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Fatigue experienced by people with cerebral palsy: a systematic review of assessment tools and decision tree

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ABSTRACT

Purpose: To conduct a systematic review of self- and proxy-report fatigue assessment tools used in studies of people with cerebral palsy (CP) of all ages, and to develop a fatigue assessment tool decision tree for clinicians and researchers.

Materials and methods: Five electronic databases (MEDLINE, PsycInfo, CINAHL, Web of Science and Cochrane) were searched to September 2021 to identify studies assessing self-reported fatigue in people with CP of any age. The assessment tools utilised were extracted and two reviewers appraised the tool characteristics, clinical utility and psychometric properties. A decision tree for selecting fatigue assessment tools was constructed.

Results: Ten assessment tools were identified across thirty-nine studies, three of which are valid and reliable for assessing fatigue severity and impact in people with CP. A four-level fatigue assessment tool decision tree was constructed. No valid and reliable tool for assessing cognitive fatigue was identified; responsiveness has not been evaluated in any tool for people with CP.

Conclusions: Physical fatigue screening and assessment tools for people with CP are available and are presented in our decision tree, however their utility as outcome measures remains unclear. Cognitive fatigue is understudied and poorly understood, further work is required in this area.

> ## IMPLICATIONS FOR REHABILITATION
> - Current measurement tools to screen and assess physical fatigue in people with cerebral palsy (CP) are valid and reliable and are presented in our 4-level decision tree to guide assessment tool selection.
> - The responsiveness of these measurement tools to screen and assess physical fatigue has not been evaluated, therefore their utility as outcome measures in people with CP is unclear.
> - Cognitive fatigue is understudied and poorly understood in people with CP.
> - Valid and reliable tools to assess cognitive fatigue in people with CP are not available.

Introduction

Cerebral palsy (CP) is an umbrella term for a heterogeneous group of neurological disorders caused by a non-progressive injury to the developing infant or foetal brain [1]. Fatigue is a common problem for people with CP with up to 40% of adults with CP reporting higher levels of fatigue than the general population [2,3]. Fatigue is known to adversely affect health-related quality of life, independence in daily activities and functional mobility, and is experienced most frequently by those with CP who have more severe motor impairment (Gross Motor Function Classification System (GMFCS) levels III-V) [4,5]. Despite its significance, the mechanisms underpinning fatigue in people with CP remain relatively poorly understood [6], which limits the development and testing of fatigue prevention strategies.

The literature on self- and proxy-report fatigue in people with CP is characterised by substantial heterogeneity in approaches to assessment. This may be attributed, at least in part, to a lack of consensus on the definition of fatigue, which is a broad construct. Fatigue is commonly described in the literature as a physical experience – for example, feelings of bodily tiredness, lack of energy for physical tasks and local muscle fatigue [7]. Less common are studies that report cognitive or mental fatigue, which presents as excessive cognitive tiredness or exhaustion in response to a demanding task or significant sensory stimulation [8]. Such cognitive fatigue is associated with a disproportionally long recovery time and is defined as an atypical or pathological response to a demanding task [8]. Studies involving people with CP demonstrate that physical and cognitive fatigue can be experienced separately

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or together, and that fatigue can be experienced in isolation or clustered with other problems, commonly depressive symptoms and pain [4]. Fatigue also has a temporal dimension, as it can be acute, for example during or after physical activity, or chronic, where excessive fatigue is experienced over prolonged periods of time without an attributable cause. Currently, there is no consensus on the definition of or criteria for identifying clinically meaningful fatigue in people with CP, which suggests that further work is required to establish cut-off scores for assessment tools.

Before considering interventions to address fatigue in people with CP, greater clarity around its measurement is required as the properties of assessment tools differ depending on their purpose. Beyond understanding its temporality and type, fatigue screening tools and tools for assessing fatigue require strong discriminative properties, whereas evaluative tools for measuring change require stability over time and responsiveness [9].

There is a need for a review of available tools and a summary of evidence in a practical format, to aid assessment tool selection for clinicians and researchers. Decision trees have been used previously to guide reliable and effective selection of assessment tools for people with CP in the domain of high-level motor skills [10], and a fatigue assessment tool decision tree which consolidates information regarding tool purpose, psychometric properties and clinical utility would be a useful addition.

The aim of this study was to conduct a systematic review of self- and proxy-report fatigue assessment tools which have been used in studies of people with CP and to develop a fatigue assessment tool decision tree for clinicians and researchers. The study was designed to identify and examine: (i) the assessment tools used; (ii) the type, domain and timeframe of fatigue measured; (iii) the population in which the measure was developed; (iv) the extent to which the psychometric properties (reliability and validity) of the tools have been evaluated in people with CP and in other clinical populations; and (v) clinical utility. This evaluation will advance understanding of which tools may be suitable for different purposes (screening, assessment, or evaluation), aid fatigue assessment tool selection for clinicians and researchers, and guide future work.

