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Surgical Correction of Unicoronal Synostosis: Fronto-orbital Distraction versus Calvarial Switch

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Short Running Head: Surgical Correction of Unicoronal Synostosis

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Abstract

Background: There is a need for a new, less invasive surgical option for unicoronal synostosis (UCS). The aim of this study was to compare the resulting morphology and symmetry in patients with UCS following fronto-orbital distraction (FOD) or calvarial switch (CS).

Methods: 79 patients with isolated UCS operated between 2005 and 2021 were analyzed. Follow-up was until 3 years of age. Angles describing orbital dystopia (ODA) and the anterior cranial fossa deviation and cant (ACFD and ACFC, respectively) were measured. Key linear dimensions, cranial cavities, and indices were calculated.

Results: 66 patients were included (14 in the FOD group and 52 in the CS group). The 3-year follow-up revealed significant improvement in all angles in both groups, with significant superiority in ODA correction following FOD (median improvement of 5.7 degrees as compared with 3.3 degrees after CS). Additionally, nasal and orbital volumes tended to be smaller, especially following CS; however, FOD resulted in a smaller absolute difference in orbital volume. Asymmetry in the orbital, nasal, and sphenoid regions also improved at the 3-year follow-up in both groups, although FOD resulted in complete normalization of the affected orbital shape and significantly improved overall asymmetry relative to that observed in the CS group.

Conclusions: This study found that FOD achieves superior overall symmetry, as well as better shape correction of the cranium, as compared with CS, while also being less invasive. These findings suggest FOD as a safe and effective alternative to correct UCS and possibly the preferred surgical method.

Introduction

Unicoronal synostosis (UCS) occurs in ~0.7 in 10 000 births and is the second most common form of craniosynostosis.¹ Patients present ipsilateral retrusion and contralateral protrusion of the forehead, orbital dystopia with elevation of the ipsilateral orbital roof, nasal root deviation towards the affected side, zygomatic asymmetries, and affected dental occlusion.²⁻⁸ Some of the functional consequences of UCS include increased intracranial pressure and ocular disorders.⁹⁻¹² Patients with UCS may also suffer from lower than average neurodevelopment.¹³⁻¹⁶

There is no consensus on the optimal treatment for UCS. The most common technique is fronto-orbital advancement remodeling (FOAR).^{17,18} Calvarial switch (CS) is a FOAR variant, where an appropriately curved bone flap from the calvaria replaces the skewed forehead.¹⁹ Although capable of improving forehead symmetry, FOAR methods have minimal effect on facial symmetry.²⁰⁻²⁵ Dynamic techniques have recently emerged as an alternative. Endoscopic strip craniectomy combined with helmet remolding produces satisfactory correction but requires consistent compliance and is best suited for early intervention.²⁶⁻²⁹ Spring-assisted surgery (SAS) is another alternative and was first described by Uejima.³⁰⁻³³ The first case report on SAS for UCS concluded that despite successful correction of facial scoliosis, issues remained concerning the unpredictability of using springs.³⁴ Fronto-orbital distraction (FOD), first described by Kobayashi et al.,³⁵ uses traditional distractors instead of springs, thereby reducing their observed issues. Despite growing interest in FOD, few case studies have been published.^{2,5,36-42} At our institution, the surgical standard has evolved to FOD from CS with positive preliminary results, including normalization of orbital dystopia and the anterior cranial fossa.² Similar results were recently reported by Park et al.⁴⁰ Several studies have employed various methods to evaluate surgical outcomes for correcting UCS. These range from assessing improvements in perioperative morbidity to ocular

symptoms; however, such methods seldom use standardized points of reference and/or lack a control group.^{3,9,17,36,38-40,43,44} In this study, we performed a direct comparison of surgical outcomes between FOD and CS in terms of improvements in perioperative morbidity and detailed asymmetry. Furthermore, this is the first study employing a cohort of non-UCS patients as a comparative control. Our aim was to analyze changes in facial symmetry before and after surgical correction of UCS to determine the optimal procedure for improving morbidity.

Methods

The study was conducted according to the principles stated in the Declaration of Helsinki. Ethical approval was obtained from the Gothenburg Ethics Committee (Dnr 784:11).

Study Design

This is a single-center, retrospective, comparative effectiveness study on all consecutive non-syndromic patients with isolated UCS treated from 22 June 2005 to 29 June 2021. The 3-year follow-up period occurred from 20 September 2007 to 18 December 2023. Surgical outcomes in the form of anthropometric measurements and morphologic analyses were compared against those in a previously published control dataset of non-pathological individuals ranging from 0 to 48 months of age ($n = 217$).⁴⁵

Participants

The patients were all treated at Sahlgrenska University Hospital in Gothenburg, Sweden, and retrospectively divided into two groups according to operation (FOD or CS) (Fig. 1). UCS diagnosis was made in the craniofacial unit through clinical examination and verified by computed tomography (CT), with follow-up CT at 3-years of age included in the analysis. Included patients were those that underwent either FOD or CS. Those with syndromic diagnoses or inadequate CT scans were excluded. FOD patients underwent CT prior to distractor removal.

Interventions

CS

From 2005 to 2018, CS was the predominant surgical method for treating UCS at our center (Fig. 2). This method has been shown to be superior in achieving forehead symmetry compared to previous techniques, such as bilateral FOAR.¹⁹ However, it still presents limitations, particularly its restriction to the supraorbital region. In short, following a bicoronal zigzag incision, subperiosteal dissection exposed the frontal bone and the calvaria. A suitably rounded bone flap was identified and harvested, and after removal of the deformed forehead, the bone flaps switched places. The supraorbital complex was removed, partitioned in the midline, and re-fixated to the new forehead. The new forehead was adjusted and fixated with sutures and resorbable plates. To enhance symmetry, barrel-staving and out-fracturing of the parietal bone on the affected side were performed.

FOD

Since 2018, FOD has been exclusively utilized for UCS regardless of severity. The general protocol includes surgery approximately two months after the initial evaluation and confirmation of diagnosis. This is followed by the distraction and consolidation phases, at the end of which all patients undergo a CT scan. An additional evaluation and CT scan are performed when the patient reaches three years of age. An anterior scalp flap was raised using a bicoronal incision to expose the orbital roof (Fig. 2). A temporal osteotomy then allowed the desired distractor placement. The osteotomy extended from the fontanel to the squamous suture along the fronto-sphenoidal suture and into the orbit through the fronto-zygomatic suture. The distractor, a 30-mm Arnaud device (KLS Martin, Tuttlingen, Germany), was affixed with eight MatrixMIDFACE™ screws at a preplanned location. Minor angle adjustments were possible at this stage but were preferably avoided, as any removal of bone to adjust the angle would inevitably reduce the total distraction capacity. Distraction

commenced immediately with three daily turns until reaching 30 mm. Distractor removal occurred after a 3-month consolidation period under general anesthesia.

Primary Outcomes

We performed morphologic analysis and three primary anthropometric measurements of angles, indices, and volumes to evaluate various aspects of cranial size, shape, and symmetry. These occurred both preoperatively and to assess changes at the 3-year follow-up.

Orbital dystopia angle (ODA), anterior cranial fossa deviation (ACFD), and anterior cranial fossa cant (ACFC)^{2,46} were measured and compared between the two surgical interventions preoperatively and at the 3-year follow-up (Figure, Supplemental Digital Content 1).

Additionally, five common cranial indices,⁴⁵ including the cephalic index (CI), upper facial index (UFI), nasal index (NI), orbital index at both affected and non-affected sides (AOI and NAOI, respectively), and one index for the sphenoid wing (SWI; defined by authors), were calculated using linear dimensions and landmarks to estimate shape changes at specific cranial regions (Figure, Supplemental Digital Content 1). Volumetric changes were quantified through measurements of intracranial volume (ICV), nasal cavity volume (NCV), and orbital volume at both affected and non-affected sides (AOV and NAOV).

Overall asymmetry was assessed through principal component analysis (PCA)⁴⁷ based on the landmark configurations⁴⁵ of 88 anatomical landmarks and 1,152 pairs of bilateral surface semilandmarks placed on the reconstructed skulls of all evaluated individuals. The function *show.asymmetry* in the R statistical environment (R Core Team, 2019) was used to calculate the amount of cranial asymmetry.⁴⁷ The surface shape variation along the first two PC scores were visualized, and the level of asymmetry in different subgroups was quantified and reported using density distribution plots.⁴⁸

Secondary Outcomes

Demographic and operative data, including operation time, perioperative bleeding and transfusion, length of stay (LOS), and complications, were collected from the Gothenburg Craniofacial Registry and medical charts. All complications were graded using the Oxford system and registered until 30-days postoperatively, including after the initial operation and following distractor removal.⁴⁹

Statistical Analysis

Analyses were conducted using SPSS (v.29.0; IBM Corp., Armonk, NY, USA).

Demographics were analyzed with a Chi-squared test, and Mann-Whitney *U* test was used to compare operative data between groups. Pre- and postoperative measurements within each group were compared using Wilcoxon signed-rank tests, and FOD and CS outcomes were compared using the Mann-Whitney *U* test. Wilcoxon signed-rank tests was used to evaluate overall asymmetry at different periods. All *P* values were two-sided, with a $p < .05$ considered significant.

