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Management of Giant Ethmoidal Osteoma with Orbital and Intracranial Extension: Illustrative Cases and Literature Review

Driss Elidrissi[1*], Youssef Lakhdar[1], Mohamed rami[1], Omar Oulghoul[1], Mohamed Chehbouni[1], Othman Benhoummad[2], Youssef Rochdi[1], Abdelaziz Raji[1]

[1]Department of ENT and HNS Surgery, Mohammed VI University Hospital, Marrakech, Morocco
[2]Department of ENT and HNS Surgery, Faculty of Medicine and Pharmacy of Agadir, Ibn Zohr University, Agadir, Morocco

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***Corresponding author:** Driss Elidrissi
Department of ENT and HNS Surgery, Mohammed VI University Hospital, Marrakech, Morocco

Abstract | **Case Report**

Sinus osteomas are benign tumors, commonly asymptomatic and discovered incidentally during radiological assessment, but they can be responsible for orbital and endocranial complications. There are 2 options for the management: clinical observation or surgery. The choice of surgical management depends on the location, size and experience of the surgeon. In most reported cases large tumors are excised by an external approach or in conjunction with an endoscopic technique. Endoscopic treatment of such tumors is a huge challenge for the operator. We report three cases of giant ethmoidal osteomas with orbital and intracranial extension. Literature is reviewed and etiopathogenic theories, diagnostic procedures and surgical approaches are discussed.

Keywords: Giant Osteoma, Paranasal Sinuses, Endoscopic Surgery, External Approach.

INTRODUCTION

Osteomas are rare benign bone tumors. They are usually seen in the craniofacial region, particularly in the paranasal sinuses [1]. These common benign tumors of the paranasal sinuses affect 1% of the population [2, 3]. Due to their slow asymptomatic growth, in most cases they are detected accidentally, in 3% of computed tomography (CT) scans and 1% of radiographs of the sinuses [4]. When they cause symptoms, surgery is required. Osteomas usually range in size from 2 to 30 mm, an osteoma with a diameter >30 mm or weighing >110 g is considered a large or giant osteoma [5]. Given its rarity, the clinical characteristics and treatment of this disease remain controversial. In this study, we report the clinical presentation and surgical methods used to treat three patients with giant osteomas of the ethmoid, combined with a literature review.

Illustrative Cases

Case 1

A 27-year-old patient, with no medical history, consulted for unilateral left nasal obstruction evolving for two years, associated with headache and left exophthalmos. On clinical examination, a hard internal canthal tumefaction was observed, associated with a irreducible, non-pulsatile left exophthalmos. Endoscopy showed a hard mass filling the entire left nasal cavity with purulent secretions. The preoperative ophthalmic examination was unremarkable.

CT scan showed a benign left fronto-ethmoido-nasal bone tumor suggestive of a giant paramedian osteoma, measuring 75x62x40mm, responsible for compression and thinning of the left papyraceous lamina reducing the lumen of the eyeball, with extension to the horizontal lamina of the ethmoid and extension to the anterior skull base (figure 1).

