Original Article

# Non-syndromic craniosynostosis: a retrospective analysis

## *Craniossinostoses não sindrômicas: uma análise retrospectiva*

PAULO VÍCTOR CUNHA COSTA [1]
MURILO SGARBI SECANHO [1]*
LEON CLERES PENIDO PINHEIRO [1]
MARCELO HANATO SANTOS [1]
ARISTIDES AUGUSTO PALHARES [1]
PEDRO TADAO HAMAMOTO-FILHO [1]

■ **ABSTRACT**

**Introduction:** Craniosynostoses are defined as premature fusions of one or more cranial sutures. They can be classified according to the sutures involved, correlated malformations and genetic alterations, and maybe syndromic or not. Its incidence is estimated at 1: 2,000-2,500 live births, with only 8% being syndromic. There are multiple surgical options for the correction of these malformations, and early referral to specialized centers for the treatment of these patients is necessary for a better therapeutic indication. The diagnosis is based on physical examination and computed tomography. The surgical treatment brings significant aesthetic and social improvements to patients. The objective is to analyze retrospectively patient data who underwent surgery to correct craniosynostosis at *HC of UNESP-Botucatu*. **Methods**: Retrospective analysis of the medical records of patients who underwent surgical correction of craniosynostosis between 2012 and 2017. **Results:** The prevalence of scaphocephaly and trigonocephaly was the same (38.5%). The mean age of the approach was 24 months; the mean surgical time was 3h48min, lower in scaphocephaly, 2h50min. All patients received blood transfusions in the perioperative period, with a mean percentage of 24.9% in relation to the preoperative blood volume. They underwent postoperative in a pediatric ICU. There were no deaths or complications, and the results were considered good by the team and family. **Conclusion:** Open surgical treatment of non-syndromic craniosynostosis is a safe procedure. Plastic surgery teams can achieve results comparable to those described in the literature, with low complication rates, good aesthetic results when performed correctly and with a multidisciplinary approach.

**Keywords:** Craniosynostoses; Surgery, Plastic; Craniofacial abnormalities; Blood transfusion; Skull.

Institution: Paulista State University, Botucatu School of Medicine, Botucatu, SP, Brazil.

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[1] Paulista State University, Botucatu School of Medicine, Botucatu, SP, Brazil.

■ **RESUMO**

**Introdução:** Craniossinostoses são definidas como fusões prematuras de uma ou mais suturas cranianas. Podem ser classificadas de acordo com as suturas envolvidas, malformações correlacionadas e alterações genéticas, podendo ser sindrômicas ou não. Sua incidência estimada em 1:2.000-2.500 nascidos vivos, sendo apenas 8% sindrômicas. Existem múltiplas opções cirúrgicas para a correção destas malformações, e o encaminhamento precoce para centros especializados no tratamento destes doentes é necessário para uma melhor indicação terapêutica. O diagnóstico é pautado no exame físico e tomografia computadorizada. O tratamento cirúrgico traz melhoras significativas estéticas e no convívio social dos pacientes. O objetivo é analisar os dados, retrospectivamente, dos pacientes submetidos à cirurgia para correção de craniossinostoses, no HC da UNESP-Botucatu. **Métodos:** Análise retrospectiva dos prontuários de pacientes submetidos à correção cirúrgica de craniossinostoses entre 2012 e 2017. **Resultados:** A prevalência de escafocefalia e trigonocefalia foi igual (38,5%), idade média de abordagem de 24 meses, o tempo cirúrgico médio foi de 3h48min, sendo menor nas escafocefalias, 2h50min. Todos os pacientes receberam transfusão de sangue no perioperatório com média percentual em relação ao volume sanguíneo pré-operatório de 24,9% e fizeram pós-operatório em UTI pediátrica. Não houve óbitos ou complicações, e os resultados tidos como bom pela equipe e familiares. **Conclusão:** O tratamento cirúrgico aberto das craniossinostoses não sindrômicas é um procedimento seguro. As equipes de cirurgia plástica conseguem alcançar resultados equiparáveis aos descritos na literatura, com baixas taxas de complicações, bom resultado estético, quando executado de maneira adequada, e com abordagem multidisciplinar.

**Descritores:** Craniossinostoses; Cirurgia plástica; Anormalidades craniofaciais; Transfusão de sangue; Crânio.

## INTRODUCTION

Craniosynostosis is defined as an abnormal growth of the skull due to the premature fusion of one or more cranial sutures. They can be classified according to the sutures involved, correlated malformations, and maybe random or linked to syndromes and genetic changes. Its estimated incidence is 1: 2,000-2,500 live births[1], with only 8% being syndromic or a family pattern[2].

