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Adenomatoid Odontogenic Tumor – A Report of Two Cases & Literature Review

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Abstract

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor. It is a benign, painless, noninvasive, and slow-growing lesion and often misdiagnosed as an odontogenic cyst on clinical examination. It usually occurs in young age group with women predilection. It is of three types follicular, extra follicular and the peripheral type. It is the most commonly seen in maxilla usually in association with maxillary impacted canine and rarely in mandible. We present here two cases of adenomatoid odontogenic tumour in the unusual location. Both the cases are having the same clinical appearance but variation in their radiographic appearances.

Background

Adenomatoid odontogenic tumor (AOT) is a relatively rare odontogenic tumor with an incidence of only 3 to 7% among all odontogenic tumors. There are three variants of AOT, the follicular type (73%); extra follicular type (24%), and the peripheral variety (3%). 1 This tumor is sometimes referred to as "two third tumor" because 2/3rd cases are seen in maxilla, 2/3rd cases are seen in young age group, 2/3rd cases are associated with an impacted tooth and 2/3rd times the affected tooth is canine. 2 In this article we present two unusual cases of AOT in the mandible, of which one is of extra follicular variety and other is of follicular variety, with an emphasis on radiographic findings, histopathologic correlation, and a literature review of this tumor. CASE REPORT-1 A sixty-one-year-old male reported to the dental hospital with a complaint of mobile teeth and associated swelling in left side of mandible since 2 months. History revealed that the swelling was gradually increasing in size with mobility in

the associated teeth. The swelling was occasionally associated with dull, aching and non-radiating pain. Medical, personal and family history was non-contributory. On extra oral examination, dome-shaped swelling was present in the mandible, extending from the left parasymphysis region till the lower border of mandible, approximately of size 3x5cm. It was firm to hard in consistency, non-tender on palpation. Intraorally, circumscribed ovoid swelling was present on the buccal aspect from 41 to 35 regions Fig. 1: Showing intraoral swelling in left mandibular canine-premolar region with buccal and lingual expansion. Obliterating the labiobuccal sulcus. The swelling was firm to hard and non- tender. Mobility was present with the associated teeth 41, 31 to 35. Expansion of labial and lingual cortical plates was evident on palpation (Fig. 1). Based on history and clinical findings a provisional diagnosis of odontogenic cyst/ tumor was taken into consideration. On radiographic investigation, mandibular occlusal radiograph, panoramic radiograph and Cone Beam Computed Tomography (CBCT) showed a unilocular radiolucency with corticated borders extending from 43 to 36 with multiple radiopaque calcifications within the radiolucency. Expansion of buccal and lingual cortical plates and external root resorption of 34, 35 was evident (Fig. 2a, 2b, 2c). All these features were suggestive of a radiographic differential diagnosis of Unicystic ameloblastoma, odontogenic keratocyst, calcifying odontogenic cyst, adenomatoid odontogenic tumor, radicular cyst and central giant cell Granuloma

Electric pulp testing revealed that the associated teeth were vital in nature. Two ml yellowish free flowing fluid was aspirated from the lesion which showed with few RBCs in a background of Neutrophils. Incisional biopsy was performed, and the specimen was subjected to histopathological examination which revealed the proliferating epithelial cells in the form of whorls, sheets, solid nodules, strands, cords, rossettes and duct like structures. Many foci of hyaline droplets were seen within the epithelial nodules. Areas of rounded and irregular calcifications were also seen within the connective tissue capsule and periphery of epithelial nodules. The lesion was diagnosed as extra follicular adenomatoid odontogenic tumor correlating with the clinical, radiographic and histopathological features (Fig. 3). The lesion was surgically excised along with extraction of the associated teeth (Fig. 4). During postoperative follow up, healing was uneventful, and patient revealed no untoward complications.

