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Improved Facial and Skull-Base Symmetry following Osteotomy and Distraction of Unilateral Coronal Synostosis

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Background: Unilateral coronal synostosis (UCS) results in a surgically demanding deformation, as the deformity is asymmetric in the calvaria but also presents with facial scoliosis and orbital dystopia. Traditional cranioplasties correct the forehead but have little effect on the face and orbits. In this article, the authors describe a consecutive series of patients operated on for UCS with osteotomy of the fused suture combined with distraction osteogenesis.

Methods: Fourteen patients (mean age, 8.0 months; range, 4.3 to 16.6 months) were included in this study. The authors measured and compared the orbital dystopia angle, anterior cranial fossa deviation, and anterior cranial fossa cant between preoperative computed tomography results and those at distractor removal.

Results: Blood loss was 6.1 mL/kg (range, 2.0 to 15.2 mL/kg), and length of stay was 4.4 days (range, 3.0 to 6.0 days). The authors observed significant improvements in the median orbital dystopia angle from 9.8 degrees (95% CI, 7.0 to 12.6 degrees) to 1.1 degrees (95% CI, −1.5 to 3.7 degrees) ($P < 0.001$), anterior cranial fossa deviation from 12.9 degrees (95% CI, 9.2 to 16.6 degrees) to 4.7 degrees (95% CI, 1.5 to 7.9 degrees) ($P < 0.001$), and anterior cranial fossa cant from 2.5 degrees (95% CI, 1.5 to 3.5 degrees) to 1.7 degrees (95% CI, 0.0 to 3.4 degrees) ($P = 0.003$).

Conclusions: Osteotomy combined with a distractor for UCS straightened the face and relieved orbital dystopia by affecting the nose angle relative to the orbits, correcting the deviation of the cranial base in the anterior fossa, and lowering the orbit on the affected side. Furthermore, this technique demonstrated a favorable morbidity profile with low perioperative bleeding and a short inpatient period, suggesting its potential to improve the surgical treatment of UCS. *(Plast. Reconstr. Surg.* 153: 447, 2024.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Therapeutic, IV.

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THERAPEUTIC

Unilateral coronal synostosis (UCS) constitutes approximately 10% of all incidents of craniosynostosis and primarily causes ipsilateral retrusion and contralateral bulging of the forehead.[1] In addition, UCS can lead to complex secondary deformities that comprise facial asymmetries with deviation of the nose toward the affected side, orbit dystopia, zygomatic asymmetries, and possible effects involving dental occlusion.[2–7] The functional consequences of UCS can include strabismus, amblyopia, and astigmatism,

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and increased intracranial pressure.[8–11] Although UCS normally appears in isolation, it can also present as part of a craniofacial syndrome.[12–14]

Surgical correction of UCS is challenging because of its extensive range of deformities. Traditionally, two major types of correction can be undertaken: fronto-orbital advancement remodeling (FOAR) and endoscopic strip craniectomy (ESC) combined with helmet therapy.[15–18] FOAR can be either unilateral or bilateral depending on the extent of forehead asymmetry.[15,16] and ESC is preferably performed early in life while the bone is still capable of being molded.[19,20] A modification of FOAR is the calvarial switch (CS), which takes advantage of a suitably rounded calvarial bone flap for corrections of the forehead.[21] Although CS can result in a significantly more symmetric forehead relative to FOAR, both methods are limited to the supraorbital level and do not involve correction of the orbits themselves or the facial skeleton below the orbits.

Comparisons of FOAR with ESC reveal that ESC shows a better morbidity profile,[22,23] improved ophthalmologic results, and equivalent aesthetic outcomes relative to FOAR[18,23]; however, actual correction of the orbit is reportedly independent of surgical method.[24] A new method for UCS correction was proposed based on osteotomy and fronto-orbital distraction (FOD).[25–29] Briefly, the fused suture is osteotomized and a distractor placed transverse to the osteotomy line, after which the forehead is distracted anteriorly and slightly downward. Following completion of the distraction and a consolidation period, the distractor is then removed. To date, case series have presented favorable results in terms of aesthetics, morbidity, and strabismus.[10,22] Moreover, reports suggest that alterations of the skull base in response to the distraction are important to the surgical outcome.[28,30]

