

Case report

Surgical treatment in a patient with ossifying fibroma in the left nostril

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Javier Isaac Molina Velásquez^{1*}, Jorge Alexander Mercado Lara², Adán Américo Fuentes Canales³

1-San Miguel Regional Hospital, Salvadoran Social Security Institute, San Miguel, El Salvador.

2-Specialty Clinic, Salvadoran Social Security Institute, San Salvador, El Salvador.

3-Self-employed, San Salvador, El Salvador.

*Correspondence

✉ djavier_molina100@hotmail.com

1.  0000-0002-0520-2707

2.  0009-0002-5243-1411

3.  0000-0001-5771-8628

Abstract

Case presentation. A 43-year-old female patient, with an 8-month history of presenting with a tumor on the nasal dorsum, which has been progressively increasing in size, presenting left rhinorrhea, and pain in the nasal dorsum region. Asymmetry is observed in the nasal dorsum, with a tumor in the left external nasal region measuring 2 x 2 cm, hard. In the computed tomography of the paranasal sinuses, a tumor is noted that affects the bone wall of the nasal fossa and the anterior and medial wall of the upper left jaw. **Treatment.** A facial degloving plus left medial maxillectomy was performed, as a finding, a left nasal tumor affecting the lateral wall of the left nostril, the bone of the nostril and the anterior and medial wall of the upper left maxilla. The biopsy described a benign osteofibrous proliferation with several osteoclast-like multinucleated giant cells, findings compatible with ossifying fibroma. **Outcome.** In the immediate post-surgery, he presented a good evolution with a progressive decrease in facial edema and improvement in pain. A week later he was followed up with good clinical progress, a decrease in facial edema was observed and no nasal bleeding was evident.

Keywords

Fibroma, Ossifying, Nose, Paranasal Sinuses..

Resumen

Presentación del caso. Paciente femenina de 43 años, con historia de presentar un tumor en el dorso nasal de ocho meses de evolución, que ha incrementado de tamaño de forma progresiva, y se caracteriza por rinorrea izquierda y dolor en región nasal. Se observa asimetría y una tumoración en región nasal externa izquierda que mide 2 x 2 cm, de consistencia dura. En la tomografía computarizada de los senos paranasales, se observa una tumoración que afecta la pared del hueso propio de la fosa nasal y la pared anterior y medial del maxilar izquierdo superior. **Intervención terapéutica.** Se realizó un *degloving* facial más maxilectomía medial izquierda, como hallazgo, una tumoración nasal izquierda que afecta la pared lateral de fosa nasal izquierda, el hueso propio de fosa nasal y la pared anterior y medial de maxilar izquierdo superior. En la biopsia se describe una proliferación osteofibrosa benigna con varias células gigantes multinucleadas tipo osteoclastos, hallazgos compatibles con fibroma osificante. **Evolución clínica.** En el posquirúrgico inmediato, la paciente presentó buena evolución con disminución progresiva del edema facial y mejoría del dolor. A la semana, se le dio seguimiento con buena evolución clínica, se observó disminución del edema facial y no se evidenció sangrado nasal.

Palabras clave

Fibroma Osificante, Nariz, Senos Paranasales..



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Tratamiento quirúrgico en una paciente con fibroma osificante en fosa nasal izquierda

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No conflicts of interests.

Introduction

Ossifying fibromas are benign fibro-osseous neoplasms that primarily affect the jaws and craniofacial skeleton.ⁱ⁻ⁱⁱⁱ Ossifying fibromas are usually benign lesions in which normal bone is replaced by fibrous tissue containing varying amounts of mineralized material resembling bone.^{iv} It occurs primarily in patients from the second to the fourth

decade of life, with a higher prevalence among women, and it affects the mandible more than the maxilla, and rarely occurs in the paranasal sinuses.^{iv}

Fibromas can be divided into the conventional form of ossification, also called cemento-ossifying fibroma, and two distinct juvenile ossifying fibromas: juvenile trabecular ossifying fibroma and juvenile psammomatoid ossifying fibroma.ⁱⁱ

