Central Asian Journal of Medical and Pharmaceutical Sciences Innovation **1**(5): 236-241 (2021)

CASE REPORT

Central giant cell granuloma of maxillary bone in pediatric patient: A case report

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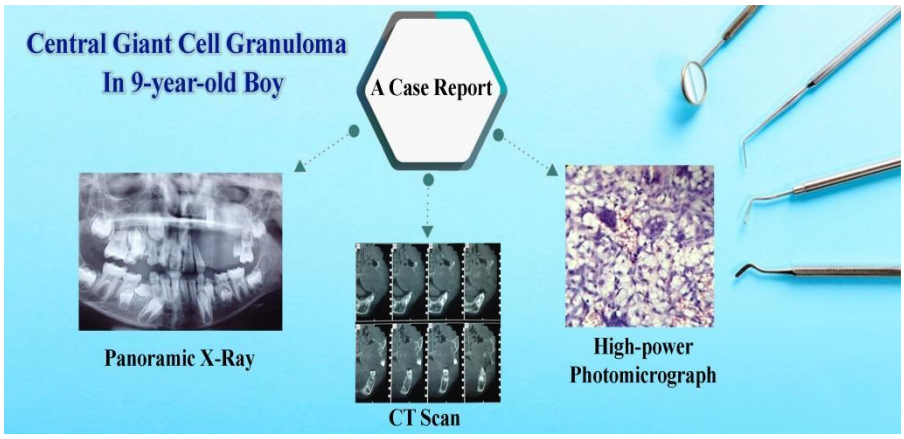
Highlights

- Central giant cell granuloma of maxilla in male pediatric patient is rare.
- To reduce the size of the lesion in the maxilla for the surgery sclerotic agent had been used.
- Giant cell lesion is in the differential diagnosis of central giant cell granuloma.

Graphical Abstract



Article Info

Receive Date: 30 August 2021
Revise Date: 07 October 2021
Accept Date: 19 October 2021
Available online: 23 October 2021

Keywords:
CGCG
Osteolytic lesions
Maxillry bone
Pediatric patient

Abstract

Central Giant Cell Granuloma (CGCG) is a benign, intraosseous osteolytic lesion which often affects the mandible and its' pathophysiology is not fully understood. Histopathology is still the dominant diagnostic method for this disorder. Clinically invasive lesions can lead to premature injury and may require invasive treatment to prevent recurrence .more than 60% of all cases occur before the age of 30. In spite of the fact that the sex ratio varies in different studies, giant cell granuloma is most commonly reported in women; and approximately 70% occur in the mandible. Radiographically, giant cell granuloma is defined as radiolucent defects that may be uni-locular or multi-locular, which have an indefinite margin Most lesions are found in the anterior part of the jaw, and mandibular lesions often cross the midline In this study, a case of invasive CGCG of maxillary bone in a 9-year-old boy with diagnostic and therapeutic challenges was presented.

10.22034/CAJMPSI.2021.05.04 **E-ISSN: 2783-0993**

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Introduction

CGCG is a rare, benign, intraosseous lesion that affects the head and neck area. Although it is a benign lesion, but can be aggressive and locally destructive and when located in the maxilla, can spread to surrounding tissues (1). More than 60% of all cases occur before the age of 30. In spite of the fact that the sex ratio varies in different studies, giant cell granuloma is most commonly reported in women; and approximately 70% occur in the mandible. Most lesions are found in the anterior part of the jaw, and mandibular lesions often cross the midline (2). Radiographically, giant cell granuloma is defined as radiolucent defects that may be uni-locular or multi-locular, which have an indefinite margin (3). Histologically, the lesions are non-encapsulated proliferations of oval and spindle-shaped mononuclear cells (MCs) and multiple multinucleated giant cells (MGCs) in a fibro vascular stroma, associated with foci of hemorrhage (4). Usual treatment for CGCG is surgery which varies from a simple curettage to an invasive jaw resection. For the management of CGCG, injection of corticosteroids into lesions had been used as an alternative non-surgical method, which was successful in the management of CGCG, providing a better prognosis and preventing surgery (1, 5).

Case Presentation

A nine-year-old boy, without any previous history of trauma who suffered left swelling in the zygomatic area, had referred to the Mashhad Dentistry School. His second primary molar tooth had been extracted without radiography, which lead to more swelling in the area. Panoramic radiographs on the left side of the maxilla showed an expansive lesion with distinct cortical boundaries, which had extended from the permanent central incisor tooth to the mesial of tooth #26, and caused the canine tooth bud to dislocate superiorly and the lateral tooth to move downward, and also it had made the follicle of tooth #25 to dislocate toward mesial of permanent first molar (Figure 1).

