

Endoscopy-assisted surgical treatment of trigonocephaly – A Technical Note

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Craniosynostosis, the premature fusion of cranial sutures, affects approximately 1 in every 1,800 to 2,000 live births and can lead to neurocognitive deficits, increased intracranial pressure, and cranial deformities. This narrative review aims to outline the endoscopic treatment of trigonocephaly, a form of craniosynostosis characterized by early closure of the metopic suture, resulting in a triangular forehead and hypotelorism. Using data from literature and a case study, this paper describes the step-by-step endoscopic surgical technique, highlighting its advantages, including smaller incisions, reduced blood loss, and shorter hospital stays compared to traditional open surgery. The patient described in the case study, a 1-month-old female, underwent successful endoscopic surgery with no significant complications, requiring only two days of hospitalization. The literature suggests that early intervention, particularly within the first three months of life, yields optimal results. Additionally, the use of cranial orthosis postoperatively is essential for long-term cranial remodeling. Endoscopic-assisted techniques offer promising outcomes, including lower morbidity, faster recovery, and reduced healthcare costs, making them a viable alternative to traditional methods for treating craniosynostosis.

Keywords: endoscopic, endoscopic-assisted, craniosynostosis, trigonocephaly.

INTRODUCTION

The infant skull is made up of bony plates that articulate with each other by means of fibrous joints called sutures. These sutures are mobile and should not be fused at birth. Craniosynostosis is a pathology characterized by the early closure of one or more cranial sutures and occurs with an incidence of one case in every 1800–2000 live births [1,2].

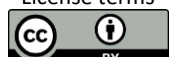
It manifests clinically with the alteration of the shape of the child's skull, presenting a typical shape depending on the cranial suture involved. This alteration in cranial growth prevents the proper accommodation of the brain inside the skull. The restriction of the intracranial space can lead to inadequate development of the brain and cause neurocognitive deficits, increased intracranial pressure, speech alterations, and potential psychological damage caused by cranial deformity and beauty standards already established in the current society [1,2].

The known forms of treatment for craniosynostosis are open reconstruction and endoscopic surgery. These surgeries aim to allow the proper growth of the brain by correcting the skull deformity. This article aims to give a step-

by-step guide for the endoscopic treatment of trigonocephaly, a type of craniosynostosis involving premature fusion of the metopic suture, which creates a triangular forehead and hypotelorism [1,2].

OBJECTIVES AND METHODS

This study aims to provide a comprehensive overview of the endoscopic-assisted treatment of metopic craniosynostosis. To achieve this, a detailed narrative review of the current literature was conducted using the PUBMED electronic database with the keywords "endoscopic" and "craniosynostosis" to gather relevant information on the topic. Furthermore, this study includes a step-by-step description of the surgical technique to ensure a thorough understanding of the procedure. To complement this review, a real-life clinical case from the author's personal archive is presented, serving to illustrate the practical application of the described method and explain the rationale behind its choice. This combined approach highlights both the theoretical foundation and the clinical implementation of endoscopic-assisted treatment for metopic craniosynostosis.



Case

The patient was a 1-month-old female, brought by her parents due to a complaint of cranial deformity noted after birth. The mother did not report complications during pregnancy and reported neuropsychomotor development within the normal range for her age. On physical examination, the patient presented phenotypic alterations suggestive of trigonocephaly, such as a triangular forehead and hypotelorism. She underwent cranial computed tomography with 3D reconstruction, which revealed the premature closure of the interfrontal suture, a frontal angle of 95° and presented an omega sign, characterizing a metopic craniosynostosis. Other cranial sutures were unaltered. The patient underwent endoscopic treatment at 2 months of age (Figure 1-5).

