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REVIEW PAPER

Caregiver stress in children with craniosynostosis: a systematic literature review

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Abstract

Objectives We present an overview of the literature on caregiver stress in children with craniosynostosis and report common trends in the literature.

Introduction Craniosynostosis occurs approximately 1 in 2500 births. As this is a diagnosis most common in infants and often requires surgical treatment, this is a significant and stressful ordeal for caregivers. Caregiver stress impacts various outcomes for the child, and little is understood and known about caregiver stress in the pediatric craniosynostosis population.

Methods A literature search for all articles pertaining to craniosynostosis and parental/caregiver stress was conducted using PubMed, Embase, PsychINFO, and CINAHL databases.

Results Seven articles on caregiver stress in craniofacial abnormalities patients and three articles on caregiver stress in pediatric craniosynostosis patients specifically were identified. Three articles on parental satisfaction after craniosynostosis repair were also identified and included in the review. Few published studies exist in the literature on caregiver stress in children with craniosynostosis and no clear trends were identified. It is evident that caregiver stress significantly affects the psychosocial outcomes of children with craniosynostosis. However, there are an equal number of studies reporting significant differences in caregiver stress in children with craniosynostosis as those reporting no significant differences.

Conclusions There is evidence that caregiver stress affects psychosocial outcomes of children with craniosynostosis, but no clear trends of either increased or decreased levels of stress were identified in caregivers of children with craniosynostosis. Additional research is needed to identify risk factors related to caregiver stress.

Keywords Craniosynostosis · Stress · Caregiver stress · Parental stress · Craniofacial abnormalities · Craniofacial repair

Introduction

Craniosynostosis is the premature fusion of one or more sutures of the skull. It occurs in approximately 1 in 2500 live births, and most commonly involves the fusion of a single cranial suture. Treating craniosynostosis often involves surgery that is routinely performed within the first year of life, typically around 3–6 months of age, which maximizes recovery potential and minimizes secondary facial deformation [1]. Since craniosynostosis is diagnosed at a young age and requires surgical intervention, the effect on the caregivers can be significant. Caregiver stress, or any stress associated with the role of caregiver, is important in understanding later developmental outcomes. Studies have shown that in children with chronic medical illnesses, caregiver stress is associated with adverse outcomes across multiple domains, including developmental and psychological [2–4]. Furthermore, surgery of a child places a significant amount of additional stress on children and their caregivers [5].

Particularly in the pediatric setting, caregiver stress has a huge impact on behavioral outcomes, parental-child relationships, feeding patterns, developmental outcomes, and physical health [6]. The effect of caregiver stress has been extensively

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studied in various pathologies, but little information exists on caregiver stress in children with craniosynostosis. This study presents a literature review on this topic in hopes to help increase the understanding of caregiver stress in the pediatric craniosynostosis population, highlights areas in need of further research, and may inform the development of evidence-based interventions for reducing caregiver stress.

Methods

Search strategy

A literature review was conducted using the PubMed, Embase, PsychINFO, and CINAHL databases. Article titles were searched within each database from the earliest available time point to December 2016. Pre-determined terms for the initial search included "craniosynostosis," AND "parental stress," OR "stress," and "parenting." Second searches included "craniofacial surgery," "craniofacial," "cranio*," AND "parental stress," OR "parent," "stress." The third and final search included "craniosynostosis" AND "parental satisfaction."

Studies published in English and involving craniosynostosis patients or craniofacial surgery patients and their caregivers were included for review. Studies were excluded if caregivers were not participants or craniosynostosis was not included in a target population of the study. For example, several articles either reported on family stress in children with cleft palates or focused on psychosocial adjustment following surgery instead of caregiver stress. After removing duplicate searches resulting from use of multiple databases, studies were screened for relevance based on their title and abstract. Further details for the study selection criteria are illustrated in Fig. 1.

Results

The initial literature search yielded 88 records for potential inclusion. After removing duplicates and those not meeting inclusion and exclusion criteria, 16 articles were identified for full-text review. Studies included retrospective, cross-sectional, and longitudinal studies with a follow-up period ranging from 8 months to 12 years. Based on our search criteria, articles fell into one of three categories: craniofacial caregiver stress [7], craniosynostosis caregiver stress [6], or craniosynostosis repair and parent satisfaction [3]. Sample sizes ranged considerably from 5 to 177 parents across all studies. Eleven studies examined parent dyads (both mothers and fathers), while five studies examined mothers only.

