Multifocal giant cell reparative granuloma involving maxilla and mandible: a rare entity

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Giant cell reparative granuloma (GCRG) is a rare lesion that is a reactive process, not a true neoplasm. It was originally coined by Jaffe to describe lesions, which he believed were a response to intraosseous hemorrhage from jaw trauma. Regardless, GCRG is much more distinct from giant cell tumor (GCT) of bone, both histologically and clinically. We report a patient who presented with multiple facial swelling involving the facial skeleton that showed a multiloculated cystic appearance on CT involving the maxilla and mandible. The patient refused surgery, but after 6 months of follow up there was no progression.

Giant cell reparative granuloma term was first coined by Jaffe in when he described the lesion as a local reparative reaction to intraosseous hemorrhage induced by trauma. This lesion was thought to be a giant cell tumor (GCT) or a GCT variant before the time of Jaffe. There has been much doubt regarding the pathogenesis of this lesion as it has been reported without having a definite history of antecedent trauma and a lack of significant elements of reparative tissue. It is mostly seen in women of younger age (F: M 2:1) and usually presents in the 2nd and 3rd decades of life in the anterior part of the jaw. The diagnosis is suggested by clinical history and examination, laboratory findings and radiological investigations.

CASE

A 30- year-old female presented with painless swelling predominantly affecting the left side of the jaw and growing insidiously over a period of 6-7 months. There was no history of trauma or tooth extraction. On examination, the lesion was firm and non-tender. There was no facial par aesthesia or nasal problem, no history of difficulty in opening the mouth. She had no systemic complaints and routine laboratory investigations were normal. The overlying skin was intact. There was no associated regional lymphadenopathy.

On examination the swelling of the lesion was bony hard, nontender in left lower jaw with an extension of swelling to the central part of jaw, with a normal appearing buccal mucosa. An orthopantomogram revealed a well-defined, multiloculated expansile and lytic lesion in the left paramedian aspect of the body of the mandible with displacement of roots of adjacent teeth. There was no evidence of sclerosis, internal calcification or resorption of teeth, and the matrix appeared clear (Figure 1). A contrast-enhanced CT scan showed a multiloculated expansile lesion with heterogeneous enhancement involving the body and adjacent part of the ramus of the mandible on the left side (Figures 2 and 3). There was evidence of bony remodeling in the form of cortical thinning with a breech at anterolateral aspect lesion with displacement of the roots of adjacent teeth (Figure 4). Also there was evidence of a similar type of lesion involving the superior alveolar arch of the maxilla on either side of the midline (Figures 5 and 6) and bony remodeling in the form of cortical thinning (Figure 7).

The patient underwent incisional biopsy to ascertain the nature of the lesion and to rule out a malignant etiology. Histopathological examination of the resected specimen revealed solid, cellular proliferation of oval to spindle fibroblasts with no cellular pleomorphism and multinucleated giant cells scattered throughout these stromal cells along with spindle-shaped fibroblasts without any atypia and scattered multinucleated giant cells consistent with the diagnosis of giant cell reparative granuloma (Figures 8 and 9). The patient was reassured and was given the option of surgical resection, but
she refused. She was in regular follow-up and 6 months after surgery there was no clinical evidence of progression of the lesion.

DISCUSSION

GCG are particularly less common lesions that are neoplastic and histologically distinct from GCTs of a bone and constitute a small proportion of masses in the region of head and neck. The incidence is highest in the second and third decade of life with a female preponderance. The reported patient ages are variable, but up to 74% of patients are under 30 years of age at presentation. Some studies show accelerated growth and recurrence of the lesion during pregnancy and in the postpartum period, which suggests that giant cell reparative granuloma may be hormone-dependent. The lesions most commonly involve the mandibular or maxillary regions and the second most common location for GCRG is in the small bones of the hands and feet, which are unusual sites for GCT, like the face. GCRG most commonly affects the phalanges of the hand, followed by the metacarpal, metatarsal, carpal, and tarsal bones and the phalanges of the foot. Multiple lesions are rare but have been reported. There is equivocal distribution in male and female for GCRG involving the hand and foot. GCG usually is a slow growing mass and therefore there is a delay in diagnosis. The patient presents with symptoms, which are largely due to a mass effect, and therefore depend on the site of involvement. Soft-tissue swelling and pain are the most common clinical findings encountered in these patients. Other clinical symptoms, including ptosis, nasal obstruction, and cranial nerve palsy, are nonspecific findings and occur in a few cases.

Gorlin-Goltz syndrome, enchondromatosis, Paget disease, and fibrous dysplasia are some of the lesions that show an association with GCRG. The radiographic findings are usually non-specific and demonstrate expansile remodeling of bone with a multiloculated appearance and thinning of the cortex; however there is usually no breech in the cortex. The radiographic appearance is similar to that of ameloblastoma, ABC, odontogenic cyst, odontogenic fibroma, and odontogenic myxoma.

GCRG affecting the hands and feet shows a bony remodeling in the form of a lytic expansile and multiloculated appearance in the metaphyseal region, which may show extension into the diaphysis. But the extension into the epiphysis is unusual and extension across an unfused epiphysis is rare. The lesions are typically a few centimeters in size approximately 3-2.5 cm in diameter, having internal trabeculation on plain radiographs. Mineralization may be seen in GCRG lesions, but is rare and typically limited in extent. The cortex is usually thin, but intact, although
more aggressive growth with soft-tissue extension has been described. The lesion may show evidence of periosteal reaction, but it is rare and a pathologic fracture may be seen. The lesion usually treated by curettage. Recurrence is seen in 25%-60% of cases, although lesion eradication typically does not require more than two excisions. There is less evidence of sarcomatous transformation or aggressive spread of the lesion.

**Conflict of interest**
The authors report no conflict of interest and no funding was received for this purpose.

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