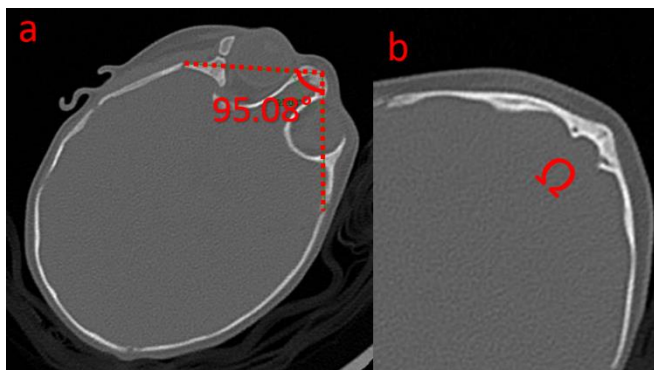


Figure 1a and 1b- The case described above had a frontal angle of approximately 95° classified as a moderate trigonocephaly and omega's sign.

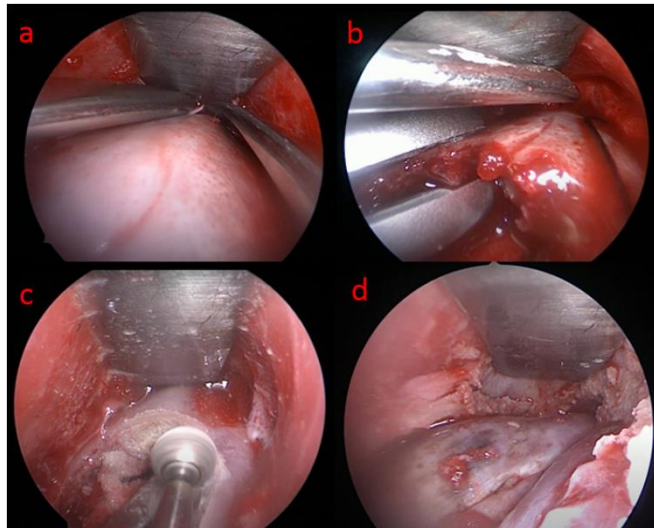


Figure 2: A- After detaching the pericranium. B C- Removal of fused metopic suture using Middleton-Jansen forceps and diamond drill. D- final aspect.

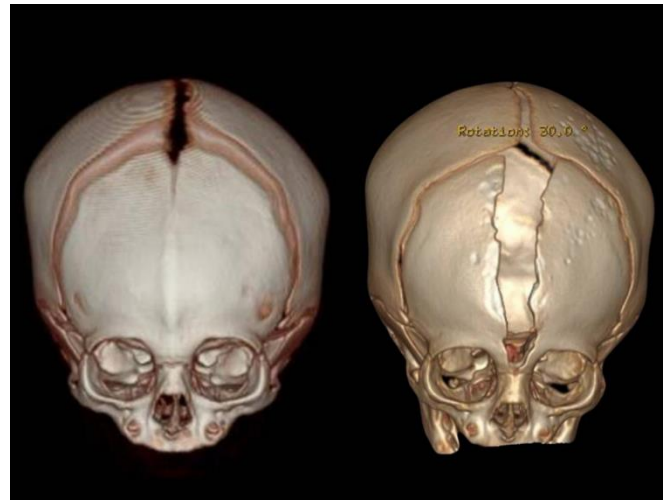


Figure 3 - 3D computed tomography scan before and after the endoscopic-assisted suturotomy.



Figure 4 - Comparison between skin incisions made during a endoscopic-assisted suturotomy (left) and a open surgery (right) to treat trigonocephaly.

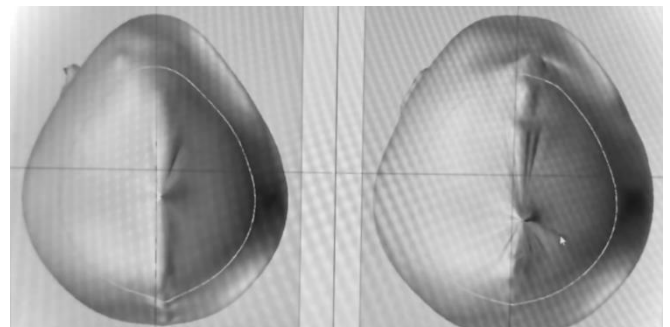


Figure 5 - Comparison between head shapes before (left) and after (right) endoscopic-assisted treatment. Images obtained by 3D scanning.

