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Factors associated with caregiver burden among family caregivers of children with cerebral palsy: a systematic review

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ABSTRACT

Objectives To identify caregiver and children factors associated with caregiver burden on primary caregivers of children with cerebral palsy (CP).

Design Systematic review

Data sources Seven electronic databases, including PubMed, Cochrane Library, Scopus, PsycINFO, Web of Science, CINAHL and Embase, were systematically searched up to 1 February 2023.

Eligibility criteria Original observational studies reporting caregiver burden and related factors among caregivers of children with CP.

Data abstraction and synthesis Two reviewers independently screened results and assessed the quality of studies. Title, abstract, full-text screening and data abstraction were done independently by two reviewers. Risk of bias was assessed using the JBI Critical Appraisal Checklist for Analytical Cross-Sectional Studies. The quality of evidence for factors was rated using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach.

Results 16 articles were included in the review. All studies were cross-sectional and examined caregiver-reported burden measures. The Zarit Burden Interview was the most commonly used questionnaire. Depression of caregiver and severity of illness in children with CP were moderate quality of evidence for factors contributing to caregiver burden.

Conclusions Higher caregiver burden is associated with more depressive feelings and worse life quality of the caregiver, and with more severe physical disability of the children. Future studies should focus on high-quality longitudinal research and appropriate assistance to reduce caregiver burden and improve the quality of caregiving for children with CP.

PROSPERO registration number CRD42021268284.

STRENGTHS AND LIMITATIONS OF THIS STUDY

- ⇒ This review was the first one to provide a comprehensive overview of caregiver burden of children with cerebral palsy.
- ⇒ The protocol was registered on the PROSPERO database (CRD42021268284) and was carried out based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines.
- ⇒ The quality of evidence for factors was judged by the Grading of Recommendations Assessment, Development and Evaluation approach.
- ⇒ Many survey factors of the total care burden were scattered and failed to provide reliable evidence.
- ⇒ The uneven quality of the included studies resulted in many methodological limitations.

INTRODUCTION

With the development of perinatal medicine and obstetric technology, the birth prevalence of cerebral palsy in high-income countries had declined to 1.6 per 1000 live births, while the prevalence in low-income and middle-income countries was as high as 3.4 per 1000 live births.[1] Cerebral palsy (CP) is defined as a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, cognition, communication, perception, behaviour and by a seizure disorder.[2] CP is the most common paediatric disability, seriously affecting the children's self-care ability and social activities.[3 4] Thus, children with CP often require lifetime extensive care and rehabilitation training, which will bring heavy psychological, physical and economic burden to their families.[5 6]

Parents, as the primary caregivers of children with disabilities, bear the arduous caregiving burden. Not only do they have to help the children's daily activities, but they also need to pay close attention to their changing health conditions, which directly leads to changes in the caregivers' lifestyle and a decline in their quality of life.[7 8] Caregiver burden has been defined as a multidimensional response to physical, emotional, psychological and financial stressors that are associated with caregiving experience.[9] Studies have shown that a high level of caregiving burden for children with CP negatively affects the mental and physical health, family functions and social interactions of caregivers, resulting in low quality of care and unmet patient needs.[10–13] Paying attention to the burden of caregivers is of great significance.

Relevant factors that affect caregiver burden should be studied in order to propose interventions to support caregivers. In the last decade, there has been an increasing number of studies on the care burden for children with CP, but a comprehensive review of caregiver factors and patient factors influencing caregiver burden is lacking. Therefore, the objective of this study was to systematically review published literature to identify caregiver and patient factors of caregiver burden among primary caregivers of children with CP.

METHODS

This systematic review was checked through the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (online supplemental appendix 1).

Search strategy

The electronic databases PubMed, Cochrane Library, Scopus, PsycINFO, Web of Science, CINAHL and Embase were systematically searched from inception to 1 February 2023. The search string contained four groups of combinations of medical subject headings and text words: Caregiver (MeSH term), care*, parents or mother; Caregiver burden (MeSH term), stress, strain or burnout; Cerebral palsy (MeSH term), CP, disability* or neurodev* disorder*; Children (MeSH term), child* or paediatr* (online supplemental appendix 2). In addition, we also searched for grey literature using the first 500 hits from Google Scholar and Open Grey. References for relevant publications were checked to make sure that no relevant papers had been missed.

Inclusion and exclusion criteria

Original observational studies reporting burden and related factors of caregivers of children with CP were taken as the primary focus of the research. Study participants need to be primary caregivers (over 18 years old) of children (0–18 years old) with CP. Burden had to be assessed with a total caregiver burden construct. Full-text articles published in English or with an English translation in peer-reviewed journals were included. Studies where caregivers of children with different diagnoses and children with adults are grouped together were excluded, unless results were reported separately for caregivers of children with CP. Intervention studies, reviews, non-original research papers and qualitative research studies were not eligible for this review.

