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TRABALHO FINAL
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Clínica Universitária de Neurocirurgia

Bilateral fronto-orbital advancement *vs* a two-step surgery to treat unicoronal craniosynostosis

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Abstract

Introduction: Unicoronal craniosynostosis (UCS) is a congenital craniofacial disorder characterized by the premature fusion of the coronal suture. It is a non-syndromic craniosynostosis, leading to retrusion of the forehead and orbital rim on the affected side and contralateral frontal bossing, orbital asymmetry and nasal deviation. Surgery is the standard treatment for UCS and aims the correction of the craniofacial scoliosis and the prevention of functional complications.

Objectives: To compare two surgical techniques for the treatment of UCS: a standard one-stage surgery with bilateral fronto-orbital advancement (FOA) versus a two-stage surgery including early distraction by springs, followed by bilateral FOA.

Methods: We retrospectively reviewed the clinical files of pediatric patients (< 18 years old) who underwent surgical correction for UCS at Centro Hospitalar Universitário Lisboa Norte (CHULN*)* between January 2012 and December 2021. We compared the outcomes of two surgical procedures using pre- and post-surgery computed tomography (CT). The specific craniometric measurements used were skull base twist, midface twist and orbital ratios.

Results: Nine out of 23 children who underwent UCS surgery at CHULN met the inclusion criteria for the study. Four children were submitted to one-stage surgery, while five children underwent a two-stage surgery. In the one-stage surgery group, the mean skull base twist changed from 12,0 to 10,48 degrees, and the mean midface twist changed from 12,33 degrees to 9,58. Children treated with a two-stage surgery had a more significant improvement in craniofacial deviation, even after the first procedure with spring-mediated distraction. Mean skull-base twist changed from 10,96 to 6,64 degrees and mean midface twist changed from 12,52 degrees to 4,76. Orbital height and width ratios did not significantly change after surgery.

Conclusion: Our study demonstrates that children who have an early diagnosis of UCS and are treated with a two-stage surgery achieve greater improvement of craniofacial scoliosis.

Keywords: Unicoronal craniosynostosis; fronto-orbital advancement; spring mediated distraction; midface twist; skull base twist; orbital ratios.

This Final Paper expresses the opinion of the author and not of the FML.

Resumo

Introdução: A craniossinostose coronal unilateral (CCU) é uma patologia craniofacial congénita caracterizada pela fusão prematura da sutura coronal. É uma craniossinostose não sindrómica, levando à retração da testa e do bordo orbitário no lado afetado e à protusão frontal contralateral, assimetria orbitária e desvio da pirâmide nasal. A cirurgia é o tratamento padrão e visa a correção da escoliose craniofacial.

Objetivos: Comparar duas técnicas cirúrgicas para o tratamento da CCU: uma cirurgia única com avanço fronto-orbitário bilateral *versus* uma cirurgia em dois tempos incluindo distração precoce com molas seguida de avanço fronto-orbitário.

Métodos: Analisamos retrospetivamente os processos clínicos de doentes pediátricos submetidos a correção cirúrgica de CCU no Centro Hospitalar Universitário Lisboa Norte (CHULN) entre Janeiro de 2012 e Dezembro de 2021. Comparámos os resultados de dois procedimentos cirúrgicos utilizando tomografias computorizadas antes e depois da cirurgia. As medidas craniométricas específicas utilizadas foram o desvio da base do crânio, desvio da face e rácios orbitários.

Resultados: Nove das 23 crianças submetidas a cirurgia no CHULN cumpriram os critérios de inclusão. Quatro crianças foram submetidas a cirurgia única e cinco a uma cirurgia em dois tempos. No grupo da cirurgia única, o desvio médio da base do crânio mudou de 12,0 para 10,48 graus, e o desvio médio da face alterou de 12,33 graus para 9,58 graus. As crianças tratadas com uma cirurgia em dois tempos obtiveram uma maior redução no desvio craniofacial, mesmo após o primeiro procedimento com distração mediada por molas. O desvio médio da base do crânio mudou de 10,96 para 6,64 graus e o desvio médio da face alterou de 12,52 para 4,76 graus. Os rácios de altura e largura orbitárias não alteraram significativamente após as cirurgias.

Conclusão: O estudo demonstra que crianças com diagnóstico precoce de CCU e tratadas com uma cirurgia em dois tempos conseguem uma maior redução da escoliose craniofacial.

Palavras-chave: Craniossinostose coronal unilateral; avanço fronto-orbitário; distração mediada por molas; desvio da face; desvio da base do crânio; rácios orbitários.

O Trabalho Final exprime a opinião do autor e não da FML.

