Juvenile trabecular skull ossifying fibroma: case report

Fibroma osificante trabecular juvenil del cráneo: reporte de caso

***Jose D. Charry MD MSc*[1 2 4]** ***Juan S. Calle-Toro MSc*[3]**
***Cristian Rincón-Guio Mg*[1]** ***Camilo Calvache MD MSc*[4 5]**
***Jorman H. Tejada MD MSc*[4 6]** ***Juan P. Solano MD MSc*[5]**

1. Research Department, Fundación Universitaria Navarra – UNINAVARRA, Neiva, Colombia.
2. Universidad de Jaén, España
3. Radiology Division, Children's Hospital of Philadelphia, Philadelphia, USA.
4. Neurosurgery Department, Hospital Universitario de Neiva, Neiva, Colombia
5. Neurosurgery Department, Clinica Medilaser, Neiva, Colombia
6. Anesthesiology Department, Universidad Surcolombiana, Neiva, Colombia

Información del artículo

Recibido: 16 de agosto de 2022. Evaluado: 5 de septiembre de 2022. Aceptado: 10 de octubre de 2022.

Cómo citar: Charry JD, Calle-Toro JS, Rincón-Guio C, Calvache C, Tejada JH, Solano JP. Juvenile trabecular skull ossifying fibroma: case report. Rev. Navar. Med. 2022;8(2):29-34. https://doi.org/10.61182/rnavmed.v8n2a4

Resumen

Antecedentes: Los fibromas óseos son lesiones intraóseas benignas muy raras que generalmente afectan las estructuras craneofaciales y suelen aparecer entre los 10 y 15 años de edad.

Presentación del caso: Paciente de 8 años con antecedente de trauma craneal por accidente de tránsito hace 2 años. Se evidenció una masa de crecimiento lento en la región frontoparietal derecha, asociada a cefalea global intermitente de un año de evolución. El servicio de neurocirugía evaluó al paciente mediante estudios de imagen diagnóstica y lo consideró candidato para manejo quirúrgico de la lesión. El informe final de patología confirmó un fibroma osificante trabecular juvenil del hueso frontal derecho. Este tipo de tumores benignos son extremadamente raros.

Conclusión: La neuroimagen es la principal herramienta para realizar el diagnóstico, y la conducta más adecuada es la resección quirúrgica, con un pronóstico generalmente favorable. Representan un reto, ya que el diagnóstico definitivo se establece mediante el estudio histopatológico.

Palabras clave

Fibroma osificante juvenil, neurocirugía, histopatología, reporte de caso.

Abstract

Background: Bone fibroids are very rare intraosseous benign lesions that generally affect craniofacial structures and onset between aged 10 and 15 years.

Case presentation: An 8-year-old patient was admitted with a history of skull trauma by a traffic accident 2 years ago, it is evidence of a mass in the right frontoparietal region of slow growth associated with intermittent global headache of one year duration. The neurosurgery service evaluates the patient with diagnostic imaging and considers candidate for surgical management of the lesion. With final report of

Keywords

Juvenile ossifying fibroma, neurosurgery, histopatholog, case report.

pathology of juvenile ossicle fibroma trabecularis of skull in right frontal bone. This type of benign tumors are extremely rare.

Conclusion: Neuroimaging is the main tool to perform the diagnosis, and the most appropriate behavior is surgical resection, having a favorable prognosis. They are a challenge because the final diagnosis is provided by the histopathology study.

Corresponding author:
Jose D. Charry MD MSc
Correo: danielcharry06@gmail.com

Background

Bone fibroids are very rare intraosseous benign lesions, usually located in the craniofacial structures such as the jaw, orbits, paranasal and frontal bones (1). Others regions that can be affected by this type of lesion are the ethmoidal, sphenoidal and temporal bones (REF). The pathogenesis of these lesions has been explained as the replacement of normal bone with encapsulated ossified fibrous stroma (2). One of the types described is the Juvenile Trabecular Ossifying Fibroma (JTOF), which normally has an asymptomatic course, can manifest with changes in the facial anatomy (3). Individuals with this type of lesions are usually between 10 to 15 years of old (4,5).

