Original Article

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Pre- and postoperative intracranial volumes in children with non-syndromic craniosynostosis using 3D volumetric technique

Gustavo Botelho Sampaio[1], Lionel Curado Valsechi[1], Renne Perez dos Santos Silva[2], Antônio Soares Souza[1]

[1] Faculdade de Medicina de São José do Rio Preto, São José do Rio Preto, São Paulo, Brasil

[2] Hospital Beneficência Portuguesa, São Paulo, São Paulo, Brasil

Gustavo Botelho Sampaio, MD

e-mail: gugsampaio@gmail.com

Available at:
http://www.archpedneurosurg.com.br/

Introduction: Corrective surgery for craniosynostosis presents challenges, particularly in gaining practical experience. The skull's complex structure, located at the cephalic end of the body, requires careful understanding. The neurocranium, which protects the brain, grows rapidly during early life. Understanding normal cranial growth is essential for monitoring, detecting abnormalities, and evaluating the long-term results of craniosynostosis surgery.
Objectives: To study cranial volume gain after surgical treatment of craniosynostosis using 3D printing technology.
Methodology: Thirty-six patients who underwent craniosynostosis surgery at Hospital da Criança e Maternidade (2019-2022) were selected; 10 were excluded for not meeting prerequisites. Preoperative, immediate postoperative, and late postoperative (3 months) tomography exams were performed. Exams were reconstructed using Blender for cranial volume calculation, and skulls were 3D printed using a Sethi 3D printer. Results were evaluated using Student's t-test for independent samples.
Results: The study included 26 patients: 10 with scaphocephaly treated with Renier's "H" cranial remodeling, 5 with trigonocephaly, 5 with plagiocephaly, and 2 with brachycephaly treated with fronto-orbital advancement (FOA). Cranial volumes increased by an average of 224 cm^3 (Renier's "H") and 138.8 cm^3 (FOA) between late postoperative and preoperative stages.
Conclusion: 3D shape and volumetric measurements indicate abnormal brain growth in single-suture craniosynostosis patients. Surgical correction improves cranial differences compared to healthy controls, suggesting less invasive techniques could utilize patients' natural volumetric gain.

Keywords: Craniosynostosis, Intracranial volume, fronto-orbital advancement surgery, Renier's h surgery

INTRODUCTION

Over the past few years, 3D printing technology has advanced considerably, resulting in reduced production costs, improved accuracy of printed objects, and increased variety of materials for printing. These improvements have enabled the creation of a variety of products and made this technology available even for domestic use. In the medical field, [2] 3D printing is increasingly being used to create models, devices, and custom implants, with the potential to enhance patient care.

Specifically in neurosurgery, 3D printing has had a significant impact, aiding in the visualization of complex anatomical structures and in the planning of delicate surgical procedures. This is crucial, as most traditional imaging methods, such as X-rays, computed tomography (CT), and magnetic resonance imaging (MRI), provide images in two dimensions (2D) or in a 3D volume in 2D slices [48]. Three-dimensional printing enables the reconstruction of anatomical structures into 3D physical models, facilitating surgical planning and education for patients and students [3 4 5].

Craniosynostoses are a group of alterations in the shape and growth of the skull, resulting from partial and premature fusion of one or more sutures in the cranial vault and its base. These changes in the sutures cause restrictions in the development of certain areas of the skull, compensated by abnormal growth in other regions. Depending on the affected sutures, different specific types may develop [7 9 16].

Surgical treatment is indicated for a significant portion of craniosynostosis patients to avoid the consequences. It is preferable for this treatment to be performed early, in the first few months of life, as it provides better aesthetic and functional outcomes and prevents brain compression. Early diagnosis of the disease is necessary to enable this early treatment.

In summary, 3D printing has demonstrated its potential in various areas of neurosurgery, including surgical planning,

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training and education, the development of surgical devices, and the advancement of tissue engineering implants. Cranial volumetry calculation has always been a major challenge; however, with technological evolution, it is now possible to perform volumetric calculation more conveniently and accurately. Given this, the proposal of this study is to use 3D printing technology to perform pre and postoperative volumetric calculation of children with non-syndromic craniosynostosis [22 23 32].

OBJECTIVE

To study the cranial volume gained after surgical treatment of craniosynostosis using 3D printing technology.

MATERIALS AND METHODS

The study enrolled 36 patients who underwent craniosynostosis surgery at the Children's Hospital and Maternity of São José do Rio Preto (HCM) between 2019 and 2022. However, 10 patients were excluded: three had syndromic craniosynostosis, two lacked suitable images for printing, three had unretrievable CT scans, and two were over 2 years old at surgery. Inclusion criteria comprised children under two years old with non-syndromic craniosynostosis, operated on at HCM between 2019 and 2022, who underwent preoperative, immediate postoperative (within 24 hours), and 3-month follow-up CT scans. Complex craniosynostosis was defined as early closure of two or more sutures. All patients underwent preoperative, immediate postoperative, and 3-month post-surgery CT scans.

