Peripheral Adenomatoid Odontogenic Tumor Masquerading as Gingival Swelling

Deepty Bansal1, Mala Kamboj2, Anjali Narwal3, Anju Devi4, Deepak Pandiar5

ABSTRACT

The adenomatoid odontogenic tumor (AOT) is an uncommon benign, non-invasive tumor also known as “two-thirds tumor,” which is either intra or extraosseous. The extraosseous AOT is also referred to as peripheral, gingival or soft tissue odontogenic tumor. Gingiva is a common site for reactive lesions, benign neoplasms and distant metastasis. Peripheral adenomatoid odontogenic tumor (PAOT) is a rare entity with an incidence of only 3% of all reported AOTs. It is primarily described as a slow-growing gingival swelling with minimum or no bone involvement. It presents highest predilection for maxillary gingiva of anterior teeth and predominantly affects children. Till date 22 PAOT’s have been reported worldwide and their behavior is similar to intraosseous AOTs. Thus, the clinician should be aware while treating them. The present case describes an additional case of PAOT on the maxillary anterior gingival region of a 15-year-old female.

Keywords: Adenomatoid odontogenic tumor, Gingival swelling, Odontogenic tumor, Peripheral.

AOTs called as "master of disguise" and peripheral ones commonly resemble gingival lesions. Gingiva is a common site for reactive and neoplastic conditions. So, care must be taken for its proper diagnosis, timely treatment and follow-up.

Journal of South Asian Association of Pediatric Dentistry (2021): 10.5005/jp-journals-10077-3091

INTRODUCTION

Adenomatoid odontogenic tumor (AOT), is infrequent (2.2% to 7.1% of all odontogenic tumors) and distinct benign (hamartomatous), slowly progressive lesion that was first described by Steensland in 1905 as "Epithelioma adamantinum". The synonyms used for AOT include ameloblastic adenomatoid tumor, adenomatoid, adenomameloblastoma, and adenoamameloblastic odontoma or pseudoamenomatous ameloblastoma. Philipsen and Birn introduced the term "Adenomatoid odontogenic tumor" in 1969 and World Health Organization in 1971 adopted Adenomatoid odontogenic tumor in classification of odontogenic tumors. Philipsen et al. described three clinic-topographic variants of AOT namely, central (intraosseous) variant which includes follicular (73%), extrafollicular type (24%) and peripheral (extraosseous/gingival) variant (PAOT). The peripheral type of AOT is rarest type comprising of 3% of total cases of AOT. Extensive literature search of indexed journals since 1958 for this pathology till date revealed only 23 cases including present case with a mean age of 13 years and ranged from 4 to 27 years. This fascinating entity camouflage as a solitary gingival swelling like fibrous epulis, since it is mostly located in the gingival mucosa. It is a cause of concern as the biological behavior of extraosseous variant is similar to that of its intraosseous counterpart.

Keywords: Adenomatoid odontogenic tumor, Gingival swelling, Odontogenic tumor, Peripheral. How to cite this article: Bansal D, Kamboj M, Narwal A, et al. Peripheral Adenomatoid Odontogenic Tumor Masquerading as Gingival Swelling. J South Asian Assoc Pediatr Dent 2021;4(3):200–202.

CASE DESCRIPTION

A 15-year-old female with the chief complaint of an asymptomatic swelling in anterior maxillary gingiva since past two months reported to the outpatient department. The swelling approximated 2 × 1 × 0.5 cm in dimension (Fig. 1A) and on palpation the consistency was soft to firm, non-reducible, non-compressible as well as non-tender. Overlying mucosa appeared normal in Color and texture. No bone loss was revealed in Intraoral periapical radiograph of left anterior maxilla (Fig. 1B). On the basis of clinico-radiological findings, a provisional diagnosis of peripheral ossifying fibroma was made alongwith fibrous epulis as differential diagnosis. The routine blood examination was carried out and the lesion was surgically excised completely under local anesthesia. Histopathological examination revealed the presence of parakeratinized stratified squamous epithelium with basal hamartia like proliferations (Fig. 2A) at areas into the connective tissue stroma. The underlying stroma disclosed an unencapsulated neoplasm of odontogenic origin exhibiting columnar to cuboidal cells with hyperchromatic nuclei arranged in a variegated pattern varying from convoluted double row, rosettes, duct-like strands (Fig. 2B) and multinodular structures. Abundant basophilic calcified areas were noted with few eosinophilic amorphous material deposits themed as tumor or hyaline droplets (Fig. 2C). Areas of intramembranous ossification and dentinoid-like material (Fig. 2D) were also seen in the vicinity.
Adenomatoid Odontogenic Tumor as Gingival Swelling

Involvement of maxilla, 2/3rd AOTs are associated with impacted teeth and 2/3rd of them are related to cuspids. Clinically, PAOT presents as asymptomatic, slowly progressive swelling in gingiva. Analogous to its central counterpart it shows female predilection (Female: Male - 2:1). The peak incidence of these tumors is reported during second and third decade of life. The present case too was reported in 15-year-old female as a firm to hard, well defined, oval swelling in the left maxillary incisor region.