Previous studies describe various ways to evaluate UCS treatment, ranging from pure aesthetic evaluation to ocular evaluation and asymmetry assessment according to three-dimensional models.[2,8,15] However, studies describing the use of distraction osteogenesis for UCS correction seldom used standardized points of reference on computed tomographic (CT) scans to evaluate the effect.[22,26,29] In addition, Choi et al.[28,30] described seven patients with UCS treated with FOD and in whom similar but not identical points of reference were used to calculate skull-base deviation.

At our institution, we switched surgical technique in 2018 from CS to FOD. In this study, we retrospectively evaluated short-term outcomes by comparing preoperative and postoperative results in terms of the facial symmetry, orbital dystopia, and skull-base morphology of the first 14 consecutive cases of UCS operated on with FOD.

PATIENTS AND METHODS

Surgical Technique

An anterior scalp flap was raised in the subgaleal plane by means of a bicoronal incision. At 10-mm above the supraorbital rim on the affected side, the periosteum was transected, with the dissection continuing subperiosteally into the orbit to expose the anterior part of the orbital roof. Osteotomy in the temporal region was then performed to enable placement of the distractor at the desired downward angle.

The closed coronal suture was osteotomized from the anterior fontanel down to the squamous suture using a rotating craniotome. The osteotomy was then extended along the frontosphenoidal suture, across the frontozygomatic suture into the orbit, and finally along the orbital roof using an ultrasonic osteotome or chisel. The lateral two-thirds of the orbital roof was osteotomized, and the dura was protected during the osteotomy of the orbital roof from the orbital side (Fig. 1). We paid particular attention to the lateral corner, which represents the connection point of the anterior fossa floor. The forehead became movable on completion of the osteotomy.

A 30-mm Arnaud distractor (KLS Martin, Tuttlingen, Germany) was placed, activated, and fixed with eight Matrix Midface screws (DePuy Synthes GmbH, Oberdorf, Switzerland) across the opened suture in a preplanned location. At this point, although minor corrections can be made if the angle requires adjusting, such corrections will inevitably lead to loss of distraction capacity. The distractor pin then penetrates the skin through either the bicoronal incision or a separate incision. If the bone quality is good, the location of the distractor can be placed at the desired angle 10 mm behind the closed suture, which will increase the likelihood that the pin will not need to penetrate the skin anterior to the bicoronal incision, thereby possibly reducing the risk of secondary skin problems at the anterior end of the distractor.

The distraction begins on the first postoperative day and continues with three turns (0.9 mm) daily until reaching 30 mm. After an additional 3-month consolidation period, the distractor is removed under general anesthesia.

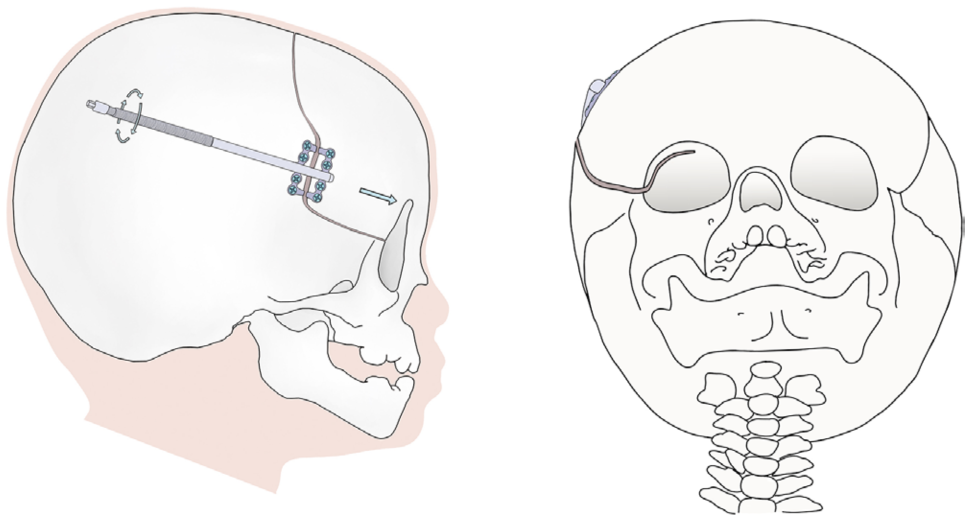

Fig. 1. Schematic presentation of the osteotomy lines from the anterior fontanel, along the closed suture, across the frontozygomatic junction, and along the lateral part of the orbital roof.

