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CASE REPORT
Reconstructive

Giant Cell Tumor of Bone, Rare Inferior Orbit Location

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Summary: Giant cell tumor of bone (GCTB) typically presents in the epiphysis of long bones and occurs in the skull in less than 1% of cases. When GCTB is diagnosed in the skull, it is primarily seen in the temporal and sphenoid bones. GCTB is an osteoclastic stromal tumor that is locally aggressive and tends to recur. We present a case of a 43-year-old woman with slowly progressive, left-sided proptosis, eyelid swelling, photophobia, epiphora, and pressure sensation. Computed tomography and magnetic resonance imaging revealed an enhancing tumor in the left inferior orbit with infiltration into the maxillary sinus roof. Histological examination was conclusive for a giant cell tumor of the maxillary bone presenting as an inferior orbital mass. Gross total surgical resection was performed via an inferior anterior orbitotomy, and the patient had resolution of symptoms without recurrence in her postoperative course to date. This report contributes to the scarce literature available on this type of tumor of skull bones, specifically presenting the first case in the maxillary bone of the floor of the orbit. *(Plast Reconstr Surg Glob Open 2024; 12:e5569; doi: 10.1097/GOX.0000000000005569; Published online 1 February 2024.)*

We present a case of giant cell tumor of the bone arising from the maxillary bone of the floor of the orbit. Primary tumors of orbital bone make up less than 2% of all orbital tumors.[1] A subtype of primary tumors known as giant cell tumor of bone (GCTB) typically presents in the epiphysis of long bones and occurs in the skull in less than 1% of cases.[2] When it does occur in the skull, it is most commonly seen in the temporal and sphenoid bones. A review of the English literature suggests that the first known case of GCTB of the orbit was reported in 1993 arising from the orbital roof.[3] To our knowledge, this is the first reported case of GCTB arising from the maxillary bone of the orbit floor. This tumor type is classified as benign; however, it can be locally aggressive with a 1%–3% chance of malignant transformation into osteosarcoma.[4] Additionally, GCTB has a tendency to locally recur and is preferentially treated with surgical intervention.[1,5]

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CASE REPORT

A 43-year-old woman presented to our clinic complaining of a 1-year history of left periorbital swelling, eye pain, headache, epiphora, and photophobia. She noted a very slow change in the appearance of her left eye, becoming more proptotic for 10 years (Fig. 1). There was no history of systemic disease, sinus disease, or facial trauma. On examination, her visual acuity was 20/20 OD and 20/30 OS. Left extraocular motility was restricted in downgaze. Hertel exophthalmometry readings demonstrated 3 mm of axial proptosis of the left eye, causing an entropion of the left lower eyelid. The remainder of the examination was unremarkable.

Magnetic resonance imaging of the orbits with and without contrast was performed which showed an enhancing lesion within the left inferior extraconal fat, measuring 1.7 × 2.8 × 1.4 cm, superiorly displacing the left inferior rectus muscle. Computed tomography (CT) of the orbit without contrast was performed, demonstrating an expansile lesion with well-corticated margins of the left orbital floor, displacing the orbital contents (Figs. 2 and 3). The lesion was noted to extend into the left maxillary sinus roof, yet

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Fig. 1. Preoperative photograph.

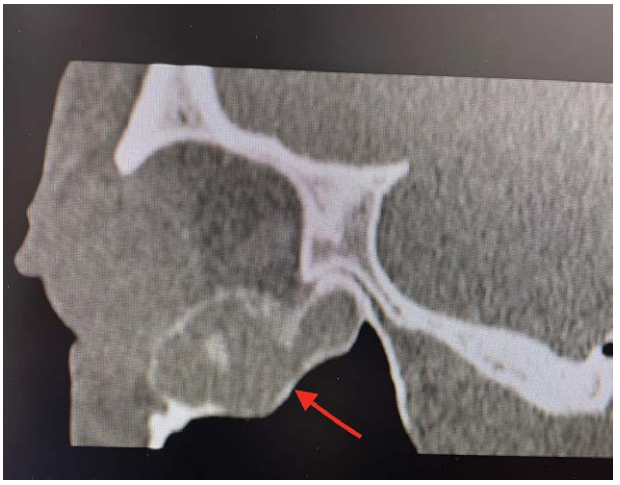

Fig. 2. CT sagittal.

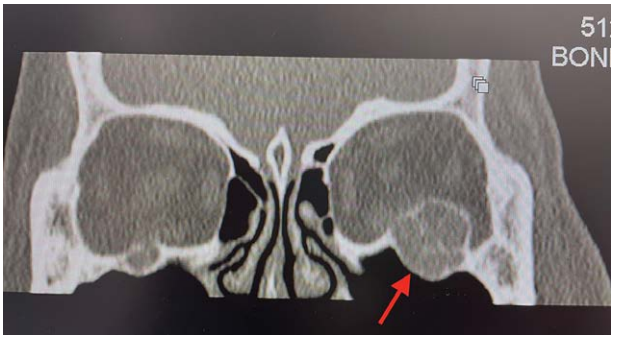

Fig. 3. CT coronal.

it was difficult to discern if the lesion was contained within the bone.

