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Health service use among adults with cerebral palsy: a mixed-methods systematic review

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ABBREVIATIONS

ANM	Andersen and Newman Model
JBI	Joanna Briggs Institute

AIM To determine the proportion of adults with cerebral palsy (CP) using health services and frequency of use, and to explore experiences and perceptions of health services for this population.

METHOD A mixed-methods systematic review was conducted using the Joanna Briggs Institute methodology. Five databases were searched to September 2020. Observational and qualitative studies were included. Two reviewers screened titles, abstracts, and full texts; extracted data; and assessed the quality of included studies. Separate meta-analyses were used to pool the proportion of adults using each service and frequency of use. A meta-aggregation approach was used to synthesize qualitative data. Quantitative and qualitative findings were integrated using the Andersen and Newman Model of health care utilization.

RESULTS Fifty-seven studies (31 quantitative, 26 qualitative) of 14 300 adults with CP were included. The proportion of adults using services ranged from 7% (95% confidence interval [CI]: 2–13%) for urologists to 84% (95% CI: 78–90%) for general practitioners. Incidence of visits ranged from 67 (95% CI: 37–123) hospital admissions to 404 (95% CI: 175–934) general practitioner visits per 100 person-years. Qualitative themes highlighted issues regarding accessibility, caregivers' involvement, health workers' expertise, unmet ageing needs, transition, and health system challenges.

INTERPRETATION Adults with CP used a wide range of health services but faced context-specific challenges in accessing required care. Appropriate service delivery models for adults with CP are required. This review emphasizes a need to develop an appropriate service model for adults with CP to meet their needs.

Cerebral palsy (CP) is characterized by atypical motor function and a range of associated impairments.[1] The incidence of CP is approximately 1.4 to 3.3 per 1000 live births.[2–4] Approximately 90% of children with CP survive to at least 19 years of age, meaning many people living with CP are adults.[5] Adults with CP experience increased risk of non-communicable disease compared to those without CP.[6] Many adults with CP also report fatigue, falls, pain, and mobility decline.[7,8] They also have an ongoing need for medical, rehabilitation, and support services.[9] However, most health services that specialize in CP are provided to children and not adults.[10]

Although adults with disabilities report difficulties accessing appropriate health services to meet their needs,[11] no systematic review of health service use among adults with CP has been conducted. This review aimed to (1) determine the proportion of adults with CP using health services and the frequency of use; (2) examine the factors relating to the environment, population, and outcomes that are associated with health service use; and (3) explore the experiences and perceptions of health services for adults with CP. The findings from this review, in combination with the lived experiences of adults with CP, may be useful in developing services for this population.

METHOD

This mixed-method systematic review was conducted following the Joanna Briggs Institute (JBI) guidelines.[12] The JBI is an internationally recognized research organization that aims to improve health outcomes by supporting use of best available evidence in health care.[13] The JBI guidelines for systematic reviews facilitates knowledge-synthesis for a broad range of research questions and study designs in health care practices.[12] This review was conducted using

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the JBI guidelines for mixed-methods reviews.[14,15] The review protocol was published[16] and registered in PROSPERO (registration number: CRD42020155380). Findings are reported according to the PRISMA,[17] Meta-Analysis of Observational Studies in Epidemiology,[18] and Enhancing Transparency in Reporting the Synthesis of Qualitative research guidelines.[19]

Study eligibility

We included studies published in English that examined health service use among adults with CP (≥18 y), or experiences and perceptions of health services for adults with CP from the perspective of adults with CP, caregivers, and service providers. Eligibility criteria are detailed in the protocol[16] and Appendix S1 (online supporting information).

Search strategy

Electronic databases (MEDLINE [Ovid], CINAHL, Embase, PsycINFO, and Cochrane Library) were searched from inception to September 2020, using keywords and subject headings relating to health services and CP (Appendix S2, online supporting information). No filters on study design, settings, or date were applied. Reference lists of included studies were searched for eligible publications.

After removal of duplicates, two reviewers (MM and JR) independently screened titles and abstracts. Where studies met the inclusion criteria, or where eligibility was unclear, full texts were retrieved and assessed independently by two reviewers. Disagreements were resolved through discussion between the reviewers or with a third reviewer.

Data extraction

Two reviewers independently extracted data from included quantitative and qualitative studies using a modified, piloted JBI data extraction tool. For quantitative studies, data extracted included participant characteristics (e.g. age, sex, motor ability), study characteristics (e.g. design, country, sample size), number of people using each health service, number of visits to each service, and factors associated with health service use.

For qualitative studies, data extracted were participant characteristics for adults with CP, caregivers, and/or service providers; study characteristics (e.g. phenomenon of interest, study design, country); and findings supported by direct quotes from study participants. Disagreements were resolved through discussion between the reviewers or with a third reviewer. Authors of six studies were contacted for further data.

JBI guidelines recommended using JBI software,[20] however the software facilitated data extraction by a single reviewer only. We conducted data extraction in MS Excel to allow data extraction by two reviewers.

Quality assessment

Included studies were independently appraised by two reviewers using the JBI Critical appraisal checklist (for each of the respective types of studies).[21,22] Disagreements were resolved through team discussion. As described in the protocol, no studies were excluded based on methodological quality.[16]

What this paper adds

- The quantitative findings indicate considerable variation in the proportion of adults using services, with a relatively low proportion of adults using certain services.
- The qualitative findings highlight the challenges experienced by adults when attempting to access and using health services.
- The integrated findings highlight the influence of environment, population, and outcome factors on health service use among adults with cerebral palsy.

Quantitative synthesis

Separate random-effects meta-analyses were conducted to pool data on the proportion of adults using each health service at least once over a 12-month period. We included studies that examined service use over 12 months in meta-analyses because 12 months was the most common time-period reported. We also conducted separate random-effects meta-analyses to pool data on the incidence rate of visits for each service, where incidence rate was number of visits divided by total person-years. We only conducted a meta-analysis if data from two or more studies were available. We assessed statistical heterogeneity using the I^2 statistic and χ^2 test.

We had to manipulate data from four included studies, relating to the proportion of adults using general practitioner, dentist, physiotherapy, occupational therapy, and speech and language therapy, in order to include it in meta-analyses.[23–26] For example, it was necessary to combine the number of individuals reporting that they used a service 'weekly', 'monthly', 'quarterly', 'half-yearly', or 'yearly' in order to identify the proportion using a service over 12 months.[24] Two studies reported adults had 'regular contact' with a service,[26] or visited a service 'occasionally/varies, occasionally to several times a week'.[23] We assumed these adults visited the service at least once over 12 months and included them in meta-analyses. However, we undertook a sensitivity analysis, by conducting the meta-analyses after excluding data from these two studies. The sensitivity analysis resulted in similar findings, with confidence intervals including the estimates obtained from the primary analysis. Details of the data that were included in meta-analyses, including description of any assumptions are available online (https://zenodo.org/record/4730672#.YWAbdtrMJPY).

All analyses were conducted using STATA IC (version 16; Statacorp, College Station, TX, USA). Data not included in meta-analyses are reported descriptively.

Qualitative synthesis

A meta-aggregation approach was used to synthesize qualitative data, informed by JBI guidelines and thematic synthesis.[27,28] The JBI meta-aggregation involves three stages which include identifying the findings, grouping into categories, and synthesizing findings.[27] However, we combined

traditional thematic synthesis (by coding, developing descriptive themes, and generating analytic themes) with the JBI guideline for meta-aggregation for robust synthesis of qualitative data.[27,28] Therefore our qualitative synthesis (conducted using N-Vivo Pro version 12; QSR international, USA) consisted of three steps using N-Vivo software. First, initial findings were independently extracted and coded by two reviewers. The two reviewers independently developed a codebook from 10 studies. Codes were compared and merged on common language within each study and across the studies. Agreed codes were then applied to all included studies and additional codes emerging from the remaining studies were identified. Codes were discussed and disagreements resolved with a third reviewer. Second, all codes and data were reread by one reviewer (MM) who categorized the codes based on similarity of meaning. Categories were discussed and agreed by the review team.

Finally, agreed categories were analysed and compared to understand relationships between them. A descriptive and analytical memo for each category was developed by one reviewer (MM), which included definitions, categories, summary of data, and deviant cases. The team derived over-arching analytical themes through discussing memos. These analytical themes form a comprehensive set of synthesized findings.