Data extraction and quality assessment

The titles/abstracts of citations retrieved using the search strategy were independently assessed by two reviewers. The full-text articles of potentially relevant studies were then independently screened by two reviewers, and any discrepancies were resolved through discussion. The number of studies excluded during each screening phase and the reasons for full-text exclusion will be described in a PRISMA flow diagram. A pre-specified form will be used for the extraction of the data from the included studies. This procedure will be completed by one reviewer, and verified by a second reviewer, with any disagreements being resolved through discussion with a third-party reviewer. We extracted the following data from each included study independently by two researchers: first authors, year of publication, sample size, study design, relevant participant demographics, key predictor and outcome variables, self-administered tools used for measurement, results and associations.

Risk of bias was assessed using the JBI Critical Appraisal Checklist for Analytical Cross-Sectional Studies (online supplemental appendix 3) by the two researchers independently. The checklists include eight questions to evaluate the overall quality of the studies from the research object, disease, measurement of influencing factors and confounding factors, data analysis and so on. The choices of answers were 'yes', 'no', 'unclear' or 'not applicable'. The overall appraisal of 'include', 'exclude' or 'seek further information' were presented in the last section of the checklist.

Data synthesis

Due to the diversity of outcome measures and factors included in the study, a meta-analysis was not possible. Bivariate associations were described in terms of different statistical analysis. Multivariate associations were described as standardised or unstandardised coefficients (β *or* b). In studies that applied a logistic regression, the OR was presented. Factors were grouped into caregiver and children characteristics subsequently thematically categorised.

Quality of evidence

We assessed the quality of evidence for each factor measured in at least three studies using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach (online supplemental appendix 4). GRADE has four levels of evidence: very low, low, moderate and high. Evidence from observational data starts at low quality. For each of study limitation in risk of bias, imprecision, inconsistency, indirectness and publication bias, researchers have the option of rating down their level of certainty in evidence. Quality of evidence also can be rated up for 'large effect' or 'dose effect'.

Patient and public involvement

Patients or public were not involved in the conduct of this systematic review.

RESULTS

Search and screening

The search identified a total of 16 754 possibly relevant articles. After the removal of duplicates and the abstract screening, a total of 80 studies were selected for full-text

