Case Report

Infantile anterior maxillary swelling: A diagnostician's dilemma

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Abstract

Anterior maxillary swellings are commonly encountered in the adolescents and adults and they represent lesions ranging from cysts to tumors which can be both benign as well as malignant. However the anterior maxillary swellings are a rare phenomenon in the infants and toddlers and they generally are indicative of an aggressive lesion. We hereby present a case of a rapidly growing infantile swelling which was histopathologically diagnosed as Peripheral Giant Cell Granuloma.

Keywords: Giant cells, maxillary anterior, swelling

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INTRODUCTION

The diagnosis of anterior maxillary swellings is a challenging task for a clinician just based on the clinical and radiological appearance of the lesion. The anterior maxillary swellings can be a result of some developmental disturbance like inclusion cysts, inflammatory in origin or can be a reactive or a neoplastic process. Although the swellings are seen throughout life, the anterior maxillary swellings in infants are usually fatal. The benign lesions do occur in infants, but the number of malignant cases exceeds than the benign ones.

These swellings can be best examined by inspection and palpation clinically, and radiographs may be an adjunct in ruling out pathologies such as abscesses and periapical inflammatory conditions. Routine panoramic radiography can help to discover bony masses arising from the maxilla. However, the final diagnosis is achieved after the histological examination.[1]

This case report is an attempt to describe an unusual presentation of peripheral giant cell granuloma (PGCG) in a 1½-year-old infant.

CASE REPORT

Parents of a 1½-year-old infant reported to the outpatient department with a chief complaint of swelling in the anterior maxillary region which was rapidly growing for 2 months. The patient was a known case of congenital acyanotic heart disease, and he was on medication for 2½ months. On examination of lesion, proper obvious facial asymmetry was noticed due to eversion of lip. Mouth opening was inadequate. Regional lymph nodes were palpable, tender and mobile. The swelling was roughly

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oval in shape, and measured about 1.5 cm × 2.5 cm approximately and had erythematous surface [Figure 1]. On palpation, it was firm, mobile, had smooth surface and was bleeding on provocation. The radiological changes were not marked and only showed superficial erosion of the underlying alveolus. A clinical diagnosis of malignant mesenchymal tumor was made. Incisional biopsy was carried out and sent for histopathological examination. The H and E stained section showed parakeratinized stratified squamous epithelium with underlying extremely cellular connective tissue stroma with numerous blood vessels and proliferation of multinucleated giant cells [Figure 2]. The giant cells were distributed throughout the stroma and had nuclei ranging from 8 to 10 in number [Figure 3]. Few areas showed the presence of hemosiderin pigment and also inflammatory cells such as lymphocytes and plasma cells [Figure 4]. Plump-, ovoid- and spindle-shaped mesenchymal cells were also seen. Based on these findings, a histopathological diagnosis of peripheral giant cell granuloma was made. The lesion was surgically excised, and the healing was uneventful.

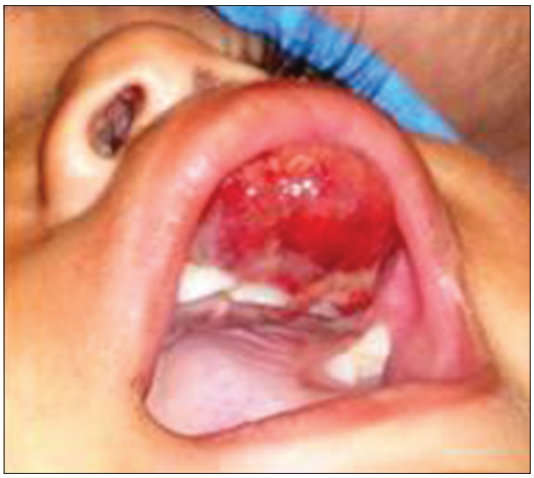

Figure 1: Clinical photograph showing a reddish lesion on the labial aspect of incisors extending onto the palate

