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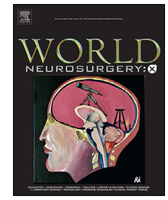

Minimally invasive craniectomy and postoperative cranial remolding orthotic treatment in infants with craniosynostosis: A multicenter prospective study

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A R T I C L E I N F O

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1. Introduction

Craniosynostosis is the premature ossification of one or more of the cranial sutures that causes an asymmetry of the skull shape. According to Virchow's law, the affected bones are fused to each other and skull growth is restricted along a direction that moves perpendicular to the sealed suture, leading to different deformities in the skull depending on the site of involvement.[1] Among the congenital abnormalities of the craniofacial area, craniosynostosis is the second most prevalent after cleft lip/palate, as it is estimated to occur in 1 out of 2500 live births.[2]

Craniosynostosis may be syndromic (when a cranial abnormality is associated with other defects or developmental delay) or nonsyndromic (more than 75% of cases). Nonsyndromic craniosynostosis have an uncertain etiology. It can be spontaneous, genetic, and familial.[1] The most common types of craniosynostosis is sagittal synostosis, which includes 40%–60% of all types of craniosynostosis. It is followed by coronal synostosis, metopic synostosis, and lambdoid synostosis with prevalence of 25%, 10%, and 1–5.5%, respectively.[3]

Treatment strategy for treating non-syndromic craniosynostosis depends on severity of the deformity and the age of the patient. Surgical treatment strategies for single-suture craniosynostosis have changed during the past years and shifted from complete cranial vault reshaping techniques[4] to more recent minimally invasive techniques applied mainly for infants younger than 6 months.[5] With the evolution of minimally invasive surgery (MIS) technique, the mortality and morbidity rate of surgery has decreased significantly and it is possible to perform such intervention for younger children. The transfusion rate in MIS is much lower than that of the extensive cranial vault remodeling surgery method.[6] Additionally, the operative time in MIS is about 45–100 min, which is significantly less than the extensive cranial vault remodeling technique (4–8 h).[7] These benefits reduce the morbidity of patients and reduce the duration of hospital stay. According to the functional matrix theory presented by Moss, after suturectomy, the growth of the brain can be an excellent internal distractor to continue the treatment process.[8] Using a post-operative cranial remolding orthosis (CRO), the distractive forces of the developing brain can be directed to the flattened areas.[9]

Data from previous studies suggest that MIS with post-operative CRO is an excellent approach for treatment of patients with single-suture craniosynostosis in North America and Europe.[6,10–12] The aims of using post-operative CRO are to allow continued reshaping of the skull and

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prevent the needs of revision surgery.[9] However, most of the previous studies in this field have been retrospective chart reviews which had inferior level of evidence compared with prospective studies and thus were subjected to numerous biases.[5] This prospective study is therefore set out to assess the results of the first cohort of patients with single-suture craniosynostosis who received MIS with post-operative CRO treatment in Iran.

2. Methods

2.1. Study design and patient's recruitment

This was a multicenter prospective study that was conducted under the approval of a research committee of Iran University of Medical Sciences (approval number 1398.178). Data gathering was performed from June 2019 to August 2021. Diagnosis of craniosynostosis was made by close cranial shape examination and measuring the anthropometric values of the skull. After clinical examination, a plain radiograph or three-dimensional computed tomography reconstruction was performed to confirm the diagnosis and surgical planning. In this study, for imaging of the patients' skull, we followed the "Guideline for Care of Patients with the Diagnoses of Craniosynostosis".[13] The CT scan is considered as the predominant imaging technique for diagnosis of craniosynostosis and preoperative planning of surgery. Because of the low mineralization of the cranium before the age of 3 months, the accuracy of plain radiograph for identifying minor sutural synostosis is suboptimal. In this study, for patients with age of <3 months, those with suspected intracranial pressure, and patients with minor sutural synostosis, a preoperative CT scan was considered.

All patients included underwent endoscopic-assisted suturectomy followed by postoperative CRO. Inclusion criteria were the following: children younger than 6 months, having non-syndromic single-suture craniosynostosis, and no prior reconstructive surgery of the skull. All patients with syndromic craniosynostosis and those who were candidates for open surgery were not included the study. Written informed consent was obtained from parents of included patients.

2.2. Surgical techniques

All included patients underwent MIS to open the prematurely fused suture. Following induction of anesthesia and preparation of the infant for surgery, depending on the type of synostosis, a surgical protocol was adopted. Our surgical techniques for MIS was similar to the same studies that were all aiming to release a fused suture with a small incision.[1,14–16]

For sagittal craniosynostosis, the procedure was conducted while the patient was lying with the head extended in a prone position. After skin preparation with povidone-iodine and drape, two transverse incisions were made with 3 cm length at 1 cm rear the anterior fontanelle and 1 cm in front of the lambdoid suture. A subcutaneous tunnel between the two incisions with complete homeostasis was created by endoscope. Burr holes were placed over the anterior and posterior sagittal sutures. Strip craniectomy was performed after dissecting the dura from the fused bone by endoscope for safety manipulation of the compromising space. The fused bone was removed from the fontanelle anteriorly to the lambda posteriorly. The width of the craniectomy site was targeted at 3 cm approximately. Following hemostasis, both incisions were routinely closed and pressure patching was performed.

