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Osteoblastoma of the nasal cavity arising from the perpendicular plate of the ethmoid bone

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Abstract
The presence of a benign osteoblastoma in the ethmoid sinus is rare and only a few cases have been reported. This is a case of a benign osteoblastoma arising from the perpendicular plate of the ethmoid bone with extension to the nasal cavity. The diagnosis and management of this unusual lesion, as well as the histopathology and the imaging characteristics are reviewed. We also review the previously reported cases of benign osteoblastomas of different origin, with nasal cavity involvement.

Key words: Osteoma, osteoid; Ethmoid sinus.

Introduction

Osteoblastoma is an uncommon benign primary bone tumour accounting for less than one per cent of all bony tumours. The skull is rarely involved, with less than 15 per cent of all cases reported affecting the skull (Lucas *et al.*, 1994). Only a few cases with ethmoid sinus and nasal cavity involvement have been reported in the literature. We present a documented report of benign osteoblastoma arising from the perpendicular plate of the ethmoid bone with nasal cavity involvement.

Case report

A 22-year-old woman was referred to our service for evaluation with a six-month history of nasal obstruction, epistaxis, left orbital pain, and progressive swelling of the left upper external nose. She did not complain of diplopia or diminished vision, and both her personal and family history were not contributory. Anterior rhinoscopy revealed a hard, tender, erythematous mass, obstructing the left nostril, in which only the anterior one-third of the inferior turbinate was seen. The right nostril was partially obstructed by the same mass. There was no deviation of the anterior part of the nasal septum. The patient had no diplopia or proptosis of the left eye. There was no evidence of lymphadenopathy, and the remainder of the physical examination was negative. A biopsy was performed under endoscopic examination and the histopathological analysis was inconclusive.

Computed tomography (CT) revealed a well demarcated, expansile mottled soft tissue mass, which contained scattered areas of calcification. The mass was centred on the perpendicular plate of the ethmoid bone, occupying both the anterior ethmoid cells and the upper parts of the nasal cavities. The left middle meatus was obliterated and there was partial opacification of the ipsilateral maxillary sinus with haemorrhagic heterogenous material. The anterior cranial fossa was not involved (Figure 1a, b).

A left lateral rhinotomy approach was undertaken. A bluish well-vascularized mass was found, which appeared to originate from the perpendicular plate of the ethmoid. The mass extended to the cribriform plates and partially eroded the nasal bones. It completely filled the anterior ethmoid cells and the left nostril, extending inferiorly to the level of the anterior one-third of the inferior turbinate. There was also an extension of the mass towards the nasal septum occupying the upper part of the right nostril to the level of the middle turbinate As the mass was removed, marked bleeding ensued, which was controlled with Surgicel and electrocautery. There was no extension of the tumour towards the skull base. In such a case, a craniofacial resection would be performed with the help of neurosurgeons. Pre-operatively, the patient was warned of the possibility of CSF leak. In respect of the anatomical location, extension and vascularization, the tumour was removed en bloc. The procedure was carried out with an estimated blood loss of 500 ml. Both nostrils were splinted with Silastic sheeting and an anterior nasal pack was placed. The recovery was uneventful, and the patient is clinically and radiologically free of tumour, twenty-two months post-operatively (Figure 2a).

Histopathology

Macroscopy

The surgical specimen consisted of multiple brown-grey fragments measuring from 1 × 1 × 1 cm to 3.5 × 3 × 1.5 cm.