Tomographic scans, with 4mm slice thickness, were conducted preoperatively, immediately postoperatively, and three months after surgery. Blender® software facilitated 3D reconstruction, with subsequent volume measurement using the program's internal algorithm. Statistical analysis employed the student's t-test for dependent samples.

The study received approval from the research ethics committee of the School of Medicine of São José do Rio Preto.

RESULTS

Scaphocephaly was observed in 38% of patients and was treated using Renier's "H" cranial remodeling technique. Another 38% presented craniosynostosis involving multiple sutures, with variations in surgical approaches. Among these cases, some exhibited trigonocephaly, plagiocephaly, and brachycephaly, and were managed with AFO and frontal remodeling techniques by Arnaud and Marchac [40]. (Figure 1).

Preoperative intracranial volume measurements in patients with plagiocephaly averaged 868.9 cm³. Postoperative volumes immediately after surgery averaged 999.79 cm³, increasing to 1136.2 cm³ in the late postoperative period, resulting in a total volume gain of 237.99 cm³. Significant differences were noted when comparing surgical volumetric gain with total volumetric gain.

For trigonocephaly cases, the average preoperative intracranial volume was 808.3 cm³. Postoperative volumes immediately after surgery averaged 958.5 cm³, increasing to 1101.7 cm³ in the late postoperative period, resulting in a total volume gain of 293.4 cm³. Significant differences were observed in comparing surgical volumetric gain with total volumetric gain.

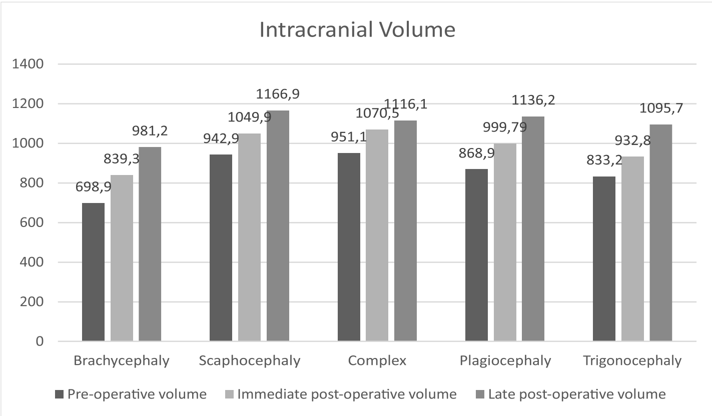

Figure 1- Disposure of pre operatory, immediate, and late post-operative intracranial volume separated by types of craniosynostosis.

In patients with complex craniosynostosis, the average preoperative intracranial volume was 951.1 cm³. Postoperative volumes immediately after surgery averaged 1070.5 cm³, increasing to 1116.1 cm³ in the late postoperative period, resulting in a total volume gain of 165.1 cm³. No significant differences were observed in comparing surgical volumetric gain with total volumetric gain.

Similar patterns were observed in cases of brachycephaly, scaphocephaly, and AFO, where significant differences were found when comparing surgical volumetric gain with total volumetric gain, indicating the influence of surgical intervention on cranial volume changes. (Figure 2).

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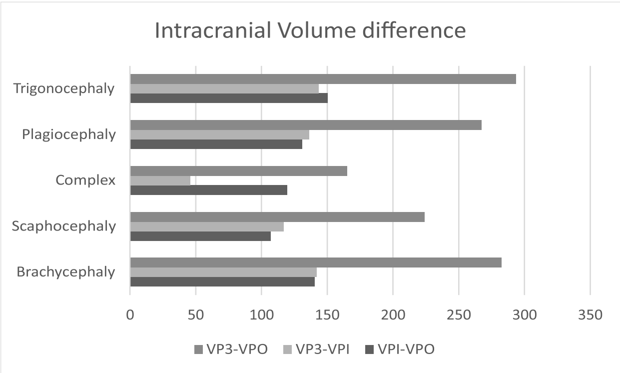

Figure 2- Difference between intracranial volume, where VPI is the pre-operative volume, VPO is the immediate post-operative volume and VP3 is the post-operative volume after 3 months. POV-VPI means the intracranial volume gained by the surgical procedure. VP3-VPO is the intracranial volume gained by the natural development of the skull and VP3-VPI is the total intracranial grain volume.

DISCUSSION

Studies conducted globally have reported a prevalence range for craniosynostosis between 1/1,700 and 1/4,000 births [12].

Abbott et al.'s study [1] investigating cranial volume using computed tomography (CT) in a pediatric cohort of 157 children revealed a progressive increase in cranial volume across infancy, ranging from 419 to 581 cm³ in 1-month-olds, 700.5 to 971 cm³ in 6-month-olds, 870.2 to 1207.2 cm³ in 12-month-olds, and 905.0 to 1255.5 cm³ in 14-month-olds.