Materials and methods

Search strategy

Initial searches were conducted in January 2022 in five electronic databases: MEDLINE, PsycInfo, CINAHL, Web of Science and Cochrane. Search terms were developed with the support of a research librarian and were tailored to each database. Initial strategies included terms that relate to CP and fatigue. Truncations (e.g., * as in fatigu* to obtain fatigue, fatiguing, fatigued), wildcards (e.g.,? to accommodate for differences between British and American spelling), and proximity operators were also used to ensure the search strategies were comprehensive and robust. Search filters were used to ensure that articles retrieved met the inclusion and exclusion criteria related to date and type of publication, and English language. Secondary searches included screening reference lists of all included articles, and publications by the authors of any included assessment tool. The search process was repeated in February 2023 to identify any newly published studies. The protocol for the review has been published (https://www.medrxiv.org/content/10.1101/2021.07.20.21260898v1).

Study screening and selection process

Following the initial search, the identified literature was screened as follows: (i) search results were imported into COVIDENCE software and duplicates were removed, (ii) title and abstract reviews were carried out by two authors (ID and RE) and conflicts were resolved via discussion, or via a third author (DC) if required, (iii) full text review was carried out by two authors (ID and RE) and conflicts resolved via discussion, or via a third author (DC) if required. Review of papers identified in secondary searches were managed using these same steps.

Inclusion and exclusion criteria

Studies were included in the systematic review if they met the following criteria: (i) original research that purported to measure any type of fatigue by self- or proxy-report in people with CP of any age, (ii) the study was written in English, (iii) the assessment tool was more than 1 item and written in English and (iv) was published between 1980 and 2021. Studies were excluded from the review if: (i) they included participants with CP but where those participants' data were indistinguishable from other neurological disorders; (ii) the publication date was prior to 1980; or (iii) the study was unpublished or identified in grey literature.

Data extraction and assessment of quality

Extracted information comprised the number of participants included in each study, demographic, and clinical characteristics of the sample of people with CP (including age, sex, CP subtype and topographical distribution, Gross Motor Function Classification System level [11], Manual Ability Classification System level [12], Communication Function Classification System level [13], and Eating and Drinking Classification System level [14] where reported), and the tool used to assess fatigue.

For each fatigue assessment tool, the following characteristics were identified: the scale type and number of items; subscales/factors; assessment method; fatigue time frame; and population in which the tool was developed. Tools were examined to determine whether items addressed fatigue in the physical domain and/or cognitive domains (for the purposes of this review "mental fatigue" was categorised as within the cognitive domain), in addition to whether items addressed fatigue severity and/or impact on daily activities.

The quality of each tool was also assessed using previously adopted appraisal methods [15] to establish: tool usability comprising ease of understanding, ease of completion and burden; clinical/research utility comprising responsiveness and scope; and general psychometric properties. The category of psychometric properties in people with CP was also added for the purposes of this review. Two reviewers graded each assessment tool in these areas as follows: 0 = unknown, 1 = poor/emerging, 2 = moderate, 3 = good; and a third reviewer was consulted in the event of conflicts. For all measures, completion time and ease of use were assessed by the author in conjunction with evidence where available.

The assessment tool purpose, domain, time-frame, development population and psychometric properties were considered to construct a decision tree for selecting fatigue assessment tools for people with CP (Supplementary Appendix S1, online supplementary material).

