

# Osteoblastoma of the ethmoid

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## ABSTRACT

Osteoblastoma is a rare, benign bony tumor of skull base. We report one such case. Clinical record. We reviewed the record of a 21-year-old-male, who presented with nasal obstruction and proptosis. The tumor had formed a large mass that displaced the frontal lobes, the orbits and projected into the nasal cavity. Imaging revealed a calcified nodular expansile lesion. The tumor could be excised near-total by bifrontal craniotomy combined with endonasal approach. Histopathology was confirmatory of osteoblastoma. The patient was discharged on the seventh postoperative day and is now on follow-up. Osteoblastoma is a rare tumor of the frontoethmoid region. Surgery requires meticulous planning and team approach.

**Key words:** Bone tumor, craniofacial, ethmoids, osteoblastoma, skull base

## INTRODUCTION

Osteoblastoma is an uncommon tumor that was first described in 1956 in separate reports by Jaffe and Lichtenstein.<sup>[1,2]</sup> The neoplasm forms osteoid tissue, and is pathologically distinct from ossifying fibroma, classical osteoid osteoma and osteoma. It commonly involves the spine, long bones and small bones of hands and feet. Less often, it involves the calvaria, patella, ribs and scapula. Skull base involvement is rare, and has been reported with involvement of orbit,<sup>[2]</sup> ethmoids [Table 1], sphenoid sinus.<sup>[3]</sup> There are only eight well-documented cases reported that involved or arose from the ethmoid region and extended into the nasal cavity [Table 1].

## CASE REPORT

A 22-year-old male was admitted with six-month-history of dull frontal headache and facial pain, feeling of nasal obstruction and mild prominence of both eyeballs. There was no history of nasal discharge, epistaxis, visual dimness or any limb weakness. Clinical evaluation revealed mild proptosis on both sides and anosmia. Anterior rhinoscopy revealed a firm nasal mass visible from both sides. The mass did not bleed on touch. Plain radiograph of the skull

outlined a faintly calcified mass in the anterior cranial fossa [Figure 1a]. No bony erosion could be appreciated. CT and MRI of brain [Figures 1b-f] showed a large extra-axial, solid, calcified, subfrontal mass extending into the nose and displacing the eyeballs. There was no cerebral edema or hydrocephalus.

Bifrontal craniotomy was done which revealed solid, friable, vascular tumor in the ethmoidal region causing upward displacement of basal dura. Tumor could be excised by curetting and suction, and the intranasal portion was removed by endoscopy assistance. Anterior cranial fossa floor was covered with pericranial flap, bone flap was anchored back and scalp was closed with subgaleal drain. Postoperative period was uneventful and he was discharged on the seventh postoperative day. He remains asymptomatic (barring anosmia) over the past one year.

Histopathology sections (H and E) showed randomly interconnected and separate trabeculae of osteoid and woven bone prominently rimmed by osteoblasts. The stroma around the trabeculae was densely populated by fibroblasts and contained many dilated and congested vessels [Figure 2]. Occasional osteoclastic giant cells were seen. No granuloma or inflammatory infiltrate were noted. Based on the clinical profile, imaging appearances and histopathological picture, a diagnosis of osteoblastoma was made.

## DISCUSSION

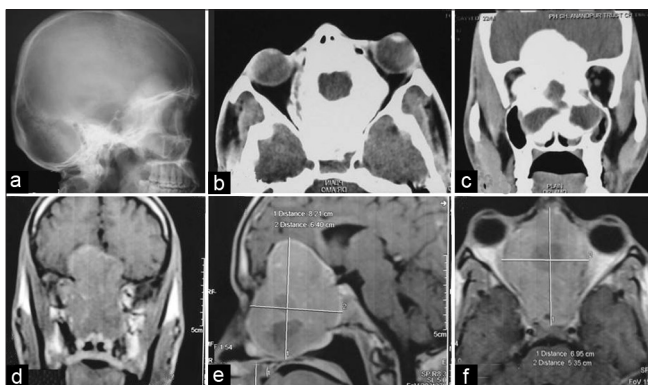
Osteoblastoma is an uncommon bone tumor, representing nearly 3% of all benign bone tumors.<sup>[4]</sup> Osteoblastoma is

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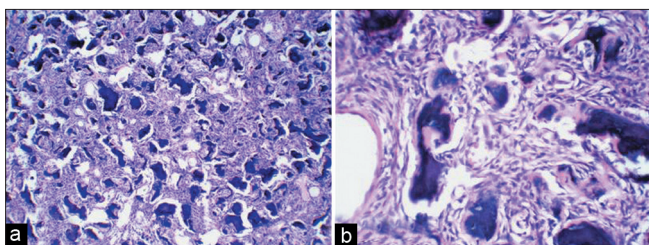
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**Table 1: Reported cases of osteoblastoma of the ethmoid**

