

Managing the Mass Effect: A Rare Case of Giant Ethmoid Sinus Osteoma with Orbital Extension —A Case Report

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Abstract

Background: Sinonasalosteomas are rare benign tumors that can cause clinical symptoms depending on their size and location. Osteomas are classified as giant osteomas when their size gets larger than three centimeters. In giant osteomas, severe symptoms and complications can occur if they expand into adjacent structures. **Aim:** In this case report, we highlight the diagnostic importance of CT scans of paranasal sinuses in diagnosing osteomas and the superiority of endoscopic surgery over an open approach in removing these benign tumors. **Case Presentation:** We report a rare case of giant ethmoidosteoma with extension to intraorbital facilities in 19 years old female which was causing a mass effect on the orbital space and discuss the appropriate management. **Conclusion:** Ethmoid sinus osteoma is asymptomatic until it increases in size producing a pressure effect on the eyes and sinusitis. CT imaging is the gold standard diagnostic modality. Surgical excision is the treatment of choice for ethmoid sinus osteoma. Due to the size of the lesion, endoscopic surgical excision is a less commonly employed modality, but it has advantages like earlier recovery and cosmetic benefits due to which it was the treatment employed in this case.

Keywords

Giant Osteoma, Ethmoid Sinus, Paranasalosteoma, Orbital Extension, Case Report

1. Introduction

Osteomas are benign tumors made up of mature, compact, or cancellous bones

that are well-differentiated [1]. Osteomas are relatively rare tumors that account for 1% of all bone tumors and 11% of benign bone tumors [2]. In the general population, these tumors are found in 3% of people [3] [4]. Ethmoid sinus osteoma made up about 15% - 25% of the total incidence of paranasal sinus osteoma [5]. A male preponderance exists, with a male-to-female ratio of 1.5—3:1, with the highest frequency occurring in the fourth to seventh decades of life [5].

Development of osteoma is attributed to congenital, inflammatory, or traumatic factors, but in most cases the cause of the osteoma is idiopathic. The ethmoid sinuses, after the frontal sinuses, are the most commonly affected paranasal sites by this benign neoplasm [2] [3] [4] [5] [6]. Most ethmoid sinus osteomas are asymptomatic [6]. In rare cases, the osteoma can grow and cause symptoms according to the site and size of the mass [7]. The term “giant” osteoma refers to an osteoma with a diameter greater than 3 cm [2]. Giant ethmoidosteomas are even more uncommon, accounting for 0.9% - 5.1% of all orbital tumors [8]. Cheng *et al.* in a literature review reported only 17 cases of giant ethmoidalosteomas until 2013 [2]. Humeniuk-Arasiewicz *et al.* did a literature review in 2018 and found only 37 cases of giant ethmoidalosteomas [9]. Moreover a recent literature research using PubMed and the terms “((giant) AND (osteoma)) AND (ethmoid sinus)” for the period of 2018 to 2022 showed only 4 new case reports of this disease. Although giant osteomas of paranasal sinus are uncommon, they can easily expand into the intraorbital or intracranial cavity, producing significant consequences [2]. Some cases have presented with dacryocystitis and metamorphopsia even [10].

Conventionally used open procedures were preferred especially for giant osteomas but Hung *et al.* proved that a nasoendoscopic approach in the treatment of these osteomas is safer and equally effective as the open procedures [11].

2. Case Presentation

A 19 years old female, with no previous medical illness, presented with a history of six months duration of blurring of vision, worsening for the past 3 months. It was also associated with proptosis of the left eye. She had no additional complaints of nasal discharge, fever, headache, vomiting, or photophobia. She also had no history of falls or trauma. Upon clinical examination, there was a protrusion of the left eyeball from the supraorbital view. No ophthalmoplegia was noted. The ear, nose, and throat examinations were normal. On nasal endoscopy, no abnormal findings were noted. For further evaluation, a contrast-enhanced CT scan of paranasal sinuses was performed, which showed a bony growth in the left ethmoid sinus with the size of $2.7 \times 3.2 \times 2.5$ cm, suggestive of giant osteoma of the left ethmoid sinus with orbital extension (**Figure 1(a)**, **Figure 1(b)** and **Figure 2**).

There was also left orbital extension displacing the medial rectus muscle and optic nerve laterally, and mild proptosis of the left globe. Endoscopic sinus sur-

gery was planned to remove the giant ethmoidosteoma under general anesthesia. The uncinete process was removed, and the ethmoid bulla was resected after identification of the maxillary sinus ostium. Intraoperative findings showed a huge, whitish bony mass in the left ethmoid sinus extending into the left orbital region (**Figure 3**).

The tumor could not be resected and removed as a whole due to its large size; hence it was removed in piece-meal using Kerrison bone punch, burr drill bits, and bone curette via the piriform aperture (**Figure 4**).

