Peripheral adenomatoid odontogenic tumor: report of a rare case

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ABSTRACT

Adenomatoid odontogenic tumor is a benign odontogenic tumor with slow growth potential and exceptionally low recurrence rate. The tumor is predominantly found in females in the second decade of life, involving the maxilla more frequently than the mandible. The tumor presents in three variants—trafollicular, extrafollicular, and peripheral. The peripheral variant is very rare and only few reports have been published. We present a rare case of peripheral adenomatoid odontogenic tumor in a 10-year-old male child in the anterior region of the mandible.

Keywords: Adenomatoid odontogenic tumor, benign, peripheral.

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a slow growing tumor of odontogenic origin which has been known by numerous names viz. adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adenomatoid, or teratomatous odontoma. Adenomatoid odontogenic tumor was first described by Steensland in 1905.¹ The term AOT was introduced by Philipsen and Brin, 1969, and adopted by World Health Organization (WHO) in their 'Histologic typing of Odontogenic Tumor, Jaw cysts, and Allied lesions', and it is now the generally accepted nomenclature.²

The lesion commonly affects the maxillary jaw of females in the second decade of life. Radiologically, the lesion frequently appears as a well-circumscribed, pear-shaped radiolucency (stem portion toward the alveolar surface) lying between divergent roots of the canine and the first premolar teeth (or between incisors or premolars); or as a unilocular, radiolucent lesion that is well-circumscribed or bounded by a radiopaque margin, lateral to or containing an impacted tooth (similar to a follicular or dentigerous cyst).³ Histologically, the tumor is usually surrounded by a well-developed connective tissue capsule and may present as a solid mass or as numerous small cystic spaces.

Of all the reported cases, 45.5% have been published from Asian countries, majority from Japan, Malaysia, and China. Asian AOT cases show a male: female ratio of 1–2.3 vs 1–1.4 for non-Asian cases.⁴

There are three clinicopathologic variants of AOT namely intrafollicular, extrafollicular, and peripheral, all with identical histologies.² The peripheral type is the rarest of them all constituting only 3% of all AOTs. We describe a rare case of peripheral AOT in a 10-year-old male child with unusual location in the anterior region of the lower jaw.

CASE REPORT

A 10-year-old male patient reported to the Department of Pediatric and Preventive Dentistry with a chief complaint of slow-growing, painless swelling of 3 months duration in the lower anterior region. On general examination, the patient was apparently healthy. On extra-oral examination, no notable findings were recorded. Intra-oral examination revealed the presence of a soft, pedunculated, firm gingival swelling in the mandibular anterior region covering two-third portion of crowns of permanent mandibular left central incisor (31), permanent mandibular right central incisor (41), and lateral incisor (42), extending into the labial vestibule (Figure 1A). Intra-oral periapical radiographs and orthopantomogram demonstrated well-circumscribed unicystic radiolucency with sclerotic border in relation to 41 and 42 (Figure 1B and C). The differential diagnosis of gingival epulis, dentigerous cyst, calcifying odontogenic cyst, calcifying epithelial tumor, odontogenic keratocyst, and unicystic

Figure 1 (A) Pre-operative clinical appearance. (B and C) Radiographs showing well-defined radiolucency in relation to 31, 41, and 42. (D) Intra-operative view after complete enucleation. (E) The excised tissue.

ameloblastoma was made and excisional biopsy of the lesion was planned.

Before surgical procedure, routine blood examinations of the patient were done which were within normal limits. Local anesthesia was administered and the lesion was removed in toto. The lesion was encapsulated with intraosseous extension and measured approximately 3 cm × 1.5 cm (Figure 1D and E). The whole lesion was fixed in formalin and was sent for histopathological examination. Iodoform dressing was given to the patient and was changed periodically to promote bone formation.

The histopathological examination showed the presence of tumor tissue with epithelium forming cystic and solid spaces. The wall was lined by odontogenic epithelium showing foci of squamous differentiation as well as rosettes and ducts like tubular structures. Trabecular and cribriform patterns were seen and eosinophilic material was also evident at places (Figure 2A–C).

Based on clinico-radiological and histological findings, a confirmatory diagnosis of AOT of peripheral type was established. The patient was recalled periodically and

follow-up examination was done up to 2 years. Within this duration, the bone defect healed completely and the lesion did not recur; however, a gingival defect was created within the region of 42 (Figure 2D and E).

