Original Article

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# An Overcorrective Modification of Cranial Vault Reconstruction for Non-Syndromic Metopic Suture Synostosis: Shiraz Technique

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**Introduction:** Growth restriction of frontal bones due to premature closure of metopic suture leads to the "Trigonon" shape of the skull, introduced by Welcker in 1862. Paul Tessier introduced cranial vault reconstruction (CVR), a common craniofacial procedure for treating this condition. The present study aims to introduce a modified version of this procedure.

**Methods:** In this study, we present our experience with a modified version of CVR, named the Shiraz technique, used as a single-stage overcorrective reconstructive method for isolated metopic suture synostosis with limited unique threesome osteotomies. We also report 35 cases treated with this technique.

**Results:** The patient population comprised 26 males (74.3%) and 9 females (25.7%) with a mean age of 8.4 months (range: 3-13). The mean duration of the operation and anesthesia was 175.0 minutes (range: 60-300). The mean intraoperative bleeding and blood transfusion volumes were 95.6 ml (range: 20-250) and 75.3 ml (range: 0-320), respectively. The main complications during the follow-up were epidural hematoma (n=3, 8.6%; one case needed reoperation), temporal hollowing (n=2, 5.7%), raised intracranial pressure (n=2, 5.7%), wound infection (n=2, 5.7%), and mortality in one of the cases, which was presumed to be due to air emboli.

**Conclusions:** The Shiraz technique, as a novel modified version of CVR, is a safe and effective technique for the treatment of non-syndromic metopic suture synostosis with low mean intraoperative blood loss and transfusion. In the long-term follow-up, the occurrence of temporal hollowing was decreased, and the outcome showed a smooth anterior skull base.

Keywords: craniosynostosis, Trigonocephaly, Cranial vault reconstruction, Metopic suture

## INTRODUCTION

Growth restriction of frontal bones due to premature closure of metopic suture leads to a keel-shaped forehead, which was named trigonocephaly by Welcker in 1862 (1-4). Hypotelorism, ethmoidal hypoplasia, and bitemporal shortening are the most concerning features usually encountered in trigonocephaly. The severity of the accompanied malformations can vary considerably in a case-based fashion. The male to female ratio of incidence in metopic suture synostosis is estimated to be between 2:1 and 6:5 (1, 2, 5-8). However, the etiology is virtually unknown and beyond the scope of this paper. For the treatment of this condition, various techniques have been suggested based on distraction osteogenesis, a modification of the classic "floating forehead technique" by Marchac (1, 7, 9).

In this study, we report our experience with a modified version of cranial vault reconstruction (CVR), a single-stage overcorrective reconstructive method, for rectifying non-syndromic metopic suture synostosis. This novel technique utilizes unique threesome osteotomies to reduce intra- and postoperative blood loss and mean operative time while shaping a smooth anterior skull base by leaving less dead bone space. Moreover, we report 35 cases treated with this technique in our center.

## MATERIALS AND METHODS

### *Study design*

Our study outlines an overcorrective technique for non-syndromic metopic suture synostosis developed in the Department of Pediatric Neurosurgery in Shiraz, a referral center in the south of Iran, as a single-stage CVR using a technique named the "Shiraz technique". We retrospectively included cases operated between August 2015 and August 2019 with this technique. Patients with multiple suture closures were excluded from the study.

### *Surgical technique*

In our technique, we start the operation with a bicoronal skin incision. Also, we favor the galeal sparing strategy



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leaving the galea attached to the bone when the skin flap is rotated. A bifrontal craniotomy beginning 1 cm superior to the superior orbital rim and extending but not reaching the coronal sutures is sketched.

Two burr holes are placed off the midline anterior to the coronal suture and just above the temporal area. Thereafter, using a craniotomy high-speed drill (πdrive® Motor, Stryker), we make two triangular bone flaps (labeled 1 and 3 in Figures 1 and 2) and one trapezoid midline bone segment (labeled 2 in Figures 1 and 2) overlaying the sagittal sinus (Figure 2). After the bone flap removal, the dura is separated from the anterior and middle fossa in the epidural plane. Then, the supraorbital bar is osteotomized; and, orbital roof osteotomy and mobilization of the supraorbital bar are performed.

In the next step, the trapezoid bone particle (labeled 2 in Figures 1 and 2) is divided into two pieces. These two pieces are reshaped using a high-speed diamond drill. The resulting two pieces are then inserted bilaterally and lateral to the supraorbital bar in order to overcorrect the frontal angle. Regarding the fronto-orbital advancement, the supraorbital bar is overcorrected using multiple mini-screws and plates (it is desired to use the absorbable screws; however, in our practice, the non-absorbable ones are used due to financial strains) (Figure 3). We advocate that an overcorrective strategy should be followed during the entire operation.

