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CASE STUDY

EXTRAFOLLICULAR ADENAMATOID ODONTOGENIC TUMOR : REPORT OF 2 CASES

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ABSTRACT

Adenomatoid odontogenic tumor AOT is a relatively uncommon, benign tumor accounting for 2.2-13% of all odontogenic tumors. It presents in three clinicopathological variants as: Follicular, Extrafollicular and Peripheral type. We present clinical features, radiological evaluation and treatmentof two rare cases of extrafollicular variant of AOT who reported to the department of oral and maxillofacial surgery.

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INTRODUCTION

Most common follicular Adenomatoid odontogenic tumor (AOT) appears as a pericoronal radiolucency. Its less common variant extrafollicular AOT, atypical radiographic appearance makes its clinical and radiographic diagnosis relatively difficult. This article will highlight two cases of extrafollicular AOT.

Case 1

A 35 year old female patient was referred with the chief complaint of enlargement of lower part of her jaw since the past three years. She had a history of extraction of her lower anterior tooth which was mobile, two years ago. Intra-oral examination revealed a single, well-defined, swelling obliterating the lower left vestibule measuring approximately 2x3cms extending mesially from distal of 41 to distal of 33 (Fig. 1). Superiorly it extended up to the incisal edge of 31 and 33. The swelling was non-tender on palpation. Tooth 33 was found to be mobile (Grade 1). OPG revealed a well-defined, unilocular, corticated, ovoid radiolucency with root displacement (Fig. 2). Electronic pulp testing (EPT) of 41 and 31 gave a normal response while a delayed response was found in 33.

Case 2

A 22 year old male patient was referred with the chief complaint of a slowly growing, asymptomatic swelling in the left upper side of face since last six months. The Intra-oral examination revealed a swelling in the upper left labial vestibule measuring 2x3cms. Antero-posteriorly it extended from the mesial of 22 upto the mesial of 24. Supero-inferiorly it extended from the canine fossa to the alveolar margin. The crowns of 22 and 23 were displaced distally and mesially respectively. On palpation the swelling was well defined, firm and non tender. CT scan showed a single, well defined, unilocular radiolucency with buccal thinning and expansion (Fig.3). However, both the teeth were found to be vital on EPT. Fine needle aspiration was negative for both the cases. With exploratory surgical approach lesions were enucleated in "to to" with intact capsule. Tooth 33 was extracted in case 1. The pathological specimens were histopathologically diagnosed as AOT.

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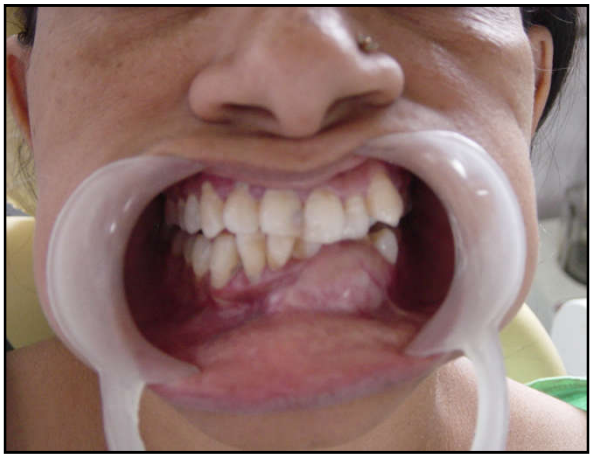

Case 1
Fig. 1. Pre-operative photograph (Intra oral)

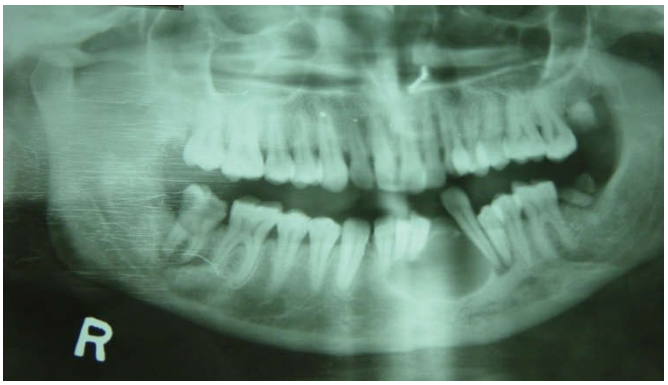

Fig. 2. Preoperative orthopantomogram of case 1

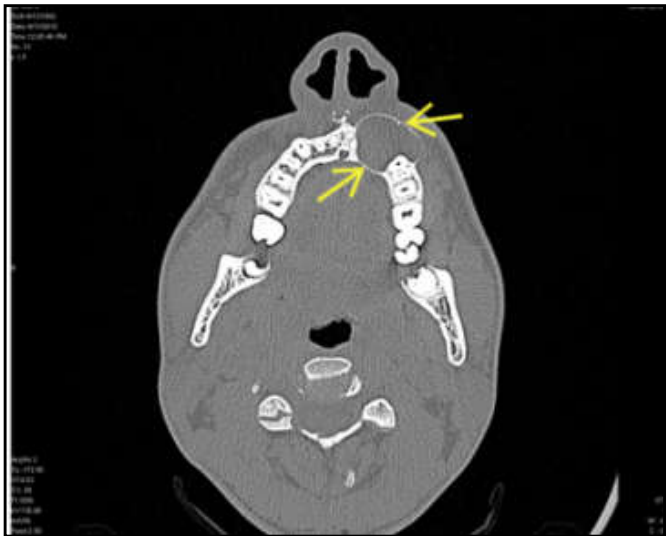

Figure 3. Preoperative CT Scan (axial section) of case 2 showing expansion of the cortex