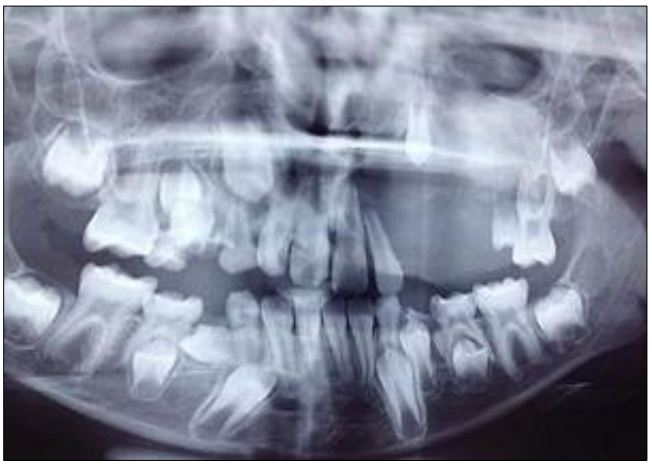

Figure 1. Panoramic X-Ray showing an expansive radiolucency.

Calcifying Odontogenic Cyst (COC) considered as differential diagnosis for this case, and also suggests an invasive CGCG or a COF, given that the lesion is mixed. In CT sections, displacement of the sinus and nasal floor is seen (Figure 2). An incisal biopsy sent for histopathology examination for pathology features and diagnosis of other lesions. Histopathologic examination showed intensive proliferation of multinucleated giant cell some of which having 20 to 30 nucleus in a fibro vascular background having some degree of chronic inflammation and trabeculae of lamellar bone, the diagnosis of CGCG was considered and The treatment intended for this patient was sclero-therapy for the lesion to make it smaller before surgery; and then, the lesion was surgically removed (Figures 3 and 4).

This procedure probably requires the removal of all involved teeth. As another option, intralesional corticosteroid injections were selected for the patient. And excised tissue sent for further histopathologic confirmation of CGCG diagnosis.

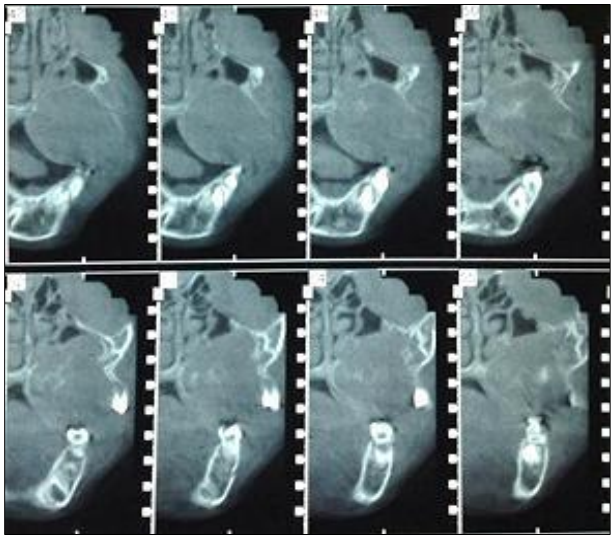

Figure 2. CT scans showing expansive lesion causing displacement of the sinus and nasal floor.

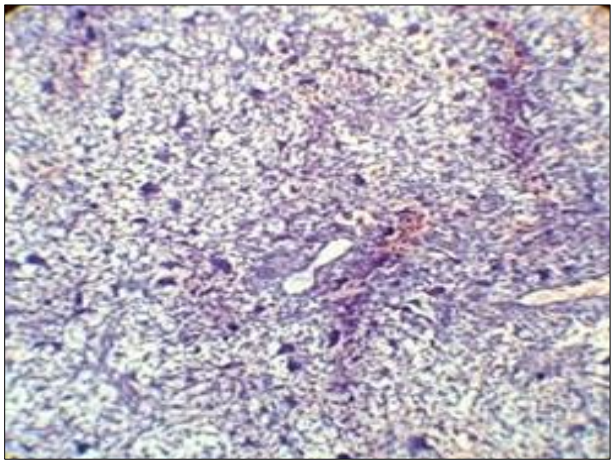

Figure 3. low-power photomicrograph shows proliferation of multinucleated giant cells in a fibrovascular background.

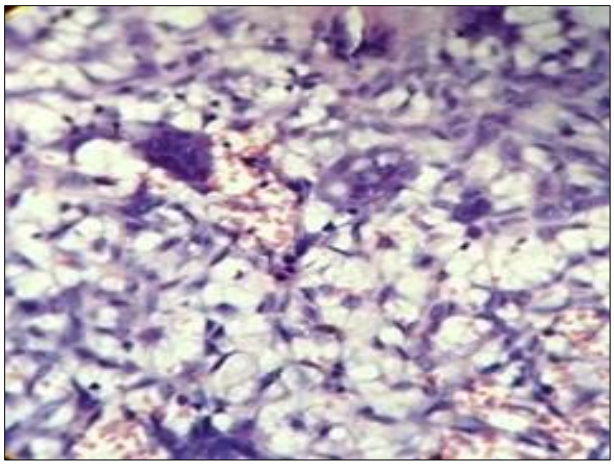

Figure 4. This high-power photomicrograph shows multinucleated giant cells having numerous nuclei.