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List of abbreviations:

- UCS: Unicoronal craniosynostosis

- FOA: Fronto-orbital advancement

- CHULN: Centro Hospitalar Universitário Lisboa Norte

- SBT: Skull base twist

- MFT: Midface twist

- OWR: Orbital width ratio

- OHR: Orbital height ratio

Introduction

Craniosynostosis is a congenital disorder characterized by premature fusion of the cranial sutures. Simple craniosynostosis is a term used when only one suture fuses prematurely. Complex craniosynostosis describe the premature fusion of multiple sutures (Kronig et al., 2021; Yang et al., 2018). When children with craniosynostosis, usually complex, also display other body malformations, this is termed syndromic craniosynostosis, frequently associated with Crouzon syndrome, Apert syndrome, Muenke syndrome and others (Wilkie et al., 2017).

Since the skull stops growing in the areas where the sutures have fused, there is overgrowth of other areas. The early fusion of the cranial sutures causes not only an abnormal growth of the skull, leading to cosmetic deformities, but may also compromise the normal development of the brain (Kronig et al., 2021; Proctor & Meara, 2019; Yang et al., 2018). In more severe cases, it can lead to increased intracranial pressure, seizures and development delay.

There are different types of craniosynostosis, according to the suture that is affected (**Figure 1**). The most common is sagittal suture craniosynostosis, followed by coronal suture craniosynostosis.

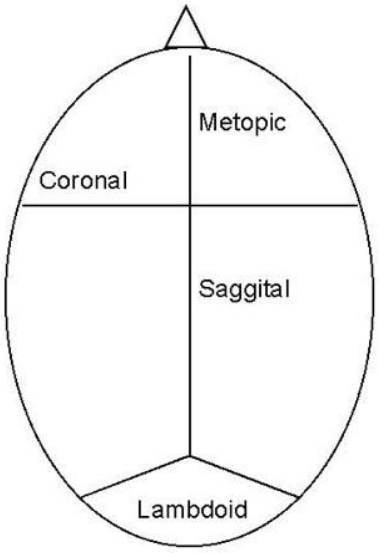


Figure 1. Diagram of a neonate's skull demonstrating the location of the sutures.

This work focuses on one specific type of craniosynostosis: unicoronal craniosynostosis (UCS). This disorder affects 1 in 10 000 births and leads to ipsilateral supraorbital retrusion, elevation of the supraorbital rim, flattening of the frontal bone, and contralateral compensatory frontal bossing (Beckett et al., 2013; Proctor & Meara, 2019). The protrusion on the unaffected side can also cause the eyelid to look as if it is hanging downwards and nasal root deviation to this side (Liu et al., 2019; Wes et al., 2017). The craniofacial scoliosis that occurs, can affect the vault, the face and the cranial base.

Surgery is the standard treatment for unicoronal craniosynostosis, aiming to achieve long-standing correction of the craniofacial asymmetry and to prevent future functional complications (Bernhardt et al., 2013; Liu et al., 2019; Mazzaferro et al., 2017). The most common treatment for UCS is one-stage surgery including fronto-orbital advancement (FOA) and remodeling of the anterior cranial vault (Liu et al., 2019). With this technique, the forehead and orbital deformity can be corrected by repositioning and remodeling the fronto-orbital bandeau (Kronig et al., 2021). It is believed that by releasing the synostosis one allows the natural correction of the nasal root deviation.

There are several methods described in the literature to evaluate the surgical outcomes after the treatment of UCS. Morphological analysis of the skull base has been used to compare deformation of the cranium before and after FOA (Liu et al., 2019; Yang et al., 2018).Pre- and postsurgical soft-tissue facial asymmetry can be analyzed using a computerized method capable of objective and spatial detailed quantification in 3-dimension (transverse, vertical, and sagittal directions), focusing on the facial region and 6 subregions (forehead, mouth, eyes, nose, cheek and chin) (Öwall et al., 2019). Although UCS surgical correction does not address nasal root deviation directly, it can be used as an indicator of facial symmetry, since this feature improves significantly over time (Wes et al., 2017). Volumetric analysis was also reported to assess symmetry of the cranium by measuring the volume of anterior, middle and posterior cranial fossa, and comparing the synostotic side to the contralateral side (Mazzaferro et al., 2017).

The craniofacial team at Centro Hospitalar Universitário Lisboa Norte (CHULN), including surgeons from the Neurosurgery Department and from the Plastic Surgery Department, has a long experience in treating children with craniosynostosis. For many years, the standard surgical procedure to treat UCS was a one-stage procedure with bilateral FOA performed after 12 months of age (Guimarães-Ferreira et al., 2004). Since 2012, a two-stage approach was developed which included suturectomy of the fused coronal suture with placement of one or two springs (**Figure 2**), in the first 6 months of age, followed by bilateral FOA around 18 months of age. This two-stage surgical approach for UCS treatment has not yet been described in the literature and the objective quantification of the surgical outcome has not been assessed.

