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A novel surgical rotation overlapping craniotomy technique for the management of non-syndromic anterior plagiocephaly

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Abstract

Background: Nonsyndromic anterior plagiocephaly is one of the most common types of craniosynostosis. Different surgical techniques to correct this deformity have been developed with dissatisfaction among many surgeons. In this study, we describe a novel surgical technique to manage this pathology. The inclusion criteria were patients presenting with non-syndromic anterior plagiocephaly below 1 year of age presenting to the Pediatric Hospital in the period between 2016 and 2019. Surgical time, blood loss, and complications were recorded. The follow-up period was at least 1 year postoperative, and cosmetic outcome satisfactory categories were reported.

Results: Seven patients were included in this study. No intraoperative complications were reported, and no blood replacement was needed in any of the patients. The parents of six patients were completely satisfied (85.7%) with the outcome and partially satisfied in 1 patient (14.3%).

Conclusion: The results of the described rotational overlapping flap technique are promising and can be considered one of the minimally invasive techniques for the correction of this pathology.

Keywords: Craniosynostosis, Anterior plagiocephaly, Pediatric

Background

Craniosynostosis is the premature fusion of one or more cranial sutures that develop an irregular head shape. It occurs approximately at 1 to 1.6 in 1000 live births [1]. Non-syndromic craniosynostosis is an isolated disorder with no genetic syndromes associated with it. Anterior plagiocephaly (AP) is a general term that denotes the unilateral flattening of the cranium's anterior quarter and is caused by unilateral coronal synostosis (UCS) [2]. The most common types of craniosynostosis after sagittal synostosis are UCS and metopic synostosis. UCS occurs in 1 out of 10,000 live births [3, 4]. The reported ratio of female to male is 68% [2].

Premature coronal suture fusion combined with the rapidly expanding infant brain results in the typical AP morphology [5]. UCS produces restriction of regional growth and compensatory expansion of adjacent regions with an evident fronto-orbital dysmorphology [2].

Different techniques for craniosynostosis have been developed since the 1960s, such as fronto-parietal suturectomy, lateral canthal advancement, and bilateral fronto-orbital advancement (FOA) [2]. Surgical intervention is targeted to correct the asymmetrical forehead and supraorbital region, with bone advancement on the affected side. This procedure is preferably performed before the patient reaches 1 year of age [6].

Most literature available on the surgical management of this pathology includes retrospective studies and case series. Consequently, there is a wide discrepancy in the opinions for the best treatment of non-syndromic AP and dissatisfaction with the existing surgical procedures among many surgeons [5].

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In this study, we describe a surgical technique to manage this pathology and the outcome and complications of this procedure are reported.

Methods

This study was conducted on cases presenting with non-syndromic AP in our University Hospital in the period between 2016 and 2019 operated by our novel surgical rotational overlapping unilateral frontal craniotomy technique. All patients were diagnosed clinically. An initial CT brain and fundus examination were done to exclude the presence of associated increased intracranial pressure. The patients were operated on once the diagnosis was done. Inclusion criteria in this study were patients presenting with non-syndromic AP below 1 year of age.

The surgical technique was analyzed regarding surgical time, blood loss, complications, and postoperative hospital stay.

The patients were followed postoperatively for at least 1 year. Clinical assessment was done by examining the forehead contour to detect bony gaps and ridges in the follow-up period. Cosmetic outcome satisfaction of the parents was reported, which was divided into 3 categories (satisfied, partially satisfied, not satisfied, and willing to do a redo surgery) after comparing the preoperative and postoperative head photographs of the patients.

In surgical technique, a coronal skin incision on the affected side is done reaching the superior temporal line on the other side with dissection and elevation of a forehead myocutaneous flap. A wide anteriorly based pericranial flap is carefully dissected to prevent tears as it will be used during closure to cover and enhance bone regrowth in the defected areas.