Integration

Quantitative and qualitative findings were integrated using a convergent segregated approach.[14] A convergent segregated approach involves synthesis of quantitative and qualitative findings separately, followed by integration of both sets of findings.[15] This methodology provides greater insights and preserves the integrity of both sets of findings.[15] We chose to narratively integrate quantitative and qualitative data at the interpretation level using the Andersen and Newman Model (ANM) of health care utilization,[29] as this provides a structure, context, and understanding of factors and relationships in health service utilization among adults with CP. The ANM is one of the most commonly used models in the health service use literature, and has been adapted and reviewed extensively over the years to understand health service use among various populations.[30] The ANM proposes relationships between factors relating to the environment, population, health behaviour, and outcomes, to health service use (Appendix S3, online supporting information).[16] We mapped the quantitative and qualitative findings to the ANM, and identified relationships between and within quantitative and qualitative findings.

Public and patient involvement

Adults with CP and service providers supported interpretation of the findings. This was achieved by presenting the findings to the two groups of adults with CP and health professionals respectively, and obtaining their interpretation of the key findings and clinical implications. Their interpretation informed the discussion of this paper. They will be involved in dissemination of findings.

RESULTS

Search results

After removal of duplicates, 18 893 titles and abstracts were screened, and 18 774 records were excluded. After screening 119 full-text articles, 57 published studies were included (Fig. S1, online supporting information). Larivière-Bastien et al. reported findings obtained from the same sample,[31,32] as did Young et al.[33,34] Therefore, we only describe participant characteristics of the samples included in Larivière-Bastien et al.[31] and Young et al,[34] and not Larivière-Bastien et al.[32] and Young et al.[33] to avoid duplication.

Included studies

Of the included 57 studies, 31 used quantitative methods and 26 used qualitative methods. Characteristics of included studies are reported in Tables 1 and 2.

Design

Of the quantitative studies, 27 were cross-sectional,[9,10,23–26,34–54] three were case–control,[33,55,56] and one was a cohort study.[57] Of the qualitative studies, four used a phenomenological approach,[58–61] two used grounded theory,[62,63] two used a narrative approach,[64,65] five reported descriptive qualitative design,[66–70] and 13 did not report study design.[11,31,32,71–80] Nineteen qualitative studies reported the views of adults with CP,[11,31,32,58,60–64,66,68,70,71,73,74,77–80] four reported views of caregivers,[59,65,72,75] one reported both caregivers' and adults' views,[67] one focused on service providers' views,[76] and one reported adults', caregivers' and service providers' views.[69]

Setting

Studies were conducted in the USA,[11,39–46,57–60,62,66–68,78,80] Canada,[32,33,48,61,64,69,72,73,79] Australia,[24,50,54,65,71,74–77] the UK,[10,25,26,37,47,53,63] France,[9,35,49] the Netherlands,[51,52] New Zealand, Republic of Korea, Singapore, Sweden, Denmark, and Taiwan.[23,36,38,55,56,70]

Participants

Studies included 14 300 people with CP. Seven studies included participants under 18 years but were included as the mean age was greater than 18 years,[25,52,55,63] or data were extracted for those aged 18 years or older only.[10,37,50] Where reported, the mean age of participants ranged from 18 years[59] to 48 years 6 months.[78]

The percentage of females ranged from 33%[53] to 100%.[44,45] Studies included adults with spastic CP,[9,23,26,36,40,45,47,50,61,63] ataxic CP,[9,36,40,45,47,50,61,63] dyskinetic CP,[9,23,36,43,45] athetoid CP,[26,40,47,50,61,63] hypotonic CP,[40] and/or mixed CP.[23,36,40,45,47,50] Gross Motor Function Classification System (GMFCS) level was often not reported. Nine studies included adults from all GMFCS

Table 1: Description of quantitative studies

Study	Country	Design	Participant characteristics				
			n	Female *n* (%)	Age mean, SD (range) y:mo	CP type and distribution *n* (%)	GMFCS level *n* (%)
Andersson et al.[23]	Sweden	Cross-sectional	221	96 (43.4)	36 (20–58)	Spastic diplegia 77 (35), spastic hemiplegia 47 (21), spastic tetraplegia 27 (12), dyskinesia 48 (22), mixed 19 (9)[a]	I 86 (39), II 23 (10), III 32 (15), IV and V 79 (37)[b]
Balandin et al.[24]	Australia	Cross-sectional	279	137 (49.1)	(30–74)	NS	NS
Bax et al.[26]	UK	Cross-sectional	45	NS	(18–25)	Spastic 36, athetoid 9	NS
Beatty et al.[39]	USA	Cross-sectional	110	NS[c]	≥18	NS	NS
Carter et al.[37]	UK	Cross-sectional	NS[d]	NS[d]	(0–24)	NS[d]	NS[d]
Cathels et al.[50]	Australia	Cross-sectional	46	NS[d]	(15–25)	Spasticity 44, athetosis 14, ataxia 1, mixed CP 7	NS
Chiang et al.[56]	Taiwan	Case–control	713	282 (39.5)	24, 4:4 (18–32:9)	NS	NS
Cornec et al.[49]	France	Cross-sectional	362	NS[d]	≥18	NS[d]	NS
Engel et al.[40]	USA	Cross-sectional	64	35 (54.6)	36:10, 13:4 (18–76)	Spastic 36 (56), athetoid 9 (14), ataxic 1 (1.5), hypotonic 1 (1.5), mixed 17 (27)	I–II 9 (14), III–V 55 (86)[b]
Fortuna et al.[41]	USA	Cross-sectional	229	94 (41)	(18 to ≥60)	NS	I–III 125 (54.5), IV–V 104 (45.4)[b]
Heller et al.[57]	USA	Cohort	111	58 (52.2)	46 (32–88)	NS	NS
Hilberink et al.[51]	Netherlands	Cross-sectional	54	28 (52)	30, 3:4 (25–36)	Hemiplegia (37), diplegia (22), quadriplegia (41)[a]	I 15 (28), II 18 (34), III 4 (7), IV 13 (24), V 4 (7)
Houtrow et al.[42]	USA	Cross-sectional	1723	840 (48.8)	(18–64)	NS	NS
McDowell et al.[10]	UK	Cross-sectional	46	NS[d]	(19–27)	NS[d]	IV–V 46 (100)
Michelsen et al.[38]	Denmark	Cross-sectional	2443	NS	(19–55)	NS	NS[e]
Morgan et al.[54]	Australia	Cross-sectional	138	66 (47.8)	41:8, 17:1 (18:0–80:6)	NS	NS
Murphy et al.[47]	UK	Cross-sectional	107	40 (37.3)	22:1, 14:5 (2–83)	Spastic 55 (51.4), athetoid 29 (27.1), ataxic 3 (2.80), mixed 13 (12.14)	NS
Murphy et al.[43]	USA	Cross-sectional	101	48 (47.5)	42:6 (19–74)	Moderate hemiparesis 9, severe hemiparesis 1, moderate diplegic 11, moderate quadriplegic 14, severe quadriplegic 14, mild dyskinesia 2, moderate dyskinesia 38, severe dyskinesia 12[a]	I–III 34 (33.6), IV–V 67 (66.3)[b]
Nandam et al.[44]	USA	Cross-sectional	118	118 (100)	(40–64)	NS	I 8 (6.8), II 29 (24.6), III 33 (28), IV 32 (27), V 16 (13.6)
Ng et al.[55]	Singapore	Case–control	32	NS[d]	19:8 (17–22)	NS[d]	I 6 (18.7), II 5 (15.6), III 7 (21.8), IV 10 (31.3), V 4 (12.5)[b]
Nieuwenhuijsen et al.[52]	Netherlands	Cross-sectional	29	10 (34.5)	28:1, 8 (16–40)	Hemiplegia 8 (28), diplegia 7 (24), quadriplegia 14 (48)[a]	I 15 (52), II 4 (14), III 3 (10), IV 7 (24), V 0 (0)
Park et al.[36]	Republic of Korea	Cross-sectional	154	61 (39.6)	40:2, 9:2 (19–69)	Spastic 63 (40.9), dyskinetic 32 (20.8), ataxic 1 (0.6), mixed 47 (30.5), do not know/no response 11 (7.1) Quadriplegia 93 (60.4), diplegia 23 (14.9), hemiplegia 23 (14.9), monoplegia 4 (2.6)[a]	I 22 (16.3), II 47 (34.8), III 10 (7.4), IV 46 (34.1), V 10 (7.4)