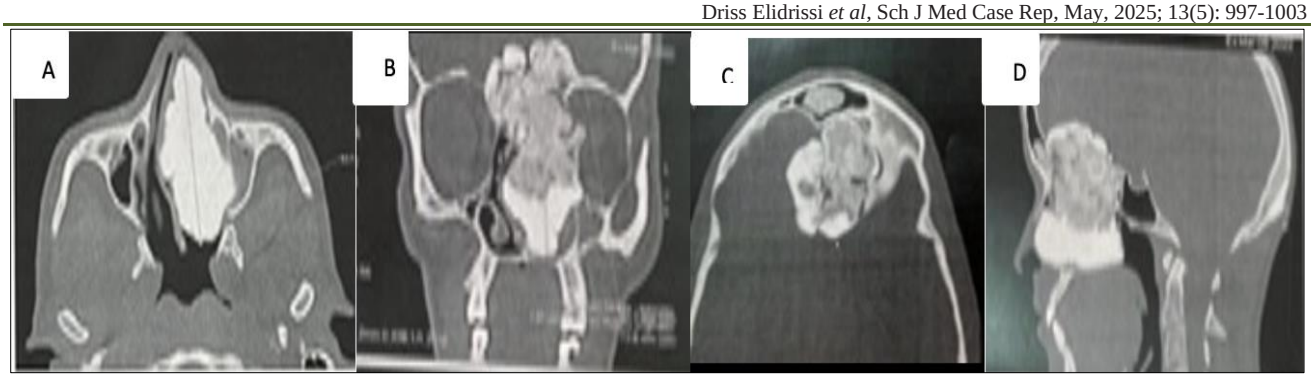

Figure 1: Axial (A, C), coronal (B) and sagittal (D) CT images showed a left fronto-ethmoido-nasal bone tumor suggestive of a giant osteoma, with thinning of the left papyraceous and extension to the anterior skull base

Osteoma was excised endoscopically using neuronavigation. The excision was carried out by drilling with excision of the entire visible part while respecting the intracranial part (figure 2).

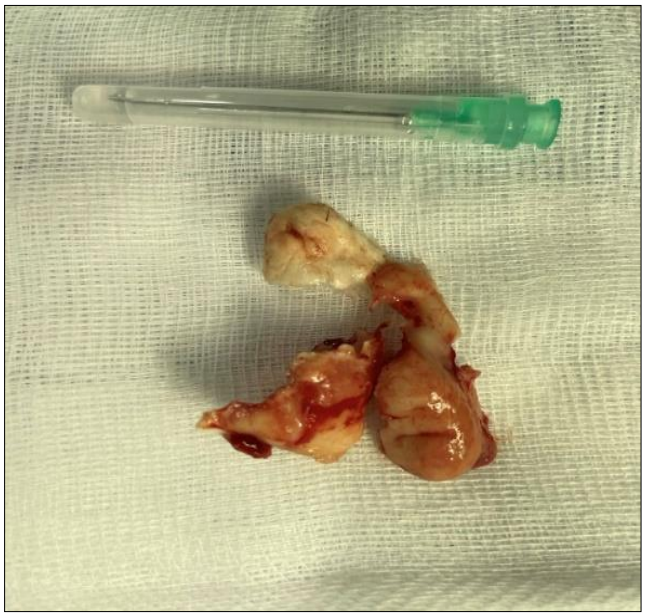

Figure 2: Piece of surgical excision of the osteoma

We opted for simple monitoring of the intracranial part and possible surgery depending on evolution. Postoperative ophthalmological examination was normal. Monitoring was clinical and radiological. After 2 years' follow up, there was no recurrence in the fronto-ethmoido-nasal region, with a stable intracranial appearance. Histological analysis confirmed the diagnosis of osteoma.

Case 2

The patient was 52-years-old women, with a 6-year history of reduced visual acuity and exophthalmos. The patient had no associated symptoms, such as epistaxis, rhinorrhea, diplopia or nasal obstruction. She had no history of surgery or trauma and was in good health. The patient was first examined in the Department of Neurosurgery and was then referred to the Department of Otolaryngology for treatment.

Clinical examination revealed irreducible, non-inflammatory right axial exophthalmos (Figure 3) with limited elevation, adduction and abduction of the eyeball, with preservation of other eye movements; in terms of visual acuity, the patient counted fingers at close range (figure 3). An endoscopic examination found no obvious lesion in the nasopharynx or nasal cavity.

Figure 3: Exophthalmos representation

CT scan revealed a voluminous hyperdense formation measuring 29x36 mm with poly-lobed contours, depending on the internal wall of the orbit, straddling the orbit and ethmoidal cells, displaces the eyeball anteriorly and causing grade II exophthalmos, compresses the optic nerve, medial and inferior rectus muscles and arrives opposite the orbital apex, without intra- or extra conical fat extension with an eyeball of normal morphology (figure 4).

