Pediatric Rheumatology

REVIEW

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A mixed-method systematic review evaluating interventions in paediatric rheumatology to address caregiver support and well-being

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Abstract

Background Paediatric rheumatology conditions are chronic autoimmune and auto-inflammatory conditions that affect predominantly the musculoskeletal systems of children and young people (CYP). About 6–7 million children are estimated to be affected worldwide. Having a rheumatological condition affects CYP as well as the wider family. Negative outcomes on psycho-social well-being, quality of life, family relationships and family functioning are commonly observed. The current review addresses interventions for caregivers of CYP with paediatric rheumatological conditions.

Methods The Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) 2020 statement guided this review. Eligibility criteria were pre-defined and registered in the International Prospective Register of Systematic Reviews (PROSPERO). Articles were included if they (1) were targeted at caregivers of CYP with rheumatological conditions, and (2) included an intervention to improve well-being of caregivers.

Results Of 1065 identified studies, 15 studies were included in the final review. A mixed-method systematic review was conducted, and included literature was assessed using the Mixed-Method Appraisal Tool (MMAT). Quantitative, qualitative and mixed methods studies, as well as review articles and abstracts investigating the effectiveness of caregiver interventions were evaluated. A third of the identified literature did not report on outcome measures. A narrative synthesis was employed to appraise interventions tailored at caregivers.

Conclusions Despite evidence suggesting that a family approach is needed to support caregivers and CYP with rheumatological conditions as well as the wider family to improve health outcomes for the child, increase family functioning, reduce family conflict, and increase psycho-social well-being, only a small number of caregiver interventions have been carried out to date. The review highlights the need for caregiver interventions to be appraised to better understand what interventions yield results that lead to better quality of life for families who are caring for a child with a chronic rheumatological condition.

Trial registration PROSPERO CRD42024524570.

Keywords Intervention, Parent, Caregiver, Quality of Life, Psychosocial Factors, Mental Health, Family

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Background

Paediatric Rheumatology conditions are chronic multisystemic inflammatory conditions affecting predominantly the musculoskeletal system [1]. Six to seven million children are estimated to have a rheumatological condition worldwide [2]. Rheumatological conditions affect children and young people (CYP) in different ways due to a heterogenous presentation of symptoms with some children experiencing joint inflammation, pain, and/or fatigue [3], whilst others present with unexplained fevers or rashes [1] or even systemic involvement which impairs a child's overall health especially when internal organs are affected [4]. Prevalence rates vary between conditions as some paediatric rheumatology conditions are common whilst others are rare. The most common condition is Juvenile Idiopathic Arthritis (JIA) which affects approximately one in 1000 children [4]. There are no known cures for paediatric rheumatological conditions, and available treatments aim to reduce as well as manage symptoms [5] with the goal to achieve inactive disease (remission) [6]. Chronic conditions (which include rheumatology conditions) have an impact on CYP's physical and social life and are associated with significant stress as well as emotional and behavioural problems [7, 8].

Living with a rheumatological condition is often associated with frequent hospital appointments which has implications on education and schooling as well as peer relationships for CYP. Adapting to a new normal and learning to cope with a chronic health condition and treatment regimens causes challenges for CYP and their families. CYP with rheumatological conditions experience higher levels of pain, fatigue, physical disabilities and psychological problems [9, 10]. A retrospective cross-sectional survey conducted in the United States looking at data from 2008 through to 2013 as part of the Medical Expenditure Panel Survey (MEPS) found that CYP with a chronic physical condition were 62% more likely to have a diagnosis of a mental health disorder [11], while other research suggests that CYP with rheumatological conditions are 15% to 65% more likely of having a mental health disorder such as anxiety or depression [12]. The additional psychological burden on CYP in turn exacerbates adverse outcomes. Mental health disorders are associated with decreased quality of life [10, 13], increased pain and disease activity [9, 13], lower physical activity levels and reduced functional ability [14], greater functional disability [9], poor disease control [15], medication non-adherence [16] and transition challenges [17, 18].

Moreover, it is well established that the impact upon the wider family can be profound, as caring for a CYP with a rheumatological condition is challenging, especially for caregivers [19]. Research showed that 82% of caregivers reported that their child's diagnosis had an impact on their own mental health [20]. Furthermore, caregivers feel that a diagnosis has major repercussions on their emotional well-being, social, economic, and work life [21]. Caring for a child with a rheumatological condition can affect a caregiver's quality of life and their mood, and is associated with higher levels of worry and decreased family functioning [22].

Caregivers tend to experience a roller-coaster of feelings throughout their child's diagnosis journey [23]. They often struggle to adjust to the 'new normal' after receiving a diagnosis [24, 25], and they experience feelings of sadness, denial [21], anxiety, and fear [26]. However, some caregivers have also expressed relief at receiving a diagnosis with caregivers having encountered disbelief relating to their child's symptoms in the beginning of their journey which led to a loss of trust in health professionals [21, 25]. Feelings of anxiety, confusion and powerlessness continue throughout the disease journey as caregivers learn to accept their child's lifelong disease [23]. Additionally, caregivers often encounter negative feelings of frustration and fatigue relating to ongoing treatments, disease flares, pain and limitations to their child's life but also admiration for their child's resilience as well as feelings of hope when things are looking up [23].