Patients

This retrospective study describes all consecutive patients with UCS who underwent surgical treatment with FOD between 2018 and 2021 at Sahlgrenska University Hospital, Gothenburg, Sweden. The patients underwent preoperative CT scanning for diagnostic purposes; another scan was obtained before distractor removal. The control program also included a CT scan at 3 years of age, which will be analyzed in a future study when the present patients have reached that age. Data regarding age at preoperative CT scan, side of fusion, age at operation and distractor removal, body weight, duration of operation, perioperative blood loss, volume of blood transfusion, other complications, and length of hospital stay following distractor placement were collected from either the Gothenburg Craniofacial Registry or medical charts. All patients underwent genetic analysis with an in silico gene panel comprising 29 to 32 genes related to craniosynostosis on a whole-exome or whole-genome platform.

Measurements

Three-dimensional reconstructions of CT scans were generated using syngo.via (v.VB30B; Siemens Healthineers, Siemens G, Erlangen, Germany), and measurements were performed in Agfa (v.8.1.5.102; Agfa HealthCare, Agfa-Gevaert NV, Mortsel, Belgium). In the frontal projection, facial symmetry was measured using the orbital dystopia angle (ODA). In the horizontal and coronal planes, we measured the anterior cranial fossa deviation (ACFD) and the anterior cranial fossa cant (ACFC), respectively.[31]

Orbital Dystopia Angle

The ODA was defined as the angle of the nasale and the line connecting the upper limits of the orbits (Fig. 2). Negative values indicated that the nasale was oriented toward the nonsynostotic side relative to the angle formed by the orbits.

Anterior Cranial Fossa Deviation

The ACFD was determined by measuring the angles between a midline from nasion to

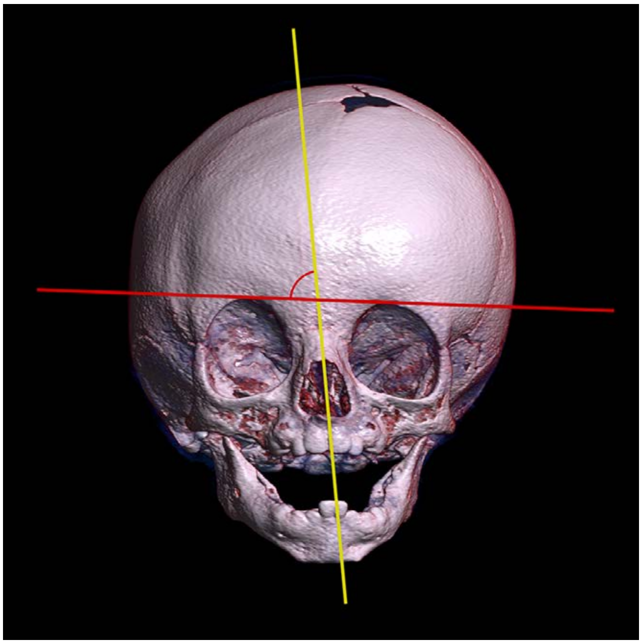

Fig. 2. The ODA was measured as the angle formed by the nasale (*yellow*) and a line connecting the upper limits of the orbits (*red*).