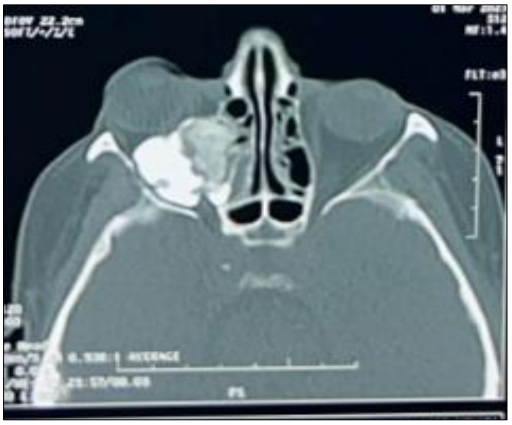

Figure 4: Axial CT scan showing ethmoido-orbital osteoma

MRI showed a condensed bone lesion measuring 24x31 mm in favor of an osteoma, with a right ethmoidal implant base and right intraorbital development with mass effect on the medial rectus muscle, optic nerve and eyeball, with no aggressivity or infiltration (figure 5).

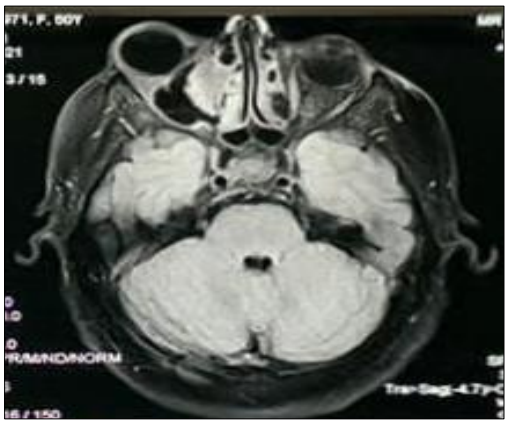

Figure 5: Axial MRI showing ethmoido-orbital osteoma

Surgery was performed in double approach, the 1st consisting of an endonasal approach using neuronavigation: First a middle meatotomy, a middle turbinectomy, an ethmoidectomy and then a drilling of the lamina papyracea of the ethmoid without opening the periorbita, thus decompressing the internal wall of the orbit.

The 2nd approach consisted of an external orbitotomy: After a right latero-orbital arciform incision and detachment down to the periosteum, we entered the orbit and discovered the osteoma, cavitated it and excised it completely (figure 6-7-8).