Results

A total of 759 articles were retrieved from the initial searches (Figure 1). Thirty-nine studies met the inclusion criteria, comprising 3172 participants with CP. Mean participant age was <18 years in
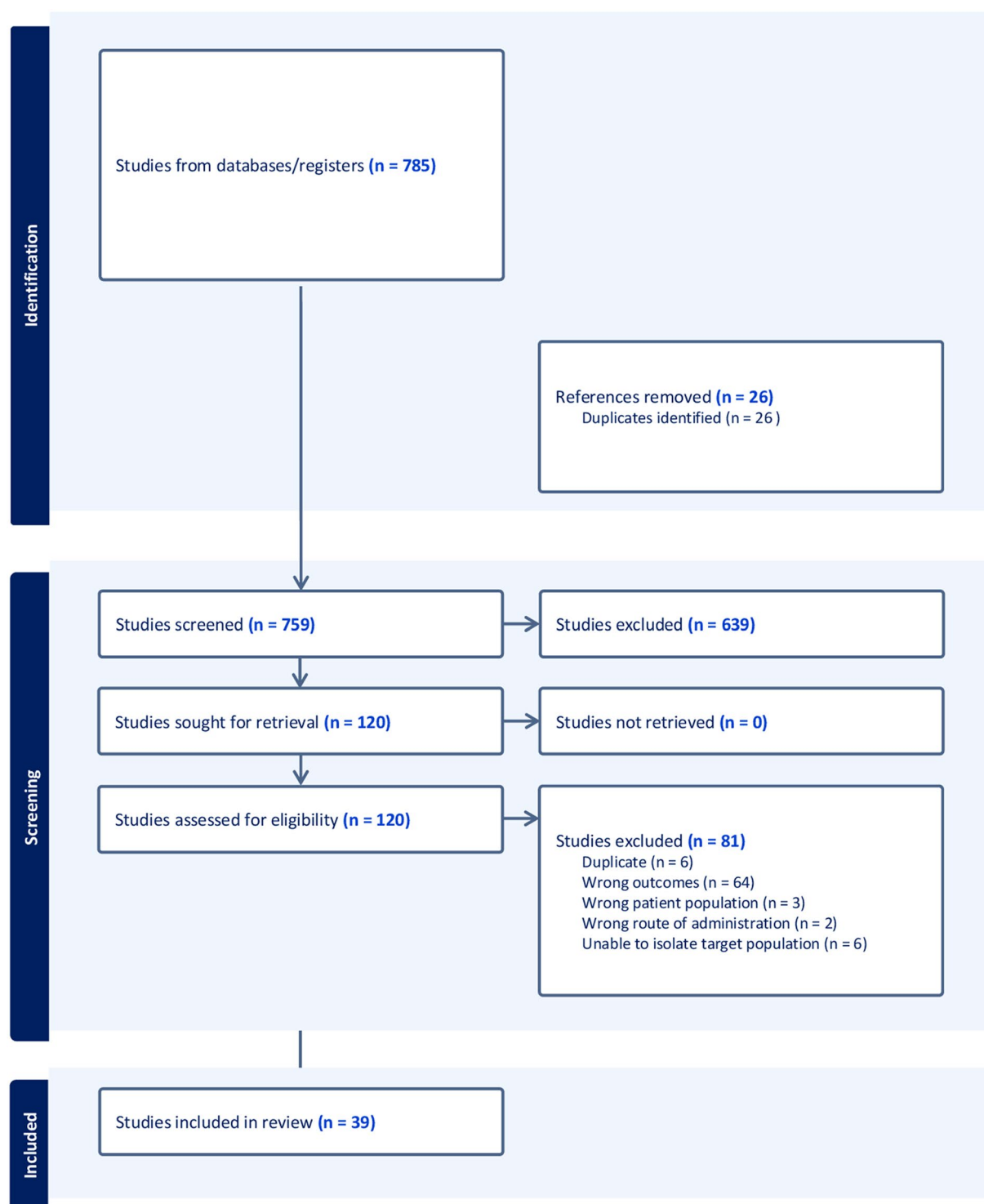
Figure 1. PRISMA flow chart: 759 studies were retrieved from five databases and screened. 120 studies underwent full-text screening. 39 were included.

12 studies; mean participant age was >18 years in 23 studies; and mean participant age was not reported in 4 studies. A list of these studies is available in Supplementary Appendix S2, in addition to the study population and fatigue assessment tool used.

Assessment tools

Ten assessment tools were identified across the 39 studies (Table 1): The Fatigue Severity Scale (FSS) (11 studies) [27], the Fatigue Impact and Severity Self-Assessment (FISSA) (7 studies) [16], the Pediatric Quality of Life Inventory Multidimensional Fatigue Scale (PedsQL MFS) (7 studies) [25], the PedsQL 3.0 Cerebral Palsy Module (PedsQL CPM) (6 studies) [19], the Patient-Reported Outcome Measurement Information System paediatric fatigue profile short form (PROMIS PSF) (4 studies) [21], the Fatigue Questionnaire (FQ) (4 studies) [23], the Checklist Individual Strength fatigue subscale (CIS) (2 studies) [24], the Global Physical Health Scale (GPHS) (2 studies) [26], the Multidimensional Fatigue Inventory (MFI) (1 study) [28], and the Fatigue Assessment Scale (FAS) (1 study) [22].

Table 1. Fatigue assessment tool properties.