One of the biggest challenges for caregivers is worrying about their child's quality of life [21], due to a lack of understanding of the condition [24] which is exacerbated by a lack of information [26] and uncertainty about what the future holds [21, 25]. Some caregivers report that as the disease progresses, CYP can demonstrate challenging behaviours such as non-adherence to medication, with caregivers then struggling to find the balance with allowing their adolescents' autonomy in relation to monitoring their medication adherence [24].

Another aspect impacting on parents' caregiving abilities when caring for a child with a chronic condition is uncertainty. Parents have expressed uncertainty relating to their child's condition and treatment management, however, the perceived uncertainty went beyond the medical aspects of their child's condition. They also expressed uncertainty relating to their role as parents and their own coping abilities [27]. Parental uncertainty relating to their child's disease is associated with parent and CYP distress [28, 29]. Furthermore, parental distress is associated with depressive symptoms in children which is further amplified if the child is experiencing illness intrusiveness which refers to a child's belief that their illness is impacting on their everyday life and functioning [30]. Child reported pain and fatigue are also negatively associated with family functioning impacting upon family activities and communication [31].

Another problem that families face relates to the lack of awareness of rheumatological diseases, and this lack of social awareness is further magnified by the lack of information that is available to caregivers [27]. Caregivers have expressed a need for support with school advocacy relating to school absenteeism due to their child's symptoms and hospital appointments [24]. Caregivers also encounter frustrations when dealing with their child's 'hidden disease' during absences of symptoms and are faced with ongoing struggles surrounding teachers' and peers' as well as wider family members' lack of understanding of the disease [23]. These school problems often lead to worries about the future transition to further education, for example, college, and work opportunities [24].

Making sure caregivers are able to cope with their child's diagnosis and understand how to better support their CYP is important to mitigate some of the challenges that CYP with paediatric rheumatology conditions and their families face on a daily basis. However, the well-being of caregivers is not always considered or prioritised. Therefore, the objective was to systematically review and evaluate existing interventions tailored at providing support for caregivers of CYP with rheumatological conditions and assess the impact of these on improving well-being.

Methods

The present systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) 2020 statement [32].

Eligibility criteria

Eligibility criteria were pre-defined and registered in the International Prospective Register of Systematic Reviews (PROSPERO, PROSPERO ID: CRD42024524570). Articles were eligible for inclusion if they (1) included caregivers of CYP with any rheumatological condition, and (2) included interventions targeted at caregivers. Articles were excluded if the interventions were not targeted at caregivers. For example, some interventions were identified in the screening process that were targeted at CYP but also tested by caregivers. If the caregiver was not the target population for the intervention, the article was not included in this review. Only articles written in English or those that included an English translation were included. Review papers, abstracts as well as grey literature were reviewed and included in the systematic review if they met the eligibility criteria.

Search strategy

Eligible articles were identified in October and November 2023 by searching electronic databases which included Embase (Ovid), Scopus, Medline (Ovid), Web of Science Core Collection and CINAHL (EBSCOhost). A Google Scholar search as was conducted in April and May 2024 and internet resources from rheumatology charities and societies were reviewed but no additional articles were identified. When screening Google Scholar results, only the first 1000 identified results have been reviewed. Google Scholar was used as an additional search to look for any potentially missed literature. Google Scholar offered a vast amount of literature to review and on average different searches have shown that beyond 10–20 pages (with 10 results per page), search results were often not relevant anymore and there was a lot of overlap with the same articles being repeatedly shown across several searches. When the literature was not matching keywords from the searched terms for more than 2 consecutive pages, the search was abandoned, and new search terms have been applied. The complete search strategy can be found in Additional file 1.

Study selection

All references from the database searches were imported into Covidence [33]. Covidence is an online platform that helps facilitate the management of the literature that was obtained from the database searches. It allows reviewers to complete the study selection process systematically by having access to the literature on one platform. The whole study selection process from sifting through titles and abstracts as well as full texts and resolving conflict can be completed via this tool. Initially, the screening was based on titles and abstracts. Two independent reviewers screened all titles and abstracts based on the inclusion and exclusion criteria listed above (always KK, and either AN, JL or PL). On occasions where consensus was not reached, a team decision was sought to decide on whether to include the article. If a decision could not be made based on the title and abstract alone, the article was included for full text review to allow a more in-depth review of the article. The same rigour was applied to the full text review with two independent reviewers assessing each article (KK, AN, JL or PL), and a team decision was sought on occasions where consensus could not be reached.

Methodological quality assessment

The Mixed Methods Appraisal Tool (MMAT) [34] was used to assess risk of bias and quality [35]. Two independent reviewers (KK & AN) assessed the quality of the 15 articles that were included in the final review. In the MMAT, there are initial screening questions relating to the research question and data collection, and these helped inform whether a study was suitable for assessment using the MMAT. Consensus was reached between

the reviewers using the MMAT, so no further actions were required.