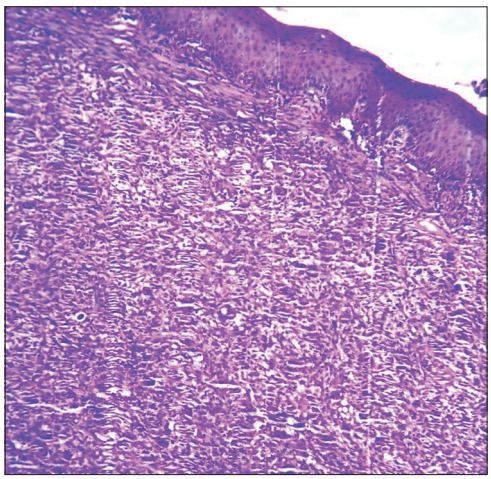

Figure 2: H&E stained section showing parakeratinized stratified squamous epithelium overlying an extremely cellular stroma (×4)

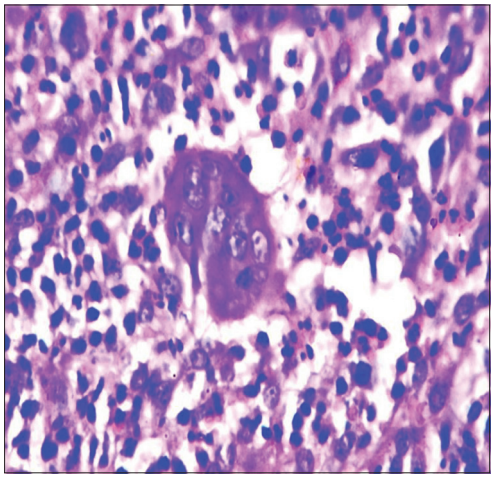

Figure 3: H&E stained section showing multinucleated giant cells in background cellular stroma (×40)

DISCUSSION

PGCG or so-called "Giant cell epulis" is a common giant cell lesion. It normally presents as a soft-tissue purplish-red nodule consisting of multinucleated giant cells in a background of mononuclear stromal cells and extravasated red blood cells. This lesion probably does not represent a true neoplasm but rather may be reactive in nature, believed to be stimulated by local irritation or trauma, but the exact etiology is not known. Although the PGCG occurs throughout life, the peak incidence is seen during the mixed dentition period and in adults during third to fourth decade of life. It is more common among females (60%). The mandible and maxilla is affected with equal frequency.

Several hypotheses had been proposed to explain the nature of multinucleated giant cells including the explanation that they were osteoclasts left from physiological resorption of teeth. Lim and Gibbins in 1995 stated that the multinucleated giant cells reacted strongly for a monoclonal antibody MB1 which reacts with lymphocytes and a proportion of T-cells and monocytes, thus laying stress on the lymphocytic–monocytic origin of these giant cells.[2]

Wülling *et al*. in 2001 revealed that the stromal cells secrete a variety of cytokines and differentiation factors, including monocyte chemoattractant protein-1, osteoclast differentiation factor and macrophage colony-stimulating factor. These molecules are monocyte chemoattractants and are essential for osteoclast differentiation, suggesting that

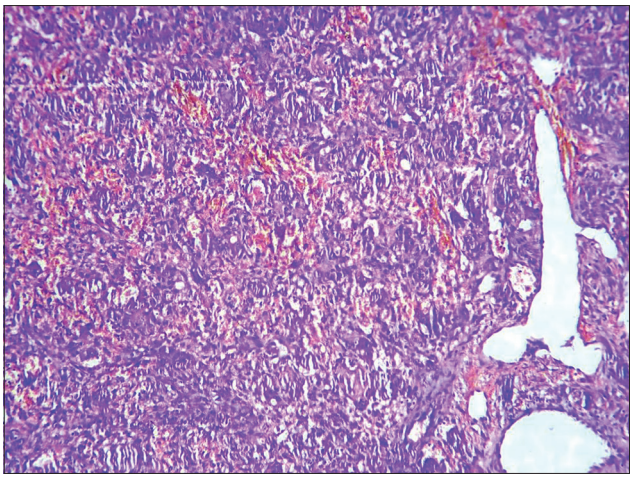

Figure 4: H&E stained section showing lots of vascular spaces and vascular channels (×10)

the stromal cell stimulates blood monocyte immigration into tumor tissue and enhances their fusion into osteoclast-like, multinucleated giant cells.[3]