Measure	Development population	Self- or proxy-report	Scale type and number of items; estimated completion time	Number of fatigue subscales/factors, as reported by assessment tool; description of dimension	Fatigue time frame	Psychometric properties		Psychometric properties in people with CP	
						Reliability	Validity	Reliability	Validity
Fatigue Impact and Severity Self-Assessment (FISSA)	People with CP N=130 Mean age (SD) = 18.9 (4.5) Median age = 17 yrs Age range = 14–31 yrs Male N=61 Female N=68 GMFCS I N=34 GMFCS II N=39 GMFCS III N=21 GMFCS IV N=18 GMFCS V N=12 Distribution of Involvement: Monoplegia N=6 Hemiplegia N=31 Diplegia N=44 Triplegia N=11 Quadriplegia N=34	Self-report	Likert scale (1–5) 31 items Estimated completion time: 15 mins	2 Factors 1: Impact of fatigue on daily living 2: Management and activity modification	Typical week (7 days)	Unknown	Unknown	Test–retest reliability, intraclass correlation coefficient (ICC) = 0.74 (95% CI 0.53–0.87)[16]	Content validity supported [16]
Pediatric Quality of Life Inventory Multidimensional Fatigue Scale (PedsQL MFS)	Paediatric cancer survivors N=220 child self-report N=337 parent-proxy report Age range = 2–18 yrs	Age-specific self-report and parent-proxy report versions available	Likert scale (1–5) 18 items Estimated completion time: 10 mins	3 subscales 1: General fatigue 2: Sleep/rest fatigue 3: Cognitive fatigue	Last month	Internal consistency, Cronbach's alpha = 0.89 child (child report), 0.92 parent report) [17]	Construct validity supported [17]	Unknown	Unknown
Fatigue Severity Scale (FSS)	People with Multiple Sclerosis N=25 Mean age (SD) = 44.8 (10.0) yrs	Self-report	Likert scale (1–7) 10 items Estimated completion time: 5 mins	Dimensions: Physical fatigue severity and impact	Last week	Internal consistency, Cronbach's alpha = 0.93 Test–retest reliability, intraclass correlation coefficient (ICC) = 0.90 (95% CI 0.81–0.95) [18]	Discriminant, Construct and Discriminative validity supported [18]	Unknown	Unknown
PedsQL 3.0 Cerebral Palsy Module (PedsQL CPM)	Children with CP and their families N=245 Mean age (SD) = 8.1 (4.4) yrs Age range 2–18 yrs Male N=134 Female N=107 GMFCS I N=28 GMFCS II N=40 GMFCS III N=86 GMFCS IV N=43 GMFCS V N=37	Age-specific self-report and parent-proxy report versions available	Likert scale (0–4) 4 items Estimated completion time: 2 mins	Dimensions: Impact of physical fatigue	Last month	Unknown	Unknown	Internal consistency, Cronbach's alpha = 0.79 (child report), 0.91 (parent report) [19]	Construct validity supported [19]

(Continued)

Table 1. (Continued)

Measure	Development population	Self- or proxy-report	Scale type and number of items; estimated completion time	Number of fatigue subscales/factors, as reported by assessment tool; description of dimension	Fatigue time frame	Psychometric properties		Psychometric properties in people with CP	
						Reliability	Validity	Reliability	Validity
Patient-Reported Outcome Measurement Information System (PROMIS) paediatric fatigue profile short form	Healthy children N=3048 Age 8–12 N=1616 Age 12–17 N=1426 Male N=1470 Female N=1578	Self-report	Likert scale (1–5) 10 items Estimated completion time: 5 mins	Dimensions: Impact and severity of physical and cognitive fatigue	Last 7 days	Internal consistency, Cronbach's alpha = 0.80 [20]	Convergent validity supported [20]	Unknown	Limited differential item functioning (range of difference = 0–0.17) [21]
Fatigue Assessment Scale (FAS)	Healthy adults N=351 Mean age (SD) males = 45 (8.4) yrs Mean age (SD) females = 43 (9.5) yrs Male N=183 Female N=166	Self-report	Likert scale (1–5) 10 items Estimated completion time: 5 mins	Dimensions: Impact and severity of physical and cognitive fatigue	"How you usually feel"	Internal consistency, Cronbach's alpha = 0.90 [22]	Convergent validity supported [22]	Unknown	Unknown
Fatigue Questionnaire (FQ)	Healthy adults N=274 Age range: 18–45 yrs	Self-report	Likert scale (1–4) 11 items Estimated completion time: 5 mins	Dimensions: Severity of physical and cognitive fatigue	Last month	Internal consistency, Cronbach's alpha = 0.88–0.90 [23]	Face validity and construct validity supported [23]	Unknown	Unknown
Checklist Individual Strength (CIS) (fatigue subscale only)	Healthy population N=2288	Self-report	Likert scale (1–7) 20 items Estimated completion time: 10 mins	Dimensions: Severity of physical and cognitive fatigue	Last 2 weeks	Internal consistency, Cronbach's alpha = 0.84–0.95 Test-retest r=0.74–0.86 [24]	Construct validity supported [24]	Unknown	Unknown
Multidimensional Fatigue Inventory (MFI)	Young adults with and without chronic health conditions N=423 Male N=183 Female N=166 Mean age: 20.0 (1.3) yrs Age range 18–25 yrs With chronic health condition N=32 Without chronic health condition N=391	Self-report	Likert scale 1–5 20 items Estimated completion time: 10 mins	5 subscales 1: General fatigue 2: Physical fatigue 3: Reduced motivation 4: Reduced activity 5: Mental fatigue	Lately (i.e., How you have been feeling lately)	Internal consistency, Cronbach's alpha = 0.53–0.93 [25]	Convergent validity supported [25]	Unknown	Unknown
Global Physical Health Scale (GPHS)	Children with CP N=91 Mean age (SD): 9.9 (4.1) yrs Age range: 2–20 yrs Male N=52 Female N=39 GMFCS I N=14 GMFCS II N=15 GMFCS III N=32 GMFCS IV N=13 GMFCS V N=12	Parent-proxy report only	Likert scale 1–5 17 items Estimated completion time: 10 mins	Dimensions: Physical fatigue severity and impact	Typical behaviour	Unknown	Unknown	Test–retest reliability, intraclass correlation coefficient = 0.94 (CI 0.84–0.98) [26]	Construct validity supported [26]

CP: cerebral palsy; GMFCS: Gross Motor Function Classification System.