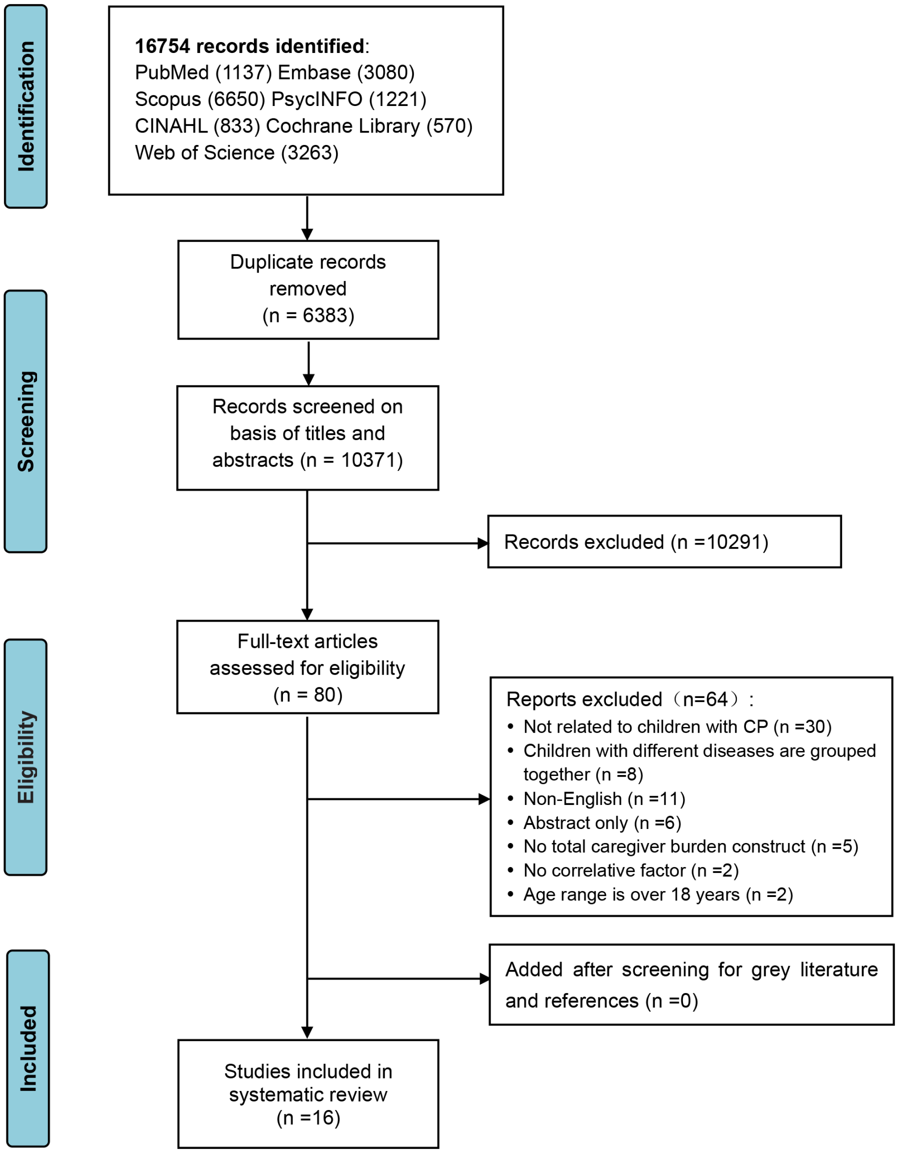

Figure 1 Flow diagram for a systematic literature review on caregiver burden in children with cerebral palsy (CP).

review. Finally, 16 studies were left for full inclusion in the review. There were no additional articles met the inclusion criteria when searching the references of the 16 articles. The PRISMA flow diagram for search and screening results are shown in figure 1.

Risk of bias

Risk of bias scores for studies ranged from 4 to 7 out of a maximum of 8 points. Some articles lacked clear information in items: study subjects and the setting, exposure factors and confounding bias. The methodological quality assessment list is listed in online supplemental appendix 5.

Characteristics of the included studies

The 16 included studies reported on overall caregiver burden and related factors of children with CP. Studies were conducted in 11 different countries between 2011 and 2022, involving in 3109 caregivers (not inclusive of controls). Of these, 14 studies investigated univariate associations; 9 studies were explored in multivariate associations. Caregivers were predominantly female, and 5 of the 16 articles had samples made up exclusively of mothers. Six articles involved fathers. The mean age of caregivers varied from 31 to 42 years. Four studies involved control populations, two of four had caregivers of healthy children as the control group. All studies were cross-sectional surveys. The most used sampling method was convenience sampling. The key characteristics of each study are presented in table 1.

Measures of burden

Seven instruments were used to measure caregiver burden: Zarit Burden Interview (n=7)[12 14–19]; Caregiver Difficulties Scale (n=4)[20–23]; Caregiver Burden Scale (n=1)[24]; The Revised Burden Measure (n=1)[25]; The Burden Interview (n=1)[26]; Daily Parenting Tasks Checklist–Parenting Burden (n=1)[27]; Viriyaprasart's questionnaire about burden of care (n=1).[8]

Table 1 Summary of included studies

Authors (year)	Country	Design	Caregiver sample size	Caregiver relation to child	Mean age of caregivers M (SD)	Age of children (years)	Measures tool
Carona *et al* (2013)[25]	Portugal	Cross-sectional study	93	Mothers (83.8%)	42.34 (5.72)	8–18	BM
Bella *et al* (2011)[26]	Brazil	Cross-sectional study	38	Mothers (–)	33.3 (7.7)	4–11	BI
Whittingham *et al* (2013)[27]	Australia	Correlational survey study	94	Mothers (90.4%)	–	2–12	DPC burden
Farajzadeh *et al* (2021)[20]	Iran	Cross-sectional study	160	Female (84.4%)	35.47 (6.59)	2–14	CDS
Boztepe *et al* (2019)[14]	Turkey	Cross-sectional study	69	Mothers (100%)	34.6 (7.3)	1–18	ZBI
Omole *et al* (2019)[21]	Nigeria	Cross-sectional study	209	Female (91.9%)	34.6 (9.2)	1–12	CDS
Terathongkum *et al* (2020)[8]	Thailand	Correlational research	75	Female (88.0%)	–	0–18	VQ burden
Farajzadeh *et al* (2020)[22]	Iran	Cross-sectional study	203	Mothers (100%)	34.48 (6.74)	4–14	CDS
Wijesinghe *et al* (2015)[23]	Sri Lanka	Cross-sectional study	375	Mothers (97%)	32.4 (7.2)	1–12	CDS
Barutcu *et al* (2021)[15]	Turkey	Cross-sectional study	109	Mothers (98.2%)	38.53 (9.62)	1–18	ZBI
Ozkan *et al* (2018)[16]	Turkey	Cross-sectional study	120	Mothers (100%)	–	2–18	ZBI
Gugała *et al* (2021)[24]	Poland	Cross-sectional study	190	Female (72.6%)	40.6 (9.1)	2–18	CBS
Marrón *et al* (2013)[17]	Spain	Cross-sectional study	62	Mothers (88.7%)	41.98 (5.64)	1–17	ZBI
Albayrak *et al* (2019)[12]	Turkey	Cross-sectional study	101	Mothers (100%)	34.93 (8.7)	0–18	ZBI
Santos *et al* (2012)[18]	Brazil	Cross-sectional study	21	Female (100%)	31 (–)	1–12	ZBI
Moriwaki *et al* (2022)[19]	Japan	Cross-sectional study	1190	Mothers (100%)	37.97 (5.28)	1–12	ZBI