Data extraction

Data extraction was based on predefined characteristics. Data extracted from the studies included first author, year of publication, study title, location (country and region of the intervention), setting (such as primary, secondary, tertiary care, community health or urban/rural/regional where applicable), population (including demographics such as age, gender, condition, socioeconomic status, ethnicity), study design, study aim(s), details of the intervention (type, aim of use, duration), outcome measures employed, and outcomes and main findings.

Data synthesis

Data were synthesised narratively due to the heterogeneity of study designs. Studies were grouped together based on type of intervention (e.g. online peer support, retreat, parent consultant programme, webpage), as well as study design (e.g. quantitative, qualitative, mixed method designs). Narrative synthesis is an approach that aims to summarise and explain results and findings from a range of different studies that employ diverse methods, populations and/or measures, and cannot be synthesised via standardised approaches such as a statistical meta-analysis or other comprehensive syntheses [35].

Results

Study selection

Fifteen studies were included in the final review. Interventions that were providing support to caregivers of CYP that went above providing solely education were deemed eligible. The search strategy identified 1054 studies for review once duplicate entries were removed (N = 11). The flow diagram for the whole search strategy can be found in Fig. 1.

Study characteristics

Table 1 provides a description of the final set of caregiver interventions that were included in the present review (N = 15). Studies were grouped together based on the type of intervention (online peer support, retreat, parent consultant programme, website). Eight of the interventions were published as an abstract [36–43]. Of those 8 abstracts, only 2 presented data [40, 43]. Eight studies have evaluated quantitative results [40, 42–48]. Of those eight studies, one did not collect any data [42], one was still collecting data [43], three reported on quantitative descriptive results [40, 44, 46], one was a non-randomised study [47], and four were randomised-controlled trials (RCT) [42, 43, 45, 48]. Two studies used mixed methods designs, analysing both quantitative and qualitative data [49, 50]. Four studies in this review focused on the same intervention (a website) that has been disseminated in different publications highlighting various milestones of the intervention [41–43, 48].

Methodological quality

It is recommended to provide an overview of all criteria instead of providing an overall score when using the MMAT [34]. Table 2 shows the outcomes of the MMAT quality review of the 15 articles. The MMAT facilitates a review and appraisal of quantitative, qualitative and mixed method study designs, but is not suitable for reviews and theoretical papers [34], 5 abstracts could therefore not be reviewed past the initial screening [36–39, 41].

Synthesis of results

Narrative synthesis was used as outlined by Popay [51] to analyse the data due to heterogeneity of results and outcome measures that were employed. Five abstracts of the 15 studies that were included in this review could not be analysed as data collection did not take place [36–39, 41]. Three of the included studies that relied on quantitative measures used non-standardised questionnaires, and relied on surveys and feedback forms as well as predefined questions [40, 46, 49]. Six studies using quantitative measures relied on a range of standardised questionnaires [43–45, 47, 48, 50]. Different outcomes measures were employed across these studies. To assess self-reported psychological distress of caregivers the Symptom Checklist- 90-Revised (SCL- 90-R) [44] was used. To measure perceived mental health symptoms the Hospital Anxiety Depression Scale (HADS) [50], or the Psychiatric Symptom Index (PSI) [45] was used. To assess parenting stress the Pediatric Inventory for Parents (PIP) was employed [43, 48], and to assess perceived disability-related stress the Parents of Children with Disabilities Inventory (PCDI) [50] was chosen. Additionally, one study relied on translated and adapted measures to assess perceived support using the Iceland Family Perceived Support Questionnaire (ICE-FPSQ) [47], to assess illness beliefs using the Icelandic-Family Illness Beliefs Questionnaire (ICE-FIBQ) [47], to assess the impact on the family using the PedsQL Family Impact Module [47], and to assess healthcare satisfaction using the PedsQL Healthcare Satisfaction Generic Module [47]. None of the studies (N= 15) relied on qualitative data only, and of the two designs that used a mixed-methods approach, only one passed the quality assessment from the MMAT as can be seen in Table 2. Therefore, no further synthesis was feasible.

