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A rare case of peripheral odontogenic myxoma of mandibular posterior lingual gingiva

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Abstract

Odontogenic myxomas are uncommon, benign mesencyhmal tumors located in the head and neck region usually sited centrally in tooth bearing areas of the jaws frequently in the mandible. Soft tissue myxomas classified as peripheral myxoma is rare than the central type. It is slow growing with less aggressive behavior compared to the central myxoma with a low recurrence rate. Cases of peripheral odontogenic myxomas in maxillary gingiva have been reported in the literature. We hereby report a rare occurrence of peripheral odontogenic myxoma (POM) in mandibular posterior lingual gingiva in a 24 year old female with a conservative surgical approach and follow up.

Keywords

odontogenic myxoma; tumors; gingiva

Introduction

Odontogenic myxoma is defined as a benign odontogenic tumor of mesenchymal origin that is locally invasive and consists of rounded and angular cells that lie in abundant mucoid stroma [1]. The origin of odontogenic myxoma is thought to be from odontogenic ectomesenchyme of a developing tooth or undifferentiated mesenchymal cells in the periodontal ligament [2]. Peripheral odontogenic myxoma or intra-oral soft tissue myxoma is a rare slow growing benign mesenchymal tumor with less than six cases manifesting as the gingival soft tissue growth reported in literature [2]. We report a case of peripheral odontogenic myxoma without evidence of bone involvement. The extreme rarity of the tumor with a still rarer site of occurrence has prompted us to report this case.

Case Presentation

A 24 year old female patient presented with a chief complaint of pedunculated soft tissue growth on the lingual gingiva in relation to lower back tooth region since 2 years (Fig 1). On intra-oral examination the patient had a localized growth on the lingual aspect of the lower left posterior tooth region. The lesion was 3x2x1cm in size, translucent white in colour extending from 44 to 46 regions. On palpation the lesion was soft, fluctuant and not associated with pain or discomfort. Based on the clinical features a provisional diagnosis of pyogenic granuloma / peripheral giant cell granuloma / myxoma was made. Radiographic examination showed no bony involvement. The lesion was excised completely under Local anaesthesia and submitted for histopathology [3] in 10% neutral buffered formalin (Fig 2) and processed. The submitted Hematoxylin & Eosin stained sections revealed parakeratinized stratified squamous epithelium of uniform thickness with absence of rete ridges overlying connective tissue stroma composed of loosely arranged myxomatous tissue with areas of well-defined collagen bundles (Fig 3a). Occasional odontogenic epithelial rests / islands were also seen which were surrounded by eosinophilic hyalinised material (Fig 3b). Cells were spindle shaped and angular with few blood vessels in the connective tissue. Inflammation was scanty with areas of extravasated RBC's. For further confirmation of mucoid areas, Alcian blue stain with a pH of 2.5 was used. The tissue showed positivity, due to the presence of mucopolysaccharides (Fig 4). Hence, the overall histopathological features were suggestive of peripheral odontogenic myxoma.

Discussion

Odontogenic myxoma (OM) of the jaw was first described by Thoma and Goldman in 1947 [2]. In Asia, Europe and America, frequency for odontogenic myxoma ranges between 0.5% and 17.7% of the odontogenic tumors [2]. OM can occur at any age from childhood to the elderly. It occurs more frequently in the second to third decade [1,2]. However it has been stated that the frequency of OM in childhood might be higher than other aggressive odontogenic tumors.

Myxomas of the jaw can be classified as odontogenic or osteogenic [1]. Odontogenic myxomas can be classified in two types, central myxomas located in the bone or peripheral myxomas located extraosseously or in the soft tissue overlying the tooth-bearing areas [4].

The origin of OM is thought to be from Odontogenic ectomesenchyme of a developing tooth or undifferentiated mesenchymal cells in the periodontal ligament [2]. The Odontogenic origin of the neoplasm is supported by its histological similarity to dental follicle ectomesenchyme, its exclusive occurrence in close proximity to the tooth bearing parts of the jaws, occasional association with missing or unerupted teeth and presence of inactive odontogenic epithelium in a few cases [2].

Myxomas of bone (central myxomas) are common in facial skeleton than in extremities, whereas peripheral myxomas or soft tissue myxomas are mostly deeply situated lesions, occurring in the skin or the subcutaneous tissues, the genitourinary tract, or in organs such as the liver, spleen or even the parotid gland. Intra-oral peripheral myxomas are rare when compared to odontogenic myxomas [2]. Also they do not show any particular radiographic features unlike central myxomas which present as radiolucent areas honeycombed by an internal trabecular pattern.