BI, The Burden Interview; BM, The Revised Burden Measure; CBS, Caregiver Burden Scale; CDS, Caregiver Difficulties Scale; DPC Burden, Daily Parenting Tasks Checklist–Parenting Burden; VQ burden, Thai version Viriyaprasart's questionnaire about burden of care; ZBI, Zarit Burden Interview.

Studied factors in relation to caregiver burden

Overviews of caregiver and children characteristics as factors of caregiver burden are presented in tables 2 and 3. The detailed description is described as follows.

Caregiver factors

Caregiver factors were grouped into five categories, including caregiver demographics, emotional functioning, physical functioning, overall health and social functioning. Caregiver demographical factors included age,[8,12,14,15,21,23] gender,[21] educational level,[14,15,21,23] monthly income,[14,15,21,23] number of caregiver,[15] socioeconomic status,[21] geographical area,[23] mother employment,[19] additional helper[14] and daily caring time.[12] Emotional functioning factors included anxiety,[15,20,24] depression,[12,14,15,17,20,22,24] mental health,[12,22,25,26] perceived stress,[20,26] beliefs of rehabilitation,[8] perceived self-efficacy[8,17] and experiential avoidance.[27] This category was included in 11 of the 15 studies. Physical functioning factors included sleep quality,[12] pain,[12,26] fatigue[12,22] and physical health.[12,22,26] Overall health factors included quality of life.[8,25] Social functioning factors included social functioning,[22,26] environment health,[22] social support[8,19,25] and coping.[23,27]

Children factors

Children factors were grouped into five categories, including children demographics, disease related, behavioural functioning, overall health and social

Table 2 Associations between caregiver factors and caregiver burden

	Factor	**Measure factor**	**Outcome caregiver burden**	**Bivariate analysis**	Bivariate association **r/t/F/β/Z**	Multivariate association **β/b (95% CI)**	**N**	**Ref**
Demographics	Age	Years	VQ burden	P	ns	–	75	8
		Years	ZBI	P or S	0.20*	–	101	12
		Years	ZBI	–	–	ns	69	14
		Years	CDS	S	ns	–	209	21
		Years	CDS	R	ns	–	375	23
		Years	ZBI	P or S	ns	–	109	15
	Gender	Male/female	CDS	T	−2.01*	–	209	21
	Education	0–4 level	ZBI	–	–	ns	69	14
		0–2 level	CDS	A	4.95**	2.61** (1.97 to 14.09)	209	21
		Low education	CDS	R	ns	–	375	23
		0–5 level (mother)	ZBI	P or S	ns	–	109	15
	Monthly income	TL	ZBI	–	–	ns	69	14
		0–4 level	CDS	A	ns	–	209	21
		Low income	CDS	R	8.65***	3.69* (–)	375	23
		0–2 level	ZBI	P or S	4.45*	–	109	15
	No of caregivers	1/2	ZBI	P or S	ns	–	109	15
	Socioeconomic status	0–4 level	CDS	A	ns	–	209	21
	Geographical area	Rural area	CDS	R	6.27**	5.35** (–)	375	23
	Mother employment	Yes/no	ZBI	U	ns	–	1190	19
	Additional helper	None or present	ZBI	–	–	ns	69	14
	Daily caring time	Hours	ZBI	P or S	ns	–	101	12