DISCUSSION

Adenomatoid odontogeneic tumor has been reported to be a progressively slow growing, asymtomatic, benign tumor often associated with an unerupted tooth, often detected on routine radiographic examination. It presents commonly in the second and third decade of life and is found to be predominant in the maxilla (max: mand, 2.6:1) with a slight female predominance (2:1) (Philipsen *et al*., 1992; Swasdison *et al*., 2008; Philipsen *et al*., 2007; Handschel *et al*., 2005). AOT has three clinicopathological variants: Follicular, Extrafollicular and Peripheral. The follicular and extrafollicular variants are both intrabony (97%), out of which 73% are follicular (Philipsen and Reichart, 1998). Follicular variant is found as a well circumscribed unilocular radiolucency associated with the crown or often a part of an unerupted permanent tooth, most commonly the canine (67.5%) followed by the incisors (21.6%), premolars (13.5%) and molars (2.7%) (Philipsen and Reichart, 1998; Philipsen *et al*., 2007). Extrafollicular variant has no association with any unerupted tooth. It appears as a radiolucency located either between, above or superimposed upon the roots of erupted permanent teeth mimicking a periapical cyst mostly in anterior region of jaws. The peripheral variant accounts for 3% of AOT's and is located in the gingival mucosa, clinically appearing as a fibrous epulis (Philipsen and Reichart, 1998). The origin of AOT remains controversial although most authors agree on its odontogenic source. Disintegration of the complex system of dental lamina gives rise to numerous epithelial remnants (ER) which persist in the jaw bones and gingiva after the completion of odontogenesis. The initiation of proliferation of ER giving rise to tumors/ hamartomatous lesions though remain speculative. Continued tumor growth with initial tooth eruption may lead to contact and later fusion between tumor and reduced enamel epithelium of erupting tooth which sooner or later is embraced by the tumor giving rise to follicular variant of AOT. If tumor develops from epithelium remnants situated at the periphery of the gubernaculaum canal or if the eruption force of the tooth is not impeded by the developing tumor then tooth may bypass the tumor mass resulting in the extrafollicular variant of AOT. The peripheral variant most likely originates from the epithelial remnants of dental lamina. The fact that all variants of AOT share almost identical histopathology strongly suggest its commom origin (Philipsen *et al*., 1992; John and John, 2010). Both our cases were clinically, radiographically and histopathologically proven to be the extrafollicular variants of AOT. They were present in second to third decade of life which is in concurrence (Tejasvi *et al*., 2010; Kanagaratnam and Maarof, 2009; Philipsen *et al*., 2002; Yilmaz *et al*., 2009). In case 1, the anterior mandibular region was involved, while in case 2, the lesion was present in relation to 22 and 23. It usually attains a size of 1.5-3cm, though larger lesions have also been reported (Konouchi *et al*., 2002; Philipsen *et al*., 1992; Philipsen *et al*., 2002; Yilmaz *et al*., 2009). We found the size to range from 2-3cm. The adjacent teeth are found to be vital on EPT with an intact periodontal ligament and lamina dura on IOPA (Tejasvi *et al*., 2010; Philipsen *et al*., 2002). This can be attributed to the fact that lesions of smaller size outside the PDL usually do not interfere with the nerve and blood supply through the root apex (Philipsen *et al*., 2002). We also found the adjoining maxillary lateral incisor and canine to be vital on EPT in the second case. However, mandibular canine showed a delayed response. Radiographic calcifications have been reported in literature, however, it was not evident in our cases. MRI is found to be most sensitive for detecting intralesional calcifications (Konouchi *et al*., 2002). Our cases mimicked a radicular cyst, but were ruled out with negatvie

aspiration and vitality. Also, on surgical exploration the tumor lining was not attached to the roots of adjacent teeth, rather the lesion had displaced the adjacent roots giving credence to the extrafollicular variant of AOT. The uniformly benign biologic behaviour of nearly all AOT and the consistent presence of a well developed fibrous capsule, a complete surgical excision usually accompanied by enucleation and curettage is the treatment modality of choice with very rare rate of recurrence (Swasdison *et al*., 2008; Kanagaratnam and Maarof, 2009; Handschel *et al*., 2005). Complete surgical excision of the lesion was performed in both of our cases. However, mandibular canine in the first case was also extracted as the patient denied root canal therapy. No recurrence have occurred since the past 6 years.

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