Peripheral odontogenic myxomas are believed to arise from odontogenic ectomesenchyme of the dental follicle. It is suggested that in a fully developed jaw remnants of odontogenic ectomesenchyme are found in periodontal ligament (PDL) and gingiva thus, ectomesenchymal cells in both these locations could possibly serve as a stem cell population for neoplastic proliferations with the microscopic features of an odontogenic myxoma [5]. Several theories concerning the pathogenesis of peripheral myxomas have been reported. According to Barnes altered fibroblasts or myofibroblasts could produce an excess of mucopolysaccharides and were commonly incapable of forming mature collagen even if some cells could retain this capacity [6]. However, histogenesis of these lesions remains obscure and further studies are necessary to clarify its origin. Pathologically, it may be difficult to differentiate from other tumours with a myxoid stroma and is occasionally misinterpreted as malignant [6].

Central Odontogenic myxomas are common whereas incidences of peripheral odontogenic myxoma are reportedly less. No information of POM's is subsequently recorded in standard text books of Oral Pathology [2]. In few cases the authors have termed the gingival soft growths as soft tissue myxoma [6]. Also presence of occasional inactive epithelial rests has been reported in literature which was also seen in our case [7]. It is also suggested that these tumors have unlimited potential to grow. Mitoses are rarely seen. Most POMs are likely to be diagnosed clinically and microscopically as irritational fibroma or fibrous epulis. These tumors grow both in the maxilla and mandible with no sex predilection. Irritational fibromas are inflamed and subsequent edematous change in them mimics the appearance of POM. Therefore, true incidence of POMs may be higher than reported. Hence, histochemical stains such as Alcian blue as performed in our case can be helpful in diagnosing the myxoid appearance of connective tissue. On the contrary, immunohistochemistry is of limited value in establishing the final diagnosis as a specific marker for cells of dental ectomesenchymal origin is lacking [5]. Conservative surgical excision is the treatment of choice and recurrences have been reported between 3-8 % [4,7].

Conclusion

The central odontogenic myxoma is a common lesion while the incidences of peripheral odontogenic myxomas are relatively few as reported in literature. The rare site of occurrence of this lesion i.e the lingual gingiva of the posterior teeth in the mandible of a female patient has been the reason for reporting the case and contributing its occurrence to the literature. Our case did not show any post – operative recurrence and the patient is under periodic follow-up.

Figures

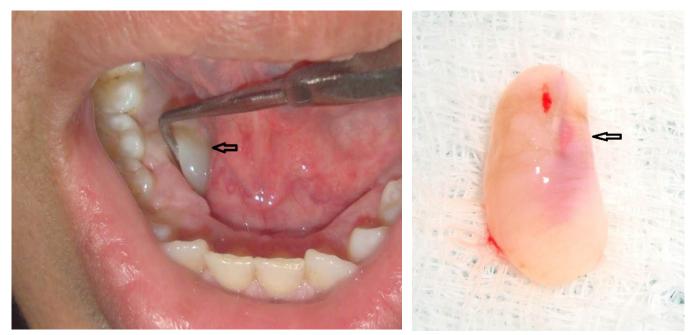


Figure 1: Lesion on mandibular lingual gingiva

Figure 2: Gross pathology

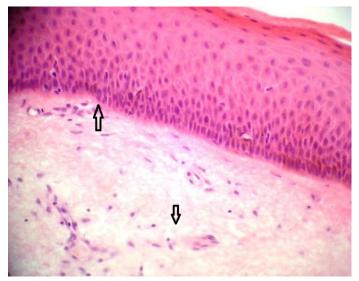


Figure 3a: Photomicrograph with overlying epithelium, flat rete ridges and loosely arranged myxoidstroma. (H & E, 400X)

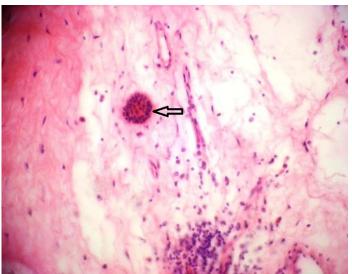


Figure 3b: Photomicrograph showing scanty odontogenic epithelial rest in myxomatous connective tissue stroma (H & E, 400X)

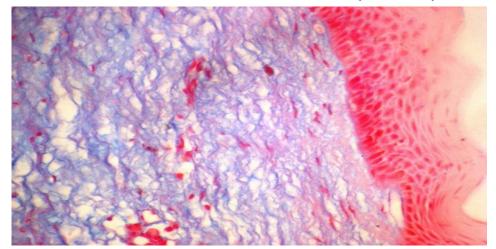


Figure 4: Photomicrograph confirming presence of mucopolysaccharides (Alcian blue stain, 400X)

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