In unilateral coronal craniosynostosis, the patient was positioned supine with the head rotated to the contralateral side. After prep and drape, a burr hole was done for accessibility of the suturectomy site. The pericranial flap was dissected by endoscope and a strip craniectomy was done 1 cm in width subperiosteally with bone scissors. The craniectomy was run from the anterior fontanelle to the zygomatic arch. After that, the incision was closed with nylon.

For metopic craniosynostosis, the patient was positioned in supine with head in neutral position using a horseshoe headrest. A 2.5–3 cm transverse incision was made in front of the anterior fontanelle behind the hairline. Two burr holes were placed on both sides of the metopic suture at the level of the skin incision. Metopic suturectomy extended from the anterior fontanelle to the nasofrontal suture with Mayo scissors. Bleeding from the ethmoidal veins to the sagittal sinus was coagulated with bipolar cautery. After that, the frontal bone was remodeled by Rongeur, and hemostatic agents were exerted over the bone margins. Finally, incisions were closed with resorbable stitches.

2.3. Postoperative CRO

All patients were referred 7 days after the surgery and after skull swelling had diminished to initiate the postoperative CRO treatment. To make a CRO, 3D image of infants' skull was obtained by a non-contact optical scanner (Vorum, SPECTRA 3D, Vancouver, Canada) with an accuracy of 0.1 mm. Parents were reassured that the scanner would not emit harmful radiation and would not adversely affect the infants. The usage of 3D scanning eliminates the need for casting, which is an advantage for infants who have undergone surgery recently and their parents. The initial scan was modified by computer-aided design software (Canfit, Vorum) to create a new shape of the baby's head with more symmetry and proportion. These modifications were made in such a way that the prominent parts of the skull were in contact with the orthosis to prevent further growth of that part. However, the flattening areas were widened so that the orthosis would direct the growth of the skull to those areas.

To supply a positive model of the infant's skull, a urethane foam block was sculptured by a computer-aided machine. After preparing the positive mold, we applied a ten mm Plastazote foam on it and vacuumed a four mm polypropylene thermoplastic over the model. After the thermoplastic sheet had cooled, the orthosis trim lines were determined and the sharp edges were polished to be smooth. Finally, the CRO was fitted for each patient.

At the time of delivery, we explained the instructions for using the CRO as well as the points that the parents had to follow while using it. These included how to use and how to clean the CRO. We asked the parents to clean the CRO after each removal with a damp cloth soaked in baby shampoo and medical alcohol.[11] To prevent forgetfulness, this instruction was also given to the parents in writing.

After fitting the CRO, we asked the parents to stay in the center for about 1 h so that in cases such as displacement or excessive pressure points, the CRO would be rectified. If there were high-pressure points, we would unload that part by removing a thin foam layer inside the CRO. For excessive displacement, a 2–3 mm thick foam was added to the posterior-inferior part and temporal extension. During this time, sleeping with CRO and breastfeeding of infants were also examined. Parents were asked to use the orthosis for 23 h a day. During treatment, all infants were monitored for checking up the CRO and head shape in an interval of 1- to 3- weeks. At each visit, if needed, we removed a thin layer of the CRO foam so that it would accommodate the growth of the infant's skull. If the CRO needed to be replaced, the baby's head was scanned again and a new CRO was made like the previous one.

To measure the compliance of the CRO, a form was provided to the parents to record the daily hours of wearing it. To assess the compliance rate, we divided the daily wearing time of CRO by the maximum suggested time multiplied by 100. To ensure that the forms were completed, the first author sent a reminder message to the parents weekly after obtaining parental permission. To evaluate the potential challenges associated with CRO wear, the parents were asked to put mark on a feedback survey with visual analog scale format.[17]

2.4. Anthropometric and clinical data extraction

Demographic, pre-operative, intra-operative, and post-operative information of each patient were recorded. Demographic information such as patient's gender, age at surgical operation, and type of craniosynostosis (sagittal, metopic, and unilateral coronal) was documented.

The pre-operative, intra-operative, and post-operative data included the amount of blood transfusion, duration of operation (including time under anesthesia), duration of hospitalization, and information about post-operative complications (such as wound infection and bleeding) were also recorded in the patients' clinical information form.

Common anthropometric cranial measurements including the cranial length, cranial width, cranial circumference, and oblique cranial diagonal diameters were measured and recorded before the surgery, at initiation of CRO treatment, and at the cessation of CRO treatment. All these measurements were performed with a meter and a caliper by one assessor. To minimize the measurement error, all measurements were repeated 3 times, and the average of measurements were considered to be analyzed. To evaluate the symmetry and proportion of the patients' skull at different stages of treatment, the values of the cranial index (CI) and cranial vault asymmetry index (CVAI) were calculated.