Continued

Table 2 Continued

	Factor	Measure factor	Outcome caregiver burden	Bivariate analysis	Bivariate association r/t/F/β/Z	Multivariate association β/b (95% CI)	N	Ref
Emotional functioning	Anxiety	HADS-A	CDS	P	0.47***	0.14*** (nr)	160	20
		BAI	ZBI	P or S	0.29**	–	109	15
		HADS-A	CBS	P	0.51****	–	190	24
	Depression	BDI	ZBI	P or S	0.70***	–	101	12
		HADS-D	CDS	P	0.51***	0.17*** (nr)	160	20
		BDI	ZBI	–	–	0.48*** (nr)	69	14
		BDI-II	CDS	P	0.64***	–	203	22
		BDI-II	ZBI	P or S	0.41****	–	109	15
		HADS-D	CBS	P	0.38****	–	190	24
		BDI-II	ZBI	–	–	0.36** (0.17 to 0.73)	62	17
	Mental health	SF-36-MCS	ZBI	P or S	ns	–	101	12
		MHI-5	BM	nr	–0.51**	–0.20* (nr)	93	25
		SF-36-RE	BI	S	ns	–	38	26
		SF-36-MH	BI	S	–0.62*	–	38	26
		WHOQOL-BREF-PsyH	CDS	P	–0.68***	–	203	22
	Perceived stress	PSQ	BI	S	0.65***	–	38	26
		PSS-4	CDS	P	0.43***	0.10*** (nr)	160	20
	Beliefs of rehabilitation	RBQ	VQ burden	P	ns	–	75	8
	Perceived self-efficacy	PSQ	VQ burden	P	–0.25*	–	75	8
		RSCSE	ZBI	–	–	–0.19** (–0.31 to –0.06)	62	17
	Experiential avoidance	AAQ-16	DPC burden	nr	0.42****	0.31** (0.52 to 1.21)	94	27
Physical functioning	Sleep quality	PSQI	ZBI	P or S	0.39***	–	101	12
	Pain	NRS	ZBI	P or S	0.36***	–	101	12
		SF-36-BP	BI	S	–0.40*	–	38	26
	Fatigue/vitality	CIS	ZBI	P or S	0.66***	–	101	12
		FSS	CDS	P	0.40***	–	203	22
		SF-36-VT	BI	S	–0.54*	–	38	26
	Physical health	SF-36-PCS	ZBI	P or S	–0.21*	–	101	12
		SF-36-PF	BI	S	ns	–	38	26
		SF-36-RP	BI	S	ns	–	38	26
		SF-36-GH	BI	S	–0.44*	–	38	26
		WHOQOL-BREF-PH	CDS	P	–0.59***	–	203	22
Overall health	Quality of life	VQ well-being	VQ burden	P	–0.50**	–	75	8
		WHOQOL-8	BM	nr	–0.39**	–0.16* (nr)	93	25

Continued

Table 2 Continued

	Factor	Measure factor	Outcome caregiver burden	Bivariate analysis	Bivariate association r/t/F/β/Z	Multivariate association β/b (95% CI)	N	Ref
Social functioning	Social functioning	SF-36-SF	BI	S	−0.61*	–	38	[26]
		WHOQOL-BREF-SR	CDS	P	−0.68***	–	203	[22]
	Environment health	WHOQOL-BREF-E	CDS	P	−0.63***	–	203	[22]
	Social support	SSS	BM	nr	−0.35**	−0.41** (nr)	93	[25]
		SSQ	VQ burden	P	ns	–	75	[8]
		Family support	ZBI	U	ns	–	1190	[19]
		Others support	ZBI	U	nr**	–	1190	[19]
		Home-visit nursing	ZBI	C	nr**	–	1190	[19]
		Home care	ZBI	C	nr*	–	1190	[19]
		Home-visit rehabilitation	ZBI	C	nr**	–	1190	[19]
		Training and treatment	ZBI	C	ns	–	1190	[19]
		Short stay	ZBI	C	ns	–	1190	[19]
		Mobility support	ZBI	C	nr*	–	1190	[19]
		Transportation services	ZBI	C	ns	–	1190	[19]
		No social support usage	ZBI	C	nr*	–	1190	[19]
		Satisfaction with home-visit nursing	ZBI	C	ns	–	1190	[19]
		Satisfaction with home care	ZBI	C	nr*	ns	1190	[19]
		Satisfaction with home-visit rehabilitation	ZBI	C	nr**	ns	1190	[19]
		Satisfaction with training and treatment	ZBI	C	nr**	ns	1190	[19]
		Satisfaction with short stay	ZBI	C	nr**	−0.29*	1190	[19]
		Satisfaction with mobility support	ZBI	C	ns	–	1190	[19]
		Satisfaction with transportation services	ZBI	C	ns	–	1190	[19]
	Coping	CHIP	DPC Burden	nr	ns	ns	94	[27]
		Individual coping	CDS	R	ns	–	375	[23]
		Seeking social support	CDS	R	−8.99***	ns	375	[23]
		Spouse support	CDS	R	−7.44***	−5.67** (nr)	375	[23]