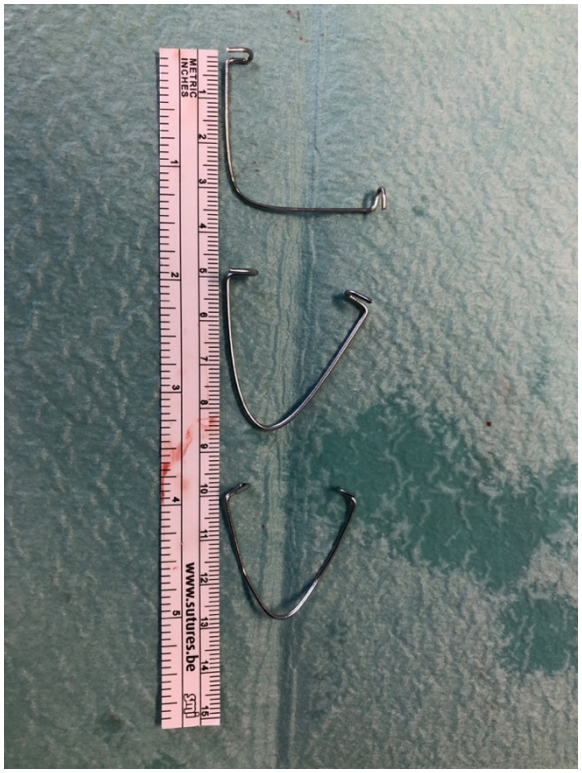


Figure 2. Three springs of different size

Objectives

The main goal of this work is to compare the outcomes of two different surgical approaches to treat UCS: a standard one-stage surgery with bilateral FOA after 12 months versus a two-stage surgery including suturectomy and springs placement before 6 months of age, followed by bilateral FOA at 18 months of age.

Methods

Study population

A retrospective review of all pediatric patients (< 18 years old) who underwent surgical correction for UCS at Centro Hospitalar Universitário Lisboa Norte (CHULN) between January 2012 and December 2021 was conducted.

We collected the following information: patient age and sex, side of affected suture, date and type of surgery and imaging data. The inclusion in the study required a pre- and post-surgery computed tomography (CT) of the head. The total number of children that were submitted to surgery during this period of time was 23, but only 9 children fulfilled the eligibility criteria (**Figure 3**).