Studies in the craniofacial caregiver stress category included heterogenous samples, parents of children with a variety of craniofacial abnormalities, including but not limited to craniosynostosis. Within the craniosynostosis caregiver stress group, the majority of studies solely analyzed caregivers of patients with single suture craniosynostosis (SSC). Only one study included all types of craniosynostosis.

Measures used to assess parental stress were more consistent, with over half of the studies utilizing the Parenting Stress Index as the primary measure [7–14]. The remaining studies utilized a variety of measures, including the Questionnaire on Resources and Stress (QRS) [15, 16] and the Life Experience Survey [17]. Additionally, the Child Behavioral Checklist (CBCL) was frequently used to assess the relationship between parenting stress and child behavior [7, 8, 14, 18, 19]. A summary of the relevant measures used can be found in Tables 1, 2, and 3.

Craniofacial abnormalities and parental stress

Seven manuscripts were identified for full text review. All seven studies were published between 1989 and 2011 with patients ranging from 15 weeks to 15 years of age. Barden et al. published one of the earliest studies examining parental stress within the craniofacial anomalies (CFA) population. This study included ten participants, five mothers of children with CFAs and five matched controls. It focused generally on effect of facial deformities on maternal attitudes and behavior, and specifically on the discrepancy between observational and self-report measures of maternal functioning. Life stress and satisfaction were measured using the Life Experience Survey (LES) and the Satisfaction with Parenting Scale (SWPS) respectively. The authors primarily found that infant attractiveness is an important predictor of the quality of mother-infant interactions, which implied that maternal responsiveness is greater when infants are more attractive. Furthermore, results showed that mothers of children with CFAs self-reported higher levels of parental satisfaction and more positive life experience than mothers of normal children. This difference was attributed to the normal disease coping process [17].

Additional studies, listed in Table 1, found that the emotional state of the caregivers greatly influences the emotional development of children with CFAs [7, 15, 16]. Three studies found caregiver stress was correlated with negative parent-child interactions [7], the presence of older children in the home, current caregiver health status, dissatisfaction with clinic staff [15], and increased anxiety and depression in the child [16]. All of the studies concluded that high caregiver stress was associated with poorer psychosocial outcomes for children that have received surgery for CFAs. Krueckeberg and Kapp-Simon examined the effect of parental factors, such as stress, on the social skills of preschool children with CFAs [10, 12]. Although the authors hypothesized that CFA parents would report higher levels of stress and have smaller social support networks than control parents, this prediction was not confirmed. The study found that 36% of variance in the children's social skills was due to the lower percentile scores on