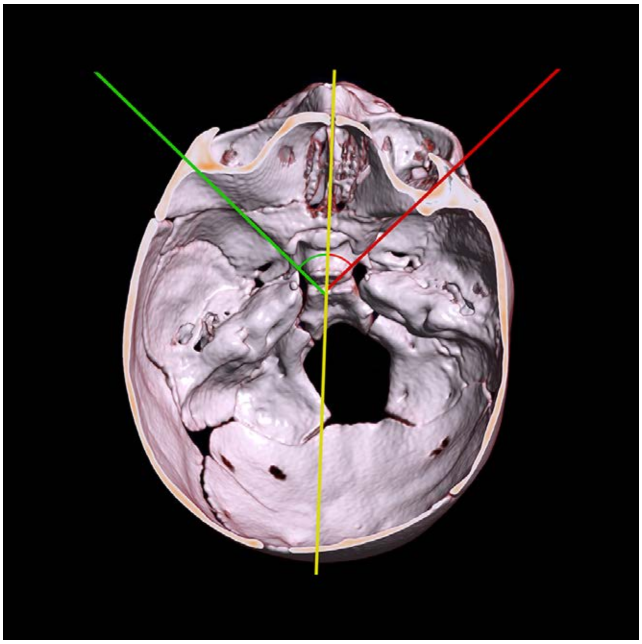

Fig. 3. The ACFD was measured as the difference between the angles formed by connecting the nasion to the sella (*yellow*) and from this line to the lesser wing of the sphenoid bone on the contralateral (*green*) and ipsilateral (*red*) sides.

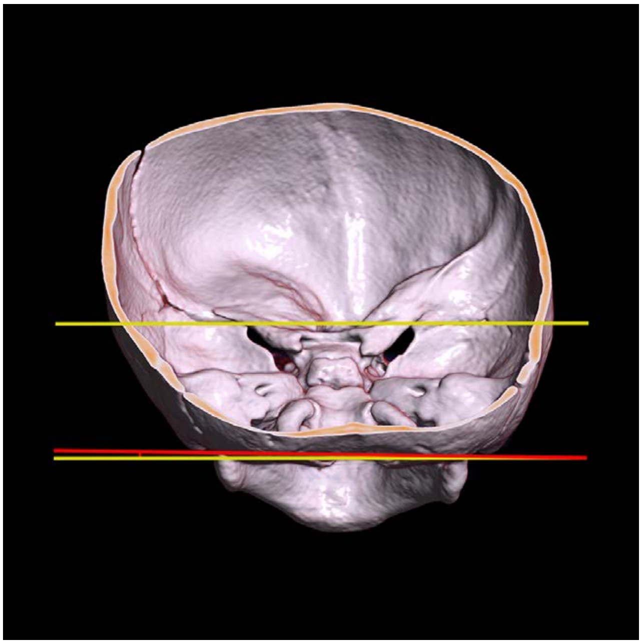

Fig. 4. The ACFC was measured as the angle formed by a line connecting the superolateral points of the superior orbital fissures (*yellow*) and a line connecting the mastoid processes (*red*).

the center of the sella turcica and the posterior rim of the lesser sphenoid wing on each side (Fig. 3).

Anterior Cranial Fossa Cant

The ACFC was measured as the angle of deviation formed by a line between the mastoid processes relative to a line between the superolateral points of the superior orbital fissures (Fig. 4). Negative values indicated that the deviation was toward the nonsynostotic side.

Complication Registration

Complications were registered until 30 days postoperatively (both after the primary operation and after distractor removal). All complications were graded according to the Oxford system.[32]

Intraclass Correlation Coefficient

All measurements were performed in triplicate by a single observer, and the mean value was used. Because the study was based on repeated measures of several different patients, we calculated the intraclass correlation coefficient (ICC) for each measurement (ODA, ACFD, and ACFC) to determine measurement variability both within the same patient and between patients.

Statistical Analysis

Statistical analyses were performed using SPSS (v.28.0; IBM Corp., Armonk, NY). Patient, operation, and postoperation data were expressed using descriptive statistics and presented as the mean and range (Table 1). Quantitative data for the angles were presented as preoperative and postoperative values, change in degrees, change in percentage, and the median and 95% confidence interval and interquartile range. Quantile regression for the 50th quantile (median) was applied to estimate the median (95% CI) for each outcome variable. The intercept (95% CI) from the quantile regression was reported as the median (95% CI) in Table 2 as descriptive statistics (summary of measurements). For comparisons between the preoperative and postoperative measurements, nonparametric tests (Wilcoxon signed rank test) were used. All P values were two-sided, and a value of $P < 0.05$ was considered statistically significant.