Scale domains and type

The PedsQL MFS, FAS, FQ and CIS comprehensively assessed physical and cognitive domains of fatigue, and the PROMIS, FSS and MFI assessed some dimensions of physical and cognitive fatigue. The remaining three tools assessed physical fatigue only. The FISSA, MFI and GPHS tools comprehensively assessed fatigue severity and impact. The PedsQL MFS, PROMIS and FAS assessed some dimensions of fatigue severity and impact, and the remaining four tools assessed fatigue severity only. All assessment tools used Likert scales (4-point $n=2$; 5-point $n=6$; 7-point $n=2$).

Time frame

The FSS and PROMIS assessed fatigue in the last week or 7 days while the FISSA assessed fatigue in a typical week. The CIS assessed fatigue in the last 2 weeks. The PedsQL MFS, PedsQL CPM and FQ assessed fatigue in the last one month. Others were less specific. The MFI assessed fatigue based on how the participant had been feeling "lately", the FAS and GPHS tools assessed fatigue based on how the participant "usually or typically feels".

Population in which assessment tools were developed

The PedsQL CPM and GPHS were developed for children and adolescents with CP aged 2–18 years. The FISSA was developed for adolescents and young adults with CP aged 14-31 years. The FSS and PedsQL MFS were developed for populations with other health conditions (paediatric cancer survivors and adults aged >18 years with multiple sclerosis, respectively). The MFI was developed for a population of young adults with and without chronic health conditions, aged 18-25 years. The PROMIS was developed for healthy children and adolescents aged 0–18 years; and the FAS, FQ and CIS were developed for healthy adults aged >18 years.

Self- and proxy report

The GPHS is a parent-proxy assessment tool only. The PedsQL MFS and PedsQL CPM have both age-specific self-report and parent-proxy report versions available. All other identified tools are self-report only,

Psychometric properties

Psychometric evaluation has been conducted with people with CP for three of the 10 identified fatigue assessment tools. The GPHS has strong internal consistency (ICC = 0.94) for people with CP [26]. The PedsQL CPM has good internal consistency (Cronbach's alpha = 0.79) [19], and the FISSA has moderate internal consistency (ICC = 0.74) [16]. Construct validity for people with CP is also supported for the PedsQL CPM [19] and GPHS [26], and content validity is supported for the FISSA for people with CP [16]. These assessment tools were found to have robust psychometric properties. The validity of the PROMIS has been evaluated for people with CP and is not strongly supported [21]. The psychometric properties of the PedsQL MFS, MFI, FSS, FAS, FQ and CIS have not been evaluated for people with CP, though these tools have been found to be valid and reliable in other clinical populations without neurological conditions (Table 1).

Table 2. Fatigue assessment tool appraisal, adapted from Whitehead, 2009 [15].

Measure	Scale usability		Clinical/research utility			
	Ease of understanding, ease to complete, burden	Assesses severity and/or impact (severity only = 1, dimensions of severity and impact = 2, severity and impact = 3)	Assesses physical and/or cognitive fatigue domains (physical only = 1, dimensions of physical and cognitive = 2, physical and cognitive = 3)	Evidence of robust psychometric properties in people with CP	Evidence of ability to act as an outcome measure responsive to change with disease progression or treatment	Evidence of ability to act as an outcome measure responsive to change with disease progression or treatment in people with CP
Fatigue Impact and Severity Self-Assessment (FISSA)	2	3	1	3	0	0
Pediatric Quality of Life Inventory (PedsQL) Multidimensional Fatigue Scale.	3	2	3	0	3	0
Fatigue Severity Scale (FSS)	3	1	2	2	3	0
PedsQL 3.0 Cerebral Palsy Module (PedsQL CPM)	3	1	1	3	0	0
Patient-Reported Outcome Measurement Information System (PROMIS) paediatric fatigue profile short form	3	2	2	0	3	0
Fatigue Assessment Scale (FAS)	3	2	3	0	1	0
Fatigue Questionnaire (FQ)	3	1	3	0	1	0
Checklist Individual Strength (CIS)(fatigue subscale only)	3	1	3	0	2	0
Multidimensional Fatigue Inventory (MFI)	3	3	2	0	2	0
Global Physical Health Scale (GPHS)	3	3	1	3	0	0

Grading as follows: 0 = unknown, 1 = poor/emerging, 2 = moderate, 3 = good. If reliability/validity were not reported in people with CP, a score of 0 was given. For all measures, completion time and ease of use were assessed by the author in conjunction with available evidence where available.