Patient Characteristics

A total of 79 patients underwent surgery for UCS between 2005 and 2021 at Sahlgrenska University Hospital. Of these, 14 underwent FOD and 65 FOAR. In the CS group, eight patients lacked adequate preoperative CT scans, and two lacked these at the 3-year follow-up for angle measurements. For linear dimensions and volumes, 10 patients lacked preoperative CT scans, and two lacked these at the 3-year follow-up. All FOD cases had sufficient CT scans for angle measurements at all stages, although the low image quality of some scans precluded analysis of linear dimensions and volumes.

As a result, this study included 66 patients (Table 1 and Table, Supplemental Digital Content 2). Mean age at the preoperative CT scan was 5.7 months (range: 0.8–15.2 months) and 6.4 months (range: 0.0–23.6 months) for the FOD and CS groups, respectively. Mean age at

operation was 8.0 months (range: 4.3–16.6 months) and 8.7 months (range: 4.6–25.5 months) for the FOD and CS groups, respectively. Mean age at distractor removal and duration of distraction in the FOD group was 12.5 months (range: 8.0–21.2 months) and 4.5 months (range: 2.6–6.2 months), respectively. Mean age at the 3-year follow-up was 38.1 months (range: 35.6–44.7 months) and 36.9 months (range: 32.5–41.5 months) for the FOD and CS groups, respectively.

Primary Outcomes

Angle Deviation

Comparison at the 3-year follow-up between the two groups identified a significant difference in correction of ODA ($P = 0.008$), with FOD improving ODA by a median of 5.7 degrees [interquartile range (IQR): 3.1 degrees] as compared with 3.3 degrees (IQR: 3.8 degrees) after CS (Table 1). ACFD improved by a median of 7.4 degrees and 5.5 degrees following FOD and CS, respectively ($P = 0.188$), and ACFC improved by a median of 2.5 degrees and 1.6 degrees following FOD and CS, respectively ($P = 0.430$). ODA and nasal root deviation in both groups consistently occurred preoperatively towards the affected side. Similarly, ACFD consistently occurred preoperatively with the non-synostotic angle greater than the synostotic angle. All but two patients (one in each group) suffered from ACFC, where the superior orbital fissure on the affected side deviated upwards preoperatively.

Changes in Size and Shape

Although both FOD and CS similarly resulted in improved nasal shape to levels comparable with measurements in the control group at the 3-year follow-up (Fig. 3 A), NCV showed different variations: NCV was similar to control values preoperatively, larger upon distractor removal, and smaller at the 3-year follow-up (Fig. 3 B). Preoperative AOI differed significantly from control values, with orbital height larger than the width. Following distractor removal, AOI in the FOD group was similar to the control group and persisted until

the 3-year follow-up (Fig. 3 C), with NAOI following a similar pattern (Fig. 3 E). Notably, measurements of AOV and NAOV were similar to control values preoperatively, with both FOD and CS resulting in overall volume reductions at the 3-year follow-up (Fig. 3 D and F). However, fewer patients showed a >10% absolute difference between AOV and NAOV at the 3-year follow-up, with absolute differences of <5% more common in the FOD group (Figure, Supplemental Digital Content 3 E). Overall changes in the shape and size of facial and calvarial regions followed similar patterns at preoperation, distractor removal, and the 3-year follow-up in both groups, resulting in measurements comparable to those in the control group (Figure, Supplemental Digital Content 3 A, B, and D). The greater sphenoid wing length on the affected side was consistently ~50% smaller than that on the unaffected side preoperatively in both groups, resulting in a low SWI. Following distractor removal, FOD resulted in an SWI closer to that measured in the control group relative to CS at distractor removal, although no apparent difference in overall improvement was observed between groups at the 3-year follow-up (Figure, Supplemental Digital Content 3 C).

Overall asymmetry

PCA revealed major asymmetry variations in the cranial morphology of all patients in both groups at different stages (PC1 explained 67.09% of the variability), with the data showing that FOD-treated patients demonstrated higher levels of similarity with the controls (Fig. 3 A–B). Results for PC2 (explaining 7.62% of the variability) indicated marginal changes primarily affecting the posterior portion of the neurocranium and with less impact on the facial complex (Figure, Supplemental Digital Content 4). PCA results suggested no significant differences in overall asymmetry between preoperation and at the 3-year follow-up in the CS group ($P = 0.256$). However, the FOD group showed significant improvements in asymmetry at distractor removal ($P < 0.001$) and the 3-year follow-up ($P = 0.029$) relative to

both preoperative measurements and as compared with the CS group (Figure, Supplemental Digital Content 5 and Table, Supplemental Digital Content 6).

Secondary Outcomes

The FOD group demonstrated significantly better outcomes than the CS group across all assessed areas. Mean duration of operation was 89.5 min (range: 63.0–121.0 min) and 153.5 min (range: 60.0–209.0) for FOD and CS, respectively ($P < 0.001$). Mean perioperative bleeding was 6.1 mL/kg (range: 2.0–15.2 mL/kg) and 22.1 mL/kg (range: 1.2–72.2 mL/kg) for FOD and CS, respectively ($P < 0.001$). Mean perioperative blood transfusion was 3.3 mL/kg (range: 0.0–14.0 mL/kg) and 14.3 mL/kg (range: 0.0–36.2 mL/kg) for FOD and CS, respectively ($P < 0.001$). Mean LOS was 4.3 days (range: 3.0–6.0 days) and 5.8 days (range: 4.0–8.0 days) for FOD and CS, respectively ($P < 0.001$).

In the FOD group, three patients received antibiotics for suspected infection (Oxford 1), and one experienced a broken distractor arm due to trauma and required reoperation (Oxford 3). After distractor removal, one patient underwent debridement for a suspected superficial abscess (Oxford 3). In the CS group, three patients presented superficial skin infections: one was managed with local wound care (Oxford 1), and the other two required intravenous antibiotics (Oxford 1 and 2). Of these cases, one also needed wound re-suturing at a primary healthcare center (Oxford 1). Additionally, one patient experienced postoperative obstipation and vomiting but fully recovered within 1 month (Oxford 2).

Discussion

Asymmetry in UCS morphology is typically assessed through various methods: linear dimensions and angles,^{2,3,6,7,18,21,38,40,50-55} three-dimensional volume comparisons,^{5,6,17,51-53,56-61} and geometric morphometric analysis.^{8,62-64} This study is the first to incorporate all three methods to directly compare FOD with CS for treating UCS and examine operative morbidity. Our findings indicated that FOD achieves significantly better asymmetry correction than CS while also being less invasive.

A general concern for FOD when used to treat older patients was that distraction would show limited success due to decreased bone pliability. However, our examination of older patients (aged 16.6 and 13.8 months at the time of operation) confirmed this concern as unwarranted. In fact, these patients tended to exhibit overcompensation in ODA and ACFD upon distractor removal. Kamel et al⁴¹ reported a significantly longer surgical time with FOD, with no significant difference in transfusion, LOS, or postoperative complications. However, the authors also reported increased frequency of dural tear and contour relapse following FOAR. Additionally, Kim et al⁶⁰ reported a significantly shorter surgical time for FOD but an increased perioperative bleeding. Tahiri et al⁴³ reported shorter surgery time, less bleeding, and shorter LOS following FOD relative to FOAR, which agreed with the present findings. Villavisanis et al⁴² reported similar findings regarding the reduction of perioperative morbidity following FOD, along with a higher degree of asymmetry correction. McKee et al⁵ reported no significant difference between FOD and FOAR regarding correction of the orbital region. Furthermore, there were no reoperations within 30 days post-surgery in either group in this study, and no major complications or permanent sequelae were observed with FOD, although there was a higher incidence of suspected skin infection in this group.

One advantage of CS is that it allows a one-stage intervention with possible instant improvement in cranial symmetry while also addressing the contralateral protruding forehead, which FOD traditionally does not. However, complete forehead symmetry might not be the end goal, given that the contralateral forehead protrudes more than a normal infant's skull, suggesting that correction might result in a bilaterally protruding forehead. The downside of CS is its status as a static procedure that takes into consideration neither the natural expansion and growth of the calvaria nor the surrounding soft tissue, often leading to progressive relapse.^{18,21,54,65-68} To compensate for this, some surgeons attempt to estimate the extent of overcompensation.^{54,55,66,68-70}

