BRAZILIAN JOURNAL OF IMPLANTOLOGY AND HEALTH SCIENCES

ISSN 2674-8169

Le Fort I Osteotomy As a Surgical Approach to Remove a Juvenile Ossifying Fibroma in The Maxillary Sinus: Pediatric Case Report

Mateus Ferreira Nogueira[1]; Oliver Renê Viana de Jesus[2]; Maysa Ferreira Nogueira[3]; Maria Carolina Cabral de Albuquerque[4]; Mateus Ferreira Oliveira[5]; Luis Philippe Lopes Donato[6]; Pedro Thalles Bernardo de Carvalho Nogueira[7]

CASE REPORT

ABSTRACT

Ossifying fibroma (OF) is a well-defined benign neoplasm formed by different fibrocellular tissue and mineralized material. The OF clinical presentation is commonly an asymptomatic slow-growing swelling, mainly affecting the posterior mandibular region. Radiographically, this condition is characterized as a well-defined lesion with radiopaque and radiolucent areas. Histopathology shows dense cellular fibrous stroma and characteristic spheroidal calcifications called psammoma bodies. Regarding pediatric patients, the Juvenile Ossifying Fibroma (JOF) is an uncommon benign fibro-osseous lesion with fast growth that impacts the facial skeleton, as it has a high-frequency potential. It occurs in early life, and approximately 79% are diagnosed before the age of 15 years. It affects both genders equally, and the maxilla is more involved compared to the mandible, there are cases also discovered in the orbit and skull. Surgical intervention to remove the tumor is the treatment of choice. The article presents a case of a pediatric patient who had the diagnosis of Juvenile Ossifying Fibroma and was submitted to surgical removal. The Le-Fort I Osteotomy approach proved to be safe, as it allows direct access to the base of the skull, allows good visualization and tumor exposure, and also allows easy tumor manipulation and curettage, enabling the preservation of facial aesthetics.

Keywords: Oral Surgery; Maxillofacial Surgery; Le Fort Osteotomy; Ossifying Fibroma.

Osteotomía de Le Fort I Como Abordaje Quirúrgico para Extirpar un Fibroma Osificante Juvenil en el Seno Maxilar: Reporte de un Caso Pediátrico

RESUMEN

El fibroma osificante (FO) es una neoplasia benigna bien definida formada por diferente tejido fibrocelular y material mineralizado. La presentación clínica de la FO suele ser una inflamación asintomática de crecimiento lento, que afecta principalmente a la región mandibular posterior. Radiográficamente, esta afección se caracteriza por ser una lesión bien definida con áreas radiopacas y radiolúcidas. La histopatología muestra un estroma fibroso celular denso y calcificaciones esferoidales características llamadas cuerpos de psammoma. Cuando se trata de pacientes pediátricos, el Fibroma Osificante Juvenil (FJO) es una lesión fibroósea benigna poco común y de rápido crecimiento que impacta el esqueleto facial, ya que tiene un potencial de alta frecuencia. Ocurre en las primeras etapas de la vida y aproximadamente el 79% se diagnostica antes de los 15 años. Afecta a ambos sexos por igual, y el maxilar está más involucrado en comparación con la mandíbula, también se descubren casos en la órbita y el cráneo. La intervención quirúrgica para extirpar el tumor es el tratamiento de elección. El artículo presenta el caso de un paciente pediátrico que tuvo diagnóstico de Fibroma Osificante Juvenil y fue sometido a extirpación quirúrgica. El abordaje de Osteotomía Le-Fort I demostró ser seguro, ya que permite el acceso directo a la base del cráneo, permite una buena visualización y exposición del tumor, y también permite una fácil manipulación y legrado del tumor, permitiendo preservar la estética facial.

Palabras-clave: Cirugía Bucal; Cirugía Maxilofacial, Osteotomía Le Fort; Fibroma Osificante.

