Case Report

Inflammatory myofibroblastic tumor in the retromolar region of an 8-year-old child: A rare finding

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Abstract

Inflammatory myofibroblastic tumor (IMT) of the oral cavity is an extremely rare finding. The etiology and pathogenesis of IMT is controversial and unclear. The tumor requires complete surgical excision and continuous monitoring of clinical consequences. The present article describes the clinical, histological, operative and immunohistochemical features of a case of IMT in the mandibular retromolar region of an 8-year-old male. Histologically, the lesion shows myofibroblastic spindle cell proliferations with infiltrative margins in an inflammatory background. Immunohistochemically, the myofibroblastic spindle cells in the present case were positive for α-smooth muscle actin and CD68 due to which the diagnosis of IMT was confirmed.

Keywords: Immunohistochemistry, inflammatory myofibroblastic tumor, retromolar

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INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is an uncommon finding in the oral cavity.[1] It has variable numbers of inflammatory cells and myofibroblastic spindle cells. It was originally described in the lung by Brunn in 1939 and was named by Umiker and Iverson in 1954 as an inflammatory pseudotumor (IPT).[2] Agrons *et al.*[3] based on the immunohistochemical analysis stated that inflammatory cells in IPT are predominantly myofibroblast, hence termed IPT as IMT, which is considered a more descriptive name. Herein, we describe the clinicopathologic features of IMT arising in the retromolar area.

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CASE REPORT

An 8-year-old male presented to the Department of Pedodontics and Preventive Dentistry with the chief complaint of a slow-growing, painless mass in the left posterior mandibular region. The patient gave a history of minor trauma 3 months back in that region, i.e. small swelling after biting hard food which increased gradually without pain or discomfort. The patient's medical history was noncontributory. The child was afebrile. Extraoral examination revealed no facial asymmetry. Intraoral examination revealed an irregular, firm, raised and sessile mass measuring approximately 1.5 cm × 1 cm in the retromolar region and tooth 36 was drifted mesially. The color was same as the surrounding mucosa with no

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bleeding or exudate. On the occlusal aspect, the presence of indentation of the upper tooth on the mass was noted [Figure 1]. The lesion was not tender on palpation.

Orthopantomograph revealed mesially drifted 36 with bone loss around the distal root [Figure 2]. A differential diagnosis of pyogenic granuloma (PG), fibroma, peripheral giant cell granuloma (PGCG) and peripheral ossifying fibroma (POF) was considered. The patient was prescribed syrup augmentin (228.5 mg/5 ml) twice daily for 7 days. On the 8th day, the patient reported pain, discomfort and increase in the size of the mass. An incisional biopsy was performed and the tissue was sent to the Department of Oral Pathology and Microbiology for further examination [Figure 3]. The test reports were suggestive of a benign soft-tissue lesion.

Complete surgical excision of the lesion was performed with the extraction of 36 under general anesthesia [Figures 4 and 5]. The excised lesion was submitted for histopathological examination.

The histological results were inconclusive due to which immunohistochemistry (IHC) was performed. The IHC report was positive for alpha-smooth muscle actin (SMA) and CD68 markers and negative for ALK-1 and S100. It also showed the presence of tumor cells arranged in interlacing fascicles with the presence of mild nuclear atypia, increased mitotic figure and no tumor necrosis suggestive of low-grade spindle cell neoplasm in subepithelial location, with ulcerated overlying squamous epithelium in certain places [Figure 6]. Considering all the above clinical, histopathological and IHC features, a final diagnosis of IMT was made.

DISCUSSION

IMT has an estimated prevalence of 0.04%–0.7% irrespective of the gender and race of the world population.[1] The most commonly documented site for IMT is the lungs. The other extrapulmonary sites reported in the literature are liver orbit, central nervous system, gastrointestinal tract, salivary glands and the oral

