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Cranial remolding orthosis for children with deformational skull deformities: A systematic review on the factors affecting success and duration of treatment

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ABSTRACT

Deformational plagiocephaly, deformational brachycephaly, and deformational scaphocephaly are the most common types of skull deformities during the first year of life. Using a cranial remolding orthosis (CRO) can have an important role in achieving a satisfactory level of improvement in symmetry and proportion of the deformed skulls. However, there is no consensus on the most important parameters for the success or length of treatment with a CRO. In this study, we did a systematic literature review in PubMed, Scopus, Web of Science, and EMBASE on January 2023. Titles/abstracts of the found studies were screened by two independent reviewers. The Newcastle–Ottawa Scale was used to evaluate the quality of the included articles. The best evidence synthesis was considered to determine the strength of the reported factors. A total of 25 articles with an accumulated sample of 7594 participants were included. Nine predictive factors, including age at initiation of CRO treatment, CRO compliance, deformity severity, deformity type, torticollis, gestational age, gestational type, delivery method, and developmental delay, were considered for CRO treatment length or success. Moderate evidence suggests that CRO treatment length is linked to a patient's age at the start of treatment and the deformity severity. Moreover, treatment success is correlated with a patient's age at the start of treatment, CRO compliance, and deformity severity. Moderate evidence indicates that there is no relationship between the presence of torticollis and gestational age with CRO treatment success.

1. Introduction

Cranial deformity refers to a series of anomalies and deviations in an infant's skull manifested as asymmetry of the head bones or lack of consistency in the form of the face. Deformational plagiocephaly, deformational brachycephaly, and deformational scaphocephaly are the most common types of skull deformities that are identified in infancy, resulting from pre- and post-natal factors. Factors such as male sex, multiple birth, large embryo, congenital anomalies, prematurity, first birth, and breech presentation are recognized in the development of cranial deformities.[1] There are huge deformative forces pressing the skull when the infant passes the delivery canal during delivery, which can deform the skull.[1] Premature infants are more prone to these deformities because of the malleability of their skull.[2–4] Congenital torticollis and imbalance of neck muscles are also presented in children with deformational plagiocephaly.[1]

Depending on the deformity type and severity, problems such as neurodevelopmental delay, cognitive problems, and learning, vision, and talking disorders, plus unattractive appearance, are more prevalent in the affected population compared to the normal population.[1,5] The prevalence of cranial deformity is 3%–48 %, which has significantly increased in the last two decades.[1,2,6] In 1992, to decrease the incidence

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of sudden infant death syndrome, the American Academy of Pediatrics (AAP) recommended that parents sleep their infant in the supine position. However, "Back to Sleep" strategy significantly increased the subsequent skull deformities from 0.3 to 48 %.[1]

The treatment methods for children with skull deformities include repositioning, physical therapy, and cranial remolding orthosis (CRO) or helmet orthotic therapy, depending on the infant's age and deformity severity.[1,7,8] CRO treatment for skull deformities is prescribed when an infant's deformed skull proportion or asymmetry maintains or does not get better despite active repositioning or physical therapy during the first three months after birth. The existing body of research on children with deformational skull deformities suggests that wearing a CRO is an effective strategy for improving skull symmetry and proportions. CROs are designed to be in contact with the bossing part of the skull while creating a gap in the flattening regions. Thus, CROs guide the skull growth toward the flattening area.

There are some controversies about the age range of using CROs to treat skull deformities. Some studies suggest that using CROs for infants aged 3–12 months leads to excellent outcomes.[9–12] As the brain and skull growth is significantly decreased after the age of 12 months, modification of the cranial deformities will be much more difficult and takes long after this age. However, some studies have reported that the age of starting to use CRO is not a major factor in treatment success.[13] In addition to age, the efficacy of CRO treatment and treatment length may be under the influence of other predictive factors such as CRO compliance, deformity severity, presence or absence of torticollis, and gestational age.[14–16]

Determining the predictive factors such as the clinical characteristics of children with skull deformity can help parents, caregivers, and the treatment team to identify the causes that might be availed from CROs. These factors indeed predict some useful information helping parents and therapists in better application of the orthoses. In this systematic review, we evaluated the most important clinical predictive factors in the success or treatment length of CRO treatment in children suffering from cranial deformities.

2. Methods

The protocol of this systematic review was registered in PROSPERO with the ID number of CRD42022302609(https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42022302609).

We reported this study in 6 stages based on PSALSAR; Protocol, Search, Appraisal and Synthesis in the method section, Analysis, and Reporting the results in separate sections.

