

Peripheral adenomatoid odontogenic tumor of mandible - A synchronous presentation or a subtype?!

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Abstract

Adenomatoid odontogenic tumor (AOT) with its unique and varied presentations histologically and clinically has always been an enigma. AOTs are multifaceted in their appearance with reports pointing out to its occurrence as a synchronous tumor, a purely cystic variant or with multiple foci; however, the three commonly encountered forms are Follicular/Coronal, Extra Follicular/Extra Coronal and Peripheral variants. Here, we present a subtype of the Peripheral AOT (PAOT) seen synchronous with an intraosseous cystic variant of AOT in a 16-year-old female in the anterior mandible. This case is unique in that a PAOT occurring adjacent to a focally aggressive intraosseous cystic AOT (not associated with impacted tooth) could either be synchronous with no connection to the intraosseous component or could actually be an erupted intraosseous cystic variant with peripheral manifestation. Features of cortical expansion, perforation and displacement of teeth without resorption were seen. Histologically, all the classical features of AOT along with abundant eosinophilic amorphous fibrinous deposits, cellular vacuolization, clearing of cells, dystrophic and reactive bone formation and a cystic lining were seen. Thus, this case of PAOT would add to the myriad presentation of AOT making it one of the most often discussed odontogenic tumor.

Keywords: Cystic, erupted intraosseous variant, mandible, peripheral adenomatoid odontogenic tumor, synchronous

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INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is the 4th common odontogenic tumor, reviewed and reported quite frequently with nearly half of the reported cases seen in Asian population.^[1]

Debated to be hamartomatous or neoplastic by factions, the WHO (2017) defines it as a benign hamartomatous lesion and has classified it under category I originating and composed of only odontogenic epithelium even though its propensity for inductive changes indicate type

II categorization.^[2] The three commonly encountered forms are, (i) Follicular/Coronal, (ii) Extra Follicular/Extra Coronal and (iii) Peripheral AOT (PAOT) variants.

The PAOT, while quite rare (2.3% of all AOTs), shows a peak incidence for ages 9 to 17 and tends to be detected early due to its clinical appearance.^[3,4] It is considered hamartomatous if found around age 3 or below and a benign tumor in older age groups.^[5] Our case shows an erupted intraosseous PAOT variant similar to the observation by Philipsen *et al.* where PAOT is described

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to have two subtypes - the classic epulis type with no intraosseous component and the second showing a definite intraosseous connection which reaches the overlying gingiva presumably through the eruptive pathway.^[6]

The presence of a cystic lining in the intraosseous component of our PAOT along with certain features such as perivascular hyalinization and osteodentin formation again puts the emphasis on the need to check whether these findings influence its biologic behavior.

CASE REPORT

The gross specimens of a 16-year-old female patient with a provisional diagnosis of ossifying fibroma (central and peripheral) were received by the department of oral and maxillofacial Pathology. As per the clinical records, the patient had come with a complaint of a slow growing swelling present on the inner aspect of her left anterior mandibular teeth for 6 months. She had no pain. On examination, the lesion was a broad based pedunculated firm gingival swelling measuring 3 cm × 2 cm × 1.5 cm with smooth margins extending from 31 to 35 and obliterated the lingual vestibule in that region [Figure 1]. Her submandibular glands were palpable but non tender. No complaints of anaesthesia or paresthesia were elicited. Past medical and dental history revealed that she had typhoid few months ago and had multiple restorations for her posterior teeth 2 years ago, respectively. No record of trauma was present.

The radiographic features from an orthopantomogram and cone beam computed tomography showed a well circumscribed lesion involving regions 31 to 35 with buccolingual cortical expansion. The lesion exhibited small radio-opacities in its superior part causing displacement of adjacent teeth (33, 34 and 35). No association with impacted or unerupted tooth was found. On surgical excision of the pedunculated peripheral lesion they found that it had perforated the lingual cortex. The exploration of the buccal cortex through a bone window revealed a cystic



Figure 1: Clinical presentation of peripheral pedunculated lesion with a broad base along with central lesion of mandible

greyish mass which was continuous with the remnants of the mucosa remaining at the site of peripheral lesion. The central and peripheral specimens were submitted to the oral pathology department.

The specimen marked P/peripheral (3 cm × 2 cm × 1.5 cm) revealed a firm whitish hard mass while the central/C specimen (2 cm × 1.5 cm × 0.5 cm) was grayish, soft and cystic [Figure 2]. Both specimens were grossed, processed and sections were taken from different levels. The peripheral lesional sections showed uniform appearing odontogenic epithelial cells proliferating in sheets, whorls and alveolar patterns with duct like spaces. Secretory structures with tall columnar cells in duct like patterns were seen surrounding dental matrix like material. Eosinophilic droplets, perivascular hyalinization and calcified materials were found.

Sections from the hard mass of the peripheral specimen showed a para keratinized stratified squamous epithelium with vacuolization of cells. The underlying connective tissue exhibited patchy distribution of inflammatory cells, dense collagen bundles and large amounts of eosinophilic fibrinous deposits between the odontogenic clear cells.

