CASE REPORT

Peripheral Adenomatoid Odontogenic Tumour Masquerading as Gingival Swelling

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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.

Key messages: 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.

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Introduction

Adenomatoid odontogenic tumor (AOT), is infrequent (2.2% to 7.1% of all odontogenic tumours) and distinct benign (hamartomatous), slowly progressive lesion that was first described by Steensland in 1905 as "Epithelioma adamantinum".[1] The synonyms used for AOT include ameloblastic adenomatoid tumour, adenomatoid, adenoameloblastoma, and adenoameloblastic odontoma or pseudoadenomatous ameloblastoma.[2,3] Philipsen and Birn introduced the term "Adenomatoid odontogenic tumour" in 1969 and World Health Organization in 1971 adopted Adenomatoid odontogenic tumour in classification of odontogenic tumours. Philipsen et al. described three clinico-topographic variants of AOT namely, central (intraosseous) variant which includes follicular (73%), extrafollicular type (24%) and peripheral (extraosseous/gingival) variant (PAOT).[4] 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 camouflages 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 behaviour of extraosseous variant is similar to that of its intraosseous counterpart.[4] Gingiva is a common site for both benign and neoplastic lesions which results in incorrect diagnosis of PAOT as a simple gingival pathology and the actual nature of lesion is disclosed only after its microscopic evaluation. A case of extraosseous variant of AOT located in anterior maxillary gingiva of a 15-year-old girl mistaken clinically as gingival epulis is described.

Case History

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 colour 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 anaesthesia. 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 termed as tumour or hyaline droplets (Fig. 2C). Areas of intramembranous ossification

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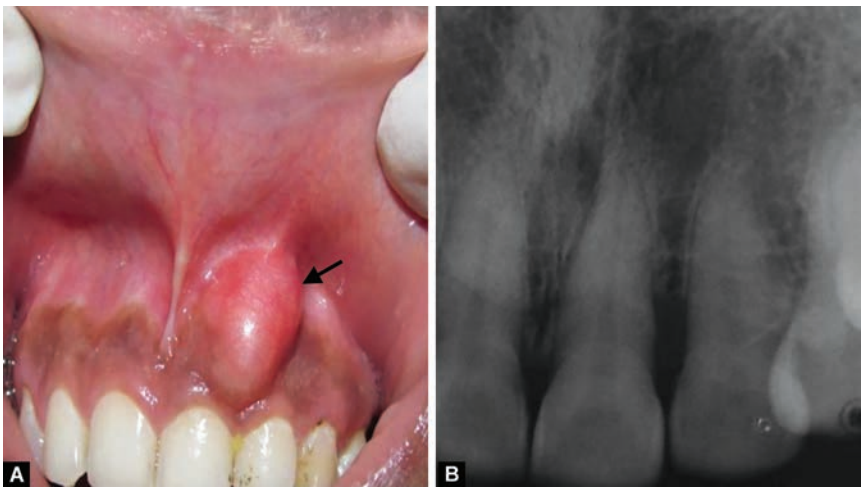

Fig. 1: (**A**): Clinical picture of the girl shows the intraoral swelling present on left side of anterior maxilla (arrow). (**B**): intraoral periapical radiograph of maxillary left central and lateral incisors shows normal bone