****p<0.0001, ***p<0.001, **p<0.01, *p<0.05.

A, one-way analysis of variance (ANOVA); AAQ-16, Acceptance and Action Questionnaire; BAI, Beck Anxiety Inventory; BDI, Beck Depression Inventory; BDI-II, Beck Depression Inventory-II; C, chi-squared test; CHIP, Coping Health Inventory for Parents; CIS, Multidimensional checklist individual strength; FSS, Fatigue Severity Scale; HADS, Hospital Anxiety and Depression Scale; HADS-A, HADS-Anxiety Scale; HADS-D, HADS-Depression Scale; MHI-5, Mental Health Inventory-short form; nr, not reported; NRS, numerical rating scale; ns, not significant; P, Pearson correlation coefficient; PSQ, Perceived self-efficacy in the care questionnaire; PSQI, Pittsburgh Sleep Quality Index; PSS-4, Perceived Stress Scale-4 item; R, univariate regression analysis; RBQ, adapted Rehabilitation Belief Questionnaire; RSCSE, Revised Scale for Caregiving Self-Efficacy; S, Spearman's linear correlation coefficient/Spearman's Rho correlation; SF-36, 36-Item Short Form Health Survey; SF-36-BP, SF-36-Bodily pain; SF-36-GH, SF-36-General health; SF-36-MCS, SF-36-Mental component summary; SF-36-MH, SF-36-Mental health; SF-36-PCS, SF-36-Physical component summary; SF-36-PF, SF-36-Physical functioning; SF-36-RE, SF-36-Emotional role; SF-36-RP, SF-36-Physical role; SF-36-SF, SF-36-Social functioning; SF-36-VT, SF-36-Vitality; SSQ, adapted Social Support Questionnaire; SSS, Satisfaction with Social Support Scale; T, independent Student's t-test; U, Mann-Whitney U test; VQ well-being, Thai version Viriyaprasart's questionnaire about well-being; WHOQOL-8, WHO Quality of Life Assessment 8 item index; WHOQOL-BREF, WHO's Quality of Life Questionnaire; WHOQOL-BREF-E, WHOQOL-BREF-Environment; WHOQOL-BREF-PH, WHOQOL-BREF-Physical health; WHOQOL-BREF-PsyH, WHOQOL-BREF-Psychological health; WHOQOL-BREF-SR, WHOQOL-BREF-Social relationships.

Table 3 Associations between children factors and caregiver burden

	Factor	Measure factor	Outcome caregiver burden	Bivariate analysis	Bivariate association r/t/F/β/Z/H/X^2	Multivariate association β/b (95% CI)	N	Ref
Demographics	Age	Years	VQ burden	P	ns	–	75	8
		Years	ZBI	P or S	ns	–	101	12
		Years	ZBI	–	–	ns	69	14
		Years	CDS	S	ns	–	209	21
		Years	CDS	R	ns	–	375	23
		1–5/6–12/>12 years old	ZBI	P or S	ns	–	109	15
		Years	ZBI	U	ns	–	1190	19
	Gender	Male/female	ZBI	–	–	ns	69	14
		Male child	CDS	T	3.02**	0.18** (1.56 to 8.34)	209	21
		Male child	CDS	R	3.60*	3.50* (–)	375	23
		Male/female	CBS	P or S	ns	–	109	15
	Going to nursery/school	Yes/no	ZBI	U	ns	–	1190	19
	Residence	Urban/rural	ZBI	U	nr****	–	1190	19
	Birth order	1st/2nd/3rd or greater	CDS	A	ns	–	209	21
		First/or not	ZBI	U	ns	–	1190	19
	No of children	3 or higher	ZBI	–	–	ns	69	14
		1/2/3	CDS	R	ns	–	375	23
		1/2/3/4 or more	ZBI	P or S	0.20*	–	109	15
		1/or not	ZBI	U	ns	–	1190	19
	No of disabled children	1/or not	ZBI	P or S	ns	–	109	15
	Body mass index	BMI	ZBI	P or S	ns	–	101	12