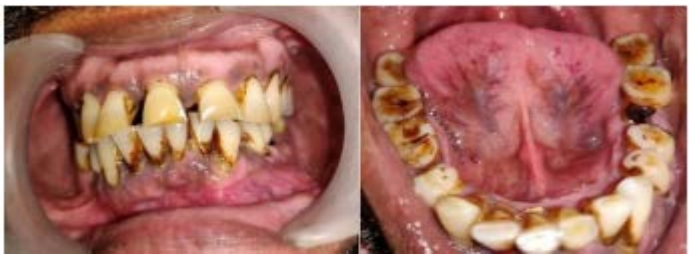
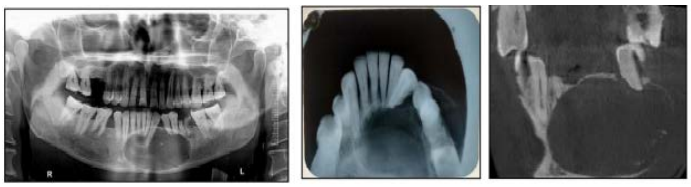


Fig. 1: Showing intraoral swelling in left mandibular canine-premolar region with buccal and lingual expansion.



2a 2b 2c

Fig. 2a: panoramic radiograph showing unilocular radiolucent lesion with corticated borders.

Fig. 2b: mandibular topographic occlusal radiograph showing buccal and lingual expansion.

Fig. 2c: Cone Beam Computed Tomography (CBCT) showing minute calcifications within the radiolucency

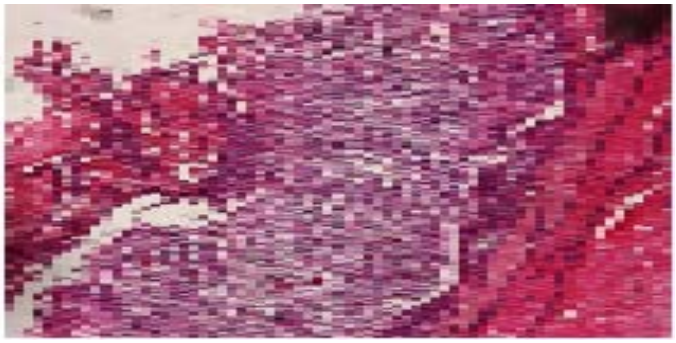


Fig. 3: Photomicrograph (H&E) 10X showing rosette & duct like

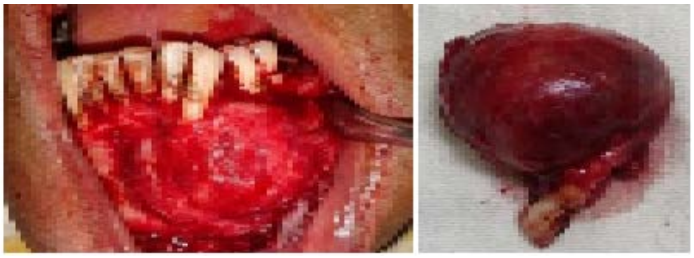
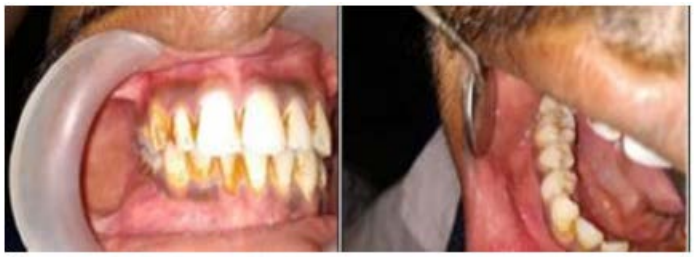


Fig. 4: Intra-operative picture and surgical specimen

Case Report - 2

Eighteen-year-old male reported to the dental hospital with a complaint of swelling on right side of mandible since a year. History revealed that the swelling was gradually increasing in size. There was no pain associated with swelling. Medical, personal and family history was non-contributory. On clinical examination, mild diffuse swelling was present, approximately 2x2cm inferior to right commissure of mouth. It was firm to hard in consistency, non-tender on palpation and overlying skin appeared normal. Intraorally, bony hard swelling was present on the buccal aspect extending from 43 to 45 obliterating the labiobuccal sulcus with the approximate size of 1.5 x 1.5 cm. On palpation, the swelling was nontender with mild expansion of labial and lingual cortical plates were evident. Over-retained deciduous 1st molar was seen on right side and missing 44. So, a provisional diagnosis of odontogenic cyst/ tumor was considered (Fig. 5)

