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OPEN AND ENDOSCOPY ASSISTED TRIGONOCEPHALUS SURGERY: REPORT OF TWO CASES

Case Report

AÇIK VE ENDOSKOPİ YARDIMLI TRİGONOSEFALİ CERRAHİSİ: 2 VAKA SUNUMU

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ABSTRACT

Introduction: Reporting two cases with trigonocephalus, treated by open and endoscopic surgery.

Materials and Methods: Craniosynostosis refers to premature fusion of one or more cranial vault sutures. Trigonocephaly is the second most frequent type of craniosynostosis and is associated with a remarkable occurance of intracranial abnormalities and neuropathology. Patients with metopic craniosynostosis are traditionally treated with fronto-orbital advancement to correct hypotelorism and trigonocephaly. Alternatively, endoscopic-assisted treatment comprises narrow osteotomy of the fused suture followed by postoperative helmet therapy.

Results: The first patient is a first-born 6-month old male to healthy parents. After he was diagnosed as trigonocephaly he underwent surgery under general anesthesia. Open surgery was performed. Postoperatively the patient was monitored in the intensive care unit and was discharged 3 days later with no complications. Second year control images revealed physilogical formation of cranium.

The second patient is a 3-month old male, in whom the vaginal delivery was at 40 weeks of gestation. After he was diagnosed as trigonocephaly he underwent surgery under general anesthesia. Endoscopy assisted microsurgery was performed. Postoperatively the patient was monitored in the intensive care unit and discharged 3 days later with no complications. Early cranial CT showed no extra complications and trigonocephalus getting better.

Conclusion: Endoscopy assisted surgery is safe and efficient while allowing shorter operative times, reduced costs due to decreased hospital stay, and fewer blood transfusions. However, minimally invasive treatment is only possible in cases with early

diagnosis and cooperation in using a postoperative helmet therapy.

Key words: Craniosynostosis, Trigonocephalus, Endoscopy Assisted Surgery

ÖZET

Amaç: Açık ve endoskopik cerrahi ile tedavi edilen ili trigonosefali vakasının sunumu.

Materyal ve Method: Kraniosinostoz, bir veya birden fazla kranyal kemik sütürünün prematür füzyonu olarak tanımlanır. Trigonosefali, sinostozun ikinci en sık görülen tipidir (insidansı, 1: 5,200) ve intrakraniyal anomaliler ve nöropatolojilerle belirgin ilişkisi vardır. Metopik kraniyosinostozu olan hastalar, hipotelorizm ve trigonosefalinin düzeltilmesi amacıyla fronto-orbital yaklaşımla tedavi edilir. Alternatif olarak; endoskopik tedavi, füzyonlu suture dar bir osteotomi yapılmasını takiben postoperatif kask tedavisini kapsamaktadır.

Bulgular: İlk hasta, sağlıklı ebeveynlerin ilk çocuğu olan 6 aylık erkek hastaydı. Trigonosefali tanısı aldıktan sonra genel anestezi altında operasyona alındı. Açık ameliyat gerçekleştirildi. Ameliyat sonrasında hasta, yoğun bakım ünitesinde monitorize edildi ve 3 gün sonra herhangi bir komplikasyon olmadan taburcu edildi. İkinci yıl kontrol görüntülemesinde kranyumun fizyolojik gelişimde olduğu görüldü.

İkinci hasta, gestasyonun 40. Haftasında vajinal doğumla dünyaya gelen 3 aylık hastaydı. Trigonosefali tanısı aldıktan sonra genel anestezi altında operasyona alındı. Endoskopik yaklaşımla mikrocerrahi operasyonu gerçekleştirildi. Ameliyat sonrasında hasta yoğun bakım ünitesinde monitorize edildi ve 3 gün sonra herhangi bir komplikasyon olmadan taburcu edildi. Erken dönem kraniyal BT'de ekstra komplikasyona rastlanmadı ve trigonosefalinin iyiye gittiği gözlendi.

Sonuç: Endoskopik yaklaşımla yapılan cerrahi güvenli ve yararlı olmasının yanında; ameliyat süresinin kısalması, daha az hastane yatış günü nedeniyle masraflarda azalma ve daha az kan transfüzyonu imkanı sunar. Ancak; minimal invazif tedavi, yalnızca erken tanı alan ve ameliyat sonrası dönemde kask tedavisine koopere olabilecek hastalarda mümkündür.

Anahtar kelimeler: Kraniyosinostoz, Trigonosefali, Endoskopi Yardımlı Cerrahi

INTRODUCTION

Craniosynostosis refers to a premature fusion of one or more cranial vault sutures. It occurs in up to 1 in 2000 live births, and may be characterized as single- or multi-suture, as well as syndromic or non-syndromic. Trigonocephaly is the second most frequent type of craniosynostosis (incidence, 1: 5,200) and is associated with a remarkable occurence of intracranial abnormalities and neuropathology.