Authors	Age/ gender	Presentation	Imaging	Findings	Treatment	Follow-up
Fu & Perzin <sup>[8]</sup>	12/F	Painless proptosis	X-ray	Nasoethmoidal mass	Local excision	Recurrence-free: 3yrs
Som <i>et al.</i> <sup>[17]</sup>	69/F	Proptosis, diplopia	X-ray	Calcified nasoethmoidal mass	Excision by lat rhinotomy	Operated for recurrence after 9 months
Freedman <sup>[7]</sup>	13/M	Epiphora	X-ray	Right nasoethmoidal mass	Excision by infraorbital approach	Not available
Ungkanont <i>et al.</i> <sup>[20]</sup>	9/M	Exophthalmos, nasal obstruction	CT, MRI	Ethmoids, sphenoid, orbit and nasal cavity	External ethmoidectomy	Not available
Imai <i>et al.</i> <sup>[10]</sup>	3/F	Bulging nose	CT, MRI, DSA	Calcified nasoethmoidal mass	Excision by bifrontal approach	Recurrence free after 8 months
Coscina <i>et al.</i> <sup>[5]</sup>	18/M	Proptosis	CT	Frontoethmoidal tumor	Excision by bifrontal approach	Not available
Velegarakis <i>et al.</i> <sup>[21]</sup>	22/F	Nasal obstruction, epistaxis	CT, MRI	Perpendicular plate of ethmoid	Excision by bifrontal approach	Recurrence free after 22 months
Park <i>et al.</i> <sup>[16]</sup>	13/M	Exophthalmos	CT, MRI	Ethmoid sinus	Transnasal endoscopy	Well after 12 months



**Figure 1:** Lateral skull radiograph showing opacified tumor (a) NCCT axial (b) and coronal (c) sections showing densely calcified ethmoid and subfrontal tumor. MRI coronal (d), midsagittal (e) and axial (f) showing the extent of tumor



**Figure 2:** Histopathology (H and E, x10) showing osteoid and loose fibrovascular stroma (a) and histopathology (H and E, x40) showing trabeculae lined by osteoblasts (b)

a benign tumor of bone that is histologically very similar to osteoid osteoma; in the past, it was sometimes known as giant osteoid osteoma. In fact, authors considered size to be an important differentiating feature, and called any tumor exceeding 1.5 cm as osteoblastoma.<sup>[5]</sup> In a series of 98 patients, only 3 were located in the skull.<sup>[5]</sup>

This tumor is generally seen in patients younger than 30 years of age.<sup>[6]</sup> Skull base involvement is extremely rare. Osteoblastoma of the anterior skull base can arise from the ethmoid region, although orbital roof

osteoblastoma<sup>[7]</sup> and a nasal osteoblastoma occurring from the nasal bone periosteum have also been reported.<sup>[8]</sup> Similar to the age pattern observed by Mirra *et al.* for osteoblastomas in the body,<sup>[6]</sup> six out of eight reported patients with nasoethmoid tumor have been in their first two decades [Table 1],<sup>[9-16]</sup> although there is one report of this tumor occurring in a 69-year-old female.<sup>[14]</sup> The patient is usually brought to otolaryngologist with nasal obstruction, visible prominence over midface or proptosis. There is significant facial pain, although pain was not a prominent feature in our case.

Radiological appearance can be in the form of non-specific, expansile, osteogenic tumor, often indistinguishable from ossifying fibroma or fibrous dysplasia. The mixed fibrous and osseous appearance tends to be more nodular in osteoblastoma.<sup>[17]</sup> Contrast MRI usually shows uniformly dense enhancement in the fibrous areas sparing the dense regions of mature bone. CT shows a densely calcified nodular lesion.<sup>[18]</sup>

Operative exposure of these tumors requires a bifrontal craniotomy for complete and radical excision. Imai *et al.* described a dismasking flap for removal of the tumor from the nasal cavity. However, we observed that the tumor can be removed with the help of endoscopy, with less morbidity. Histologically, the classical features are long interanastomosing trabeculae of osteoid or woven bone in a loose stroma of fibrovascular tissue. The trabeculae are lined by single row of osteoblasts and there is irregular deposition of osteoid. Highly vascularized tissue lies between the osseous trabecular network. The main differential diagnosis includes osteosarcoma, osteoid osteoma and aneurismal bone cyst. However, in osteoblastoma, the stromal cells are small and slender and do not resemble the sarcomatous spindle cells. Mitoses are rare, anaplastic cells are absent, there is

no cartilage formation, and the cells within the osteoid matrix are smaller and inconspicuous. Presence of sheets of osteoblasts with destructive permeation of surrounding tissue, however, confirms the diagnosis of osteosarcoma.<sup>[14]</sup> As compared to osteoid osteoma, osteoblastoma has more abundant fibrous stroma, many multinucleated giant cells, extravasated blood. Giant cell tumor can be excluded by the presence of calcification and osteoid, and formation of bone in osteoblastoma.

These findings can be identical to those of osteoid osteoma; what differentiates it from the latter is the size, which is more than 2 cm in case of osteoblastoma. Osteosarcoma is the other important tumor to be differentiated from osteoblastoma.

Osteoblastoma is a benign lesion, but local recurrence can occur following subtotal resection. Aggressive osteoblastomas have been reported, with multiple recurrences despite radiation.<sup>[19]</sup> Lesions of the calvarium can be excised *en bloc*, while skull base lesion can be curetted. Malignant transformation has been reported, both spontaneously and following radiotherapy.<sup>[20]</sup>

## CONCLUSION

Skull base osteoblastoma involving the ethmoid is a rare tumor that presents with nasal obstruction in young patients. Imaging reveals a densely calcified solid tumor with areas of vascularity. Large tumors need bifrontal craniotomy and endoscopy-assisted removal. Histopathology shows characteristic features in the form of interanastomosing trabeculae, lined by thin layer of osteoblasts and absence of mitoses or invasion. Long-term prognosis is good with excision.

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