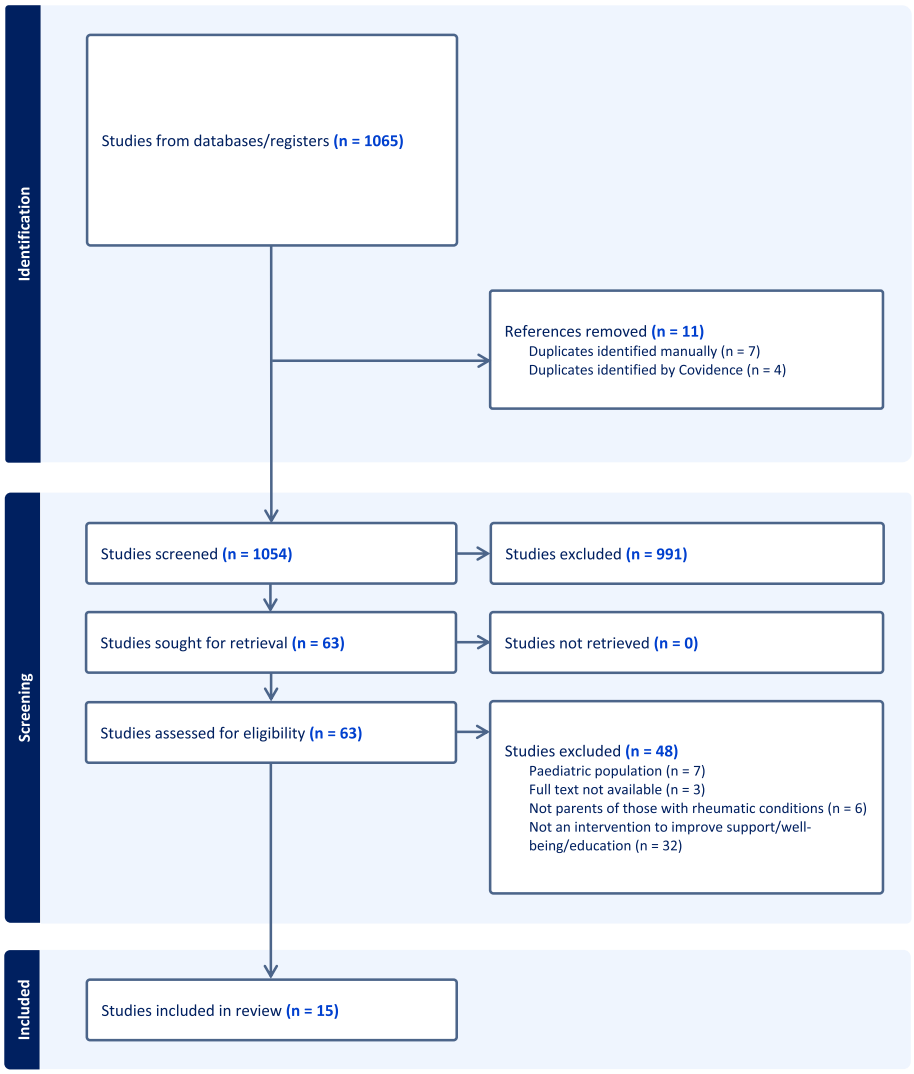

Fig. 1 Flow diagram of article search strategy

Discussion

This review highlights the need for interventions tailored at caregivers of CYP with rheumatological conditions. Our findings show that there is limited support offered to caregivers in the first instance, and the few interventions that were identified in this review have not always been suitably assessed. There are various means in which support was offered to caregivers which involved peer support (via Facebook groups, meet ups, residential workshops, or in clinics), websites and tailored information sessions, and on one occasion a Family Strengths Oriented Therapeutic Conversation (FAM-SOTC) intervention. However, a third of the studies that were identified in this review have not been formally assessed.

Understanding the implications of caregiver interventions is important to help mitigate some of the challenges that families face daily, but also to improve outcomes for CYP and their families. A mixed-method systematic review by Knafl et al. [52] that evaluated 29 articles showed a moderate positive association between caregivers' and children's psychological functioning highlighting the complementary relationship between caregivers' and CYP's well-being. They also found negative associations between family conflict and psycho-social well-being of children as well as parental depression and children's physical functioning [52]. This suggests that by supporting the caregiver and improving family functioning, positive implications for both the caregiver as well as the CYP should be observed and may have further advantageous implications on the wider family. Moreover, even though some interventions that were evaluated in this review have

Table 1 Extracted information of included literature ($N = 15$)

First author, Year of publication [ref.], Study title, Location	Setting Population	Study Design Study aim(s)	Details of the intervention Outcome measure(s) employed	Outcomes and main findings
Markus, 2014 [36] *The Network of Parents with Children Who Have Arthritis in Germany.* Germany	*Not specified* *No demographic information available*	*N/a not a research study* The German Network – Deutsche Rheuma-Liga Bundesverband - provides support to caregivers of children with rheumatological conditions	The German Network has several representatives for various associations nationwide and the network offers a range of different information and support services such as workshops, telephone hotlines and newsletters *No outcome measures employed.*	*N/a, no data collected*
Marchal, 2016 [37] *Info Sessions for (Future) Parents with RMDs and for Parents with Children with Juvenile Arthritis.* Belgium	*Not specified* *No demographic information available*	*N/a not a research study* To offer information for parents of a child with a rheumatic condition (RMD) to help families when they receive a diagnosis. The information session was also tailored at young people with an RMD with a child wish.	ReumaNet, a Flemish organisation for people with RMDs carried out two sessions to provide tailor made information. The first session was a discussion session where families discussed their concerns. During the second session health professionals provided information and gave advice on how to support their child better. *No outcome measures employed.*	*N/a, no data collected*
Eveleigh, 2018 [38] *Parent support via facebook group* Nottingham, UK	Online facebook page, recruitment via a paediatric rheumatology clinic ($N = 1$) Caregivers ($N = 33$)	To create a facebook page to offer peer support	A clinical nurse specialist (CNS) had informal discussions with families about creating a social media platform and a facebook page for peer support was established. *No outcome measures employed.*	*N/a, no data collected*
Earle, 2019 [39] *A parent-led peer support network across England and Wales* North and West London, UK	N/a	To create a peer network for families of CYP with JIA	A local JIA Matters North and West London closed Facebook group was created, and families from the area were invited to join. A local, face-to-face meet-up for families was arranged with participants from the group. *No outcome measures employed.*	*N/a, no data collected*