Table 1. Continued

Study	Country	Design	Participant characteristics				
			n	Female *n* (%)	Age mean, SD (range) y:mo	CP type and distribution *n* (%)	GMFCS level *n* (%)
Pons et al.[9]	France	Cross-sectional	282	119 (42.5)	37:5, 12:8	Unilateral spastic 37 (13.5), bilateral spastic 126 (46), dyskinetic 42 (15.3), ataxic 26 (9.5), other 43 (15.7)	I (7.8), II (14.2), III (17.7), IV (29.1), V (31.2)
Roquet et al.[35]	France	Cross-sectional	282	NS[d]	(18 to ≥40)	NS[d]	I–III 112 (39.7), IV–V 170 (60.2)[b]
Russell et al.[25]	UK	Cross-sectional	57	22 (38.5)	(16–50)	NS	I 26 (45.6), II 4 (7), III 7 (12.2), IV–V 24 (42.1)[b]
Stevenson et al.[53]	UK	Cross-sectional	42	14 (33.3)	(20–22)	NS[d]	NS
Turk et al.[45]	USA	Cross-sectional	63	63 (100)	37:7, 12:7	Spastic diplegia 18 (29), dyskinesia/posturing 16 (25), spastic hemiplegia 11 (18), other (ataxia/ hypotonia/mixed) 10 (16), spastic quadriplegia 7 (11)[a]	NS
Whitney et al.[46]	USA	Cross-sectional	5555	2900 (52.2)	42:3, 13:9	NS	NS
Young et al.[48]	Canada	Cross-sectional	199	NS[c]	22	NS	III–V (58.9)
Young et al.[33,34,f]	Canada	Case–control	477	202 (42.3)	26:3, 2:6 (23–32:9)	NS	I–II (36), III (19), IV–V (45)

[a]CP subtypes presented as reported by the authors. [b]Estimated GMFCS based on ambulatory status data. [c]Not stated as per CP condition. [d]Not stated for adults with CP. [e]Unable to calculate GMFCS level from the ambulatory status data of different groups presented. [f]Young et al. described findings obtained from the same sample.[33,34] Therefore, we only describe participant characteristics of the samples included in Young et al.[34] and not Young et al.[33] to avoid duplication. CP, cerebral palsy, GMFCS, Gross Motor Function Classification System; NS, not stated.

levels.[9,33,36,44,51,52,66,70,73] Two studies included adults in GMFCS levels IV to V[10,69] and another study included those in GMFCS levels II to III.[77]

Caregivers were aged between 42 years[59] and 75 years,[65] with between 75%[67] and 100% being female.[59] Service providers were aged 27 years to 62 years, worked in various settings, and included physiotherapists, support workers, nurses, paediatricians, and physicians.[69,76]

Study quality

Quality of included studies is presented in Appendix S4 (online supporting information). Of the 27 cross-sectional studies included, six did not describe inclusion criteria,[24,35,39,43,47,54] three did not describe study participants and settings,[24,43,50] six did not describe outcomes measured,[24,26,39,40,43,50] and one did not clearly describe appropriate analysis.[40] The three case–control studies did not clearly report cases and control studied.[33,55,56] Reporting issues in relation to recruitment, outcomes measured, and incomplete follow-up were noted in the cohort study.[57]

Of the 26 qualitative studies, 10 lacked clear evidence between the stated philosophical perspective and methodology employed.[11,60,62,63,67,71,72,74,76,77] Six lacked clarity on congruity between methodology and study objectives and did not describe data collection processes.[60,62,71,72,74,77] Five studies lacked clear agreement between methodology and analysis[60,63,71,74,80] and interpretation of findings was unclear in four.[11,60,71,80] Seventeen studies did not locate the researchers' position clearly either culturally or theoretically[11,31,32,59,60,62,63,65,66,68,71,72,74–77,80] and 17 did not address reflexivity.[11,31,32,59,60,62–64,66,68,70,71,73,75–77,80] Participant voices were not adequately represented in two studies.[60,76] Four studies did not provide clear evidence of ethical approval.[11,65,80,81] A clear conclusion was provided by all included qualitative studies.

Health service use

Data from two quantitative studies are not included in this section as they did not report the proportion of adults using a service or frequency of visits to services.[42,57]

Data from 12 studies were included in meta-analyses examining the proportion of adults with CP using health services.[23–26,36,38,45,46,50–53] The results from nine studies are reported descriptively only.[9,10,33,35,39–41,43,44] Eight studies had data that were included in both the meta-analysis and descriptive summary.[24,36,38,45,46,50,51,53] Eight studies were included in meta-analyses of incidence rates of visits to each service.[33,38,45,46,48,54–56] The incidence rate is reported descriptively for two studies.[41,51] Two studies were included in both the meta-analyses and descriptive summary.[33,55]

Table 2: Description of qualitative studies

			Participant characteristics				
Study	Country	Design and analysis type	*n*	Female *n* (%)	Age mean, SD (range) y:mo	CP type and distribution *n* (%)	GMFCS level *n* (%)
Bagatell et al.[66]	USA	Descriptive design Thematic	Adults with CP 9	5 (55.5)	(19–33)	NS	I 4 (44.4), II 1 (11.1), IV 2 (22.2), V 2 (22.2)
Balandin et al.[71]	Australia	Thematic[a]	Adults with CP 10	5 (50)	48 (35–61)	NS	NS
Brandon et al.[69]	Canada	Descriptive design Content	Adults with CP 4, caregivers 4, service providers 4	Adults with CP 2 (50)	21	NS	IV 3 (75), V 1 (25)
Burkhard et al.[59]	USA	Phenomenological	Caregivers 11	11 (100)	52 (42–62)	NS	NS
Carroll et al.[58]	USA	Phenomenological	Adults with CP 9	6 (66.6)	(19–25)	NS	I–II 2 (22.2), III 3 (33.3), IV–V 4 (44.4)[b]
Darrah et al.[72]	Canada	Content[a]	Caregivers 39	NS	(19–23)	NS	NS
DiFazio et al.[67]	USA	Descriptive design Content	Adults with CP 5, caregivers 8	Adults with CP 3 (60), caregivers 6 (75)	Adults with CP 32, 8:6 (24–44), caregivers 62, 8:5 (50–72)	NS	NS
Hanes et al.[73]	Canada	Thematic[a]	Adults with CP 16	7 (43.7)	26, 3 (17–29)	NS	I 4 (25), II 3 (18.7), III 3 (18.7), IV 3 (18.7), V 1 (6.2)
Hemsley et al.[65]	Australia	Narrative approach	Caregivers 8	NS	66 (60–75)	NS	NS
Hemsley et al.[74]	Australia	Thematic[a]	Adults with CP 6	NS	(38–58)	NS	NS
Hemsley et al.[75]	Australia	Thematic[a]	Caregivers 5	4 (80)	66 (60–68)	NS	NS
Hemsley et al.[76]	Australia	Thematic[a]	Service providers 6	NS	(27–62)	NS	NS
Horsman et al.[61]	Canada	Phenomenological	Adults with CP 12	6 (50)	41 (25–58)	Spastic diplegia (6), spastic quadriplegia (2), ataxia (2), spastic double hemiplegia/ataxia (1), athetoid quadriplegia (1)[c]	I–III 5 (41.6), IV–V 7 (58.3)[b]
Iezzoni et al.[62]	USA	Grounded theory	Adults with CP 3	NS	NS	NS	NS
Iezzoni et al.[68]	USA	Descriptive design Content	Adults with CP 4	NS	NS	NS	NS
Kroll et al.[60]	USA	Phenomenological	Adults with CP 10	NS	NS	NS	NS
LariviAre-Bastien[31,32,d]	Canada	Thematic[a]	Adults with CP 14	7 (50)	20:9 (18–25)	NS	I–II 2 (14.2), III–IV 4 (28.6), V 8 (57.1)[b]
Moll et al.[64]	Canada	Narrative	Adults with CP 9	6 (66.6)	(26–70)	NS	I–II 2 (22.2), III–IV 3 (33.3), V 4 (44.4)[b]
Morgan et al.[77]	Australia	Thematic[a]	Adults with CP 6	5 (83.3)	45:8 (35–52)	NS	II 3 (50), III 3 (50)