Unilateral frontal craniotomy in the affected side is performed extending medially 1 cm from the midline, laterally to the squamous suture, posteriorly to the coronal suture, and anteriorly just above the supraorbital

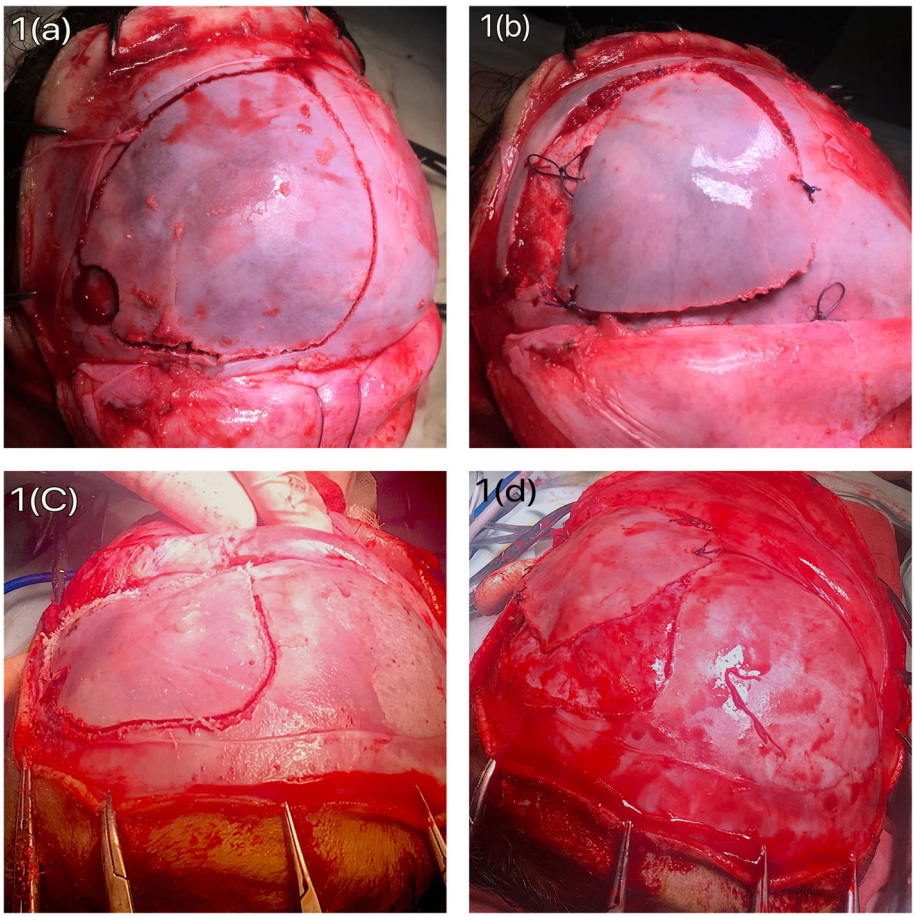

Fig. 1 **a**) case number 1 with right anterior plagiocephaly, intraoperative anterior view of a right frontal craniotomy with the initial burr hole placed over the squamous suture. **b**) Case number 1 with right Plagiocephaly, intraoperative anterior view photo after clockwise rotation and overlapping of the frontal craniotomy flap over the frontal bone and the supraorbital bar creating the new forehead contour. **c**) case number 2 with left anterior plagiocephaly, intraoperative posterior view photo of a designed left frontal craniotomy **d**) case number 2 with left anterior plagiocephaly intraoperative posterior view photo after the anticlockwise rotation of the craniotomy flap to correct the skull deformity

rim. Using a high speed drill, a single burr hole is done at the middle of the exposed squamous suture and the frontal craniotomy is followed (Fig. 1).

