CASE REPORT

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Peripheral adenomatoid odontogenic tumour

Satish R Balwani, SR Barpande, JV Tupkari

Department of Oral Pathology and Microbiology, Government Dental College and Hospital, GHATI, Aurangabad - 431 001, Maharashtra, India

Correspondence Address:
S R Barpande
Department of Oral Pathology and Microbiology, Government Dental College and Hospital, GHATI, Aurangabad - 431 001, Maharashtra
India
Login to access the email ID

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Abstract

The adenomatoid odontogenic tumour (AOT) is a benign (hamartomatous), non-invasive lesion with a slow but progressive growth. It occurs in intraosseous as well as in peripheral forms. The peripheral variant is still by far the most rarely reported type constituting only 4.4% of all AOT cases. This article presents the clinical, radiological and microscopic features of a peripheral AOT occurring in the soft tissue overlying the tooth-bearing areas of the anterior maxilla in a 19-year-old female patient.

Keywords: Adenomatoid odontogenic tumour, gingival lesions, peripheral odontogenic tumours

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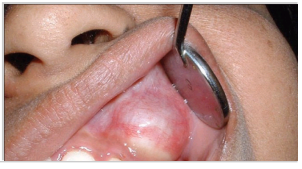
Figures and Tables



Introduction

The adenomatoid odontogenic tumour (AOT) is a benign (hamartomatous), non-invasive lesion with a slow but progressive growth. It occurs in intraosseous as well as extraosseous forms. AOT accounts for 2.2% to 7.1% of all odontogenic tumours. The central AOT accounts for approximately 96% of all AOTs of which 71% are of the follicular type. [1]

Peripheral odontogenic tumours (POTs) are tumours that demonstrate the histologic characteristics of their intraosseous counterparts but occur solely in the soft tissue covering the tooth-bearing portion of the mandible and maxilla. These lesions are also known as extraosseous odontogenic tumours, soft tissue odontogenic tumours or odontogenic tumours of the gingiva. [2],[3],[4]

POTs are rare with mostly single case reports or a small series of cases reports reported in the literature. Odontogenic tumours, described as originating in the gingiva, include ameloblastoma, calcifying epithelial odontogenic tumour, squamous odontogenic tumour, calcifying cystic odontogenic tumour (calicifying odontogenic cyst), AOT, ameloblastic fibroma, odontoma, odontogenic fibroma and odontogenic myxoma, some of which are exceedingly rare. [3]

In this article, we present the clinical, radiological and microscopic characteristics of a peripheral AOT occurring in the soft tissue overlying the tooth bearing areas of the anterior maxilla in a 19-year-old female patient.

The AOT was first described by Stafne in 1948 as an 'epithelial tumour associated with developmental cysts of the maxilla'. He reported three cases, occurring in the anterior mandible, two related to impacted teeth (follicular AOT) and a third lesion unassociated with an unerupted tooth (extra-follicular AOT). [5] A variety of terms have been used to describe this lesion including cystic complex composite odontome, adenoameloblastoma and ameloblastic adenomatoid tumour amongst many others, of which the adeno-ameloblastma was in common use for many years since the tumour was considered a histological variant of the ameloblastoma. [1],[5]

In 1967, the term odontogenic adenomatoid tumour was suggested by Abrams *et al*. In 1969, Philipsen and Birn presented a review based on 76 cases of AOT, which showed the tumour to be an entity clearly distinguishable from the solid or multicystic ('classical') ameloblastoma. Philipsen and Birn introduced the term adenomatoid odontogenic tumour, to be adopted by the WHO in their "Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions" and it is now the generally accepted nomenclature. [1]

A review of the literature by Giansanti and coworkers in 1970 revealed more than 100 cases of intraosseous AOTs and a second review by Courtney and Kerr in 1975 raised the number to almost 150 cases. The occurrence of the lesions within soft tissues is rare. [6]

In 1990 Philipsen reviewed 499 cases of AOT and provided a comprehensive database on this tumour. The AOT has been divided into peripheral (extraosseous) and central (intraosseous) groups. The central lesion is further divided into follicular (related to an unerupted tooth) and extra-follicular (not related to an unerupted tooth). [5],[7] In their series the central variant was the most common type, accounting for 97.2% of all lesions, 73% of which were follicular. [5] Philipsen and Reichart (1998) surveyed the literature of AOTs and identified 412 cases in which 394 (95.6%) were central and 18 (4.4%) peripheral. [3]