Despite the limited number of cases, no significant differences in volumetric gains were observed in brachycephaly. However, when comparing surgical procedure-induced volumetric gains to total volumetric gains, a highly significant difference emerged (p=0.008). Similarly, significant differences were noted when contrasting patient-induced volumetric gains with total volumetric gains (p=0.02).

In Gault's study examining intracranial volume in 104 children with craniosynostosis, it was concluded that most children exhibited age-appropriate volumetric growth. Notably, girls with scaphocephaly displayed significantly smaller cranial volumes compared to boys, who showed no significant variations [15 16].

Utilizing Renier's H surgical technique for scaphocephaly treatment [40], a highly significant difference was detected when comparing surgical procedureinduced volumetric gains to total volumetric gains. Similarly, significant differences were observed when contrasting patient-induced volumetric gains with total volumetric gains. Notably, patients with single-suture craniosynostosis exhibited larger volumes and altered shape metrics compared to age-matched controls, both pre- and post-surgery [43].

Existing studies on intracranial volume in sagittal synostosis have yielded inconsistent results. Lee et al. found variations in intracranial volume across different age groups, with male patients exhibiting below-normal volumes before 6 months, normal to slightly elevated volumes between 7 and 12 months, and decreased volumes in older age groups. Conversely, Anderson et al. reported significantly larger intracranial volumes in both male and female patients with untreated sagittal synostosis [29].

The analysis of volumetric gains following AFO surgery compared to total volumetric gains revealed a highly significant difference, underscoring the procedure's impact on cranial development. This aligns with Shukriyah et al.'s findings, [50] which documented notable increases in cranial volume post-cranial expansion surgery for craniosynostosis. While Shukriyah et al. noted no statistical differences in radiological and clinical outcomes, they speculated on the longterm correlation between volumetric changes and neurological development improvement [50].

In our study, significant differences were observed when comparing volumetric gains from AFO surgery to total volumetric gains in trigonocephaly patients. Similarly, highly significant differences were noted when contrasting patient-induced volumetric gains with total volumetric gains. In comparison, Shukriyah et al. reported an average increase of 191.5 mL (28%) in intracranial volume within an average of 101 days post-surgery. Our study showed a volumetric increase of 136.4 mL in 3 months following AFO surgery for plagiocephaly, representing 51% of the total gain. Additionally, patients with trigonocephaly undergoing AFO exhibited an average gain of 162.9 mL, 26.5 mL larger than those with plagiocephaly undergoing the same procedure.

Overall, comparisons of volumetric gains between various craniosynostosis types and surgical interventions highlight significant differences, emphasizing the multifaceted nature of cranial development and the need for tailored treatment approaches. Further research incorporating larger patient cohorts and advanced imaging

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modalities will provide deeper insights into craniosynostosis pathophysiology and optimize therapeutic outcomes.

CONCLUSION

The most common craniosynostosis is scaphocephaly, followed by plagiocephaly and trigonocephaly. The average age of children undergoing surgical correction was 9 months, while the average gestational age at birth of the patients was 37 weeks and 5 days. On the other hand, the gestational age of children with complex craniosynostosis was 32 weeks.

The volumetric gain from the surgical procedure is important and is associated with the AFO technique used for correction of plagiocephaly and trigonocephaly and the H technique of Renier for correction of scaphocephaly.

The volumetric gain considering natural postoperative growth is important in patients undergoing correction of complex craniosynostosis, scaphocephaly, and trigonocephaly and was not as significant in plagiocephaly.

The cranial volume of patients with craniosynostosis depends on the type of craniosynostosis, with complex craniosynostosis and scaphocephaly having the highest cranial volume at the time of diagnosis, whereas the final cranial volume is higher in scaphocephaly.

DISCLOSURES

Ethical approval

This study was performed in line with the principles of the Declaration of Helsinki. Approval was granted by the local Ethics Committee, number: 6.703.943.

Consent to participate

The patients gave consent to use their information and images for research purposes. *Consent for publication*

The patient gave consent to use his information and images for publication.

Conflict of interest

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Artificial intelligence

Yes. During the preparation of this work the author(s) used CHAT GPT-6 in order to improve translation, correct errors and improve formatting.. After using this tool/service, the author(s) reviewed and edited the content as needed and take(s) full responsibility for the content of the publication

CONTRIBUTIONS

-**Gustavo Botelho Sampaio**: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing
-**Lionel Curado Valsechi**: Data curation, Investigation, Methodology
-**Renne Perez dos Santos Silva.**: Methodology, Software
-Antônio Soares Souza: Conceptualization, Project administration, Supervision, Validation, Visualization, Writing – review & editing

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