**DISCUSSION**

AOTs are rare benign, non-invasive, epithelial tumors of odontogenic origin exhibiting slow growth. This tumor is also termed as "two-thirds tumor", as 2/3rd cases reported in females, 2/3rd involvement of maxilla, 2/3rd AOTs are associated with impacted teeth and 2/3rd of them are related to cuspids. Clinically, PAOT presents as asymptomatic, slowly progressive swelling in gingiva. Analogous to its central counterpart it shows female predilection (Female: Male - 2:1). The peak incidence of these tumors is reported during second and third decade of life. The present case too was reported in 15-year-old female as a firm to hard, well defined, oval swelling in the left maxillary incisor region.

PAOT most commonly presents in labial anterior mucosa, a usual site for other reactive and odontogenic lesions of gingiva.

Figs 1A and B: (A): Clinical picture of the girl shows the intraoral swelling present on left side of anterior maxilla (arrow). (B) Intraoral periapical radiograph of maxillary left central and lateral incisors shows normal bone.

Figs 2A to D: (A): Photomicrograph reveals overlying epithelium (white arrow) with basal hamartia (black arrow) like proliferation into connective tissue (20X, H/E). (B): Columnar to cuboidal cells arranged in convoluted double row and duct-like strands (20X, H/E, arrow). (C): Abundant basophilic calcified areas along with few eosinophilic hyaline droplets (20X, H/E, arrow). (D) Areas of intramembranous ossification and dentinoid-like material (20X, H/E, arrow).
The prevalence of reactive lesions involving the gingiva is 47.61% for inflammatory gingival hyperplasia, 27.16% pyogenic granuloma, 15.34% fibrous hyperplasia, 1.66% peripheral giant cell granuloma (PGCG) and 8.19% peripheral ossifying fibroma (POF). The incidence of peripheral odontogenic tumors reported in literature is, 40% for peripheral odontogenic fibroma and peripheral calcifying cystic odontogenic tumor (PCCOT), 6% peripheral calcifying epithelial odontogenic tumor (PCEOT) and peripheral ameloblastic fibroma (PAF) and mere 3% is PAOT. Radiographic picture of PAOT varies from minimal to no loss of cortex alveolar bone, similar to reactive and peripheral odontogenic lesions. There were no pathognomonic radiographic findings related to the present lesion too. Hence, clinic-radiographically PAOT shows features similar to reactive gingival lesions. Due to its uncommon occurrence and clinical resemblance with reactive lesions, often clinician does not consider it in differential diagnosis of epulis like gingival growths.

The common gingival lesions most often confused with PAOT are other peripheral odontogenic tumors, POF and PGPG. The most frequently reported peripheral odontogenic tumor is peripheral odontogenic fibroma and clinically it may present as sessile or pedunculated, red or pink along with an intact or ulcerated surface mucosa. On palpation it is normally firm and non-tender. Histopathologically it is comprised of dense connective tissue fibres with variable quantity of calcifications as well as odontogenic epithelial islands.

Peripheral ameloblastoma is a neoplasm of odontogenic epithelium which appears as a sessile painless exophytic thin mass with surface texture of smooth to granular, frequently involving canine–premolar area of mandible. Microscopically most common features include nest and strands of epithelial cells in fibrous matrix. The peripheral cells are ameloblast-like with central stellate reticulum like cells exhibiting microcyt formation.

Peripheral calcifying cystic odontogenic tumor clinically presents as a normal to red in Color firm painless gingival outgrowth with ulcerated surface. The most frequent site for this lesion is anterior and premolar region of mandible. Histologically it is comprised of epithelial islands of polyhedral tumor cells which shows prominent intercellular desmosome junctions and pleomorphic enlarged nuclei in the fibrous connective tissue stroma.