Microscopy

The tumour consists of a variable number of inter-anastomosing trabeculae of osteoid and primitive bone, within a highly vascular connective tissue stroma (Figure 3a). The osteoblasts are present in abundance. At the

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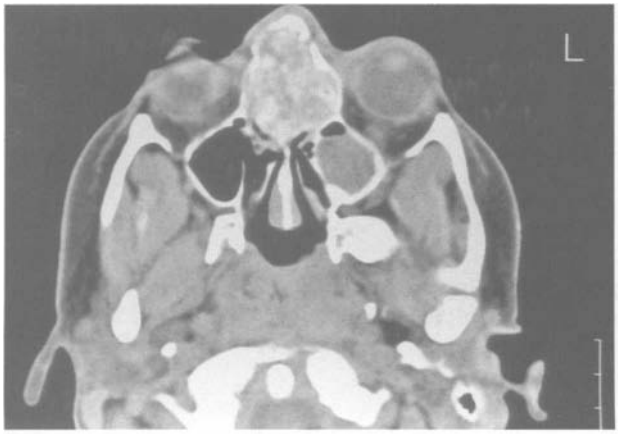

FIG. 1a

Axial CT scan shows a nasoethmoid expansile mass arising from the perpendicular plate of the ethmoid bone.

border of osteoid and primitive bone trabeculae they are larger and more active in appearance, but they do not show abnormal mitotic activity or nuclear atypia. Within the highly vascular connective tissue stroma, there are also a variable number of multinucleated giant cells of the osteoclastic type (Figure 3b).

Histological features indicative of the aggressiveness of the osteoblastoma (large epithelioid osteoblasts, bizarre cells, prominent mitotic activity, and a multifocal growth pattern), as described by Lucas *et al.*, 1994, were not identified in our specimen.

Discussion

Benign osteoblastoma was first described by Virchow in 1863 (Virchow, 1863). In the English literature, Jaffe and Mayer first described this entity in 1932 (Jaffe and Mayer, 1932). The long bones and vertebral column are the commonest sites. The ratio of male to females is 2 to 1, and most cases occur in the second decade (McLeod *et al.*, 1976). The differential diagnosis of benign osteoblastoma includes lesions such as osteoid osteoma, osteosarcoma, giant cell tumour, fibrous dysplasia, aneurysmal bone cyst and cartilaginous lesions. Regarding the benign lesions, osteoid osteoma is more related to osteoblastoma (Katsantonis *et al.*, 1981; Loizaga *et al.*, 1993), but its natural history is different (Greenspan, 1993). More often osteoblastoma can be mistaken for an osteosarcoma.

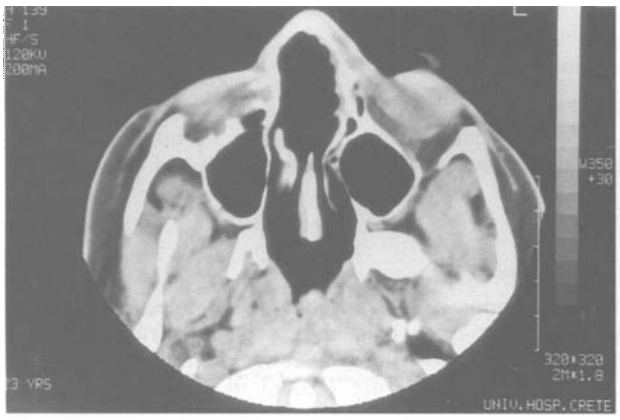

FIG. 2a

Post-operative axial CT scan demonstrating total removal of the tumour without any evidence of recurrence. The left maxillary sinus appears totally clear.