We observed that FOD corrected ODA significantly better than CS, demonstrating the efficacy of distraction in the orbital region. Additionally, both ACFD and ACFC demonstrated greater degrees of improvement after FOD as compared with CS. Moreover, FOD resulted in smaller differences between NOAV and AOV, as well as similarities with volume and shape measurements in controls, which is in line with previous studies.^{5,51-54,56,57,61,62,71-73} At the 3-year follow-up, both orbital shape and volume remained similar to controls to a larger extent following FOD relative to CS. Furthermore, FOD resulted in greater postoperative improvements in overall symmetry according to PCA. Öwall et al⁷⁴ found that only one in 11 patients with minimal preoperative symmetry achieved facial symmetry comparable with that measured in a normal control after FOAR. By contrast, the patients in our cohort presenting preoperative asymmetry demonstrated improvements in ODA, ACFD, and ACFC following FOD (Table, Supplemental Digital Content 7 and 8). Although FOD is utilized in various centers worldwide, there are differences in surgical approach, including osteotomy placement, distraction regimes, and the number of distractors employed. Choi et al^{40,50,75} described a one-piece bilateral coronal osteotomy with two distractors that resulted in improved skull base symmetry but required a relatively long mean surgery duration (322 min). A recent study by Kim et al⁷⁶ compared the bilateral one-piece approach with a technique similar to that described in the present study. Their results showed that this method improved both morphology and operative morbidity (mean operation time: 168 min vs. to 89.5 min in the present study), and that it may be unnecessary to distract the unaffected suture. Notably, the technique described by Kim et al⁷⁶ differs from that employed in the present study by its use of two distractors and an osteotomy involving the naso-frontal suture. Additionally, the technique described by Taylor et al^{11,39,43} involved an osteotomy placed inferiorly along the orbital roof to facilitate horizontal expansion of the orbit, and they performed a contralateral vertical osteotomy at point along the transition of the forehead

deformity. The authors reported a mean operation time ranging from 111 min to 127 min along with a favorable perioperative bleeding profile. Brandel et al⁵⁹ employed a combination of anterior cranial vault reconstruction and release of a fronto-orbital bandeau with distraction osteogenesis. They reported more perioperative bleeding than that observed in the present study (mean 14.17 mL/kg vs. 6.1 mL/kg) but showed distraction osteogenesis as effective for volume expansion. At our center, we use a single continuous osteotomy line extending from the anterior fontanel to the lateral third of the orbital roof along with a single distractor. Despite the absence of additional osteotomies at the orbital rim in the present study, orbital symmetry developed favorably. In the literature, distraction lengths range from 18 mm to 30 mm. Our protocol includes a 30 mm distraction in all cases. However, given the slight undercorrection in facial symmetry observed in the current study, further investigation into the use of a 40 mm distractor would be valuable.

Limitations

This study has several limitations. The small sample size is notable but difficult to avoid, given the infrequency of UCS diagnosis (~8 children in Sweden annually). As a result, this cohort represents the majority of UCS patients among the Swedish population, given that our center handles ~80% of cases in Sweden. Additionally, because we compared FOD with CS as a remodeling technique, further studies comparing other FOAR variations would be highly relevant. Although the primary disadvantage of FOD is the requirement for distractor removal, Corkum et al⁷⁷ reported more unplanned reoperations following FOAR as compared with FOD.

Conclusion

The results of this case-control study showed that facial and skull symmetry were significantly better following FOD as compared with CS at the 3-year follow-up, with significant improvements observed in the orbital region. Additionally, FOD resulted in shorter

surgical time (89.5 vs. 153.5 min), less perioperative bleeding (6.1 vs. 22.1 mL/kg), and shorter LOS (4.3 vs. 5.8 days). Furthermore, FOD achieved superior cranial symmetry relative to CS while being significantly less invasive. Given the continuing debate regarding the optimal surgical approach to UCS, these findings could promote a definitive shift in pediatric surgery.

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Figure 1. Patient enrollment and follow-up.

Figure 2. Perioperative photographs and schematic illustrations of FOD (left) and CS (right).

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Figure 3. Changes in selected cranial indices based on linear dimensions and volumes.

(A) NI = nasal breadth / nasal height \times 100. (B) NCV. (C) AOI = affected orbital height / affected orbital width \times 100. (D) AOV. (E) NAOI = non-affected orbital height / non-affected orbital width \times 100. (F) NAOV. All regression curves (using linear or third-order polynomial functions) were computed based on the control dataset (n = 217, age: 0 to 48 months) and reported with 95% confidence intervals. See Figure, Supplemental Digital Content 3 for detailed information on additional calculated cranial indices and volumetric changes in cranial cavities.

Figure 4. PCA scatterplot (A) and density distribution plots (B) describing post-surgical asymmetry changes.

Surface shape variations along the PC1 axis are shown with yellow crania. Three individuals demonstrating extreme fluctuating asymmetry conditions (left and right), and normal asymmetry (along the 0 line) are shown in cyan. The dashed yellow line in the density plot represents the preoperative FOD group, while the dotted line represents the preoperative CS group. See Figure, Supplemental Digital Content 5 for the density distribution plots of each group and Table, Supplemental Digital Content 6 for all *P* values.

Figure, Supplemental Digital Content 1. Angle Measurements and Landmark Configuration

Used for Linear Dimensions. See Mellgren et al.² for details concerning the angle measurements. See Liang et al.⁴⁵ for the definition of the 88 anatomical landmarks.

Table, Supplemental Digital Content 2. Detailed patient information.

Figure, Supplemental Digital Content 3. Changes in additional cranial indices based on linear dimensions and volumes.

(A) CI = maximum cranial width / maximum cranial length \times 100. (B) UFI = upper facial height / bizygomatic breadth \times 100. (C) SWI = length of frontal and parietal margin of the greater sphenoid wing on the synostotic side / length of frontal and parietal margin of the greater sphenoid wing on the non-synostotic side \times 100. (D) ICV. (E) Absolute difference between AOV and NAOV (10% difference; red-dotted line). All regression curves (using linear or third-order polynomial functions) were computed based on the control dataset (n=217, age: 0 to 48 months) and reported with 95% confidence intervals.

Figure, Supplemental Digital Content 4. Post-surgical variations in head shape according to PCA. Left-lateral (above) and antero-superior (below) views. PC2 analysis revealed marginal changes that primarily affected the posterior portion of the neurocranium, with less impact on the facial complex.

Figure, Supplemental Digital Content 5. Density distribution plots describing post-surgical asymmetry changes.

Table, Supplemental Digital Content 6. *P* values (Wilcoxon signed-rank test) from group comparisons of asymmetry values.

Table, Supplemental Digital Content 7. Measurements of angles preoperatively and at the 3-year follow-up.

Table, Supplemental Digital Content 8. Measurements of angles in the FOD group.

Demographic data	FOD group	CS group	P^a
No. of cases	14	52	
Sex			
Female	7	32	0.640
Male	7	20	
Affected side			
Right	8	33	0.900
Left	6	19	
Operative and postoperative data			
Age at preoperative CT (months)	5.7 0.8–15.2	6.4 0.0–23.6	0.610
Age at operation (months)	8.0 4.3–16.6	8.7 4.6–25.5	0.370
Duration of operation (minutes)	89.5 63.0–121.0	153.5 60.0–209.0	< 0.001
Perioperative bleeding (mL/kg)	6.1 2.0–15.2	22.1 1.2–72.2	< 0.001
Perioperative transfusion (mL/kg)	3.3 0.0–14.0	14.3 0.0–36.2	< 0.001
Length of stay (days)	4.3 3.0–6.0	5.8 4.0–8.0	< 0.001
Age at distractor removal (months)	12.5 8.0–21.2	NA	NA
Duration of distraction (months)	4.5 2.6–6.2	NA	NA
Age at 3-year follow-up (months)	38.1 35.6–44.7	36.9 32.5–41.5	0.250
Comparison of measurements			
No. of cases	11	42	
Degrees changed (median ± IQR)			
ODA	5.7 ± 3.1	3.3 ± 3.8	0.008

ACFD	7.4 ± 4.9	5.5 ± 10.7	0.190
ACFC ^b	2.5 ± 3.6	1.6 ± 2.0	0.430

Operative and postoperative data are presented as the mean with range, and comparison of measurements are presented as median with IQR.

^aDemographic data was analyzed with the Chi-square test, operative and postoperative data and the comparison of FOD and CS were made with the Mann-Whitney *U* test.

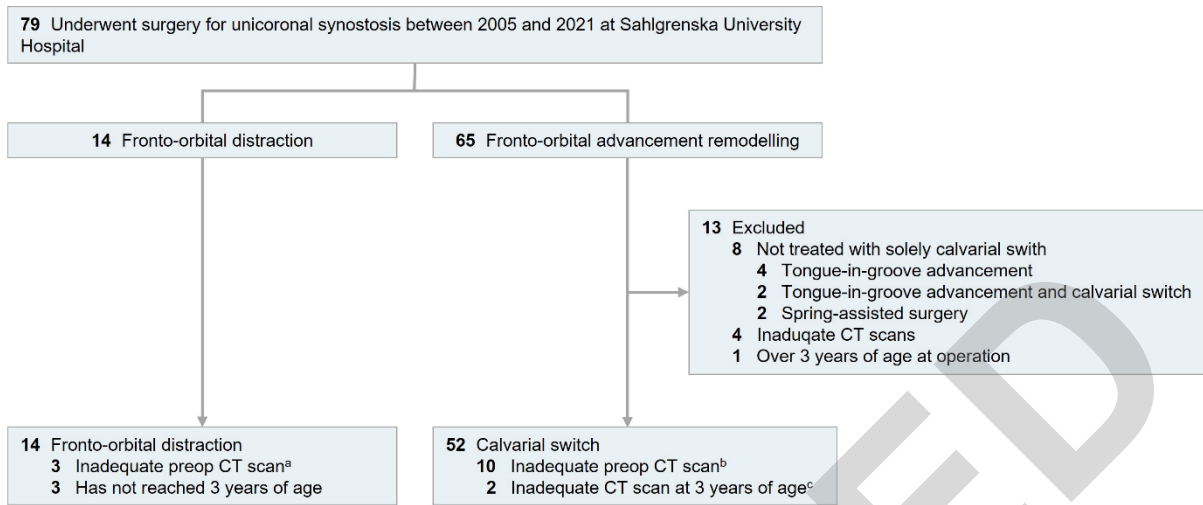
^bThe CT scan on one case (#61) did not capture the mastoid processes and was not included in the analysis.