DISCUSSION

Adenomatoid odontogenic tumor represents 3–7% of all odontogenic tumors and ranks fourth or fifth among the ‘top 10’ of odontogenic tumors, surpassed only by odontomas, cementomas, myxomas, and ameloblastomas.⁴ Adenomatoid odontogenic tumor affects females more frequently than males in a ratio of 1.9:1.² This ratio may be even higher as reported in Asian populations, the highest incidence being observed in Sri Lanka, 3.2:1⁵ and Japan, 3:1.⁶ The lesion appears most often in the second decade of life. The tumor is twice more commonly located in the maxilla than in the mandible. Case reports have been published with the tumors present in uncommon locations like anterior mandibular

Figure 2 (A) Cyst wall lined by odontogenic epithelium showing peripheral palisading (marked by blue arrow) and loose stellate reticulum (shown by red arrow H&E 40×). (B) Higher power view of the same (H&E 100×). (C) Foci of squamous differentiation and duct-like structures (marked by arrow H&E 100×). (D) Follow-up orthopantogram after 2 years. (E) Follow-up clinical photograph after 2 years.

region^{7–9} and mandibular posterior region.¹⁰ It is sometimes referred to as ‘two-thirds tumor’ because it occurs in the maxilla in about two-third cases, about two-third cases arise in young females, two-third cases are associated with an unerupted tooth, and two-third affected teeth are canines.

Based on clinical and radiologic findings, three variants of AOT have been described— intraosseous follicular, intraosseous extrafollicular, and peripheral. Follicular type (73%) appears as dentigerous cyst and is associated with the crown of an embedded tooth whereas extrafollicular type (24%) has no association with the crown of an embedded tooth. Peripheral variant is rare (3%) and is attached to the gingival structures. Histologically, all variants of AOT are well-encapsulated and show an identical benign behavior with similar histological features. World Health Organization defined AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degree of changes in the connective tissue. The tumor may be cystic and eosinophilic and uncalcified, amorphous material

called ‘tumor droplets’ can be found. The histological findings in our case were consistent with that reported in the literature.

Conservative surgical enucleation and curettage of the lesion is the treatment of choice because the tumor is well-encapsulated in almost all the cases, showing an identical, benign biological behavior.⁴ Recurrence of the tumor is very rare and the prognosis is excellent. In our case, the tumor was completely enucleated along with its capsule. Follow-up of the patient was done up to 2 years. During that duration, the patient remained asymptomatic and the lesion did not showing excellent prognosis.

Our case was diagnosed as a peripheral variant of AOT which is very rare and only few case reports have been published in literature. Moreover, 10:1 maxilla mandible ratio is reported in the peripheral variant. However, in this case, the tumor was present in the anterior mandible which is a very rare site for the tumor development. In our case, the tumor was attached to the buccal gingiva of 31, 41, and 42 which extended to the buccal cortical bone. The peripheral

type seems to be considerably more common in females than in males, but in our case, the tumor was reported in a male child of 10 years age. Thus, this case has some unusual characteristics of AOT like rare occurrence in the anterior mandible in a male child with an intraosseous extension.

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REFERENCES

1. Steensland HS. Epithelioma adamantinum. *J Exp Med* 1905; 6:377–89.
2. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumor: facts and figures. *Oral Oncol* 1998;35:125–31.
3. Motamedi MHK, Shafeie HA, Azizi T. Salvage of an impacted canine associated with an adenomatoid odontogenic tumour: a case report. *Br Dent J* 2005;199:89–90.
4. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu QX. Adenomatoid odontogenic tumor: biologic profile based on 499 cases. *J Oral Pathol Med* 1991;20:149–58.
5. Mendis BR, MacDonald DG. Adenomatoid odontogenic tumour: a survey of 21 cases from Sri Lanka. *Int J Oral Maxillofac Surg* 1990;19:141–3.
6. Toida M, Hyodo I, Okuda T, Tatematsu N. Adenomatoid odontogenic tumor: report of two cases and survey of 126 cases in Japan. *J Oral Maxillofac Surg* 1990;48:404–8.
7. Yilmaz N, Acikgoz A, Celebi N, Zengin AZ, Gunhan O. Extrafollicular adenomatoid odontogenic tumor of the mandible: report of a case. *Eur J Dent* 2009;3:71–4.
8. Bhullar RPK, Brar RS, Sandhu SV, Bansal H, Bhandari R. Mandibular adenomatoid odontogenic tumor: a report of an unusual case. *Contemp Clin Dent* 2011;2:230–3.
9. Anegundi RT, Radhika R, Patil S, Sahana BA. Adenomatoid odontogenic tumor: an uncommon location. *Pediatr Dent* 2011; 33:437–9.
10. Marx RE, Stern D. In: *Oral and Maxillofacial Pathology: a Rationale for Diagnosis and Treatment*. Hanover Park: Quintessence Publishing 2003:609–12.