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ORIGINAL ARTICLE

Modified endoscopic strip craniectomy technique for sagittal craniosynostosis: provides comparable results and avoids bony defects

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Abstract

Purpose This study describes a modified technique addressing bony defects and incomplete ossification after endoscopic strip craniectomy (ESC) for SC followed by postoperative helmet therapy (PHT). The study aims to delineate quantitative and qualitative outcomes of this modified ESC technique followed by PHT and discern the optimal duration of PHT following ESC. A secondary aim is to address the effects of the technique on bony defects.

Methods Patients undergoing ESC followed by PHT between 2017 and 2021 were included. Patient sex, age at surgery, duration of surgery, red blood cell transfusion, length of hospital stay, PHT duration, cephalic index (CI) at multiple time points, and bony defect information were collected. Descriptive and correlative analysis was done.

Results Thirty-one patients (25 male, 6 female) were operated in study period. Mean age at surgery was 12.81 weeks, mean duration of surgery was 57.50 min, average transfused RBC volume was 32 cc, mean length of hospital stay was 1.84 days, mean PHT duration was 33.16 weeks, and mean follow-up time was 63.42 weeks. Mean preoperative CI was 70.6, and mean CI at the end of PHT was significantly higher, being 77.1. Maximum improvement in CI (CI_{max}) took place at week 22.97. PHT duration did not have a correlation with CI at last follow up. There were no bony defects.

Conclusion Modified ESC technique is effective in successful correction of sagittal craniosynostosis. CI_{max} already takes place, while PHT is continuing, but there is no certain time point for dishelmeting. The technique avoided bony defects and incomplete ossification.

Keywords Sagittal craniosynostosis · Endoscopic strip craniectomy · Postoperative helmet therapy · Bony defect · Incomplete ossification · PHT duration

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Introduction

Craniosynostosis, defined as the premature closure of one or more cranial sutures, occurs in an estimated 3.1 to 6.4 per 1000 live births, 40% to 58% of which involve sagittal suture [1, 2]. Isolated sagittal craniosynostosis (SC) is reported to be the most common type of craniosynostosis (1 in 5000 children), accounting for 40–55% of all non-syndromic craniosynostosis cases with a strong male predilection [3]. SC leads to a characteristic head shape with an increase in anteroposterior diameter, biparietal narrowing, compensatory frontal, and occipital bossing together with ridging of sagittal suture, namely, scaphocephaly.

Surgery of SC aims to mitigate raised intracranial pressure, neurocognitive deficits, and neuropsychological effects and improve aesthetic form [4–7]. Various surgical strategies have been employed for SC ranging from suturectomies to cranial vault remodeling (CVR). Endoscopic strip craniectomy (ESC) followed by postoperative helmet therapy (PHT) has gained wider appeal over CVR owing to the brevity of surgery, decreased rate of blood loss and transfusion requirement, shorter hospital stays, and limited surgical scars [8–12]. Recently, ESC with PHT was reported to be an effective treatment in non-syndromic single suture SC with comparable head growth and aesthetic outcomes [13]. The overall goal of PHT is to augment the passive reshaping of the deformed cranium. It allows cranial expansion in recessed areas and compression in areas of overcompensated growth, a process driven by the rapid underlying brain growth in the first postnatal year [10, 11]. While there is a significant body of literature documenting the efficacy of PHT in achieving optimal head shape following ESC, there is still much obscurity when it comes to defining the optimal duration for PHT [9, 11, 12]. Setting these parameters is of multifaceted benefits such as alleviating the financial burden as well as revamping the quality of life and morbidities associated with manufacturing and wearing these helmets.

There is little information regarding bony defects following ESC for SC, although revision surgeries have been reported [14, 15]. There are inconsistencies about the terminology for incomplete ossifications, and little detail is reported about the events observed. Underdocumentation of the subject is argued [16]. It is obvious that incomplete ossification must be addressed when treating SC, considering the fact that revision surgeries may be needed and family concerns must be taken into account.