Discussion

Giant Cell Granuloma is considered a non-neoplastic intraosseous lesion. Some lesions show aggressive behavior similar to that of neoplasms. The lesion was first reported by Jaffe in 1953 as a "Giant Cell Repair Granuloma". To date, the cause of giant cell granuloma has not been determined (6).

This lesion may be seen in patients aged 2 to 80 years, although more than 60% of all cases occur before the age of 30 (7). In spite of the fact that the sex ratio varies in different studies, giant cell granuloma is most commonly reported in women; and approximately 70% occur in the mandible (7, 8). Most lesions are found in the anterior part of the jaw, and mandibular lesions often cross the midline (9, 10). Most of the time they are asymptomatic and are first seen during a routine radiographic examination or as a result of painless expansion of the damaged bone. However, a few cases may be associated with pain, paresthesia, or perforation of the cortical plate. Based on clinical and radiographic features, these jaw lesions may be of two types: invasive (characterized by pain, rapid growth, cortical bone perforation, root resorption, and show a significant tendency for recurrence after treatment); non-invasive (most cases, have few or no symptoms, and show slow growth).

Radiographically, giant cell granuloma is defined as radiolucent defects that may be uni-locular or multi-locular, which have an indefinite margin. Radiographic findings are not definitely diagnostic; small uni-locular lesions may be confused with periapical granulomas or cysts. Multi-locular Giant cell granuloma lesions cannot be radiographically distinguishable from ameloblastoma or other multi-locular lesions (1, 2).

CGCG can easily be confused with giant cell tumor (GCT) of bone before surgery, because the histological differences between CGCG and GCT may be negligible. Pathological features of CGCG include multinucleated giant cells clustered around hemorrhagic foci, while multinucleated giant cells of GCT tend to be uniformly distributed. In GCT multinucleated giant cells are larger and may have up to 50 nuclei, compared to the 10 to 15 nuclei typically seen in CGCG. New bone formation and collagen deposition are characteristic of CGCG but not present in GCT (6).

The disease is likely to be clinically misdiagnosed due to its low prevalence, lack of specific features in terms of clinical manifestations, and nonspecific radiological features. Patient age at the onset of the disease, clinical manifestations, radiological findings, pathology and response to treatment should be considered in the clinical diagnosis. The role of hormonal changes in the pathogenesis of these lesions should not be underestimated, as it may be a possible cause of more occurrences in female patients.

Some other osseous lesions such as brown tumor of hyperparathyroidism which are due to persistent disease radiographically appear uni-locular or multi-locular radiolucencies. Although in brown tumors, other osseous changes such as osteitis fibrosa cystica can be seen. Hyperparathyroidism is histopathologically identical to the central giant cell granuloma of the jaws. Both lesions are characterized by a proliferation of multi- nucleated giant cells in a background of vascular granulation tissue (2). Researches show that CGCG causes more tooth displacement than root resorption, because more time is required for root resorption and young patients are diagnosed with the lesion before tooth resorption occurs (11).

Surgery is the most common treatment and can range from simple curettage to block removal. In invasive cases, surgical treatment may cause deformity, especially for lesions located in the maxilla. Alternative treatment with corticosteroid infusion has been performed with satisfactory results. These are used as definitive or preoperative treatments to reduce complications (1). Radiographic and histopathologic examinations of the lesion are not sufficient to diagnose. Laboratory tests, including serum calcium, phosphate alkaline phosphatase, parathyroid hormone (PTH) and, radiographic examination of other bones such as the wrist, pelvis, and femur are needed to make a more definitive diagnosis (12).

According to the recent studies pharmacological agents in clinical cases of invasive CGCG that affect the maxilla in pediatric patients are clinically promising (1, 13-15). The study of the expression of calcitonin and glucocorticoid receptors for treatment planning is very useful in making decisions for the management of invasive CGCG. In other studies, denosumab (16, 17) treatment for CGCG has been considered successful, especially in invasive lesions. The distinction between invasive and non-invasive CGCGs is very important because it leads to a change in a person's treatment protocol. Early diagnosis such as perforation or thinning of the cortical bone, cortical bone expansion and the presence of root resorption helps in lower recurrences after treatment (18, 19).

Conclusion

Our case demonstrated that by using sclerotic agent the size of the lesion can be decreased before surgical removal. And since the aggressive and expansive lesion was in the maxillary of a pediatric patient this method has to be used to minimize the size of surgery as much as possible.

Acknowledgement

The authors thank Dr. Samare Mortazavi (Assistant Professor of Oral & Maxillofacial Radiology, Department of Oral and Maxillofacial Radiology, School of Dentistry, Mashhad University of Medical Sciences) for differential radiographic diagnosis.

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How to cite this paper:
Shahabinejad M, Mohtasham N, Mohajertehran F, Mohammadi M. Central giant cell granuloma of maxillary bone in pediatric patient: A case report. Cent Asian J Med Pharm Sci Innov 2021; 1(5): 236-241.