Over the past decade, the use of minimal incision craniectomy for craniosynostosis has increased as an alternative to full exposure craniectomy or calvarial vault reconstruction. Differences in utilization and opinions regarding outcomes of these techniques differ among surgeons specializing in the care of these conditions. Outcome variables debated include magnitude and durability of head shape improvement, cost, neurodevelopmental trajectory, burden of care to patient, and intra- and postoperative complication rates. Although a number of studies have reported various outcomes in endoscopic or open craniosynostosis surgeries, few have directly compared results of the two on a large cohort of patients.

CASE REPORT

Case 1

The first patient is a 6-month old male, first-born to healthy parents. Birth weight was 3250 gr and his body length was 50 cm. There were no complications during his birth. Head circumference measured 32 cm. Craniofacial Computerized Tomography (CT) scan with 3D reconstruction revealed metopic suture total synostosis (Figure 1a-d). After he was diagnosed as trigonocephaly, he underwent surgery under general anesthesia. A zigzag skin incision was made from the anterior portion of one tragus to the other. The scalp was dissected up to both temporal sides and supraorbital region. The dissection was performed using frontozygomatic and frontonasal sutures 2 cm posterior of the orbital ceilings while preserving supraorbital nerves bilaterally. The temporal muscles were dissected up to squamous sutures. A bone drill was used to perform bifrontal craniotomy and bilateral supraorbital rim osteotomy and the dura mater was dissected from the floor of the anterior fossa , while the supraorbital rim was fused to form a triangular shape. The frontal bone was also cut in half, flipped 180^0, and then sutured to the parietal and supraorbital bones to ensure anterior cranial widening (Figures 1e, f). The scalp was closed with a pericranial flap in its original position. Postoperatively the patient was monitored in the intensive care unit and discharged 3 days later without any complications. Early cranial CT showed no extra complications. Two year later he was followed up with a control computerized cranial tomography (Figures 1g-i). Second year follow-up control images revealed physiological formation of cranium.

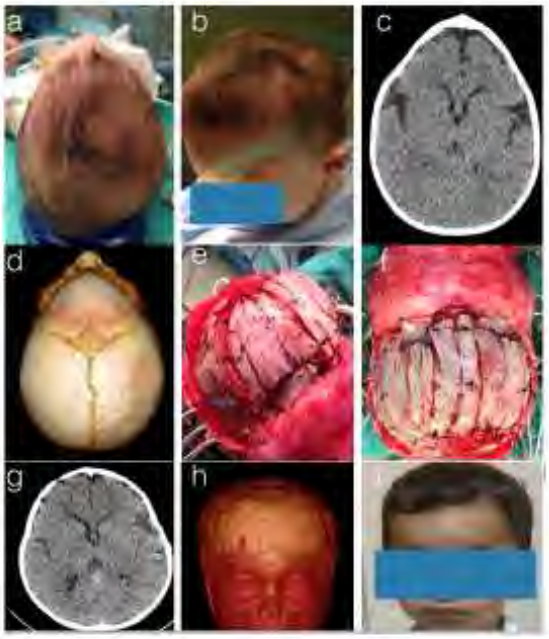

Fig1
a-b: Preoperative cranial images of Case 1
c-d: Craniofacial Computerized Tomography (CT) scan with 3D reconstruction revealed metopic suture total synostosis
e-f: Surgical pictures of Trigonocephalus. Bifrontal craniotomy and bilateral supraorbital rim osteotomy and the dura mater was dissected from the floor of the anterior fossa, while the supraorbital rim was fused to form a triangular shape.
g-i: Second year follow-up control CT images (g-h) and photo (i) revealed physiological formation of cranium.

Case 2:

The second patient is a 3-month old male, born at 40 weeks of gestation via vaginal delivery, birth weight being 3170 gr, body length at 51 cm, and head circumference measuring 30 cm, with an Apgar score of 10/10/10. His clinical examination suspected trigonocephaly without other birth defects, the diagnosis was confirmed by a cranial CT scan and the brain parenchyma presented to be normal (Figures 2a-e). The patient was kept supine with his head in a soft donut head holder. A 2- to 3-cm incision was placed just behind the hairline, perpendicular to the metopic suture, followed by a subgaleal dissection performed with the endoscope and a malleable extension on the cautery so the galea may be dissected all the way to the nasion.
A lighted retractor helped to protect the scalp from cautery injury. A single burr hole was placed over the metopic suture and was widened slightly with a Kerrison

rongeur. A 1-cm trough of bone was cut to the fontanelle. Piecemeal removal of the suture with large bone pituitary rongeurs was performed. The endoscope presented to be very useful in securing the dura not to be getting caught in the rongeurs. Proceeding down the suture, visualization became more difficult. It is also useful to switch downward curved rongeurs. The strip craniectomy needed to be dutifully performed to the nasion to ensure correction from the molding helmet. Both instruments were used for his patient. Once completed, coagulation of bone edges was performed to minimize postoperative bleeding. Thickness of the bone itself and the piecemeal removal of bone promoted greater blood loss. Postoperatively the patient was monitored in the intensive care unit and discharged 5 days later with no complications. Postoperatif control 3D computerized cranial tomography (Figures 2f,g) and third month follow-up images (Figures 2h,i) revealed physiological formation of cranium.