Table 2. Continued

Study	Country	Design and analysis type	Participant characteristics				
			n	Female *n* (%)	Age mean, SD (range) y:mo	CP type and distribution *n* (%)	GMFCS level *n* (%)
Morris et al.[78]	USA	Thematic[a]	Adults with CP 5	3 (60)	48:6 (36–69)	NS	NS
Mudge et al.[70]	New Zealand	Descriptive design Thematic	Adults with CP 28	14 (50)	Median 47 (37–70)	Diplegia (2), hemiplegia (5), quadriplegia (18), unspecified (3)[c]	II 5 (17.8), III 8 (28.5), IV 12 (42.8), V 3 (10.7)
O'Day et al.[80]	USA	NS	Adults with CP 18	NS	NS	NS	NS
Racine et al.[79]	Canada	Thematic[a]	Adults with CP 14	7 (50)	20:9 (18–25)	NS	I–II 2 (14.2), III–IV 4 (28.6), V 8 (57.1)[b]
Redmond et al.[63]	UK	Grounded theory	Adults with CP 26	15 (57.6)	(17–22)	Spastic quadriplegia 13, hemiplegia 1, diplegia 6, athetoid 4, ataxic 2[c]	NS
Scheer et al.[11]	USA	Thematic[a]	Adults with CP 10	NS	NS	NS	NS

[a]Studies only reported analysis type and not study design. [b]Estimated GMFCS based on ambulatory status data. [c]CP subtypes presented as reported by the authors. [d]Larivière-Bastien et al. reported findings obtained from the same sample.[31,32] Therefore, we only describe participant characteristics of the samples included in Larivière-Bastien et al.[31] and not Larivière-Bastien et al.[32] to avoid duplication. CP, cerebral palsy, GMFCS, Gross Motor Function Classification System; NS, not stated.

Medical services

Use of medical services are summarized in Table S1 (online supporting information). Thirty-three per cent (95% confidence interval [CI]: 23–44%; I^2=98.7%; p<0.01) of adults were admitted to hospital during a 12-month period.[36,38,45,46,53] Twenty-eight per cent (95% CI: 17–40%; I^2=98.7%; p<0.01) of adults used the emergency department over 12 months,[38,45,46] with other studies reporting 10% of adults visited the emergency department in the last 3 months[40] and 38% visited the emergency department in the past 2 years.[41]

Pooled analysis of three studies showed that 84% of adults (95% CI: 78–90%; I^2=82.2%; p<0.01) visited a general practitioner over 12 months[24,38,45,50] (Fig. 1). The proportion of adults with CP that attended a general practitioner over alternative[10,33,39,41] or unspecified[9,35] time-periods ranged from 39%[10] to 77%.[39] One study reported 40% of adults visited a general practitioner or nurse practitioner for pain treatment in the past 3 months.[40] Between 26.1% and 53.2% of adults received medical,[36] physical,[24] or disability[53] assessment check-ups during varying time-periods.

Pooled analysis of four studies indicated that 11% (95% CI: 2–20%; I^2=87.7%; p<0.01) of adults visited a neurologist over 12 months.[24,50–52] Three single studies reported between 25% and 37% of adults visited a neurologist over different time-periods.[35,38,41] Pooled analysis showed that 16% (95% CI: 6–26%; I^2=86.4%; p<0.01) of adults visited an orthopaedic surgeon over the past 12 months.[24,50–52] Estimates from single studies over various time-periods ranged from 21% to 36%.[10,38,41] Pooled analysis of two studies indicated that 7% (95% CI: 2–13%; I^2=0.0%; p=0.01) of adults visited urology services in the past 12 months.[51,52] Although this review focused on adults, one study reported 26% of participants visited a paediatrician over an unspecified time-period[50] whereas another study reported 4% of adults visited a paediatrician over a 9-year period.[38] Medical services for female health issues ranged from 18% to 92% of the population.[44,45] Pooled analysis of five studies showed that 65% (95% CI: 54–75%; I^2=80.1%; p<0.01) of adults visited a dentist over 12 months.[24–26,45,50] The proportion of adults attending services for visual impairment ranged from 2% over 6 months to 53% over 12 months.[10,24,35] Seven percent of adults attended an audiologist over 12 months.[24]

Adults had 67 (95% CI: 37–123) hospital admissions per 100 person-years (I^2=99.9%; p<0.001).[33,46,48,54,56] This equates to 67 visits among 100 adults with CP observed for 1 year. Adults had 81 (95% CI: 34–194) emergency department visits per 100 person-years (I^2=100%; p<0.001).[33,38,46,48] One study reported 28.8% of adults had 'one or two emergency department visits' and 9.6% had 'three or more emergency department visits' over 2 years.[41]

Adults had 404 (95% CI: 175–934) general practitioner visits per 100 person-years (I^2=100%; p<0.001; Fig. 2).[33,38,45,55] One study reported 33.6% of adults had 'one or two primary care visits' in the past 2 years.[41] Single

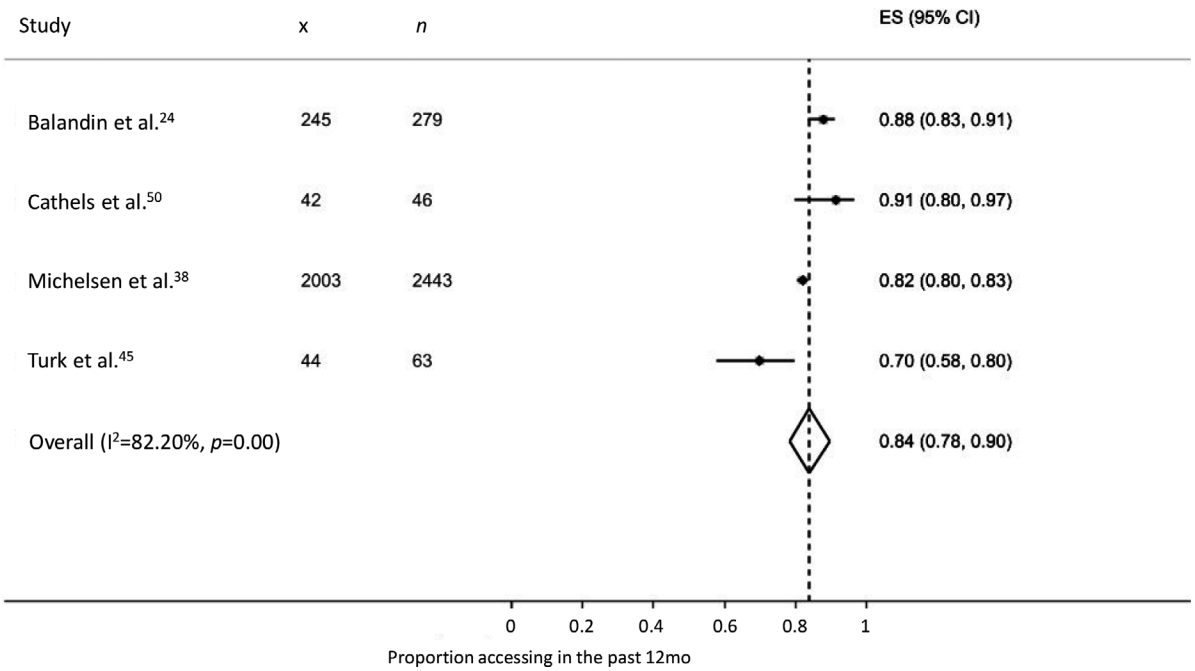


Figure 1: Proportion of general practitioner use. x, number of people used; *n*, total sample; ES, effect size (proportion); CI, confidence interval.

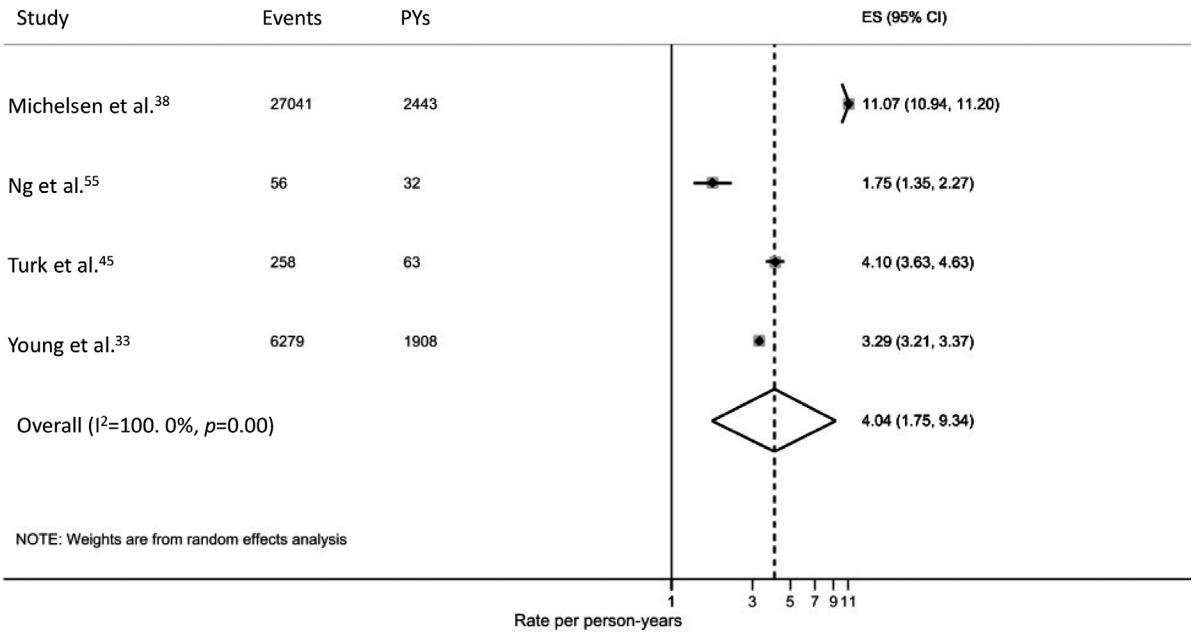


Figure 2: Rate of general practitioner visits. PYs, person-years; ES, effect size (rate); CI, confidence interval.