2.1. Protocol

The PICO model was used to construct the study criteria. PICO refers to patient/population (children with non-syndromic and non-synostosis cranial deformities), intervention (CRO), comparison (if available, compared with no treatment), and outcomes (treatment length or treatment success).

2.2. Search strategy

We did a systematic literature review in PubMed, Scopus, Web of Science, and EMBASE (via Ovid) in January 2023 to find articles that assessed the factors affecting treatment length and success of CRO treatment in children with cranial deformities. The following search strategy or keywords was utilized for searching articles in PubMed: ((plagiocephaly OR scaphocephaly OR brachycephaly OR "skull deformity" OR "cranial deformity") AND ("cranial remodeling orthosis" OR helmet OR "cranial remodeling device" OR ortho*)) AND (outcome OR predict* OR effect* OR "predictive factor"). We used the same strategy to search the other databases. A search in Google Scholar was done using "Allintitle" to track down further related articles. Moreover, the reference lists of the included articles were assessed to ensure all eligible studies were reviewed. Only articles that were published in English were included. This review was performed according to the preferred reporting items for systematic reviews and meta-analysis (PRISMA) 2020 guideline (Fig. 1).

2.3. Appraisal

Two independent reviewers (H.H. and M.B.) screened the titles/abstracts of the found studies to select the potentially related articles. A consensus with a third reviewer (T.B.) was done whenever required. After omitting duplicates, the remaining titles and abstracts were screened. Afterward, we applied the inclusion criteria on the full texts of the potentially relevant papers. Studies were included if CRO treatment was completed up to the 18th month. Conference summaries, review studies, case reports, and studies that assessed patients with syndromic or synostosis cranial deformities were excluded.

We used the Newcastle–Ottawa Scale (NOS) to evaluate the quality of the included articles. This tool has eight items that are distributed in three sections of selection, comparability, and outcome. The items of the selection and outcome sections can get a maximum of one star. The whole comparability section can receive two stars. The overall score of this test can be from 0 (lowest quality) to 9 (highest quality) stars. The quality of articles based on the NOS can be low (0–3 stars), moderate (4–6 stars), and high (7–9 stars). Two authors (T.B. and H.H.) independently scored the articles, and the results were shared. In case of any discrepancy, they reached a consensus for a final decision.

Best evidence synthesis was performed based on previous studies to determine the strength of the reported factors.[17,18] The following ranking was used: 1) strong evidence was obtained in the case of consistent findings (>75 % of the studies showed the same association) and scored a high quality on the NOS; 2) moderate evidence was obtained in the case of consistent findings (>75 % of the studies showed the same association) and scored a moderate or low quality on the NOS; 3) limited evidence was obtained in the case of consistent findings in one high quality in all the subdomains of the NOS, or two moderate or low-quality studies in one or more subdomains of the NOS; 4) conflicting evidence was obtained in the case of non-consistent findings (<75 % of the studies showed the same direction of association); and 5) no evidence was obtained when no studies could be found.

2.4. Synthesis

Two reviewers (H.H. and T.B.) independently extracted the data from the full texts. Any discrepancy was discussed and resolved in a session. We create a data extraction table to record the following information found in the included studies: authors (year), study design, age at initiation of treatment, sample size, type of cranial deformity, deformity severity, CRO wearing time, treatment length, improvement rate, and main findings (Table 1).

3. Results

3.1. Search analysis

The results of the database searches are shown in Fig. 1. After deleting duplicate and unrelated articles, 65 articles remained. Among them, 25 met the eligibility criteria for this study.

The sample sizes of included studies were between 45 and 1050 patients, with a total sample of 7594 patients. The participant's age at the initiation of treatment ranged from 19 days to 21.6 months, and the duration of CRO treatment was between 6.3 and 39.7 weeks (Table 1).

The quality of the 25 included studies was moderate, ranging from 4 to 6 stars based on NOS (Table 2). Eighteen studies were retrospective in nature,[9,11,14,19,21–28,30,31,33,34,37] and seven studies had prospective designs.[10,20,29,32,35,36,39]