Afterward, one of the triangular bony particles (labeled 1 in Figures 1 and 2) is placed into the middle of the forehead. Besides, the other triangular bony particle (labeled 3 in Figures 1 and 2) is halved, rotated, and inset into both sides of the first triangular bone flap forming a smooth forehead. Also, an extra bandeau is made in some of the cases, based on the surgeon's decision (labeled 4 in Figure 1). The extra bandeau is needed when some empty spaces are remained, which should be filled. This piece is reshaped and cut into pieces in order to fill those spaces. In Figure 1, the remaining segment of the bandeau is inset into its initial location. Figure 4 demonstrates the preoperative and postoperative 3D skull computed tomography (CT) scans of a patient treated with the Shiraz technique in our center.

When discharging the patients, the parents are instructed to expect postoperative edema of the face, which will subside as time passes. Also, wearing a child-friendly helmet is suggested – it is reported that helmet can be helpful to modify the skull growth in all dimensions (10); however, further studies evaluating the efficiency of helmet therapy after CVR is needed.

### *Follow-up*

Consultation with a pediatric plastic surgeon has been carried out for further management of the patients. We have followed our cases with a 3D skull CT scan as they grow up (6 and 12 months following the operation) (Figure 5).

Ethical approval

Written informed consent for participation and publication was obtained from the patients' parents/guardians prior to the operation.

## RESULTS

In our study, 35 patients, including 26 males (74.3%) and 9 females (25.7%) with a mean age of 8.4 months (range: 3-13), were recruited. No family history of craniosynostosis was reported in the patients, only one had a history of cardiovascular comorbidity. The mean hospital stay was 7.5 days (range: 2-60). Also, the mean duration of the operation and anesthesia were respectively 175.0 minutes (range: 60-300) and 240.6 minutes (range: 105-420). The mean intraoperative bleeding was 95.6 ml (range: 20-250). Blood transfusion was required in 31 cases (mean=75.3 ml; range: 0-320) (Table 1).

**Table 1-** Operation details

| Variable | Mean (Range) |
|---|---|
| Age at operation (months) | 8.4 (3-13) |
| ICU admission duration (days) | 2.1 (1-14) |
| Hospital stay (days) | 7.5 (2-60) |
| Duration of surgery (minutes) | 175.0 (60-300) |
| Duration of anesthesia (minutes) | 240.6 (105-420) |
| Intraoperative bleeding (ml) | 95.6 (20-250) |
| Blood transfusion (ml) | 75.3 (0-320) |

ICU: intensive care unit

Three cases (8.6%) presented epidural hematoma, one of them needed reoperation. In the follow-up, two patients (5.7%) presented temporal hollowing. In two cases (5.7%), post-operative development of increased intracranial pressure necessitated the insertion of a ventriculoperitoneal shunt. These two individuals had bilateral plagiocephaly. One patient (2.9%) was expired after the surgery due to air emboli, and two (5.7%) developed postoperative wound infection, which was completely resolved after an antibiotic therapy course.

## DISCUSSION

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There are various modifications, based on distraction osteogenesis, in the literature regarding the treatment of metopic synostosis. Marchac's "floating forehead technique" contemplated the distraction osteogenesis for CVR (1). Nonetheless, Paul Tessier made aesthetic aspects of what is now called CVR more vivid (1). Besides, David et al. advocated that there is no outstanding merit in utilizing the endoscopic assisted minimally invasive techniques in the management of metopic synostosis. The reason is the expansion of a tangled web between the brain parenchyma, the dura mater, and the bony skull base in the expansion of the cranial vault (11); however, some other studies have reported different conclusions regarding this issue (12, 13). Herein, we introduced a novel modified version of CVR, named the Shiraz technique, and reported the experience of using this procedure in our center. For the Shiraz technique, we prefer to proceed with the operation when the patient`s age is between 6 and 9 months old. In our method, utilization of threesome osteotomies results in the reduction of intra- and postoperative blood loss and mean operative time; also, the aesthetic outcome was acceptable by shaping a smoother anterior skull base with leaving less dead bone space. The nature of the overcorrective attitude in our technique would provide a higher probability to correct the frontal angle and trigonocephalic feature of the skull in the long term. The great advantage of this technique is its lower chance of developing temporal hollowing. Moreover, two cases (5.7%) experienced raised intracranial pressure following the surgery, which is comparable to previous studies.

Mean bleeding volume was 95.6 ml (range: 20-250) in our 35 cases, which is remarkable. The intraoperative blood loss volume is much higher in other studies; mean volume of 458.3 ml in the hinge technique by Magoon et al. (14) and median volume of 220ml and less than 255 respectively in studies by Kelleher et al. (15) and Engel et al. (16). Subsequently, the blood loss volume and, as a result, the needed transfusion volume were significantly lower in our method. Besides, the mean operation duration of 175 minutes (range: 60-300) is comparable to other techniques; a mean of 183.4 minutes in the cranial orbital buttress technique by Seal et al. (17) and 159 minutes in the hinge technique by Magoon et al. (14).