studies reported adults had 1169 outpatient physician visits per 100 person-years[48] and 24 specialist visits per 100 person-years.[55] One study reported 121 visits to a specialist physician, nine visits to a paediatrician, 2.3 visits to a pregnancy care office, and 54 visits to a psychiatrist per 100 person-years.[33]

Rehabilitation services

Use of rehabilitation services are summarized in Table S2 (online supporting information). Pooled analysis of four studies indicated that 22% (95% CI: 9–36%; I^2=90.3%; p<0.01) of adults visited a rehabilitation specialist over 12 months.[24,50–52] Single studies reported that 52% to

59% attended similar services over unspecified time-periods.[9,35]

Pooled analysis of seven studies indicated that 44% (95% CI: 36–51%; I^2=70.2%; p<0.01) of adults visited a physiotherapist over 12 months.[23,24,26,50–53] The proportion of adults visiting physiotherapy services over other or unspecified time-periods[9,10,35,36,41] varied from 28% to 87%. Hippotherapy and hydrotherapy services were used by 9% and 28% of adults respectively.[51]

Similar to the findings of individual studies,[9,10,35,36,41] pooled analysis showed that 27% (95% CI: 12–41%; I^2=87.7%; p<0.01) of adults visited an occupational therapist over 12 months.[24,26,51,52] Pooled analysis of four studies indicated that 16% (95% CI: 3–28%; I^2=90.9%; p<0.01) of adults visited a speech therapist/pathologist over 12 months.[24,26,51,52] However, for individual studies over various[10,41] or unspecified time-periods,[35,36] the proportion varied from 1.3% to 26%.[10,36] Pooled analysis of three studies indicated that 7% (95% CI: 4–9%; I^2=0%; p=0.90) of adults visited a dietician over 12 months.[24,51,52] One study reported 28% of adults attended a podiatrist over 12 months,[24] with another reporting that 12% visited a podiatrist over 2 years.[41] Pooled analysis of three studies indicated that 11% (95% CI: 1–20%; I^2=85.4%; p<0.01) of adults visited a psychologist or psychiatrist over 12 months,[24,51,52] with single studies over variable[10,38,40,41] or unspecified time periods[9,35,36] reporting that 1.3% to 21%[9,36] of adults attended such services.

A single study reported assistive equipment services were used by 68.2% of adults over 12 months.[39] Other single studies reported 74% of adults used aids and equipment,[53] 35% to 100% used augmentative and alternative communication aids,[24,43] 9% to 69.5% used orthoses,[9,35] 72% to 78% used mobility aids,[9,35] 30% used a trunk supporting device, 5% used a standing device, and 21% used a specialized foam mattress over an unspecified time-period.[35] One study reported 50% of adults visited formal and informal respite care, 35% visited a social worker, and 48% visited a support group in the past 6 months.[10]

Single studies reported 484 physiotherapy visits per 100 person-years,[55] 204 occupational therapist visits per 100 person-years,[55] and 65 allied health professionals visits per 100 person-years.[51]

Factors and outcomes associated with health service use

Nine studies reported factors associated with health service use.[9,33,35,36,42,44,48,51,52] Sex and ethnicity were related to health service use: females had higher rates of service use compared to males[33,36,48] and adults from ethnic minority groups were less likely to be hospitalized or attend the emergency department compared to those of white ethnicity.[42] Those receiving national basic livelihood security/medical aid in the Republic of Korea were more likely to have frequent hospital visits, periodic medical follow-ups, and rehabilitation treatment compared to adults not receiving aid.[36]

GMFCS level was associated with medical[9,36,52] and rehabilitation service use.[9,36,51,52] One study found adults in lower GMFCS levels (I–II) were more likely to visit rehabilitation physicians, orthopaedic surgeons, physiotherapists, occupational therapists, and speech therapists compared to adults in higher GMFCS levels (III–IV).[52] However, other studies reported adults in higher GMFCS levels had more frequent hospital visits and medical follow-ups, and were more likely to use rehabilitation treatment, use of medications, orthotic devices, general practitioner visits, and rehabilitation physician input compared to those in a lower GMFCS level.[9,36] Adults in higher Manual Ability Classification System levels (IV–V) were less likely to attend mammogram screening in the previous 2 years.[44] Adults with difficulties in activities of daily living were more likely to attend the emergency department compared to those with no difficulties in activities of daily living.[42]

For ambulatory adults, increased age was associated with decreased use of psychiatrist visits, orthoses, trunk supports, rehabilitation specialist follow-ups, physiotherapist, occupational therapist, speech therapist, psychomotor therapist, and orthoptist services.[35] However, for non-ambulatory adults, increased age was associated with increased use of mobility aids, rehabilitation specialist follow-ups, psychiatrist visits, general practitioner visits, neurologist visits, occupational therapy, psychomotor therapy, and orthoptist services,[35] and decreased use of orthosis, trunk supports, physiotherapist services, and speech therapist services.[35]

Adults with pain or epilepsy were more likely to attend rehabilitation physicians and neurologists respectively, than adults without pain or epilepsy.[51] Adults with cognitive limitation were less likely to visit the emergency department than those without cognitive limitation.[42] One study reported weak associations between cognitive level and use of physiotherapy, occupational therapy, hippotherapy, and hydrotherapy, however the direction of association was unclear.[51]

Unmet needs in the areas of information, mobility, social activities, work or training, voluntary organizations, housing, and formal social services were positively associated with GMFCS level, which was in turn positively associated with the use of rehabilitation physicians and physical therapists.[52] Adults with bilateral CP were more likely to have unmet needs in the areas of information, finances, and formal social services, and they were also more likely to use rehabilitation physicians and physical therapists in comparison to people with unilateral CP.[52]

Qualitative synthesis

Six themes were developed from the 26 included qualitative studies.

Transition challenges faced by adults with CP

The transition from paediatric to adult services posed challenges for adults with CP in 10 of the 26 included studies.[31,32,58,61,64,66,67,69,72,73] Studies from the USA and Canada reported abruptness in transition, which adults with CP described as a loss of a trusted relationship and a

feeling of abandonment.[32,64,66,67,77] Young adults were not prepared for the transition which made them feel anxious and overwhelmed to take on health care management responsibilities.[32,66] During transition, both adults and caregivers reported emotional upheaval; adults with CP also reported fear and sadness.[32,67] To ease the transition process, some adults proactively took steps to communicate with service providers before each appointment.[67] Some adults had high expectations of adult services, which the transition process then failed to meet.[58]

> The word abandonment is exactly how I feel. I feel like 'they' just took us and just threw us out there to an adult provider …[67]
>
> (p. 22)

During the transition to adult services, adults and caregivers faced challenges in navigating appropriately trained providers to meet their needs.[32,58,67,69] Service providers reported a need for training and support from a wider team for a smooth transition process.[69] Lack of information and continuity of care during and after transition frustrated both caregivers and adults with CP.[32,67,69] Adult and paediatric service providers failed to coordinate with each other, causing transition to be even more challenging for young adults with CP.[32,67,69] In addition, adults with CP were disappointed with the brief consultations provided within adult services, reportedly due to lack of resources, compared to longer paediatric appointments.[58,61,67,69] Some caregivers of adults with CP worked around the transition barriers by accessing previous paediatric services.[67]