Dados da publicação: Artigo recebido em 27 de Dezembro e publicado em 07 de Fevereiro de 2024.

DOI: https://doi.org/10.36557/2674-8169.2024v6n2p650-658

Autor correspondente*: Oliver Renê Viana de Jesus -* *vianawork@hotmail.com*

Introduction

Ossifying fibroma (OF) is a well-defined benign neoplasm formed by different fibrocellular tissue and mineralized material. Female patients aged between 20 and 40 years are the most affected. The OF clinical presentation is commonly an asymptomatic slow-growing swelling, mainly affecting the posterior mandibular region. Radiographically, this condition is characterized as a well-defined lesion with radiopaque and radiolucent areas. Histopathology shows dense cellular fibrous stroma and characteristic spheroidal calcifications called psammoma bodies [6, 7].

Regarding pediatric patients, the Juvenile Ossifying Fibroma (JOF) is an uncommon benign fibro- osseous lesion with fast growth that impacts the facial skeleton, as it has a high-frequency potential [4]. It occurs in early life, and approximately 79% are diagnosed before the age of 15 years [4]. It affects both genders equally, and the maxilla is more involved compared to the mandible, there are cases also discovered in the orbit and skull. Usually, the studies reveal that approximately 85% of cases were found in the face, while the calvarium and extracranial sites accounted for 12% and 3% [4].

According to the newly released 2017 WHO classification, there are 3 variants of ossifying fibromas: cemento-ossifying fibroma, juvenile trabecular ossifying fibroma, and juvenile psammomatoid ossifying fibroma [7, 8]. JOF is locally aggressive and associated with cortical osteolytic modifications, being able to get involved in closely related anatomical structures too. JPOFs are rare benign tumors, but they can be aggressive and invade vital structures such as the orbit and the cranial base [8].

Surgical intervention with excision to remove the tumor is the treatment of choice, the scientific evidence also reports that a radical surgical approach has proven better at decreasing the level of recurrence compared with conservative surgery [6, 7, 8, 9, 10, 11]. This article presents a case of a pediatric patient who had the diagnosis of Juvenile Ossifying Fibroma and was submitted to surgical removal utilizing the Le Fort I osteotomy to access the tumor after the diagnosis.

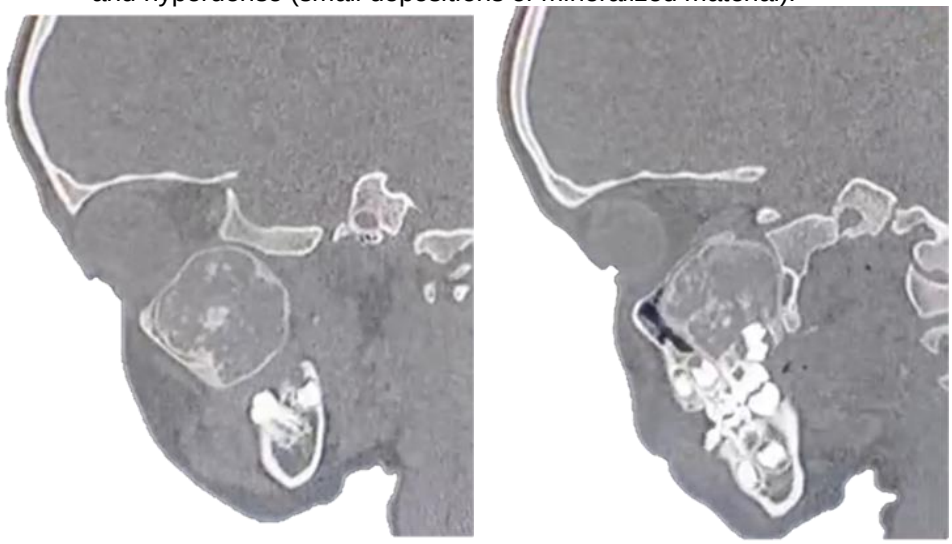
Case Report

A 7-year-old female was referred to the Department of Oral and Maxillo-Facial Surgery outpatient clinic at the Hospital in Maceió, Brazil. The responsible for the patient reported the first signs of a possible unknown pathology, as she presented to the service with complaints of blurred vision, and increased volume in the left periorbital region and left maxilla, respectively. During the examination, all the needed physical exams were executed and all the clinical exams were requested to begin the diagnosis process.