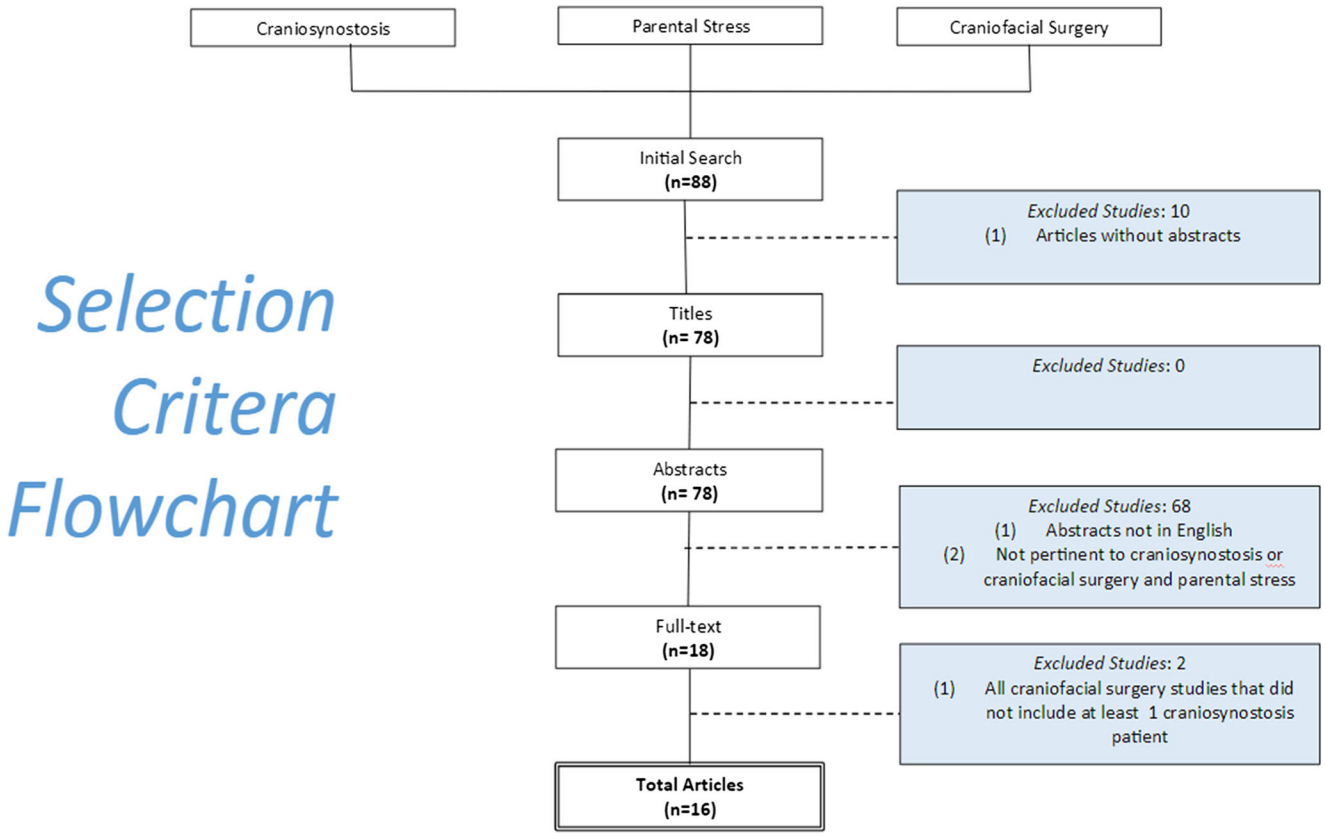

Fig. 1 Selection criteria for articles assessed in the study illustrates the selection criteria used to identify the 16 articles included in the final review

the Parenting Stress Index (PSI) assessments and suggested that high levels of caregiver stress negatively impact outcomes for children with CFAs [12].

In a more recent study conducted by Pope et al., children with craniofacial anomalies were assessed to identify a correlation between parenting stress during infancy and later child psychosocial adjustment during toddlerhood. This study utilized a retrospective chart review to evaluate parent stress during infancy and toddlerhood. Caregivers had a Parent Stress Index/Short Form (PSI/SF) completed when children were 24 months old or younger and a PSI/SF and Child Behavior Checklist completed when children were toddlerhood and between 24 and 46 months. The results showed no difference in total stress means from the PSI/SF between times one and two; however, a larger-than-expected percentage of the sample scored above the 90th percentile at both time points showing elevated levels of parental stress. This suggested that increased stress levels may be present in parents of children with CFAs. Furthermore, persistently elevated levels parenting stress was correlated with higher levels of maladjustment in the children. The authors concluded that parental stress remains stable over time and that persistently increased levels of parenting stress during infancy and beyond can negatively impact the children's development [18].

As more studies hinted at a correlation between caregiver stress and psychosocial development in children with craniosynostosis, Roberts and Shute recognized the need for a consistent measure of stress specific to patients with CFAs and their caregivers. The authors created the Craniofacial Experiences Questionnaire (CFEQ) to measure both stressors and positive aspects of living with a CFA. During the course of developing the CFEQ, they found that caregivers of children with syndromal craniosynostosis, or complex craniofacial conditions, had significantly higher CFEQ scores for the positive aspects scale than for caregivers of children with other craniofacial pathologies, such as neoplasms. This finding demonstrates possible positive aspects related to the presence of a CFA that may not have been considered in previous studies [19].

The article published by Barden et al. paved the way for additional research to be conducted on the impact of parental stress in CFA patients. After the review of all the seven articles, two concurrent themes were noticed; maternal stress negatively impacts psychosocial outcomes of children with CFAs, and no significant differences in stress levels existed in parents of children CFAs and controls. Although the studies did not indicate a higher level of caregiver stress in CFAs patients, it was clear that parenting stress leads to poorer psychosocial outcomes in children, regardless of medical condition [7, 12, 15–19].