Table 1 (continued)

First author, Year of publication [ref.], Study title, Location	Setting Population	Study Design Study aim(s)	Details of the intervention Outcome measure(s) employed	Outcomes and main findings
DeNardo, 1995 [49] *Parents of children with rheumatic disease as peer counselors* Massachusetts, Rhode Island & Connecticut, USA	Paediatric rheumatology clinics (*N* = 7) *N* = 354 caregivers Ethnicity: 86% White, 5% Black, 7% Hispanic, & 5% Asian Mean age = 38.8 (range 19.0–50.8) Mothers (86%), married (91%), education level above high school graduate (77%)	Mixed-method study Evaluation of a parent consultant programme	Parent consultants (*N* = 10) met with families several times and provided information and support for general concerns, school issues, parent support groups and networks, communication with health professionals and parent's role. Surveys were sent within 6 months of the initial meeting with a parent consultant.	Due to an admin error, questionnaires were sent to 257 caregivers, and 84 (33%) were completed, quantitative and qualitative data was collected. Quantitative data assessed usefulness and satisfaction of the intervention and 87% felt the interaction with the parent consultant was helpful. Qualitative results showed that being able to talk to a peer has been comforting, and helpful due to overcoming issues and learning strategies to help with difficult situations.
Brown, 1998 [40] *Highly positive impact of parent consultant program in pediatric rheumatology clinic* San Diego, California, USA	Paediatric rheumatology clinic (*N* = 1) *N*= 23 caregivers of CYP with JRA (*N*= 14), Spondyloarthritis (*N*= 4), SLE (*N*= *2*), DMS (*N*= 1), MCTD (*N*= 1), & Raynaud's phenomenon (*N*= 1)	Quantitative descriptive study Evaluation of a parent consultant programme	A parent consultant met with families between 2 to > 6 times for 15–30 minutes to provide information and discuss general concerns, as well as support with specific problems and concerns relating to school, medications, physical/occupational therapy, community services and expectations. A survey was conducted at the end of 6 months to evaluate understanding of the disease, coping, loneliness and well-being.	Caregivers reported having a better understanding of the condition, coping better with frustrations, feeling less alone, sad and depressed as well as less anxious as a result of the parent consultant programme with caregivers of newly diagnosed CYP benefitting from the programme the most.

Table 1 (continued)

First author, Year of publication [ref.], Study title, Location	Setting Population	Study Design Study aim(s)	Details of the intervention Outcome measure(s) employed	Outcomes and main findings
Hagglund, 1996 [44] *A family retreat as a comprehensive intervention for children with arthritis and their families.* Missouri, USA	3-day retreat Families attending retreat (*N* = 39) 17 families completed baseline and follow ups for both caregiver and CYP Attrition rate: Caregivers *N* = 35 completed baseline questionnaires and *N* = 21 completed follow up; CYP *N* = 27 completed baseline and follow up	Quantitative descriptive study To evaluate the efficacy of a 3-day retreat	A retreat to provide education, social and emotional support, and recreational activities for CYP, caregivers and siblings. Fourteen hours of educational and therapeutic sessions were provided, and 14-hours of scheduled family-oriented recreational activities. Measures were completed at baseline and 6-months post retreat: Child Behaviour Checklist (CBCL), Paediatric Pain Questionnaire (PPQ), Symptom Checklist- 90-Revised (SCL- 90-R) to assess psychological distress of caregivers, and 2 questions rated on a 5-point-likert scale – "How much strain does your child's illness place on you while you are working?" & "How much does your child's illness interfere with family leisure/recreation time?"	Reduction on strain on caregivers' work and leisure activities and improvement in CYP's internalising behaviour problems (emotional distress) were observed.
Turner, 2001 [50] *Residential workshop for parents of adolescents with juvenile idiopathic arthritis: a preliminary evaluation.* UK	Residential workshop Caregivers of CYP with JIA Workshop group (*N* = 23, 14 mothers (61%)) Control group (*N* = 28, 27 mothers (96%)) **please review the article for a detailed list of demographics*	Non-randomised intervention To evaluate a three-day residential workshop in relation to parental perception of disease related stress and psycho-social wellbeing	A workshop over 3 consecutive days was offered to caregivers to provide information, education and social and emotional support. Outcome measures: Parents' concerns about child's health, and child's pain were measured; Parents of Children With Disabilities Inventory (PCDI) to assess perceived disability-related stress, Hospital Anxiety and Depression Scale (HADS) to assess parents' psychosocial well-being Questionnaires were completed before and three months post intervention (for both groups) and interviews were conducted 1 month after the intervention (for workshop group only).	Quantitative data: A reduction in the frequency of stressful events was observed in the workshop group as well as an improvement in their mental health Qualitative data: Three main themes were identified: Communication/understanding: improved communication with their child, better understanding of child's problems and more tolerance Positive feelings: Increase in positive mood, and a more positive outlook for the future Action/advocacy resources: new resources and information to fight for child's rights and access support