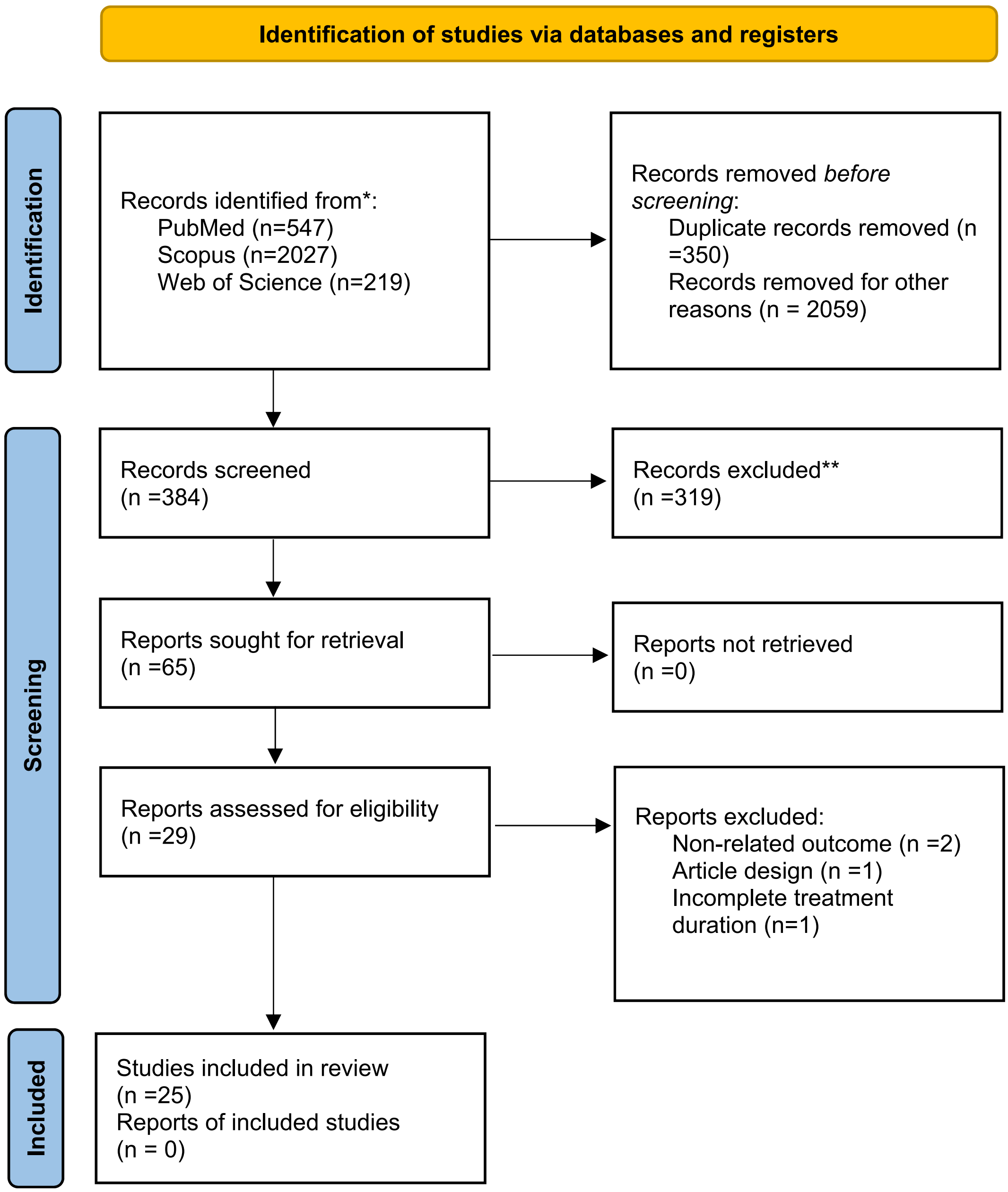

Fig. 1. PRISMA 2020 flow diagram for new systematic reviews, which included searches of databases and registers only.

Studies have identified several factors influencing the duration and success of CRO treatment. These risk factors can affect the treatment length, its success, or both (Supplementary file), which are listed under two headings. Obviously, if a study has measured both the outcome of treatment duration and success, it is mentioned separately in each section.

3.2. Risk factors for treatment success

Eight predictive factors, including age at initiation of orthotic treatment, orthotic compliance, deformity severity, torticollis, gestational age, gestational type, method of delivery (cesarean or vaginal), and developmental delay had been considered for evaluating CRO treatment success in the included studies. Twenty-one moderate quality studies examined the relationship between the age at initiation of orthotic treatment and treatment success.[9–11,14,24,21–23,25,27,31,33,20,26,30,37,29,32,35,36,38] Eighteen of them stated that this factor is negatively related to orthotic treatment success.[9–11,14,24,21,25,23,31,33,20,26,37,29,32,35,36,38] However, four of them concluded that the age at initiation of orthotic treatment is not an influential factor in the treatment success.[22,27,30,38] Twelve moderate quality studies evaluated the relationship between deformity severity and treatment success[9,11,14,21,23,27,33,34,26,29,32,39]; ten reported a negative relationship.[11,14,21,23,27,33,26,29,32,39] Still, two of them stated no relationship between these two factors.[9,34]

Three moderate quality studies evaluated the effect of CRO compliance on treatment success.[9,23,36] All of them agreed that there is a positive relationship between them. In terms of presence of torticollis, the results of three moderate-quality studies revealed that there is no relationship between this factor and CRO treatment success.[9,14,21] Three moderate-quality studies evaluated the effect of gestational age on treatment success.[14,21,29] Their results were not significant at the $p = 0.05$ level. The effect of gestational type, delivery type, and developmental delay on CRO treatment success was assessed in only one moderate-quality study.[9] Its results showed that there is no association between these parameters and treatment success with a CRO.