TECHNICAL NOTE

Surgical Technique

The patient was positioned in supine position under general anesthesia. Anesthetic preparation required blood pressure monitoring by means of an intra-arterial catheter, two peripheral venous accesses, and bladder catheterization for volume management. After careful aseptic preparation, the skin was marked to indicate a median linear incision with a transverse orientation, measuring about 3 cm, and located posterior to the hairline.

From the beginning of surgery, thorough hemostasis is required due to the lower tolerance of these patients for blood loss during the procedure.

The underlying tissues were dissected at the level of the areolar stratum, initially with the naked eye and later with the aid of the endoscope for wide exposure from the bregma to the nasium. A monopolar cautery with a needle-like tip and 0-degree optics is used at this stage of the procedure, in addition to an Aufricht retractor for better exposure of the accessed area.

After dissection, the pericranium was detached from the exposed area with the assistance of the endoscopy. After this phase, the osteotomy can be started.

Two parasagittal trepanations were performed, one on each side, to detach the superior sagittal sinus. The trepanation holes were communicated and widened using Kerrison. Thereafter, the dura mater was detached from the area that will be manipulated, initially under the naked eye, but in deeper areas with the help of the endoscope.

First, scissors-assisted resection was performed towards both the bregma and the nasium. This osteotomy towards the bregma may or may not need to be assisted by endoscopy. Towards the nasium, the osteotomy can be done initially with scissors, but its deepest portion will require a diamond drill or Middleton-Jansen forceps. Osteotomy was completed when it was possible to identify the sagittal suture or anterior fontanelle posteriorly and the internasal suture anteriorly. At this point, palpation may reveal greater mobility in the frontal bones.

After the osteotomy, the surgical site was reviewed to identify any pointed or irregular bone keels that must be removed, in addition to rigorous hemostasis of the cavity. Closure was completed by planes using absorbable sutures. The wound was covered with sterile dressing with compressive bandaging. After surgery, the patient was referred to an intensive care bed for monitoring of vital signs.

After being discharged from the hospital on the 2nd postoperative day, the patient underwent a 3D scan of the skull on the 7th postoperative day and started using the

cranial orthosis on the 10th postoperative day (23-hour per day regimen).

The injury to the superior sagittal sinus is the most severe complication that can occur during the endoscopic-assisted procedure. Its occurrence can be prevented through careful dissection of the dura mater and the application of hemostatic agents on its surface. The endoscopic suturectomy technique described above is currently the only endoscopic method available to treat metopic craniosynostosis in the medical literature.

DISCUSSION

Based on our research, we were able to identify that endoscopy-assisted surgery for craniosynostosis began to be used in 1998, with the objective of using smaller incisions, less tissue manipulation of the patient, less transfusion of blood products, lower cost, and shorter surgical time [3].

The rationale for choosing the endoscopic-assisted surgical technique as the patient's treatment was based on the age group compatible with the best results found in the literature, early treatment of craniosynostosis with the least possible tissue damage, clarification of doubts and agreement by parents on the use of cranial orthosis and general care in the postoperative period.

The surgery of the patient in question lasted about 1 hour, a linear incision of about 3 cm was used, her hospital stay was 2 days, with only 1 day spent in intensive care. The patient did not need blood transfusion nor during the procedure or after. No other notable complications were encountered.

The main objectives of craniosynostosis treatment are the correction of cranial deformity (aesthetic outcome) and allowing adequate brain growth (functional outcome), both by cranial bone remodeling. In a study by Magge et al., no statistically or clinically significant differences in cognitive outcomes were found among school-age children or in the type of surgical procedure (open or endoscopically assisted) used to repair non-syndromic sagittal craniosynostosis [4].