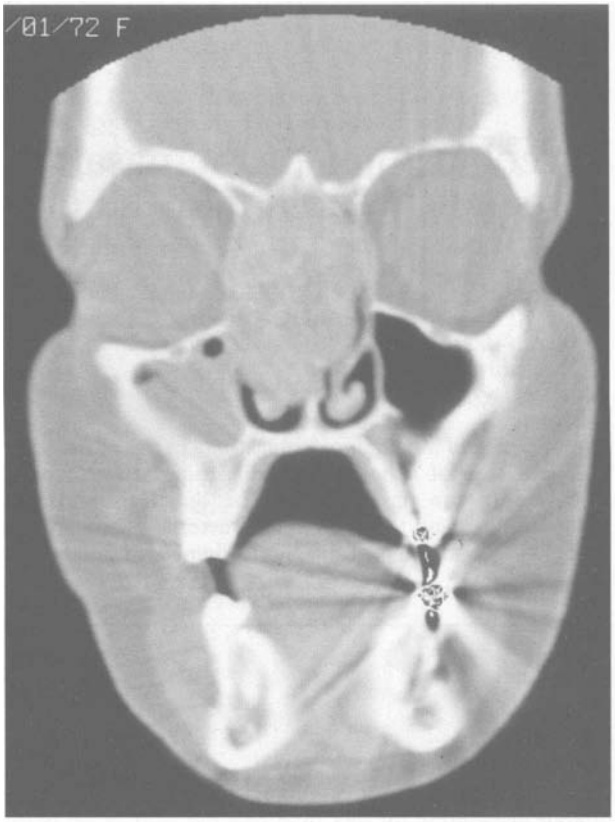

FIG. 1b

Pre-operative coronal CT scan reveals the superior extent of the tumour.

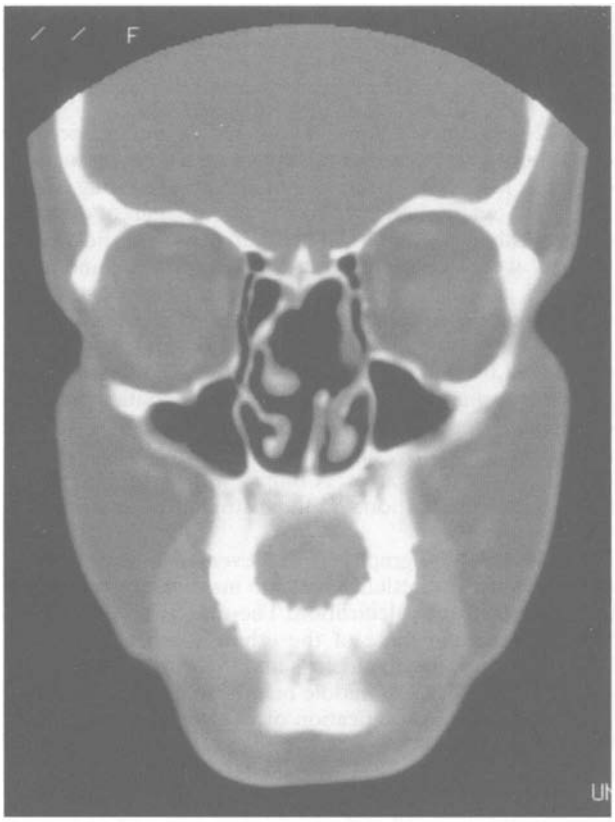

FIG. 2b

Coronal CT scan demonstrating the surgical result 22 months after the operation.

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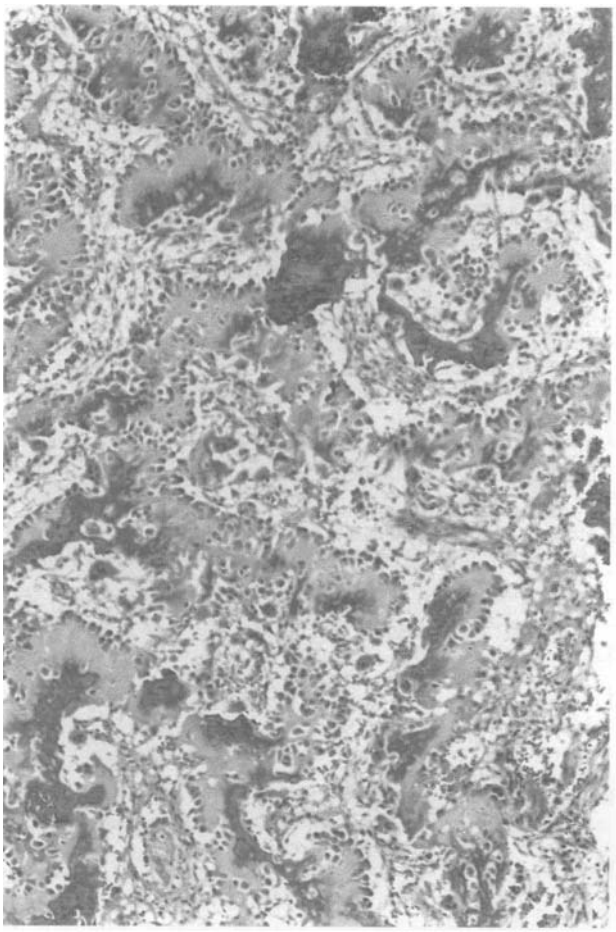