Table 1 (continued)

First author, Year of publication [ref.], Study title, Location	Setting Population	Study Design Study aim(s)	Details of the intervention Outcome measure(s) employed	Outcomes and main findings
Ireys, 1996 [45] *A social support intervention for parents of children with juvenile rheumatoid arthritis: Results of a randomized trial* Baltimore, Maryland, USA	Paediatric rheumatology clinic (*N* = 1) Mothers (*N* = 48) Mean age = 36.8 (range 24 - 55) Married (87.2%), education level at least high school graduate (91.7%), work outside of home at least part-time (64.6%)	A Randomised Controlled Study A 15-month social support intervention for mothers of children with JRA using a mentoring scheme	The intervention was focusing on three types of support: informational support, affirmational support and emotional support. The programme used different approaches and the mentors interacted with the families via telephone conversations every 2 weeks, and face-to-face meetings every 6 weeks which also included informal sessions such as picnics and lunches. Outcome measures: Psychiatric Symptom Index (PSI) to measure mental health Social support was assessed by measuring perceived availability of support as well as using one item from the Impact on Family Scale	A reduction of maternal mental health symptoms was observed in the intervention group.
Vale, 2019 [46] *Integrating peer support for young people and parents into routine practice.* London, UK	Paediatric rheumatology clinic (*N* = 1) CYP with JIA, SLE or lupus like inflammatory conditions were invited alongside their caregivers CYP *N* = 35 (25 female and 10 male) *No information relating to the parent sample available*	Quantitative descriptive study Assessing integrated peer support as part of a rheumatology clinic	Three clinics were held in the evening, facilitated by a youth worker and supported by a young adult expert at two of the clinics. A separate session for parents was facilitated by an expert parent. Three short talks were provided on topics voted on by the group prior to attending. Parents participated in a facilitated question and answer session and informal discussion. CYPs had a one-to-one 15-minute structured conversation with a health professional, guided by the Ready Steady Go questionnaire, and encompassing goal setting. Outcome measure: Feedback form	Parents provided feedback at two of the clinics. Meeting other parents was most valued with a feedback average of 9.1/10. Parents valued sharing experiences with others. CYPs valued the structured one-to-one conversation the highest with a feedback average score of 8.8/10. Meeting others was scored with an average of 8.3/10 and received the most number of positive comments. Team building activities received an average feedback score of 8.3/10, and information sessions were rated with an average of 8.0/10.

Table 1 (continued)

First author, Year of publication [ref.], Study title, Location	Setting Population	Study Design Study aim(s)	Details of the intervention Outcome measure(s) employed	Outcomes and main findings
Svavarsdottir, 2020 [47] *The impact of family strengths oriented therapeutic conversations on parents of children with a new chronic illness diagnosis.* Reykjavik, Iceland	Children's Hospital Mothers (*N* = 31) of CYP with chronic conditions (including but not limited to JIA)	Non-randomised study To evaluate the benefit of two sessions of a Family Strengths Oriented Therapeutic Conversation (FAM-SOTC) intervention	Two sessions of a Family Strengths Oriented Therapeutic Conversation (FAM-SOTC) intervention were offered to families. The sessions were 4–10 weeks apart. Questionnaires were collected pre- and post-intervention. Outcome measures: The Iceland Family Perceived Support Questionnaire (ICE-PFSQ), Icelandic-Family Illness Beliefs Questionnaire (ICE-FIBQ), the PedsQL Family Impact Module and the PedsQL Health care Satisfaction Generic Module	Families reported higher levels of family support, an increase in quality of life, greater conviction about illness beliefs and greater satisfaction with health care services.
Melville, 2014 [41] *Development of a comprehensive support website for parents of children with juvenile idiopathic arthritis.* London, UK	*N/a*	*N/a, not a research study* To provide additional support to caregivers in managing their child's JIA and provide support strategies	*N/a, this is the first stage of a wider study, a website was created, the next steps will be to evaluate it*	*N/a, no data collected; a website was developed, and the next step will be a RCT to evaluate its efficacy*
Whitelaw, 2017 [42] *Evaluation of a Website for Parents of Children with Juvenile Idiopathic Arthritis (JIA) –WebParC.*	*N/a; website*	A RCT that is currently recruiting families to the intervention To introduce WebParC, a RCT currently recruiting families for the intervention	Comparing the effectiveness of using the website alongside standard of care (intervention arm) to standard of care alone (control arm). Measures include parental stress, parental self-efficacy in managing their child's illness, parental mood, and child health-related quality of life.	*None to date.*
Mulligan, 2020 [43] *Website for parents of children with juvenile idiopathic arthritis reduces parenting stress.* England, UK	Tertiary care; paediatric rheumatology clinics (*N* = 16) Caregivers of children with JIA (*N* = 220) Mothers (*N* = 183, 83%) Intervention (*N* = 106, 48%)	A Randomised Controlled Study The aim is to evaluate a web-based tool (WebParC) for caregivers of children with a recent diagnosis of JIA	The website content was developed as part of a wider multi-disciplinary team which included various health professionals. It includes information about the disease and treatments as well as a "toolkit" for caregivers. Outcome measure(s): Primary outcome measure: Pediatric Inventory for Parents (PIP) to measure parenting stress; outcome measures were completed prior to randomisation, and at 4- and 12-month post randomisation	The website helped reduce parenting stress in the intervention group.