Abbreviations: FOD, fronto-orbital distraction; CS, calvarial switch; ODA, orbital dystopia angle; ACFD, anterior cranial fossa deviation; ACFC, anterior cranial fossa cant; NA, not available.

Table 1. Patient information and measurements.

ACCEPTED

Figure 1



^aOne case had movement disturbances and two had incomplete CT scans.

^bEight cases had inadequate CT scans and two cases had irretrievable CT scans.

^cOne case had an incomplete CT scan and one had an irretrievable CT scan.

Figure 2

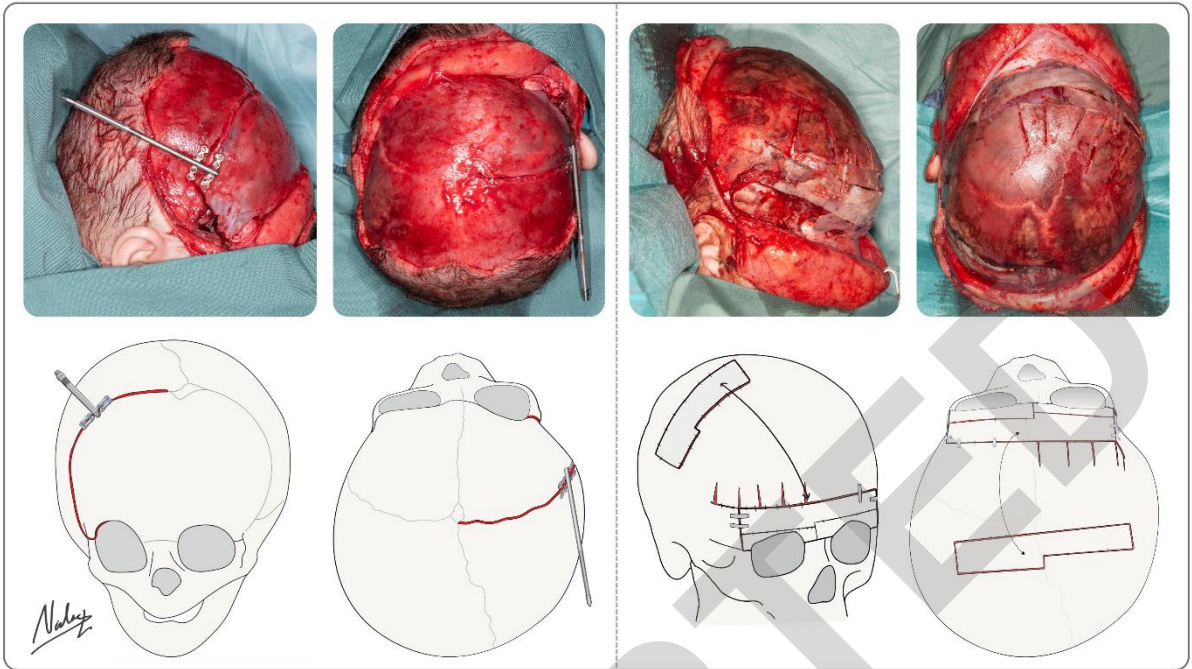


Figure 3

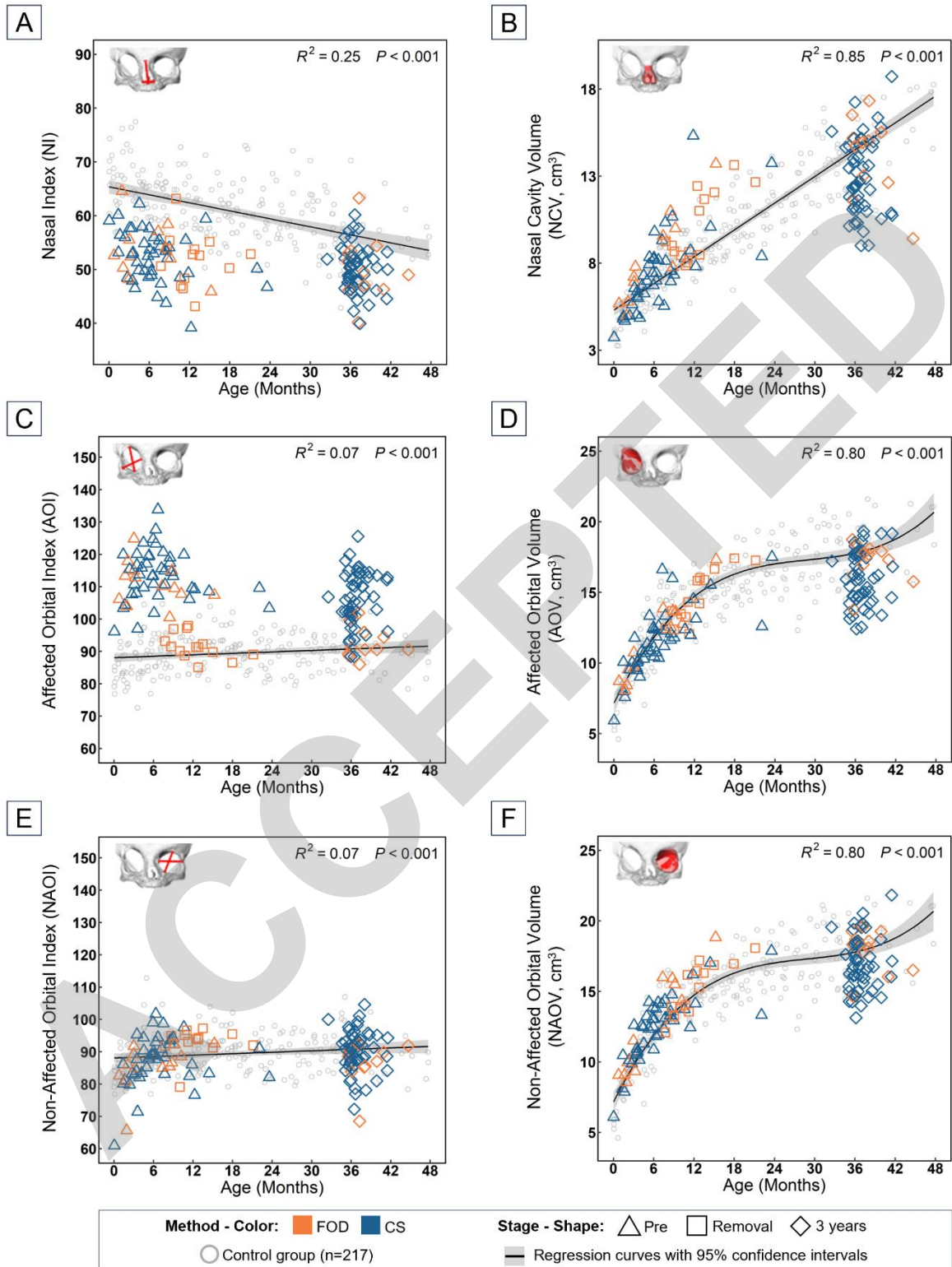
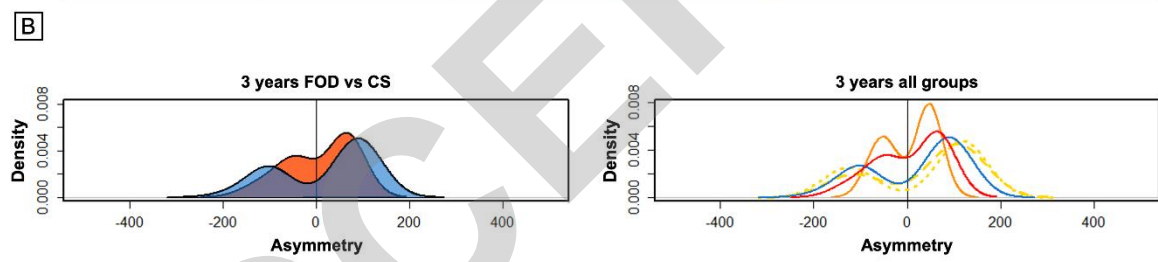
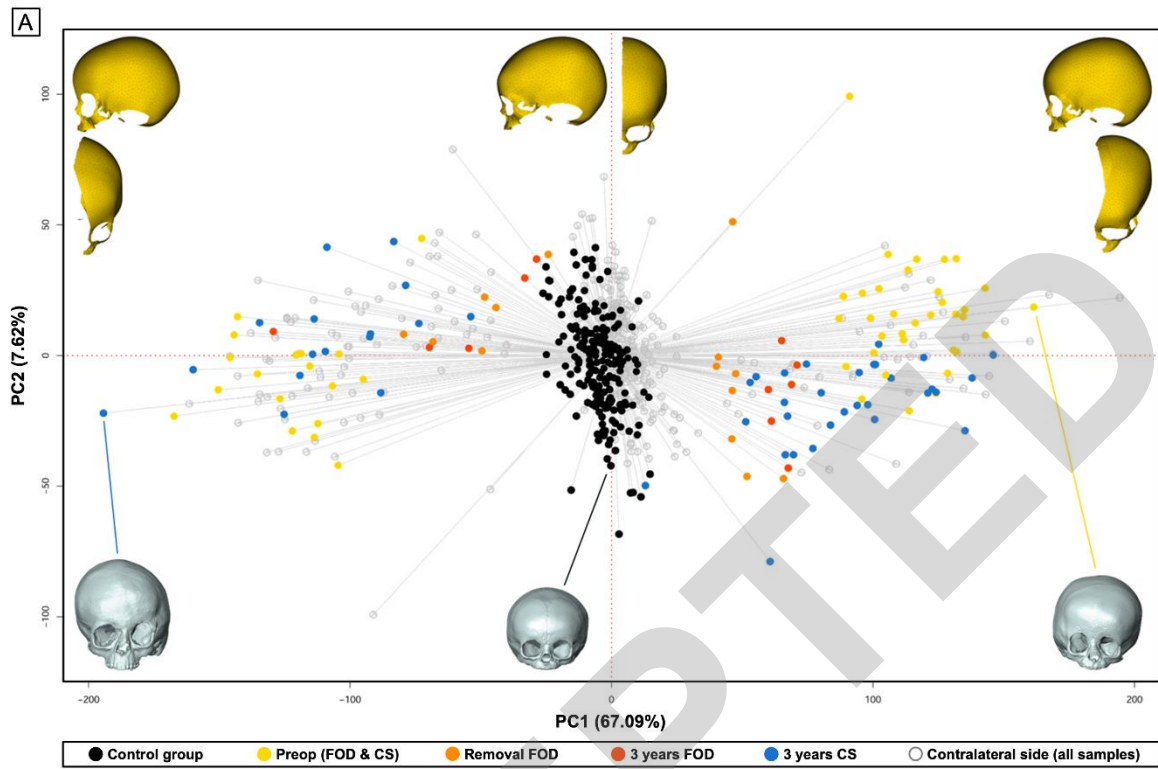
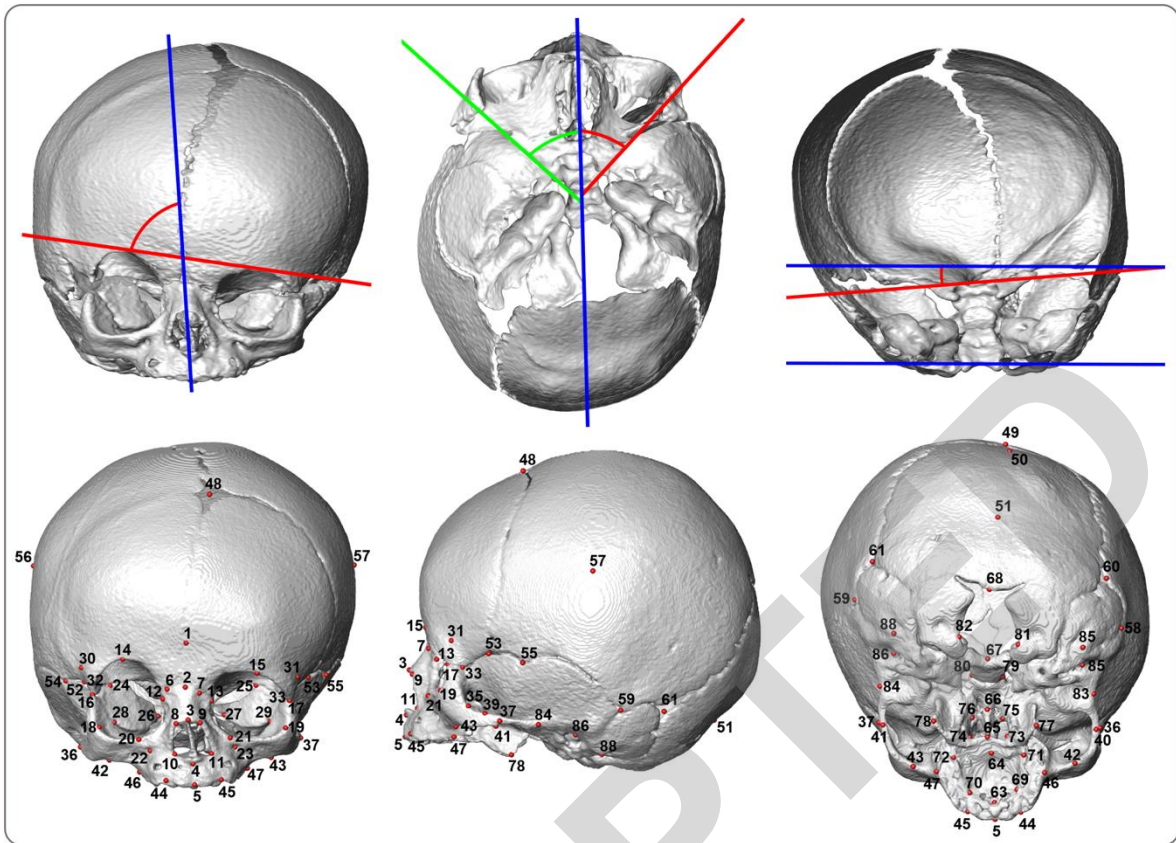