The aim of this study is to delineate the quantitative outcomes of ESC with PHT for SC using the described technique and discern the optimal duration of PHT following ESC required to realize optimal aesthetics. A secondary aim is to address the effects of the technique on bony defects and incomplete ossification.

Materials

Patient selection

Patients with non-syndromic single suture sagittal craniosynostosis, operated at Marmara University Department of Neurosurgery with the described technique, were selected. A total of 31 consecutive cases from January 2017 to January 2021 were included. Inclusion criteria for this study consisted of the following: (1) operated under the age of 6 months, (2) diagnosed preoperatively with computed tomography (CT), (3) operated with described technique, (4) obtained 3 dimensional imaging with STARscanner™ laser acquisition system pre or early postoperative, (5) PHT was ended at the time of the study, and (6) followed up with STARscanner™ laser acquisition system until PHT was ended or beyond. Patient sex, age at surgery, duration of surgery, length of hospital stay, red blood cell transfusion, PHT duration, and cephalic indices (CI) at multiple time points were collected. This retrospective study is conducted under Marmara University Institutional Ethics Committee approval.

Surgical technique

Patients were positioned supine with head being lateral on a gel horseshoe headrest. After skin preparation and draping, a 4-cm-wide horizontal incision was made posterior to the anterior fontanel, and a midline burr hole is placed which is then widened 2 cm in each side. Craniectomy was completed up to the anterior fontanel using Kerrison Rongeurs. The second 4-cm-wide horizontal incision was made anterior to lambda, and craniectomy was carried out to lambdoid sutures in a similar fashion. Under endoscopic assistance, a 4-cm-wide strip craniectomy was accomplished in the remaining mid-section, and meticulous hemostasis was done. Barrel staves were not used.

The excised bone was then thinned to multiple 1-cm-wide strips and replaced in two pieces over sagittal sinus from anterior fontanel to lambda (Fig. 1). The bone graft was always in two pieces, and their total length depended on the length from bregma to lambda on each patient individually. The grafts were bended to accommodate the curvature of cranial vault, and greenstick fractures were used when necessary. Tissue adhesives were used to reliably secure replaced bones in place and help hemostasis. Skin incisions were closed in anatomic fashion. No drains were used.

Anthropometric measurement

CI was used as anthropometric measurement. It is calculated as biparietal diameter divided by anteroposterior diameter, multiplied by 100. All CI measurements were collected using 3-dimensional imaging with STARscanner™ laser acquisition system, while CT was avoided. The acquired 3D image was sectioned by a software into 10 equally distant parallel cross sections, with Sect. 0 crossing the sellion and both tragions, and Sect. 10 passing through vertex. Then, Sect. 3 measurements were used to calculate CI [17] (Fig. 2). CI measurements were taken every 2 weeks whenever possible throughout the PHT period. These serial measurements allowed us to determine the CI at various time points, including preoperatively (CI_{pre}), when maximum improvement in CI was achieved (CI_{max}), when PHT was discontinued ($CI_{post\ PHT}$), and at last follow-up (CI_{final}).