The sagittal suture is most commonly affected in non-syndromic craniosynostosis, and the cause is not known. Possibly spontaneous mutations in a gene occur, but other risk factors involved are: fetal constriction (nulliparity, multiple pregnancy, and macrosomia), low birth weight, preterm birth, maternal use of valproate, and hydrocephalus with shunt[3,4].

When a suture closes early, the vault has restricted perpendicular growth, growing only parallel to this suture. This fact is known as Virchow's law[5]. The classification of non-syndromic craniosynostosis is based on the affected suture. The sagittal synostosis, known as scaphocephaly, determines that the skull has the shape of a "boat" with the increase of the anteroposterior dimension. The bicoronal synostosis, known as brachycephaly, produces a flattened head that is determined by the increase in biparietal diameter. Metopid synostosis determines the skull in a triangular shape or trigonocephaly. Plagiocephaly is the general term that denotes asymmetry in the coronal plane. It may be anterior when the synostosis is unilateral coronal (right or left) or posterior when the synostosis is lambdoid (right or left), this one being a rarer form[5].

There are multiple surgical options for the correction of these malformations. Early referral to a specialized center for these patients' treatment is necessary for a better therapeutic indication. The basis for diagnosis is physical examination. However, there

is often a delay in identifying this disease, especially in mild cases, when the shape of the head is not clearly abnormal, in symmetrical cases, with postnatal fusion, or due to the lack of knowledge of the attending professional[6]

After physical examination using craniometric measurements, we proceeded to the image diagnosis when there is a clinical suspicion of craniosynostosis. It is based on computed tomography with three-dimensional reconstruction, evaluating the fused suture, ventricular size, corpus callosum defects, and signs suggestive of increased ICP (intracranial pressure), such as "thumbprinting" or "beaten silver" patterns, loss of swollen folds and blunt cisterns. The increase in ICP is more common in multiple suture fusions or syndromic craniosynostosis[7-9]

Failure to treat patients can generate severe psychosocial losses for children when interacting with others during development, with repercussions in adult life. The increase in ICP is controversial in non-syndromic craniosynostosis cases, but some studies demonstrate this increase with rates varying between 4.5-24% of those affected[10,11].

The literature points to several treatment philosophies, such as the total open reconstruction of the skullcap, minimally invasive craniectomy using a postoperative helmet, minimally invasive craniectomy using springs, and cranial distractors[12-14]. Each procedure has positive and negative points. Our service's choice is due to the open reconstruction technique, which allows the removal of deformed portions, remodeling, and bone repositioning, with the possibility of extensive repairs, various osteotomies, and a single surgical time. The shape of osteotomies depends on the deformity presented and is discussed on a case-by-case basis.

With the proper care and perioperative management, it is possible to perform these complex procedures with low rates of complications[15].

## OBJECTIVE

The objective of this work is to retrospectively analyze data from patients with non-syndromic craniosynostosis surgically treated at the *Hospital das Clínicas of UNESP-Botucatu* together by the plastic surgery and neurosurgery teams.

## METHODS

Data collection was performed by reviewing the medical records of patients diagnosed with non-syndromic craniosynostosis, carried out between 2012 to 2017 at the *Hospital das Clínicas da Faculdade de Medicina de Botucatu*.

The data collected were age, type of craniosynostosis, date, type and duration of surgery, blood transfusions, length of stay in the ICU, postoperative complications, neuropsychomotor development (NPMD).

We assessed if there was a delay in the NPMD to analyze each age group's expected frames according to the child's card and recommendations from the Ministry of Health[16].

All surgeries were performed with sinuous bicoronal access for adequate exposure of the osteotomy areas. Osteosyntheses were performed with steel wires, not being necessary in cases of isolated scaphocephaly. Patients underwent the postoperative period in a pediatric ICU bed, and all required blood transfusion.

The institution's ethics committee approved the work through report 3,524,698.

## RESULTS

Patients' mean age at surgery was 24 months, with a median of 16 months, a standard deviation of 24.63, and a confidence interval of 0.693 ($p > 0.05$).

The types of craniosynostosis found were five scaphocephaly (38.5%), five trigonocephaly (38.5%), two plagiocephalies (15.4%), and an association between plagiocephaly and scaphocephaly (7.6%) (Figures 1 to 5).

