Case Report: Giant Cell Reparative Granuloma of Ethmoid Sinus

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Giant Cell Reparative Granuloma of Ethmoid Sinus
Mazyad Marji, Jihan Al Maddah

Aims: Giant cell reparative granuloma is a non-neoplastic lesion rarely occurs and histologically is difficult to differentiate from giant tumor. We add our case to the few reported cases in the literature.

Methods: To report a case of a giant cell reparative granuloma of the ethmoidal region and to review the literature of all reported cases.

Results: Our case presented with only proptosis and lacrimation with recurrent infection but no history of trauma. Imaging study narrowed the differential diagnosis. Biopsy was diagnostic. Endoscopic complete resection of the Mass was performed with final histology of Giant cell reparative granuloma.

Histologically, giant cell reparative granuloma can be difficult to differentiate from giant cell tumor. Prior to Jaffe report on 1953 any bone lesion containing giant cells was thought to be a giant cell tumor or variant thereof. It represents a local hyperplastic reparative process after injury or secondary granulomatous lesion developing in the setting of infection or inflammation complicated by hemorrhage.

Conclusions: Giant cell reparative granuloma (GCRG) is a rare condition affecting the mandible and maxilla. There are few of documented GCRG involving the facial bones or paranasal sinuses. Giant cell reparative granuloma should be considered when an expansile lesion is seen within the paranasal sinuses, bony orbit, or calvaria and MR shows isointensity with gray matter typical of a fibrous lesion, especially in a patient in the first two decades of life. Histopathological diagnosis is mandatory, surgical excision is the treatment of choice while radiotherapy is used in inoperable and recurrent cases.

Keyword: Giant Cell Reparative Granuloma, Ethmoid Sinus, Granuloma, Fibroosseous lesion

CASE REPORT
A 35-year-old woman presented to ophthalmology clinic at Prince Sultan Medical Military City with left-sided proptosis and increased lacrimation for 8 weeks. Contrast CT of the orbit showed heterogeneous soft tissue density enhancing mass seen involving the left ethmoid air cells measuring about 2.3 x 1.9 cm with erosions and remodeling of ipsilateral cribiform plate and lamina papyracea which is protruded laterally into the extraconal space of left orbit with lateral displacement of medial rectus muscle and subsequent mild left eye proptosis (Fig. 1a). Patient then was referred to our otolaryngology department. On further questioning patient had no history of trauma or significant nasal or sinus infection, family and past history were unremarkable. Proptosis and lateral displacement of the eye were evident by physical examination, scopic examination of the nose showed lateral nasal mass. MRI outside our institution and thin cuts CT scan done (Fig. 1b).
MRI showed a well-defined mass in the left ethmoid air cells with mixed solid and cystic component, low to intermediate intensity on T1 (Fig. 2a), intermediate to high intensity on T2 (Fig. 2b) and has heterogeneous enhancement in postgadolinium images (Fig. 2c).

Measuring 2.4 x 3.3 x 2.9 cm in its transverse, craniocaudal and anteroposterior dimensions respectively, it has a mass effect along the adjacent structures which bulged into the extraconal space of left orbit with lateral displacement of medial rectus muscle and mild proptosis of left eye, focal inflammation seen in the anterior inferior aspect of left frontal sinus. Imaging studies were inconclusive regarding nature of disease cystic versus soft tissues lesion.

The main differential diagnosis was ethmoidal neoplastic lesion and fibro-osseous lesion. Patient underwent two endoscopic surgeries. Biopsy taken from lesion and sent to histopathology and result showed Giant cell reparative granuloma (central giant cell granuloma). Endoscopic surgery done a week later, the mass was excised completely and sent for histopathology with final pathological diagnosis of Giant Cell Reparative Granuloma of Ethmoidal Sinus. Patient symptoms resolved postoperatively with clear nasal cavity on telescopic examination.

Patient did not keep her follow-up appointments. Six months later was referred from the antenatal clinic with recurrence of symptoms MRI imaging showing recurrence of lesion. Revision endoscopic sinus surgery with navigation done for patient with improvement of symptoms. Patient followed for six months clinically and radiologically (Fig. 3a,b) with no recurrence.
DISCUSSION

Giant cell reparative granuloma (GCRG) was first reported on 1953 by Jaffe as a reaction to intra-osseous hemorrhage found exclusively in the jawbones. [1]