After receiving the first exam (Face CT), was possible to notice the modified tissue on the midface region characterized by a delimited lesion with hypodense and hyperdense aspects. This tumoral lesion was found extending throughout the region of the left maxillary sinus, causing expansion of the bone cortical, forming a volume increase in the face, and raising the orbit floor.

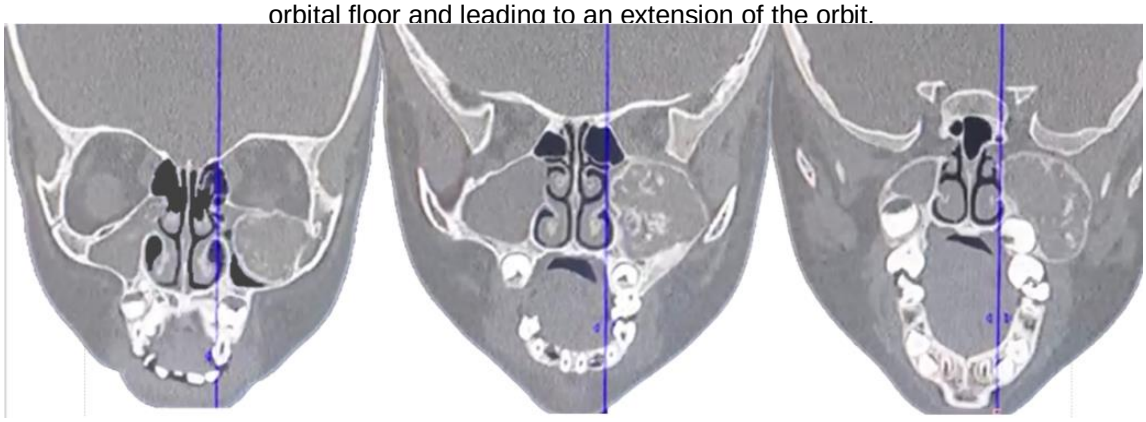
The pathology also caused a growth of intraocular pressure. The tomographic exam demonstrates a nitid lesion expansion in different anatomic sections showing the orbital expansion and other important aspects that demanded a crucial surgical method (**figures 1, 2, and 3**). The treatment management, significantly, initiates with the understanding of how the surgical technique that will be applied can impact the prognosis of the face development, considering the pediatric patient's quality of life.

Figure 1. The sagittal section reveals a delimited lesion with mixed aspect hypodense and hyperdense (small depositions of mineralized material).



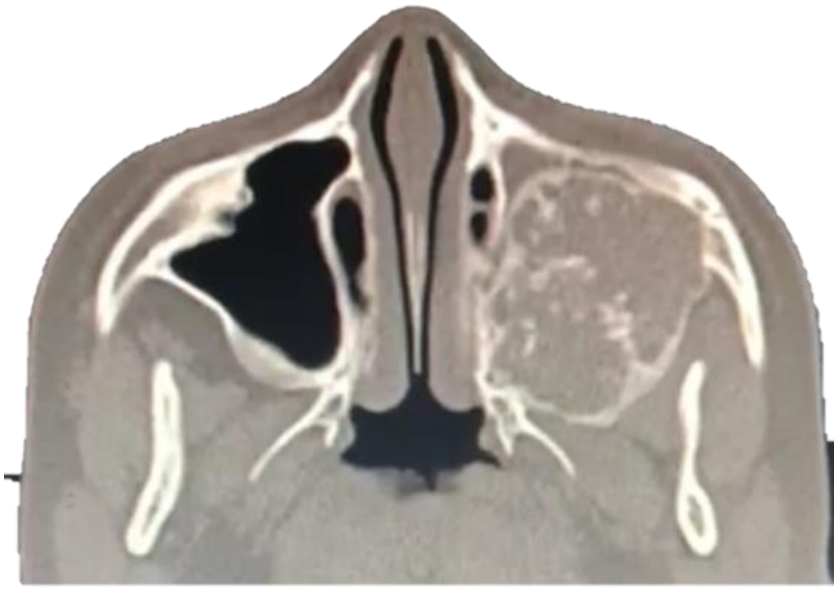
Source: authors, 2023.