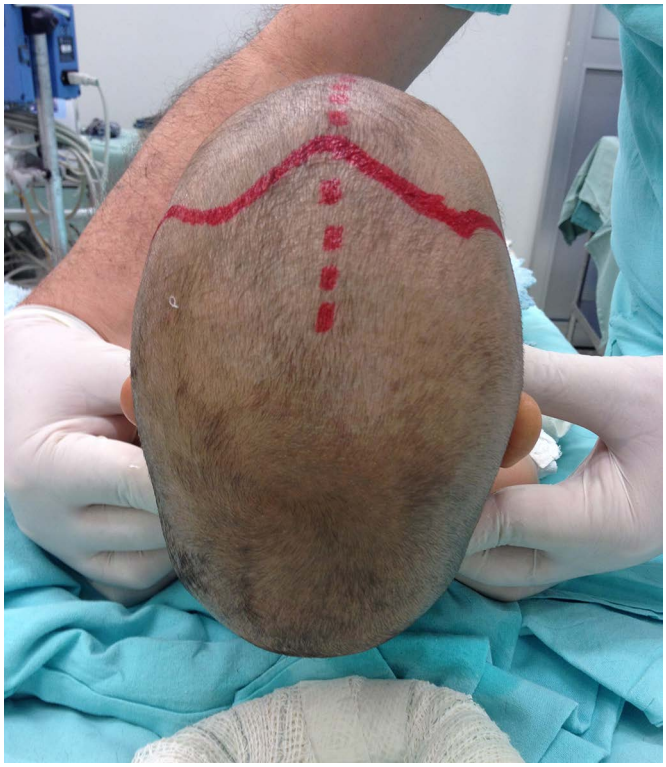

**Figure 1.** Preoperative marking of the incision for surgical treatment of scaphocephaly.

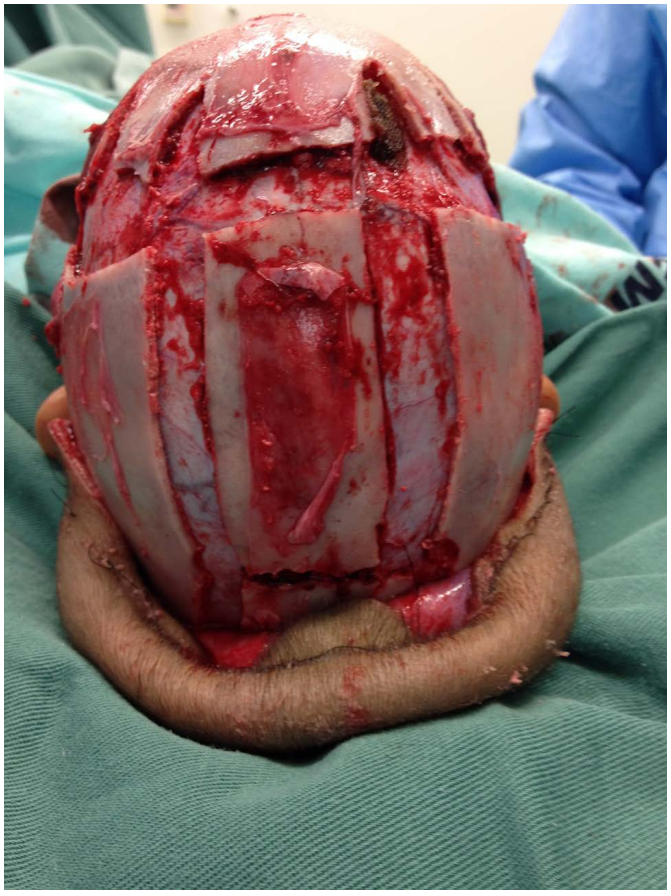

**Figure 2.** Intraoperative: osteotomy for surgical treatment of scaphocephaly.

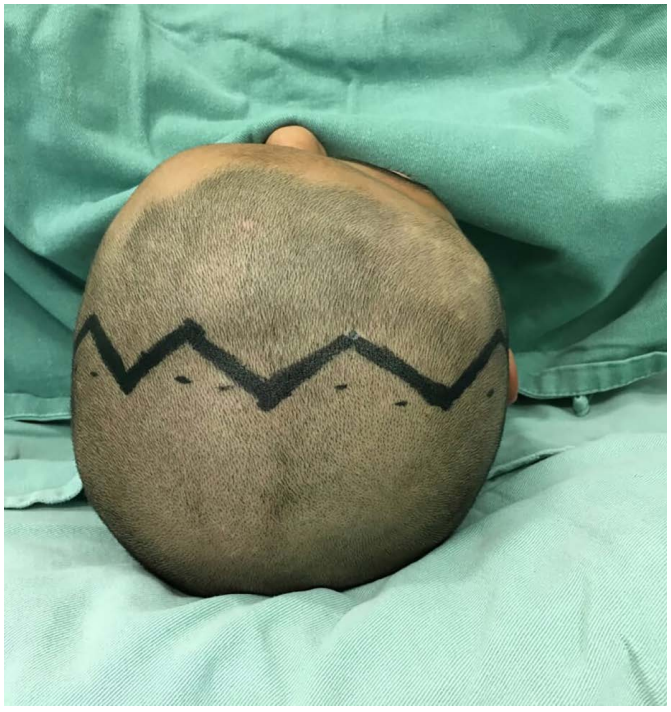

**Figure 3.** Incision marking for surgical treatment of brachiocephaly.

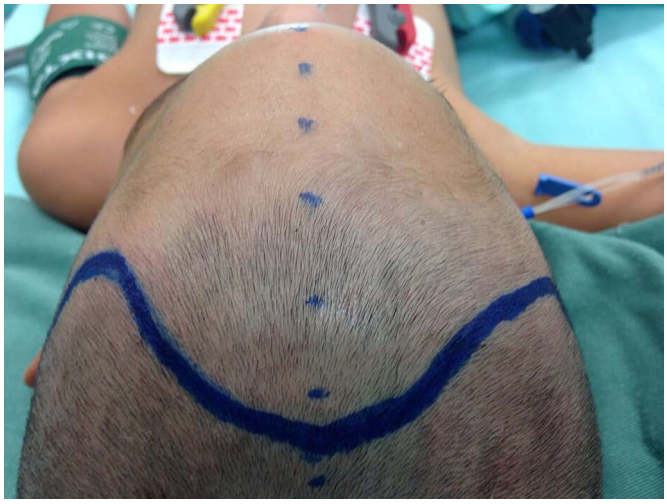

**Figure 4.** Marking of incision lines for surgical treatment of trigonocephaly.

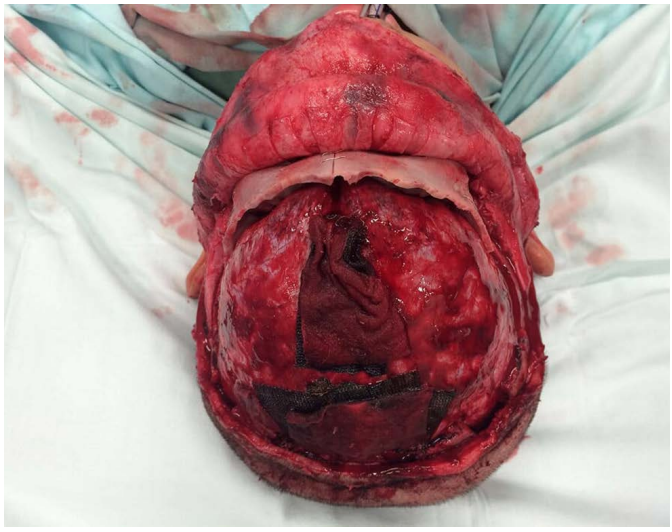

**Figure 5.** Intraoperative surgery correction for trigonocephaly.

The surgical time varied between 2h30min and 5h20min, with an average of 3h48min, and the scaphocephaly required shorter procedure time, with variation between 2h30min and 3h30min (Figure 6).

Postoperative mortality and postoperative complication rate were 0%.

Blood transfusions were performed in all procedures, with a mean percentage of transfused blood of 24.9% in relation to the preoperative blood volume (Table 1).

The children's intensive care unit's stay ranged from 1 to 5 days, with an average of 2.6 days of stay (Figure 7).

As for the aesthetic result and the need for surgical revision, considering the Whitaker classification, 12 were classified as level I and one as level II. The latter is awaiting a new procedure[17].