Figure 4





Figure, Supplemental Digital Content 1. Angle Measurements and Landmark Configuration Used for Linear Dimensions. See Mellgren et al.² for details concerning the angle measurements. See Liang et al.⁴⁵ for the definition of the 88 anatomical landmarks.

Table, Supplemental Digital Content 2. Detailed patient information.

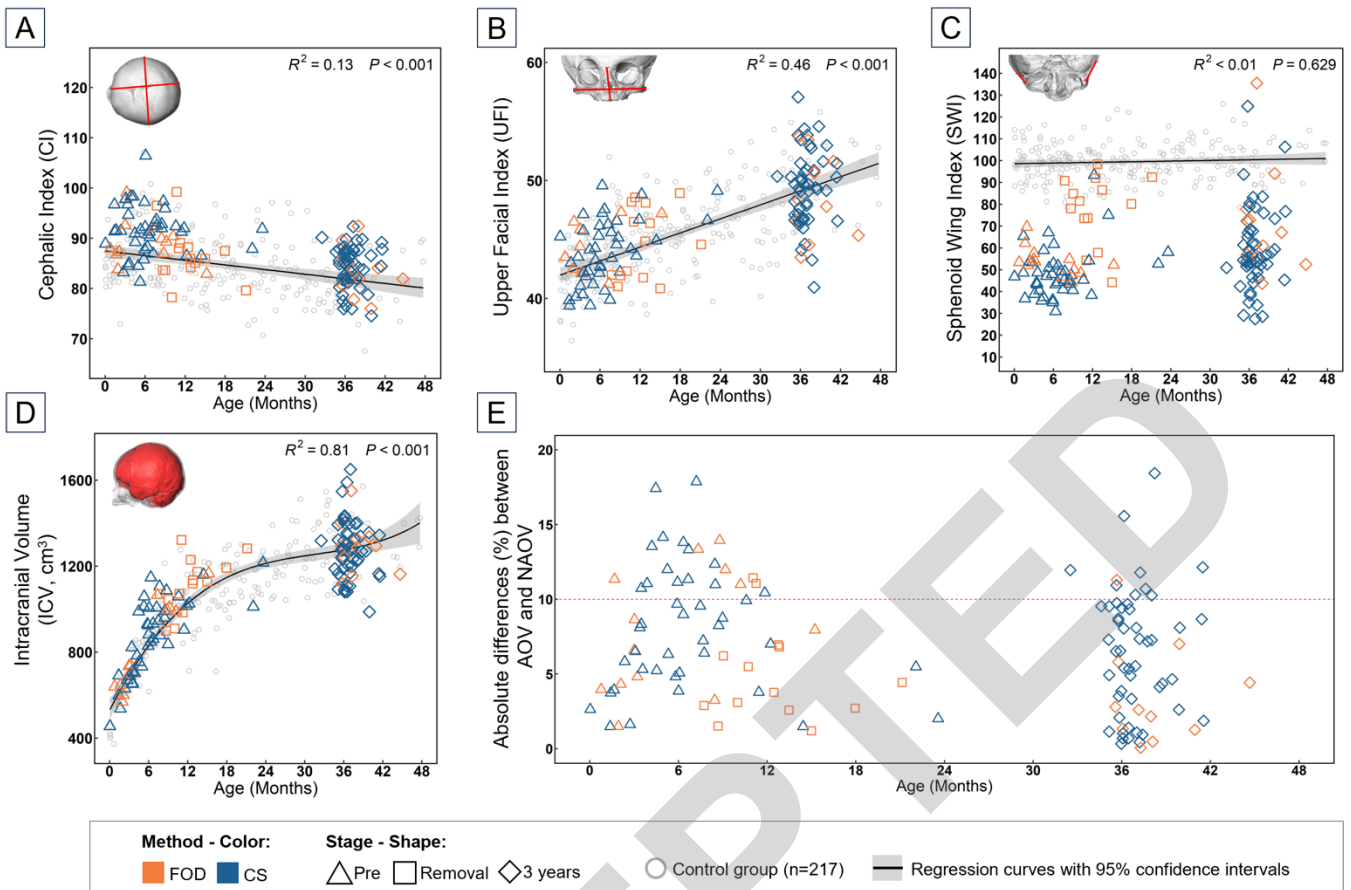
Case	Sex	Side	Age at preoperative CT (mo)	Age at operation (mo)	Duration of operation (min)	Perioperative bleeding (mL/kg)	Perioperative transfusion (mL/kg)	Length of stay (d)	Age at distractor removal (mo)	Duration of distraction (mo)	Age at 3-year follow up (mo)
CS group											
1	F	R	6.5	9.1	NA	39.4	NA	6	NA	NA	36.0
2	M	R	8.7	8.8	NA	12.2	NA	6	NA	NA	36.9
3	F	R	11.4	11.5	NA	18.8	NA	6	NA	NA	36.5
4	F	L	6.0	6.0	NA	30.0	NA	6	NA	NA	35.1
5	F	R	9.0	9.0	NA	31.7	NA	6	NA	NA	36.5
6	M	L	23.6	23.6	NA	25.4	NA	7	NA	NA	39.9
7	M	L	6.1	6.2	NA	27.8	NA	5	NA	NA	37.2
8	F	R	6.7	6.7	NA	16.3	NA	6	NA	NA	38.6
9	M	R	11.8	11.9	NA	27.8	NA	7	NA	NA	35.8
10	M	R	7.7	7.7	NA	29.9	NA	6	NA	NA	36.6
11	F	L	4.8	7.8	NA	48.8	NA	6	NA	NA	32.5
12	F	R	7.2	7.3	NA	22.3	NA	7	NA	NA	37.6
13	F	L	8.5	8.7	NA	39.0	NA	7	NA	NA	NA
14	M	R	12.2	13.2	NA	13.0	NA	6	NA	NA	35.6
15	F	R	7.5	9.3	NA	16.1	NA	6	NA	NA	37.3
16	F	R	5.3	8.7	NA	56.1	NA	6	NA	NA	36.0
17	F	R	1.8	4.6	NA	22.9	NA	7	NA	NA	38.0
18	M	L	5.3	6.6	NA	19.6	NA	7	NA	NA	41.5
19	F	L	3.5	6.0	NA	27.4	NA	5	NA	NA	35.2
20	F	R	3.5	5.8	NA	12.2	NA	8	NA	NA	41.5
21	M	L	10.6	11.7	NA	25.3	NA	5	NA	NA	35.2
22	F	L	6.7	6.8	NA	15.5	NA	5	NA	NA	38.0
23	M	R	4.2	6.0	NA	16.1	NA	8	NA	NA	35.8
24	M	R	10.9	14.4	NA	25.3	NA	5	NA	NA	35.7