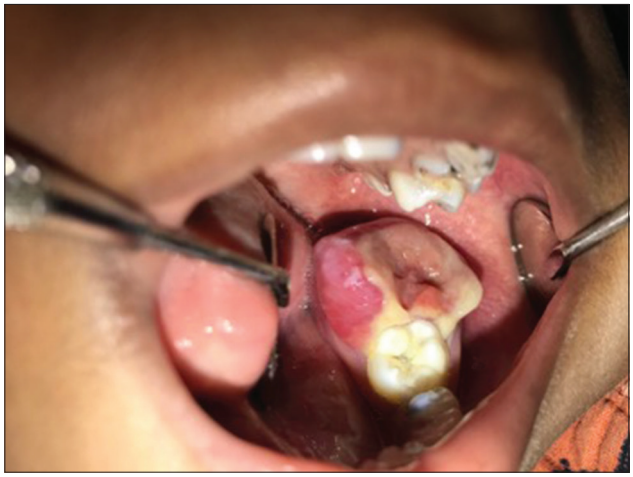

Figure 1: Preoperative view

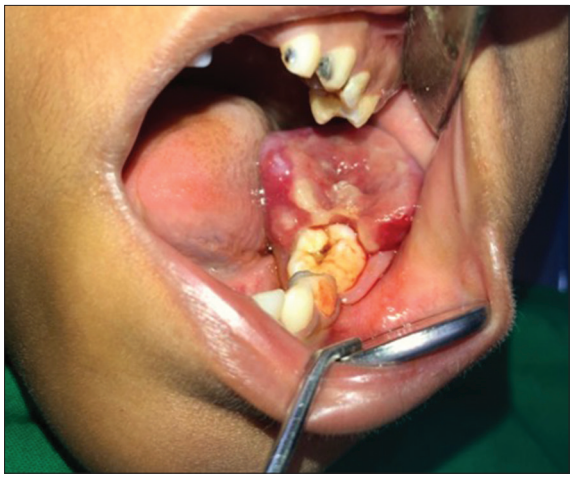

Figure 3: Lesion showing increase in the size/incisional biopsy performed

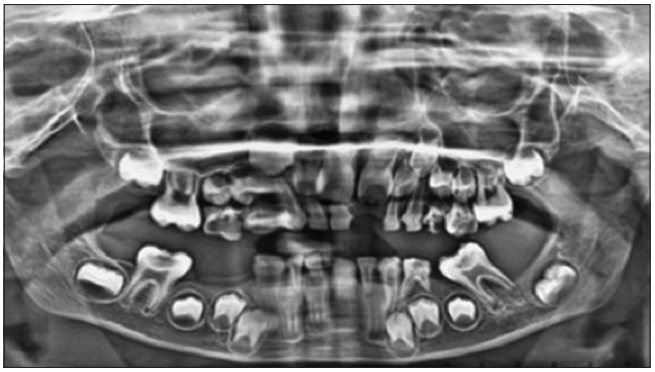

Figure 2: OPG-Orthopantomogram

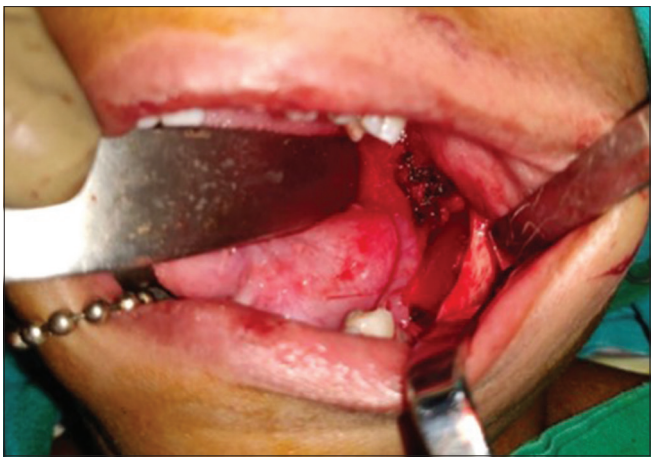

Figure 4: Postoperative view

cavity.[2] IMT in the oral cavity is a relatively infrequent finding and has been most commonly reported in the buccal mucosa, tongue, floor of the mouth and retromolar area in adults.[4]

Our literature search has revealed that in children <14 years of age, the presence of IMT is very rare. The evidence of oral IMT in children was reported by Liston *et al*.[5] where they described three cases 4 years, 2 years and 6 years, involving the buccal mucosa. Rautava *et al*.[6] reported the case of an 11-year-old child with IMT involving the gingiva (maxilla buccal mucosa of 23 and 24) and Satomi *et al*.[7] reported the case of a 14-year-old child with IMT involving the gingiva. To our knowledge, this is the first case report in the child where IMT was observed on the retromolar area.

The etiology of IMT remains unknown and controversial. Binmadi *et al*.[4] have suggested that the lesion maybe infectious, autoimmune, syndromic or traumatic in origin. In the present case, the patient had reported to us with a history of biting on hard food 3 months back after which the swelling continued to increase in size.

The clinical manifestation of IMT shows varying features. Liston *et al*.[5] in their case report stated that the lesion had increased in size in spite of the medications prescribed (oral penicillin). Similarly, we also noticed a rapid increase in size from 1.5 cm × 2 cm to 4 cm × 4 cm in the 1st week of medication. They also reported that the lesion is hard, ulcerated and firmly adhered to the mandible which was also observed in the present case. Histopathological results revealed the presence of ulceration overlying the squamous epithelium at certain places. The lesion in the present case was painless despite the size of the lesion which was in accordance with all three cases reported by Liston *et al*. (1981). Satomi *et al*.[7] and Rautava *et al*.[6] in their case reported painful lesion. In addition, Liston *et al*. (1981) reported that the child's temperature and white cell count were either normal or slightly elevated. In the present case, none of the findings with the elevated level was observed. Satomi *et al*. (2010) in their case reported that the patient was having low but constant fever.

Radiographically, there was extensive bone loss present in the distal aspect of 36 and the tooth was mesially drifted. In case of children, similar bone loss was observed by Rautava *et al*. (2013) and Satomi *et al*. (2010).