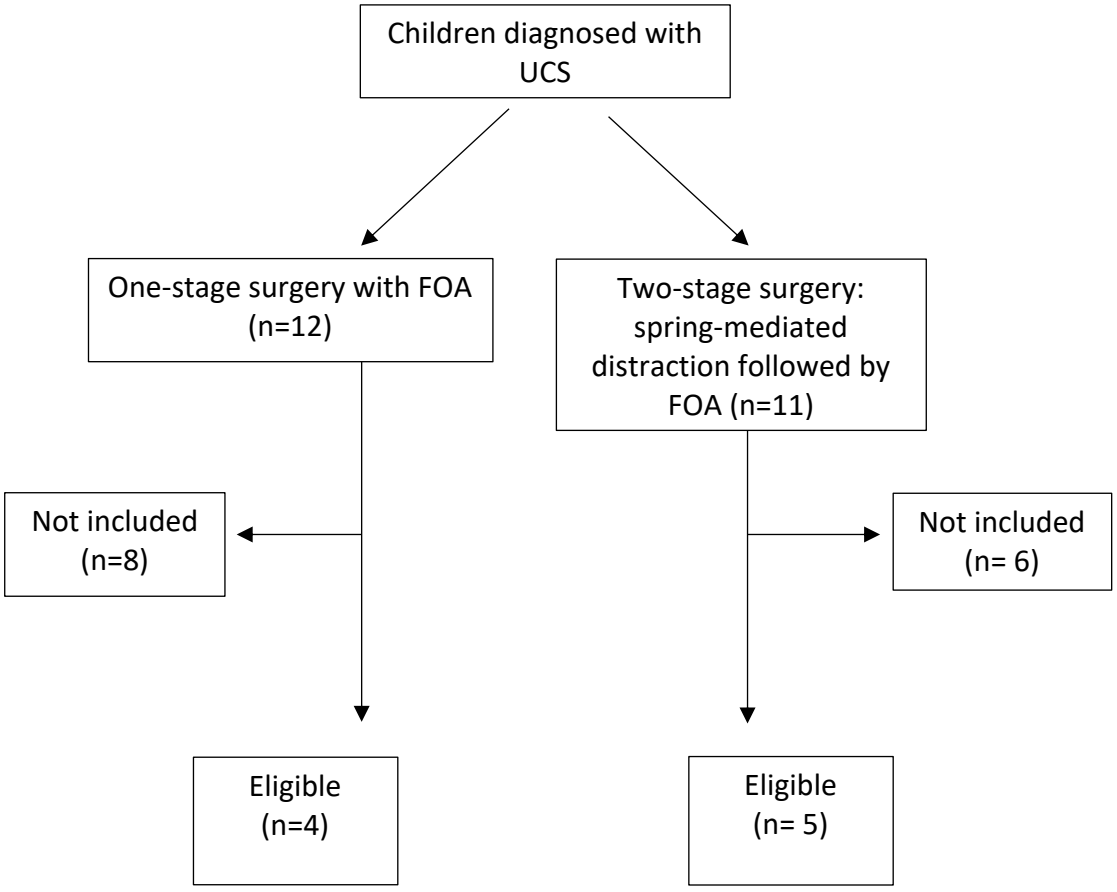


Figure 3. Flowchart representing the patients included and excluded in the study

Two patients had the first surgery in 2021 and are waiting for the second procedure to remove the springs and to perform the FOA. Two children recently submitted to a FOA are waiting for the post-operative head CT scan. Ten children had performed pre-operative scans outside the CHULN and the exams were not available for evaluation.

Operative Technique

One-stage surgery: fronto-orbital advancement

A bicoronal zigzag incision was performed to expose the skull up to the supraorbital bar. The supraorbital bar was removed, remodeled to become symmetric and repositioned, in order to be more advanced and lowered on the fused side. To obtain a forehead symmetric and with a nice contour, bone from the temporo-parietal region was transplanted to the appropriate position, over the supraorbital bar (**Figure 4**) (Guimarães-Ferreira et al., 2004; Maltese et al., 2014; Taylor et al., 2015).

Figure 4. Bilateral fronto-orbital advancement in a left unicoronal craniosynostosis. (A) Frontal view and (B) Right lateral view.

Two-stage surgery: spring mediated distraction followed by fronto-orbital advancement

The first stage of the surgery was performed when children had between 3 to 6 months of age. It consisted in the opening of the closed suture (suturectomy) and placing one or two costume made springs, aiming at promoting bone distraction over the next months (**Figure 5**). The second stage of the surgery was performed when children were around 18 months of age and consisted of springs' removal followed by a FOA, as described above.

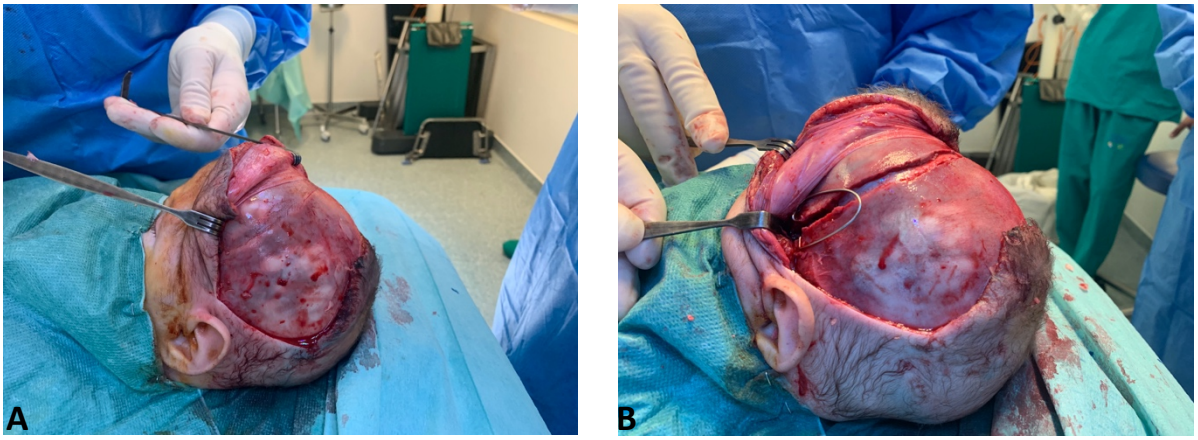


Figure 5. Left coronal synostosis (A) treated with suturectomy and placement of a spring (B).

Craniometric Analysis

To determine the craniofacial symmetry before and after one-stage and two-stage surgeries, we have used specific craniometric measurements (skull base twist, midface twist and orbital ratios) using the bone window of 3D CT scans, as previously published by Mengyuan T. Liu, *et al*.

Skull Base Twist

The skull base twist was measured on an axial section of the CT scan that crossed the petrous part of the temporal bone. Two lines were drawn: one from the opisthion until the most anterior point of the sella turca and another between the sella turca and the nasion (**Figure 6**). The angle made by these two lines was measured and called *skull base twist* (SBT) (Liu et al., 2019).