Table 1 Articles on craniofacial abnormalities and caregiver stress

Study	Study type	Study size	Population	Relevant measures of stress	Pertinent findings
Barden et al. 1989	Case-control	Single-center	-5 mothers of infants with CFAs-5 mothers with normal infants	-Life Experience Survey (LES) -Satisfaction with Parenting Scale (SWPS)	-Mothers of infants with CFAs were consistently less nurturing toward their infants. -Mothers of infants with CFAs claimed they were more satisfied with parenting than mothers of normal infants. -No significant differences existed between groups in terms of social support and general life satisfaction.
Pillemer and Cook 1989	Case-control	Single-center	-25 craniofacial patients	-Parent-Caretaker Questionnaire-QRS	-High caregiver stress levels were associated with increased anxiety and depression in children. -Children with higher stress caregivers demonstrated lower self-concept.
Coulter et al. 1991	Case-control	Single-center	-27 parents of children with CFAs	-Questionnaire on Resources and Stress (QRS) -Maternal Social Support Index	-High caregiver stress levels were associated with dissatisfaction with clinical staff, presence of older children at home, and current health status of the caregiver(s).
Krueckeberg and Kapp-Simon 1993	Case-control	Single-center	-30 families of children with CFAs-22 families of children without CFAs	-PSI-Social Relationship Scale-Social Skills Questionnaire (SSQ)	-Caregiver stress was strongly associated with negative impacts on social outcomes for children with CFAs. -No significant differences in parenting stress scores were found in families of children with or without CFAs.
Campis et al. 1995	Case-control	Single-center	-77 mothers of children with CFAs	-CBCL-PSI	-Both parent and child domains of the PSI were linked significantly to maternal reports of anxiety and depression-according to CBCL and PSI scores, maternal stress is associated with child maladjustment.
Pope et al. 2005	Retrospective	Single-center	-47 parents of children with CFAs	-PSI-Short Form (PSI-SF) -CBCL	-No significant differences in caregiver stress levels was found when measured during child's infancy and later childhood.
Roberts et al. 2011	Case-control	Single-center	-50 adolescents with CFAs-55 parents of children with CFAs	-CBCL-Craniofacial Experiences Questionnaire (CFEQ)	-The CFEQ was newly developed to measure stressors and positive experiences of young people with CFAs. -No significant differences were found in parental stressors of children with or without CFAs.

The table shows studies on craniofacial abnormalities and caregiver stress included in the review, number of participants, and relevant stress measures utilized in the studies

CBCL Child Behavior Checklist, *CFA* craniofacial anomaly, *CFEQ* Craniofacial Experiences Questionnaire, *LES* Life Experience Survey, *PSI* Parenting Stress Index, *SI-SF* Parenting Stress Index-Short Form, *SSC* single-suture craniosynostosis, *SSQ* Social Skills Questionnaire

Table 2 Articles on craniosynostosis and parental stress

Study	Study type	Study size	Population	Relevant measures of stress	Pertinent findings
Sarimski, 1998	Case-control	Single-center	-41 parent dyads of children with Apert syndrome	-PSI-Child Behavior Checklist (CBCL)	-Dissatisfaction with child's appearance, low-parental self-esteem, and child behavioral problems were associated with higher caregiver stress levels.
Kapp-Simon et al. 2005	Prospective	Multicenter	-100 mothers of infants with SSC	-PSI	-No significant correlations were found between child developmental scales and caregiver stress.
Rogers, 2007	Case-control	Single-center	-115 mothers of SSC patients -100 fathers of SSC patients -94 mothers and 82 fathers matched controls respectively	-Parenting Stress Index (PSI)	-Mothers of children with SSC had increased levels of stress than control group mothers.
Rosenberg et al., 2011	Case-control	Single-center	-246 parent dyads of infants with SSC -253 matched controls	-PSI	-No significant differences in caregiver stress was found between child with or without SSC. -Mothers of children with SSC reported higher levels of stress than father counterparts.
Cloonan et al., 2013	Case-control and cross-sectional	Multicenter	-24 mothers of children with craniosynostosis- 124 controls	-PSI-CBCL	-No significant differences in caregiver stress was found between children with or without SSC.
Gray et al., 2015	Prospective	Multicenter	-247 mothers of children with SSC -211 fathers of children with SSC -254 control mothers -220 control fathers	-PSI	-No significant differences in caregiver stress was found between children with or without SSC. -Mothers of children with SSC reported higher levels of stress than father counterparts.