In this way, the giant osteoma was successfully removed using an endoscopic approach. There were no perioperative complications like damage to the cribriform plate, hemorrhage, or cerebrospinal fluid rhinorrhoea. Post-operatively, the patient recovered well. Histopathological examination revealed irregular trabeculae of woven bone with osteoblastic rimming. Pagetic cement lines and eosinophilic osteoid deposition were also visible in some areas. These findings were consistent with an osteoid osteoma. During a follow-up appointment, an endoscopic examination showed minimal scarring at the postoperative site. Her

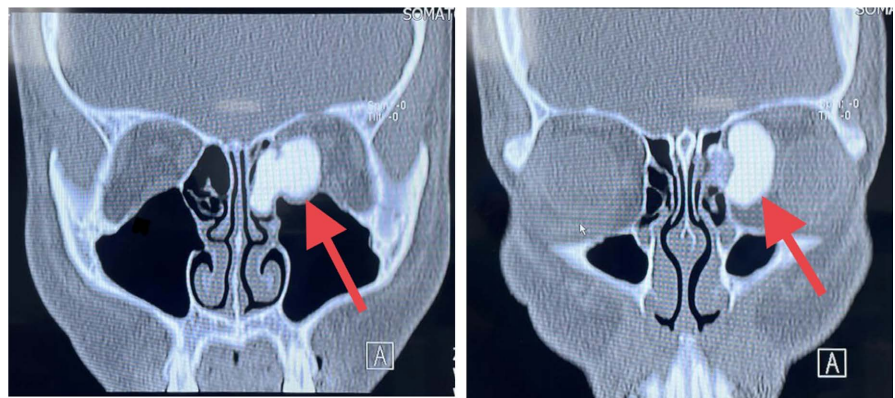


Figure 1. Coronal views of CT paranasal sinus showing a lesion extending from left ethmoid sinus into left orbital cavity (red arrow).

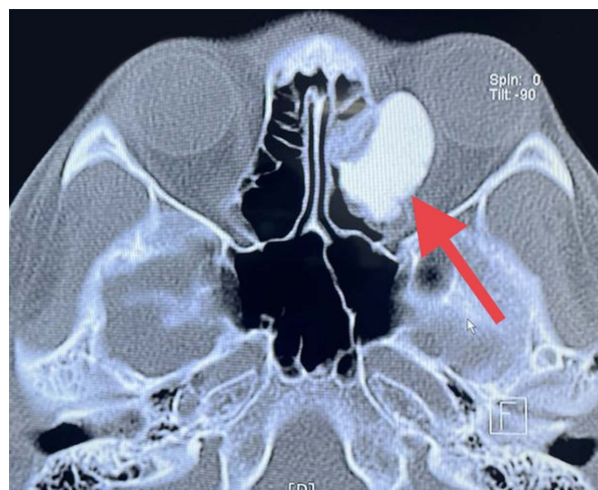


Figure 2. Axial view of CT paranasal sinus showing lesion extending from left ethmoid sinus into left orbital cavity (red arrow).

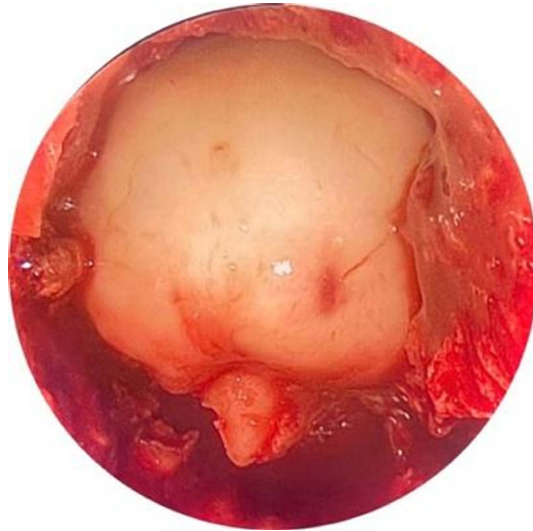


Figure 3. Endoscopic view of intra-operative finding showing a bony mass in the left ethmoid sinus.

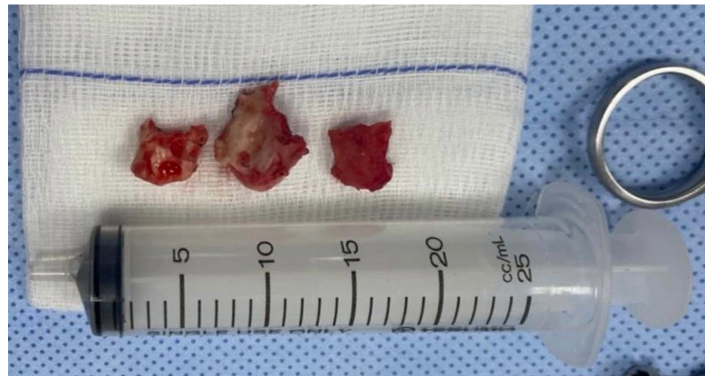


Figure 4. The osteoma was resected in piece-meal.

proptosis was resolved and the patient is asymptomatic and up-to-date.