**Table 1.** Weight of patients and volume transfused.

| Pacient | Classification | Weight (KG) | Transfusion (Ml) | Percentage in Relation to Initial Blood Volume |
|---|---|---|---|---|
| 1 | Trigonocephaly | 8.1 | 120 | 18.50% |
| 2 | Plagiocephaly | 14.5 | 289 | 24.90% |
| 3 | Trigonocephaly | 13.1 | 200 | 20.30% |
| 4 | Scaphocephaly | 8 | 240 | 37.50% |
| 5 | Trigonocephaly | 8.3 | 100 | 15% |
| 6 | Scaphocephaly | 9.6 | 100 | 13% |
| 7 | Trigonocephaly | 15 | 218 | 19.40% |
| 8 | Plagiocephaly + Scaphocephaly | 7 | 376 | 67.10% |
| 9 | Plagiocephaly | 27 | 244 | 12.00% |
| 10 | Scaphocephaly | 9 | 150 | 20.80% |
| 11 | Scaphocephaly | 9.2 | 346 | 47.00% |
| 12 | Trigonocephaly | 13 | 149 | 15.30% |
| 13 | Scaphocephaly | 9.4 | 102 | 13.50% |

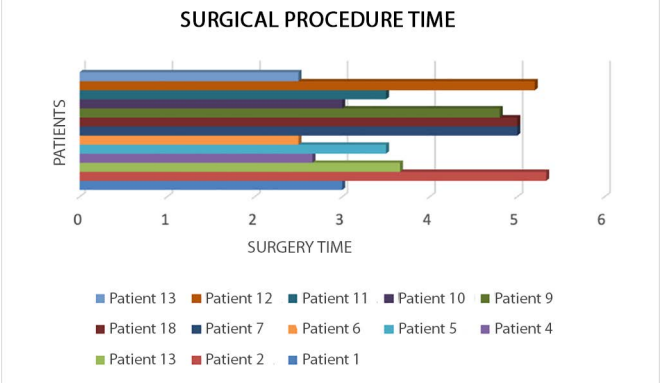


**Figure 6.** Time of surgery procedure.

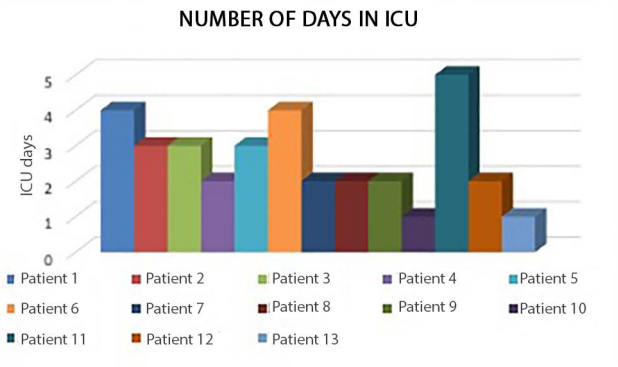


**Figure 7.** Necessary length of stay in the ICU bed.

NPMD delay occurred in 4 out of 13 patients, with a slight degree of delay (30%), in 2 of them related to speech.

## DISCUSSION

The craniofacial surgical procedures and the possible morbidities associated with surgical techniques and general anesthesia are of great concern and discussion among specialists. Blood loss and subsequent change in coagulability are still the main mortality factors in children's surgeries due to lower blood volume and higher energy expenditure. Regarding anesthesia, the risk of laryngeal and bronchospasm is higher in this population. However, with advances in surgical techniques and anesthetic care, the literature has shown that craniosynostosis surgeries are safe, with reduced rates of complications and mortality[18-20].

With a mean of 24 months ($p > 0.693$), the patients' age of approach was above the reports in the literature. This fact occurred due to a surgery performed on a child who was eight years and three months old, as he missed outpatient follow-up and returned at an advanced age, wanting surgery for aesthetic reasons. But even with a high average, the results were considered good by the surgical team[5,21-23].

There was the same prevalence between scaphocephaly and trigonocephaly in our case series, with 5 cases (38.5%), differing from the literature that reports a higher prevalence of scaphocephaly[1,2,15].

There has been an increase in the discussion about treatment with minimally invasive procedures, which would require less surgical time and less blood loss; however, there is a need for two or more approaches[17,24]. The treatment proposed in this series of cases showed good results for evaluating family members and staff and being reported as regular in only

one scaphocephaly case, which kept the frontal region not very prominent, with quantified improvement by 70% by family members and discussion of a second future intervention. Therefore, according to Whitaker's classification of surgical revision and result, where level I does not need new approaches, II the patient is submitted to soft tissue or small osteotomy correction, in III it is necessary osteotomies or larger bone grafts, and in IV, a new craniotomy and/or fronto-orbital remodeling is indicated; 12 patients are at level I and one of them at III, awaiting a new procedure[17].