Scale usability

Scale usability was rated highly for all but one assessment tool (FISSA), which was scored as moderate (Table 2). Assessment tools were easy to understand. Burden was low for all tools and the number of items ranged from 4 to 31. Time to completion was poorly reported, however expected time frames ranged from <2 min to approximately 15 min. This time may vary greatly for people with CP who have associated impairments of cognition or communication.

Clinical/research utility

The PedsQL MFS, PROMIS and FSS rated highly for responsiveness to change when used with other clinical populations, the CIS and MFI assessment tools were rated as moderate, and the remaining five tools rated poorly. In none of the identified assessment tools had responsiveness to change for people with CP been evaluated.

Decision tree

Appraisal of the identified assessment tools informed the construction of a 4-level fatigue assessment decision tree (Supplementary Appendix S1). The four levels considered were: (1) the dimension of fatigue to be measured (physical, cognitive or both); (2) the components measured (fatigue severity, impact on daily activities or both); (3) clinical utility and (4) psychometric properties in people with CP.

Discussion

This systematic review identified 39 studies and 10 assessment tools that have been used to measure self- or proxy-report fatigue in people with CP. While 3 of the 10 identified assessment tools are valid and reliable for assessing the severity and impact of physical fatigue experienced by people with CP, no valid and reliable tools were identified to assess cognitive fatigue in this population. Responsiveness to change for people with CP has also not been evaluated for any of the identified tools, and there was also substantial heterogeneity in the populations for which the assessment tools were developed and the timeframes of assessment. Given the putative prevalence and significance of fatigue experienced by people with CP [2,5], these findings are surprising, but signal clear directions for future research.

The PedsQL CPM, GPHS and FISSA are valid and reliable for use in descriptive assessment of physical fatigue for people with CP. Each of these tools has robust psychometric properties and is suitable for screening and assessment purposes. However, it is important that they are used for this specific purpose as their responsiveness to change has not been evaluated. As such, their clinical and research utility as evaluative outcome measures remains unclear. The developers of the FISSA have discussed their interest in future analysis of the extent to which this tool is appropriate for evaluative use, which would be an important contribution [29].

Disappointingly, valid and reliable measures to assess cognitive fatigue in people with CP are not available, which may explain why this experience remains poorly understood. One comparative study reported no difference in the level of cognitive fatigue between adults with CP and the general population [2], suggesting that fatigue in CP may be primarily physical in origin. However, in that comparative study, cognitive fatigue was assessed using the mental fatigue subscale of the FQ which comprises only 6 items, and for which the reliability and validity have not been evaluated for people with CP. These findings should therefore be interpreted cautiously and the potential for cognitive fatigue to be a common, even debilitating problem for people with CP cannot be discounted until further work is undertaken. In fact, people with CP have qualitatively described the presence and effect of cognitive fatigue on daily tasks [30]. A promising solution may be the modified Mental Fatigue Scale (MFS) [8], which is yet to be utilised with people with CP beyond psychometric evaluation and was therefore not included in this review. This scale proposes a cut-off score for 'problematic fatigue' and demonstrated construct validity with a small sample of people with CP (n = 10) [8]. Interestingly, 8 out of the 10 participants with CP who participated in this validation study reported levels of cognitive fatigue higher than for the proposed cut-off for "problematic fatigue", even though the presence of fatigue was not an inclusion criterion [8]. Further evaluation of the MFS is required with larger samples to further understand reliability and responsiveness to change.

Three tools that had been developed in people with CP were identified: the GPHS, PedsQL CPM and the FISSA. There was substantial heterogeneity in the age and impairment levels of the development populations. The GPHS and PedsQL CPM were both developed for younger people with CP (2–20 years and 2–18 years, respectively) [31]. The GPHS is a parent-proxy report assessment tool only, whereas there are both age-specific self-report and parent-proxy report versions of the PedsQL CPM. While it is recommended that children with CP should self-report on health-related constructs wherever possible [32], these tools offer promising insights into select domains of fatigue for children with CP who are unable to self-report. The FISSA was developed for adolescents and adults with CP aged 14–31 years. While all scales rated highly for usability, without evaluation, it cannot be assumed that these assessment tools will be age-appropriate across such a wide age range. Furthermore, all three development populations included the full spectrum of people with CP (GMFCS I–V). Within this range there is significant heterogeneity in typical physical activity levels, motor impairment distribution and severity and prevalence of other health-related factors [33] which may influence fatigue (e.g., pain, depression, medication). Given the variety of factors which are likely to influence fatigue across the spectrum of people with CP, further evaluation of assessment tools by age or impairment severity is indicated.

There was also heterogeneity in the timeframes assessed – ranging from the last week to the last month. The lack of specificity in certain timeframes (e.g., "usually" or "lately") permits only an indication of whether fatigue is a commonly experienced problem in daily life, and precludes understanding of whether fatigue symptoms are acute or chronic, or may be influenced by other factors. Rate of fatigue onset or recovery is not comprehensively assessed by the identified tools. One recently published study advocates for real-time monitoring of fatigue, which may account for these issues, and would eradicate the need for accurate recall and the potentially difficult task of averaging perceived fatigue over time [34]. This concept warrants further evaluation and, overall, greater attention to the temporal dimension of fatigue is needed to guide assessment and the development of management strategies.