POF is more commonly reported in 5 to 25 years of age with the peak incidence at 13 years with equal predilection for maxilla and mandible. Clinically it appears as a well-demarcated focal gingival tissue mass either sessile or pedunculated, coral pink or red with intact or surface ulceration. Histologically overlying epithelium could be intact or ulcerated with the stroma displaying calcification in the form of osteoid tissue, compact or cancellous bony trabeculae along with plump, proliferating fibroblasts.

PGCG is a reactive gingival growth pedunculated or sessile, haemorrhagic in appearance with frequent surface ulceration. Histologically it consists of unencapsulated tissue mass with delicate reticular and fibrillar connective tissue stroma, abundant fibroblasts and multinucleated giant cells. Numerous blood capillaries are evident at periphery of the lesion. Foci of hemorrhage, with liberation of hemosiderin pigment are characteristic features.

Based on the clinical and radiographic findings of 23 peripheral AOTs reported so far, most of them are considered as reactive lesions of gingiva and were completely excised under local anesthesia. Generally, PAOTs are indolent and the most recommended treatment is complete surgical excision. Their prognosis is usually excellent and their recurrence rate is very low (0.2%). Similar approach was practiced in our case and after a regular follow up of two years, patient has shown no signs of recurrence.

Recent studies have unveiled the immunohistochemical reactivity of AOT for CK5, CK17, CK19 and ki67. The minimal expression of ki67 by AOT, correlates the low cellular proliferation and decreased tendency to recur.

AOTs by themselves are rare odontogenic tumors and within its classification extraosseous tumors (PAOT) are rarer. This tumor is truly called as “master of disguise” as it resembles other frequently occurring gingival lesions of oral mucosa. It is very habitual to excise harmless epulis-like growths before any incisional biopsy which may later prove to be a neoplasm on histopathological diagnosis. The final surgical excision should be done post histopathological report rather than employing overzealous treatment protocols since peripheral odontogenic tumors are very much uncommon but do manifest as gingival lesions.

**References**

1. Steensens HS. Epithelioma adamantinum. J Exp Med 1905;6(6):377–389. DOI: 10.1084/jem.6.4.6.377
2. Stafne EC. Epithelial tumors associated with developmental cysts of the maxilla: a report of three cases. Oral Surg Oral Med Oral Pathol 1948;1(10):887–894. DOI: 10.1016/0030-4220(48)90114-5
3. Phillipsen HP, Birn H. The adenomatoid odontogenic tumor. Ameloblastic adenomatoid tumor or adeno-ameloblastoma. Acta Pathol Microbiol Immunol Scand 1969, 75(3):375. PMID: 5801660
4. Phillipsen HP, Kongkhunthiang P, Reichart PA. The adenomatoid odontogenic tumor: an update of selected issues. J Oral Pathol Med 2016;45(6):394–398. DOI: 10.1111/jop.12418
5. Reddy Kundoor VK, Maloth KN, Guguloth NN, et al. Extrafollicular adenomatoid odontogenic tumor: an unusual case presentation. J Dent (Shiraz). 2016(Dec);17(4):370-374. PMID: 27942555; PMCID: PMC5136418.
6. Ide F, Mishima K, Saito I, et al. Rare peripheral odontogenic tumors: report of 5 cases and comprehensive review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008; 106(4):22–28. DOI: 10.1016/j.tripleo.2008.05.064
7. Babu B, Hallikeri K. Reactive lesions of oral cavity: a retrospective study of 659 cases. J Indian Soc Periodontol 2017;21(4):258–263. DOI: 10.1016/j.tripleo.2008.05.064
8. Philipsen HP, Birn H, Hallikeri K. Reactive lesions of oral cavity: a retrospective study of 659 cases. J Indian Soc Periodontol 2017;21(4):258–263. DOI: 10.1016/j.tripleo.2008.05.064
9. Babu B, Hallikeri K. Reactive lesions of oral cavity: a retrospective study of 659 cases. J Indian Soc Periodontol 2017;21(4):258–263. DOI: 10.1016/j.tripleo.2008.05.064
10. Vidyasagar GV, Pahalia YK, Kushwah AP. Surgical management of peripheral variant of adenomatoid odontogenic tumor: a case report with review. J Contemp Entomol 2015;Jan;6 (1):128–130. DOI: 10.4103/0976-237X.149309.
11. Prakash SM, Vidisha G, Nagaraju K, et al. Peripheral adenomatoid odontogenic tumor: a rare case report. Ann Clin Case Rep 2017;2:1235.

**ORCID Id**

Mala Kamboj ♦ http://orcid.org/0000-0001-5626-2622