ISSN : 2350-0743

www.ijramr.com

International Journal of Recent Advances in Multidisciplinary Research

Vol. 03, Issue 07, pp.1639-1641, July, 2016

REVIEW ARTICLE

INFLAMMATORY MYOFIBROBLASTIC TUMOR OF MANDIBLE: A RARE OCCURANCE

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ARTICLE INFO

Article History:
Received 17th April, 2016
Received in revised form
26th May, 2016
Accepted 14th June, 2016
Published online 31st July, 2016

Keywords:

Inflammatory Myofibroblastic Tumor,
Inflammatory pseudo-tumor,
Mandible.

ABSTRACT

Inflammatory Myofibroblastic Tumor is a rare neoplasm of unknown etiology and pathogenesis. It clinically manifest as mass with rapid development that may occur at almost any site of body, but rarely arise in oral cavity. The lesion has a potential for recurrence, persistent local growth, progression to frank sarcoma and metastasis. We hereby describe a case of Inflammatory Myofibroblastic Tumor of mandible occurring in a 45 year old lady. Because of some aggressive clinical, histological, and radiological features, this lesion may be confused with a malignant tumor. Therefore, its correct recognition is important to avoid unnecessary extensive and radical therapeutic approaches.

INTRODUCTION

Inflammatory Myofibroblastic tumor (IMT) is rare neoplasm of unknown cause. It was first observed in lung and described by Brunn in 1939. It was called inflammatory pseudo-tumor until 1994. (Narla *et al*., 2003 and Binmadi *et al*., 2011). The term inflammatory myofibroblastic tumor (IMT), was given in the classification of World Health Organization established in 2002 (Ishihara *et al*., 2010). The common sites of involvement include the lung, liver and orbit, but it has been reported to occur in nearly every site of the body, including the major salivary glands and the oral cavity (Narla *et al*., 2003; Binmadi *et al*., 2011 and Shek *et al*., 1996). Head and neck lesions represent 14 to 18 percent of extrapulmonary IMT cases; amongst these, oral lesions are uncommon (Brooks *et al*., 2005; Brooks *et al*., 1997).

Case Report

A 45 year old female patient reported to our unit with a painless swelling on the right side of the jaw since 2 years. The swelling started as a peanut size and gradually grew to the present size. The lesion grew rapidly over the last six months. Oral examination revealed an approximately 3 centimeter (cm) in its greatest dimension, hard and sessile well circumscribed large mass of the same colour as the surrounding mucosa on the right gingivobuccal sulcus (Figure1). There was no bleeding or exudates in the area. There was no regional lymphadenopathy. The physical examination was normal. The patient's medical and social history was non contributory.

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She was not on any medications. The patient had a history of betelnut chewing for the past 20 years. Radiographic examination was unremarkable. Based on the history and clinical examination, differential diagnosis of fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma and gingival epulis were made. All hematological and biochemical parameters were within normal limits. The lesion was excised under local anaesthesia and specimen was sent for histopathological examination (Figure 2).

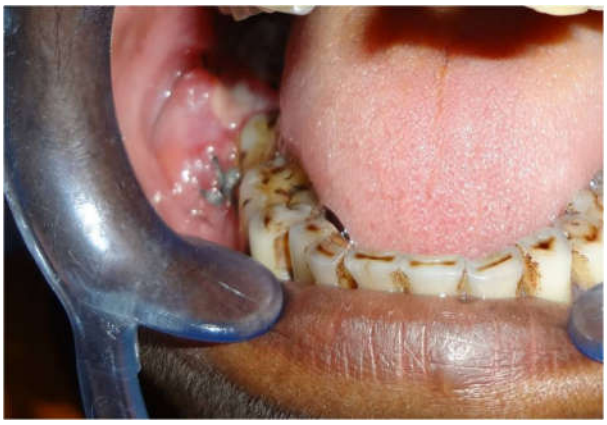

Figure 1. Preoperative Lesion

Macroscopically, the excised specimen measured approximately 3 × 2 × 2.5 cm. It had an irregular shape, grayish-white color and firm consistency. Microscopically, the lesion was composed of myofibroblastic spindle cells, fibroblasts, histiocytes (Figure 3). The inflammatory infiltrate also contained neutrophils, lymphocytes and macrophages.