Regarding the aesthetic outcome, in a study conducted by Domínguez et al., the 34 patients who underwent endoscopic-assisted surgery were followed-up on an outpatient basis for 12 months and were classified as Whitaker I during evaluation. This category is used for those patients who did not require a new corrective surgery [5,6].

Temporal hollowing is a frequent issue encountered after surgeries involving the frontal region and the temporal muscle. It arises from a combination of osteotomies performed in the area, varying degrees of osteolysis, and temporal muscle atrophy. In children, this can result in an uncomfortable and undesirable aesthetic outcome, highlighting the need to develop increasingly less invasive

surgical techniques that prioritize both functional and cosmetic results [7,8].

Objective information specifically addressing temporal hollowing and comparing the outcomes between endoscopic and open techniques was not found in the literature. This gap highlights an opportunity for future studies. However, it is suggested that the endoscopic technique may be superior in this regard, as the areas commonly manipulated during open surgery are preserved in the endoscopic procedure.

The consensus holds that the ideal age for treatment of craniosynostosis is very young, since most brain growth occurs in the first months of life. Ingraham et al. and Shillito et al. recommend that treatment of craniosynostosis should occur between 4 and 6 weeks of age. Jimenez et al. reported that their best results were obtained when patients underwent the procedure within 3 months of age. Berry-Candelario et al. recommend that endoscopic-assisted suturectomy be performed as early as possible, balancing factors such as general health conditions and weight, noting that the ideal time would be at 3 months or just before 3 months [9–12].

Domínguez et al. showed that the surgical time was significantly shorter in patients undergoing endoscopy-assisted surgical treatment [6]. This finding was similar to the results of other studies and systematic reviews [3,14,15]. A multicenter study that included 1382 children less than 12 months of age described a mean surgical time of 70 minutes for endoscopy-assisted surgery and 115 minutes for open surgery [16]. The smaller skin incisions and en bloc osteotomies are factors that influence the reduction of the time required to perform endoscopy-assisted surgery [3,6,14,17].

The estimated volume of blood loss had a median of 38 cc in endoscopy-assisted surgery and 165 cc in open surgery in a study conducted by Domínguez et al. The same study revealed that the use of blood products was necessary in 8% of patients undergoing endoscopy-assisted surgery, while in patients undergoing open surgery, the rate is 79.2%. These data are in agreement with data found in other studies that reported the need for blood transfusion between 4% and 10% of patients undergoing endoscopic-assisted surgery [6,11,13,14,18]. In the study conducted by Mendonça et al., none of the 17 patients who underwent endoscopic-assisted surgery required transfusion [15]. The authors suggested the preoperative use of erythropoietin and iron supplementation associated with the use of tranexamic acid during anesthetic induction. To assist in hemostasis during the surgical procedure, gelfoam fragments soaked in a tranexamic acid solution are prepared.

The use of cranial orthosis is initiated by 1 week after performing the surgical procedure. Subsequent adjustments of the cranial orthosis are critical to the success of endoscopic treatment of craniosynostosis. Brain growth acts

as an internal distraction, reshaping the skull in all three dimensions. The use of the helmet must be rigorous to obtain the best result. Cases in which the helmet is not used properly are associated with inferior results in the correction of cranial deformity. The use of cranial orthosis is suspended as soon as the desired cranial phenotype is obtained, which most often happens when the infant reaches around 1 year of age.

In an analysis of the costs involved in the treatment procedures for craniosynostosis, even after adding the cost of obtaining and adjusting cranial orthosis and the work time that is lost by the families, using minimally invasive techniques substantially reduces costs. Comparing 10 patients treated with minimally invasive techniques and 10 patients treated with open reconstruction, the total costs of endoscopic techniques accounted for, on average, 42% of the costs of open techniques [13].