The craniotomy frontal bone flap is then rotated and replaced to create the new forehead using the more symmetrical curved postero-medial surface of the craniotomy flap. In order to do this, the flap is rotated clockwise for correction of right AP and anticlockwise for the left cases. The rotated frontal craniotomy flap is fixed overlapping the supraorbital bar anteriorly and the anterior part of the frontal bone medially to equalize the contralateral frontal bossing. The new positioned bone flap is fixed in place using non-absorbable silk sutures 3/0 through drill holes in both the craniotomy flap and the surrounding cranium (Fig. 1).

The preserved pericranial flap is then used to cover the whole craniotomy site including the areas with bone defect to enhance bony healing and growth. As the temporalis muscle was not detached from the skin, it is replaced in-block during the closure of the skin. A drain is left in place and the wound is closed in layers.

Results

Seven female cases with non-syndromic AP were surgically treated by our modified surgical correction technique in our department during the study period. The age of the cases ranged from 3 to 11 months; the mean age was 7 months.

Increased intracranial pressure was detected preoperatively in one case. She presented delayed milestones and silver-beaten appearance of the skull in the preoperative radiological imaging. The right AP was present in 4 cases (57.1%), and the left AP was present in 3 patients (42.9%). The decision for surgical correction was done once the condition was diagnosed, and the preoperative investigations were done.

Surgical time ranged from 83 to 110 min with a mean time of 95.3 min. No intraoperative complications were reported, and no blood replacement was needed in any of the patients. No postoperative intensive care unit (ICU) admission was needed in our cases. As scheduled preoperatively, all patients were discharged after 2 days.

Remodeling of the overlapped frontal bone was detected in the follow-up period. Within 3 months

