Adenomatoid Odontogenic Tumor of Mandible: A Rare Case Report
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Case Report

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Abstract: Adenomatoid Odontogenic Tumor is a rare tumor constituting 3-7% of all odontogenic tumors. It is a benign neoplasm which arises from the odontogenic epithelium. We present a case of 22 year female patient with a well defined, unilocular radiolucent lesion containing an impacted tooth seen radiographically. On microscopic examination, sections comprise of cyst wall composed of fibrous connective tissue with areas of haemorrhage. Whorls and lobules of spindle shaped cells were seen on luminal aspect with interspersed duct like structures. Based on the above histopathological features final diagnosis of Adenomatoid Odontogenic Tumor was made. The treatment of choice is conservative surgical removal through simple curettage or enucleation. Recurrence is very rarely seen.

Keywords: Odontogenic, radiolucent, adenomatoid.

INTRODUCTION
Philipsen et al., [1] proposed the term “Adenomatoid Odontogenic Tumor” (AOT). It is a benign neoplasm which arises from the odontogenic epithelium accounting for 3-7% of all odontogenic tumors [2]. The tumor is common in younger age group between 10-30 years and around two-thirds of all cases are reported when patient is between 10-19 years of age [2, 3, 4]. The incidence is uncommon in patients older than 30 years. It commonly occurs in the anterior portion of the jaw (95%) and is found twice as often in the maxilla (65%) than in the mandible. Females are affected about twice as often as males [2, 3, 5].

AOT has three clinical presentations: intra-osseous (associated with impacted teeth and also known as “follicular”; 70% of cases); intra-osseous (present between erupted teeth and also known as “extra-follicular”; 25% of cases); and extra-osseous (on the gingiva, also known as “peripheral”; 5% of cases) [5]. The treatment of choice is conservative surgical removal through simple curettage or enucleation [6, 7]. Recurrence is very rarely seen [6-9].

CASE REPORT
A 22 year female patient presented to the Department of Dentistry of our institute with chief complaint of pain in left lower jaw anterior region for past 5-6 months. On examination, intraorally a swelling was palpated in the region of lower left incisor. The swelling was firm in consistency and non-tender. Radiographically, a well defined, unilocular radiolucent lesion containing an impacted tooth was seen. Any internal septation or sclerotic area was not seen (Figure-1). The provisional diagnosis of dentigerous cyst was made. The lesion was surgically removed and sent for histopathological examination. The specimen received was grossly, globular in shape, firm in consistency with an embedded tooth. Cut section showed a cystic cavity containing tooth with papillary growth like structures lining the cavity (Figure-2). On microscopic examination, sections comprised of cyst wall composed of fibrous connective tissue with areas of haemorrhage. Whorls and lobules of spindle shaped cells were seen on luminal aspect with interspersed duct like structures (Figure-3a &b). Based on the above histopathological features final diagnosis of Adenomatoid Odontogenic Tumor was made.
Fig-1: Panoramic radiograph showing impacted permanent mandibular right canine in a well-defined radiolucency, expansion and thinning of the inferior border of the mandible, displacement of the anterior teeth

Fig-2: Gross appearance of grey brown to tan cystic tumor mass with embedded root of canine teeth

Fig-3: Adenomatoid odontogenic tumor. (a) Low-power view demonstrating a capsule surrounding the tumor. (20X, H&E) (b) Higher magnification showing the duct-like epithelial structures. (100X, H&E)

DISCUSSION

The differential diagnosis of jaw swelling in a young patient include both non neoplastic and neoplastic causes. The common non neoplastic causes are an apical cyst, dentigerous cyst, calcifying odontogenic cyst, odontogenic keratocyst, and central giant cell granuloma [2, 10, 11], whereas common neoplastic causes are an adenomatoid odontogenic tumor, unicystic ameloblastoma, calcifying epithelial odontogenic tumor (CEOT), ameloblastic fibroma, and ameloblastic fibroodontoma [2, 11, 12].

Adenomatoid odontogenic tumor (AOT) is an odontogenic epithelial tumor commonly affecting young female patients between 10 and 19 years of age. The maxilla being the more common site compared to the mandible. The size of the lesion ranges from 2 to 7cm with a slow growing pattern which results in a painless expansion of the jaws. The canine is the most common tooth which is impacted in AOT [3, 10, 13]. In this case, a 22-year-old female presented with a well-demarcated radiolucent lesion of 2.5cm diameter in the anterior mandibular region associated with an impacted permanent canine. The tumor progressively enlarged over a period of 5-6 months.

Since the lesion was well-demarcated and radiolucent and associated with the crown of impacted teeth like in this case, possibility of apical cyst, calcifying odontogenic cyst, odontogenic keratocyst, and central giant cell granuloma was ruled out. Further an apical cyst is usually associated with an endodontic-treated or mortified pulptooth. Calcifying odontogenic cyst, odontogenic keratocyst, and giant cell granuloma were ruled out because usually they are not related to the crown of an impacted tooth, and they are mostly multilocular. Dentigerous cyst, unicystic ameloblastoma, ameloblastoma, and ameloblastic fibroma are most frequent in the posterior region of the mandible and are associated in most cases with the third molar. However, adenomatoid odontogenic tumor
occurs mostly in the anterior maxillary region and is associated in 60% of cases with a canine [14, 15].

Mostly, AOTs are the central follicular type and appear as well-demarcated radiolucent lesions. AOT usually surrounds an unerupted tooth, and it looks as a corticated radiolucency with small radiopacities, but there are cases where the lesion has no radiopaque component, and in such cases, a dentigerous cyst is the preferred differential diagnosis. However, an AOT often appears to envelop the crown as well, when we divide the specimen to show the relation of the lesion to the tooth, unlike the dentigerous cyst which does not surround the roots [8, 13, 15]. In this case, the lesion surrounded the entire crown and overlapped the root of the lateral incisor. Few cases of AOT, like this case, were described in the literature in association with a mandibular canine.

The histological typing of the WHO defined the AOT “like a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor is well encapsulated and shows an identical benign behaviour [7, 13].” The treatment of choice was a surgical management of this tumor. Enucleation of tumor along with the associated impacted tooth and simple curettage is the treatment of choice. Conservative treatment is adequate because the tumor is not locally invasive, is well encapsulated, and can be easily separated from the bone [3].

CONCLUSION
AOTs are usually asymptomatic lesions that sometimes may cause cortical expansion and displacement of the adjacent teeth. Early diagnosis of and early enucleation prevents an excessive destruction of bone and it should be differentiated from close differential diagnosis of dentigerous cyst.

REFERENCES