In the case of juvenile psammomatoid ossifying fibroma, it usually occurs in the maxilla, whereas juvenile psammomatoid ossifying fibroma has a predilection for the paranasal sinuses.ⁱⁱ

Among the most common locations are the frontal sinus and the ethmoid sinus.^v In the study by Magboul *et al.*, where seven patients were evaluated, nasal obstruction was reported in all cases (100 %). Headache was present in five patients (71.4 %), while other clinical symptoms, including sneezing, snoring, periorbital swelling, proptosis, visual defects, and seizures, were detected in one case each.^v

In the study by Dong D. *et al.*, 44 patients with ossifying fibroma were included, 20 were men (45.5 %) and 24 were women (54.5 %) with the age of onset being 24.5 (5-62) years.^{vi} The patients had an age of onset younger than 20 years, in 38.7 %, and those with an age of onset younger than 30 years, accounted for 59.1 %. Thirty patients (68.2 %) were treated for craniofacial malformation, and eight patients (18.2 %) were treated for ocular symptoms.^{vi}

It is important to note that juvenile psammomatoid ossifying fibroma is a rare benign fibro-osseous tumor that occurs in a wide range of age groups, with its most common sites being the orbital bones and paranasal sinuses, followed by the maxilla and mandible.^{vii}

Even though ossifying fibromas are considered benign neoplasms, they can invade local structures, including the orbits, causing a variety of signs and symptoms depending on the compressed structures. Occasionally, visual loss may occur as a result of optic nerve compression due to the tumor spreading within the orbit, which can even aggressively extend to the jaw, especially the molar branch. Facial swelling and asymmetry are the most common clinical findings. The prognosis is good provided the tumor is completely removed, depending on the location and size of the mass.^{viii-x}

Ossifying fibromas are mainly found in patients of European descent, followed by those of African descent. However, their pathogenesis remains unknown and congenital conditions, infections and trauma, mainly in childhood, are described as the main causative agents.^{xi}

Computed tomography (CT) is the gold standard for the evaluation of sinonasal osteoma, where they appear well circumscribed, as dense masses that can be homogeneous or heterogeneous depending on the histological subtype.^{xii} MRI findings are also influenced by the presence of fibrous tissue in the mass; for example, in ivory-type

osteoma, hyperintense areas may appear on T1-weighted images, while mixed or mature subtypes may show a lack of signal in all sequences.^{xii} The images allow some differentiation between osteomas and their two most common mimics, fibroma ossificans and fibrosaxii dysplasia. The ossifying fibroma, on the contrary, appears in CT well defined with a peripheral border, while a dense border and heterogeneous center can be distinguished.^{xii}

Fibrous dysplasia may have a lytic or ground-glass appearance on CT, and hypointense on T1-weighted MRI with gadolinium contrast enhancement.^{xii} On T1-weighted MRI, the ossifying fibroma will have a hypointense periphery and a hyperintense/intermediate core, while on T2-weighted MRI, the sequence will show a hyperintense signal throughout the entire zona.^{xii}

Transnasal endoscopic surgical resection is the main treatment of nasal ossifying fibroma. Because of its location, the tumor has a rich blood supply, which increases the risk of intraoperative hemorrhage and makes it difficult to accurately localize the tumor and determine boundaries intraoperatively.^{xiii} In children, the narrow nasal cavities make it difficult to achieve complete tumor resection.^{xiv}

Endoscopic sinus surgery with image navigation and lateral nasal butterfly incision resection has been considered available as a therapeutic option.^{xv} Lateral nasal butterfly incision has less bleeding and shorter operation time, but with slight swelling of the nasal face after surgery, and nasal endoscopy is a surgical method with less damage.^{xv}

Case presentation

A 43-year-old female patient with a medical history of eight months due to the presence of a tumor on the nasal dorsum, with progressive increase in size, accompanied by left-sided rhinorrhea and pain in the nasal dorsum region, with no epistaxis. She was also diagnosed with stage 5 chronic kidney disease and was on ambulatory peritoneal dialysis.