Comparing craniosynostosis treatment techniques and the analysis of the use of hospital resources, it is possible to state that endoscopy-assisted surgery had the lowest rate of use of hospital resources, while open surgery with distraction had the highest use of hospital resources [19]. In order to corroborate these data, the average length of hospital stay after endoscopy-assisted surgery was 1 day, while patients who underwent open surgery required an average of 4 days of hospitalization [6]. Other studies have also shown similar data [3,14,15]. It is worth noting that the reduced cost is the result of reduced hospitalization time associated with shorter surgical time, less intraoperative drug administration, less transfusion of blood products, and shorter postoperative intensive care unit stay [6].

In a study by Barone et al. on the endoscopic treatment of scaphocephaly as compared with open surgery, the mean operative time (1.6 hours versus 3.5 hours), estimated blood loss (43 cc versus 168 cc), hospital costs (US\$ 11,671 versus US\$ 36,685) and length of hospital stay (1.16 days versus 5.1 days) were shorter with the use of the endoscopic technique [20]. There is a lack of data in the Brazilian medical literature regarding the cost comparison between open and endoscopic approaches for the treatment of metopic craniosynostosis. This gap could serve as a valuable focus for future research.

The hypotelorism commonly found in these patients results from the approximation of both orbital cavities confirmed by a reduced frontal angle. As described by Oi and Matsumoto, the frontal angle is defined as the angle between the two lines drawn through the pterium bilaterally and the nasium. The frontal angle is considered normal when it measures 104° or more. Trigonocephaly, on the other hand, can be classified as mild when it has an angle between 96° and 103°, moderate when the angle is between 90° and 95°, or severe when the angle is less than 89° [7,21]. The case described above had a frontal angle of approximately 95°.

In a study conducted by Jimenez et al., 108 (76.6%) patients had hypotelorism (represented by an interpupillary distance divided by the interfrontozygomatic distance with a result of less than 0.4). Of the 108 patients with hypotelorism, 100 (93%) achieved a value above 0.4 after endoscopy-assisted surgical correction. Correcting the deformity caused by metopic synostosis is a slow process. Once the fused suture is removed, the cranial orthosis helps to correct the deformity in order to guide the bone molding induced by brain growth, maintaining the anteroposterior distance and allowing the frontal bones to grow forward and laterally [14].

Treatment of patients with metopic craniosynostosis is a challenge, as the work does not end once surgery is completed. A review on open surgical correction revealed favorable immediate results after surgery but found a significant decline in aesthetic outcomes over time [14,22].

The rationale for the development of endoscopic procedures for craniosynostosis is quite sound. Any option that reduces the risks, morbidity, and costs of reconstruction compared to traditional cranial remodeling should be vigorously pursued [23].

CONCLUSION

The literature indicates that endoscopic-assisted surgical technique allows adequate correction of cranial deformity similar or superior to the open technique, since it has the advantages of shorter surgical time, smaller skin incision, lower estimated blood loss, lower blood transfusion rate, and faster hospital discharge. Given such a narrow age treatment window, this knowledge should be disseminated among neurosurgeons, through theoretical and practical training, and among other health professionals, parents, and caregivers, through training, so that this treatment can become increasingly available and more patients to benefit.

DISCLOSURES

Ethical approval

This study was performed in line with the principles of the Declaration of Helsinki. This study does not require approval from a Research Ethics Committee as it is a narrative review based on previously published literature and publicly available data, without direct involvement of human subjects or personal data.

Consent to participate

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

The authors affirm that no artificial intelligence tools were used in the writing, editing, or content generation of this manuscript. All work was conducted manually, based on thorough research and academic expertise.

CONTRIBUTIONS

-Emmanuel de Oliveira Sampaio Vasconcelos e Sá: Conceptualization, Formal Analysis, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing

-Marcos Devanir Silva da Costa: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing

-Sérgio Cavalheiro: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing

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