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Case report

Peripheral adenomatoid odontogenic tumour: Case report and review of literature

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ABSTRACT

The peripheral adenomatoid odontogenic tumour is an uncommon subtype of adenomatoid odontogenic tumour (AOT).It has a female preponderance commonly in theage group of 3–25 years. Anterior maxilla is themost frequent site of involvement. Clinically, it is manifested as soft tissue mass on gingiva with infrabony pocket and minimum bone involvement. So this lesion is easily misdiagnosed by the clinician as simple gingival growth.Histopathological features are identical to that of their intra-osseous counterpart.We present a case of 27 years old female who had her lesion on the gingiva of right maxillary canine region which is notausual site of involvement. Few cases have been reported in the literature but all exclusively involved the gingiva of maxillary incisors. In this context extensive study is needed to figure out the exact site distribution and clinical presentation of the disease.

1. Introduction

In 1905, Steenland first described about a group of Adenomatoid odontogenic tumour (AOT) like lesions as “epithelioma adamantinum”. Two years later, similar lesion was described by Dreibladt as “psudoadeno ameloblastoma”. Harbitz, in 1915, also mentioned about a cystic adamantinoma. Stafne, in 1948, first considered this lesion as a distinct entity. Unal et al. produced a list of nomenclature for this lesion before naming it as adenomatoid odontogenic tumour (AOT). The names were adeno ameloblastoma, adenoameloblastic odontoma, ameloblastic adenomatoid tumour, epithelial tumour associated with developmental cyst, adamantinoma, psudoadenomatous ameloblastoma, epithelioma adamantinum and teratomatous odontoma. The widely accepted name AOT was proposed by Philipsen and Birn in 1969 [1]. WHO accepted the name and included it in the classification system in 1971. It was retained in the second edition in 1992 and the latest edition in 2005 with inclusion in the first group of classification of odontotogenic tumours under the heading of “tumours of odontogenic epithelium without ectomesenchyme”. The reasons for this were the absence of ectomesenchyme in immunohistochemical staining and absence of dysplastic dentine respectively. Regarding histogenesis, AOT is a benign neoplasm or a harmartomatous growth which is still debaTable Some investigators consider it as being a metaplastic process rather than epithelial-ectomesenchymal interaction [2]. Which specific stimulus triggers the proliferation of the progenitor cells of AOT is still unknown. Evidence suggests that follicular AOT arise from the reduced enamel epithelium which lines the follicles of unerupted teeth [3]. Philipsen et al., in 1992, strongly suggested that AOT develops from remnants of dental laminae in gubernaculum dentis or gubernacular canal. All the topographical variants having unique histological characteristics are explainable as unified source of origin within the gubercular canal [4]. AOT is a benign harmartomatous slow growing lesion and occurs in both extra osseous and intra osseous locations on the tooth bearing area of both the jaws. AOT accounts for approximately 2.2–7.1% of all odontogenic neoplasms. Out of all the AOTs, approximately 96% is intra-osseous while only 4% is extra-osseous (peripheral) [5]. This data signifies that the peripheral variety is rare. The present article describes about a case of peripheral variety of AOT in an unusual location clinically.

2. Case report

A 27 years old female reported for histopathological examination at Department of Oral and Maxillofacial Pathology of Dr. R. Ahmed Dental College & Hospital from Oral Surgery Department of the same institution after excision of a small gingival growth on mesial marginal and attached gingiva of upper right canine tooth. As per case history sheet available with the specimen, the lesion was approximately

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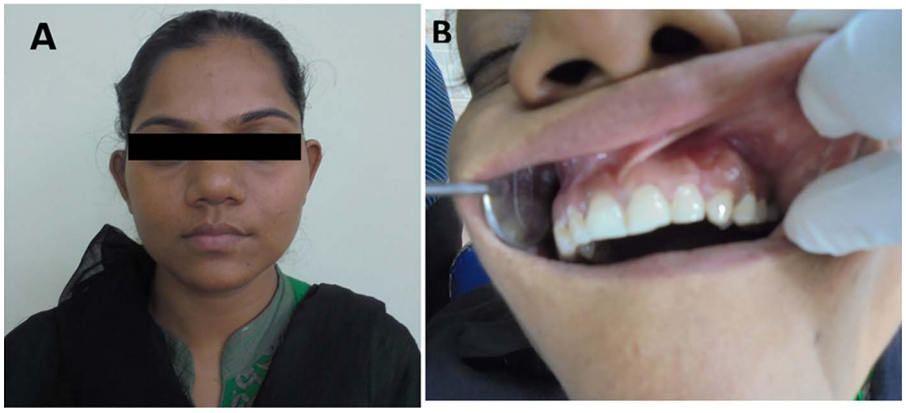

Fig. 1. A. Extra-oral appearance B. Intra-oralPost-operative clinical image of lesion site.

(3 × 1.5 × 1.5) cm in size and was located on the attached gingiva, about 0.7 cm away from the marginal gingiva (Fig. 1).