The CI was measured by dividing the cranial width by the cranial length multiplied by 100. The CVAI was defined as the difference between the long diagonal and short diagonal diameters of the cranium divided by the long diagonal diameter multiplied by 100.[18] The normal rate of CI value was reported to be between 82% and 84%.[19] Moreover, a CVAI of less than 3.5% was reported to be an acceptable rate for the symmetry of the skull.[18]

The Total volume (TV), anterior hemisphere volume (AV), and AV to TV were also acquired with aforementioned scanner. After setting the anatomical landmarks at the exocanthion-tragion line of the images, the TV was calculated above this line.[17] To determine the posterior and anterior skull volume, we divided the cranium into two anterior and posterior sections at the tragi point. The proportional volume of the AV to the TV was computed to preclude the normal growth from confounding the measurement.

2.5. Statistical analyses

Statistical data analyses were performed by IBM SPSS Statistics version 24 (IBM Corp, USA). Normality of data was assessed using Shapiro–Wilk test. A one-way repeated measures analysis of variance was conducted to compare the mean values of CI and CVAI in different stages of treatment (before surgery, at initiation of CRO treatment, and after cessation of CRO treatment). A paired-samples *t*-test was conducted to evaluate the impact of the treatment on patients' skull volume. Regarding age at surgery, we categorized patients into two groups of ≤3 months and 4–6 months. An independent sample *t* test and a Mann–Whitney *U* test for values of CI and CVAI at final follow-up were conducted between two groups. A p-value of <0.05 was deemed to be the significance level.

3. Results

38 patients in total (15 with sagittal craniosynostosis, 10 with unilateral craniosynostosis, and 13 with metopic craniosynostosis) were included in the study. The cohort comprised 21 boys and 15 girls, with an average age of 3.40 months (range 1.64–6.00 months) at the time of surgery. Totally, there were 26 cases (68.4%) with age of ≤3 months and 12 cases (31.6%) with age of 4–6 months at surgery. The average operative time including anesthesia time was 95.38 min (range 30–180 min). The average length of hospital admission was 2.5 days (range 2–4 days) (Table 1). Four patients were twins and three were in breech position. Sutural abscess occurred for 3 patients (8.33%) that required oral antibiotic drugs. A total of 6 patients (16.6%) received a blood transfusion (80–120 ml of allogenic blood).

The mean treatment length with CRO for sagittal, unilateral, and metopic craniosynostosis was 6.70, 7.07, and 7.70 months, respectively. The average follow-up after cessation of CRO treatment was 4.68 months (range 2–8.55 months). None of the patients needed second surgery until final follow-up. The changes of head shape and anthropometric data in each group of included patients are highlighted in Table 2 and Figs. 1, 2, and 3). From this data, we can see that there is a statistically significant difference at the $p < 0.05$ level in CI values for the patients with sagittal craniosynostosis. Moreover, the improvement in CVAI values were statistically significant in all three groups of participants ($p < 0.001$). Regarding age at surgery, there were no significant differences in values of CI and CVAI at final follow-up between patients with age of ≤3 months and those with ages of 4–6 months ($p > 0.05$) (Fig. 4).

Results of paired-sample *t*-tests showed statistically significant improvements in AV and TV from initiation to cessation of CRO treatment in all three groups of participants ($p < 0.001$). Regarding the AV to TV ratio, there was a significant difference from initiation to the cessation of CRO treatment in sagittal and metopic groups of participants ($p < 0.05$) (Table 2).

Table 1
Preoperative, intraoperative and postoperative data of included patients (n = 38).

Variables	Mean ± SD (range)
Age at time of surgery (month)	3.40 ± 0.21 (1.64–6.00)
Operation time including anaesthesia time (min)	95.38 ± 41.40 (30.00–180.00)
Admission time (day)	2.50 ± 0.67 (2.00–4.00)
Cephalic Index (%)	78.72 ± 1.85 (64.12–93.24)
Circumference (cm)	40.28 ± 0.49 (36.00–45.00)
Total blood transfusion (ml)	54.61 ± 15.06 (30.00–80.00)

Table 2
Descriptive statistics for parameters of interest during the different treatment phases.

Variables	Sagittal (n = 15)	Unilateral coronal (n = 10)	Metopic (n = 13)
	Mean ± SD	Mean ± SD	Mean ± SD
Age at initiation of CRO (year)	3.40 ± 1.11	3.42 ± 1.77	3.50 ± 1.16
Age at cessation of CRO (year)	10.56 ± 1.26	10.70 ± 2.16	10.69 ± 1.45
Duration of CRO treatment (months)	6.70 ± 0.84	7.07 ± 0.79	7.70 ± 0.67
Cephalic Index (%)			
Before surgery	69.41 ± 3.60	89.09 ± 6.64	84.61 ± 3.85
At initiation of CRO	71.07 ± 3.92	88.22 ± 6.22	84.91 ± 4.17
At cessation of CRO	80.83 ± 3.40	87.55 ± 3.26	85.24 ± 2.88
P	<0.001	0.65	0.71
Cranial Vault Asymmetry Index (%)			
Before surgery	4.09 ± 1.90	8.64 ± 3.67	4.62 ± 3.04
At initiation of CRO	2.53 ± 2.57	7.01 ± 2.96	4.42 ± 3.65
At cessation of CRO	0.74 ± 0.46	1.46 ± 1.02	0.89 ± 0.54
P	<0.001	<0.001	0.006
Anterior volume (cm^3)			
At initiation of CRO	544.91 ± 105.84	475.63 ± 85.14	516.38 ± 84.24
At cessation of CRO	753.73 ± 95.17	737.63 ± 86.96	752.18 ± 109.97
P	<0.001	<0.001	<0.001
Total volume (cm^3)			
At initiation of CRO	1078.20 ± 184.65	920.40 ± 118.36	1088.09 ± 145.84
At cessation of CRO	1416.99 ± 143.86	1356.922 ± 84.65	1410.78 ± 171.01
P	<0.001	<0.001	<0.001
Anterior to total cranial volume (cm^3)			
At initiation of CRO	49.17 ± 3.62	51.62 ± 5.12	47.44 ± 4.36
At cessation of CRO	52.31 ± 3.66	54.50 ± 7.02	53.295 ± 3.95
P	0.001	0.19	0.001