Physical examination revealed an asymmetric nose at the nasal dorsum, with a firm, 2 x 2 cm tumor in the left external nasal region. The oral cavity revealed a hyperemic pharynx with no posterior drainage. There was no evidence of lesions. A nasopharyngolaryngoscopy was performed, which revealed no tumor in the left nasal fossa. A CT of the paranasal sinuses was also performed, revealing a tumor affecting the wall of the nasal fossa and the anterior and medial walls of the left maxilla (Figure 1).

Tests were indicated prior to the surgery (Table 1). She was evaluated by the nephrology specialty, who prescribed peritoneal dialysis the day before surgery.

A biopsy of a left nasal tumor was taken two days after the consultation. It was described bulging of the middle meatus and the lateral nasal wall above the inferior turbinate, from the nasal vestibule, of hard consistency, said tumor involving the nasal dorsum and the external lateral portion of the left nose (Figure 2). The frozen biopsy was positive for malignancy with diagnosis by paraffin.

A new review of the biopsy was requested, where a benign osteofibrous proliferation was subsequently reported with several multinucleated osteoclast-like giant cells, where there was no atypia, mitoses, or

necrosis, the findings being compatible with ossifying fibroma in the left nasal fossa. component.

Treatment

The patient was scheduled for surgery and facial degloving plus left medial maxillectomy is performed six months after the consultation (Figure 3).

A nasolabial incision was made from canine to canine, columellar and intercar-tilaginous incision bilateral, with subperiosteal dissection to expose the left nasal tumor, having as findings a tumor left nasal, affecting the lateral wall of the left nostril, the bone of the nostril and the anterior and medial wall of the left maxillary superior, with dimensions of 5 x 5 cm. (Figure 4).

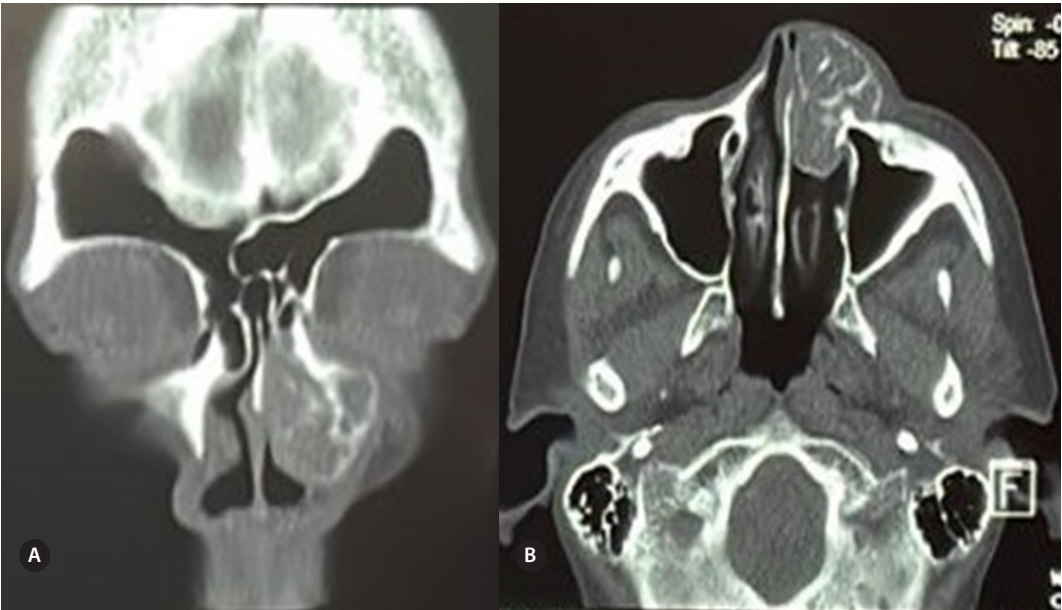


Figure 1. Computed tomography of the paranasal sinuses A. Coronal CT of the paranasal sinuses. B. Axial CT of the paranasal sinuses showing an ossifying fibroma in the left nostril

Table 1. Laboratory tests.