Table 1
Characteristics of included studies.

Reference	Study design	Age (months)	Sample size	Deformity type	Severity of deformity	Orthotic treatment length (months)	Follow-up (months)	Improvement rate	Outcome assessment
Couture et al.[19]	Retrospective	0.63 to 21.6	1050	Plagiocephaly	Argenta Type II–V	3 to 6	6.3	81.6 %	Treatment length
Graham et al.[20]	Prospective	6.6	159	Plagiocephaly	DD:1.13	4.2	Not provided	0.71 cm for DD	Treatment success
Graham et al.[14]	Retrospective	2 to 17	499	Plagiocephaly	CVAI: 3.1 %–16.1 %	Not provided	Not provided	Final CVAI: 0.1 %–10.1 %	Treatment length, treatment success
Graham et al.[21]	Retrospective	3 to 18	500	Brachycephaly	CVAI: 3.5 % to >11 % CI: 90 % to >97 %	Not provided	Not provided	44.60 %	Treatment success
Grigsby et al.[22]	Retrospective	7.08 to 10.05	58	Plagiocephaly	CVAI: 9.29 %–11.32 %	3.62 to 6.51	Not provided	72.13 %–53.03 %	Treatment length, treatment success
Hinken et al.[23]	Retrospective	7.82 to 8.88	1050	Plagiocephaly Brachycephaly, combination	CVAI:11.34 %–11.49 % CI: 98.73 %–95.60 %	5.25 to 6	Not provided	Final CVAI: 2.94 %–7.08 % Final CI: 4.17 %–8.22 %	Treatment length, treatment success
Kelly et al.[11]	Retrospective	6.5	258	Plagiocephaly	CVA: 8.8 mm, SBA: 6.2 mm, OTDA: 4.4 mm	4.1	Not provided	Final CVA: 3.3 mm Final SBA: 3.2 mm Final OTDA: 2.4 mm	Treatment success
Kluba et al.[10]	Prospective	4.1 to 10.7	62	Plagiocephaly	CVAI 13.6 % and 13.1 %	4	Not provided	60.6 %–75.3 %	Treatment length, treatment success
Çevik et al.[24]	Retrospective	6.5	89	Plagiocephaly, Brachycephaly	CVAI: 9.9 % CI: 95.8 %	1.5 to 9.9	Not provided	23 %–43 %	Treatment length, treatment success
Han et al.[25]	Retrospective	6.14	310	Plagiocephaly	CVAI: 9.7 %	5.51	Not provided	CVAI: 43.4 %–67.9 %	Treatment length, treatment success
Freudlsperger et al.[26]	Retrospective	7.1	213	Plagiocephaly	CVAI: 9.8 %	4.675	Not provided	33 %	Treatment success
Kim et al.[27]	Retrospective	3 to >6	200	Plagiocephaly	CVAI: 12.73 %	3.35	Not provided	Final CVAI: 4.91 ± 0.8 %–5.5 %	Treatment length, treatment success
Choi et al.[28]	Retrospective	3 to 14	207	Brachycephaly	CVAI: 8.11 % CI: 95.55 %	5.02	Not provided	Final CI: 88.87 ± 1.07 %	Treatment length
Kunz et al.[29]	Prospective	7	239	Plagiocephaly	CVAI: 8.69 %–9.58 %	5.1	Not provided	Final CVAI 4.63 % to 5.03 %	Treatment length, treatment success
Teichgraeber et al.[30]	Retrospective	5.8	125	Plagiocephaly	CVA: 8.53 mm OTDA: 3.12 mm CBA: 7.08 mm	4.5	6 to 12	CVA: 40.23 % OTDA:18.72 %).	Treatment success
Lam et al.[31]	Retrospective	5.8 months	543	Plagiocephaly, Brachycephaly, Combination	ODD: 125 mm CI: 1.007	Not provided	Not provided	ODD: 37 %	Treatment success
Kunz et al.[32]	Prospective	6.8	144	plagiocephaly	CVAI: 8.57–8.84 %	5.15	Not provided	7 %–83 %	Treatment length, treatment success
Mackel et al.[33]	Retrospective	6.4	45	Plagiocephaly	CVAI: 8.5 %	4	Not provided	Final CVAI: 3 %	Treatment length, treatment success
Peethambaran et al.[34]	Retrospective	5.89	70	Plagiocephaly, Brachycephaly, Combination	Not provided	4.99	Not provided	CVAI: 28.8 %–41.4 % CR: 2.60 %–4.66 %	Treatment length, treatment success
Seruya et al.[35]	Prospective	4.2 to 11.2	346	Plagiocephaly	CVAI: 10–12 mm	1.9 to 3.25	Not provided	0.41 mm–0.93 mm	Treatment length, treatment success