It was firm in consistency and of same colour as the adjacent gingiva, being confined on alveolar mucosa exclusively. The mass slowly grew in size, was symptomless and present since last 1 year approximately. Intra oral periapical X-ray revealed no bone loss in the periapical, interdental as well as inter-radicular areas (Fig. 2). In spite of that, alveolar crestal bone loss is evident which is not related with the disease process described herein but in all likelihood is the result of underlying periodontal problem.

It was provisionally diagnosed as pyogenic granuloma. We proceeded for histopathological evaluation by following standard laboratory protocol. Tissue section stained by hematoxylin and eosin showed loose butthick collagenous connective tissue stroma within which the polyhedral to spindle shaped tumour cells were arranged in varying patterns form solid islands, nests to rosette like structures. In the lumen of the rosette like and duct like structures eosinophilic amorphous material was found (Fig. 3). The overall histopathological features are corroborative to Peripheral Adenomatoid Odontogenic Tumor.

3. Discussion

PAOT is a rare lesion of the oral cavity and constitute only 3.4% of the odontogenic tumours [6]. Histological presentations do not differ between central and peripheral AOT. Peripheral AOT occurs in the soft tissue which overlies the tooth bearing areas of the jaws [7]. They typically present as soft masses of the gingiva like an epulis, as in most of the other peripheral odontogenic tumours. PAOT has a female predilection in the ratio of 2:1 and it is seven times more common in the maxilla than in the mandible [8]. Its most common site is the labial mucosa of anterior maxilla, but our case was located on labial mucosa of left maxillary premolar region, which in itself is also a rarity.

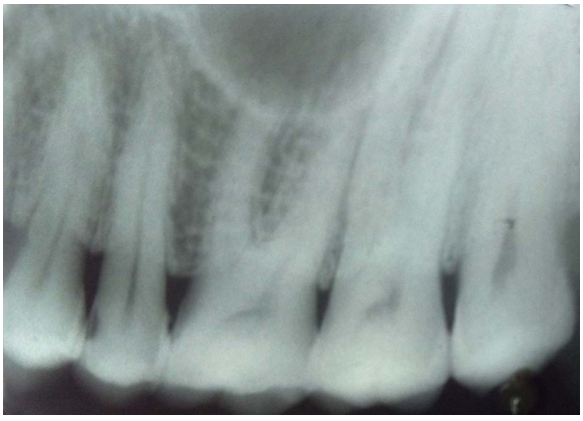

Fig. 2. Intra oral periapical X-ray showing no cortical bone loss.

The summary of data from PAOT from available literature is summarized in Table 1. The ages ranged from 4 to 21 years with the mean of 14.3 years. However in our case the age was 27 years, higher compared to the previous published cases. Out of 17 reviewed cases 13 were observed in female and 4 were observed in males indicating a clear female predilection. Our presented case was also female. Peripheral lesion mainly occurred in maxilla (82%) while 18% occurred in mandible. Among the reviewed cases 71.4% of the lesion occurring in the upper central incisor region and 21.4% in the canine region and 7.2% lesions were observed in the premolar area. Radiographic features were available only in 10 cases but only one showed no bone involvement which was similar to our presented case. Duration of the disease was ranges from 3month to 4 years in reviewed cases.

The differential diagnosis for these peripheral epulis-like lesion ranges from fibroma, pyogenic granuloma, peripheral giant cell granuloma and neurofibromasto the rarer connective tissue neoplasms likeperipheral odontogenic neoplasms. The most interesting part of this lesion is that most of the times they are diagnosed when excised and histopathologically examined presuming them to be an innocuous growth like epulis.

Macroscopically AOTs appear as an encapsulated spherical soft tissue mass. Microscopically, proliferation of odontogenic epithelium with formation of duct like structures of variable sizes and solid nests of cuboidal or columnar cells were noted in the connective tissue. Diffuse calcification may be present in some cases. The presented case was not showing any connective tissue capsule but areas of diffuse calcification were noted.

Although it is very slow growing, some lesions have been reported in literature which even attained a size of 6–7 cm. Extension into the intracranial space of a recurrent tumour has been reported also.

The treatment recommended is enucleation or excision followed by curettage. When thick connective tissue capsule is present, it facilitates the enucleation procedure. Prognosis of such treatment protocol is usually excellent and the risk of recurrence is extremely low. Malignant transformation has never been reported.

4. Conclusion

Based on currently available evidence and the findings ofthe present case of peripheral AOT we reiterate that it is very rare. Herein the site and age of occurrence are also uncommon. This peripheral AOT was initially provisionally diagnosed as a case of pyogenic granuloma. It was excised and finally histopathologically diagnosed as peripheral AOT. It is a common practice to excise innocuous epulis like lesions without going for incisional biopsy beforehand which later are histopathologically proved to be peripheral odontogenic tumours. Caution should be maintained regarding such presumptive and overzealous