The patient underwent an uncomplicated two-hour anterior, inferior orbitotomy with bone removal and tumor resection with 25 mL of blood loss. Intraoperatively, there was a very thin layer of bone completely surrounding the soft-tissue mass in the floor of the orbit. The bone

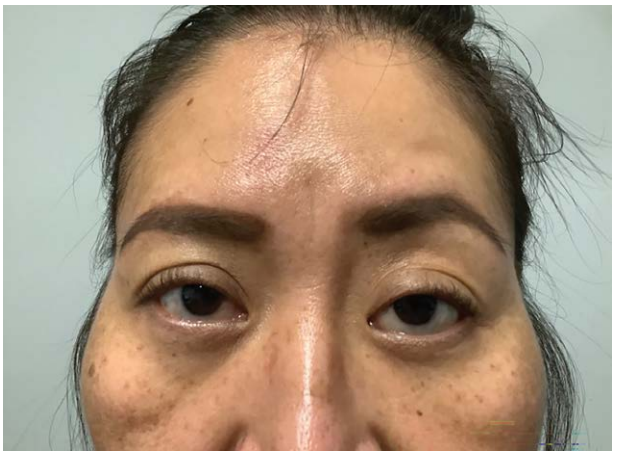

Fig. 4. Postoperative photograph.

over the superior portion of the tumor was removed from the intraorbital approach to access the soft-tissue mass within the bone cortex. Gross total tumor resection was accomplished. The inferior orbit wall had expanded into the maxillary sinus, likely from years of soft-tissue tumor growth. The floor of the orbit was repaired with a KLS Martin Resorb X orbital floor implant. Pathology showed osteoclastic giant cell tumors scattered among mononuclear cells with ovoid to spindled morphology. (**See figure, Supplemental Digital Content 1**, which shows a pathology slide, **http://links.lww.com/PRSGO/D46**.) No cytological atypia or mitoses were seen to suggest malignancy. A diagnosis of GCTB was made.

Postoperatively, the patient experienced initially worse diplopia on downgaze due to inferior recuts muscle paresis. At her 9-month postoperative visit, her vision was 20/25 OD and 20/25 OS. Extraocular motility was full in all directions with complete resolution of her diplopia. The entropion was resolved and her exophthalmos had resolved (Fig. 4). No additional postoperative medications or adjuvant therapy was required. The CT scan done 14 months after surgery showed no evidence of recurrence. We plan to follow up with the patient in 1 year with orbital CT imaging to monitor for recurrence.

DISCUSSION

GCTB is a neoplasia of mesenchymal tumor cells that aids in recruitment and formation of osteoclasts. Research has found that there are three common cell types of GCTB: multinucleated giant cells; morphologically round cells resembling monocytes; and spindle-shaped, fibroblast-like stromal cells. This research suggests that the neoplastic component of GCTB is the stromal cell, which secretes monocyte chemoattractants, stimulating monocyte fusion into multinucleated, osteoclastic-like, giant cells.

GCTB classically presents in the epiphysis of long bones such as the ulna, radius, distal femur, proximal tibia, and fibula. The rare skull bone cases of GCTB in the literature reveal that the sphenoid, ethmoid, frontal,

and temporal bones are most involved. In the orbit, there are a few reported cases of GCTB mainly in the roof and lateral wall, making our case unique in the maxillary bone of the floor of the orbit. There is a mild predisposition in females over males.[6,7] GCTB has a peak incidence in the second to fourth decades of life.[6] GCTBs of the orbit are typically associated with findings of visual field deficits, proptosis, diplopia, decreased visual acuity, ophthalmoplegia, and headache.[8]

Imaging findings for GCTB are nonspecific. Currently, there are no magnetic resonance imaging features sufficiently characteristic to allow for a preoperative diagnosis. Nonspecific findings of GCTB may include an enhancing, well-circumscribed lesion with low-to-intermediate signal intensity on T1-weighted images and intermediate-to-high intensity on T2-weighted images.[9] On CT imaging, GCTB tends to demonstrate a radiolucent, osteolytic lesion with well-demarcated margins. Differential diagnosis of this orbit tumor included: osteosarcoma, intraosseous hemangioma, osteoma, Langerhans cell histiocytosis, and metastatic disease.

Current treatment of choice for GCTB is gross total surgical resection.[3,10] Radiotherapy and subcutaneous injections of denosumab can be used for inoperable and/or recurrent cases.[2] Long-term follow-up is of maximal importance due to the risk of recurrence.

CONCLUSIONS

GCTB involving the orbital bones is extremely rare. To our knowledge, there have been no reported cases of GCTB primarily arising from the maxillary bone of the inferior orbital floor. Although extraordinarily rare, GCTB should be considered in the differential diagnosis of orbit tumors involving the bone. Gross total resection is the treatment of choice, paired with long-term follow-up to monitor for recurrence.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

PATIENT CONSENT

The patient provided written consent for the use of her image.

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