Table 1 (continued)

First author, Year of publication [ref.], Study title, Location	Setting Population	Study Design Study aim(s)	Details of the intervention Outcome measure(s) employed	Outcomes and main findings
Mulligan, 2022 [48] *The Effects of a web-based tool for parents of children with juvenile idiopathic arthritis: randomized controlled trial.* England, UK	Tertiary care; paediatric rheumatology clinics (*N* = 16) Caregivers of children with JIA (*N* = 220) Mothers (*N* = 183, 83%) Intervention (*N* = 106, 48%)	A Randomised Controlled Study The aim is to evaluate a web-based tool (WebParC) for caregivers of children with a recent diagnosis of JIA	The website content was developed as part of a wider multi-disciplinary team which included various health professionals. It includes information about the disease and treatments as well as a "toolkit" for caregivers. Outcome measure(s): Primary outcome measure: Pediatric Inventory for Parents (PIP) to measure parenting stress; outcome measures were completed prior to randomisation, and at 4- and 12-month post randomisation **please review the article for a detailed list of secondary outcome measures for caregivers and CYP*	The website helped reduce parenting stress in the intervention group in relation to communication, managing medical aspects of your child's care and carrying out everyday family and social activities. **please review the article for a detailed list of secondary outcomes for caregivers and CYP*

Table 2 MMAT assessment [36–50]

First author, Year [ref.]	Screening questions		1. Qualitative Studies					2. Randomized Controlled Trials					3. Non-Randomized Studies					4. Quantitative Descriptive Studies					5. Mixed Methods Studies				
	S1	S2	1.1	1.2	1.3	1.4	1.5	2.1	2.2	2.3	2.4	2.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5
Markus 2014 [37]	No	No																									
Marchal 2016 [38]	No	No																									
Eveleigh 2018 [39]	No	No																									
Earle 2019 [40]	No	No																									
Melville 2014 [48]	No	No																									
Brown 1998 [42]	Yes	Yes																Yes	No	Can't tell	Can't tell	Yes					
Hagglund 1996 [43]	Yes	No																Yes	Yes	No	No	Yes					
Vale 2019 [46]	Yes	Yes																Yes	Yes	Can't tell	Yes	No					
Svavarsdottir 2020 [47]	Yes	Yes											Yes	Yes	Yes	Yes	Yes										
Ireys 1996 [45]	Yes	Yes						Yes	No	Yes	Can't tell	Yes															
Whitelaw 2017 [49]	Yes	Can't tell						Yes	Can't tell	No	Can't tell	Can't tell															
Mulligan 2020 [50]	Yes	Yes						Yes	Yes	Yes	Can't tell	Yes															
Mulligan 2022 [51]	Yes	Yes						Yes	Yes	Yes	Can't tell	Yes															
DeNardo 1995 [41]	Yes	Yes	Yes	No	No	No	Can't tell											Yes	No	No	No	Yes	Yes	No	No	No	No
Turner 2001 [44]	Yes	Yes	Yes	Yes	Yes	Yes	Yes						Yes	Yes	Yes	Can't tell	Yes						Yes	Yes	Yes	Can't tell	Yes

Screening Questions
S1. Are there clear research questions?
S2. Do the collected data allow to address the research questions?

1. Qualitative studies
1.1. Is the qualitative approach appropriate to answer the research question?
1.2. Are the qualitative data collection methods adequate to address the research question?
1.3. Are the findings adequately derived from the data?
1.4. Is the interpretation of results sufficiently substantiated by data?
1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?

2. Randomised Controlled Trials
2.1. Is randomization appropriately performed?
2.2. Are the groups comparable at baseline?
2.3. Are there complete outcome data?
2.4. Are outcome assessors blinded to the intervention provided?
2.5. Did the participants adhere to the assigned intervention?

3. Non-Randomized Studies
3.1. Are the participants representative of the target population?
3.2. Are measurements appropriate regarding both the outcome and intervention?
3. (*continued*)
3.3. Are there complete outcome measures?
3.4. Are the confounders accounted for in the design and analysis?
3.5. During the study period, is the intervention administered (or exposure occurred) as intended?

4. Quantitative Descriptive Studies
4.1. Is the sampling strategy relevant to address the research question?
4.2. Is the sample representative of the target population?
4.3. Are the measurements appropriate?
4.4. Is the risk of bias low?
4.5. Is the statistical analysis appropriate to answer the research question?