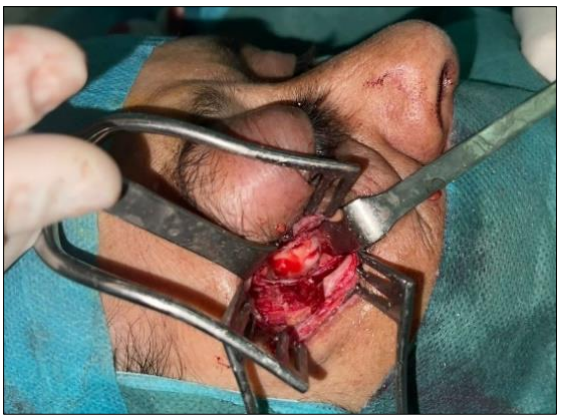

Figure 6: Discovery of osteoma by external approach

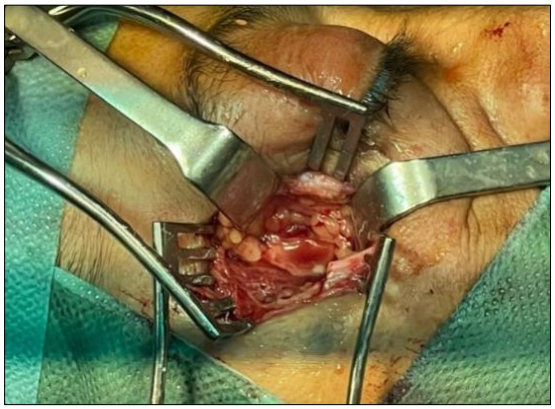

Figure 7: Operative view after taking off the osteoma

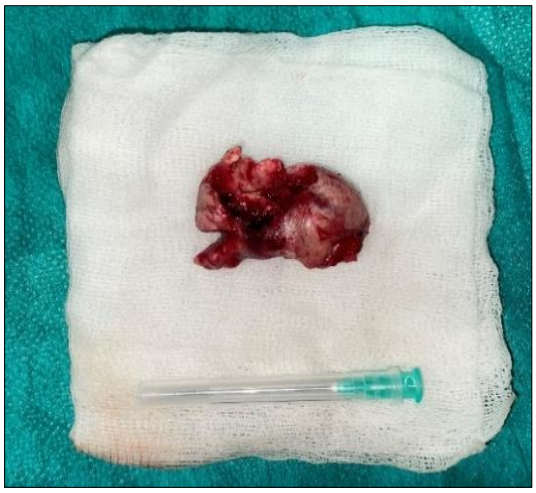

Figure 8 : Excision piece

Post-operative follow-up was straightforward, with no neuro-meningeal or ophthalmic signs observed, and the patient reported a slight improvement in visual acuity. The anatomopathological study was in favor of an osteoma with a necrotic center and no signs of malignancy.

Case 3

The patient was 51-year-old women, with a 4-year history of right unilateral nasal obstruction associated with purulent rhinorrhea, anosmia, and homolateral exophthalmos without epistaxis. Clinical examination revealed a hard internal canthal tumefaction, associated with right axial exophthalmos with inferior palpebral edema. An endoscopic examination showed a hard, white mass filling the entire right nasal cavity, with left septal deviation.

The CT scan showed an osteocondensing right ethmoido-nasal lesional process measuring 50x25x53 mm, responsible for displacement of the internal wall of the orbit with a deformed appearance of the papyraceous lamina, with endorbital extension responsible for grade 1 exophthalmos, and infiltration of the right sphenoidal sinus without endocranial extension figure (9).

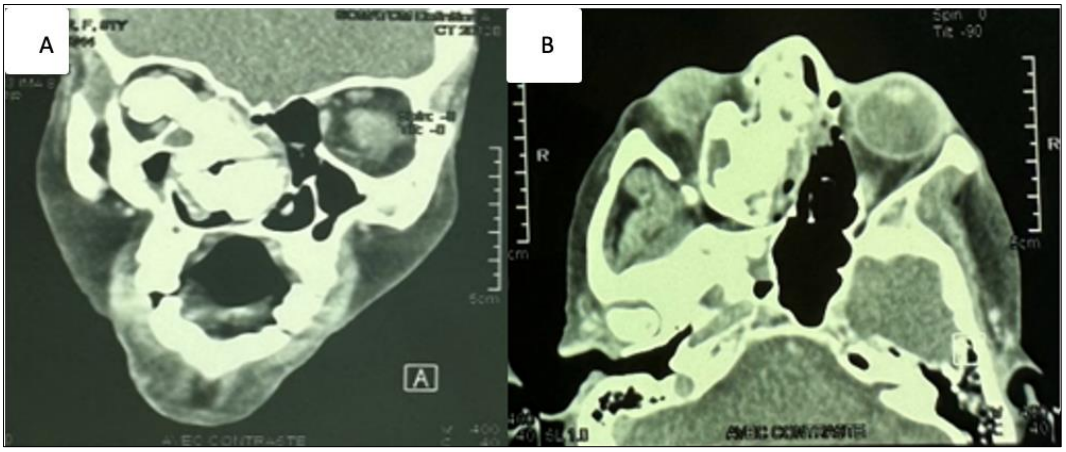

Figure 9: Coronal (A) and Axial (B) CT scan showing ethmoido-orbital osteoma

MRI revealed a limited right ethmoido-nasal lesional process with low T1 and T2 signal, heterogeneously enhancing after contrast injection, responsible for displacement of the nasal septum, with homolateral endorbital extension responsible for grade 1 exophthalmos (figure 10).