The table shows studies on craniosynostosis and parental stress included in the review, number of participants, and relevant stress measures utilized in the studies

CBCL Child Behavior Checklist, *PSI* Parenting Stress Index, *PSI-SF* Parenting Stress Index-Short Form, *SSC* single-suture craniosynostosis

Table 3 Articles on craniosynostosis and patient satisfaction

Study	Study type	Study size	Population	Relevant measures of stress	Pertinent findings
Kim et al. 2008	Case-control	Multicenter	-22 parents of patients using minimally invasive technique -25 parents of patients using open technique	-PSI-SF	-Parent satisfaction scores were higher for children having undergone minimally-invasive repair (vs. open repair).
Kluba et al. 2016	Retrospective	Single-center	-46 parents of children with craniosynostosis	-Quality of communication -Questionnaire on esthetic outcome	-Majority of parents were satisfied with undergoing surgery for their child and would opt for surgery in retrospect.
Wong-Gibbons et al. 2009	Case-control	Population-based	-82 mothers of children with craniosynostosis	-Structured Clinical Interview	-Majority of mothers reported to be satisfied with surgery results.

The table shows studies on craniosynostosis and patient satisfaction included in the review, number of participants, and relevant stress measures utilized in the studies

PSI-SF Parenting Stress Index-Short Form

Craniosynostosis and parental stress

Six studies primarily involving craniosynostosis and caregiver stress were identified and shown in Table 2. Four studies examined both mothers and fathers of children with craniosynostosis [9, 13, 14, 20] while the remaining two examined only mothers [8, 10]. Four studies included only single-suture craniosynostosis (SSC) cases [9, 10, 13, 20]. Of the remaining two studies, Sarimski included only children with Apert syndrome while Cloonan et al. included children with all types of craniosynostosis [8, 14].

Among the six studies, there were varied findings with regard to caregiver stress. Three studies found significant differences in caregiver stress between craniosynostosis patients and matched controls [13, 14, 20], while the latter three studies reported minimal to no differences between groups [8–10].

Rogers et al. found that parents of children with SSC were more likely to report higher levels of child-related stress. More specifically, mothers had increased levels of stress when compared to the control group mothers. Despite showing similar stress levels as control counterparts, fathers of children with SSC were more likely to report higher levels of child-related stress than mothers of children with SSC [20]. This higher reporting among fathers was seen again in the Rosenberg et al. study [13].

Rosenberg et al. and Sarimski provided more insight on contributing factors to increased caregiver stress levels. Both studies found that mothers reported greater stress if their child's condition was more noticeable in appearance, suggesting increased stress caused by worry over the visibility of their child's condition. No significant differences were noted in the fathers. In addition, Sarimski alluded to low parental self-esteem and child behavior problems as contributing factors to higher caregiver stress levels [13, 14].

With respect to studies that found no significant differences in stress levels of caregivers of children with craniosynostosis, there were other significant differences within each studies' patient cohorts. Cloonan et al. found no differences in caregiver stress between caregivers of children with SSC and controls, but parents of children with SSC were found to have lower health-related quality of life scores than control counterparts [8]. Furthermore, Gray et al. found that mothers report higher stress than fathers on parent-related stress, but ultimately concluded that parents of children with SSC have similar levels of stress to the control cohort [9]. Lastly, Kapp-Simon et al. found no significant correlations between child developmental scales and parenting stress levels based on PSI assessments [10].

Craniosynostosis repair and patient satisfaction

In addition to the stress and coping involved in taking care of a child with craniosynostosis, the surgical repair is also a very

Table 4 CFA severity, comorbidities, and cognitive functions' effects on caregiver stress

Study	Factors assessed	Findings
Krueckeberg and Kapp-Simon 1993	Severity of CFA and parental behavior	• Parents of children with visible defects found social networks more helpful. Children with visible defects had parents with more nurturing and less restrictive parenting behaviors.
Campis et al. 1995	Effect of comorbidities on caregiver stress in CFA patients	• Children with most severe comorbid medical conditions had significantly higher CBCL scores. • No significant differences in CBCL were found in caregivers with mild or moderate comorbid medical conditions.
Sarimski 1998	Cognitive functioning and caregiver stress	• Fathers of children with Apert syndrome and moderate/severe learning disabilities demonstrated higher stress due to various behaviors (vs. fathers of children with craniosynostosis and borderline/normal learning capacity).