FIG. 3a

Interanastomosing trabeculae of osteoid and primitive bone, within a highly vascular connective tissue stroma (H & E; × 100).

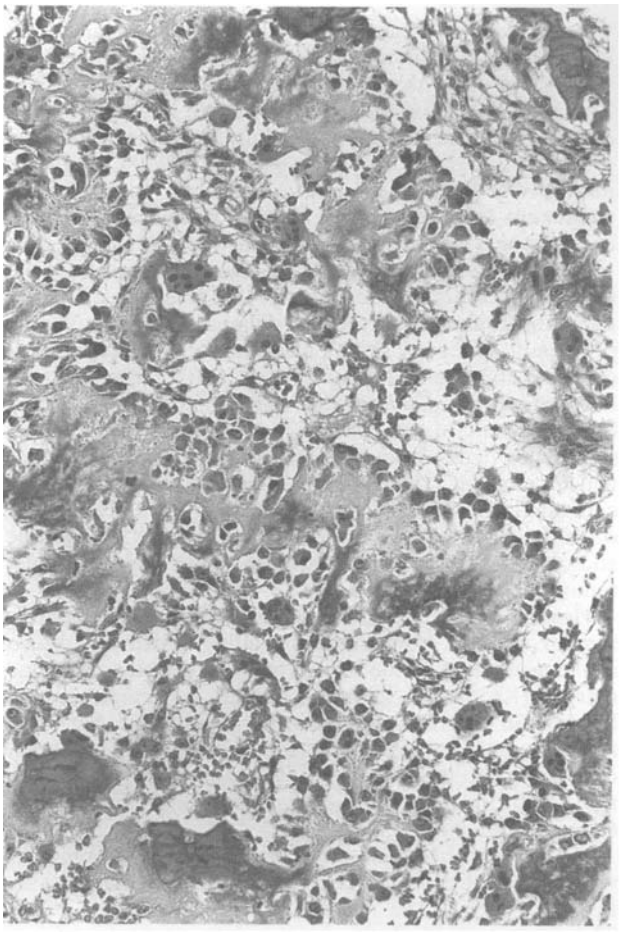

FIG. 3b

Trabeculae of osteoid and primitive bone with intense osteoblastic activity. Highly vascular connective tissue stroma with the presence of multinucleated giant cells of osteoclastic type (H & E; × 150).

There are only a few previously reported cases of benign osteoblastoma involving the ethmoid sinus and nasal cavity (Table I). The first reported case involved a 12-year-old girl, who presented with a painless displacement of an eye, and a mass that involved the right ethmoid cells (Fu and Perzin, 1974). The second case involved a 13-year-old boy who had a history of painless epiphora of the right eye and diplopia on left medial and upward gaze (Freedman, 1975). The lesion was found at the lamina papyracea of the ethmoid bone, without any extension to the nasal cavity. The third case involved a 69-year-old lady, presenting with proptosis of the right eye and anosmia for four months (Som *et al.*, 1979). In this case the tumour also involved the ethmoid cells. The fourth case involved an 18-year-old male with benign osteoblastoma centred on the left ethmoid sinus with nasal cavity involvement (Coscina and Lee, 1985). The fifth case involved a 12-year-old girl, who presented with epistaxis and a left nasal mass without bone destruction, which seemed to have arisen from the periosteum of the middle turbinate (Ducastelle *et al.*, 1985). The sixth case involved a 14-year-old boy, who had a two year history of right nasal swelling with a mass that arose from the nasal bones (Sooknundun *et al.*, 1986). The seventh case involved a 19-year-old man, who presented