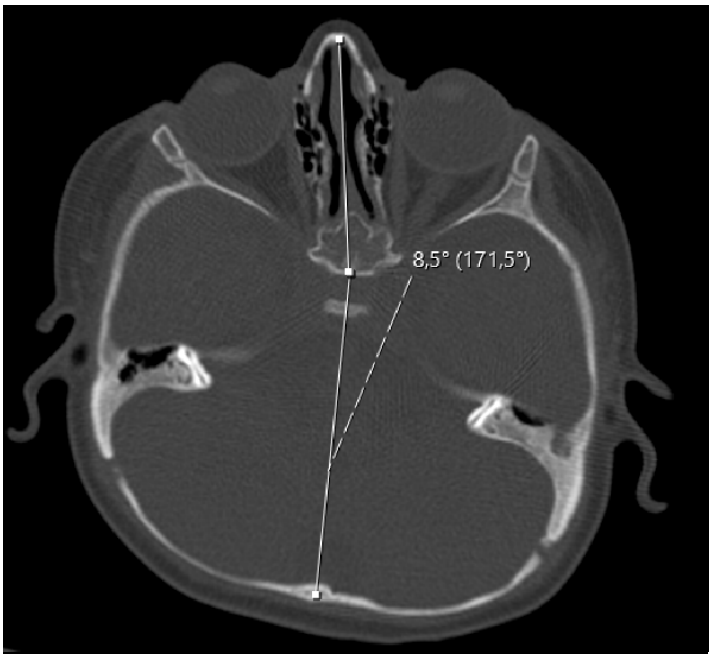


Figure 6. Example of a skull base twist measurement

Midface Twist

The midface twist was determined using a coronal section including the nasal septum. A line was drawn from the most inferior to the most superior point of the nasal septum and another from the latter until the metopic suture (**Figure 7**). This angle was measured and name *midface twist* (MFT) (Liu et al., 2019).

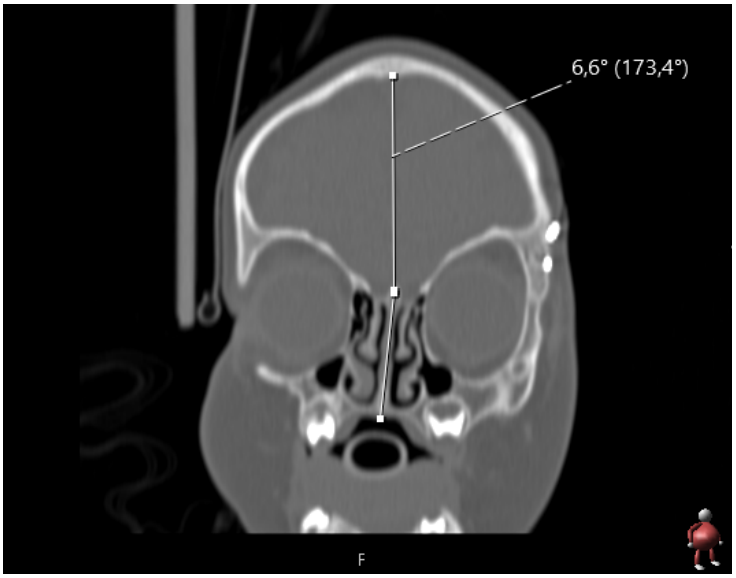
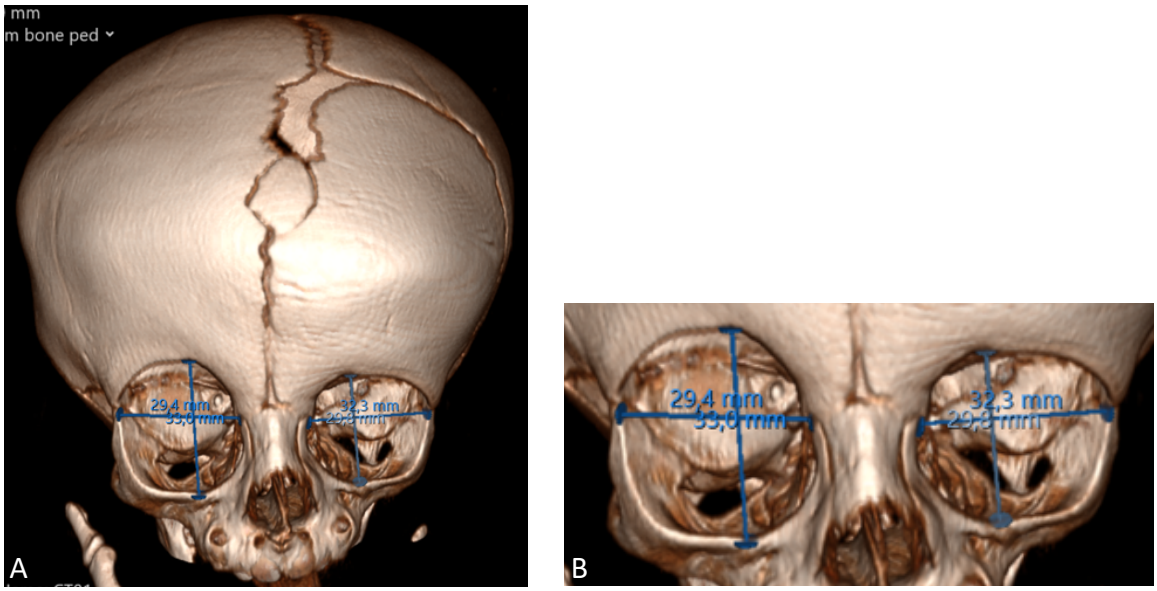