(*continued on next page*)

Table 1 (*continued*)

Reference	Study design	Age (months)	Sample size	Deformity type	Severity of deformity	Orthotic treatment length (months)	Follow-up (months)	Improvement rate	Outcome assessment
Steinberg et al.[9]	Retrospective	7.1	997	Plagiocephaly, Brachycephaly	CI: 0.99, DD: 12.8 mm	Not provided	Not provided	87 %–95.7 %	Treatment success
Thompson et al.[36]	Prospective	6	116	Plagiocephaly	Severity score: 14.7	4.9	Not provided	27 %	Treatment success
Yoo et al.[37]	Retrospective	9	108	Plagiocephaly	CVA:16.0 mm CVAI: 10.68 %	6.56	Not provided	Final CVA: 4.7 mm Final CVAI: 3.00 %	Treatment length, treatment success
Teichgraeber et al.[38]	Retrospective	6	144	Brachycephaly Plagiocephaly	CI: 0.923 to 0.937 DFN: 0.143 to 0.2 FD: 9.4 to 9.5 OD: 3–3.4 mm	4.5 ± 1.8	Not provided	DFN: 0.007 to 0.080 FD: 4.6 mm–5.6 mm OD:1.2 mm–1.8 mm	Treatment success
Kluba et al.[39]	Prospective	6.3	62	Plagiocephaly	CVAI: 13.3 %	3.9	Not provided	CVAI: 9.2 %	Treatment success

Table 2
The scoring of included studies based on Newcastle–Ottawa Scale.

The Newcastle–Ottawa Scale's items	Representativeness of the exposed cohort	Selection of the non-exposed cohort	Ascertainment of exposure	The outcome of interest was not present	Comparability of cohorts	Assessment of outcome	Follow-up long enough for outcomes to occur	Adequacy of follow up of cohorts	Total score
Hinken et al.[23]	*	–	*	*	*	*	*	–	6
Çevik et al.[24]	*	–	–	*	*	*	*	–	5
Han et al.[25]	*	–	–	*	–	*	*	–	4
Freudlsperger et al.[26]	*	–	–	*	–	*	*	–	4
Kunz et al.[29]	*	–	–	*	*	*	*	–	5
Kunz et al.[32]	*	–	–	*	*	*	*	–	5
Mackel et al.[33]	*	–	–	*	–	*	*	–	4
Kluba et al.[10]	*	–	–	*	–	*	*	–	4
Couture et al.[19]	*	–	–	*	–	*	*	*	5
Graham et al.[40]	*	–	–	*	–	*	*	–	4
Graham et al.[14]	*	*	–	*	*	*	*	–	6
Graham et al.[21]	*	–	–	*	*	*	*	–	5
Grigsby et al.[22]	*	–	–	*	–	*	*	–	4
Kelly et al.[11]	*	–	–	*	*	*	*	–	5
Kim et al.[27]	*	–	–	*	–	*	*	–	4
Choi et al.[28]	*	–	–	*	*	*	*	–	5
Teichgraeber et al.[30]	*	–	–	*	–	*	*	*	5
Lam et al.[31]	*	–	–	*	*	*	–	–	4
Peethambaran et al.[34]	*	–	–	*	–	*	*	–	4
Seruya et al.[35]	*	–	*	*	–	*	*	–	5
Steinberg et al.[9]	*	–	*	*	*	*	*	–	6
Thompson et al.[36]	*	–	*	*	*	*	*	–	6
Yoo et al.[37]	*	–	*	*	–	*	*	–	5
Teichgraeber et al.[38]	*	–	–	*	–	*	*	–	4
Kluba et al.[39]	*	*	–	*	–	*	*	–	5