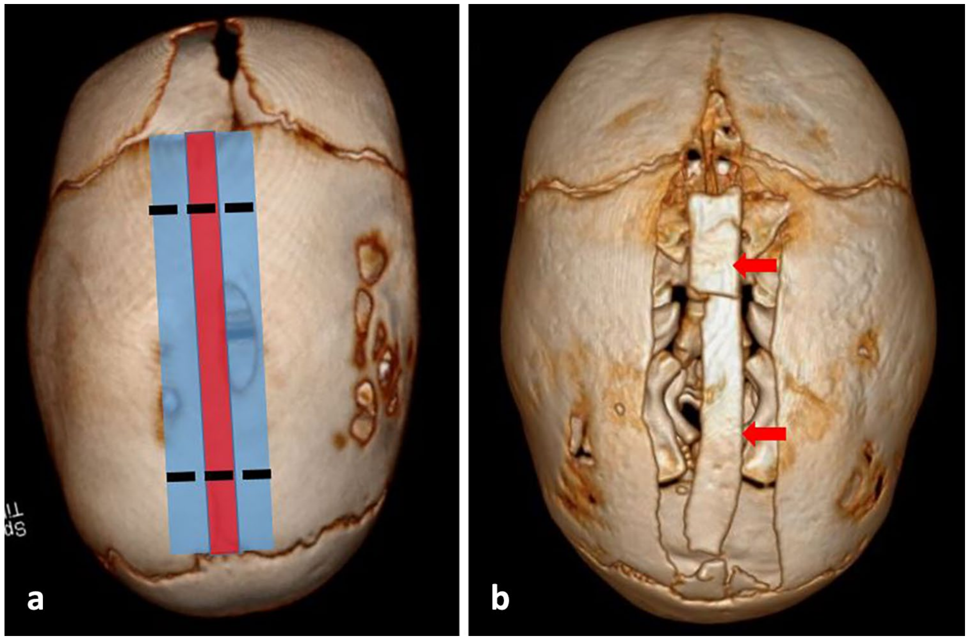

Fig. 1 **a** Depiction of the surgical technique (blue, craniectomy site; red, replaced bone strip; black dashed lines, incisions). **b** Final composition on post-operative CT (red arrows, replaced bone strip)

Helmet protocol

Postoperative helmet therapy was usually initiated in 2 weeks after surgery following regression of scalp swelling and wound healing. Custom made molding helmets were used to direct calvarial growth into a normal shape by limiting frontal and occipital poles to promote biparietal expansion. Helmets were worn 23 h a day and aimed to be used until age 1 during rapid growth of the head and brain, as described in the literature [11, 18]. During PHT course, 1 to 2 helmets were used depending on serial measurements and clinical course of the patient.

Follow-up

In the PHT period, outpatient clinic visits took place every 2 weeks whenever possible, intended for CI measurement. After PHT was ended, follow-ups took place at 6 months intervals. Neurological examination, head circumference, and finger palpation over ESC areas were the major concerns together with general appearance of head shape. Further anthropometric measurements were left to parents' will if no problems were encountered. CT was reserved for cases of strong suspicion of incomplete ossifications.

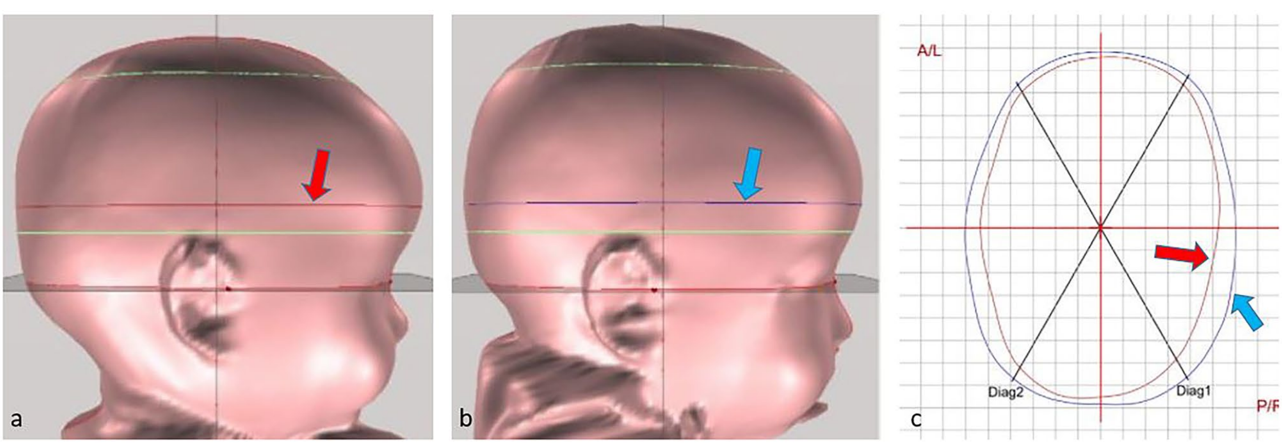

Fig. 2 A 3D photography with laser acquisition system in different time points. **a** Preoperative scanning (red arrow showing section 3). **b** Eighteenth week scanning (blue arrow showing section 3). **c** Calculated axial depictions (red arrow showing preoperative calculation representing section 3 in a, and blue arrow showing 18th week calculation representing section 3 in b)