Table, Supplemental Digital Content 2. Detailed patient information “(continued).”

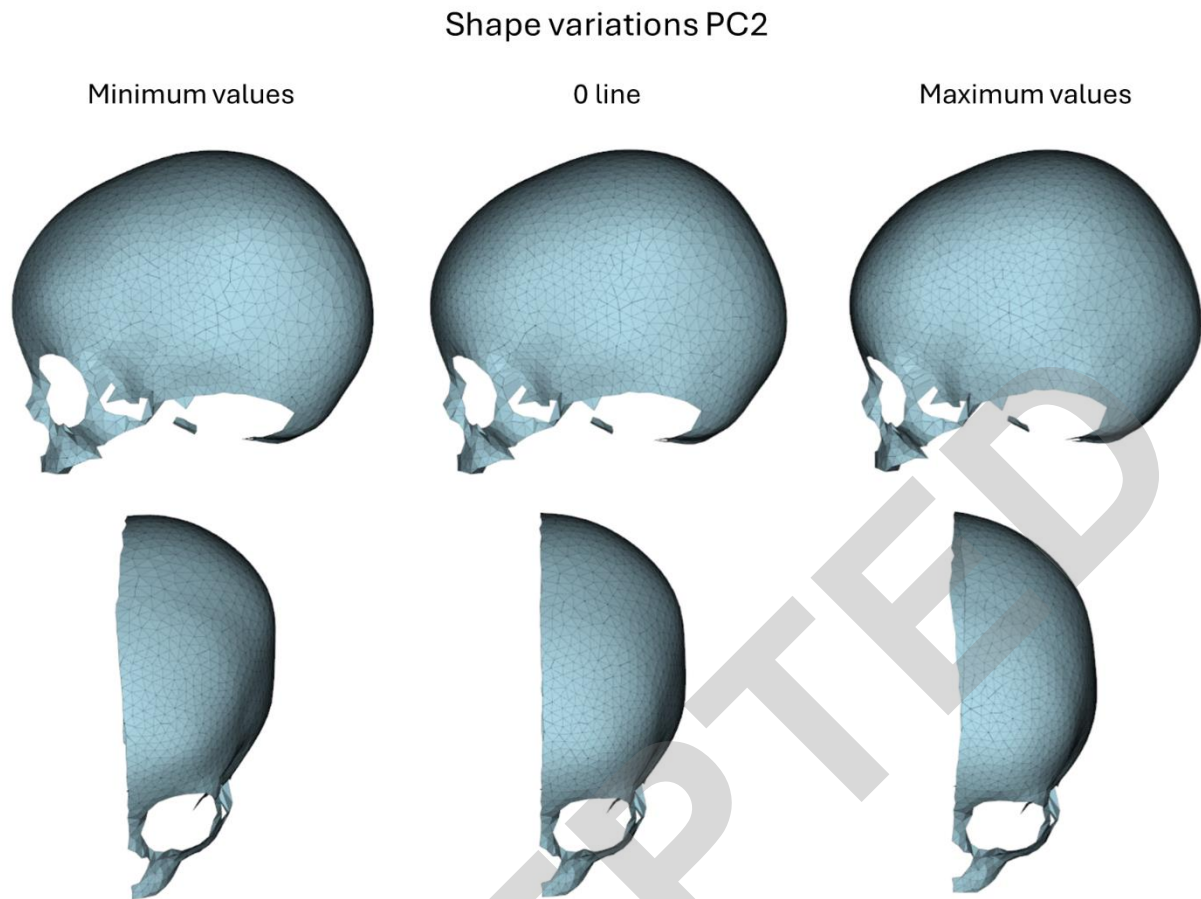
Case	Sex	Side	Age at preoperative CT (mo)	Age at operation (mo)	Duration of operation (min)	Perioperative bleeding (mL/kg)	Perioperative transfusion (mL/kg)	Length of stay (d)	Age at distractor removal (mo)	Duration of distraction (mo)	Age at 3-year follow up (mo)
CS group											
25	F	L	22.1	25.5	110	8.8	7.4	5	NA	NA	35.2
26	M	R	6.3	8.6	NA	11.9	NA	5	NA	NA	37.0
27	F	L	8.5	8.5	NA	40.5	NA	7	NA	NA	37.4
28	F	R	3.1	6.5	NA	72.2	NA	7	NA	NA	37.2
29	F	L	3.5	5.8	NA	25.6	NA	6	NA	NA	39.9
30	M	R	1.4	6.1	NA	41.3	NA	5	NA	NA	37.7
31	M	R	0.0	7.1	NA	24.7	NA	6	NA	NA	36.2
32	M	R	5.9	8.4	NA	11.4	NA	7	NA	NA	36.4
33	F	R	1.8	6.1	NA	16.4	NA	6	NA	NA	36.2
34	M	R	6.8	8.9	162	9.1	10.8	6	NA	NA	35.9
35	F	R	5.8	6.7	NA	16.0	NA	5	NA	NA	36.1
36	M	L	3.4	7.4	96	24.4	29.3	5	NA	NA	36.1
37	F	L	3.9	6.4	154	11.2	13.4	6	NA	NA	36.9
38	F	L	14.4	19.3	203	10.4	0.0	5	NA	NA	36.5
39	F	R	6.2	11.3	121	11.7	15.5	4	NA	NA	35.8
40	M	L	5.0	5.3	209	21.4	18.2	5	NA	NA	37.0
41	F	R	6.3	10.4	104	5.7	10.3	5	NA	NA	35.6
42	F	R	1.4	6.2	192	36.8	36.2	5	NA	NA	36.0
43	F	R	7.7	9.8	172	9.8	0.0	5	NA	NA	41.4
44	F	L	5.9	8.3	182	19.2	18.6	6	NA	NA	38.8
45	M	L	0.6	5.5	201	14.5	13.5	6	NA	NA	37.2
46	M	R	5.3	5.7	159	16.5	23.6	5	NA	NA	35.8
47	F	R	2.7	6.3	151	9.4	10.0	6	NA	NA	38.2
48	F	L	2.4	5.5	154	9.9	15.6	6	NA	NA	36.7

Table, Supplemental Digital Content 2. Detailed patient information “(continued).”