3.3. *Risk factors for treatment length*

Six predictive factors, including age at initiation of orthotic treatment, orthotic compliance, deformity severity, torticollis, gestational age, and deformity type, were considered for orthotic treatment length in the included studies. Twelve moderate quality studies examined the relationship between the age at initiation of orthotic treatment and treatment length[10,14,24,28,22,23,25,27,34,29,32,35]; eleven of them stated that there is a positive relationship between them,[10,14,24,28,22,23,25,27,29,32,35] and one study revealed a negative relationship.[34] Six moderate quality studies evaluated the relationship between deformity severity and treatment length, and reported that there is a positive relationship between the two factors.[14,28,19,37,29,32] Two moderate quality studies examined the relationship between gestational age and treatment length, one of them showed a positive relationship,[29] and another revealed no association.[14]

To evaluate the relationship between compliance and CRO treatment length, one moderate quality study found a negative relationship between these two factors.[37] The effect of torticollis on CRO treatment length was examined in one moderate quality study. There was a significant positive correlation between the two factors.[14] Two moderate quality studies individually evaluated the association between deformity type with treatment length and stated that there is no relationship between these two factors and treatment length.[28,23]

4. Discussion

This systematic review aimed to assess the predictive factors of treatment length and success rate of helmet orthotic therapy in children with skull deformities. Results of 25 included moderate quality studies showed that there are nine clinical parameters, including age at initiation of treatment, compliance, deformity severity, deformity type, torticollis, gestational age, gestational type, method of delivery, and developmental delay that have a role on length and success of CRO treatment in children with skull deformities. We found that age at initiation of treatment and deformity severity are the most important risk factors for treatment length and success of children with skull deformities. These findings are discussed in more detail in the following sections.

4.1. Age at initiation of treatment

Data from previous studies suggest that early diagnosis and applying appropriate management of newborns with cranial asymmetry are essential to reduce facial asymmetry and the following psychological burden on patients and their parents.[41] According to 11 moderate quality studies, moderate evidence showed that there is a positive relationship between age at initiation of CRO therapy and treatment length.[10,14,24,28,22,23,25,27,29,32,35] In these studies, the authors indicated that children younger than eight months need a shorter period of CRO treatment than those older than eight months. Moreover, there is moderate evidence for a negative relationship between age at initiation of orthotic therapy and treatment success.[9–11,14,24,21,25,23,31,33,20,26,37,29,32,35,36,38] Therefore, using a CRO for younger-aged children may provide excellent aesthetic results and require less treatment duration.

A key aspect that should be noted while using a CRO is considering brain development in different months of life. A baby's brain grows to 200 % of its birth size by six months old and only grows an additional 50 % over the next 24 months. At two years old, the brain is approximately 70 % of its adult size, and the remaining growth occurs gradually over the next four years. Choi et al.[28] reported that treatment length would increase by 0.391 months for every month of increase in age at initiation of treatment. It should be noted that children with advanced age can still benefit from CRO wear, but they will need more extended time periods.[35] It has been observed that children with cranial deformities can benefit from CRO treatment until 18 months old.[35] This finding contradicts the widely held belief that CRO use after one year of age is ineffective due to the cranium becoming too stiff or stationary. However, the course of treatment seems restricted until the 18th month; this may be due to the child's unwillingness to wear the orthosis, caregiver tiredness, or a lack of further progress. Consequently, it appears that orthotic therapy should be considered between the 3rd to 18th months of a child's life.

4.2. Compliance

Our findings show moderate evidence for a positive association between parent's compliance with CRO therapy and treatment success. However, the evidence for a relationship between CRO compliance and treatment length was conflicting. The patient's low adaptability to CRO therapy can reduce the chances of complete deformity correction and prolong the treatment period. The child's refusal of wearing the orthosis, slow recovery, and caregiver exhaustion are the most significant problems that increase the CRO treatment length. A possible explanation might be that most parents are commonly concerned whenever their child undergoes CRO treatment. Therefore, to prevent these problems and achieve the expected therapeutic results, it seems necessary to increase parental satisfaction and reduce their stress and worries about treatment.

The CRO compliance has traditionally been assessed using subjective approaches such as inquiring from the child's parents or nurse and reviewing clinical data.[23,37,36] Nevertheless, because they rely on the parents' assessments, these subjective approaches are imprecise and biased toward overestimation. Monitoring CRO compliance with pressure or heat sensors, on the other hand, can provide more accuracy in terms of daily CRO wearing time. What is required now is a study to assess objective compliance of CRO treatment in this population.