Figure 7. Example of a midface twist measurement

Orbital Ratios

To determine the symmetry of the orbits we have assessed the height ratio and the width ratio.

The orbital height ratio (OHR) was calculated by the distance from the supraorbital notch to the zygomaticomaxillary suture of the orbit on the affected side, over the orbit on the contralateral side (unaffected). For the orbital width ratio (OWR), the same logic was applied, but measuring the distance from the dacryon to the zygomaticofrontal suture (**Figure 8**). Thus, the closer the ratio is to 1, the more symmetric the height or width of the orbits (Liu et al., 2019) .



Figures 8. Example of orbital height and width measurements.

Statistical Analysis

The measurements were made manually using the imaging computer system at the hospital (CHULN).

In the case of the four children who underwent only FOA, measurements of the previously mentioned vectors and distances were made on the scans taken before and after surgery and a comparison was made in each individual case, establishing an average for each value.

In the case of the five children operated in two stages (placement of springs and FOA afterwards), the measurements were made at three different times (before surgery, between the first and second surgery and after the second surgery), making an individual comparison of the evolution of each case and a group average value.

Results

Twenty-three children underwent UCS correction at CHULN between January 2012 and December 2021, but only nine met the inclusion criteria of having pre- and post-surgery head CT scans.

Four children, two boys and two girls, were submitted to one-stage surgery with FOA between the ages of 9 months and 15 months (mean age = 12,4 months). These children were born between 2012 and 2015 and were diagnosed with a right UCS.

On the other hand, five children (two girls and three boys) underwent a two-stage surgery. They were born between 2016 and 2020 and they were aged between 5 and 8 months (mean age = 6,1 months) by the time of the springs' implantation. Three had a left UCS, while two had a right suture fused.

Skull Base and Midface Twists

Tables 1 and **2** summarize the pre-and post-operative degree of skull base twist and midface twist, in patients treated with one-stage surgery and two-stage surgery, respectively.

Table 1 – Skull base twist and midface twist in patients treated with one-stage surgery (fronto-orbital advancement)

	Skull base twist (degree)	Midface twist (degree)
Preoperative	12,0 (8,3 – 13,7)	12,33 (8,0 – 17,6)
Postoperative (after FOA)	10,48 (7,7 – 14,3)	9,58 (5,1 – 13,5)

*values are averages of a total of 4 patients (range)

Table 2 - Skull base twist and midface twist in patients treated with a two-stage surgery

	Skull base twist (degree)	Midface twist (degree)
Preoperative	10, 96 (7,7 – 13,7)	12,52 (6,7 – 18,5)
Postoperative 1 (after springs)	7,06 (4,1 – 10,1)	5,38 (2,0 – 7,0)
Postoperative 2 (after FOA)	6,64 (3,7 – 9,3)	4,76 (1,5 - 7,8)

*values are averages of a total of 5 patients (range)

The preoperative skull base twist in the two surgical groups was identical and above 10 degrees on average (**Tables 1** and **2**). The postoperative evaluation of the skull base twist revealed there was always an improvement after each surgical intervention, but with different impacts. In children who underwent one-stage surgery with FOA, there was a decrease in skull base deviation by 1.52 degrees. A much greater improvement in deviation occurred in children who underwent a two-stage surgery, with a decrease in deviation of the skull base by more than 4 degrees, after the complete treatment. It is worth mentioning that even after springs-mediated distraction, the correction was already superior to the one-stage FOA. An example of the evolution of the skull base deviation after each surgical procedure is demonstrated in **Figure 9**.