5. Mixed Methods Studies
5.1. Is there an adequate rationale for using a mixed methods design to address the research question?
5.2. Are the different components of the study effectively integrated to answer the research question?
5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?
5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?
5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?

shown to have a positive impact, these interventions are not available outside of the research context. For example, the WebParC website is no longer available to families. This is mostly due to the challenging steps required to have an intervention adopted into clinical care which often require further time, and funding being awarded. Having viable interventions that are not accessible to caregivers after the initial testing phase means that the outcomes are only short-lived and cannot be rolled out on a larger scale.

Caregiver roles and relationships change as a result of receiving a paediatric rheumatology diagnosis and having to care for a child with a chronic condition. Adjusting to the new normal and learning to adapt to new responsibilities are essential. Caregivers need to learn to facilitate CYP's treatment management and exercise regimens which can take an emotional toll on caregivers and may lead to changes in relationships between caregivers and their child [53]. Moreover, relationships between caregivers and other family members are also affected which frequently stems from family members' lack of understanding of the disease [23]. The literature also highlights the potential impact on workplace relationships, which can be significant as well [53]. Therefore, interventions aimed at caregivers are necessary to improve outcomes for caregivers so that they are able to better support their CYP.

Caring for a CYP with a rheumatological condition can be a very lonely and isolating experience. Peer support for caregivers such as informal support groups and family weekends were found to be a good way of providing reassurance and lessening the impact of a perceived sense of isolation [54], and loneliness [40]. While interventions aimed at providing caregiver peer support are available and have been included in this review, the impact of peer support needs to be analysed and evaluated to understand the positive effects of those interventions.

Interventions should also be tailored depending on caregiver demands, level of understanding of CYP and caregivers, and stage of child's disease journey. For example, CYP and caregivers prefer interventions that are adapted to individual needs and different support should be provided depending on factors such as disease severity and developmental age [54]. Caregivers also want positive and easy to understand information without complex jargon and terminology when dealing with their child's 'unpredictable' disease [55]. Support should be offered to the wider family and family-based education tailored to individual needs [56]. Newly diagnosed CYP and their families are often in shock, denial and disbelief when receiving a diagnosis and require different support at the beginning of their journey. By comparison, when CYP have been diagnosed for longer, these more experienced caregivers face challenges in relation to continued treatment management and unpredictability of flares, and report that they would benefit from monitoring checklists to be able to record symptoms over time and favour support relating to coping with flares by having access to relevant resources and guidance [55]. Additionally, caregivers often struggle in supporting their child in relation to pain management and procedural responsibilities [52], so this should be addressed in future interventions as well.

Addressing the need for more information and reliability of sources is also important. Research suggests that caregivers prefer to obtain information from physicians (98.8%), and websites (47.9%), as opposed to seminars (3.5%), and books (1.7%) [57]. However, even when caregivers felt that they have been well informed by their medical teams relating to their child's disease and treatment, the need for more information prevailed [58]. Parents reported wanting more information on topics that they already have been given information on as well as other topics such as complementary and alternative therapies, psychological support as well as educational and vocational rehabilitation for their child [58].

Conclusions

The present review highlights the need for tailored interventions for caregivers of CYP with rheumatological conditions. The review has shown that caregivers value meeting peers and hearing other people's stories as they provide empathic and relatable sources of support, and reduce feelings of loneliness and isolation. However, support offered to caregivers needs to be evaluated to better understand the mechanisms and benefits of currently available interventions, as well as to guide future interventions. There is limited evidence from this review that caregivers receive adequate support. Considering the full impact of a chronic paediatric rheumatological diagnosis upon the whole family, this review justifies the need for better interventions to improve the quality of life of families.

Abbreviations

BRC	Biomedical Research Centre
CNS	Clinical Nurse Specialist
CYP	Children and young people
DMS	Dermatomyositis
FAM-SOTC	Family Strengths Oriented Therapeutic Conversation
GOSH	Great Ormond Street Hospital for Children NHS Foundation Trust
HADS	Hospital Anxiety Depression Scale
ICE-FIBQ	Iceland – Family Illness Beliefs Questionnaire
ICE-FPSQ	Iceland – Family Perceived Support Questionnaire
JIA	Juvenile Idiopathic Arthritis
JRA	Juvenile Rheumatoid Arthritis
MCTD	Mixed Connective Tissue Disease
MEPS	Medical Expenditure Panel Survey
MMAT	Mixed-Method Appraisal Tool

NIHR	National Institute for Health and Care Research
PCDI	Parents of Children with Disabilities Inventory
PedsQL	Pediatric Quality of Life Inventory
PIP	Pediatric Inventory for Parents
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-analyses
PROSPERO	International prospective register of systematic reviews
PSI	Psychiatric Symptom Index
RCT	Randomised-controlled trial
RMD	Rheumatic and musculoskeletal diseases
SCL- 90-R	Symptom Checklist- 90-Revised
SLE	Systemic Lupus Erythematosus

Supplementary Information

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Additional file 1.

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PL, KK, AN and JL were involved conceptualization, design and screening of the literature. HC and KK conducted the search strategy of the literature. KK and AN performed the quality assessment using the MMAT. KK prepared the manuscript. All authors have edited and approved this manuscript.

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No datasets were generated or analysed during the current study.

Declarations

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Not applicable.

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Not applicable.

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