CRO, cranial remolding orthosis; SD, standard deviation.

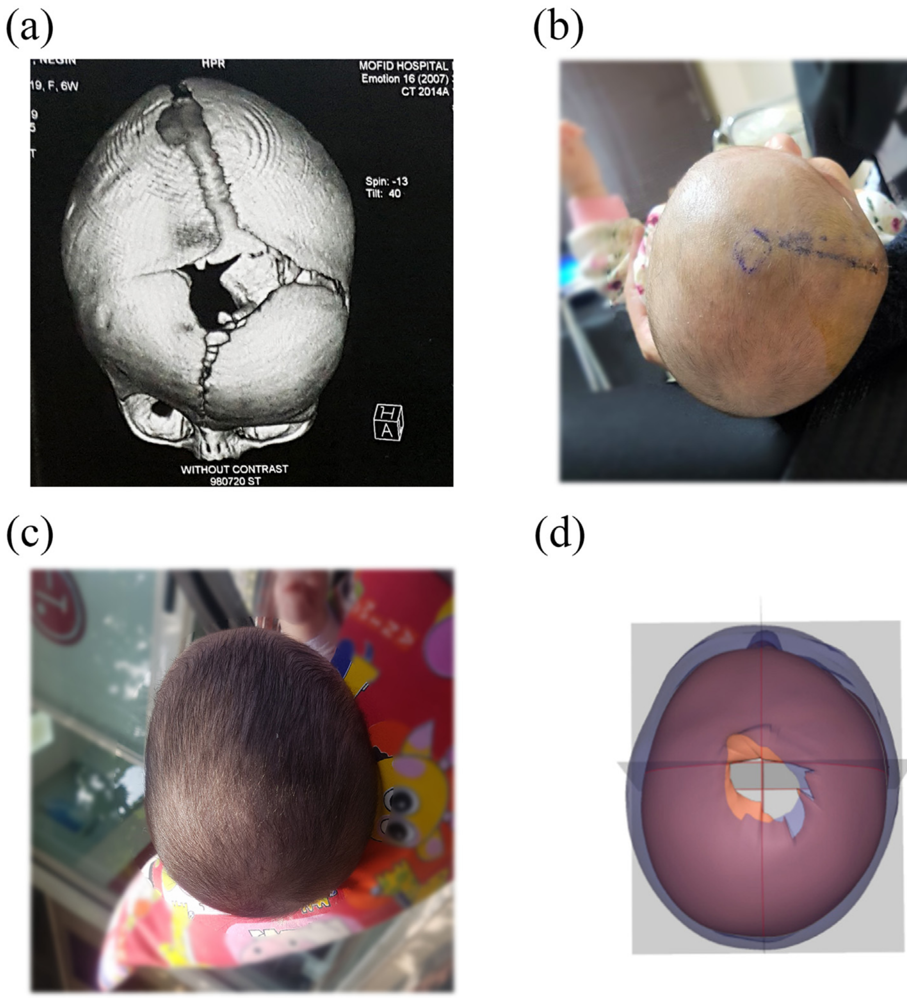

Fig. 1. Skull images of a patient with unilateral coronal craniosynostosis. (a): Preoperative CT scan shows the fusion of the right coronal suture. (b): Post-operative image of the patient's skull. (c): The patient's skull at cessation of cranial remolding orthosis treatment. (d): The semitransparent 3D scan of the patient's skull at the initiation and cessation of cranial remolding orthosis treatment.

A total of 81 CROs were manufactured (average 2.38 for each patient). The cost of making each CRO was $60. The average daily hours of CRO wearing was 18.54 h (range 16.53–20.61 h) based on the parents' reports. Parents' satisfaction with the surgical intervention and CRO treatment was 89% (Table 3). Receiving negative feedbacks from others (54%) while using CRO was the main challenge of the patients' parents (Table 3).

4. Discussion

Premature closure of skull sutures and subsequent skull deformity is mainly caused by compensatory overgrowth in the area of adjacent sutures.[15] For this reason, immediately after the diagnosis of craniosynostosis, the patient undergoes surgery to prevent further progression of cranial deformity. This study was set out with the aim of assessing the effect of MIS and postoperative CRO treatment on cranial symmetry of infants with craniosynostosis. After applying this method of treatment to 38 cases, including most prevalent forms of single-suture craniosynostosis, we found that endoscopic-assisted craniectomy followed by postoperative CRO is a safe strategy with satisfactory results. Based on findings of the present study, it would be advised that MIS technique should be accompanied by CRO treatment in patients with any single-suture craniosynostosis under the age of 6 months.