Panoramic and mandibular occlusal radiograph were taken. The radiographs revealed an unilocularradiolucency with impacted 44 displaced towards the inferior border of mandible with expansion of buccaland lingual cortical plate. Root resorption with 43, 45 and over-retained 84 was also evident. Hence radiographic differential diagnosis of dentigerous cyst, Unicystic ameloblastoma, Odontogenic keratocyst, adenomatoid odontogenic tumor, Calcifying odontogenic cyst were taken into consideration (Fig. 6a & 6b). On histopathological examination odontogenic epithelium was cuboidal with hyperchromatic nuclei and 2-4 cell layer thick showing variable degree of intraluminal proliferation in the form of small and large nodules and duct-like structures. Nodules were composed of densely arranged spindle shaped cells forming whorls and rosettes. Few areas of dystrophic calcifications are also present within the sheets of epithelium. All these features were suggestive of adenomatoid odontogenic tumor (fig 7). The lesion was surgically excised with extraction of impacted tooth (fig 8). Healing was uneventful during the follow-up period.



6a 6b

Fig. 5: Intraoral picture showing swelling from 43 to 45 with obliteration of labiobuccal vestibule.

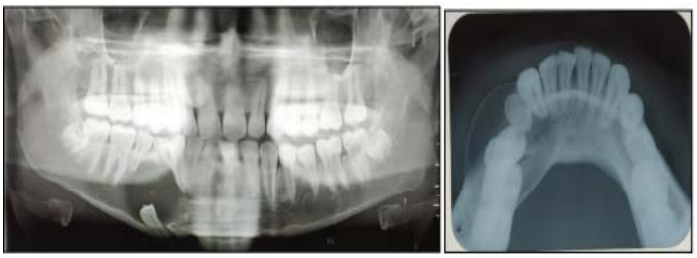


Fig. 6a: Panoramic radiograph showing radiolucent lesion with impacted 44 Fig. 6b: Mandibular occlusal radiograph showing buccal and lingual calcification