Although there are studies in the literature concluding that untreated craniosynostosis may lead to raised intracranial pressure, a few studies have been reported regarding the rise in intracranial pressure after the surgical treatment of craniosynostosis (18, 19). A systematic review by Christian et al. in 2015 reported that intracranial hypertension occurs following the operation with incidences of 5% in sagittal synostosis and 4% in all forms of non-syndromic craniosynostosis. Also, they mentioned that the exact incidence should be further evaluated because of the possibility of underreporting (20). Among our cases, two (5.7%) complicated postoperative raised intracranial pressure, which needed ventriculoperitoneal shunt insertion. Further studies should be obtained concerning this complication.

Natghian et al. looked into the Oxford practical knowledge in isolated metopic synostosis over a period of 22 years. They concluded that temporal hollowing and forehead outline defects were confronted more frequently than desired with an incidence of 17.8% as a late postoperative complication (21). There are certain downsides to our technique, such as bone breaches to be filled by future bone growth with a reasonable risk of bone defect in the long term. However, the strong point of this technique is its lower chance of developing temporal hollowing (n=2, 5.7%). This statement is mainly due to less manipulation in the temporalis muscle region and a subsequent lower chance of temporalis muscle atrophy (Figure 6). Besides, it is worth mentioning that the judgment for the presence of temporal hollowing is subjective leading to underestimation or overestimation of its diagnosis. Table 2 demonstrates the advantages and disadvantages of the Shiraz technique.

**Table 2**- The advantages and disadvantages of our modification of cranial vault reconstruction, named Shiraz technique

| Advantages | Disadvantages |
|---|---|
| Low blood loss volume | Risk of bone defect in the long-term |
| Relatively low operative duration | Risk of postoperative hematoma formation in the vicinity of frontal dead spaces |
| Low risk of temporal hollowing | |
| Smooth anterior skull base | |

There was mortality in our case series presumed to be due to air emboli; however, we do not know the exact explanation and pathophysiology for the occurrence of air emboli in this case. Generally, venous air embolism is reported to be more common in pediatric neurosurgery operations, especially when the head is above the heart during the operation (22). Besides, the occurrence of air emboli has been reported as a complication after the treatment of isolated metopic synostosis (21). So, we cannot justify whether the air emboli in that case correlated with our surgical technique. On the other hand, we leave the galea intact and attached to the skull during the primary dissection of the skin. This may help reducing the chance of air emboli formation.

We avoid putting a drainage tube at the end of the operation because we believe it may induce a negative pressure over the bare dura matter, which may trigger potentially fatal bradycardia via the trigeminal nerve (trigeminal dural reflex) (23).

Limitations of the present study

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The sample size was small, and larger studies can be performed. Also, longer follow-up can be done, reporting the aesthetic outcomes of the patients. The purpose of this study was to introduce our novel modification and to present the cases treated with this method; however, we did not compare our results to the results of other techniques performed in our center. So, we suggest multicentric studies, especially clinical trials with long-term follow-up, regarding the aesthetic features, clinical outcomes, and complications of our method in comparison to other common surgical methods for trigonocephaly.

## CONCLUSION

In conclusion, our novel modified version of CVR, the Shiraz technique, is a safe and effective technique for the treatment of non-syndromic metopic suture synostosis. The mean intraoperative blood loss and blood transfusion were significantly low in our method. Also, we demonstrated a relatively low operative duration. The occurrence of temporal hollowing was decreased, and the outcome showed a smooth anterior skull base. We reported 35 cases who underwent this technique; however, multicentric evaluations should be obtained to compare the clinical outcomes and complications of this technique with other common techniques for this condition.

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## DISCLOSURES

### *Ethical approval*

Written formed consent was obtained from the patients prior to the surgery. Also, written consent formed was obtained from the patients regarding the publication of this study. The present study was approved by the Medical Ethics Committee of Shiraz University of Medical Sciences (Ethics code: IR.SUMS.MED.REC.1398.202).

### *Consent to participate*

The patients gave consent to use their information and images for research purposes. *Consent for publication*

The patient gave consent to use his information and images for publication.

### *Conflict of interest*

The authors declare no conflicts of interest with respect to the content, authorship, and/or publication of this article.

The authors report no conflict of interest

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## CONTRIBUTIONS

-**Mohammad Sadegh Masoudi:** Conceptualization, Formal Analysis, Investigation, Methodology, Validation, Visualization, Writing – original draft, Writing – review & editing
-**Ali Ansari**: Writing – original draft, Writing – review & editing
-**Sanaz Taherpour**: Writing – original draft, Writing – review & editing
**-Reza Shahriarirad:** Writing – original draft, Writing – review & editing
**-Reza Taheri:** Data Gathering

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