Case Report

Peripheral adenomatoid odontogenic tumour

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Abstract

The Adenomatoid Odontogenic Tumour (AOT) is more than 100 years old, known by different names and the term AOT was coined in 1969 by Philipsen and Birn. AOT frequently occurs in intra-osseous location in gnathic skeleton and rarely seen in peripheral forms. These are rare odontogenic tumours, accounting for 2.2--7.1% of all odontogenic tumours, whereas the central or intraosseous variant of AOT in follicular and extra-follicular presentation, accounts for 95.6% of all AOTs. The peripheral variant presents as a gingival mass, which may cause slight saucerization of alveolar bone or might not affect the bone at all. These peripheral variants constitute 4.4% or less, of all AOTs of the gnathic skeleton. Here, we present a rare case of a peripheral variant of AOT in a 35-years-old female, 1.5 to 2 cm in dimension seen on the labial gingiva in right canine-premolar area. The lesion showed marked bone loss in 13 and 14 teeth region, causing mobility of 13. Histopathology showed a well encapsulated tumour mass with metaplastic bone in the capsule. A conventional AOT with ductal and rosette pattern was seen with tumour droplets and amyloid like material. The case clinically presented a moderately aggressive behaviour.

Keywords: Capsule with metaplastic calcification, gingiva, induction-tumour droplets, interdental bone loss, odontogenic tumour, peripheral AOT

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INTRODUCTION

The term Adenomatoid odontogenic tumour (AOT) was given by Philipsen and Birn in 1969. It has been described by numerous authors under diverse terminology like "adamantoma", "epithelial odontome", "cystic adamantoma", "adenoameloblastoma", "tooth germ (or chorioblastomatous) cyst of the jaw", "epithelial tumours associated with developmental cysts of the maxilla" and several more dating names and terminology from 1877.[1-7] AOT is a benign odontogenic tumour belonging to the category of odontogenic epithelial tumours occurring with a relative frequency of 0.6--38.5%. The tumour commonly presents in an intra-osseous location in the maxillary anterior segment in follicular and extra follicular types accounting for more than 95.6% of reported cases. Rarely, we get to see the peripheral variant (4.4%),[1,2] attached to the gingiva of the maxillary anterior teeth causing little or no erosion of the alveolar crest bone. The peripheral variant is also called as extra-osseous tumour, soft tissue odontogenic tumour, and odontogenic tumour of the gingiva.[8] Histopathologically, they demonstrate the same pattern as the intra osseous or conventional AOTs with a well-formed capsule, tumour droplets, and other inductive tissues. Here we report a rare peripheral

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variant of AOT, measuring around 2--2.5 cm in diameter, causing destruction of interdental bone, mobility of the canine and histopathologically showing a rare finding of metaplastic bone formation in the tumour capsule. The present epithelial tumour showed a classic appearance of cuboidal and spindle cells, arranged in rosettes and ducts with tumour droplets.

CASE REPORT

A 38-years-old female reported to the Department with a painless gingival mass in the maxillary anterior segment. The patient presented with a gingival mass of 2—2.5 cm diameter on the maxillary right quadrant involving the gingiva of right canine (13) extending to the premolar area. A history of slow growth of the lesion in the past 3 months was given. Asymptomatic and painless, the mass caused discomfort during mastication and was unesthetic. The gingival mass was covered by normal coloured mucosa [Figure 1a].

On palpation, a well-defined, demarcated soft tissue mass 2 cm in diameter, soft to firm in consistency, and sessile in nature was noted. Examination of teeth revealed grade III mobility in 13. The roentgenograph showed vertical and horizontal loss of interdental bone in between the canine (13) and premolar (14) teeth, extending more than 2/3rd the root length. An irregular horizontal bone loss was noted in between 13 and 14 and right lateral incisor (12) and 13 area [Figure 1b].

A provisional diagnosis of a reactive lesion-pyogenic granuloma was made and a conservative excision was carried out under local anaesthesia along with the extraction of 13.

On grossing, the gingival mass was well demarcated, homogenous, off-white in colour on a cut section and was soft to firm in consistency. Few areas of small specks of haemorrhage were noted. The stereo zoom image of the gross specimen showed the well encapsulated lesion to be located below the gingival epithelium [Figure 2a and 2b]. The radiograph of excised specimen showed soft tissue shadow.

On histopathological examination, mild pseudoepitheliomatous hyperplasia of the surface epithelium was seen with deeper connective tissue showing a well encapsulated tumour [Figure 3a]. The capsule was mature, well-formed and showed discontinuous patches of linear metaplastic bone formation [Figure 3b]. The metaplastic bone was seen distributed linearly along the capsular tissue. The tumour displays various epithelial patterns in a solid area [Figure 3c]. Numerous solid cell balls; cellular epithelial sheets; cells arranged in ductal pattern, rosettes, and reticular pattern with eosinophilic coagulum like