Another hypothesis suggests that giant cells may arise from the fusion of endothelial cells of proliferating capillaries.[4]

The differential diagnosis for the most common lesions of the maxillary anterior region includes reactive hyperplasias, related to a number of chronic irritant stimuli. However, there are a number of entities that have a predilection for the gingivae, which are much less common in other parts of the oral cavity.

1. Pyogenic granuloma constitutes 85% of all reactive gingival swellings, representing a profuse mass of vascular granulation tissue. In oral cavity, it occurs most commonly in the maxillary buccal and interproximal gingiva. It is usually highly vascular, fast-growing, elevated, lobular, sessile or pedunculated and commonly ulcerated or covered by pseudomembrane. The color varies from red to pink with tendency to bleed on slight irritation. It occurs at any age and sex with a slight predilection for young females.[5] The size ranges from few millimeters to centimeters but neither cause alveolar bone loss nor do they disturb tooth positioning[4]
2. Peripheral ossifying fibroma - It originates from the periodontal ligament or the periosteum. This lesion has a predilection for females and is most common in young patients between 1 and 19 years of age.[6] It occurs exclusively on the gingiva, especially in the anterior gingiva, with slight predilection to the maxilla and rare presentation in primary teeth.[7] It usually presents as a well-demarcated sessile nodules, often ulcerated. It is of the same color as normal mucosa or red if ulcerated, with firm to hard consistency, depending on the amount of ossification and calcifications[4]
3. Sarcomas – rhabdomyosarcoma and fibrosarcoma
   - Rhabdomyosarcoma is a malignant neoplasm of skeletal muscle origin accounting for 4%–8% of all malignant diseases in children <15 years of age. It primarily occurs in the first decade of life with a peak incidence between 2 and 6 years with slightly more predilection in males. The tongue, palate and cheek are the most common locations in the mouth.[8] Gingiva is the additional site of growth.[4] The clinical appearance ranges from a small nodule to an extensive mucosal outgrowth. It may present as a painless, yet occasionally painful, facial swelling[8]
   - Infantile fibrosarcoma is a rare malignant neoplasm of fibroblast origin. They usually develop within the first 2 years of life with slightly more male predilection. Only 10% occur in the head and neck area, including the oral cavity,[9] with common site of occurrence in the buccal mucosa, palate, lips and periosteum of the mandible and maxilla. Fibrosarcomas present as fast-growing, fleshly, exophytic ulcerated lesions with tooth displacement and bone and tooth resorption[9]
4. Other less aggressive mesenchymal (infantile myofibromatosis and aggressive fibromatosis) lesions can also be considered when a lesion recurs in such a short period of time and especially in a child. They present as locally aggressive, firm, painless and poorly demarcated masses, that may be either rapidly growing or slowly growing[4]
5. Lymphoma and leukemia have a nonspecific clinical presentation but often present with swelling and reddening of the gingival tissues. Advanced cases are likely to be accompanied by bone loss and tooth mobility. More localized swelling may be mistaken for pyogenic granuloma or giant cell epulis.[10]

CONCLUSION

Although the present case in the anterior maxillary region was growing at an alarming pace and was causing discomfort to the patient, the diagnosis for the same turned out to be a reactive lesion. Wide local excision was carried out, and the healing was uneventful. Hence, a careful histopathological examination is mandatory for all oral lesions for proper treatment planning.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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