For ICC calculations, a two-way random model was applied. The study was approved by the Gothenburg ethics committee (approval no. 784:11).

RESULTS

Patients

A total of 14 patients were included in the study with at a gender ratio of 1:1 (Table 1). Of

Table 1. Patient Information and Perioperative and Postoperative Data

	Age at Preoperative Evaluation (Mo)[a]	Age at OP (Mo)	OP Duration (Min)	Perioperative Bleeding (mL/kg)	Transfusion Volume (mL)	Length of Hospital Stay (Days)	Age at Distractor Removal (Mo)	Duration of Distraction (Mo)
Mean	5.7	8.0	89.5	6.1	42.6	4.4	12.5	4.5
Range	0.8–15.2	4.3–16.6	63.0–121.0	2.0–15.2	0–200	3.0–6.0	8.0–21.1	2.6–6.2

OP, operation.
[a]Includes CT scan, photographic documentation, neurologic status, and cosmetic evaluation.

these patients, 57% presented issues with the right coronal suture ($n = 8$) and 43% with the left coronal suture ($n = 6$). Mean age at operation and distractor removal was 8 months and 12.5 months, respectively. Two patients harbored a variant of uncertain significance in the ETS2 repressor factor (case 5) or twist family basic helix-loop-helix transcription factor 1 (case 12) gene, respectively.

Surgery and Complications

Mean perioperative blood loss was 6.1 mL/kg body weight (range, 2.0 to 15.2 mL/kg), with five patients requiring a blood transfusion. Mean duration of hospital stay was 4.4 days (range, 3.0 to 6.0 days) (Table 1).

Complications up to 30 days after the primary operation included three cases treated with antibiotics for suspected infection (Oxford 1) and one reoperation because of a broken distractor arm (Oxford 3). The complications up to 30 days after distractor removal included one reoperation because of a superficial abscess (Oxford 3). The complete range of complications (also those >30 days postoperatively and after distractor removal) included another five cases of Oxford 1 complications [three wounds (exposed distractor), one superficial infection at the point where the distractor arm penetrated the skin, and one case treated with antibiotics because of suspected infection]. In all patients, we observed overbridging ossification of the gap at the time of distractor removal.

FOD Improved Postoperative Outcomes

ODA improved in all cases, with a median improvement of 87.5% ($P < 0.001$) at distractor removal and even overcompensation in three cases. In one of these three cases (case 10), the deviation became greater than that measured preoperatively but instead was contralateral to the synostotic side. In addition, ACFD improved in all cases, with a median improvement of 68.3% ($P < 0.001$) and overcompensation occurring in two cases (but not becoming greater than the original deviation). Moreover, ACFC improved in all cases, with a median improvement of 41.4% ($P = 0.003$). Similar to ACFD, overcompensation developed in two cases but did not advance more than the initial deviation. In one case (case 12), the preoperative CT scan did not capture both of the mastoid processes required to determine the ACFC (Table 2). Table 3 presents the ICC and the respective 95% confidence intervals. For all measurements, the variability between patients was larger than that within the same patient. In Figure 5, the change in all three angles and the concomitant change in facial appearance is demonstrated.

DISCUSSION

Correction of UCS is challenging, and the extensive range of methods available for this activity suggests that none stands out as preferable. A recent study of facial soft-tissue asymmetry indicated that considerable asymmetry remained following surgical correction with bilateral craniotomy of the frontal bone and unilateral advancement of the supraorbital rim.[33] In the present study, we evaluated data from a consecutive series of UCS cases operated on using the new technique FOD, which demonstrated a beneficial morbidity profile and shorter operation time, and resulted in less perioperative bleeding than the more extensive CS. In addition, patients operated on with FOD did not require postoperative admission to the intensive care unit and stayed in the hospital for shorter durations. Furthermore, FOD corrected the orbital dystopia and straightened both the nose and the skull base in the anterior cranial fossa of all operative patients.