Finally, it is known that people with CP who have significant physical impairments (GMFCS IV and V) are most likely to experience problematic fatigue [4] and the prevalence of comorbidities, including intellectual disability, is significantly higher for this group [35]. However, it is unclear whether the available tools may be

valid for people with CP who have intellectual disability, who were not included in samples used for psychometric evaluation of the PedsQL CPM, GPHS or FISSA. The prevalence of impairment of vision, hearing or communication is also significantly higher in people at GMFCS level IV and V [36], and the adapted use of fatigue assessment tools for people with complex communication needs also warrants further evaluation. This again brings importance to proxy-reporting. Several identified assessment tools in particular are proxy-report (GPHS) or have proxy-report versions available (PedsQL CPM and PedsQL MFS), however all were developed for the purposes of assessing fatigue in children unable to self-report. None were developed for the purposes of proxy-reporting in adults with intellectual disability or other comorbidities preventing self-report, and this is an area requiring further development.

This review has two main limitations. First, the scope was limited to self- and proxy-reported fatigue and therefore studies objectively measuring physical fatigue were excluded. It is possible that a combination of self-report and objective measurement may provide a broader understanding of fatigue, however this methodology appears to be rare. Second, the review is unable to systematically address the complex relationship between fatigue and other concepts (e.g., pain, wellbeing, activity, physical fitness) due to the heterogeneity in assessment methods. Studies evaluating the interaction between fatigue and other concepts, using robust tools and considering physical and cognitive dimensions of fatigue, and will enable this analysis in the future.

Conclusion

Fatigue is a widely recognised problem for many people with CP, particularly those with moderate to severe physical impairments. We have reviewed the available self- and proxy-report fatigue assessment tools, developed a decision tree to aid tool selection for clinicians and researchers, and identified areas for development. The FISSA, GPHS and PedsQL CPM are suitable tools for screening and descriptive assessment of physical fatigue in people with CP, however reliable and valid tools for assessment of cognitive fatigue are not available. Future work is also required to determine the responsiveness to change of currently available tools to establish their clinical and research utility as outcome measures. Further evaluation by age or impairment severity is indicated, as is evaluation of the adapted use of fatigue assessment tools for people with complex communication needs or intellectual disability. Further research will advance understanding of the experience of fatigue for people with CP which will aid the development and evaluation of interventions.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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Data availability statement

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

References

[1] Bax M, Goldstein M, Rosenbaum P, et al. Proposed definition and classification of cerebral palsy, april 2005. Dev Med Child Neurol. 2005;47(8):571–576.

[2] Jahnsen R, Villien L, Stanghelle JK, et al. Fatigue in adults with cerebral palsy in Norway compared with the general population. Dev Med Child Neurol. 2003;45(5):296–303.

[3] Jacobson DNO, Löwing K, Tedroff K. Health-related quality of life, pain, and fatigue in young adults with cerebral palsy. Dev Med Child Neurol. 2020;62(3):372–378.

[4] Van Der Slot WMA, Nieuwenhuijsen C, Van Den Berg-Emons RJG, et al. Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy. Dev Med Child Neurol. 2012;54(9):836–842.

[5] Brunton LK. Descriptive report of the impact of fatigue and current management strategies in cerebral palsy. Pediatr Phys Ther. 2018;30(2):135–141.

[6] Puce L, Pallecchi I, Chamari K, et al. Systematic review of fatigue in individuals with cerebral palsy. Front Hum Neurosci. 2021;15(March):1–16.

[7] Brunton LK, Rice CL. Fatigue in cerebral palsy: a critical review. Developmental Neurorehabilitation. 2012;15(1):54–62.

[8] Bergqvist L, Öhrvall A-M, Rönnbäck L, et al. Evidence of construct validity for the modified mental fatigue scale when used in persons with cerebral palsy. Dev Neurorehabil. 2020;23(4):240–252.

[9] Mokkink LB, Terwee CB, Patrick DL, et al. The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: an international Delphi study. Qual Life Res. 2010;19(4):539–549.

[10] Clutterbuck GL, Auld ML, Johnston LM. High-level motor skills assessment for ambulant children with cerebral palsy: a systematic review and decision tree. Dev Med Child Neurol. 2020;62(6):693–699.

[11] Palisano RJ, Rosenbaum P, Bartlett D, et al. Content validity of the expanded and revised gross motor function classification system. Dev Med Child Neurol. 2008;50(10):744–750.

[12] Eliasson A-C, Krumlinde-Sundholm L, Rösblad B, et al. The manual ability classification system (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. Dev Med Child Neurol. 2006;48(7):549–554.