Table 1 Demographics and cephalic indices following ESC and PHT in 31 patients

Variable	Value
Mean age at surgery ± SD (weeks)	12.81 ± 2.54
Mean duration of surgery ± SD (minutes)	57.50 ± 16.75
Mean blood transfused ± SD (cc)	32.00 ± 39.08
Mean hospital stay post ESC (days)	1.84 ± 0.93
Mean PHT duration ± SD (weeks)	33.16 ± 10.85
Mean follow-up duration ± SD (weeks)	63.42 ± 18.89
Mean $CI_{pre\ op}$ ± SD	70.6 ± 3.3
Mean CI_{max} ± SD	77.9 ± 3.4
Mean $CI_{post\ PHT}$ ± SD	77.1 ± 3.5
Mean CI_{final} ± SD	76.0 ± 3.6

Statistical analysis

Statistical analysis was performed using SPSS (version 22, IBM Corp.). Using standard descriptive statistics, CI values at discrete time points were analyzed. CIs were compared using the paired *t*-test. Normally distributed data were analyzed for correlation between several variables using the Pearson correlation test.

Results

Patient population

A total of 31 patients (25 male, 6 female) satisfied the eligibility criteria. The mean age at surgery was 12.81 ± 2.54 weeks (range 8 to 21 weeks). The mean duration of surgery was 57.50 ± 16.75 min (range 30 to 95 min). The average overall transfused red blood cell (RBC) volume was 32.00 ± 39.08 cc (range 0 to 180 cc), and 19 patients (61.3%) required intra-operative blood transfusion. Postoperative helmet therapy (PHT) lasted an average of 33.16 ± 10.85 weeks (Table 1). There were two complications. One patient needed simple scar revision shortly after ESC. The other patient had a dural injury, which was repaired primarily during the operation, needed a scar revision, and a through dural repair in two other surgeries.

Cranial anthropometric outcome

Comparison of our CI_{pre} to gender- and age-specific normative cephalic index values revealed that all but 3 male patients (88%) and all female patients (100%) had CI_{pre} below the minimum normal values as defined by Haas [19]. Those 3 male patients had CI_{pre} within the normal range. Postoperatively and after completing their PHT course, all patients had a substantial increase in CI, which falls in the normal range for their age and gender.

Overall, the mean cephalic index was 70.6 ± 3.31 (range 63.7 to 77.6), 77.1 ± 3.59 (range 71.1 to 85.8), and 76.0 ± 3.66 at CI_{pre}, $CI_{post\ PHT}$, and CI_{final} time points, respectively. CI_{max} took place at week 22.97 ± 9.17 postoperatively, with a mean CI_{max} of 77.9 ± 3.38 (range 72.6 to 86.3) (Table 1 and Fig. 3). $CI_{post\ PHT}$ and CI_{final} time points were 33 weeks and 63 weeks postoperatively, respectively (Fig. 3).

Paired sample *t*-test showed that the 6.5% mean increase in CI from CI_{pre} to $CI_{post\ PHT}$ and the 5.4% mean increase in CI from CI_{pre} to CI_{final} were both statistically significant