Transition often required adults with CP to deal with insurance companies, bureaucracy, and extensive paperwork and requires them to self-advocate for their needs.[67,72] Additionally, transferring health records from paediatric to adult services was challenging with long waiting times and delays in processing reports.[32,61,66,67] Each agency involved in transition had its own set of processes to follow, but adults with CP perceived there to be a lack of guidance on navigating those processes.[66]

Adults with CP and caregivers recommended transition should begin early with a clear roadmap and honest communication of the transition process by their service providers.[32,67] Service providers also agreed that coordination between paediatricians and adult physicians should begin early with clear communication.[69] However, some paediatricians reported challenges in coordinating with adult services, which lacks a collaborative model of care.[69]

> It's very difficult for a primary care physician who is in practice on their own to be able to provide all that care. If there was a nice connection to resources in the community, that would be even better.[69]
>
> (p. 6)

A clinical care coordinator was required during the transition process to facilitate sharing of adequate information.[31,32,67] A model similar to paediatric services, with appropriately trained providers available under one roof, was recommended by adults with CP and their caregivers.[67,73] Adults, caregivers, and service providers suggested adequate resources were required for adult services to meet the needs of adults during the transition process.[31,58,61,69]

Caregivers' collaborative roles and responsibilities for adults with CP

The importance of caregivers' collaborative roles and responsibilities was evident in 11 studies.[31,58,59,65,71,73–76,79,80] Of these, four were outputs from an Australian study focusing on perspectives of adults with CP, caregivers, and service providers in relation to hospital experience.[65,74–76] Primary caregivers felt their caring role for adults was an obligation as a parent,[31,58,59,65,71,73–76] although it affected their lives both physically and emotionally.[59,65,75,76] Caregivers accompanied adults for all their health care visits, viewing that as part of their role.[58,79] Some studies reported that the caregivers' role involved encouraging adults to be independent.[59,79] Some adults found caregivers presence useful for appointments, however, some adults perceived caregivers as overprotective and preferred to be independent and advocate for themselves.[73,74,79,80]

> I want to move out of my place but there are no houses adapted for me, and I do want to make my life without my mother and she does not want to, I find it hard to do my life alone, I just cannot, and my mother says, 'you are not allowed because of your disability'. Gosh, I can … I am able to watch over my own health … I find it difficult and have to put up with this.[79]
>
> (p. 875)

Most of the time, primary caregivers advocated for the care needs of the adult with CP,[58,59,65,73–76] as they understood those needs and acted in the adult's best interest[59,65,73–76,80] and considered themselves responsible for ensuring that the voice of the adult with CP was heard.[59,65,75] This usually involved developing partnerships with health care staff, which was both a positive and negative experience for caregivers.[65,74–76] In particular, caregivers intervened, or acted as advocates, to protect adults from emergency situations or get the necessary attention from service providers.[59,65,75] Some studies suggested this created friction between caregivers and service providers within hospital settings.[65,75] However, service providers acknowledged and valued caregivers' intermediary role in communicating the needs of adults with CP.[76] Adults with CP reported that service providers were stressed when the adult was not accompanied by caregivers for hospital admissions.[31]

> I learned that she has one advocate … She has no one's voice but mine and if I don't get loud and I don't act boldly on her behalf, no one will. You need to trust your gut as a caregiver and mom.[59]
>
> (p. 360)

Caregivers played an important role in supporting adults who had speech impairments in hospital settings.[65,71,74–76] They also felt a responsibility to care for the needs of these adults in the hospital because they had expertise in caring for the adult, or because they had concerns about the quality of hospital care.[65,75]

Caregivers were often challenged to accommodate care needs and multiple roles and responsibilities in life.[59,65,75] Therefore, primary caregivers took support from a spouse, grandparents, and siblings who also played an integral role.[59,65] Some older parents additionally suffered from chronic age-related medical conditions themselves.[59,65,75] Lack of support from service providers or challenges in navigating services added emotional and physical demand to caregivers.[59,65,75] Some caregivers were worried about their absence in the future and the impact of that on adults' care management.[59,65]

Caregivers' experiences with hospital service providers were described as challenging and negative.[65,75] Both caregivers and adults with CP reported that hospital service providers did not have the expertise to care for adults with CP who had additional communication needs.[65,74,75] Lack of expertise meant some service providers were perceived to be not flexible or patient enough to care for adults with CP in the hospital setting.[65,74,75] In addition, some caregivers felt that their expertise was never acknowledged, and opinions were ignored by hospital service providers.[65,75] However, some caregivers took additional steps in building a collaborative relationship with service providers in relation to addressing the care needs of adults with CP and appreciated any opportunities to collaborate.[65]

> I stay with her because I don't expect the staff to be able to give her the time, they wouldn't know how to look after her. And I used to push her down to the bathroom and wash her hair and everything on my own because they didn't know how to do it.[65]
>
> (pp. 245–246)

Needs associated with ageing in adults with CP

Ageing in people with CP has a significant impact on adults' physical and mental health; this was evident in 9 of the 26 included studies.[32,58,61,62,64,66,70,73,80] Age-related changes included increased fatigue, reduced energy, mobility or functional decline, reduced balance, stiffness, and pain in middle age.[61,64,66,70,73,80] Adults also reported that they experienced pain, spasms, and anxiety affecting sleep, work, and daily living functions.[73] Ageing changes affected adults' identity and mental health, causing loneliness and isolation and consequently demonstrating a need for counselling services.[61,70]

Lack of information and expertise on ageing among service providers were consistently reported to cause frustration in adults with CP.[61,62,66,80] Some adults were prepared for these changes either because of experiencing it themselves or by anticipating it for the future.[58,61,64,66,70,80] Adults frequently described fear, anxiety, and worry in relation to future health care needs due to ageing.[70,73]

> My fear is it's going to be harder – that's my fear, and I'm going to have some person who comes by and dresses me and puts me together and but … there's this determination there that no, it's not to be that way – I'm going to stay as fit and as mobile as I possibly can till the day I cark it.[70]
>
> (p. 6)

A sense of loneliness was a contributing factor from lack of having a relationship or family for themselves.[61] However, some service providers ignored adults' needs and desire to have a family.[66] Family members' lack of understanding of ageing needs impacted adults seeking help from their family caregivers.[70] Adults preferred to be independent as much as possible and depend on their caregivers only when necessary.[70] They faced challenges in securing funds to recruit paid caregivers to meet their needs.[61] Some adults used coping strategies to deal with ageing such as acceptance of ageing, self-management with diet and exercise, using alternative therapies, and using wheelchairs to save energy or reduce fatigue.[61,70] Most adults preferred discussing adaptations for their ageing needs or coping strategies with service providers who understood ageing needs in CP.[61,66,70,80]

Access – the ability to attend health services

In 16 of the 26 qualitative papers, adults experienced challenges in their ability to access health services due to the affordability,[11,59,77,80] transportation issues,[11,77] or environmental barriers within the service[11,31,68,69,77,79] particularly for people who used assistive devices.[11,65–67,69,71,73–76]

In studies conducted in the USA, the ability to afford services was a concern for adults or their caregivers.[11,59,80] Insurance payments and eligibility for therapy or equipment services were perceived as an important limitation in accessing services,[11,80] despite some health professionals' efforts to lobby insurance companies.[80] Adults in Australia experienced a financial burden in order to pay for health care services, which hindered access to services.[77] Lack of public transport services and the cost involved in arranging transport for appointments also impacted accessibility.[11,77]

In both the USA and Canada, environmental barriers within the health services contributed to inaccessibility.[11,31,68,69,77,79] This was often related to lack of appropriate equipment such as height-adjustable examination tables or diagnostic equipment, wheelchair-accessible weighing scales, and manual handling equipment for transfers.[11,31,68,69] In addition, several studies highlighted that hospital, physician, diagnostic, and gym services lacked accessible examination rooms, disabled access toilets, wheelchair-accessible entrances, accessible car parking locations, or had only stair access that further limited accessibility.[31,68,69,77,79]

Studies from the USA and Canada reported that some health services were inaccessible for adults with assistive

mobility devices such as a wheelchair.[11,68,69] Adults perceived that services were designed for non-wheelchair users.[11,68,69]

> How many physicians have accessible mammogram machines? You are in a wheelchair and they make you stand up. You've got cerebral palsy, you can hardly hold on, let alone keep your breast still, so even though you have insurance and access, you don't have access to appropriate care. You don't have access to a high–low examining table, so you are too frightened to get a Pap because you feel you are going to fall off the table.[11]
>
> (p. 225)