Considering the above factors, the clinical differential diagnosis of PG, PGCG, fibroma and POF was given. Clinically, all these lesions are proliferative gingival lesions that can show very similar characteristics.[8] PG was ruled out because it is a highly vascular lesion and exhibits a tendency to bleed, there was no bleeding seen in the present case in spite of its size and presence of indentation of the maxillary molars. PGCG was ruled out because it appears as a bluish-purple nodular mass which is not more than 2 cm. POF was ruled out because it usually emanates as an ulcerated lesion from the interdental papilla most commonly affecting the maxillary anterior region (incisor-cuspid) of not more than 2 cm. Fibroma was ruled out because it is most commonly seen along the bite line and the size ranges from few millimeters to not as large as 1.5 cm in diameter.[9,10]

Histopathology report

IMT has varying clinical features and poor specificity of radiographic images; therefore, the diagnosis of IMT is based on the histopathological immunohistochemical findings.[11]

In the present case, a low grade of spindle cells was seen in subepithelial location, with ulcerated overlying squamous epithelium in certain places. IMT of the oral cavity is known to exhibit a high degree of spindle cells with the presence of inflammatory background as a

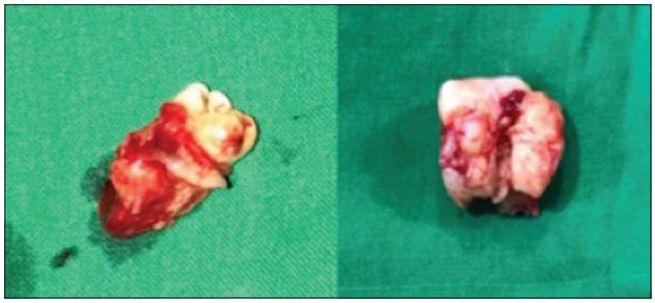

Figure 5: Extracted tooth and excised mass

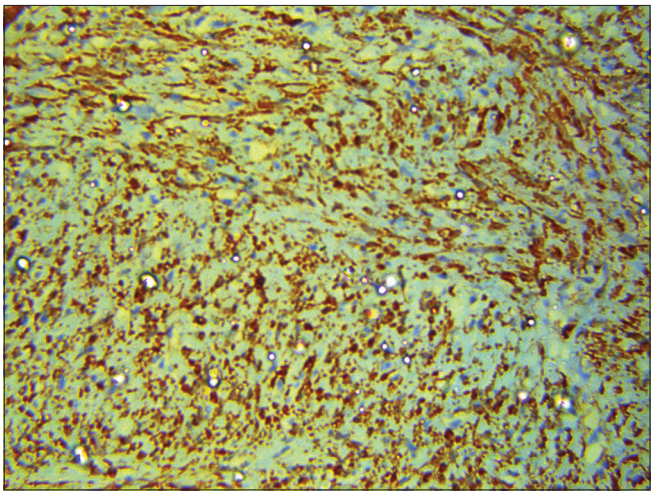

Figure 6: Immunohistochemistry

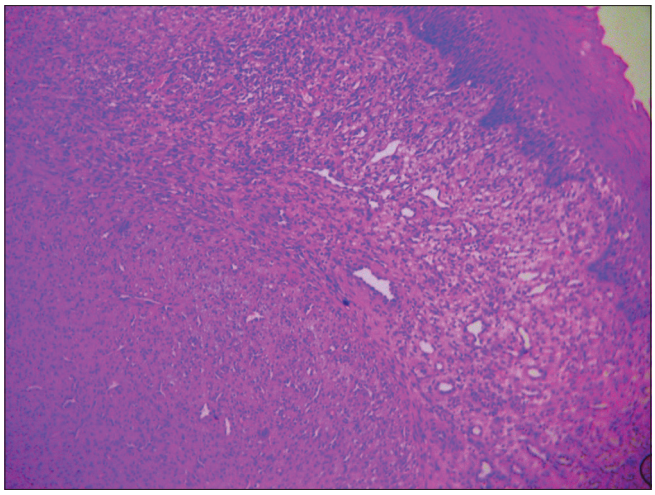

Figure 7: Histopathology

distinctive microscopic feature. Tumor cells were arranged in interlacing fascicles, the presence of mild nuclear atypia, increased mitotic figure and no tumor necrosis seen suggestive of low-grade spindle cell neoplasm [Figure 7]. IMT has been associated with high degree of cellular atypia and increased mitotic figure.

Since the histopathological findings were inconclusive and suggested low-grade spindle cell neoplasm, IHC was performed. The IHC report was positive for alpha-SMA and CD68 markers and negative for ALK-1 and S100. Considering all the above clinical, histopathological and IHC factors, a final diagnosis of IMT was made and complete surgical excision of the lesion was performed. Recall after 2 weeks of surgery showed satisfactory wound healing and recall at 6 months showed no evidence of recurrence.

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 initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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