A principal difference between FOD and traditional open cranioplasty techniques (eg, FOAR and CS) is that despite the limited osteotomies in FOD, the distractor-mediated effects can occur not only locally in the temporal region but also on the orbits, facial skeleton, and skull base. By contrast, FOAR and CS effects are limited to the supraorbital level, enabling their correction of the forehead while leaving several of the secondary deformities of the orbits and facial skeleton

Table 2. Summary of Measurements

	ODA (Deg)[a]				ACFD (Deg)[b]				ACFC (Deg)[c]			
Case	Preoperative	Distractor Removal	Change	Change (%)	Preoperative	Distractor Removal	Change	Change (%)	Preoperative	Distractor Removal	Change	Change (%)
1	10.4	3.0	7.4	71.2	15.0	1.0	14	93.3	2.3	2.0	0.3	13.0
2	7.6	4.4	3.2	42.1	14.3	10.0	4.3	30.1	2.5	–2.4	4.9	196.0
3	8.3	1.1	7.2	86.7	16.9	8.0	8.9	52.7	2.3	0.4	1.9	82.6
4	9.8	0.7	9.1	92.9	6.5	4.7	1.8	27.7	4.5	–0.1	4.6	102.2
5	10.1	1.1	9.0	89.1	8.2	2.6	5.6	68.3	2.4	1.1	1.3	54.2
6	8.6	1.7	6.9	80.2	12.9	5.5	7.4	57.4	2.2	0.7	1.5	68.2
7	9.5	3.3	6.2	65.3	14.8	7.2	7.6	51.4	4.6	3.7	0.9	19.6
8	3.9	–3.8	7.7	197.4	13.2	7.7	5.5	41.7	–0.5	–0.2	0.3	60.0
9	11.7	1.5	10.2	87.2	8.8	1.5	7.3	83.0	4.8	3.5	1.3	27.1
10	6.2	–7.0	13.2	212.9	6.4	–1.0	7.4	115.6	4.6	4.5	0.1	2.2
11	11.4	–3.9	15.3	134.2	10.1	0.9	9.2	91.1	2.0	1.6	0.4	20.0
12	1.6	0.2	1.4	87.5	6.6	–5.8	12.4	187.9	N/A	4.7	N/A	N/A
13	12.8	1.6	11.2	87.5	32.8	2.6	30.2	92.1	3.1	2.4	0.7	22.6
14	10.5	0.4	10.1	96.2	10.0	7.5	2.5	25.0	2.9	1.7	1.2	41.4
Median	9.8	1.1	9.0	87.5	12.9	4.7	7.4	68.3	2.5	1.7	1.2	41.4
IQR	3.5	2.8	3.7	27.7	7.1	6.6	4.8	53.6	2.3	3.3	1.4	55.6
95% CI	7.0–12.6	–1.5 to 3.7	5.4–12.6	40.2–134.8	9.2–16.6	1.5–7.9	3.3–11.5	36.8–99.8	1.5–3.5	0.0–3.4	–0.4 to 2.8	8.8–74.0
P	<0.001				<0.001				0.003			

N/A, not available; IQR, interquartile range.
[a]Negative values indicate the nasale pointing toward the unaffected suture.
[b]Negative values indicate that the synostotic angle was greater than the nonsynostotic angle.
[c]Negative values indicate the contralateral fossa cant.

Table 3. Intraclass Correlation Coefficient

Measurement	ICC (%)	95% CI
ODA		
Preoperatively	93.2	84.5–97.6
Distractor removal	97.8	94.9–99.2
ACFD		
Preoperatively	97.0	92.9–99.0
Distractor removal	93.3	84.7–97.6
ACFC		
Preoperatively	94.6	87.1–98.2
Distractor removal	98.6	96.8–99.5

uncorrected. Notably, ESC combined with helmet therapy demonstrates similar modes of action to FOD. In ESC, the osteotomies allow for a gradual correction mediated by the helmet, which is similar to how the distractor works in FOD.[19] Comparisons of surgical results, including measurements of forehead symmetry, indicated significantly better outcomes after CS relative to FOAR, and a more favorable morbidity profile, shorter operation time, decreased blood loss, and shorter hospital stay for FOD relative to both CS and FOAR.[21] Furthermore, studies show that unilateral and bilateral FOAR are equally effective, in particular when postoperative temporal hollowing is determined.[15,16] In general, ESC shows good performance in terms of morbidity and the extent of surgical correction.[9,23] These findings together with those of the present study suggest that a combination of endoscopic techniques, followed by placement of the distractor, might be optimal for UCS correction.