Case presentation

A 8-year-old male patient was admitted with a history of head and facial trauma, loss of consciousness secondary to a motor vehicle accident 48 hours before. Initially, he was treated at a first level center where he underwent observation and no radiological studies were performed. One month after the accident, he presented with a mass in the right Fronto-Parietal region, which had a slow growth rate, associated with intermittent global headache. The mother of the patient refers that last year he had problems at school with lower academic performance, periods of physical and verbal aggression, encopresis and fever. Because of this, she consulted a second level hospital where it was found a deformity in the right frontoparietal region without systemic inflammatory response syndrome (SIRS) and without any other neurological sign. It was found there in a skull X-Ray a radiolucent Fronto-Parietal right lesion. Then, he was referred to a fourth-level hospital for a computerized tomography (CT) scan of the head. This image showed a right frontal intraparenchymal hemorrhage associated with mass effect (Figure 1). The Neurosurgery was consulted and evaluated the patient and orders a contrasted enhanced magnetic resonance image (MRI), which showed an hyperdense lesion in the right frontal region with ipsilateral compression of the brain, also associated passage of an hyperdense tissue into the ocular cavity, with an avid enhancement with the contrast medium and no surrounding edema. Based on these findings, a patient with a neoplastic bone lesion invading the orbital roof with neovascularization (meningioma) was considered. This case was

then presented in a committee and was decided to perform a panangiography with embolization prior to surgery to reduce the risk of bleeding. Oncology, ophthalmology and otorhinolaryngology were also involved to determine paranasal sinus involvement. Otorhinolaryngology and Ophthalmology did not find abnormalities, Oncology group considered resection of the tumor pertinent, then neurosurgery group proceeded with the treatment. Cerebral panangiography showed intracranial projections, displacement to the left of the anterior cerebral arteries in relation to a large lesion occupying the right frontal space that did not receive pial irrigation. The injection of the right external carotid artery showed a giant lesion at the right basal frontal level, with high vascularization, nourished by branches of the middle meningeal artery, anterior meningeal and ethmoidal branches. There were abundant intratumoral venous lakes. Subsequently, the embolization was performed where devascularization of the lesion was achieved with preservation of all the vascular territories.

Surgical procedure:
An hemisoutar extended incision was made. Dissection by planes controlling the bleeding of the hypervascularizated radicle was performed. The external table was exposed and craniotomy was performed around the tumor region, the complete pathological piece was resected and extracted. Following epidural hemostasis was performed in the anterior and middle fossa with orbital roof milling to achieve complete removal of the tumor portion that compressed the orbit region. It was removed the portion of the tumor with duramater that was partially located in the lower right of the sphenoid wing. Partial durosinangiosis was found in the basal right frontal parenchyma, the synangiosic meningeal vessels were ligated and hemostasis was supplemented with fibrillar surgicell. The area extirpated was replaced with a 7.5x7.5 cm of dura mater that was resected using the surplus. After this, mesh for orbitoplasty was placed, fixed with 3x4 mm screws until enough support was achieved. Multiple escape points for cerebrospinal fluid were found despite the anterior, and it was decided to apply a fibrin sealant to close the cerebrospinal fluid fistula; cranial flap was replaced and the planes were closed in a conventional manner.

Macroscopic findings:
Infiltrating tumor from the right frontal bone table to the middle fossa including the roof of the orbit, extrinsic compression of the right orbit and its contents, infiltration and hyperplasia with excessive vascularization of the underlying dura and marked collapse of the encephalon by mass effect (Figure 2).