3. Discussion

Osteoma is the most common benign tumor of the paranasal sinuses [5]. Despite being benign, they can also be locally invasive [12]. They are most commonly unilateral, although, in less than 10% of cases, they can also be bilateral [12]. Greater than 3 cm are considered “giant osteomas” [6].

Osteomas have a controversial etiology. The origins of traumatic, infectious, and developmental injuries have all been considered. Sinusitis or trauma can cause osteoblast growth in the mucoperiosteum of the sinuses, which can lead to tumor formation. Many osteomas appear to emerge near the confluence of the ethmoid and frontal sinus, a place where membranous and cartilaginous tissues converge during embryonic life, according to developmental theory [13].

Osteomas are round or oval in shape, hard, yellowish-white in color, and well-circumscribed, with a broad base or a small stalk attaching them to the adjacent bone [1]. Osteomas are characterized histologically into three types: ivory,

mature, and mixed. Ivory osteomas are made up of mature, thick lamellar bone with a little fibrous stroma. Mature osteomas are made up of enormous trabeculae of mature, lamellar bone with a more fibrous stroma, and they may or may not be surrounded by osteoblasts. The mixed kind includes tumors that have both ivory and mature characteristics [14]. Histopathological examination of our patient's lesion revealed irregular trabeculae of woven bone with osteoblastic rimming. Pagetic cement lines and eosinophilic osteoid deposition were also visible in some areas.

Depending on where the mass is located, osteomas can induce various signs and symptoms [6]. Common symptoms due to the pressure effect include headaches in the location of the osteoma, facial pain or deformity, and feeling of a mass. Symptoms also occur as a sequelae of sinusitis and includerhinorrhea, anosmia, and epistaxis [15]. Due to the limited space in the ethmoid region and, as a result, the rapid expansion, ethmoidosteomas can induce symptoms considerably sooner than those seen in the frontal sinuses [16]. For example, ocular symptoms like exophthalmos, diplopia, orbital cellulitis, epiphora, and proptosis may develop if the giant osteoma is located near the orbit or if there is an intraorbital extension [17] [18] [19] [20]. Intraorbital consequences from an osteoma are expected to occur in just 0.9% to 5.1% of all orbital tumors [2] [13]. Our patient presented with left orbital extension displacing the medial rectus muscle and optic nerve laterally, and mild proptosis of the left globe. Symptoms due to the pressure effect such as headache and diplopia were present however there were no symptoms due to sinusitis *i.e.* sinusitis was not present.

On imaging, the well-circumscribed, dense bone appearance is frequently diagnostic [21]. A CT scan of the paranasal sinuses is a crucial tool that not only allows for accurate diagnosis but also helps the surgeon determine the appropriate surgical intervention [6]. In our case a contrast-enhanced CT scan of paranasal sinuses was performed, which showed a bony growth in the left ethmoid sinus with the size of $2.7 \times 3.2 \times 2.5$ cm and helped us in diagnosing the condition. Because of the low morbidity rate, aesthetic advantages, and more experience of surgeons, endoscopic sinus surgery has been the procedure of choice for the excision of paranasal sinus osteomas in recent years [9]. For symptomatic giant ethmoidosteomas, surgical intervention is the preferred treatment [6]. The limited soft tissue dissection, avoidance of face bone disruption, and lack of a facial incision are the significant advantages of this approach. The use of endoscopes, which allow for magnification and multiple angled views, may facilitate the excision of osteoma with minimal morbidity [15]. The patient in our case successfully underwent endoscopic surgery for the removal of the osteoma. There are even fewer cases of giant ethmoidalosteomas being removed by endoscopic approach because of the larger size and therefore the tumor in our case could not be resected and removed as a whole; hence it was removed in piece-meal using Kerrison bone punch, burr drill bits, and bone curette via the piriform aperture. Humeniuk-Arasiewicz *et al.* showed that only 31.58% of the patients had

resection done by endoscopic approach. The results of open surgical versus endoscopic removal, however, did not differ in cases of giant ethmoidalosteomas [9].

Although osteomas are not known to transform into malignancy, the goal of surgical resection is to prevent further complications (particularly intraorbital and cerebral expansions) without aggravating the current condition [6]. Surgery for giant osteoma has a good prognosis, with very few recurrences, no malignant transformation, and minimal symptoms that persist [2]. To rule out recurrence post-operatively, patients must be constantly monitored and undergo periodic imaging examinations [19]. During a follow-up appointment of our patient an endoscopic examination showed minimal scarring at the postoperative site.

4. Conclusion

Ethmoid sinus osteoma is an asymptomatic benign neoplasm until it increases in size and displaces the surrounding structure and causes ocular complications. CT imaging is the gold standard diagnostic modality. Surgical excision is the treatment of choice for ethmoid sinus osteoma, despite the size of the lesion. Endoscopic surgical excision is a less commonly employed modality, but it has advantages like earlier recovery and cosmetic benefits due to which it was the treatment employed in this case.

Consent

The informed consent from the patient was obtained for this case report.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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