In this study, we showed that FOD was capable of simultaneously correcting several UCS deformities. Specifically, the retruded forehead on the affected side underwent anterior and caudal distraction, thereby improving both the facial scoliosis and orbit dystopia. In addition, medial, downward pressure on the facial skeleton and anterior skull base both straightened the nose and resolved orbital dystopia, which was reflected in all three measurements (ODA, ACFD, and ACFC). The most obvious of the measured effects was normalization of the nasal structures, which when combined with correction of the orbit dystopia resulted in recovered facial symmetry. Whether forehead asymmetry is equally corrected using this technique remains to be confirmed.[34] Furthermore, although we identified residual deviations in all measurements, their statistical difference from normal variations remains unclear and warrants further studies. Median improvement in orbital dystopia was 87.5%, and three patients were overcompensated, with one (case 10) showing an even greater deviation than that measured preoperatively.

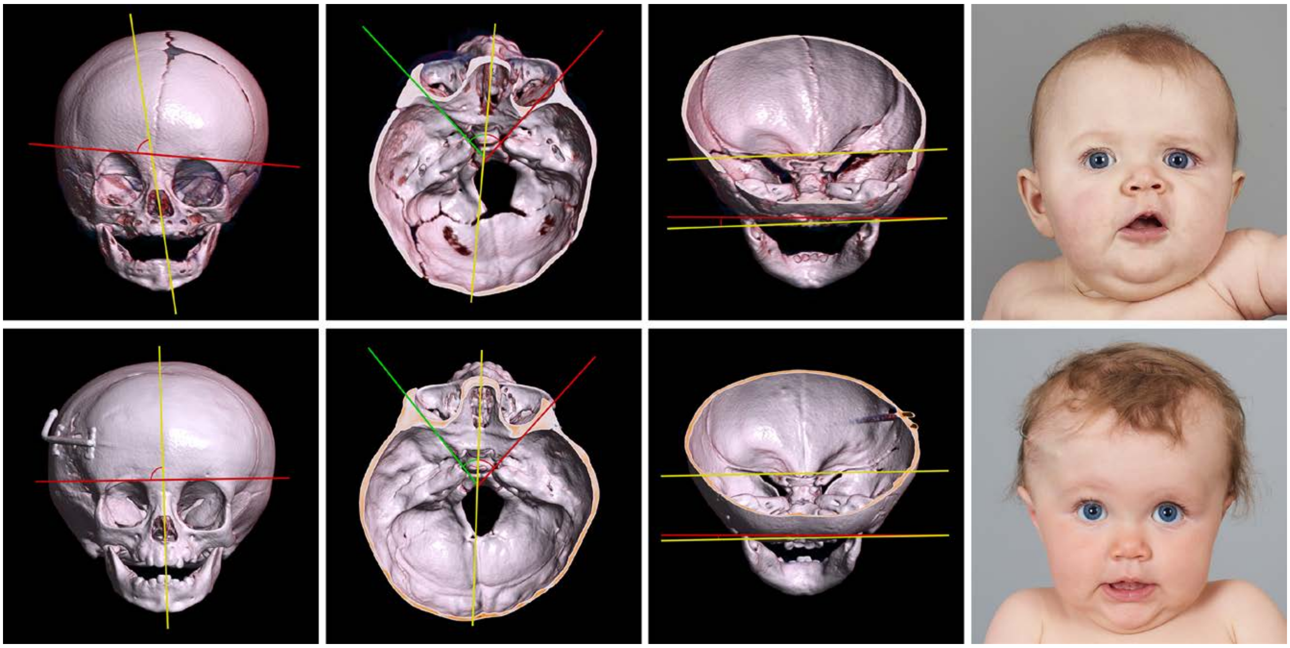

Fig. 5. Preoperative three-dimensional CT scans (*above*) and those at the time of distractor removal (*below*) along with preoperative and postoperative photographs of the patient (female) with UCS of the right coronal suture. Scans show changes in the ODA (*left*), the ACFD (*center*), and the ACFC (*right*) before (*above*) and after (*below*) FOD. Aesthetically, the nasal root deviation and orbital dystopia were corrected by distraction treatment.