Microscopic findings:
It was evidenced a proliferation rich in polyhedral and fusiform cells arranged in a pattern of short beams and swirls between collagen fibers with scarce mitotic figures. Some areas were accompanied with immature osteoid trabecular cell tissue focally surrounded by osteoblasts and multinucleated giant cells. The lesion was surrounded on the periphery by a thin capsule of cortical bone. Dense fibro-connective tissue was found in the dura mater, and in one of the faces, it was found a proliferation of spindle cells with high cellularity, indicating formation of bony trabeculae (Figure 3). The diagnosis made was juvenile trabecular ossifying fibroma of the skull in the right frontal bone.

Postoperative course:
Patient had an optimal recovery, he withdrew from the intensive care unit without ventilatory support, did not present any neurological deficit. Postoperative images were performed including CT scans that where only showed local hemorrhages in the surgical bed, preserved midline and permeable base cisterns. Ophthalmology assessed the patient again and concluded absence of visual deficits. Oncology decided to continue with follow-ups. Neurosurgery team decided to discharge, without neurological deficit and follow-up. Six months later, the patient returned to his control appointment where good healing of the surgical area was evident and did not present any symptoms or complications related to the surgery.

Discussion
JTOF is an aggressive lesion, with a high rates of recurrence after surgical excision, with previous reported frequencies up to 56% (6,7), therefore making follow-up a very important part of the management (8). It is necessary to take into account the different differential diagnoses that usually have very similar clinical presentations where the only difference is the histological findings, some of these may be the conventional ossifying fibroma, fibrous dysplasia. Radiographically FOJT denotes a clear demarcation, which helped us to make the differential diagnosis with fibrous dysplasia (9,10). Treatment of choice is the surgical resection, weather complete or more conservative local resection should be performed is still controversial. Conservative resection is generally recommended because these are usually young patients with success rates that are similar to an aggressive lesion (11,12,13). Radiotherapy has not been shown to be effective reducing tumor size, but instead increases the risk of malignant conversion (4).

Conclusions
JTOF are extremely rare benign tumors, affecting usually skull bones. Neuro-Radiological images are the main studies that help to identify, describe, and define surgical approach. Diagnosis is very challenging for the clinician because the definitive diagnosis is provided by pathology due to its unique and characteristically histopathological findings.

Declarations

Ethics approval and consent to participate:
Not applicable

Consent for publication:
Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of data and material:
Not applicable

Competing interests
The authors declare that they have no competing interests

Funding
This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Acknowledgements
Not applicable.

References

1. Cañón OL, Rodríguez MJ. Fibroma osificante juvenil: reporte de un caso. MedUNAB. 2003;6(17):102-6. Disponible en https://revistas.unab.edu.co/index.php/medunab/article/view/261

2. Speight PM, Carlos R. Maxillofacial fibro-osseous lesions. Curr. Diagn. Pathol. 2006;12(1):1-10. doi: 10.1016/j.cdip.2005.10.002

3. Noffke CE. Juvenile ossifying fibroma of the mandible. An 8 year radiological follow-up. Dentomaxillofac Radiol. 1998;27(6):363-6. doi: 10.1038/sj/dmfr/4600384

4. Barrena López C, Bollar Zabala A, Úrculo Bareño E. Cranial juvenile psammomatoid ossifying fibroma: case report. J Neurosurg Pediatr. 2016;17(3):318-23. doi: 10.3171/2015.7.PEDS1521

5. Wang M, Zhou B, Cui S, Li Y. Juvenile psammomatoid ossifying fibroma in paranasal sinus and skull base. Acta Otolaryngol. 2017;137(7):743-749. doi: 10.1080/00016489

6. Williams HK, Mangham C, Speight PM. Juvenile ossifying fibroma. An analysis of eight cases and a comparison with other fibro-osseous lesions. J Oral Pathol Med. 2000;29(1):13-8. doi: 10.1034/j.1600-0714.2000.290103.x.