Similarly, in studies from the USA and Australia, adults with speech assistive devices experienced challenges accessing hospital and community services.[65–67,71,74–76,80] However, four of these studies were from a single larger study on the experiences of adults with CP, caregivers, and health professionals within the hospital environment.[65,74–76] Adults with speech impairment had difficulty communicating with the service providers, so they relied on assistive devices or interpreters to access health services.[65–67,71,74–76,80] Interpreters for adults with speech impairments were often family caregivers who used various methods such as alternative communication boards to enhance access to health services.[65,67,75,76] However, accessibility for adults was limited by a lack of understanding among service providers.[65,67,71,76]

> I had to explain to them, they'd ask him a question, and even though every change of shift they would be told he cannot talk, and he can't answer your questions … so then because [my son] uses a talking machine, I got the old pointing thing [communication board] for them to try and help them.[75]
>
> (p. 263)

Alternatively, some adults from one study had a positive experience in accessing health services with the assistance of an interpreter or when nurses used good communication strategies.[71]

Knowledge and experience impact care relationships and service provision for adults with CP

The knowledge and experience of service providers in relation to CP, were perceived to impact the relationship between adults with CP or caregivers, and service providers in 25 studies.[11,31,32,58–68,70–80] Service providers' lack of understanding of CP and their needs was a concern for both adults with CP and caregivers.[11,32,59,62,65,66,70,71,74,75,77,79,80] Some studies reported inappropriate treatment provided from lack of knowledge about CP and its characteristics.[32,71,77,80] This sometimes resulted in getting conflicting information from service providers.[60] In addition, some service providers were perceived to be unwilling to learn from adults with CP, due to either time constraints or considering themselves experts.[65,70,74–76,78]

Lack of expertise in providing basic care or interacting with adults with CP with complex communication needs in the hospital setting was frustrating for both caregivers and adults with CP.[31,65,71,75] Studies reported that when health workers stereotyped adults with a physical disability as having an intellectual disability, it affected their care and relationship.[70,75,79,80] This was evident when staff ignored adults with CP and directly interacted with caregivers, or were impatient and used medical jargon.[31,70–72,74,75,79] Some adults with CP experienced offensive comments and rude attitudes from service providers causing dissatisfaction with the services received.[31,80]

> The thing is too I think they see people in a wheelchair and they immediately think that they don't have a brain.[75]
>
> (p. 263)

Within the hospital settings, adults felt that a lack of knowledge and experience from service providers increased their length of stay and delayed their recovery.[71,74] Adults also experienced negative attitudes towards disability from service providers in making referrals, where service providers' assumptions about disability overshadowed adults' needs.[11,80] Both service providers and adults agreed on a need for more training, knowledge, and experience about CP to provide better care.[31,74,76] Some adults had a positive experience when service providers listened to adults' needs and spent more time working in partnership with adults.[62,66,70,74,78]

For adults with CP, knowledge and understanding of their own needs was an essential element in self-managing their health and educating others. The ongoing nature of CP meant that adults needed to communicate and educate service providers or caregivers about their disability and needs.[58,70,74] However, adults were not always well-prepared to self-advocate for their needs, potentially because of a lack of training or support received over the years.[67] For some adults, self-advocacy arose from frustrating experiences with service providers or caregivers' lack of knowledge.[74,77]

> But what's urgent to you and what's urgent to them often don't meet. So you have to fight, fight, fight.[77]
>
> (p. 286)

Self-advocacy skills often required in-depth knowledge of the adult's own condition, perception of their self-identity, and their identity among others. For some adults, living with CP as an identity was a challenge because of social attitudes and stigma associated with disability.[61,66,70,73] Rather than focusing on disability, most adults valued their identity based on their roles and responsibilities in life, which helped in maintaining control of their health and wellbeing.[61,63,64,70,73,74,77]

Health system challenges faced by adults with CP

Health system challenges were evident in 16 of the 26 included studies.[11,31,32,59,60,62–65,68,72,73,76,77,79,80] Both adults with CP and caregivers discussed a mismatch

between the availability of services, and the needs and expectations of adults with CP.[11,31,32,59,62–64,68,73,77,80] In Australia and the USA, studies reported inadequate community-based services for adults with CP.[59,77] Lack of rehabilitation, physiotherapy, and hydrotherapy services were of particular concern and led to a reported decline in mobility and functional abilities among adults with CP.[63,64,73,77] Challenges in navigating mental health services among adults with mild CP affected their mental wellbeing[73] and poorly managed hospital services caused an unpleasant experience for some adults with CP.[31]

A fragmented health system for adults with CP was illustrated by both adults and caregivers.[32,65,77,80] Having a consistent service provider was important for adults with CP: frequent changes in physiotherapists or doctors created mistrust.[77,80]

> Well, you go to the doctor one time, you see him for two months, and then he's shipped somewhere else – you don't know who you will see. He doesn't know you from beans. That's the biggest problem.[80]
>
> (p. 47)

Some adults acknowledged the complexity and time involved in their care but expressed a sense of dissatisfaction with the fragmented adult health system.[32,77,80] Information exchange was consistently a challenging experience for both adults and caregivers within the adult health system.[60,65,72,79] Studies reported that the lack of information from service providers after a discharge from hospital, or dearth of information on available services within the community, frustrated caregivers.[65,72] Some adults perceived lack of time as a contributing factor for this limited information sharing.[60] Other studies highlighted that a lack of communication between service providers, and not involving adults with CP and their caregivers in decision making, made coordinating their care challenging within the adult health system.[60,79]

> One is doing their thing and the other one is doing theirs, and there's no communication between them about what's going on … but they don't follow-up … there's no-cross communication between them about what's going on.[60]
>
> (p. 1111)

Some adults preferred the paediatric health system model, where adequate time, resources, and communication were perceived to have made care coordination easier.[32] Adults recommended a partnership and collaborative model to meet their needs, funded by the public health system, with adequate resources and staff to reduce system-level challenges.[77]

Integration

Quantitative and qualitative findings were mapped to the ANM (Fig. 3). The relationships between components as described in the original ANM are shown in Appendix S3. We identified associations between the environment, population characteristics, outcomes, and adults' use of services. The associations identified in this review are described in the next sections.

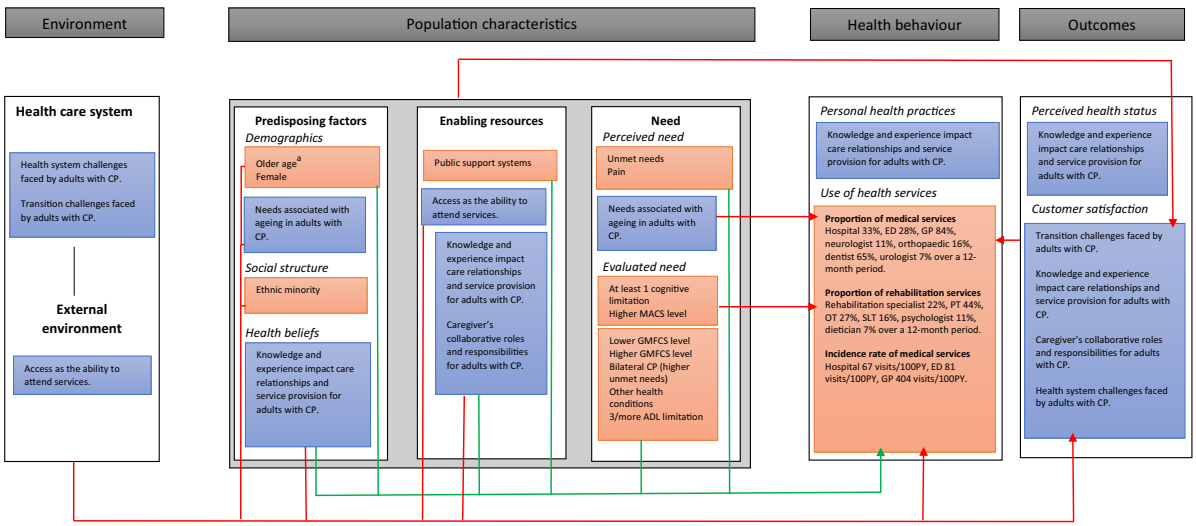


Figure 3: Integration of quantitative and qualitative findings using Andersen and Newman Model of health care utilization. Quantitative findings are in orange boxes. Qualitative findings are in blue boxes. Positive associations or facilitators of health service use are illustrated using green arrows. Negative associations or barriers to health service use are illustrated using red arrows. [a]Age and Gross Motor Function Classification System (GMFCS). CP, cerebral palsy; ADL, activities of daily living; MACS, Manual Ability Classification System; ED, emergency department; PT, physiotherapist; OT, occupational therapist; SLT, speech and language therapist; GP, general practitioner; PY, person-years. [Colour figure can be viewed at wileyonlinelibrary.com]

Health behaviour

Use of medical services over 12 months ranged from 7% for a urologist to 84% for a general practitioner. Use of rehabilitation services over 12 months ranged from 7% for a dietician to 44% for a physiotherapist. Visits ranged from 67 hospital admissions per 100 person-years, to 404 general practitioner visits per 100 person-years. Qualitative findings highlighted that service providers' lack of knowledge about CP resulted in adults having to self-manage their condition without adequate guidance.