Continued

Table 3 Continued

	Factor	Measure factor	Outcome caregiver burden	Bivariate analysis	Bivariate association r/t/F/β/Z/H/X^2	Multivariate association β/b (95% CI)	N	Ref
Disease related	Type of CP	5 types	CDS	A	3.15*	–	209	[21]
		Spastic quadriplegia	CDS	R	10.62***	–	375	[23]
	Severity of illness	GMFCS	ZBI	P or S	0.30**	–	101	[12]
		Severity of illness scale	ZBI	–	–	0.29* (nr)	69	[14]
		GMFCS-ER	CDS	A	6.87***	0.29*** (4.42 to 11.23)	209	[21]
		nr	ZBI	–	–	0.28* (0.04 to 0.35)	62	[17]
		3 levels	ZBI	H	nr**	ns	1190	[19]
	Tube/intravenous nutrition	Yes/no	ZBI	C	nr*	ns	1190	[19]
	Aetiology	7 types	CDS	A	2.49*	–	209	[21]
	Additional physical illness	Absent/present	ZBI	–	–	ns	69	[14]
	Associated conditions	0/1/2/3/4 or more	CDS	A	3.21*	–	209	[21]
		0/1 or more	CDS	R	12.75***	ns	375	[23]
	Functional impairments	No of functional deficits	CDS	R	3.97***	2.64*** (nr)	375	[23]
Behavioural functioning	Functional status	FIM	ZBI	P or S	ns	–	109	[15]
	Behavioural problems	SDQ	BM	nr	0.24*	0.29**	93	[25]
		SDQ	DPC burden	nr	0.38****	0.27** (0.06 to 1.43)	94	[27]
Overall health	Quality of life	KIDSCREEN-10	BM	nr	ns	–	93	[25]
		PedsQL	ZBI	P	0.40***	–	120	[16]
Social environment	Social support	SSSS	BM	nr	ns	−0.19* (nr)	93	[25]
	Environment	Space for child locomotion	ZBI	P	nr*	–	21	[18]

****p<0.0001, ***p<0.001, **p<0.01, *p<0.05.

A, one-way analysis of variance (ANOVA); BMI, Body Mass Index; C, chi-squared test; CP, cerebral palsy; FIM, Functional Independence Measure; GMFCS, Gross Motor Function Classification System; GMFCS-ER, Gross Motor Function Classification System–Expanded and Revised; H, Kruskal-Wallis H; KIDSCREEN-10, The shortest version of Kidscreen questionnaires; nr, not reported; ns, not significant; P, Pearson correlation coefficient; PedsQL, Pediatric Quality of Life 4.0 Generic Core Scales; R, univariate regression analysis; S, Spearman's linear correlation coefficient/Spearman's Rho correlation; SDQ, Strengths and Difficulties Questionnaire; SSSS, Satisfaction with social support scale for children and adolescents; T, independent Student's t-test; U, Mann-Whitney U test.

environment. Children demographical factors included age,[8 12 14 15 21 23] gender,[14 15 21 23] going to nursery/school,[19] residence,[19] birth order,[21] number of children,[14 15 23] number of disabled child[15] and body mass index.[12] Disease-related factors included type of CP,[21 23] severity of CP,[12 14 17 21] tube/intravenous nutrition,[19] aetiology,[21] additional physical illness,[14] associated conditions[21 23] and functional impairments.[23] This category was included in 5 of the 15 studies. Behavioural functioning factors included functional statu[15]wehavioural problems.[25 27] Overall health and social environment factors included quality of life,[16 25] social support[25] and environment.[18]

Quality of evidence for potential factors

Quality of evidence for potential factors of caregiver burden are presented in online supplemental table. The criteria for compelling evidence are that each factor was measured in at least three studies that reported significant relationships. The details are as follows.

Caregiver factors

The results of evidence synthesis for caregiver factors using the GRADE criteria were that 'depression' was the moderate quality of evidence for factor of caregiver burden. 'Age', 'anxiety' and 'fatigue' of caregivers was the low quality of evidence for factors of caregiver burden. Very low quality of evidence was found for the relationship between caregiver burden and 'education', 'monthly income', 'mental health', 'physical health' and 'social support' of caregivers.

Children factors

The synthesis of the evidence for children factors resulted in moderate quality of evidence between caregiver burden and 'severity of illness'. The factors for 'age' and 'gender' of children, and 'number of children' were the low quality of evidence for the relations with caregiver burden. Few of the included studies have explored children factors, and fewer factors have been able to perform evidence synthesis.

DISCUSSION

Caring for children with CP is a stressful and difficult task for primary caregivers, especially for their parents. Caregivers of children with CP were under higher pressure than caregivers of healthy children.[12 28 29] There were many factors that affect the caregiving burden for children with CP. In our systematic review, we focused on both caregiver characteristics and children characteristics as factors of caregiver burden. There are fewer studies involving children-related factors than caregiver-related factors, possibly because caregiver-related factors are more likely to modify. This review revealed that anxiety and depression of caregiver, and severity of illness in children with CP were moderate quality of evidence for factors contributing to caregiver burden. However, our ability to draw conclusions on remaining caregiver and children characteristics as factors is limited because of low to very low quality of evidence.