Figure 2. Coronal section: expansion of the lesion is observed, causing the rise of the orbital floor and leading to an extension of the orbit.



Source: authors, 2023.

Figure 3. Axial section: the lesion is inside of the entire left maxillary sinus.



Source: authors, 2023.

The incisional biopsy was performed 30 days before the tumor removal. During the biopsy access using a left vestibular approach, an osteotomy and curettage were made to do the histopathological study, which detected the final diagnosis classifying the pathology as a Juvenile Ossifying Fibroma (JOF). The chosen treatment was total resection of the lesion using Le-Fort I Osteotomy with the assistance of Piezo Ultrasonic.

Tumor resection and curettage were performed to preserve the orbital floor, to avoid hypophthalmia, as it is one of the most common complications associated with orbital trauma (even if the fracture was induced to access a tumor on the midface region). Additionally, the Le Fort I osteotomy was made higher

than usually performed to reduce the risk of trauma at the permanent teeth germs and prevent problems during the dental change and eruption. The prognosis showed a positive patient recovery and the technique used was able to prevent another sort of radical approach.

Discussion

Ossifying fibromas are benign fibro-osseous neoplasms impacting the jaws and the craniofacial skeleton. Most JOFs, including both variants, revealed bone expansion, were painless, and presented no cortical perforation and no secondary aneurysmal bone cyst. The surgical resection of JOFs resulted in a virtual absence of recurrence. Recurrence of these lesions has been associated with an incomplete excision due to the infiltrative nature of the tumor borders. The studies express that enucleation and curettage had a considerably high recurrence rate, regardless of the anatomical location or the variant type of the lesion [11]. Although the surgical resection of JOFs resulted in a virtual absence of recurrence, enucleation followed by peripheral osteotomy/curettage should be the treatment of choice for both JOF variants to avoid the disfigurement usually associated with surgical resection [11].

Le Fort I osteotomy consists of 2 horizontal osteotomies above the dental apex from the piriform fossa to the pterygomaxillary fissure, associated with the osteotomy of the pterygoid plate, the nasal septum, and the medial wall of the maxillary sinus. There is no consensus about how to approach better skull base, midface, and rhinopharynx posterior portion tumors. Localization, size, involved areas, adjacent soft tissues, and surgeon skill lead to an adequate surgical approach for each patient. Other advantages of this technique include wide surgical exposure with straight visualization of resection margins, reconstruction possibilities with bone grafts, orthognathic corrections, easy bleeding control, surgical reoperation, and low rates of complications [1, 2, 3].

The nasopharynx, pterygopalatine fossa, and nasal fossa are difficult areas to gain wide surgical access. The transverse maxillary osteotomy with down fracturing of the entire palate and inferior maxilla has recently been adopted as a surgical option. Simultaneous bilateral wide surgical exposure is achieved in the maxillary, ethmoidal, and sphenoidal sinuses, nasal fossa, clivus,

pterygopalatine fossa, and medial portion of the infratemporal fossa. Compared with other popular techniques, the transverse maxillary osteotomy provides excellent exposure for angiofibromas, clivus tumors, and other tumors of the central base of the skull and midface regions [5, 6, 8, 9].