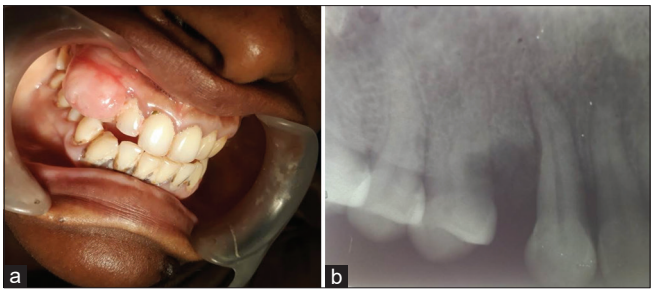

Figure 1: (a) Soft tissue mass in the upper maxillary right canine and premolar region, measuring approximately 2 cm in diameter. (b) Roentgenograph showing irregular horizontal bone loss in right canine premolar region

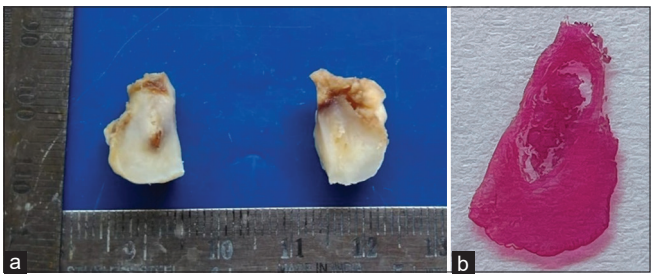

Figure 2: (a) Gross specimen- cut section showing homogenous off-white area with specks of hemorrhage. (b) Steromicroscopic image of the H&E stained tissue section in relation to the cut surface of the gross shows peripheral surface epithelium and well circumscribed tumour mass in the underlying connective tissue of the gingiva

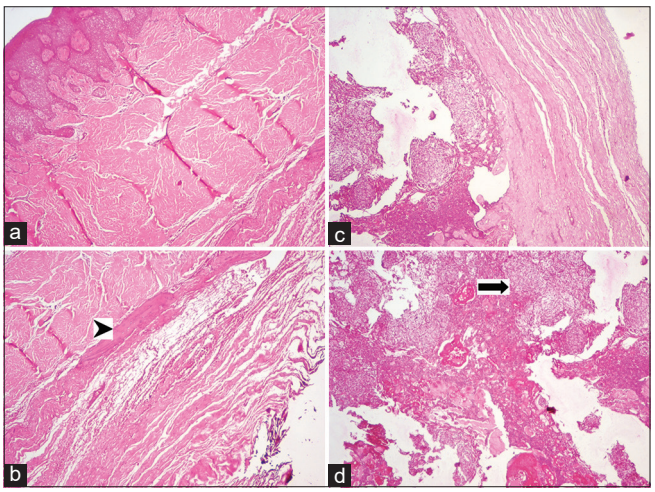

Figure 3: (a) Surface epithelium exhibits pseudoepitheliomatous hyperplasia [H&E stain, x40]. (b) The capsule of the lesional tissue exhibiting linear bone formation (black arrow head) walling off the tumour from surrounding connective tissue [H&E stain, ×40]. (c) Scanner view exhibiting well encapsulated tumour mass with odontogenic epithelial cells arranged in different patterns [H&E stain, ×40]. (d) Scanner view shows odontogenic epithelial cells arranged in solid cell ball pattern (black arrow), sheets, ducts, rosettes, reticular pattern and eosinophilic coagulum with scanty intervening connective tissue stroma [H&E stain, ×40]

areas were seen with scanty intervening connective tissue stroma [Figure 3d]. Many areas showed the presence of tumour droplets, small inductive eosinophilic areas in relation to the epithelial cells [Figure 4a], and areas of basophilic calcifications [Figure 4b]. Predominantly formed by rosettes, ductal pattern [Figure 4c] and solid cell balls of varying sizes, the solid odontogenic tumour was diagnosed as a peripheral variant of Adenomatoid Odontogenic Tumour (PAOT).

DISCUSSION

AOT a benign epithelial odontogenic tumour is frequently called by the name of "two-third tumour" as 2/3rd of the cases are associated with females, 2/3rd in the maxillary bone, 2/3rd are associated with impacted tooth predominantly or 2/3rd times with the maxillary canine. AOT is frequently seen in women in the ratio of 14:1 as a intraosseous tumour in the canine-premolar area of young individuals.[1-4,8-10] The peripheral variant is the rare variant with 2.3--4.4%[8] occurrence rate. The conventional AOT is seen in an age group of 13--19 years with a peak in 2nd decade. The mean age of occurrence of PAOT is around 13 years, presenting at an earlier age than the follicular and extra follicular intra osseous variants. The present case was seen in female patient and is in unison with the literature of various reports.[1-3,8,10] But in stark contrast to other case reports of PAOT, where they are seen in earlier decades or in adolescents, the present case was seen in a 35-years-old individual.

The follicular and extra-follicular variants are predominantly seen in the maxillary canine- premolar area. The peripheral variants are also frequently reported in anterior maxillary gingiva as described by Philipsen & Reichart[1,2,9] and Gomez *et al*.[10,11] The present case is in harmony with this finding and was seen involving the maxillary labial gingiva in the canine-premolar (13 to 15) region.