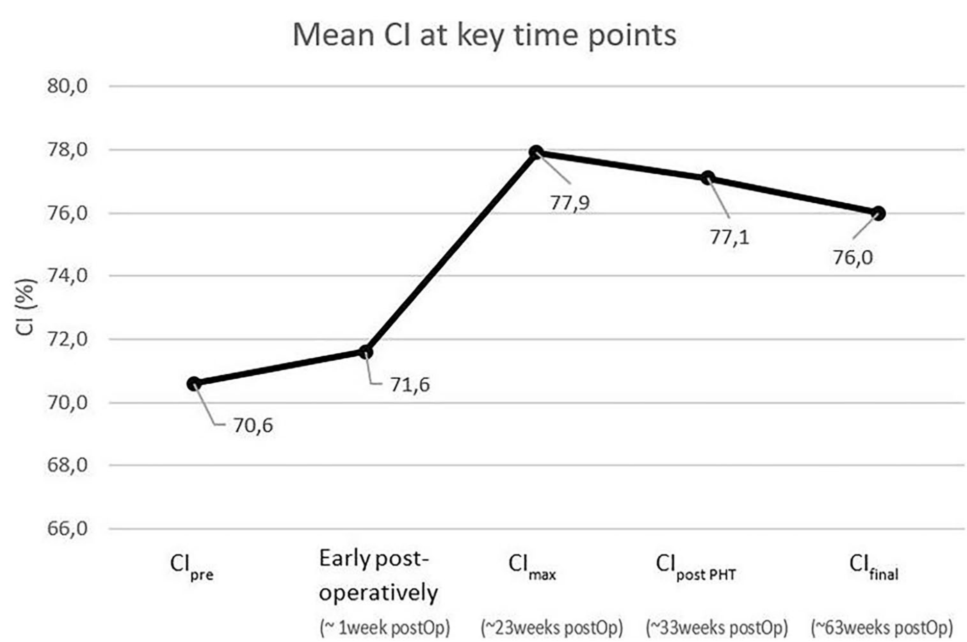

Fig. 3 Line graph of mean CI values measured at key time points for 31 patients (CI_{max} around 23 weeks postoperatively, $CI_{post\ PHT}$ around 33 weeks postoperatively which also indicates PHT duration and CI_{final} around 63 weeks postoperatively which also indicates mean last follow-up)

(both; $p < 0.001$). Moreover, an average regression of 0.8% was observed between CI_{max} and CI_{PHT}. Regression in CI continued in the time frame between PHT discontinuation ($CI_{post\ PHT}$) until last follow-up (CI_{final}) with an average CI decline of 1.1%.

No patients were found to have postoperative cephalic index values equal to or below the preoperative measurements.

Statistical analysis

Several statistically significant correlations where identified using the Pearson correlation test, most notably a correlation between CI_{pre} on one side and all from CI_{max} (p value < 0.001; $R = 0.707$), $CI_{post\ PHT}$ (p value < 0.00001; $R = 0.723$), and time to reach CI_{max} (p value = 0.013; $R = -0.497$). CI_{pre} was not correlated with the change from CI_{pre} to $CI_{post\ PHT}$. There was no correlation between PHT duration and the change from CI_{pre} to $CI_{post\ PHT}$. Likewise, no correlation was seen between the age at surgery and any of the above-mentioned variables (Table 2).

A negative correlation was detected between the chronology of surgery date and the corresponding intra-operative blood transfusion volume ($p = 0.012$; $R = -0.454$).

Furthermore, there were no significant inter-gender differences with regard to CI_{pre} ($p = 0.949$), CI_{max} ($p = 0.495$), $CI_{post\ PHT}$ ($p = 0.765$), and the CI change from CI_{pre} to $CI_{post\ PHT}$ ($p = 0.400$).

Follow-up

In a cohort of the first 22 patients with at least 2 years follow-up, there were no significant bony defects or incomplete ossifications by finger palpation. Patients with lesser follow-up times did not show signs of incomplete ossification, either. No revision surgeries were needed in any patient for cosmesis; no CT was required.

Discussion

Surgical treatment of SC includes a wide range of surgical techniques from open cranial vault remodeling to endoscopic suturectomy. Both aim to achieve multiple benefits including prevention of raised intracranial pressure and neurocognitive and neuropsychological deficits and allow for a better aesthetic form [4–6]. Endoscopic techniques have gained wide acceptance after being invented by Jimenez et al. in the late 1990s, since they have the advantage of shorter hospital stay, shorter surgery duration, less blood loss, and decreased morbidity and mortality [8–11, 20, 21].