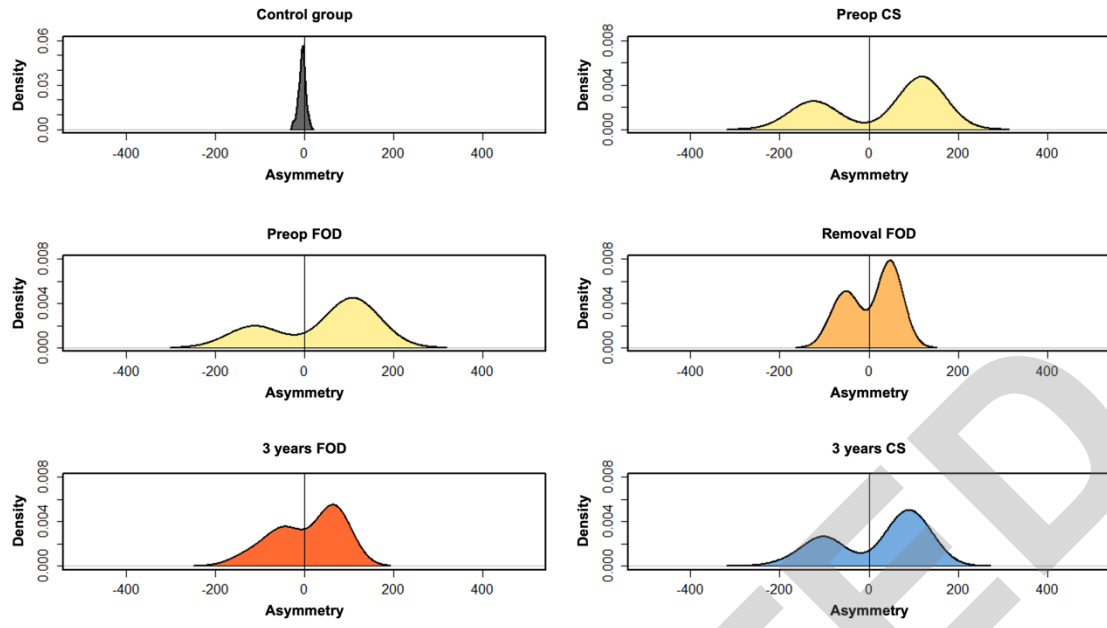
Case	Sex	Side	Age at preoperative CT (mo)	Age at operation (mo)	Duration of operation (min)	Perioperative bleeding (mL/kg)	Perioperative transfusion (mL/kg)	Length of stay (d)	Age at distractor removal (mo)	Duration of distraction (mo)	Age at 3-year follow up (mo)
CS group											
49	F	R	1.6	6.7	144	14.1	13.0	6	NA	NA	34.6
50	F	R	4.5	9.8	180	23.8	23.8	5	NA	NA	39.4
51	M	R	4.5	6.1	60	1.2	0.0	5	NA	NA	36.5
52	F	R	3.6	6.8	163	12.0	12.0	4	NA	NA	36.1
Mean			6.4	8.7	153.5	22.1	14.3	5.8			36.9
(range)			0.0–23.6	4.6–25.5	60.0–209.0	1.2–72.2	0.0–36.2	4.0–8.0			32.5–41.5
FOD group											
53	F	R	3.0	6.4	101	6.7	9.1	6	10.7	4.4	38.0
54	M	R	5.7	8.9	121	7.8	10.7	4	12.8	3.9	44.7
55	F	L	8.8	9.9	89	15.2	0.0	4	15.2	5.3	36.1
56	M	R	7.3	7.8	110	6.4	0.0	5	12.5	4.7	35.6
57	F	L	15.2	16.6	112	5.4	0.0	4	21.2	4.6	39.9
58	F	R	3.0	5.5	90	12.0	14.0	5	11.3	5.7	35.8
59	M	R	9.2	9.6	107	4.9	0.0	4	13.5	3.9	35.7
60	F	L	1.9	4.5	63	2.5	0.0	4	10.2	5.7	37.3
61	M	L	10.2	13.8	80	3.8	0.0	4	17.9	4.2	38.1
62	M	L	8.4	9.3	83	2.0	0.0	4	12.8	3.4	41.0
63	M	R	0.8	4.8	68	4.8	12.0	4	11.1	6.2	37.1
64	F	R	2.1	4.3	81	8.1	0.0	5	8.7	4.4	NA
65	M	R	3.2	5.4	66	3.5	0.0	4	8.0	2.6	NA
66	F	L	1.7	5.1	82	2.7	0.0	3	9.0	3.9	NA
Mean			5.7	8.0	89.5	6.1	3.3	4.3	12.5	4.5	38.1
(range)			0.8–15.2	4.3–16.6	63.0–121.0	2.0–15.2	0.0–14.0	3.0–6.0	8.0–21.2	2.6–6.2	35.6–44.7
Abbreviations: CS, calvarial switch; CT, computed tomography; FOD, fronto-orbital distraction.											



Figure, Supplemental Digital Content 3. Changes in additional cranial indices based on linear dimensions and volumes. (A) CI = maximum cranial width / maximum cranial length \times 100. (B) UFI = upper facial height / bizygomatic breadth \times 100. (C) SWI = length of frontal and parietal margin of the greater sphenoid wing on the synostotic side / length of frontal and parietal margin of the greater sphenoid wing on the non-synostotic side \times 100. (D) ICV. (E) Absolute difference between AOV and NAOV (10% difference; red-dotted line). All regression curves (using linear or third-order polynomial functions) were computed based on the control dataset (n=217, age: 0 to 48 months) and reported with 95% confidence intervals.



Figure, Supplemental Digital Content 4. Post-surgical variations in head shape according to PCA. Left-lateral (above) and antero-superior (below) views. PC2 analysis revealed marginal changes that primarily affected the posterior portion of the neurocranium, with less impact on the facial complex.



Figure, Supplemental Digital Content 5. Density distribution plots describing post-surgical asymmetry changes.

Table, Supplemental Digital Content 6. *P* values (Wilcoxon signed-rank test) from group comparisons of asymmetry values.

	Preoperative CS	3-year CS	Removal FOD	Preoperative FOD	3-year FOD
Control Group	0.001	0.001	0.001	0.001	0.001
Preoperative CS	NA	0.256	0.001	0.097	0.001
3-year CS	NA	NA	0.001	0.021	0.001
Removal FOD	NA	NA	NA	0.001	0.005
Preoperative FOD	NA	NA	NA	NA	0.029
3-year FOD	NA	NA	NA	NA	NA

Table, Supplemental Digital Content 7. Measurements of angles preoperatively and at the 3-year follow-up.

CS group	ODA				ACFD				ACFC			
	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)
1	NA	2.4	NA	NA	NA	11.6	NA	NA	NA	1.2	NA	NA
2	5.6	4.1	1.5	27.0	12.3	13.5	-1.2	-9.8	5.6	2.0	3.6	63.9
3	6.1	4.3	1.8	30.0	2.3	8.2	-5.9	-256.3	6.6	3.8	2.7	41.8
4	2.5	0.6	1.9	76.8	8.8	8.9	-0.1	-1.4	6.0	4.3	1.7	27.9
5	9.4	6.9	2.5	26.4	11.6	4.9	6.7	58.2	4.4	4.1	0.3	6.4
6	5.2	2.4	2.8	53.4	6.6	7.0	-0.3	-5.1	3.9	3.1	0.9	21.9
7	1.2	NA	NA	NA	18.7	NA	NA	NA	0.1	NA	NA	NA
8	8.3	8.2	0.0	0.6	15.2	5.1	10.1	66.7	2.8	-0.7	3.5	124.2
9	12.3	8.3	4.0	32.3	18.0	5.0	13.0	72.3	1.4	1.6	-0.2	-15.6
10	8.6	6.2	2.5	28.6	8.4	20.4	-12.0	-142.6	4.0	3.7	0.3	7.1
11	4.2	1.8	2.4	56.7	12.1	7.7	4.4	36.5	5.7	1.8	3.9	69.0
12	11.5	7.4	4.1	35.3	18.8	12.9	5.9	31.2	3.8	1.6	2.3	58.7
13	8.2	NA	NA	NA	11.5	NA	NA	NA	2.9	NA	NA	NA
14	12.4	6.1	6.3	51.0	26.0	8.6	17.4	66.8	4.5	2.9	1.6	35.6
15	7.3	4.2	3.2	43.2	21.8	4.0	17.8	81.7	1.2	1.4	-0.2	-15.7
16	13.7	8.4	5.3	38.8	2.6	9.2	-6.6	-254.7	8.3	6.8	1.5	18.0
17	NA	3.3	NA	NA	NA	17.2	NA	NA	NA	7.1	NA	NA
18	NA	4.4	NA	NA	NA	8.0	NA	NA	NA	4.2	NA	NA
19	NA	3.6	NA	NA	NA	23.3	NA	NA	NA	1.6	NA	NA
20	14.2	4.3	9.8	69.4	5.2	-5.2	10.4	201.0	1.0	-2.2	3.2	326.5
21	9.0	8.9	0.1	1.2	15.4	9.5	5.8	38.0	1.5	2.5	-1.0	-66.0
22	9.9	10.0	-0.1	-1.1	3.1	4.2	-1.1	-36.9	3.3	1.3	2.0	59.6
23	13.5	6.4	7.0	52.3	14.6	7.6	6.9	47.6	4.8	2.7	2.1	43.7
24	NA	2.2	NA	NA	NA	18.4	NA	NA	NA	-3.6	NA	NA
25	7.3	2.6	4.6	63.9	17.9	11.3	6.6	36.7	-0.4	0.6	-1.0	242.9
26	10.4	7.4	2.9	28.4	17.0	14.6	2.5	14.4	3.9	2.3	1.7	41.9
27	7.9	5.0	2.9	37.2	6.4	0.9	5.5	86.1	5.0	3.9	1.1	21.1
28	13.5	10.0	3.6	26.4	20.6	5.7	14.9	72.1	4.7	1.1	3.6	76.5
29	10.2	1.2	9.0	88.0	11.7	8.1	3.6	30.8	0.9	0.3	0.6	64.8

Table, Supplemental Digital Content 7. Measurements of angles preoperatively and at the 3-year follow-up “(continued).”