4.3. Severity of deformity

Anthropometric skull measurements provide a basis for clinical support of the correction or progression of the skull deformity. The relationship between cranial width and length as well as cranial diagonal dimensions are commonly used to determine the proportional association of the skull. Routinely, craniofacial experts use the cephalic index (CI), cranial vault asymmetry (CVA), and cranial vault asymmetry index (CVAI) to evaluate the value of asymmetry among skulls of various dimensions or between the same skull in different periods when growth takes place. The CI value can be estimated when we divide the cranial width by length multiplied by 100. CVA is the absolute value of the cranial diagonal difference, and CVAI is CVA divided by the longer diagonal diameter multiplied by 100. The CI values from 79 % to 84 % are considered to be within a normal range in infants aged from 0 to 12 months. The CVAI value should be lower than 3.5 % to represent a symmetric skull.[40,42]

In this systematic review, moderate evidence from a total of six moderate quality studies has revealed that there is a link between deformity severity and CRO treatment length.[14,28,19,37,29,32] In cases with similar final head shapes, less severity of deformities was related to shorter treatment length. Asymmetries ranging from moderate to severe had a more significant improvement with CRO than those that were from mild to moderate. In addition, moderate evidence obtained from ten moderate quality studies showed that there is a negative relationship between deformity severity and treatment success.[11,14,21,23,27,33,26,29,32,39]

4.4. Type of deformity

Deformational plagiocephaly is the most prevalent skull deformity that presents with unconformity of the right and left sides of the skull and face. Deformational brachycephaly and scaphocephaly are other deformities of the skull. Their prevalence is less common than that of plagiocephaly. These deformities are present with disruptions of the skull proportion. Many children may have both plagiocephaly and brachycephaly, needing a treatment strategy to provide a skull with more symmetry and proportion.[43] Wilbrand et al.[42] proposed a clinical classification system according to percentile curves of the normal cranial vault growth in the first 12 months of life. Using CI and CVAI, children with skull deformities were categorized into three groups of positional plagiocephaly, brachycephaly, and combined positional plagiocephaly and brachycephaly.

Among the included studies in this systematic review, most studies included children with plagiocephaly, brachycephaly, and both plagiocephaly and brachycephaly. No study had evaluated the success and length of treatment with CRO in children with deformational scaphocephaly. The role of deformity type on the outcome of CRO treatment has only been studied in two moderate studies.[28,23] Teichgraeber et al.[38] found that CI value improves significantly after CRO therapy in children with plagiocephaly. However, the improvement rate is not significant in children with brachycephaly. This difference might be attributed to the developing of the cranial vault and base. The cranial vault expands in response to the growing brain mass, whereas the cranial base expands due to endocranial absorption and ectocranial deposition. Cranial base development may be more compromised in brachycephaly than in plagiocephaly. The effectiveness of CRO treatment in children with plagiocephaly or with a combination of plagiocephaly and brachycephaly is higher than those with brachycephaly. According to Hinken et al. and Choi et al.'s studies,[28,23] the treatment length with CRO for children with brachycephaly is longer than that of the other two groups.

4.5. Torticollis

Congenital muscular torticollis and neck muscle imbalance are common findings in infants with deformational plagiocephaly.[2] Neck muscle imbalance leads to deviation of the skull positioning and provides unbalanced loading over the growing skull structures.[44] According to one moderate quality study, the existence of torticollis might lengthen treatment time.[14] However, the evidence for this relationship is conflicting. Moreover, moderate evidence suggests that torticollis does not appear to impair the chance of newborns to attain full cranial correction, and it does not appear to be a risk factor for CRO treatment success.

4.6. Gestational age (prematurity)

Skull deformation may initiate in utero. Prematurity is an influencing factor for the occurrence of cranial deformities.[45] Premature infants are exposed to skull distortion associated with the pliability of the immature skull structures. Premature infants have less neck muscle tone, meaning their heads are usually held in a side- or back-lying positions.[46] Moderate evidence revealed that gestational age does not affect the final outcome of CRO treatment, and premature infants can achieve complete correction if treatment is applied in a timely manner, and the treatment compliance would be high. We found conflicting evidence regarding the effect of prematurity on CRO treatment length. In two moderate quality studies, Graham et al.[14,21] stated that prematurity is not a strong predictor in attaining complete correction of skull deformity in CRO. In this study, the role of prematurity has been assessed on treatment outcomes of CRO treatment for infants with brachycephaly. The authors considered patients as premature in case they were born at 37 weeks of gestation or before. The age for initiation of CRO treatment was corrected by considering the number of weeks of prematurity minus the postpartum age and then rounded it to the closest half month. The study results have shown that corrected age at the initiation of CRO treatment has considerable impact on treatment outcomes. Overall, rate of deformity correction in children with younger ages is higher than that of those with older ages. In addition, in their cohort study, Kunz et al.[29] stated that infants with a lower gestational age would experience a shorter course of CRO treatment. Premature babies may respond faster to treatment because they have more growth potential in the first months of life.