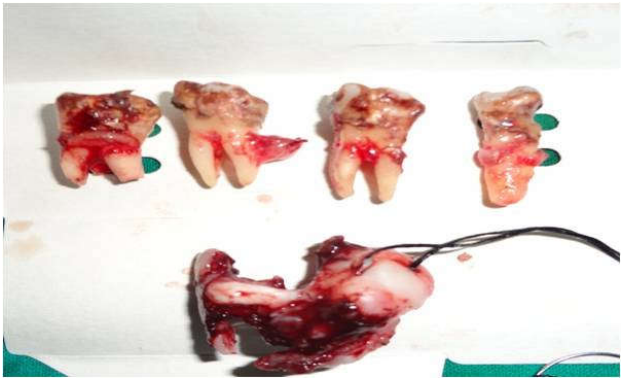

Figure 2. Excised specimen sent for Histopathology

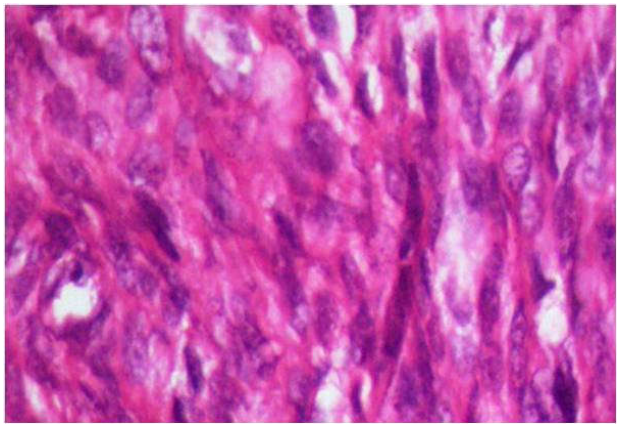

Figure 3. Histopathology Slide

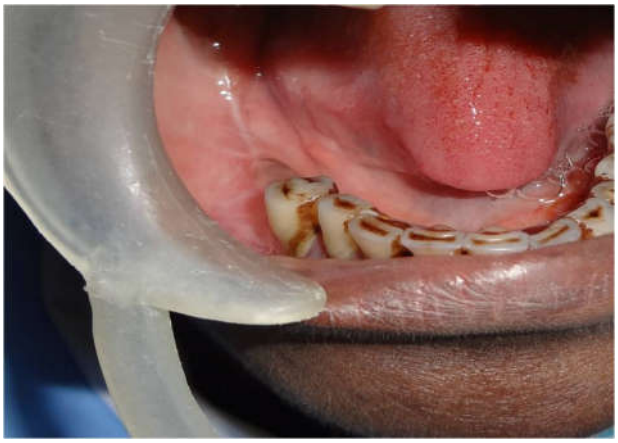

Figure 4. Postoperative Healing

Based on these findings, a diagnosis of Inflammatory Myofibroblastic Tumor of Mandible was made. The patient was followed up for 18 months with no recurrence (Figure 4).

DISCUSSION

Inflammatory Myofibroblastic Tumor is a chronic inflammatory mass which presents as a benign clinical course. IMT also known as plasma cell granuloma, myoblastoma or inflammatory pseudotumor is a lesion charecterised by proliferation of myofibroblastic spindle cells with mixed inflammatory infiltrates of plasma cells, lymphocytes, eosinophils and histiocytes. This tumor was earlier thought to be reactive in nature, but is now considered to be a true neoplasm with potential for recurrence and multifocality (McKinney *et al*., 2007). However the exact etiology and pathogenesis of this spectrum of inflammatory disease is largely unknown. It has been suggested that the host defense mechanism against infectious agents, micro-organism, neoplasm, foreign bodies and trauma may play a role in etiology of this type of tumor. Trauma and viral infection i.e. herpes simplex virus type 8 (HSV 8) and Epstein-Barr Virus (EBV), as well as autoimmune reaction have also been suggested as etiological factors. Nature of this tumor is controversial. Because of its rarity, current conception is altered from benign reactive process to an intermediate neoplasm (McKinney *et al*., 2012).

According to World Health Organisation (WHO), IMT is classified as tumor of intermediate biological potential due to tendency of local recurrence and small risk of distant metastasis (Binmadi *et al*., 2011). The majority of IMT lesions present in lungs. Extrapulmonary sites include the abdomen, head, neck and CNS. The most common sub sites in the head and neck include orbit, oral cavity, tonsils, larynx, thyroid, parapharyngeal space, parotid and lacrimal glands (Ide *et al*., 1998). IMT can present with variable clinical manifestations. Therefore, clinical manifestations of IMT or radiographic images have poor specificity to make a diagnosis, as it was in the present case also. Diagnosis of IMT is based on histopathological and immunohistochemical findings according to the WHO classification of soft tissue tumors. IMT of the oral cavity and head and neck region have been described priorly as benign lesions in various case reports. Therefore, it has been suggested that these lesions should be treated as low grade mesenchymal neoplasm. Because of confusion in management of IMT, different treatment modalities, such as steroid therapy, excision biopsy, radical surgery, radiotherapy and chemotherapy can be offered. IMT are tumor with unpredictable clinical behaviour, requiring complete surgical excision and continuous monitoring of clinical consequence. A long term follow up protocol should be instituted to prevent complications.

Conclusion

IMT in oral cavity may be confused with other malignant tumor on clinical, radiographic and histologic appearance. Therefore, it is important to recognize the distinction among them in order to provide better guidelines for treatment and outcome.

Acknowledgement

The case presented in this article was treated at the Department of Oral and Maxillofacial Surgery, Rajarajeswari Dental College and Hospital, Bangalore. I express my heartfelt gratitude to my colleagues and staff for their support. Last but not the least I would like to thank my patient who extended full cooperation.

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