Laboratory tests	Result
Hemoglobin	10.3 g/dL
Leukocytes	6.74 x10 ³ /mm ³
Neutrophils	85.7 %
Platelets	214 x10 ³ /mm ³
Urea nitrogen	78 mg/dL
Creatinine	14.4 mg/dL
Glomerular filtration rate	2.7 mL/min/1.73

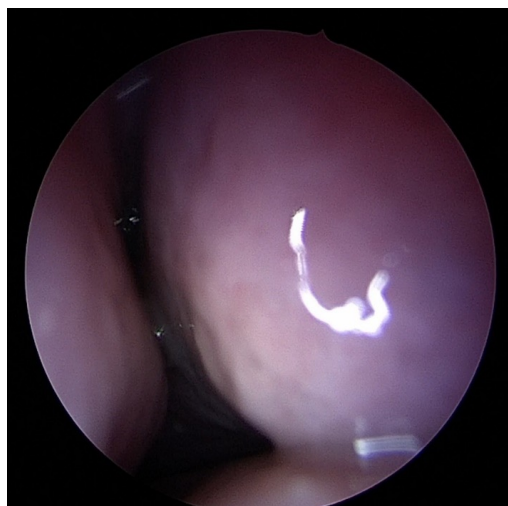


Figure 2. Endoscopic view of a tumor with bulging of the lateral wall of the left nasal fossa.

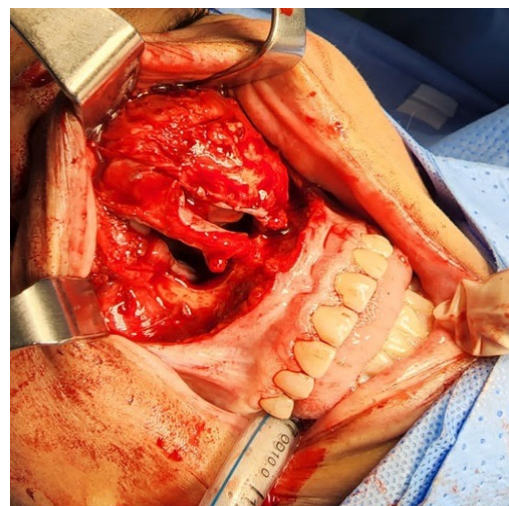


Figure 3. Facial degloving plus Left Medial Maxectomy with exposure of ossifying fibroma.

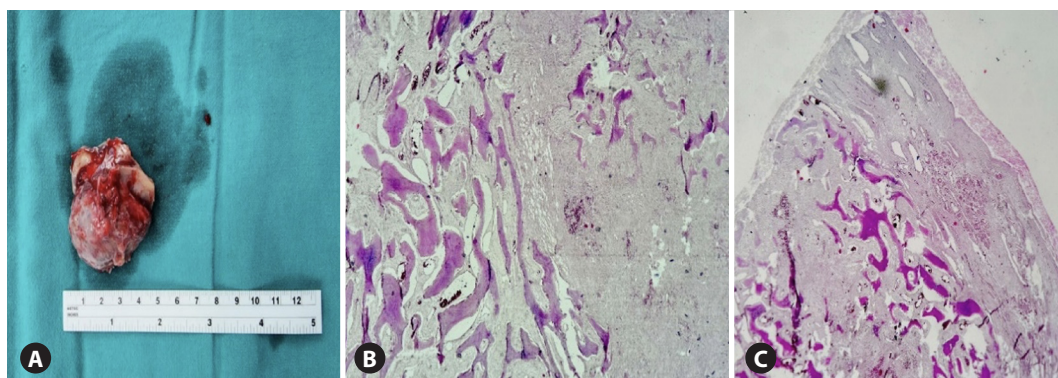


Figure 4. A. 5 x 5 cm tumor found during surgical procedure. B. Completely calcified multinodular fragment. C. Calcified sample with respiratory epithelium.

No abnormality was observed in the right nostril. It is concluded with the excision of the tumor in block plus left medial maxillectomy.