[13] Inter-rater reliability of the communication function classification system (CFCS) for adults and adolescents with cerebral palsy. Dev Med Child Neurol. 2016;58:87–87.

[14] Tschirren L, Bauer S, Hanser C, et al. The eating and drinking ability classification system: concurrent validity and reliability in children with cerebral palsy. Dev Med Child Neurol. 2018;60(6):611–617.

[15] Whitehead LPMN The measurement of fatigue in chronic illness: a systematic review of unidimensional and multidimensional fatigue measures. J Pain Symptom Manage. 2009;37(1):107–128.

[16] Brunton LK, Bartlett DJ. Construction and validation of the fatigue impact and severity self-assessment for youth and young adults with cerebral palsy. Dev Neurorehabil. 2017;20(5):274–279.

[17] Varni JW, Burwinkle TM, Katz ER, et al. The PedsQL™ in pediatric cancer: reliability and validity of the pediatric quality of life inventory™ generic core scales, multidimensional fatigue scale, and cancer module. Cancer. 2002;94(7):2090–2106.

[18] Rossi D, Galant LH, Marroni CA. Psychometric property of fatigue severity scale and correlation with depression and quality of life in cirrhotics. Arq Gastroenterol. 2017;54(4):344–348.

[19] Varni JW, Burwinkle TM, Berrin SJ, et al. The PedsQL in pediatric cerebral palsy: reliability, validity, and sensitivity of the generic core scales and cerebral palsy module. Dev Med Child Neurol. 2006;48(6):442–449.

[20] Lai J-S, Stucky BD, Thissen D, et al. Development and psychometric properties of the PROMIS pediatric fatigue item banks. Qual Life Res. 2013;22(9):2417–2427.

[21] Coster WJ, Ni P, Slavin MD, et al. Differential item functioning in the patient reported outcomes measurement information system pediatric short forms in a sample of children and adolescents with cerebral palsy. Dev Med Child Neurol. 2016;58(11):1132–1138.

[22] Michielsen HJ, De Vries J, Van Heck GL. Psychometric qualities of a brief self-rated fatigue measure: the fatigue assessment scale. J Psychosom Res. 2003;54(4):345–352.

[23] Chalder T, Berelowitz G, Pawlikowska T, et al. Development of a fatigue scale. J Psychosom Res. 1993;37(2):147–153.

[24] Worm-Smeitink M, Gielissen M, Bloot L, et al. The assessment of fatigue: psychometric qualities and norms for the checklist individual strength. J Psychosom Res. 2017;98:40–46.

[25] Varni JW, Limbers CA. The pedsql™ multidimensional fatigue scale in young adults: feasibility, reliability and validity in a university student population. Qual Life Res. 2008;17(1):105–114.

[26] Haley SM, Chafetz RS, Tian F, et al. Validity and reliability of physical functioning computer-adaptive tests for children with cerebral palsy. J Pediatr Orthop. 2010;30(1):71–75.

[27] Krupp LB, LaRocca NG, Muir-Nash J, et al. The fatigue severity scale: application to patients with multiple sclerosis and systemic lupus erythematosus. Arch Neurol. 1989;46(10):1121–1123.

[28] Smets EM, Garssen B, Bonke B, et al. The multidimensional fatigue inventory (MFI) psychometric qualities of an instrument to assess fatigue. J Psychosom Res. 1995;39(3):315–325.

[29] Brunton LK, Bartlett DJ. Profiles of fatigue severity and variability among adolescents and young adults with cerebral palsy. Fatigue. 2017;5(1):5–14.

[30] Sandström K. The lived body—experiences from adults with cerebral palsy. Clin Rehabil. 2007;21(5):432–441.

[31] Sawyer SM, Drew S, Yeo MS, et al. Adolescents with a chronic condition: challenges living, challenges treating. Lancet. 2007;369(9571):1481–1489.

[32] Varni JW, Burwinkle TM, Sherman SA, et al. Health-related quality of life of children and adolescents with cerebral palsy: hearing the voices of the children. Dev Med Child Neurol. 2005;47(9):592–597.

[33] Reid SM, Carlin JB, Reddihough DS. Using the gross motor function classification system to describe patterns of motor severity in cerebral palsy. Dev Med Child Neurol. 2011;53(11):1007–1012.

[34] Verschuren O, Benner JL, Balemans ACJ, et al. Real-time daily fatigue, sleep, physical activity, and health-related fitness in adults with cerebral palsy. Dev Med Child Neurol. 2022;65(4):509–516.

[35] Compagnone E, Maniglio J, Camposeo S, et al. Functional classifications for cerebral palsy: correlations between the gross motor function classification system (GMFCS), the manual ability classification system (MACS) and the communication function classification system (CFCS). Res Dev Disabil. 2014;35(11):2651–2657.

[36] Shevell MI, Dagenais L, Hall N, REPACQ Consortium Comorbidities in cerebral palsy and their relationship to neurologic subtype and GMFCS level. Neurology. 2009;72(24):2090–2096.