4.1. *Treatment results*

In this study, the mean age at time of surgery was 3.40 months and all cases had an age of <6 months at surgery. Regarding age at craniosynostosis surgery, some authors recommend to do MIS by the age 3–4 months.[20] However, according to a systematic review and meta-analysis, most authors prefer to conduct the procedure of MIS at an age of <6 months.[21] Evidence suggests that patients undergoing MIS infrequently need a blood transfusion. Transfusion rate for our studied patient population was 16.6% which was higher than that of reported by Jimenez et al.'s studies (1.7%–6.7%)[10,12,22] and lower than that of Honeycutt's study (23%).[23] Still, It is encouraging to compare this finding with the results of a systematic review and meta-analysis which revealed that the transfusion rate for MIS method is 32.3% (215/665).[21]

Anthropometric cranial assessment establishes a reference for clinical proof of refinement or deterioration of the cranial deformity. Common clinical calculations for evaluation of cranial symmetry and proportion include CI and CVAI.[9] In sagittal craniosynostosis, the amount of scaphocephalic shape correction can be measured by evaluating CI changes. In this study, the value of CI was changed significantly from initiation of the surgery (69%) to cessation of the CRO treatment (81%) and reached the normal value reported in previous study.[19] In their study, Jimenez et al found that more than 96% of patients with sagittal craniosynostosis who underwent MIS and CRO treatment showed good to excellent CI results.[6] Other researchers reporting similar results, found that the final

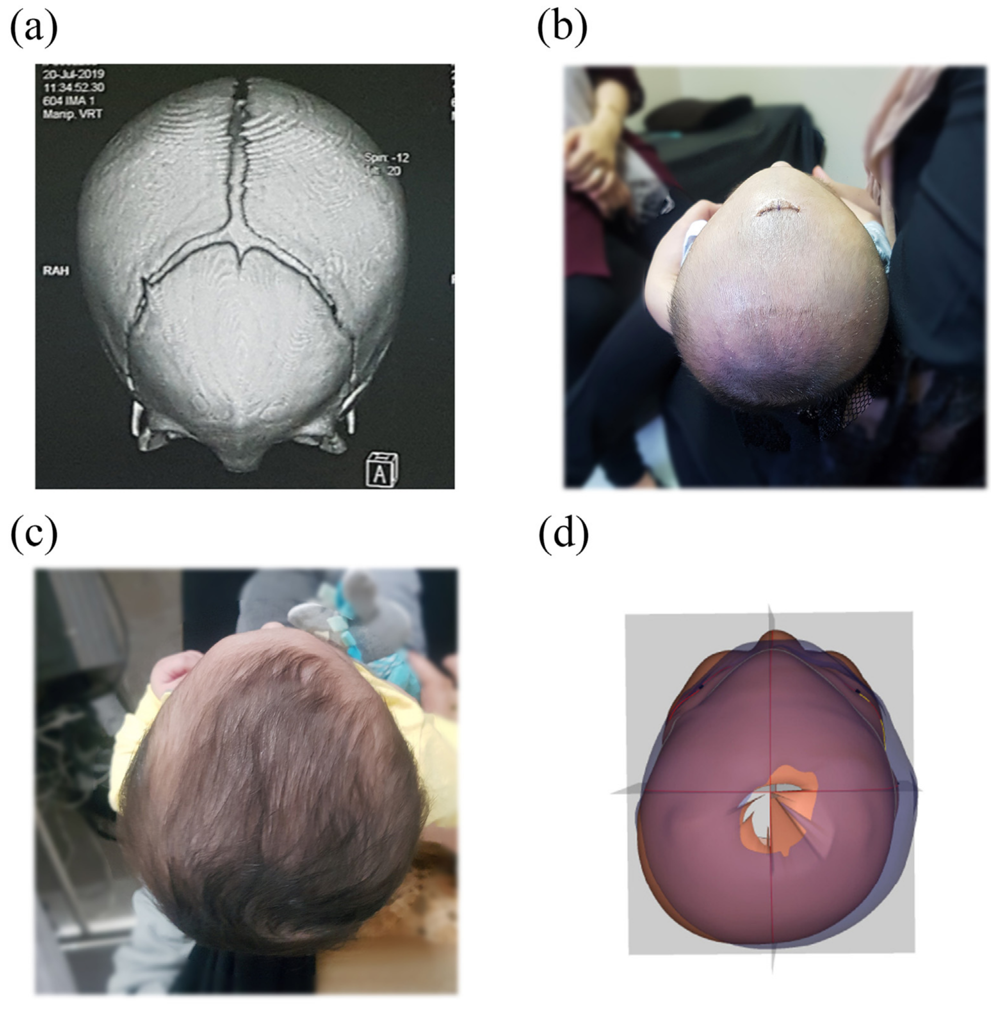

Fig. 2. Skull images of a patient with metopic craniosynostosis. (a): Preoperative CT scan shows the fusion of the metopic suture. (b): Post-operative image of the patient's skull. (c): The patient's skull at cessation of cranial remolding orthosis treatment. (d): The semitransparent 3D scan of the patient's skull at the initiation and cessation of cranial remolding orthosis treatment.