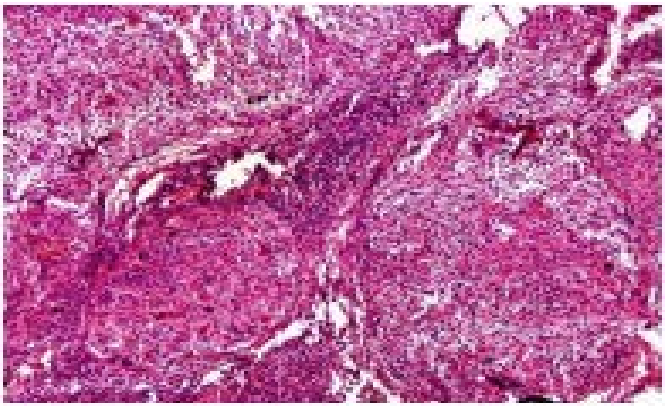


Fig 7: Photomicrograph (H&E) 10X

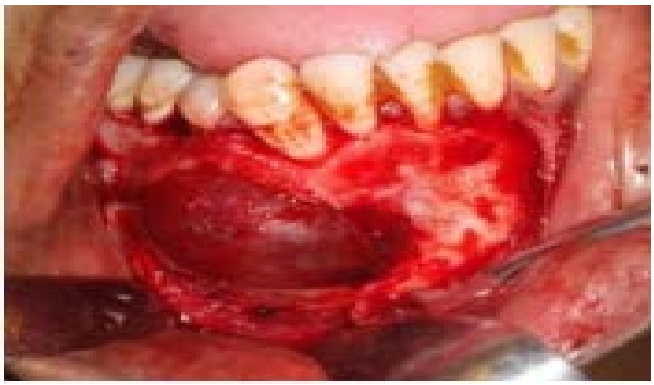


Fig 8: Intraoperative picture

Discussion

Adenomatoid odontogenic tumor (AOT) is an uncommon tumor accounting for 3 to 7% among all odontogenic tumours.2 Many authors consider it as benign tumor, while others have categorized it as a hamartomatous malformation due to the limited size and lack of recurrence in most cases.2 According to WHO "Histological typing of odontogenic tumors" AOT has been histologically defined as a tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumour may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm. The worldwide incidence showed AOT as the fifth most prevalent lesion among the odontogenic tumours.3 Even though the age range of patients with AOT varies between 3 to 82 years, 2/3rd of the cases are diagnosed at 11-14 years of age. It has a female predilection. 4 This female predilection is even more marked in Asian population, Sri Lanka (3:2:1) and Japan (3:1).4 Its occurrence is more common in maxilla than mandible and anterior portion of jaw is involved more than the posterior jaw. Almost two-third cases are associated with an impacted canine. 5 AOT is classified as intraosseous follicular (73%), intraosseous extra follicular (24%) and extra osseous peripheral variant (3%).4, 5 The tumors are usually asymptomatic, seldom exceeding 1.5 to 3cm in diameter. However, some large tumors have been reported.4 The slow growing nature of the lesion may cause cortical plate expansion leading to a painless hard swelling, asymmetry of the face, and displacement of the teeth, as was evident in our case analysis. Also the growth is only confined to the jaw bone with no invasion of the soft tissue. Radiographically, unilocular radiolucency with a corticated border is seen. There is displacement of teeth while root resorption is rare. AOT may show multiple minute variable shaped, well-defined calcifications or radiopaque foci, which may appear like a cluster of small pebbles.7 The follicular type shows a well-defined, unilocular (round or ovoid) radiolucency associated with the crown and part of the root of an unerupted tooth thus mimicking a dentigerous or follicular cyst 4 Extra follicular type is not associated with unerupted teeth and is sub-divided into four subtypes- E1: without relation to the tooth structures neither erupted nor unerupted; E2: inter-radicular; adjacent roots diverge apically due to tumour expansion; E3: superimposed at root apex level (radicular/periapical); E4: superimposed at mid-root level. The peripheral variant appears on the labial gingiva as a gingival fibroma or epulis. Radiographically, it shows slight erosion of the alveolar bone crest. Both our cases were intraosseous type, first case, the extra follicular (E2) variant and the second case, the follicular type mimicking

dentigerous cyst. There are dentinoid materials derived from root sheath epithelium. These calcifications are best appreciated on the intraoral radiograph, as is evident in the intraoral radiograph of the extra follicular variant in the present case. Occasionally a typical target appearance may be seen, in which a radiolucent circumferential halo surrounds a dense, central radiopaque mass. Dare et al showed that intraoral periapical radiograph gives more correct interpretation of AOT than panoramic radiograph.8 M Jiang et al reported cone beam computed tomography possess better potential in diagnosing AOT as compared to the panoramic radiography.9 Mutalik VS et al10 showed as many as 20 different histological patterns of AOT that have been described in the literature; AOT with CEOT-like areas, Convulated cords or bodies mimicking invaginations, Cribriform pattern, Duct like structures, Interlacing strands of cells, Luminal proliferations into the cystic lumen, Nests-like pattern, Rosette-like arrangement, Sheets of tumor cells, Trabecular arrangement, Tubular arrangement 4 . Cystic variants forming mural lining11 , Sieve-like pattern, Solid nodules of cells12, arrangement of cells in layers, ribbon-like pattern, ring like pattern of tumor cells, whorled spheroidal masses of tumor cells 13, peripheral strand of smaller cells which form net like proliferations 14 and cystic variant of AOT 15. Our cases revealed, the proliferating epithelial cells in the form of whorls, sheets, solid nodules, strands, cords, rossettes and duct like structures. Areas of rounded and irregular dystrophic calcifications are also seen within the connective tissue capsule and periphery of epithelial nodules. AOT should primarily be differentiated from calcified odontogenic tumors, as it may contain calcified foci, as was the extrafollicular case in the present study, and from cysts with radiological similarity. In addition, ameloblastoma, ameloblastic fibroma, and ameloblastic fibro-odontoma are lesions that should be considered in the differential diagnosis Considering the non-aggressive and encapsulated nature of the lesion, conservative surgical excision is the treatment of choice for AOT.3 In the present cases, local surgical excision along with extraction of associated teeth were done. Recurrence of AOT is rare and prognosis is good.

Interestingly, our present cases had some unusual clinical and radiographical features that distinguished it from normal types of AOT. Firstly, AOT are usually seen in young age and in females while male patients reported in both the cases. More common in maxilla, both the cases are seen in mandible. Secondly, radio graphically root resorption is seldom seen with AOT while in both our cases root resorption was seen. There are many few studies that showed root resorption.

Conclusion

The present study described two different varieties of AOT with respect to localization of the lesion in mandible and unusual findings. These variations in findings should be considered while differentiating it from other lesions of the jaw.

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