CS group	ODA				ACFD				ACFC			
	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)
30	22.6	6.3	16.3	72.1	11.6	3.7	7.9	68.3	3.8	2.9	0.9	24.2
31	9.7	21.5	-11.8	-122.3	20.2	17.2	3.1	15.1	4.8	6.5	-1.7	-34.5
32	11.0	7.1	4.0	35.9	25.5	13.6	11.9	46.6	1.2	0.3	0.9	72.0
33	NA	6.3	NA	NA	NA	5.2	NA	NA	NA	4.1	NA	NA
34	5.9	7.0	-1.1	-18.1	10.9	16.4	-5.6	-51.1	3.1	2.1	1.0	33.3
35	8.1	4.5	3.5	43.7	20.7	15.0	5.6	27.3	4.4	1.3	3.1	70.7
36	9.3	5.2	4.1	44.1	13.1	7.6	5.5	41.8	3.0	1.5	1.4	48.5
37	11.8	7.8	4.0	33.6	10.4	1.0	9.4	90.3	3.9	3.2	0.7	17.9
38	3.2	3.3	-0.1	-2.2	10.7	15.2	-4.5	-41.8	3.5	0.7	2.8	79.4
39	NA	4.8	NA	NA	NA	10.7	NA	NA	NA	1.0	NA	NA
40	8.3	3.1	5.2	62.2	8.8	2.2	6.7	75.5	3.5	3.2	0.4	10.8
41	11.7	9.3	2.4	20.8	10.9	8.2	2.8	25.5	2.4	1.3	1.1	43.9
42	14.5	9.1	5.4	37.4	16.2	5.9	10.3	63.7	5.4	3.6	1.8	33.5
43	12.3	5.6	6.7	54.4	14.1	8.3	5.8	41.0	3.1	1.1	1.9	63.0
44	15.6	-1.8	17.4	111.2	11.5	20.1	-8.6	-75.0	3.0	-0.6	3.6	118.2
45	NA	1.5	NA	NA	NA	6.7	NA	NA	NA	6.5	NA	NA
46	8.5	10.4	-1.9	-22.4	17.2	35.4	-18.2	-105.6	3.0	6.6	-3.6	-119.7
47	12.7	5.9	6.8	53.8	19.2	0.9	18.4	95.4	3.2	3.0	0.2	5.1
48	7.9	7.1	0.8	9.7	22.1	12.1	10.0	45.4	4.3	4.1	0.1	3.1
49	12.7	5.3	7.4	58.3	22.6	34.1	-11.5	-51.0	3.3	1.2	2.1	63.5
50	10.6	9.2	1.4	13.5	18.8	14.5	4.3	22.8	1.4	-0.6	2.1	144.8
51	8.7	5.3	3.4	38.8	17.7	12.8	4.9	27.8	5.7	2.0	3.7	64.8
52	6.2	8.8	-2.6	-41.0	2.3	11.2	-8.9	-393.8	4.0	1.7	2.3	56.5
Median	9.4	5.7	3.3	36.5	13.6	8.8	5.5	33.8	3.7	2.0	1.6	42.8
IQR	4.9	4.5	3.8	35.0	8.3	8.9	10.7	83.3	1.8	2.5	2.0	49.7
95% CI	8.0 : 10.8	4.5 : 7.2	2.3 : 4.5	25.8 : 48.6	11.4 : 16.9	6.4 : 11.4	2.4 : 8.6	7.5 : 65.4	3.1 : 4.5	1.3 : 2.7	0.9 : 2.3	27.4 : 59.9
P	< 0.001				0.009				< 0.001			

Table, Supplemental Digital Content 7. Measurements of angles preoperatively and at the 3-year follow-up “(continued).”

FOD group	ODA				ACFD				ACFC			
	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)	Preoperative ^a	3-year follow up ^a	Δ^b	$\Delta^{b,c,d}$ (%)
53	10.4	2.5	7.9	76.0	15.0	2.1	12.9	86.2	2.3	3.1	-0.8	-34.8
54	7.6	2.3	5.3	69.3	14.3	6.9	7.4	51.5	2.5	-1.3	3.8	152.0
55	9.8	0.4	9.4	95.6	6.5	-0.1	6.6	101.5	4.5	-2.0	6.5	144.4
56	8.3	6.1	2.2	26.1	16.9	7.0	9.9	58.4	2.3	0.3	2.0	88.4
57	3.9	-3.5	7.4	188.9	13.2	5.4	7.8	58.8	-0.5	-1.3	-0.8	-160.0
58	8.6	3.6	5.0	57.8	12.9	3.8	9.1	70.8	2.2	-0.7	2.9	131.8
59	9.5	5.8	3.7	39.3	14.8	5.7	9.1	61.5	4.6	4.4	0.2	3.6
60	10.1	5.3	4.8	47.3	8.2	6.6	1.6	19.0	2.4	-0.7	3.1	127.1
61	1.6	-5.6	7.2	450.0	6.6	-0.7	7.3	110.6	NA	3.2	NA	NA
62	6.2	0.5	5.7	91.9	6.4	4.4	2.0	31.3	4.6	5.3	-0.7	-15.2
63	11.7	-0.2	11.9	101.7	8.8	4.6	4.2	47.7	4.8	1.3	3.5	72.9
64	11.4	NA	NA	NA	10.1	NA	NA	NA	2.0	NA	NA	NA
65	12.8	NA	NA	NA	32.8	NA	NA	NA	3.1	NA	NA	NA
66	10.5	NA	NA	NA	10.0	NA	NA	NA	2.9	NA	NA	NA
Median	9.7	2.3	5.7	76.0	11.5	4.6	7.4	58.8	2.5	0.3	2.5	80.7
IQR	3.5	5.5	3.1	54.4	7.1	4.6	4.9	38.5	2.3	4.5	3.6	155.1
95% CI	7.0 : 12.6	-2.2 : 6.8	2.0 : 9.4	-87.0 : 238.9	9.2 : 16.6	1.6 : 7.6	3.0 : 11.7	23.6 : 94.1	1.5 : 3.5	-2.5 : 3.1	0.0 : 5.8	-35.3 : 212.1
P	0.003				0.003				0.025			

Angle measurements are presented in degrees.

^aNegative values indicate contralateral deviation.

^bNegative values indicate worsening.

^cValues >100 indicate overcorrection.

^dValues >200 indicate severe overreaction resulting in even greater deviation than preoperation.

Abbreviations: FOD, fronto-orbital distraction; CS, calvarial switch; ODA, orbital dystopia angle; ACFD, anterior cranial fossa deviation; ACFC, anterior cranial fossa cant; CI, confidence interval; IQR, interquartile range.

Table, Supplemental Digital Content 8. Measurements of angles in the FOD group.

FOD group	ODA			ACFD			ACFC		
	Preop	Removal ^a	3-year	Preop	Removal	3-year	Preop	Removal	3-year
53	10.4	3.0	2.5	15.0	1.0	2.1	2.3	2.0	3.1
54	7.6	4.4	2.3	14.3	10.0	6.9	2.5	-2.4	-1.3
55	9.8	0.7	0.4	6.5	4.7	-0.1	4.5	-0.1	-2.0
56	8.3	1.1	6.1	16.9	8.0	7.0	2.3	0.4	0.3
57	3.9	-3.8	-3.5	13.2	7.7	5.4	-0.5	-0.2	-1.3
58	8.6	1.7	3.6	12.9	5.5	3.8	2.2	0.7	-0.7
59	9.5	3.3	5.8	14.8	7.2	5.7	4.6	3.7	4.4
60	10.1	1.1	5.3	8.2	2.6	6.6	2.4	1.1	-0.7
61	1.6	0.2	-5.6	6.6	-5.8	-0.7	NA	4.7	3.2
62	6.2	-7.0	0.5	6.4	-1.0	4.4	4.6	4.5	5.3
63	11.7	1.5	-0.2	8.8	1.5	4.6	4.8	3.5	1.3
64	11.4	-3.9	NA	10.1	0.9	NA	2.0	1.6	NA
65	12.8	1.6	NA	32.8	2.6	NA	3.1	2.4	NA
66	10.5	0.4	NA	10.0	7.5	NA	2.9	1.7	NA
Median	9.7	1.1	2.3	11.5	3.7	4.6	2.5	1.7	0.3
IQR	3.5	2.8	5.5	7.1	6.6	4.6	2.3	3.3	4.5
95% CI	7.0 : 12.6	-1.5 : 3.7	-2.2 : 6.8	9.2 : 16.6	1.5 : 7.9	1.6 : 7.6	1.5 : 3.5	0.0 : 3.4	-2.5 : 3.1
P	< 0.001	0.420		< 0.001	0.660		0.003	0.110	

Angle measurements are presented in degrees.

^aNegative values indicate contralateral deviation.

Abbreviations: FOD, fronto-orbital distraction; ODA, orbital dystopia angle; ACFD, anterior cranial fossa deviation; ACFC, anterior cranial fossa cant; CI, confidence interval; IQR, interquartile range.