The table shows findings from three studies that respectively assessed the effect of CFA severity, patient comorbidities, and cognitive function on caregiver stress

CBCL Child Behavior Checklist, *CFA* craniofacial anomaly

significant and stressful event for the both the child and his or her parent(s). Gewalli et al. study surveyed the parents of 26 children who underwent surgical repair of craniosynostosis and found that parents' decisions to pursue surgical repair were based on concerns for brain damage and/or esthetic appearance. By pursuing surgical treatment, parents ultimately hoped that their child's developmental prognosis would improve [21].

In addition to the importance of operative success, the parents' satisfaction with the procedure is also an undeniably influential factor that is related to both parental stress and the care of the child. Kluba et al. and Wong-Gibbons et al. investigated parent satisfaction with regard to their child's postoperative outcomes. A majority of the interviewed parents from both studies were satisfied with the outcomes; greater than 75% of parents were "very happy" or gave ratings of "good or very good" of the operative results. Of the fraction of parents who were dissatisfied and felt additional surgeries were needed, specific reasons for dissatisfaction included bone defects, asymmetric postoperative head shaping and scarring, and alopecia [22, 23].

Notably, Kim et al. looked at patient satisfaction comparing open and minimally invasive techniques in craniosynostosis repair. The studies recorded parental responses using the PSI/SF, and it was found parents of open-repaired cases scored lower in the parent-child dysfunctional interaction, difficult child, and total stress domains of the assessment. When controlled for confounders, it was also found that parents of female children generally scored lower than those of male children [11].

Other relevant remarks included the parents' satisfaction toward the medical services and support from their providers. The majority of parents expressed satisfied with regard to the services and support received from their providers even if they were not pleased with the esthetic outcome of the surgery [22]. Additional details on the aforementioned studies on craniosynostosis repair and patient satisfaction are shown in Table 3.

Craniosynostosis repair and surgeon satisfaction

Only one article, Kluba et al., provided details on both patient and surgeon satisfaction of the craniosynostosis repair. The majority of physician and parental gave ratings of either "good" or "very good" on the surveys. The mean parental and surgeon ratings respectively were 1.6 and 1.44 out of a scale of 0 to 5 with a score of 1 correlating to a "perfect rating" and a score of 5 correlating to a "deficient" rating. Due to the lack of a sizeable cohort, Kluba et al. were unable to conduct an analysis comparing parental and surgeon satisfaction based on type and form of craniosynostosis [22].

CFA severity, comorbidities, and cognitive functions' effects on caregiver stress

Kruckeberg and Kapp-Simon was the only article that assessed the relationship between the severity of a CFA and parental behavior. Parents of children with visible defects (vs. CFAs not easily visible or apparent) found social networks more helpful and favored parenting styles that were more nurturing and less restrictive [12].

With respect to the effect of comorbidities on caregiver stress in CFAs patients, Campis et al. found caregivers of children "most severe comorbid medical conditions" had significantly higher CBCL scores, indicating greater stress, while also reporting more adjustment problems. No significant differences in CBCL were present in caregivers with children with mild and moderate comorbid medical conditions [7].

Furthermore, with respect to cognitive functioning and caregiver stress, Sarimski found that fathers of children with Apert syndrome and moderate or severe learning disabilities felt more stress due to unacceptable behaviors, adaptability, distractibility/hyperactivity, and mood compared to fathers of children with craniosynostosis and borderline or normal learning capacity [14]. Findings of the three aforementioned studies are further detailed in Table 4.

Discussion

The diagnosis and treatment of craniosynostosis in one's child may cause long-lasting psychological effects in caregivers, but the current literature on the subject is inconclusive. Several studies report increased levels of caregiver stress following surgery that alleviates over time. Other studies find no differences in levels of caregiver stress between parents of children with craniosynostosis and controls. However, very little research has been conducted on the topic and, of the research that does exist, very few studies have measured caregiver stress prior to treatment and throughout the course of post-operative care. Furthermore, as is common in the general population, studies on mothers of children with craniosynostosis patients were more prevalent in the literature. These differences could be related to traditional distribution of caregiver responsibilities or could be due to the fact that a larger proportion of mothers have been studied within this population to date. The question remains whether there are truly meaningful gender differences between caregiver stress.