Several studies indicate that overcorrection is desired to achieve better results at long-term follow-up[35–37]; therefore, it will be interesting to evaluate how these residual deviations develop at the 3-year follow-up.

Evaluation of the data suggested that FOD resulted in a slight undercorrection in the ODA, in which case a more pronounced distraction would be desirable. However, we also observed overcorrection of the ODA in three of the 14 cases. To better individualize the treatment, using a longer distractor and obtaining an additional CT scan at 20 to 25 mm of distraction could offer insight into how much farther the distraction should proceed (with the aim of approaching 0 degrees). Given the difficulty in justifying additional radiation exposure based on the current data, results of long-term follow-up at 3 years of age may clarify this matter.

The theory associated with the involvement of the coronal ring could possibly contribute to an increased understanding of the variations observed in both preoperative status and postoperative outcomes. Previous studies report various morphologic changes, depending on the precise distribution of the synostosis (ie, purely frontoparietal or a combination of frontoparietal and frontosphenoidal).[38–40] Moreover, there is some inconsistency in the literature as to what combination results in the most pronounced changes. However, it is possible that detailed anatomical localization of the extent of the synostosis could contribute to future simulations of both the angle at which to place the distractor and how far the distraction should proceed.

There occasionally exist difficulties with familial acceptance of aspects of the FOD technique. In our experience, family members quickly adapt to the turning of the distractor pin; however, challenges arise in the event of complications. In the present study, three cases presented wounds related to distractor penetration. In all cases, the distraction was allowed to proceed, usually in the absence of antibiotic treatment, resulting in distractor removal at the desired date with no long-term effects beyond a minor scar on the side of the forehead. To reduce skin problems at the head of the distractor, we began positioning the distractor approximately 10 mm posterior to the closed suture (assuming adequate bone quality). Whether this minor adjustment alleviates the healing problems associated with skin penetration remains to be elucidated.

This study has some limitations. Although we used several previously identified points of reference, their application still proved to be a limitation, as the references were occasionally hard to define because of anatomical variations between the patients. In addition, this proved challenging when measuring the same patient because of their natural development and the effect of distraction on the growing skull. Specifically, the anatomy of the lesser wing of the sphenoid bone varied considerably, with a more prominent arch observed in some patients, which made it difficult to estimate a straight angle. Regarding skull base cant, because of the natural development of the mastoid processes, which are absent at birth but grow rapidly during the first year, the extent of ACFC was sometimes hard to calculate, especially using preoperative CT scans.[41–43] Moreover, given that these measurements represent visual estimates and are subsequently prone to inconsistencies, a potential compensation could involve several observers performing the same measurements. However, ICC analysis showed that the variability within the same patient was much lower than that between patients, suggesting the reproducibility of the measurements and the adequacy of the previously defined reference points used for the measurements. Another limitation is the lack of direct comparison of the morphometric outcomes relative to those obtained using other techniques. Because the previously described traditional treatments do not include a follow-up CT scan at 4.5 months postoperatively (during which the distractor is removed), a direct comparison at this stage is not possible. However, we plan on performing this comparison for the current cohort following acquisition of data on the patients reaching 3 years of age.

In summary, our retrospective analysis revealed that FOD to correct UCS presents a highly favorable morbidity profile and results in excellent correction of facial symmetry. Further studies evaluating the long-term results of FOD will determine its suitability to replace FOAR and CS for correction of UCS.

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DISCLOSURE

The authors have no conflicts of interest to declare.

PATIENT CONSENT

Parents or guardians provided written informed consent for use of the patient images.

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