Moderate quality was found for the association between caregiver burden and depression symptoms of the caregiver. Feelings of depression are positively correlated with caregiver burden of the caregiver. However, the factor for mental health predicting psychological well-being of the caregiver as well was rated as very low quality of evidence. This may be due to the limited number of studies that did not yield reliable evidence. Three studies included in this review revealed that caregivers, who experience poor psychological condition, are more likely to experience high caregiver burden. This correlation between mental health of caregiver and caregiver burden is consistent with the findings in other children with neurodevelopmental diseases, such as epilepsy and autism spectrum disorder.[30–32] Mother caregivers constitutes the majority of caregiving. Long-term care of children with CP and restricted social activity for the mother caregiver predict more psychological conditions such as anxiety, depression and distress in them.[33] Caregivers who feel depressed experience a greater burden of care, thereby affecting CP children's quality of life.[16] A study revealed that psychological interventions such as Stepping Stones Triple P therapy, and Acceptance and Commitment Therapy could improve parental mental well-being of parents of children with CP. It is crucial to develop more interventions that focus primarily on the psychological well-being of the parents.[34]

Low quality of evidence was found for the relationship between caregiver burden and fatigue of the caregiver. Fatigue of caregivers are positively related to caregiver burden. Children with CP are often accompanied by intellectual and physical disabilities. Daily life care and assistance with rehabilitation exercises mean more physical strength and energy for the caregiver, so that caregivers often experience problems such as body pain, fatigue and sleep problems,[12 35–38] which seriously affect the physical well-being and reduce the quality of life of the caregiver.[39 40] Few studies have focused on interventions for caregivers' physical conditions. We should actively pay attention to the physical health of the caregiver and take feasible interventions to improve the physical functioning of caregivers of children with CP.

In our systematic review, it is not sufficiently concluded that social support for caregivers is a positive factor in reducing the burden. However, many previous studies have explored the correlation between social support and parenting stress in caregivers of children with CP. Negative associations were found between the levels of stress among parents and social support perceived by them.[41–44] Many families faced financial difficulties and restricted access to healthcare services.[45 46] Social support from spouses, social organisation and medical institution help prevent and decrease the stressful situation of the family's experience in providing care to children with

CP.[43 47] Although parenting stress and caregiver burden have similar meanings, this review focuses on caregiver burden and involves fewer relevant articles about the association between social support and care burden, so more evidence is needed to determine their relevance.

The factor for severity of illness in CP children as moderate quality of evidence is positively correlated with the burden of caregiver. Gross Motor Function Classification System score is most commonly used to measure the severity of disability of children with CP. Children with more severe disabilities require the more support of their caregivers to carry out the activities of daily living. Caregivers will spend more time and physical strength to meet the caregiving demands,[38 48] and the caregiver burden will be heavier. In addition, there was no significant correlation between the age of CP children and the caregiver burden. Also, the gender of children with CP may affect the caregiver's burden. The burden of care for male child is greater than that of female child, probably because of the greater mobility of male children. At present, there are many researches on motor function intervention for children with CP. A guideline synthesised approaches to functional exercise in walking ability and hand mobility in children, indicating whole-task practice combined with assistive devices could increase independence and reduce caregiver burden.[49]

This systematic review offers insight into factors related to caregiver burden and guides the supportive interventions aiming to reduce caregiver burden, but more additional research into factors associated with caregiver burden is needed.

Strengths and limitations

This review was the first one to provide a comprehensive overview of caregiver burden of children with CP. The protocol was registered on the PROSPERO database and was carried out based on the PRISMA guidelines. The quality of evidence for factors was judged by the GRADE approach. There were also some limitations of the review. First, the included literature used different measurement tools, and the survey factors of the total care burden with/without the burden of each dimension were scattered. Second, due to the uneven quality of the included studies, the quality of the literature is at a medium level, and convenience sampling is often used, which has many methodological limitations.

CONCLUSION

Caregivers of children with CP generally have a heavy burden of caregiving, which has affected the physical, psychological, social and economic conditions of the caregivers. We summarised factors related to caregiver burden from the aspects of caregiver factors and children factors. The results revealed that depression of caregiver and severity of illness in children with CP were moderate quality of evidence for factors contributing to caregiver burden. At present, the relevant literature on the factors affecting the caregiver burden of children with CP are mostly cross-sectional studies, and there is a lack of longitudinal studies with high demonstration efficiency. In the future, we should focus on carrying out high-quality longitudinal research and verifying the relevant influencing factors of caregiver burden of children with CP. Moreover, by identifying all the factors, healthcare professionals can provide appropriate assistance to relieve caregiver burden and improve the quality of caregiving for children with CP.

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