Based on a recent literature review, Le Fort I osteotomy can be used safely as an approach for tumor resection in areas of difficult access, allowing a wide exposition with direct visualization of lesion margins and promoting total resection. The low rates of recurrences and complications prove the efficacy of this technique which allows repetitions, reconstructions, and orthognathic corrections at the same surgical time. It can be considered uncomplicated access, which could be better performed by a multidisciplinary surgical team, such as Oral and Maxillofacial Surgery, Otorhinolaryngology, Neurosurgery, and Head and Neck Surgery, aiming for a better outcome, with patient resolution without aesthetical sequel on the face. [1, 2, 3, 6, 9]

Final Considerations

This procedure has worldwide acceptance for orthognathic surgery and is easily adapted to head and neck oncologic surgery. The Le-Fort I Osteotomy approach proved to be safe, as it allows direct access to the skull's base, good visualization and tumor exposure, easy tumor manipulation, curettage, and enables the preservation of facial aesthetics, respectively, furthermore, all the patient appearance must be preserved and using the surgical technique has provided this. Complications Attributable to the Le Fort I osteotomy reported by authors are minimal, but as a surgery like any other, there is always a possibility of interferences.

Conflict of interest: the authors declare no conflict of interest.

Disclosure: none.

Disclaimers: none.

References

1. SAILER, Hermann F.; HAERS, Piet E.; GRÄTZ, Klaus W. The Le Fort I osteotomy as a surgical approach for removal of tumours of the midface. **Journal of Cranio-Maxillofacial Surgery**, v. 27, n. 1, p. 1-6, 1999.

2. LEWARK, Tyler M. et al. Le Fort I osteotomy and skull base tumors: a pediatric experience. **Archives of Otolaryngology–Head & Neck Surgery**, v. 126, n. 8, p. 1004-1008, 2000.

3. QUIRINO, Lilian Caldas et al. Le Fort I Down-Fracture for Management of Midface Tumors. **Journal of Craniofacial Surgery**, v. 31, n. 6, p. e608-e612, 2020.

4. FOMETE, Benjamin et al. Juvenile aggressive psammomatous ossifying fibroma of the maxilla: A report of three cases and review of the literature. **Sahel Medical Journal**, v. 25, n. 1, p. 28, 2022.

5. BELMONT, Judson R. The Le Fort I osteotomy approach for nasopharyngeal and nasal fossa tumors. **Archives of Otolaryngology–Head & Neck Surgery**, v. 114, n. 7, p. 751-754, 1988.

6. STUDART SOARES, Eduardo Costa et al. The use of Le Fort I Approach in the resection of an extensive ossifying fibroma. **Oral and maxillofacial surgery**, v. 17, p. 209-212, 2013.

7. SARODE, Sachin C. et al. Juvenile psammomatoid ossifying fibroma: a review. **Oral oncology**, v. 47, n. 12, p. 1110-1116, 2011.

8. NGUYEN, Sally et al. Juvenile psammomatoid ossifying fibroma: a radiolucent lesion to suspect preoperatively. **Radiology Case Reports**, v. 14, n. 8, p. 1014-1020, 2019.

9. ADHAM, Marlinda; DEWI, Dwi Juliana. Comparison radical surgery versus conservative surgery to decrease post-operative recurrence in ossifying fibroma: systematic review. **Journal of Oral Medicine and Oral Surgery**, v. 26, n. 4, p. 44, 2020.

10. RABELO, Nicollas et al. Orbit ossifying fibroma–Case report and literature review. **Surgical neurology international**, v. 11, 2020.

11. CHRCANOVIC, B. R.; GOMEZ, R. S. Juvenile ossifying fibroma of the jaws and paranasal sinuses: a systematic review of the cases reported in the literature. **International journal of oral and maxillofacial surgery**, v. 49, n. 1, p. 28-37, 2020.