The complication rate was extremely low, with no case of infection in the postoperative period, with 0% mortality. Only one patient presented temporary convergent strabismus, returning to normal without the need for intervention and keeping our rates comparable to those found in the literature concerning mortality and lower in complications[15,22,25,26].

The fixation of bone grafts can be performed with non-absorbable or absorbable materials to allow the growth of the skullcap[15]. The use of steel wires is the approach used in our service. Possible complications are extrusion, palpation, and intracranial translocation. The latter occurs due to the internal board's bone resorption, with deposition on the external one during cranial development. However, they have a low incidence without related symptoms. Associated with these factors, the high cost and the learning curve for using absorbable materials justify our choice for steel wires. Isolated scaphocephaly was treated without fixation after osteotomies[27].

Bleeding, a source of great concern occurs after the incision and comes from the scalp, skullcap, and dura. The presence of dural bone adhesions and the possible laceration of venous sinuses during craniotomy are imminent risk factors for difficult to control hemorrhage[6,28]. Blood transfusions were performed in all procedures, with an average transfused volume rate of 24.9% in relation to the patient's blood volume, preoperatively; this rate is lower than studies that analyze the repairing technique craniosynostosis with open surgery[22,29,30].

Although there are reports in the CSF fistula literature, we did not have this complication in our surgeries. The main associated risk factors are re-approaches due to fronto-orbital adhesions and distractions, which explains the absence in the sample presented, since such procedures were not performed[31,32].

Most specialists recommend the postoperative period performed in a pediatric ICU. We consider it necessary for patients' best control, with fine adjustment of the hydro electrolytic balance and ventilatory weaning [5,15,21,22]. The unit's stay ranged from 1 to 5 days, with an average of 2.6 days, longer than the one found, one day[33].

The average surgical time of 3 hours and 48 minutes is above that reported in the reviewed publications. However, in the articles analyzed, the decrease in time is due to the fixation of biodegradable materials using ultrasound, which can reduce this step's duration by 50%[22,34]. Another factor influencing the procedure's time is that the surgeries were performed by residents, still on a learning curve[35].

Several techniques are described for osteotomies in each specific type of craniosynostosis. The patients' planning and approach are made by the craniofacial surgery team, which has neurosurgeons and plastic surgeons. Each case is evaluated, and the osteotomies are individually planned against the defect presented by the patient. We believe that in this way, the best results are obtained.

The patients' follow-up varied between 10 months and six years, with a great psychosocial impact on the children approached. The follow-up to adulthood will answer the final repercussion of craniosynostosis correction in patients' interpersonal relationships.

Much is discussed about the neurological repercussions in non-syndromic craniosynostosis, increased ICP, and delayed neuropsychomotor development, especially concerning disorders related to speech language[1,2,7,11,36]. In our retrospective analysis, we noted a considerable prevalence of NPMD delay, with 4 out of 13 patients showing a mild degree of delay (30%), in two of them related to speech. Three were older than 24 months. Two underwent surgical correction for trigonocephaly, one for scaphocephaly, and one for plagiocephaly, corroborating the findings by Kljajic et al. in 2019[34].

This study's limitations were mainly the low number of cases analyzed, but this may reflect the difficulty of diagnosis and even access to consultations and referrals, which patients find in the public network. Another limitation is the fact that this study is retrospective, based on the analysis of medical records.

## CONCLUSION

The conventional open treatment of non-syndromic craniosynostosis brings good results, comparable to those existing in the literature, with low rates of complications when appropriately performed, showing to be a safe technique. Even older children can benefit from the surgical procedure.

Despite a small sample, the article shows good results compared to articles already published and denotes the importance of standardization of accesses and techniques, with multidisciplinary involvement and discussion.

## COLLABORATIONS

| | |
|---|---|
| **PVCC** | Analysis and/or data interpretation, Conception and design study, Data Curation, Methodology, Project Administration |
| **MSS** | Analysis and/or data interpretation, Conception and design study, Data Curation, Methodology, Writing - Original Draft Preparation |
| **LCPP** | Analysis and/or data interpretation, Data Curation |
| **MHS** | Data Curation |
| **AAP** | Supervision, Writing - Review & Editing |
| **PTHF** | Supervision, Writing - Review & Editing |

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***Corresponding author:** **Murilo Sgarbi Secanho**
Avenida Professor Mário Rubens Guimarães Montenegro, Unesp Campus de Botucatu, Botucatu, SP, Brazil.
Zip Code: 18618-687
E-mail: murilosecanho@gmail.com