PAOT infrequently show any pathognomonic radiographic findings, but a few cases may show shallow erosions or bone loss.[1-3,8] In contrary to this finding, our case showed extensive horizontal bone loss interdentally between 12--13 and 13--14 leading to mobility of the permanent right maxillary canine. The bone loss in relation to the canine was confirmed while doing the excisional procedure along with extraction of 13.

The histogenesis of AOT is associated with dental lamina cells and its remnants. The other suggested theories include role of enamel organ, reduced enamel epithelium and stratum intermedium. In case of histogenesis of the peripheral variant, the lesion can be categorized as an hamartoma or a benign tumour. Some variants are also categorized as hybrid variant by Philipsen *et al*.[3,4,10,12] Lesions of early age are generally regarded as hamartoma and those that occur later in life as tumours. Our case is in agreement to the tumour form, as it is seen in third decade and has presented with well-formed tumour capsule.

AOT on histopathological analysis has a very classic and unique pattern of arrangement of tumour cells with a well-formed distinct capsule. Some of the PAOTs might not show the presence of a capsule[8,10] but demonstrate the similar pattern of tumour epithelial cells[8] as in the intra osseous variant. The characteristic histopathological presentation includes presence of cuboidal or columnar cells with single layer of cells forming ductal pattern, small and large rosettes along with numerous spindle cells. Various other patterns that are seen in AOT include reticular, solid cell balls, cystic areas, plexiform, and trabecular patterns.[9,10] Amorphous homogenous eosinophilic areas seen in between the tumour cells called as tumour droplets and homogenous dystrophic calcification are a common finding. All these patterns are supported by a very delicate and scanty connective tissue stroma with vascular elements.

The histopathology of the present PAOT relates to conventional intraosseous AOT. The rare finding in our case was the presence of a well-formed capsule with

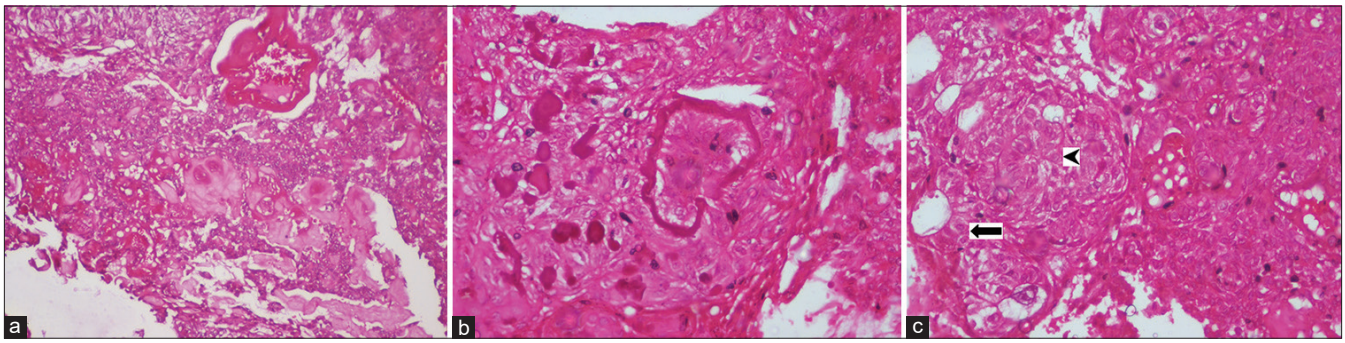

Figure 4: (a) Photomicrograph shows tumour droplets and eosinophilic areas interspersed between the tumour cells [H&E stain, ×100]. (b) Photomicrograph shows basophilic calcifications [H&E stain, ×100]. (c) High power view of the ducts and rosettes [H&E stain, ×100]

metaplastic bone formation. Not all cases of PAOT demonstrate a capsule but here we saw a well capsulated PAOT with discontinuous rim of linear metaplastic bone formation in the mature hyalinized capsule. Reports of similar finding were not found in the English literature. The arrangement of epithelial cells in ductal, rosette, and cell ball pattern were similar to reports by Philipsen & Reichart, Gomez *et al*., Sadasivan *et al*. and Muniswamappa *et al*.[1-3,8,9,13]

The clinical presentation of PAOT can simulate other common gingival enlargements which are reactive lesions or tumours and have to be considered as differentials. Pyogenic granuloma, peripheral ossifying fibroma, peripheral giant cell granuloma are frequently the provisional clinical diagnosis.[8,10] The histopathological revelation in the present case clears out all the differential diagnosis since it presented the classic histopathological features of an AOT.

A well encapsulated AOT---intra osseous or peripheral variants are well managed by a conservative surgical approach, as was done in the present case. The follow up is uneventful after 2 yrs.

CONCLUSION

AOT is a rare odontogenic tumour and the peripheral variant is still rarer. The documentation of the present case participates to enrich the literature. The report highlights the occurrence of the lesion in a middle-aged woman, exhibiting moderately aggressive behaviour, destroying the interdental bone to great extent causing mobility of the canine, and growing to the size of 2 to 2.5 cm. It also directs our attention to be aware of this pathology and consider it as a differential for the mass that occur in anterior gingiva in a middle-aged woman.

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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