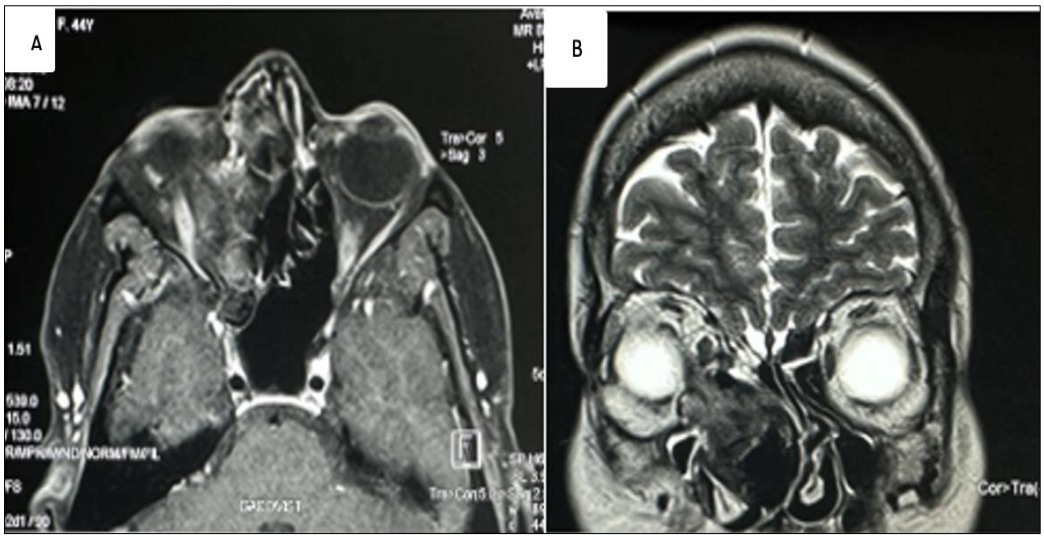

Figure 10: Axial (A) and coronal (B) MRI showing ethmoido-orbital osteoma

Osteoma was excised endoscopically with neuronavigation. Surgical exploration revealed an osteoma filling the entire right nasal cavity, arising from the middle turbinate and ethmoid (figure 11). A middle meatotomy was performed by thinning the osteoma, removing a portion attached to the middle turbinate, then drilling and thinning the second portion, located in the ethmoid, frontal sinus and middle meatus. Excision was subtotal, with a residual portion on the inner wall of the orbit (figure 12). Monitoring was clinical and radiological. After one year's follow up, there was no recurrence in the ethmoido-nasal region and the residual portion on the inner wall of the orbit remained stable.

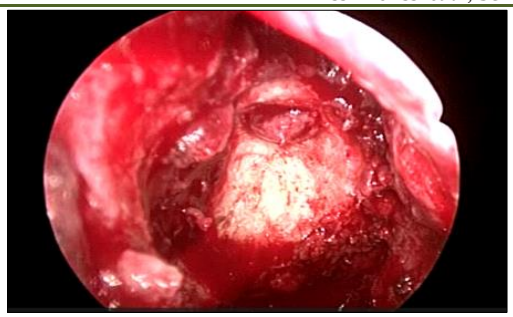

Figure 11: Endoscopic intraoperative view of osteoma

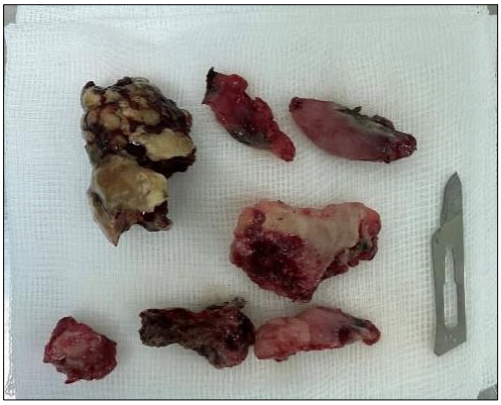

Figure 12 : Excision piece

DISCUSSION

Osteoma is a benign osteoblastic lesion and constitutes 1% of all bone tumors and 11% of benign bone lesions [1]. It's generally located in the frontal sinus (57-80%) followed by the ethmoidal sinuses (16-25%) [6]. They are more frequent in the male sex and in the 4th decade of live [7]. Tumors larger than 30 mm in diameter are considered giant tumors [5].