4.7. Gestational type, delivery method, and developmental delay

Conflicting evidence suggests that there is no relationship between gestational type, delivery method, or developmental delay and CRO treatment success. Delivery method may be a risk factor for developing cranial deformities. Vaginal delivery causes substantial deformity of the fetal skull while it moves through the birth canal. Assisted delivery is another predisposing factor for the occurrence of skull deformities.[47] Scholars have debated the impact of the delivery method on the treatment outcome with CROs. Steinberg et al.[9] indicated that vaginal delivery and multiple gestations are protective against repositioning therapy or physical therapy treatment failure. However, they discovered no link between these variables and CRO therapy success.

Repositioning treatment or physical therapy failure might increase if neuromuscular development is delayed. It is also in line with the etiopathogenesis of skull deformations postulated. Infants may be unable to overcome deforming pressures due to delays in muscle development.

Only one study investigated the relevance of this parameter to CRO treatment success. Steinberg et al. discovered no link between developmental delay and helmet therapy success. The helmet can provide a fixed-shape environment and control skull growth while isolating extrinsic deforming influences like torticollis and developmental delays. Thus, it may mitigate the impacts of these conditions.

4.8. Limitations

When interpreting the findings of this systematic review, there are numerous limitations that should be considered. First, there is significant evidence that CRO compliance is positively linked with treatment success and inversely associated with treatment duration. However, there is inadequate research examining the effect of compliance on CRO therapy results and duration in infants with skull deformities. Second, little research has looked into the effects of gestational type, delivery method, and developmental delay. The relevance of all clinical indicators that may have a role in helmet treatment results was emphasized in this systematic review. However, from the included studies Eighteen were retrospective, and seven had prospective designs. Hence our findings are based primarily on data from retrospective studies, which are prone to various biases that might mask variations in predicting variables and helmet treatment results. Third, the meta-analysis was hampered by the heterogeneity of various measuring methodologies, definitions of treatment outcomes, assessment processes, and reporting methods across the included studies. Finally, the findings were limited to articles written in English. It's possible that adding in other languages might change the outcome.

5. Conclusions

According to moderate evidence, CRO treatment length is linked to a patient's age at the start of treatment and deformity severity. Moreover, treatment success is correlated with patient's age at the start of treatment, CRO compliance, and deformity severity. It indicates that younger infants with less severe deformities will have a shorter treatment duration. Moderate evidence indicates no relationship between the presence of torticollis and gestational age with CRO treatment success. There is conflicting evidence regarding the relationship between deformity type, gestational type, delivery method, or developmental delay and CRO treatment success. This data may help clinicians in determining CRO treatment decisions for infants with skull deformities. Further longitudinal prospective studies are needed to evaluate all possible predictors of helmet treatment effects.

CRediT authorship contribution statement

Hoda Hashemi: Conceptualization, Formal analysis, Methodology, Writing – original draft, Writing – review & editing. **Taher Babaee:** Conceptualization, Formal analysis, Methodology, Project administration, Supervision, Writing – review & editing. **Vahideh Moradi:** Conceptualization, Methodology, Writing – review & editing. **Mahtab Bagheri:** Conceptualization, Methodology, Writing – review & editing. **Mohammad Javad Moghadam:** Conceptualization, Methodology, Writing – review & editing. **Maryam Ashkar:** Conceptualization, Methodology, Writing – review & editing. **Behnaz Tavakoli:** Conceptualization, Methodology, Writing – review & editing. **Amir Ali Gordahani:** Conceptualization, Methodology, Writing – review & editing. **Zohreh Habibi:** Conceptualization, Methodology, Writing – review & editing.

Declaration of competing interest

The author declares that the article content was composed in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Appendix A. Supplementary data

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Abbreviations and Acronyms

MIS: minimally invasive suturectomy
CRO: cranial remolding orthosis
CI: cephalic index
CVAI: cranial vault asymmetry index
DD: diagonal difference
RDD: reductions in diagonal difference
CHOA: Children's Healthcare of Atlanta
CVA: cranial vault asymmetry
SBA-CBA: skull base asymmetry-cranial base asymmetry
OTDA: orbitotragial depth asymmetry
DP: deformational plagiocephaly
AB: symmetrical brachycephaly
ODD: oblique diagonal difference
CR: cephalic ratio
DFN: divergence from the norm
FD: forehead asymmetry
OD: orbital asymmetry
PCAI: posterior cranial asymmetry index