CI value between patients who underwent MIS and CRO treatment and those who underwent extensive cranial vault remodeling were almost similar and had no significant difference.[24,25] In this study, the mean duration of using post-operative CRO in patients with sagittal craniosynostosis was 6.7 months. According to the study of Iyer et al, the optimal duration of using post-operative CRO for patients with sagittal craniosynostosis was reported to be between 7 and 9 months,[26] which was longer than that of the present study. Although the optimal CRO treatment duration can be affected by the CRO compliance rate,[22] the role of this variable on the duration of CRO treatment was not considered in the study of Iyer et al. In the present study, compliance rate of CRO treatment was 80.60%.

Unilateral coronal craniosynostosis causes plagiocephaly and is associated with frontal, supraorbital and orbital asymmetry. The most important radiological and clinical characteristic of this group of patients is backward and upward deviation of the supraorbital region, which is known as “Harlequin orbit".[1] The purpose of CRO treatment for this group of patients is to prevent the growth of the contralateral frontal bone and to direct the growth to the ipsilateral plagiocephalic frontal bone. After opening the fused suture and during the use of CRO, the growth of the brain causes the forehead area to expand in the anterior-inferior direction, and thus causes the orbit of the involved side to descend and move forward.[10] In unilateral coronal craniosynostosis, the skull symmetry can be measured by evaluating CVAI changes to evaluate the improvement of anterior plagiocephalic shape of the patients' head. In the present study, the value of CVAI at initiation of the surgery was 8.64% that reached 1.46% at cessation of the CRO treatment. This finding is consistent with that of Wolfswinkel et al.[27] It has previously been observed that the improvement rate of plagiocephaly in unilateral coronal craniosynostosis is related to the severity of the deformity and the patient's age at the time of surgery.[10] Therefore, the final results of MIS with CRO treatment may be suboptimal for older patients with more severe deformities.[22]

In metopic craniosynostosis, the skull becomes triangular and a middle anterior calvarial ridge is formed on the forehead.[1] Consequently, the overall frontal volume is reduced and therefore requires early surgical intervention to allow the normal growth of the brain and skull.[12] In this patient population, the amount of correction of the trigonocephalic shape can be measured by evaluating the changes of the AV.[28] Our results showed that the AV at the beginning of CRO treatment was 516.4 cm^3, which reached 752 cm^3 at the end of the treatment, consistent with the value obtained in the previous study (750 cm^3).[28] We also found that the TV at the beginning of the CRO treatment was 1088 cm^3, which reached 1410 cm^3 at the end of the treatment, which is slightly lower than that reported by Meulstee et al for normal 12-month-old children (1477 cm^3).[29] This result may be explained by the fact that the age of