The origin of osteomas is unknown. Different theories exist concerning the factors responsible for their formation. A combination of traumatic, inflammatory and embryologic hypotheses is the most widely accepted at present [8].

Osteomas are characterized by slow asymptomatic growth that can take years until the first symptoms of the disease appear. Most commonly they include headaches, facial deformities, vertigo, sinusitis, disorders of nasal obstruction. The symptoms of the disease appear when normal sinus drainage becomes impaired due to the obstruction of its ostium by the tumor growth. Ocular and central nervous system symptoms result from the spread of osteomas located in the fronto-ethmoid region and can cause exophthalmos, dacryorrhea, retrobulbar pain, double vision [4].

Histologically, osteomas can be divided into ivory, mature or mixed types. Almost all osteomas contain different proportions of the three types, suggesting that they grow outwards from the center, with increasing maturation at the periphery of the tumor. This may explain why partial resection leaving residual peripheral tissue does not often lead to recurrence [9].

CT scan demonstrates a sharply defined, lobulated, homogeneously dense ossified tumor that grows by taking the shape of the sinus of origin. On MRI, ivory osteomas and mature osteomas have low signal intensity in all sequences. However mature osteomas may show similar signal intensities to bone marrow unfrequently [1].

Osteoblastoma and osteoid osteoma are usually the major differential diagnostic. Other benign fibro-osseous and cartilaginous lesions include fibrous dysplasia, ossifying fibroma, or chondroma. A biopsy of the lesion is required if the clinical and radiological presentation is unusual [8].

Indications for surgical treatment of osteomas are ambiguous. The treatment of asymptomatic patients is controversial in the literature. Because of slow growth rate (average, 1.6 mm/year), many authors have proposed clinical observation consisting of physical and radiological examinations for asymptomatic or small osteomas. Surgery is preferred when asymptomatic lesions obliterate >50% of the sinus volume, grows rapidly, shows intracranial or intraorbital extension [11].

The decision of surgical technique depends on the size, location and extension of the tumor, the presence of any complication, the surgeon's experience, and must cause as little aesthetic damage as possible. There are 3 approaches: external, endoscopic, or

combined. Considering the slow growth and rarity of recurrence, some authors suggest incomplete resection of the tumor. Recurrence rate after incomplete resection may be up to 10% [12]. If asymptomatic and small, the lesion may be left alone and observed.

Technological advances in endoscopic sinus surgery and increasing surgical experience have expanded the indications of endoscopic approaches in osteoma surgery. The major advantages of this approach are better exposure that leads to preserving vital structures such as the orbit or brain, maintenance of the natural endonasal drainage pathways, lower amounts of bleeding, reduced postoperative morbidity, shorter hospitalization time, and better cosmetic results. Major disadvantages are inadequate control of bleeding and tumor margins, greater risk of intracranial complications, and longer learning curve of the surgery [11].

On the other hand, external approaches have several disadvantages such as lower patient compliance, cicatrization, frontal pain or numbness, cosmetic deformity, higher morbidity, and longer hospital stay. The external approach has been considered as a standard procedure especially for frontal sinus and giant osteomas.

However, combined endoscopic and external procedures provide wide view of giant tumors and can be excised easily with less cosmetic defects.

Postoperative monitoring must be regular and rigorous, based on clinic and CT scan. It makes it possible to detect infectious complications, but especially recurrences.

CONCLUSION

Osteomas are rare tumors, often discovered accidentally or following intracranial or orbital complications. They are evaluated radiologically and treated surgically on extensive or symptomatic osteomas. Advances in endonasal surgery now allow a less invasive therapeutic approach, with less aesthetic sequelae.

Ethical Statement:
Compliance with Ethical Standards: The study was conducted in compliance with ethical standards.

Funding: This research received no external funding.

Conflict of Interest: There are no conflicts of interest to declare related to this research.

Ethical Approval:

While formal ethical approval was not obtained for this study, we ensured that all aspects of the research were conducted ethically and with respect for the rights and well-being of the participants.

Informed Consent:

Informed consent was obtained from all participants involved in the study, and this information has been appropriately included in the manuscript.

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