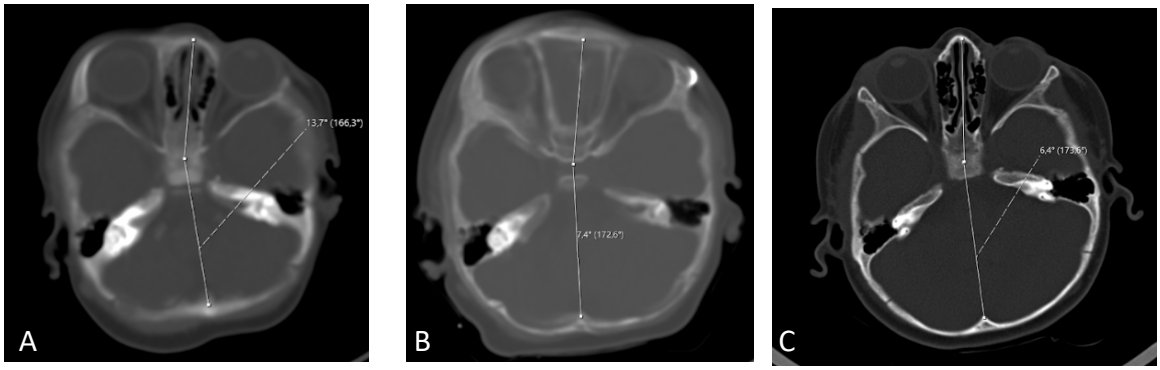


Figure 9. Example of skull base twist (SBT) measurement in a two-stage surgery.
A – Before surgical intervention (SBT = 13,7 degrees)
B – After suturectomy and implantation of springs (SBT = 7,4 degrees)
C – After FOA (SBT = 6,4 degrees)

For the midface twist, the differences in both surgical groups were even greater (**Tables 1** and **2**). Both groups showed a similar degree of midface twist before the interventions (above 12 degrees). The postoperative evaluation showed a significant improvement with the two-stage surgery (decrease in deviation of 7.76 degrees), when compared to the one-stage surgery (deviation decrease in 2.75 degrees). An example of the evolution of the midface deviation after each surgical procedure is demonstrated in **Figure 10**.

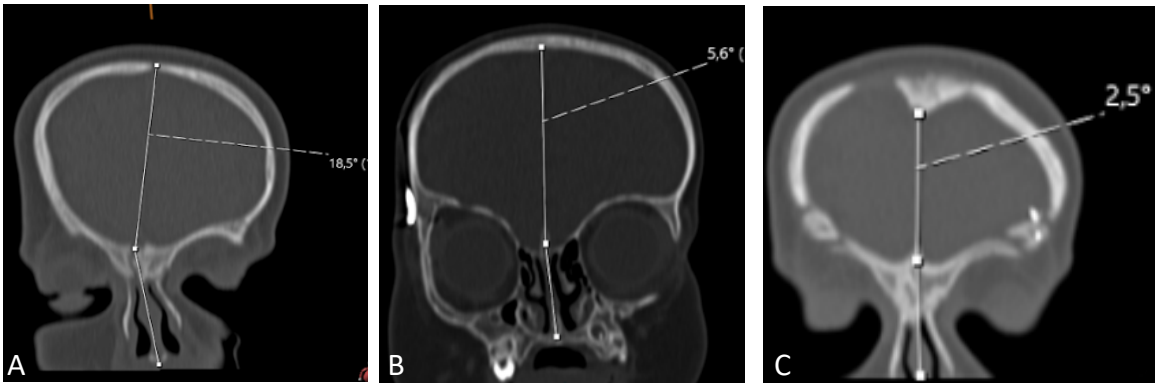


Figure 10. Example of midface twist (MFT) measurement on a two stage-surgery.
A – Before surgical intervention (MFT = 18,5 degrees)
B – After suturectomy and implantation of springs (MFT = 5,6 degrees)
C – After FOA (MFT = 2,5 degrees)

Orbital height and orbital width ratios

Tables 3 and **4** summarize the pre-and post-operative orbital height ratio and orbital width ratio, in patients treated with one-stage surgery and two-stage surgery, respectively.

Table 3 – Orbital Height Ratio and Orbital Width Ratio in patients submitted to one-stage surgery with FOA

	Orbital Height Ratio (OHR)	Orbital Width Ratio (OWR)
Preoperative	1,08 (1,01 – 1,14)	0,92 (0,85 – 0,93)
Postoperative	1,16 (1,08 – 1,35)	0,9 (0,85 – 1)

*values are averages of a total of 4 patients (range)

Table 4 **-** Orbital Height Ratio and Orbital Width Ratio in patients submitted to a two-stage surgery

	Orbital Height Ratio (OHR)	Orbital Width Ratio (OWR)
Preoperative	1,08 (1,02 – 1,12)	0,91 (0,85 – 1,07)
Postoperative 1 (after springs)	1,04 (0,98 – 1,11)	0,9 (0,86 – 0,96)
Postoperative 2 (after FOA)	1,04 (0,96 – 1,12)	0,91 (0,87 – 0,93)

*values are averages of a total of 5 patients (range)

Since OHR is calculated dividing the orbital height on the affected side by the contralateral orbital height, it is possible to notice that the height of the orbit on the affected side is consistently greater than on the contralateral side, even after surgery (OHR >1). The opposite happens for orbital width, since OWR is constantly under 1.