ESC with PHT is reported to have comparable results with cranial vault remodeling. In two different series including different techniques, an increase in CI from 65.0 to 76.4 with reverse total calvarial reconstruction and from 67.9 to 78.5 with open calvarial vault remodeling is reported [22, 23]. Jimenez et al. reported 87% success rate (CI > 75) with 139 patients having mean preoperative CI of 67, with 12 months PHT duration [11]. Although having shorter PHT durations, Iyer et al. reported a CI increase from 68 to 78, and Ridgway et al. reported a CI increase from 69 to 76 [9, 12]. Studies comparing ESC with cranial vault remodeling have also found comparable CI increases between these techniques both retrospectively and prospectively [13, 24]. These data are similar to our experience with an improvement from CI_{pre} of 70.6, to CI_{max} of 77.8, $CI_{post\ PHT}$ of 77.1 and CI_{final} of 76.0. Mean PHT duration was 33.16 weeks. Our results show that ESC with replaced bone strip over sagittal sinus is an effective surgical technique which provides CI correction comparable to various other techniques.

Table 2 Correlation matrix

	Age at Sx	CI_{pre}	CI_{max}	$CI_{post\ PHT}$	CI_{final}	Time to CI_{max}	PHT duration	Δ CI_{pre} to $CI_{post\ PHT}$
Age at Sx	1							
CI_{pre}	.084	1						
CI_{max}	.065	.707**	1					
$CI_{post\ PHT}$	.021	.723**	.972**	1				
CI_{final}	.191	.664	.894**	.942**	1			
Time to CI_{max}	–.247	–.497*	–.412*	–.312	–.102	1		
PHT duration	.019	–.219	–.401*	–.442*	–.219	.446*	1	
Δ CI_{pre} to $CI_{post\ PHT}$	–.127	–.206	.517**	.527**	.762*	–.047	–.386	1

Sx = Surgery, Δ = Change

*Correlation is significant at the 0.05 level (2-tailed); **Correlation is significant at the 0.01 level (2-tailed)

While the effectiveness of ESC with PHT is now well recognized, there is still much debate on the optimal duration of a PHT course. Early reports advocated a PHT course of 12 months or up to age 1, probably taking into account that rapid brain and calvarium growth takes place in the first year of life, which makes sense [11, 12]. Number of helmets during PHT course differed among reports from single helmets to 2 to 4 for a single patient [9, 11, 14, 25]. Considering the high-tech materials used, final costs of these custom molded helmets restrict availability of such an approach for most families, especially in low-income countries. Efforts up to now failed to demonstrate a clear picture for optimal duration of PHT, but a regression after reaching a CI_{max} is strongly pronounced [9, 12, 25]. Avoiding radiation exposure with 3-dimensional laser acquisition systems, serial measurement became available and revealed that CI_{max} is already achieved while PHT course is still continuing. Iyer et al. found that the average maximum CI is reached at 8.4 months of PHT. Furthermore, they described two discrete subgroups reaching a CI_{max} at around 6 months and 11 months [9]. Therefore, they suggested that helmeting beyond CI_{max} did not improve final anthropometrics. They also found that there was no difference between their subgroups regarding the regression once CI_{max} has been reached [9]. Pickersgill et al. also reported that the so-called aCI (adjusted CI, taking into account the ideal euryon locations instead of actual biparietal width) reached its maximum within the first 6 months of surgery, which then began to decline. There was no discussion about the optimal duration of PHT [25]. Our results indicate similar findings with Iyer et al. [9] with a mean CI_{max} of 77.9 at around 23 weeks postoperatively. However, there were no correlation between PHT duration and all of CI_{max}, $CI_{post\ PHT}$, and delta change in CI_{pre} to $CI_{post\ PHT}$. A consistent regression was observed thereafter to the end of PHT and beyond. The average regression from CI_{max} to $CI_{post\ PHT}$ was 0.8%, and average regression from $CI_{post\ PHT}$ to CI_{final} was 1.1%. These results are in line with the ranges reported in the literature [9, 12, 25]. Regression in CI is not only seen after ESC; it is also reported after CVR for SC. And with these findings, it is now obvious that regression in CI is not initiated after dishelmeting. Although there is evidence that CI_{max} is reached somewhere around 6 to 8 months, it is not clear that helmeting after reaching CI_{max} is necessary or discontinuing will have adverse effects on the final outcome. Prospective randomized studies are needed to compare the effects of duration of PHT and set an optimum.