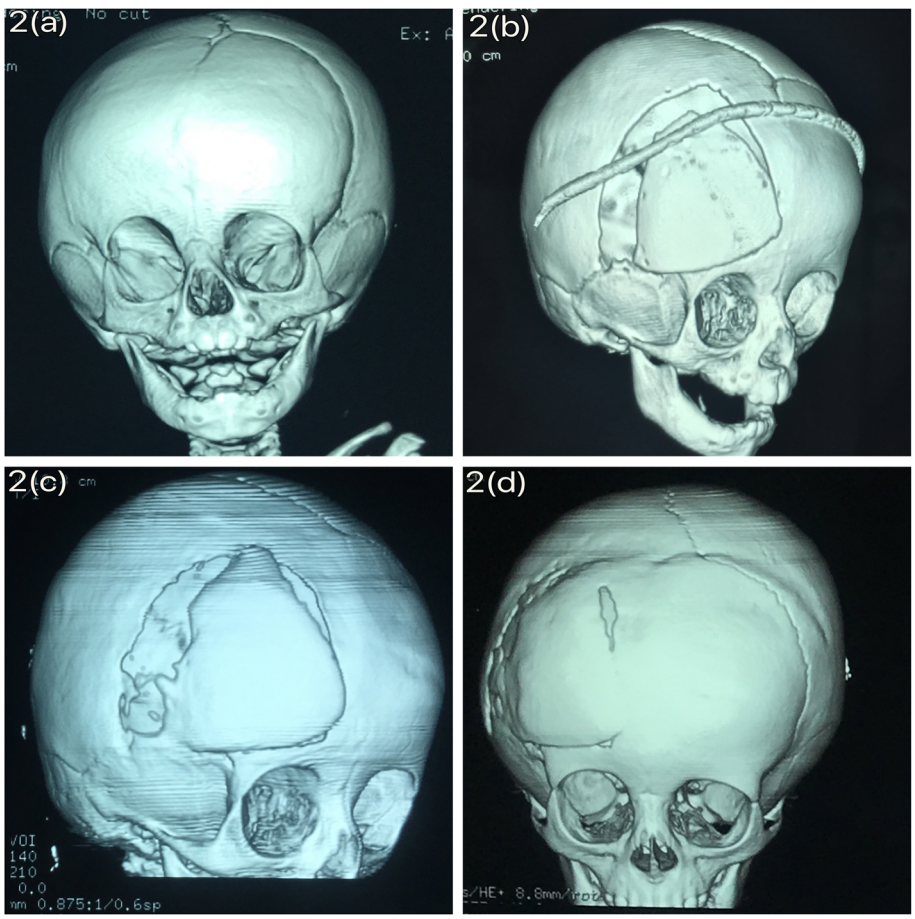

Fig. 2 **a**) 3D skull reconstruction CT showing right anterior Plagiocephaly **b**) immediate postoperative image showing the rotated overlapped frontal flap **c**) 3 months postoperative image **d**) 1-year postoperative image showing bone remodeling and bone regrowth in the defect

postoperatively, a smooth forehead contour, without any bony ridges, and a normal eyebrow appearance were achieved in all patients. Bone regrowth and filling of the forehead bony defects took an average of 1 year postoperatively. Temporal depression was not observed in the follow-up period in any of the study cases (Fig. 2).

Postoperative seizures were reported in one patient who presented preoperatively with increased intracranial pressure and delayed milestones. Antiepileptic drugs were prescribed for 1 year to control the postoperative seizures.

Clinical outcome

The follow-up period ranged from 12 to 44 months postoperatively (mean 27.4 months). The cosmetic outcome satisfaction reported by the parents was completely satisfied in 6 patients (85.7%) and partially satisfied in 1 patient (14.3%). No parents of any of the cases reported dissatisfaction with the cosmetic outcome and the need for a re-do surgery (Figs. 3 and 4).

Discussion

Over many years, different techniques have been developed to treat AP due to non-syndromic UCS, but the lack of quality level evidence supporting a technique's superiority over another has resulted in practical discrepancies. The lack of both evidence-based medicine and agreement between surgeons are regarded as the major obstacles in decision making in UCS management [5].

AP surgical repair has gone from strip craniectomy to the classic open FOA through a great advancement due to the inconsistent results and the reported high rate of recurrence of the strip craniectomy technique [7].

Despite global indications and implementation, all open FOA techniques have some disadvantages. It requires a long ear-to-ear scalp incision, blood transfusion necessity due to significant blood loss amount, and a prolonged hospital stay [7]. Several complications were reported in literature following open FOA techniques such as intracranial bleeding and hematomas formation, wound infections, dural tears, and cerebrospinal fluid leaks [8, 9]. Residual asymmetries, deformity relapses, and contour irregularities are also not uncommon and have been demonstrated in several studies [8–12].

The goals for efficient craniosynostosis treatment are adequate intracranial volume, sufficient for normal brain expansion, and to reduce the cognitive sequelae and obtain a normal cranium shape [13]. From this background, we innovated the rotational overlapping flap (ROF) technique for the correction of non-syndromic AP and reported its complications and outcome. We believe that our new technique has several similar advantages to the standard FOA, compared to minimal invasive techniques, regarding the immediate correction of the skull deformity and providing an adequate volume for the expanding brain. However, the ROF has several advantages over FOA in terms of simplicity, duration of surgery, the need for blood transfusion and postoperative hospital stay. It also has the advantage that the

Fig. 3 (**a** & **b**) Preoperative photographs of a 5 months old girl showing right anterior plagiocephaly. 3 (**c** & **d**) Photographs 1 months postoperative showing smooth forehead contour and symmetrical forehead appearance. 3 (**e** & **f**) Photographs of the patient 48 months postoperative showing normal forehead contour and normal orbital appearance with no temporal depression observed

Fig. 4 (**a** & **b**) Preoperative photographs of a 3 months old girl showing left anterior plagiocephaly. 4 (**c** & **d**) Immediate postoperative photographs. 4 (**e** & **f**) Photographs of the patient 48 months postoperative showing normal forehead contour and normal orbital appearance with no temporal depression observed

postoperative remodeling helmet is not needed as in several minimal invasive corrective techniques.