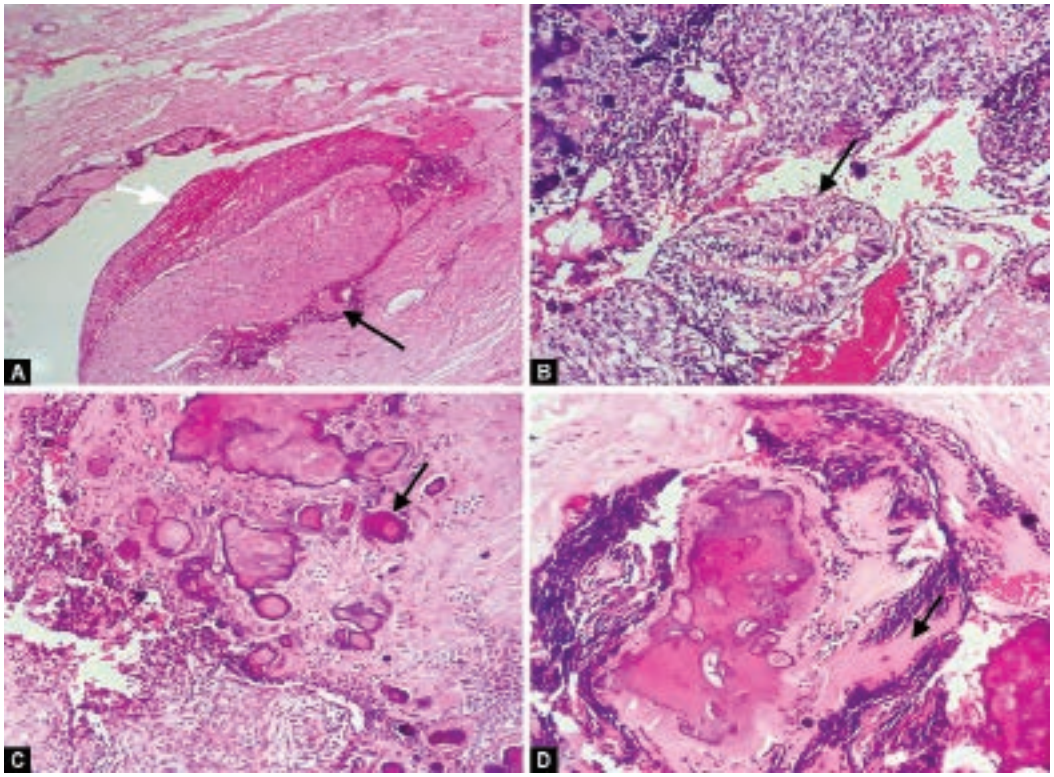

Fig. 2: (**A**): Photomicrograph reveals overlying epithelium (white arrow) with basal hamartia (black arrow) like proliferation into connective tissue (20X, H/E). (**B**): Columnar to cuboidal cells arranged in convoluted double row and duct like strands (20X, H/E, arrow). (**C**): Abundant basophilic 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)

and dentinoid like material (Fig. 2D) were also seen in the vicinity of the tumour mass. Based on the histopathological evidences a final diagnosis of peripheral adenomatoid odontogenic tumour of gingiva was made. No recurrence has been noticed even after two years of its excision.

Discussion

AOTs are rare benign, non-invasive, epithelial tumours of odontogenic origin exhibiting slow growth. This tumour is also termed as "two-thirds tumour", 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.[5] 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 tumours is reported during second and third decade of life.[6] 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. 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).[7] The incidence of peripheral odontogenic tumours reported in literature is, 40% for peripheral odontogenic fibroma and peripheral calcifying cystic odontogenic tumour (PCCOT), 6% peripheral calcifying epithelial odontogenic tumour (PCEOT) and peripheral ameloblastic fibroma (PAF) and mere 3% is PAOT.[8] Radiographic picture of PAOT varies from minimal to no loss of cortex alveolar bone, similar to reactive and peripheral odontogenic lesions.[6,8] There were no pathognomonic radiographic findings related to the present lesion too. Hence, clinico-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.[3,4]

Peripheral ameloblastoma is a neoplasm of odontogenic epithelium which appears as a sessile painless exophytic tiny 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 microcyst formation.[3,9]

Peripheral calcifying cystic odontogenic tumor clinically presents as a normal to red in colour 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 tumour cells which shows prominent intercellular desmosome junctions and pleomorphic enlarged nuclei in the fibrous connective tissue stroma.[3]

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 alongwith plump, proliferating fibroblasts.[3]

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 haemorrhage, with liberation of hemosiderin pigment are characteristic features.[6]

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 anaesthesia.

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%).[10] 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.[5,11]

AOTs by themselves are rare odontogenic tumours and within its classification extraosseous tumours (PAOT) are rarer. This tumour 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 tumours are very much uncommon but do manifest as gingival lesions.

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