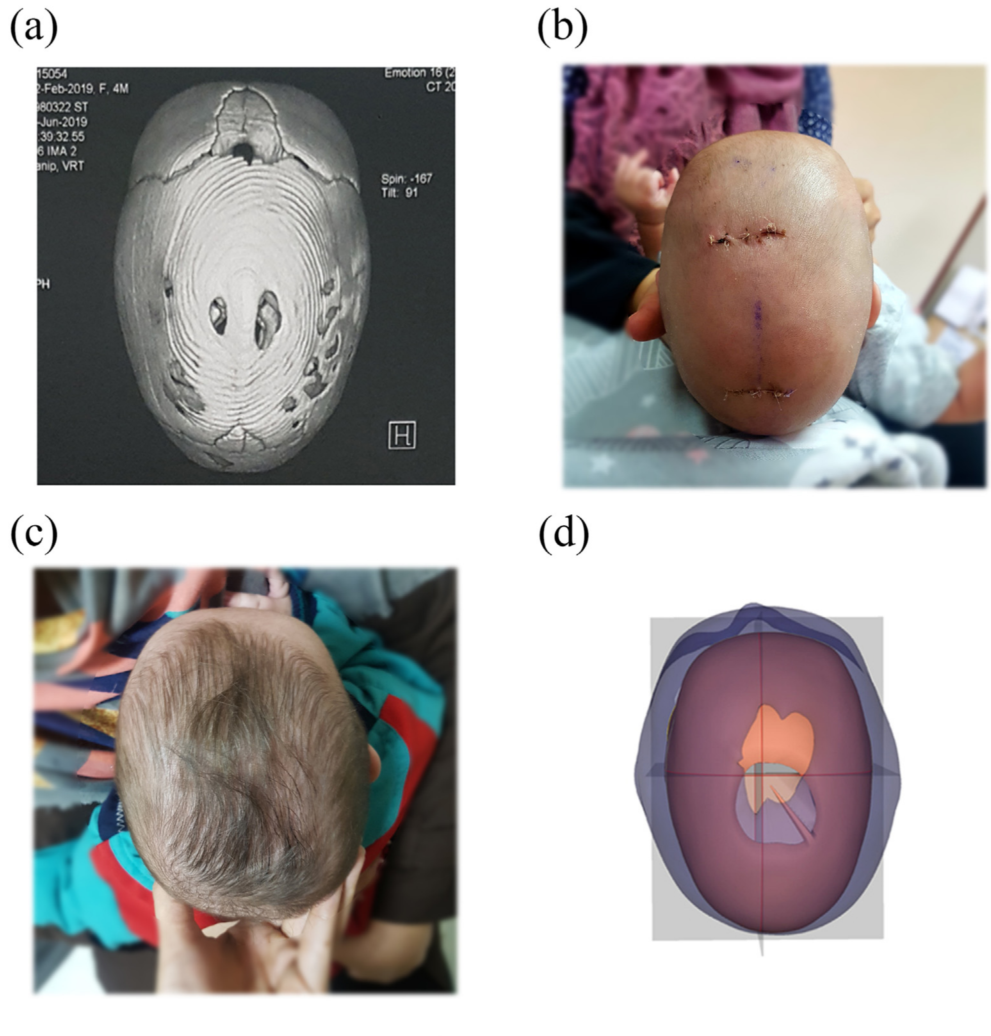

Fig. 3. Skull images of a patient with sagittal craniosynostosis. (a): Preoperative CT scan shows the fusion of the sagittal suture. (b): Post-operative image of the patient's skull. (c): The patient's skull at cessation of cranial remolding orthosis treatment. (d): The semitransparent 3D scan of the patient's skull at the initiation and cessation of cranial remolding orthosis treatment.

completion of CRO treatment in the present study was 10.7 months and the skull volume was calculated at that time. However, in the study of Meulstee et al,[29] the skull volume was reported for children aged 3, 6, 9, 12, 15, 18, and 24 months, and information about the normal skull volume was not reported for children aged 10–11 months. In the present study, the TV in patients with metopic craniosynostosis was the same as the value reported for normal children.[29] A possible explanation for this might be that following the closure of the metopic suture, the biparietal region of the skull undergoes compensatory widening, and therefore this compensatory deformation causes the TV of the skull not to change compared to normal children.[30] Therefore, to investigate the skull volume changes of this patient population, considering anterior skull volume may provide more accurate results.

In this study, the ratio of AV to TV increased in all three studied groups, which was significant for the group of patients with sagittal and metopic craniosynostosis. These results are likely due to the bilateral temporal widening of the skull in these two groups of patients while using CRO, which can change this ratio. These results were also confirmed in previous studies.[30,31]

4.2. The CRO-related problems

The outcome of MIS is thoroughly tied to the use of CRO.[5] However, some authors considered this part of treatment as the main handicap or complication of treatment.[15] In this study, using a very light custom-made CRO (about 150 g), the complication was low. We measured the complications associated with CRO using a survey in VAS format. The survey items covered all areas that may be affected by CRO. We found that the rate of pressure sores or contact dermatitis was very low. Only 13% of parents reported pressure sore or contact dermatitis at the temporal extension skin interface while using the orthosis. In this regard, we asked the parents to stop using the CRO for 2 to 3 consecutive days until the symptoms disappear. It should be noted that all parents reported that CRO was well tolerated by their child and none of the parents stopped using CRO due to their child's intolerance. These results were in line with other studies.[14,15]

In this study, the most significant challenge that parents experienced while using CRO was receiving negative feedback from others. This forced some parents to temporarily cut off their communication with others in order to finish the CRO treatment more comfortably. More than half of the parents of our patients stated that they were exposed to many questions from others during the treatment of their child and sometimes in social interactions, the meaningful looks and gestures of others made them worried and anxious. Studies have shown that appearance plays an important role in social interactions. People with different appearances are subjected to more unwanted attention than people with normal