7. Shekhar MG, Bokhari K. Juvenile aggressive ossifying fibroma of the maxilla. J Indian Soc Pedod Prev Dent. 2009;27(3):170-4. doi: 10.4103/0970-4388.57098

8. Abuzinada S, Alyamani A. Management of juvenile ossifying fibroma in the maxilla and mandible. J Maxillofac Oral Surg. 2010;9(1):91-5. doi: 10.1007/s12663-010-0027-6

9. Eversole LR. Craniofacial fibrous dysplasia and ossifying fibroma. Oral Maxillofac Surg Clin North Am. 1997;9(4):625-42. doi:10.1016/S1042-3699(20)30355-1

10. Rai S, Kaur M, Goel S, Prabhat M. Trabeculae type of juvenile aggressive ossifying fibroma of the maxilla: Report of two cases. Contemp Clin Dent. 2012;3(Suppl1):S45-S50. doi: 10.4103/0976-237X.95104

11. Espinosa SA, Villanueva J, Hampel H, Reyes D. Spontaneous regeneration after juvenile ossifying fibroma resection: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006;102(5):e32-5. doi: 10.1016/j.tripleo.2006.03.027

12. Slootweg PJ, Müller H. Juvenile ossifying fibroma. Report of four cases. J Craniomaxillofac Surg. 1990;18(3):125-9. doi: 10.1016/s1010-5182(05)80329-4

13. Saiz-Pardo-Pinos AJ, Olmedo-Gaya MV, Prados-Sánchez E, Vallecillo-Capilla M. Juvenile ossifying fibroma: a case study. Med Oral Patol Oral Cir Bucal. 2004;9(5):456-8.

Figure legends

Figure 1. Right frontal intraparenchymal hemorrhage with mass effect.

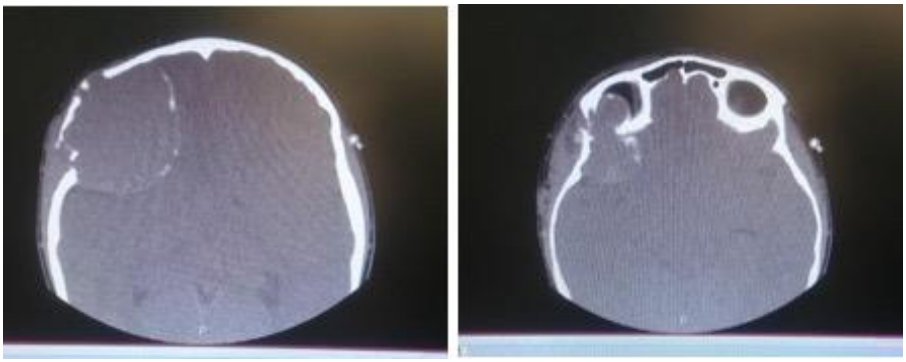

Figure 2. Tumor extracted.

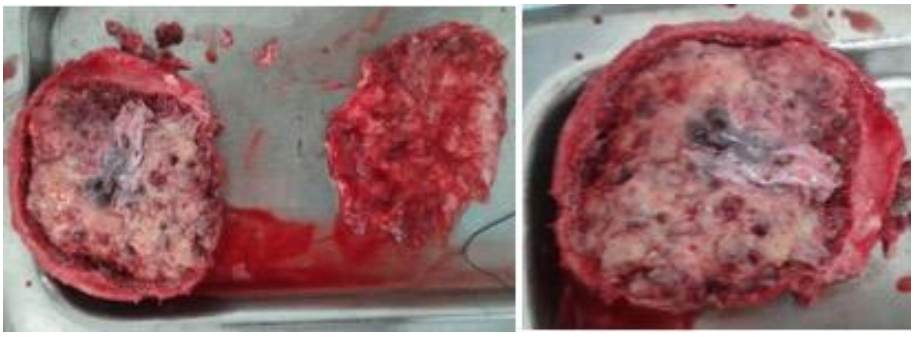

Figure 3. Dense fibro-connective tissue and spindle cells.