Hirschl and Kratz first described GCRG of cranial bone in 1974. [2,3] In the head and neck region its incidence is reported to be 0.00011%. [2] It is not a true neoplasm but rather a reactive process; its origin could be triggered by trauma or inflammation. [1,2] In our case, the cause of the lesion was not discernible from the patient history. GCRG affects children and young adults, predominantly females, in the 2nd and 3rd decades of life. [3,4] In our case patient was female and in 3rd decade. GCRGs are classified, according to location, as central or peripheral, occurring, respectively, in bone or gingival soft tissues. [5] GCRG occurs in many bones with the mandible, maxilla, and the small bones of the hand and feet being the most common sites. [6,7] Could be presented rarely as a mass lesion in the orbit, paranasal sinuses (most commonly the sphenoidal) temporal bone. [8-10] In our case lesion was in paranasal sinus (ethmoidal region) which is rarely reported. Reported symptoms are relatively nonspecific and depend on the location of the lesion. Local pain, periorbital swelling, and diplopia may be identified when involvement of the orbital structure is present. [10] In our case, symptoms resulted from impression of the mass on the orbital soft tissues causing proptosis. The radiologic manifestations of GCRG are nonspecific with CT scan show lytic expansile multi-loculated lesion which is due to slow growth that cause thinning of cortex but usually intact. [3] There is usually opacification of the affected sinus due to the soft tissue component of the mass. Also, they may appear as heterogeneous soft tissue masses with occasional hemorrhagic or cystic foci. In this sinus, GCRGs can also appear quite aggressive with bony destruction through either the ethmoid or sphenoid bone, involving the clivus or cribriform plate, and extending intracranially. [11] Osteoid is occasionally identified within the lesion on CT. [10] In our case CT showed same finding of heterogeneous soft tissue density enhancing mass involving the left ethmoid air cells with erosions and remodeling of ipsilateral al cribiform plate. MR findings of GCRG include low to intermediate T1 signal, high T2 signal (depending on the amount of hemosiderin deposition in the lesion) with hypointense septations and variable contrast enhancement. [11] In our case same finding were seen. Differential diagnoses of our lesion included giant cell tumors, brown tumor of hyperparathyroidism (osteitis fibrosa cystic), aneurysmal bone cysts and fibrous dysplasia.

Radiologically, GCT and GCRG are indistinguishable. On CT, both usually appear as nonspecific lytic lesions. On MR imaging, most lesions show areas of low signal intensity T1 and T2-weighted imaging, corresponding to the areas of fibrous and/or hemosiderin. Both tumors enhance, with the degree of enhancement ranging from slight to strong. [12,13] Histologically, giant cell reparative granuloma can be difficult to differentiate from giant cell tumor. Both show multinucleated giant cells in a connective tissue stroma; in the reparative granuloma, however, the giant cells are scattered, mitotic figures are rare, and the stromal cells show a cytoplasmic predominance, as opposed to those seen in giant cell tumors, which show a nuclear predominance. [8,14,15] GCT can be distinguished based on following: it mostly observed between the ages of 20 and 40 years, most commonly involves the epiphyses of major long bones and within the skull, the sphenoid and temporal bones are the most commonly affected whereas GCRG commonly involve mandible and maxilla. [5,13] GCT has a higher incidence of recurrence 45%-60% than GCRG 10%-20% and may undergo malignant transformation and metastasize whereas GCRG does not undergo malignant transformation and metastasis has not been reported. [12,13] A brown tumor can also be difficult to differentiate from a giant cell reparative granuloma.
Laboratory values are extremely important in the setting of brown tumors in showing elevated serum calcium, alkaline phosphatase, and parathyroid hormone levels and depressed serum phosphate. [8,10,15] Aneurysmal bone cysts histologically reveal histologically thin-walled blood-filled sinuses separated by a thin, fibrous septa and have characteristic radiologic features demonstrating a multilocular "soap bubble" or honeycomb radiolucency. On MRI images the classic findings of aneurysmal bone cyst are those of multiple cysts with fluid-fluid levels. [16,17] Fibrous dysplasia can be excluded because of the radiologic features demonstrating a ground-glass appearance on CT scans and low signal intensity on all MR imaging sequences with intense enhancement. [16,17]

Treatment modalities for GCRG usually consist of curettage, surgical excision, or radiation therapy. [17] Complete surgical excision has been advocated over simple curettage, because of the high incidence of recurrence with incomplete removal. [17] When these tumors are not amenable to surgical resection or recurrence, radiation therapy has been advocated. 17 Sarcomatous degeneration after treatment of this lesion with radiation therapy has been described, but de novo malignant transformation is not seen. [11]

CONCLUSION

GCRG is a rare benign condition that may involve the paranasal sinus. It should be considered when an expansile lesion is seen within the paranasal sinuses. Imaging are non-specific, MR shows include low to intermediate T1 signal, high T2 signal with hypointense septations and variable contrast enhancement especially in a patient in the 2nd and 3rd decades of life. Histologic analysis is required for diagnosis. Optimal management of GCRG is complete surgical excision.

REFERENCES