Assessment of caregiver stress was primarily conducted using the PSI, a measure created for parents of typically developing children. This measure may not be sensitive to the unique circumstances a caregiver of a child with craniosynostosis faces and could be missing domains of stress related to a medical environment. Furthermore, additional research is needed to identify risk factors related to increased caregiver stress. There is reason to believe socioeconomic status, education-level, surgical procedure, and the presence of older children in the home could be related to caregiver stress, but future research is needed to determine if this is the case.

In the majority of studies included in this review, parents of children with SSC were assessed, making comparisons between the different diagnoses of craniosynostosis difficult. Only one study was found that evaluated surgical type (minimally invasive vs. open surgery) and further evaluation of this interaction is needed. In addition, only one out all identified and included studies reported and compared parental and surgeon satisfaction after craniosynostosis repair, which also demonstrates the lack thereof and need for additional investigation.

The majority of studies dealing specifically with caregiver stress and craniosynostosis have been conducted in the past decade, suggesting an increased interest in the topic. While there remains little research in the craniosynostosis population, caregiver stress has been more closely looked in other illnesses such as traumatic brain injuries (TBI). For example, a study by Wade et al. focusing on varying severities of pediatric TBI discovered higher rates of parental and caregiver stress in children with severe TBI than moderate TBI [24]. Furthermore, Pinquart conducted a meta-analysis looking at parenting stress in caregivers of children with a chronic physical condition found significantly higher levels of caregiver stress in children with HIV infection or AIDs, spina bifida, cerebral palsy, or cancer. A correlation between low parental mental health and child behavioral problems was also identified [25]. Nonetheless, little has been reported on caregiver stress in surgically treated conditions in fields including neurosurgery, orthopedics, and ENT. There is a lack of longitudinal research in this area, which is surprising because almost all articles argue for the necessity of a longitudinal design.

Limitations and future research

The limited amount of literature on caregiver stress in the pediatric craniosynostosis population did not allow for a more in-depth and complete assessment of the topic. Due to the limited number of articles identified, no summative correlations, differences, or conclusions were made on the impact of caregiver stress of children with or without craniosynostosis. The original objective of this review was to highlight the main findings of caregiver stress in children with craniosynostosis, but it also identified the lack of research and the need for more studies on the subject matter.

The present review reveals several potential areas of improvement in future research. In the 16 included and reviewed studies, a variety of methods were utilized for measuring and defining stress. There is some consensus on measures of parent-related and child-related stress in terms of outcomes, but little to no discussion was found dealing with medically related stress. This suggests the potential need for measures more sensitive to the environment of these caregivers with special attention paid to stress caused by being in a medical environment. Furthermore, the effect of having a child with a lower psychosocial outcome may also reciprocally lead to greater caregiver stress has also not been studied in the literature in the craniosynostosis population, thus highlighting another area in need of further attention.

Additionally, there is a need for longitudinal research. Repeated assessment with longer time frames between measurements is necessary to follow parents prospectively through the different phases of treatment, from diagnosis through recovery. It is recommended that a consensus be determined on the optimal time points to assess stress in parents following the diagnosis of craniosynostosis in their child. This would facilitate the comparison of results. As it stands now, it is difficult to compare the current literature in the field due to the different measurements used and the vast array of time points at which the measures of parental stress were conducted.

Conclusions

Through our review of current literature on caregiver stress in the pediatric craniosynostosis population, it was evident that

parental stress significantly impacts the psychosocial outcomes of children. Although no consistent differences in stress levels were found between caregivers of craniosynostosis or normal children, the limited amount of literature on this topic demonstrates the need for further research and studies.

Contributorship statement Dr. Lim conceptualized and designed the review, collected data, summarized and interpreted the findings, drafted the initial manuscript, and reviewed and critically revised the manuscript.

Ms. Davis conceptualized and designed the review, collected data, summarized and interpreted the findings, drafted the initial manuscript, and reviewed and critically revised the manuscript.

Mr. Tang participated in data collection and reviewed and critically revised the manuscript.

Dr. Shannon conceptualized and designed the review, interpreted the findings, and reviewed and critically revised the manuscript for important intellectual content.

Dr. Bonfield conceptualized and designed the review, interpreted the findings, and reviewed and critically revised the manuscript for important intellectual content.

Dr. Lim, Ms. Davis, Mr. Tang, Dr. Shannon, and Dr. Bonfield all agree to be accountable for all aspects of the work in ensuring that questions related to accuracy or integrity of any part of the work are appropriately investigated and resolved.

Compliance with ethical standards

Conflict of interest The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in the manuscript.

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