The H & E sections of the central lesional tissue showed an odontogenic cystic epithelial lining, single to multi-layered with an underlying fibro-cellular stroma. Discontinuous, fragmented, tall columnar cells with areas showing stellate reticulum like appearance were seen within the stroma along with areas of dystrophic calcification and trabeculae of reactive bone formation. Deeper sections also showed discrete epithelial islands with definite whorls, rosettes and duct-like patterns [Figure 3]. Taking into account the clinical, histological and radiological features, a diagnosis of PAOT as an Erupted Intraosseous Cystic Variant was given.

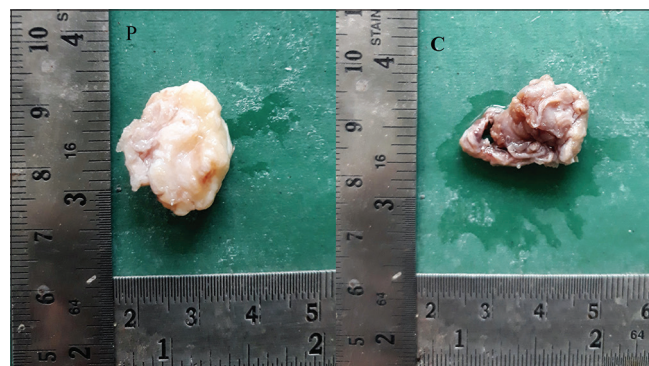


Figure 2: (P)-showing the peripheral excised specimen. (C)-Showing the central excised specimen

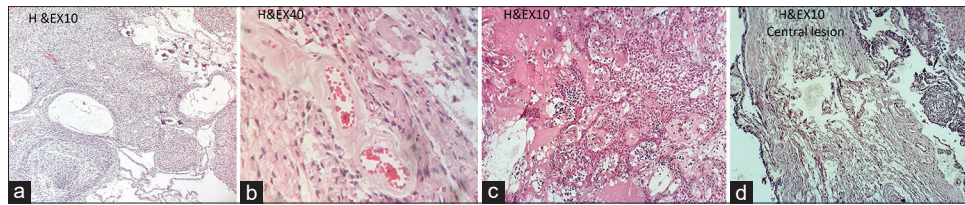


Figure 3: Left to right. (a) Peripheral lesional tissue section showing whorls, tubular and rosette like pattern and dystrophic bone formation (H&E, $\times 10$). (b) Peripheral lesional tissue section showing perivascular hyalinization (H&E, $\times 40$). (c) Peripheral lesional tissue section showing inflammatory cells, dense collagen bundles, large amounts of eosinophilic fibrinous deposits and odontogenic clear cells (H&E, $\times 10$). (d) Central lesional tissue section showing cystic lining and discrete epithelial islands (H&E, $\times 10$)

DISCUSSION

Cases of PAOT arise either *de novo* on the gingiva with minimal bone involvement or exhibit notable bone loss with an intraosseous origin and an association with erupting tooth. The later were called Hybrid Variants^[3] we have avoided the term “Hybrid” in accordance to the observations made by Philipsen *et al.*^[6] as we were unable to observe a combination of histopathological features of two or more different odontogenic tumours or cysts” Ide *et al.*^[7]

Among AOTs, the PAOTs tend to be detected early due to its clinical appearance but show similar predilection for females (1:5.3)^[8,9] and maxilla (10:1).^[10] However, recent literature has pointed out a paradigm shift in demography and biological nature exhibited by AOTs. An increasing mandibular involvement (69% anterior and 27% posterior quadrant) with cortical involvement is seen when the patient is 16 years or more.^[11] Studies even consider cystic AOTs as a separate variant with a few arising from unclassifiable odontogenic cyst having no relation to an impacted tooth.^[2]

Cystic AOTs generally have more predilection for males (1.6:1) and maxilla (1.6:1). While tending to be larger than their solid counterparts, there has not been any significant difference in their behavior and prognosis. However, assumptions on their aggressiveness have been attributed to the presence of perivascular hyalinization, osteodentin formation and mandibular involvement in few cases.^[2]

Among the 11 Aggressive AOTs reported so far in the literature 8 were in the mandible similar to ours, the mandibular lesions are seen to be larger, crossing the midline and predominantly in older individuals.^[12,13] We were able to find these similarities mirrored in our case with respect to the age, jaw, bone involvement and presence of a cystic lining not associated with erupting or impacted tooth and a greater degree of bone involvement. AOTs capacity to be proliferative and its aggressive behavior has been proved by their positive

proliferating cell nuclear antigen and p53 findings through immunohistochemical.^[14] Cases of multiple AOTs involving different foci at varied times are almost always associated with impacted supernumerary tooth. Though mostly nonsyndromic, AOTs associated with Schimmelpenning syndrome characteristically are multiple.^[15] Our case reports two foci of nonsyndromic AOTs in the mandible, an intraosseous and a peripheral manifestation without any association to impacted or supernumerary tooth. While radiological and histopathological findings point to two independent lesions, we assume that our PAOT could be an erupted intraosseous cystic variant based on the clinical findings during the surgical procedure.

Thus, in conclusion, the AOT continues to show greater diversity in its clinical presentation and though its histology has been remarkably distinct and original in all cases, there has been changes in its biologic nature that needs to be taken note of.

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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