As for the ideal timing of the surgical management of craniosynostosis, interfering before the age of 1 year is usually recommended [14]. Most surgeons operate on the patient as soon as possible [13]. Earlier surgical correction benefits from the thinner and softer bones which are easier to remodel and the rapidly growing brain, thus minimizing consequent skull deformities and facial compensatory changes induced by brain growth. In older children, the bones become harder to remodel and lose the ossification properties and bone grafting is usually required [15]. This time window is crucial in our technique to obtain the best cosmetic results as it involves the overlap of the relatively thin bony edges to compensate for the deficient bone size, instead of the more aggressive bone advancement techniques.

Prolonged surgical time and anesthesia exposure have been related to several complications in pediatric patients. Naumann and colleagues [16] demonstrated inverse relations between neurodevelopment in children with craniosynostosis and the amount of anesthesia exposure. They found an average decline in developmental test scores from 1.1 to 2.9 for every 30-min increase in anesthesia duration. They concluded that this decline in neurodevelopmental scores may be attributed to the

exposure to anesthesia, unspecific surgery effects, or unmeasured variables that correlate with the duration of surgery.

The duration of surgery in our series ranged from 83 to 110 min with a mean time of 95.3 min. This duration was markedly shorter than the duration of surgery for open corrective procedures reported in the literature. Guzman and colleagues [17] reported a mean operative time of 210 min in their series, and a hospital stay of 4.5 days compared to 2 days in our study. Zakhary and colleagues [8] reported an average surgical time of 216.7 min. Hassanpour and colleagues [18] also reported an average anesthetic time of 397.72 min in their study and an average hospital stay of 7.84 days.

Management of intraoperative unavoidable blood loss presents the greatest anesthetic challenge for craniosynostosis surgical correction [19]. Significant blood loss can occur from the subgalial tissues, bony edges, and dural sinuses breeching. Postoperative ICU admission and elective postoperative ventilation can be considered with prolonged surgical duration and hemodynamic instability [19, 20]. No intraoperative complications were reported in our study, and no blood transfusion was needed in any of the patients due to minimal surgical blood loss. Postoperatively, no ICU admission was recorded in any of the study cases and the patients were admitted to the ward. No wound infections or CSF leakage was reported in the follow-up period. This is probably attributed to the simplicity of the procedure, short operative and anesthetic time, and early hospital discharge besides the small number of patients.

For postoperative evaluation, several methods have been proposed for the analysis of the results of different surgical correction procedures for craniosynostosis such as repeated CT scans and different classification and scorings systems [6]. In the present study, we used standardized photographs to evaluate postoperative results based on the preoperative deformities of AP. The advantage of this simple method in comparison to other suggested methods [21–25] is that photographs can be taken easily routinely in the follow-up clinics, and therefore, the data are readily available. Furthermore, it is not time-consuming and does not involve radiation exposure. This method was previously used by Hilling and colleagues [6] in their evaluation of the long-term aesthetic results of fronto-orbital correction for AP.

A theoretical disadvantage of using photographs is its exaggeration of revealing deformities compared with the given real-life three-dimensional impression [6]. Therefore, we added the subjective satisfactory cosmetic outcome reported by the parents of the patients to our outcome evaluation.

Follow-up Ct brain with 3D skull reconstruction was done only in the first 2 cases of the study after 3 months and 1 year postoperatively to confirm bone remodeling and the occurrence of bony fusion.

A very promising cosmetic outcome satisfaction was reported by the parents of the operated patients in the present study. Complete satisfaction was reported by the parents of 6 patients (85.7%) while the parents of 1 patient were partially satisfied (14.3%). Their partial satisfaction was due to the presence of post-operative epilepsy and not related to cosmetic outcomes. No dissatisfaction or the need for a re-do surgery was reported by any of the parents. Our results revealed that our surgical technique produced appreciable improvement in all the presenting features of AP. The forehead shape was the most obvious preoperative deformity; therefore, our technique was mainly directed towards its correction. This was achieved by the rotation of the frontal craniotomy flap to use the more symmetrical curved surface of the postero-medial part of the frontal bone to replace the deficient flattened frontal part creating the new forehead shape. Moreover, augmentation of the frontal bone was done by overlapping the edges of the craniotomy flap with the anterior frontal bone medially to equalize the exaggerated contralateral frontal bossing.