The group of children submitted to one-stage surgery showed an increase in asymmetry of the height of the orbits, since the OHR went from 1,08 preoperatively to 1,16 postoperatively (**Table 3**). OWR remained stable, from 0,92 preoperatively to 0,9 postoperatively.

In the group of patients who underwent a two-stage surgical approach, there was a small decrease in the asymmetry of orbital height and a minor change of the OWR (**Table 4**).

Discussion

Unicoronal craniosynostosis (UCS) if left untreated can lead to severe craniofacial scoliosis and compromise brain development. The primary intention is to improve both functional and aesthetical problems, which remains a challenge for craniofacial teams.

The craniofacial surgeons at CHULN, including Neurosurgeons and Plastic Surgeons, have been treating craniosynostosis for decades. Regarding UCS, the most common surgical intervention was a one-stage operation consisting on a bilateral FOA, usually performed after children were older than 12 months. More recently, a new technique was introduced consisting on a two-stage procedure. The first intervention occurs usually before 6 months of age and includes suturectomy of the fused suture and implantation of springs that will promote distraction over the next 6 months. The second intervention is usually planned around 18 months and includes removal of the springs and bilateral FOA to further correct the remaining craniofacial deviation. To our knowledge, this two-stage procedure to treat UCS has not yet been reported in the literature. Although surgeons had the subjective impression that the outcomes with the two-stage surgery were better, mainly by comparing photographs of the patients before and after the procedures, a quantitative measurement of the craniofacial deviation has not been performed. Therefore, we used craniometric measurements already published in the literature, including skull base twist, midface twist and orbital ratios, to compare the outcomes of the two types of surgical treatment for UCS.

We have observed that all patients in the study had a preoperative skull base deviation greater than ten degrees. This shift of the anterior fossa occurred to the side of the craniosynostosis because of the compensatory expansion on the non-fused side. After surgical intervention in both groups there was an improvement in this parameter, but it was noted that the greatest increase occurred in the group treated with a two-stage surgery.

Among the four children who had only one intervention, the correction of the skull base twist was modest. On the other hand, the five children treated initially with spring-mediated distraction significantly decreased the angle of skull base deviation. Importantly, the second procedure with bilateral FOA further improved the deviation in 4 out of 5 patients. We conclude that the distraction with springs largely corrects the deviation of the skull base, most likely due to the continuous distraction forces that simulate the natural growth of the bone until the age at which FOA can be performed.

The midface deviation is another characteristic of UCS. Although the skull base deviates to the side of the craniosynostosis, the midface twist occurs to the contralateral side. This happens due to the compensatory growth of the skull, causing contralateral movement of the bones of the nose and infraorbital region. In our study, the greatest correction of the midface twist was obtained with the two-stage surgery. Similarly to the skull base twist, the degree of improvement was higher upon distraction with springs and less pronounced after the second surgery.

In addition, the ratios for orbital height and width are in theory a very good predictor of symmetry between the orbital cavities. In all cases from our study we have preoperatively observed what was already expected: a higher and narrower orbit on the side of the synostosis, compared to the other side.

The average values of OHR and OWR in children who were submitted only to fronto-orbital advancement, showed that the difference in width and height between the orbits increased with surgical correction. However, all the changes that occurred were very subtle. In the other group that was submitted to a two-stage surgery, there was an improvement in the orbital height difference with the placement of springs and no significant differences in the orbital width ratio.

Although the changes in orbit height and width ratios have been minimal and, in many cases, have worsened, it should be kept in mind that orbit symmetry depends on other variables than just height and width, particularly their position and shape in the face - parameters that were not taken into account in this study.

Limitations

There were several limitations to the study that should be taken into consideration. Although the number of children surgically intervened for UCS correction between 2012 and 2021 was considerable (n = 23), only 9 were able to be included in the study. The main reason for exclusion was the absence of imaging studies, specifically head CT scans, before and after each intervention.

Sometimes head CT scans were not of high enough quality, which made 3D measurements of the distance of the orbits difficult since there were specific limits to the distance.

In addition, all data was collected manually, and no specific system was used for measurement or calculation.

Conclusion and future prospects

In conclusion, our study shows that children who have an early diagnosis of UCS and are treated in a two-stage surgery, are able to achieve greater symmetry of the face and skull base.

Further studies should include more patients in each group, evaluate other craniometric parameters and, importantly, compare blood transfusion rates and complications of the two surgical approaches.

We aim to publish our results in a peer review journal in the field of craniofacial disorders and we anticipate the two-stage surgery may be adopted in other reference centers around the world for the treatment of UCS.

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