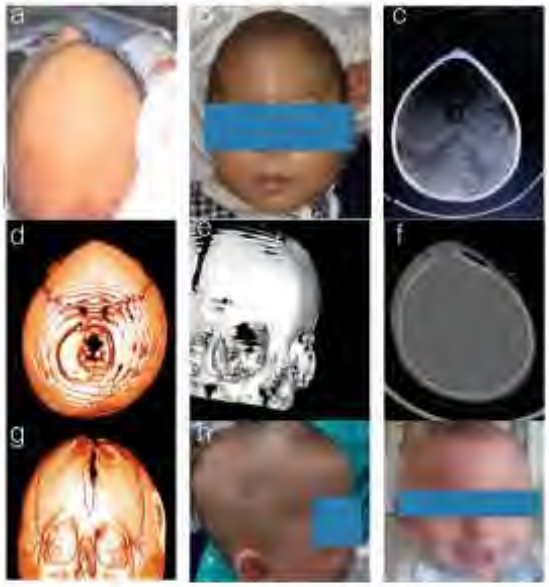

Fig2
a-b: Preoperative cranial images of Case 2
c-e: Craniofacial Computerized Tomography (CT) scan with 3D reconstruction revealed metopic suture total synostosis
f-g: Early postoperative cranial CT control images
h-i: Third month follow-up images revealed physiological formation of cranium.

DISCUSSION

The goal of craniosynostosis surgery is to remove the abnormal suture, then allow brain growth and aid reshaping the skull utilizing postoperative molding helmet. As head-shape correction occurs slowly after surgery, helmet therapy is just as important as the surgery. Surgical management of craniosynostosis has shifted from open craniectomy to minimally invasive endoscopic approaches by Barone and Jimenez (1, 2). The endoscope is used to perform the traditional strip craniectomy using very small incisions with minimal blood loss. In addition, this allows for minimal tissue disruption, and the bone generating dura and periosteum to remain largely untouched (3). Benefits of the endoscopic approach over more traditional open techniques include comparable safety and efficacy while allowing shorter operative times, reducing costs due to decreased hospital stays, and fewer blood transfusions (4, 5). However, minimally invasive treatment is only possible in cases with early diagnosis and cooperation in using a postoperative helmet therapy to allow proper manipulation of the thin bones in young infants is granted (5). Most data comparing long-term results of endoscopic versus open treatment of craniosynostosis concern isolated sagittal synostosis and resultant scaphocephaly. The use of strip craniectomies for coronal, metopic, or lambdoid sutures is a newer phenomenon (6). Due to these constraints, surgeons vary widely in their opinions regarding the proper timing and indications for choosing an endoscopic versus open approach, or whether to offer an endoscopic approach at all (7). Patients at our center are generally offered the endoscopic technique if they are under 6 months of age at time of surgery, but other factors including comorbidities, syndromes, and anticipated difficulties with postoperative helmet molding must be considered as well. While some centers perform minimally invasive craniosynostosis surgery in children aged 6 to 9 months, our practice has been to limit the use of these endoscopic

procedures to children under 6 months of age (8). Chan et al. directly compared endoscopic versus open craniosynostosis surgery by examining a cohort of 21 open and 36 endoscopic patients receiving treatment for all types of craniosynostosis. They found a mean age at surgery of 10.56 months for open patients and 4.74 months for endoscopic patients (8). After making this decision other factors have to be considered as well, such as possible complications of the surgery. Barone and Jimenez reported no infections, air emboli, intraoperative durotomies, intraparenchymal injuries, postoperative hematomas, seizures, or intraoperative deaths in their endoscopic sagittal synostosis series. However, they did describe superficial skin irritation along the incisions in 5 patients. In their endoscopic coronal synostosis series, Barone and Jimenez found no infections, sagittal sinus injury, postoperative hematomas, visual or ocular injuries, seizures, conversion to open procedures, or deaths. They reported 2 intraoperative durotomies, 2 minor scalp irritations, 3 calvarial defects, and 2 venous air emboli with a change in Doppler tones and decrease in end-tidal carbon dioxide, but no changes in blood pressure or oxygenation and of no clinical significance (9). Gociman et al. presented a series of 46 patients with non-syndromic sagittal synostosis who received endoscopic treatment and experienced no conversions to open approaches, reoperations, air emboli, cerebral parenchymal injuries, postoperative infections, hematomas, coagulopathies, CSF leaks, seizures, or deaths. They described 2 intraoperative durotomies that were repaired, and 8 patients who displayed pyrexia (10). It is important to note that there are veins bridging the dura and the bones in this region, and bipolar cautery should be used to coagulate the veins in advance of the bone removal. In our cases, none of the patients had any complications.

CONCLUSION

Minimally invasive endoscopic techniques are excellent alternatives to open craniofacial repairs, and this is now supported by nearly two decades of literature. Endoscopy assisted surgery is safe and efficent while allowing shorter operative times, reduced costs due to decreased hospital stay, and fewer blood transfusions. However, minimally invasive treatment is only possible in cases where early diagnosis and cooperation in using a postoperative helmet therapy to allow proper manipulation of the thin bones in young infants is granted.

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