Despite that our technique does not include correction of any accompanying orbital dystopia, improvement in the orbital appearance and normalization of the horizontal orbital position were probably achieved by the release and correction of the frontal supraorbital region. This observation was also reported by Hilling and colleagues [6] without addressing orbital dystopia correction in their surgical correction technique. In addition, we believe that the augmentation of the supraorbital region by the intended overlapping of the frontal craniotomy flap anteriorly over the supraorbital bar added to the normalization of the orbital appearance and consequently to the cosmetic outcome.

Temporal depression is a common observation following open FOA techniques. Several causes have been attributed to this cosmetic complication such as temporalis muscle suspension and atrophy, superficial temporal fat pad atrophy, and inadequate correction and advancement [26, 27]. Different techniques to avoid this problem have been proposed, such as temporal muscle elevation with the scalp flap [28], advancement and resuspension of the temporalis muscle [29, 30], and musculo-osseous advancement flap [31, 32]. However, these techniques have failed to prevent this characteristic complication [31–33]. Ak Oh and colleagues [34] suggested that the inferior coronal gap between the advanced frontal bone and the parietal bone is the main cause of this finding causing a lack of structural continuity and support for the advanced temporalis muscle. They assumed that ossification of the wide bony gap occurs efficiently before dural expansion is complete with

a resultant bony depression. This is markedly obvious as the infantile temporalis muscle bulk is insufficient to hide the underlying depression [34].

In our study, temporal depression was not observed in any of the cases in the follow-up period. We believe that this was attributed to many factors. Our technique followed all the recommendations to ensure a healthy postoperative temporalis muscle to prevent its atrophy. A myocutaneous flap was done in all cases, and no advancement and re-suspension of the temporalis muscle were needed. Unlike the FOA techniques, the ROF technique prevented the development of a significant structural bony discontinuity with an adequate underlying bony support for the temporalis muscle. Moreover, our technique prevented the creation of the wide inferior coronal gap and its assumed role in the development of the postoperative temporal bony depression.

Conclusion

The described ROF technique can be considered one of the minimally invasive techniques for the correction of non-syndromic AP with the advantages of the more aggressive corrective procedures. The initially reported results of this technique are very promising with a lot of advantages. A larger scale of patients is required in future studies for more evaluation of this technique.

Abbreviations

AP: Anterior plagiocephaly; CT: Computed tomography; FOA: Fronto-orbital advancement; ICU: Intensive care unit; ROF: Rotational overlapping flap; UCS: Unilateral coronal synostosis

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Not applicable

Declarations

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Authors' contributions

Prof. MR and MA were the operator and professor of the study. OY collected and analyzed the data and interpreted it and followed-up the patients with prof. MR. The authors have read and approved the manuscript and ensure that this is the case.

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Availability of data and materials

All the data is available.

Ethics approval and consent to participate

This is study is ethically approved by the Neurosurgery Department Cairo University (Prof Mohamed Hafez Ramdan in (7/11/2018); since the reference number is not applicable, we just take approval from our department. This study does not need participation consent as its experiments do not involve humans. It just shows our experience and outcome of surgical treatment of anterior plagiocephaly which was first described by Rougerie in 1972 with a lot of modification since this year.

Consent for publication

Patient photographic authorization and release are signed by the guardian (father) for the two cases.

Competing interests

The authors have no personal, financial, institutional interest, or industry affiliations in any of the drugs, materials, or devices described in this article. The authors declare that they have no competing interests.

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