Bony defect or incomplete ossification after ESC is another topic with less exposure, although it may affect aesthetic outcome and increase the risk of brain injury. Bony defects or incomplete ossification has been well discussed following extensive surgeries or with patients operated after 1 year of age [26]. On the other hand, there are few reports of incomplete ossification following ESC techniques. In a long-term follow-up study, 14 out of 32 (44%) patients were found to have over 4 cm^2 calvarial defects, 3 of which (9.4%) required reoperation [14]. They also observed that incomplete ossifications were commonly located over strip craniectomy areas rather than lateral edges and speculated that wide vertex craniectomy was the cause. In another quantitative study, Thenier-Villa et al. reported a reoperation rate as 5.6% in a mix cohort including patients with open surgeries and endoscope-assisted procedures [15]. In their report, after studying different configurations, they hypothesized that the maximum distance between the bone plates perpendicular to the major defect plays a substantial role; the wider the defect, the lesser the ossification. Senior author's (A.D.) previous personal experience led us to modify our ESC technique by replacing a 1-cm-wide autologous bone strip over the superior sagittal sinus. This method not only reduces the maximum distance between bone plates, but it may also serve as a solid protection cover over the superior sagittal sinus. After completing the PHT course, we scheduled follow-up visits with 6-month intervals, which included finger palpation and family perception. Regarding incomplete ossification, CT was reserved for significant suspicion. We identified a cohort of 22 patients having at least 2 years of follow-up. With a mean follow-up of 35.95 months (range 24 to 52 months) in this cohort, we did not have any patients with high degree suspicion of incomplete ossification; hence, no CT was required. In two well-documented long-term follow-up series, 9.4% and 5.6% reoperation rate was reported in the literature by Persad et al. and Thenier-Villa et al. respectively [14, 15]. Our modified technique resulted in no reoperation in the provided follow-up of 35.95 months. This modified technique addresses the problems speculated as the cause of incomplete ossification by these articles, including maximum defect area and maximum distance between the bone plates perpendicular to major defect. Although no patients in this group had revisions and even did not necessitate CT imaging, long-term follow-up is still recommended for claiming persistent results. Another issue, which can be regarded as a limitation for this study, is secondary stenosis. Reducing the size of the created defect may seem as a possible theoretical cause for reducing the effectiveness of the surgery at first sight. On the other hand, there is a wide variety of ESC techniques presented in the literature as effective surgeries, which described craniectomies ranging between 2 and 6-cm wide. Optimal craniectomy width has not been set yet. Although we did not face any sign of secondary stenosis in any patients up to now, longer-term results have to be waited to draw a clear picture about the issue.

Conclusion

ESC with PHT has wide acceptance as a surgical treatment option for single suture non-syndromic SC. Replacing a narrow strip over superior sagittal sinus does not hinder the results. It is effective in correcting CI values and provides comparable outcomes to different ESC techniques as well as CVR.. CI_{max} takes place at around 6 months after ESC with PHT, and regression in CI starts, while PHT is ongoing. The duration of PHT is not correlated with all of CI_{max}, $CI_{post\ PHT}$, and CI_{final}. These results may indicate that longer PHT courses might be unnecessary, howbeit a certain time point is not evident. Future prospective randomized trials are needed to clarify the optimum duration of PHT. The described modified technique may play a role in reducing incomplete ossification rates and revisions, although long-term follow-up is needed.

Declarations

Ethics approval Approval was obtained from the Ethics Committee of Marmara University. The procedures used in this study adhere to the tenets of the Declaration of Helsinki.

Consent to participate Not applicable.

Consent of publication Not applicable.

Conflict of interest The authors declare no competing interests.

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