Environment

Factors relating to the environment, and their influence on health service use, were only described in the qualitative literature. Environmental factors negatively impacted use of health services and satisfaction with health services. People with CP experienced a fragmented, poorly resourced health care system, which prevented them from identifying and accessing appropriate services to meet their needs and potentially resulted in reduced service use. Adults faced system-level challenges, such as poor care coordination, bureaucracy, funding, and organizational issues, from the beginning of the process of transitioning to adults' services and throughout adulthood. Adults' use of services was also negatively impacted by physical barriers, such as lack of appropriate equipment, examination rooms, and transportation issues.

Population characteristics

Quantitative and qualitative findings described the relationship between population characteristics, health service use, and satisfaction with health services.

Predisposing characteristics. Demographics, social structure, and health beliefs were positively and negatively associated with health service use. Qualitative findings described how adults perceived that their knowledge of their condition and ability to self-advocate were positively associated with service use. Adults who lacked an understanding of the impact of ageing on their physical or mental health and people with a negative attitude towards health services were less likely to use services.

Enabling resources. Adults that could avail of publicly funded support systems were more likely to use services. Health service use was enabled when service providers had previous experience with adults with CP, and when caregivers supported adults to access services. Lack of resources, service providers' lack of previous experience, negative attitude towards disability, and lack of support to caregivers from providers all negatively impacted health service use.

Need. There was evidence of positive and negative associations between service use and perceived and evaluated need. Pain, unmet needs, bilateral CP, activities of daily living limitations, and associated health conditions were associated with an increased likelihood of service use. There was conflicting evidence regarding the direction of association between GMFCS level and service use. People with cognitive limitations and in higher Manual Ability Classification System levels (IV and V) were less likely to use services. Qualitative evidence highlighted that a perceived lack of understanding of the ageing needs of adults on the part of service providers negatively impacted service use.

Outcomes

We did not identify quantitative data describing associations between service use and outcomes such as customer satisfaction, perceived health status, or evaluated health status. Qualitative findings showed that service providers' lack of knowledge about CP negatively influenced adults' perceived health status, which impacted negatively service use. Dissatisfaction with health services was also associated with decreased service use. Health system challenges experienced led to poor satisfaction. Service providers' lack of expertise in CP and caregivers' disappointment with service providers also negatively impacted satisfaction. The review findings suggest that satisfaction with services mediates the negative association between environmental factors, population characteristics, and use of health services.

DISCUSSION

The findings indicate that the general practitioner is the most commonly and most frequently visited health professional among adults with CP. Health service use was affected by demographics and needs such as sex and functional ability, although the direction of association between functional ability and service use was inconsistent. We identified six key themes relating to experiences of health services, which encompassed individual and health system challenges. Integration of quantitative and qualitative findings highlighted the influence of contextual factors on health service use among adults with CP.

Although most of the people living with CP are adults, most research and services for CP have focused on children. The quantitative findings from this review indicate the proportion of adults who use services varies considerably depending on the type of service, with a relatively low proportion of adults using specialist and rehabilitation services. Previous studies of children with CP reported that 19% to 59% used rehabilitation services such as physiotherapy, occupational therapy, and speech and language therapy, higher than the proportions estimated for adults in this review.[82,83] Other studies have demonstrated that the proportion of people with CP who use specialist and rehabilitation services decreases sharply between childhood and adulthood.[10,33,35,38] When compared to other neurological conditions with a similar prevalence, such as multiple sclerosis or Parkinson disease, the proportion of adults with CP accessing specialist services is much lower.[84,85]

We found that the largest proportion of adults (84%) and the highest rate of visits (404 visits per 100 person-years) were for general practitioner services. A recent report documented lower rates of general practitioner visits for children with CP.[86] About half of people with CP in the UK are discharged from children's services to their general practitioner because there are no dedicated adult services to refer to.[87] Absence of rehabilitation specialists that support services for adults with CP could partially explain increased use of general practitioner services. However, in this review, use of rehabilitation physician services by adults with CP was much lower compared to adults with acquired brain injury (22% and 68% respectively).[88] Thus, although there is lack of adult rehabilitation services for adults with CP, it remains unclear if this is due to lack of CP specialists, or if existing adult rehabilitation services are focused more on people who acquire a brain injury in adulthood. The fact that dedicated services exist for individuals whose condition typically arises in adulthood (e.g. multiple sclerosis, Parkinson disease, and acquired brain injury) suggests that service commissioners are either not aware, or not prioritizing the health care service needs of adults with a 'paediatric condition'. Although we did not specifically seek studies that examined transition to adult services, the challenges faced by people as they transition from a well-established paediatric multidisciplinary service to a fragmented adult service was a clear theme from qualitative studies. Interestingly, despite the National Institute for Health and Care Excellence (NICE) guideline on transition recommending that young people should have a 'named person to facilitate an effective transition process',[89] very few people with CP have a key worker at this time.[90] Our findings emphasize the need for a key worker to coordinate care during transition and in adulthood.

Health service use does not indicate service need or adequacy. Qualitative findings suggest that the needs of adults are not being met by current services. A lack of person-centred care at both individual and system level, with fragmented or poorly developed adult services, were the key challenges faced by adults with CP and caregivers. While the qualitative studies did not explore recommendations for improving services, or optimal service delivery models, our findings echo recent NICE guidelines for adults with CP,[91] highlighting the need for coordinated care provided by a multidisciplinary team for adults with CP. Adequate training for adult service providers to understand and meet adult needs is important for effective service delivery. A collaborative working relationship with paediatric services is key for successful transitioning to adult services. There is a need for system-level changes with clear care pathways and resources to improve accessibility for adults with CP, supporting implementation of the NICE guideline.

The ANM suggests that inequitable access to services occurs when health service use is determined by poor social structures, health beliefs, and enabling resources.[29,92] Our findings highlight limited availability of and access to services. Globally 1 billion people have a disability, therefore ensuring services are accessible to everyone, including people with CP, is a priority.[93]

Several methodological difficulties and gaps in the published literature were encountered when conducting this review. A lack of consistent reporting between studies, particularly regarding the time-period studied, made synthesis difficult. There were limited data on factors associated with service use. Further, the direction of association between factors and service use was not always reported. According to the ANM, education, marital status, family income, area of residence, and social support influence service use;[29] none of the included studies examined associations between these factors and service use. Similarly, there was a lack of quantitative data on the associations between outcomes, such as satisfaction and perceived health status, and service use.[29] Studies included in this review were all from high-income countries. Health service use in other regions will likely vary from the findings reported here. Other evidence gaps included a lack of qualitative studies exploring range of service providers' perspectives, experiences of mental health services, and adults' recommendations for an ideal service delivery model.

Limitations

Relatively few studies were included in each meta-analysis and there was significant heterogeneity associated with the meta-analyses. This may be due to differences in population, country, and methodology. We were unable to perform a meta-regression to explore factors that contributed to this heterogeneity because of the small number of studies. As a result, the findings from pooled analyses should be interpreted with caution. However, we believe that providing a summary statistic to indicate service use, where possible, is more helpful than providing a narrative description of findings.

CONCLUSION

Adults with CP used a range of medical and rehabilitation services. The proportion of adults using services and the frequency of use varied considerably between services. This systematic review unfolds the context-specific challenges faced by adults with CP to access necessary services. The integrated findings demonstrated the complexities of health service use among adults with CP, and highlighted the variation and lack of equity in service provision for adults with CP. This review provides evidence of the need to develop appropriate service delivery models and educate service providers to meet the needs of adults with CP.

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DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: PRISMA diagram.

Table S1: Medical service use

Table S2: Rehabilitation service use

Appendix S1: Eligibility criteria.

Appendix S2: Search strategy.

Appendix S3: Andersen and Newman Model of health care utilization.

Appendix S4: JBI quality appraisal of studies.

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