Outcome

In the immediate postoperative period, an anterior nasal packing was placed with wicks, for which he received hospital treatment with ceftriaxone 1 g every 12 hours, tramadol 50 mg every eight hours and dexamethasone 8 g every eight hours, all via intravenous and for two days, noting a progressive decrease in facial edema and improvement of pain.

Hospital discharge was indicated on the third day after the surgery. Then, one week later, the patient was scheduled for a control evaluation where she presented a good clinical evolution, in which a decrease of the facial edema was observed, and no bleeding was evidenced nasal.

She was followed up for one year at multiple controls, and to date, there is no presence of a tumor in the nasal dorsum.

Clinical diagnosis

Clinical evaluations through physical examination, imaging and histological studies allowed the diagnosis of ossifying fibroma in the left nasal fossa.

Discussion

Ossifying fibroma is a benign, well-circumscribed, slowly expanding fibro-osseous tumor characterized by a solitary lesion exhibiting focal replacement of normal bone by a variable mixture of dense connective tissue, mineralized tissue, and cementum.^{xvi} It is important to note that the lesion usually develops in the third to fourth decade of life and has a predilection for females.^{xvi}

Cases of ossifying fibroma have been described in a 12-year-old boy in the sphenoid sinus, where the tumor was removed by sphenoidotomy without median turbinectomy.^{xiv} Postoperative imaging studies in this patient confirmed that the sphenoid sinus had cleared and the tumor did not recur for two years.^{xiv}

The predominantly affected sites are the head and neck region, with the mandible being the most frequent (70 - 80 % of cases), followed by the maxilla, orbit and paranasal sinuses.^{xvi} The literature mentions that the etiology of ossifying fibroma is unknown, but it is thought to arise from incomplete migration of blast cells multipotent mesenchymal cells which they give rise to the periodontal ligament.^{xvi}

In the case of this patient, it is important to the presence of chronic kidney disease, which may predispose to the development of fibroma ossificans. Trauma is also mentioned that it could play an etiological role, at least in some cases.^{xvi}

Computed tomography is the gold standard for the evaluation of os sino-nasal theoma. They appear well circumscribed described as dense masses that can be homogeneous or heterogeneous depending on the histological subtype.^{xii}

Transvarar endoscopic surgical resection is the main surgical procedure for nasal ossifying fibroma, taking into account that, due to its location, the tumor has a rich blood supply, which increases the risk of intraoperative hemorrhage.^{xiii}

Fibro-osseous lesions are a differential diagnosis when evaluating a patient with long-standing otorhinolaryngological symptoms; in this case, the differential diagnoses of an ossifying fibroma are fibrous dysplasia, osteoma, osteoblastoma, osteosarcoma, among others; therefore, for proper management, it is important to have a preoperative image that reveals the involvement of the surrounding tissue when planning treatment, whether conservative management or surgery, being computed tomography of paranasal sinuses, the ideal tool.

These lesions should be managed according to clinical appearance, radiographic features, and related morbidity, all due to the intricate anatomical positions of such tumors within the orbits and skull base, as well as the increasing propensity surrounding nearby neurovascular pathways, making histopathologic examination and surgical excision of such tumors difficult.ⁱⁱⁱ

It is important to mention that transnasal endoscopic surgical resection is the main treatment for the resection of nasal ossifying fibroma. Endoscopic surgery of the nasal sinuses with image navigation and resection with lateral nasal butterfly incision has been considered available; which assures the total tumor excision and at the same time maintains the patient's esthetics.^{xv}

The prognosis of ossifying fibroma is good, and malignant transformation or metastasis has never been described. It is

important to note that it has a recurrence rate of 30 % to 56 %, so close long-term clinical and radiological follow-up is essential, even in the case of microscopically confirmed complete resection.^{xvi,xvii}

Ethical aspects

For the publication of this case, informed consent was obtained from the patient and the responsible person, both for the hospital care in which the physical examination was performed and for the publication of this article, with the commitment to maintain the patient's privacy, as established in the Helsinki Declaration.

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