TABLE I

LOCALIZATION OF BENIGN OSTEOBLASTOMA IN ETHMOID SINUS AND NASAL CAVITY

Author(s)	Year	Sex	Age	Presentation	Location
Fu and Perzin	1974	F	12	Displacement of the right eye	Ethmoid, orbit, nasal cavity
Freedman	1975	M	13	Epiphora of the right eye	Ethmoid, orbit, lacrimal duct
Som *et al.*	1979	F	69	Proptosis of the right eye, nasal obstruction, anosmia	Ethmoid, maxillary, sinus, nasal cavity
Coscina and Lee	1985	M	18	Proptosis of the left eye	Ethmoid, maxillary, frontal sinus, nasal cavity
Ducastelle *et al.*	1985	F	12	Epistaxis, left nasal obstruction	Nasal cavity
Sooknundun	1986	M	14	Right nasal obstruction	Nasal bone
Chen *et al.*	1993	M	19	Right nasal obstruction, epistaxis	Nasal cavity
Ungkanont *et al.*	1996	M	9	Right exophthalmos and nasal obstruction	Ethmoid, sphenoid sinus, orbit, nasal cavity
Present study		F	22	Nasal obstruction, epistaxis and left orbital pain	Ethmoid, nasal cavity

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with a history of bulging in the right nasal bone area, and epistaxis for a six-month period. The tumour arose from the periosteum of the middle turbinate (Chen *et al.*, 1993). The eighth case involved a nine-year-old boy with benign osteoblastoma of the ethmoid sinus suffering from slow progressive right exophthalmos, with extension to the nasal cavity (Ungkanont *et al.*, 1996). All these patients underwent local excision of the lesions except the sixth case in which an enucleation was carried out.

Computed tomography (CT) can easily demonstrate the origin and extent of the tumour, the presence of matrix mineralization and the tumour delineation, depicted as a peripheral, thin bony shell. Furthermore, CT is superior in demonstrating remodelling and erosion of the adjacent bony structures, a remarkable imaging in cases of benign bone tumours. In addition, CT can show the surrounding soft tissue oedema and muscle wasting. Magnetic resonance imaging (MRI) does not have any benefits compared to CT in the evaluation of osteoblastomas, since the signal intensities on MR images are not specific and do not differ from those in other primary bone tumours.

However, MRI is of great value in differentiating tumour margins from the surrounding oedema and also in the evaluation of the involvement of the adjacent bone marrow (Kroon and Schrumans, 1990). Arteriography can be helpful in the pre-operative assessment but only approximately half of the cases exhibit tumour vascularity or a dense capillary blush. However, angiography is often not helpful in determining the true nature of the lesion (Kroon and Schrumans, 1990).

Benign osteoblastoma is not usually life threatening. As well as the ethmoid, other paranasal sinuses may also be involved, such as the maxillary sinus (Tom *et al.*, 1980). The treatment of choice is complete surgical excision. However, sometimes it is difficult to judge whether or not to sacrifice important anatomical structures. There have been reported also some cases of malignant transformation (Dalinka and Chunn, 1972; Lucas *et al.*, 1994), as well as spontaneous regression following incomplete excision (Som *et al.*, 1979; Leone *et al.*, 1988). Post-operative radiotherapy may be a predisposing factor for transformation to osteosarcoma (Jackson, 1978) and it does not alter the clinical course of the disease. Lifelong follow-up is obligatory.

We believe that otolaryngologists must be aware in their practice of the entity of benign osteoblastoma with nasal cavity involvement, and consider this diagnosis based on the correlation of the clinical appearance, imaging, and histopathology data.

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