluding those unable to comply with effort-dependent lung function measures such as spirometry. Subsequently, these studies do not inform feasible ways to assess and evaluate those most at risk of respiratory morbidity and mortality.[8] Further research to address this challenge is essential to reduce health care burden and improve quality of life for wider populations with CP.

In contrast, OMIs mapped to 'Participation and quality of life' were implemented across a wide range of cohorts and contexts but the domain was considered to have least consensus. Registered trials of ongoing studies to date will only add to this variability, proposing new measures such as 'days missed from school because of respiratory illness', 'days missed off work by caregivers', and the Beach Center Family Quality of Life survey,[81] the Child Health Utility 9D,[82] the EQ-5D-Y,[82] the Canadian Occupational Performance Measure,[83] and CP CHILD.[83] These ongoing studies also add new 'Activity and performance' OMIs, such as goal attainment scale and advanced technology of activity monitoring devices,[83] adding to the existing challenge of meta-analysis within systematic reviews. Authors call for urgent measurement agreement, as experimental research interest grows, to establish effective management strategies for best practice.

Participant characteristics of motor impairment informed a subquestion for this review because of its evidence-based association with respiratory health.[3-5,9] Many more studies recruited children with milder motor impairments, and those that recruited 'all' levels of motor impairment represented a proportionate sample of more severely impaired children, reflecting wider population distribution estimated at 25.5% to 34.3%.[45] Arguably, research should prioritize recruitment of children classified in GMFCS level V, representing a higher health care burden population with poorer health outcomes.[3,5] However, children with milder CP impairments also remain at risk of poorer respiratory health, presenting with lower lung function and respiratory muscle strength compared to typically developing controls,[84–86] supporting research to address the entire spectrum of this condition.

Findings revealed children with complex comorbidities or those unable to follow instructions were underserved in this review. Although many studies reported phrases such as 'cognitive and language abilities insufficient to fulfil respiratory test'[84–88] in their exclusion criteria, only one study employed an IQ level under 60 threshold,[75] limiting interpretation of how a child's ability to follow instructions was determined. This participant characteristic is important to address in future research, as 1 in 2 children with CP have a learning disability, increasing to two-thirds in those with greater motor impairment.[7] Researchers acknowledge such characteristics increase sample heterogeneity and risk of adverse events during treatment, whilst limiting application of 'criterion standard' measures such as spirometry. However, these exclusions limit application of research to a multimorbidity, high health care burden population, who engage differently with assessment and intervention, and remain most vulnerable to respiratory illnesses.[1,3,7] These exclusions risk perpetuating health and health care disparities, informing public health research priority for underserved populations.[89]

In this review, few research designs featured mixed cohorts of children and adults, despite respiratory illness persisting as a primary cause of death in a growing population of young adults with CP.[90] This is in part due to the eligibility criteria imposed by authors. Extending research beyond the traditional paediatric population presents new challenges such as conducting validated child-focused measures in adult cohorts and applying research into health practice that remains vastly different. However, core literature strives to encompass children and young people up to 25 years, both with CP[1] and chronic neurodisability,[8] prompting authors to propose parallel research exploring respiratory outcomes of young adults, to support challenging transitions to adult health care services.[91]

The review findings suggested a relationship between context and measurer characteristics, highlighting risk of participant burden in research. Objective OMIs were primarily conducted in laboratories or controlled outpatient facilities, measured by clinicians, researchers, with or without a device, introducing participant burden through travel, transportation, lack of access to supportive equipment and familiar networks for participants and their families.[92] In contrast, patient/proxy-reported OMIs were conducted in more familiar environments, such as school or home, greatly reducing barriers to participation. Yet, the self-reporting nature of these measures introduces response error and burden, namely for parents.[93] Caregiver burden is well reported in CP-focused literature,[94] which is heightened in families of low socioeconomic background,[95] and should inform the selection of OMI in research, with efforts to mitigate burden through relevant stakeholder engagement in codesign.

The review findings also revealed significant underrepresentation of the child as the measurer. Factors of age, prevalence of learning and communication impairment in CP, or the outcome domain (e.g. sleep) may have contributed to this lower volume of measures. However, adopting inclusive and flexible methods such as alternative augmented communication or familiar communicators can support and engage children with CP as respondents in data collection measures, aligning with best practice guidelines.[1] Increased use of these methods in future research will promote children's voices, building meaningful and authentic research capacity led by those most affected by respiratory illness.

Incomplete reporting limits full analysis of OMI psychometric properties, and findings are interpreted with caution. Studies implementing validated OMIs (e.g. Paediatric Evaluation of Disability Inventory) often failed to report this in publication.[36] Studies that did report measurement properties presented varied level of detail, ranging from a

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single citation to embedding psychometric property development within their design. Specific measurement development studies examining validity and reliability[34,35] excluded children with significant motor, learning, and respiratory impairment, limiting the application of findings to a population at risk of poorer respiratory health. Further research is required to examine health measurement properties and the quality of underlying developmental studies for OMIs featured in this review, before informing recommendations for use across the population with CP.

Limitations

For this review, it was necessary to undertake a second stage of screening to refine the condition of CP, based upon the vast number of retrieved articles and widely varying neurological presentations. However, the authors acknowledge this is a deviation from the original protocol. The research team selected CP as the exemplar condition, because of its prevalence within chronic neurodisability, its well-defined presentation, and widely established association with respiratory-related morbidity and mortality.[8] However, the authors note children with other chronic neurodisability conditions remain at risk of respiratory illness, encouraging future exploration in a separate review.

The authors acknowledge the vast number of included studies limited the in-depth analysis and quality assessment of individual studies. However, review findings were able to conclude that existing experimental designs were limited in number and size, and lacked consensus of outcomes, aligning with conclusions from robust systematic reviews in the last decade.[19,20] Health measurement properties were not feasible to examine in depth for this scoping review. For example, multiple-item patient-reported outcome measures have not been subject to in-depth content analysis and have been categorized based on their 'dominant' ICF domain. As a result, the review may overestimate the number of OMI's mapped to 'Participation and quality of life'.

Conclusion

This review revealed no consensus in respiratory health outcome domains and their associated instruments in children with CP over the past 50 years. Inconsistency of these outcomes has become a major barrier to determining best practice with regards to early assessment and delivery of effective interventions. Furthermore, the body of research in this review reveals inadequate representation of the full spectrum of CP, presenting a biased evidence base, which could perpetuate ongoing respiratory health and health care disparities in children with more severe forms of CP.

There is an urgent need to establish 'what to measure', prioritizing respiratory health domains that are important to stakeholders. By defining these domains, research can then examine 'how to measure', with an initial focus on feasibility and acceptability in children with CP most vulnerable to respiratory morbidity and mortality. Measurement consensus can inform high-quality inclusive research design and a minimum agreement of outcomes, facilitating comparison and synthesis across studies, in this underserved research population. It can also inform measures of anticipatory monitoring in practice, closer to home, aligning with best practice guidelines set out by other paediatric chronic respiratory diseases.[23–25] These long-term research and practice transformations have potential to determine effective interventions that can be delivered in a timely manner, before irreversible lung damage occurs, minimizing escalation to emergency hospital care, further morbidity, and even mortality.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

The following additional material may be found online:
Appendix S1: JBI systematic review title registration form.
Appendix S2: PRISMA-ScR checklist.
Appendix S3: Search strategy example.
Appendix S4: Data extraction tool template.
Figure S1: PRISMA flow diagram of primary searches.
Figure S2: PRISMA flow chart presenting rerun of searches (24th February 2023).
Table S1: Study characteristics of included studies.
Table S2: Summary of outcome measures.

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