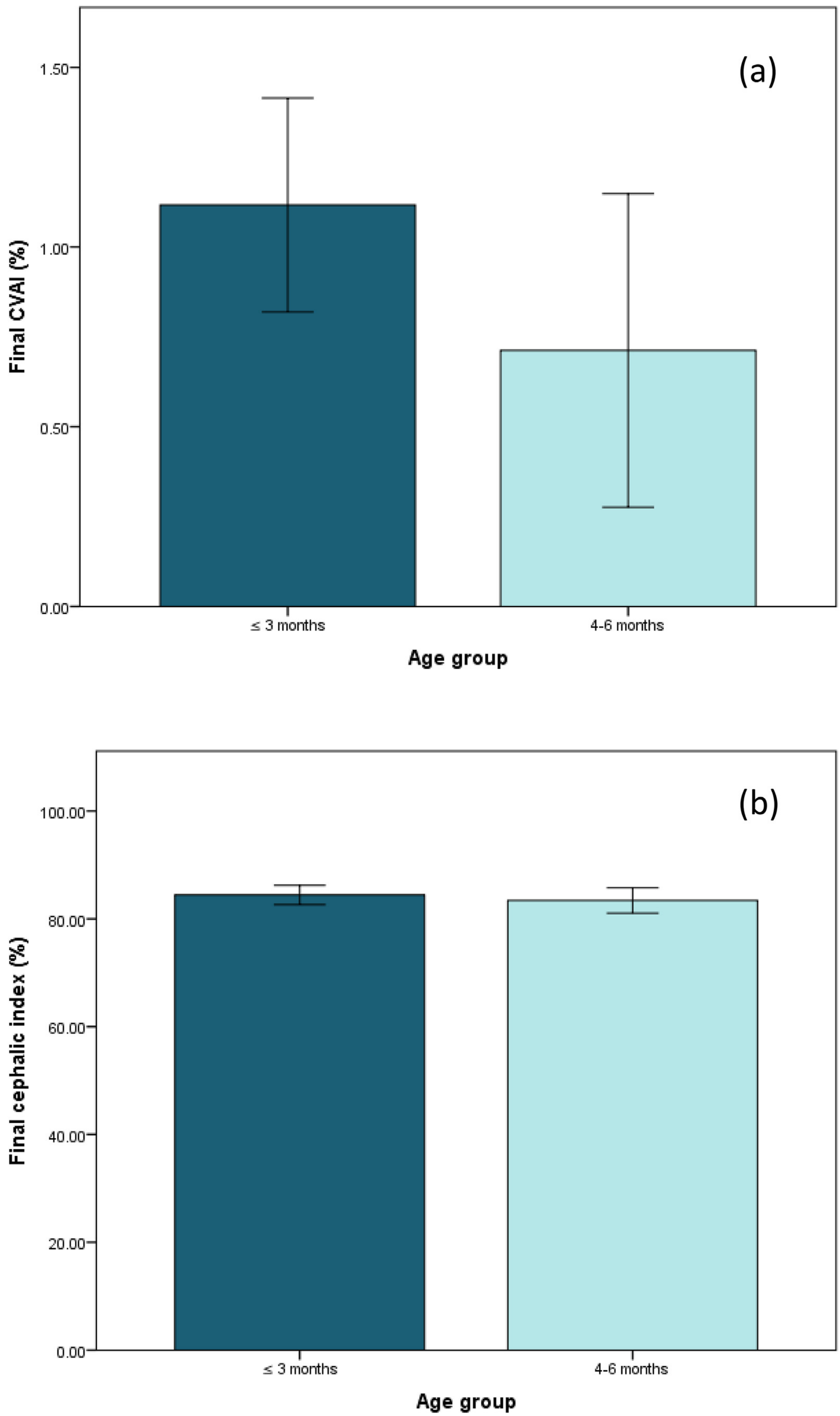

Fig. 4. Comparison of cranial vault asymmetry index (a) and cephalic index (b) values at final follow-up between two groups of patients with age of ≤3 and 4–6 months at surgery.

Table 3
The average recorded percentage of visual analog scale in Parental feedback study outcomes.

Questions	Average percentage (%)
Satisfaction with final appearance of baby's head	89
Problems with contact dermatitis or pressure sores while using the CRO	13
Problems with perspiration while using the CRO	31
Problems with CRO displacement on baby's head	27
Challenges with donning and offing of the CRO	22
Received negative feedback from others while using the CRO	54
Problem with baby's sleeping while using the CRO	28

CRO, cranial remolding orthosis.

appearances.[32] This can lead to increased levels of anxiety and avoidance of social situations.[33]

4.3. Limitations

This study had some limitations that should be considered. Firstly, one blind spot which could have affected the accuracy of the data is the movements of the patient during the head scan. In this study, in order to minimize measurement errors, if the patient moved during the measurements, the scanning was repeated.

Secondly, children affected by single-suture craniosynostosis can show an adverse neurodevelopmental outcome, especially concerning visual, language, and motor domains.[1] In this study, we did not evaluate these neurodevelopmental outcomes of the studied patient population. Future research into this topic is recommended. Thirdly, CRO compliance is an important factor for the success of treatment in patients with skull deformities.[34] In this study, CRO compliance was assessed subjectively. However, this method of compliance measurement may be prone to overestimation and depend on whether the parents record the daily CRO wearing time or not. The objective compliance measurement method using temperature sensors combined with a data logger may provide more accurate information regarding daily wearing hours of the CRO. Further research needs to examine the links between CRO compliance and the improvement rate of skull deformities more precisely. Finally, a relatively small sample size in each group of craniosynostosis and short follow-up duration was another limitation of this study. Further investigation with longer follow-up duration and larger sample size in each craniosynostosis group should be undertaken to determine long-term results of treatment with MIS and post-operative CRO in patients with craniosynostosis.

5. Conclusion

Early endoscopic-assisted removal of the fused sutures accompanied by custom-made CRO treatment is an effective strategy to treat children diagnosed with single-suture craniosynostosis.

CRediT authorship contribution statement

Zahra Taheri: Conceptualization, Data curation, Investigation, Methodology, Writing – original draft, Writing – review & editing. **Taher Babaee:** Conceptualization, Formal analysis, Investigation, Methodology, Project administration, Supervision, Writing – original draft, Writing – review & editing. **Ehsan Moradi:** Conceptualization, Investigation, Methodology, Resources, Validation, Writing – review & editing. **Behnam Hajiaghaei:** Conceptualization, Methodology, Resources, Software, Writing – review & editing. **Hassan Reza Mohammadi:** Conceptualization, Methodology, Resources, Validation, 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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Abbreviations and Acronyms

MIS: minimally invasive suturectomy
CRO: cranial remolding orthosis
CI: cephalic index
CVAI: cranial vault asymmetry index
TV: total cranial volume
AV: anterior hemisphere cranial volume