Some authorities feel that, having the slow growth and circumscribed growth pattern of the lesion, it is best

classified as a hamartoma rather than a true neoplasm. Although there is evidence that the tumour cells are derived from enamel organ epithelium, investigators have also suggested that the lesion arises from remnants of dental lamina. [8]

In regard to the histogenesis of the peripheral AOT, Abrams and co-workers (1968) suggested that their three cases were associated with unerupted teeth. As normal eruption progressed, they speculated, the tumour was pushed peripherally and to the side and only those portions that were not destroyed by the erupting tooth remained as a residual AOT. [6] Yazdi and Nowparast (1974), on the other hand, suggested that the basal cell layer of the surface epithelium is the source of origin of the peripheral AOT. They also suggested that the peripheral variant is rarer because some lesions undergo atrophy after eruption of the tooth. This is unlikely because in most cases, the appearance of the lesion generally is several years post-eruption. [9]

Case Report

A 19-year-old female patient reported with the complaint of a painless swelling in the anterior region of upper jaw on the left side since four to five months. The swelling was initially soft and gradually it became firm to hard in consistency. Extraorally, small diffuse swelling was seen around the ala of the nose of left side.

Intraoral examination revealed the presence of firm to hard well-defined, roughly oval swelling in 22, 23 and 24 region. It measured approximately 3 cm × 2.5 cm × 1.5 cm. The swelling extended superoinferiorly from 0.5 cm away from gingival margin of 22, 23 and 24 to upper labial vestibule and anteroposteriorly from the mesial of 22 to distal of 24. It was of same color as that of adjacent mucosa. The surface was smooth [Figure - 1].

Intraoral periapical radiograph (IOPA) revealed faint circumscribed radiolucency with radiopaque outline in 22, 23 and 24 region with no bone involvement [Figure - 2].

Hematoxylin and eosin stained sections of the excised mass showed the presence of cystic cavity along with fibrous connective tissue wall. The proliferation of tumour cells was seen in the cystic cavity [Figure - 3]. Tumour cells were polyhedral to spindle in shape and arranged in varying patterns from solid islands, nests to thin anastomosing strands (plexiform pattern) [Figure - 3],[Figure - 4],[Figure - 5]. Duct-like structures with lumina of varying size with some basophilic non-homogeneous material were present. These duct-like structures were lined by a single layer of ameloblast-like cells with an eosinophilic rim inside [Figure - 6]. Eosinophilic material was seen in between the thin anastomosing strands of tumour cells as well as in the duct-like structures [Figure - 4],[Figure - 5]. Calcification was also present within the tumour parenchyma [Figure - 7].

Based on the clinical, radiological and histopathological findings, the case was diagnosed as a "peripheral adenomatoid odontogenic tumour".

Discussion

Peripheral odontogenic lesions are considered rare within the odontogenic tumour classification. The clinical differential diagnoses of these lesions infrequently include a peripheral odontogenic tumour, but usually favor gingival fibroma, pyogenic granuloma, peripheral giant cell granuloma or other reactive hyperplasia. For the most part, it is not until the tumour has been excised and examined histologically that

the true nature of the lesion is known. [10]

The peripheral AOT is still by far the most rare type constituting only 4.4% of all AOT cases. Of the 18 cases reported so far, the mean age at time of operation was 13 years which is 3 and 10 years earlier than the corresponding mean ages for AOT of follicular and extrafollicular types, respectively. The distribution by sex (female: male=14: 1) makes this type of AOT quite unique. [1] Peripheral lesions present as a gingival-colored mass that ranges from 1 to 1.5 cm in diameter (the size was listed in only 4 of 18 reported cases). They are 10 times more prevalent in the maxillary gingiva than in the mandibular gingiva; all but 3 of the 18 reported cases were located adjacent to an incisor-usually the maxillary central incisor. [11] Two cases found in infants have recently been reported. [1]

The present case was reported in a 19-year-old-female patient. Firm to hard, well-defined, roughly oval swelling was present in 22, 23 and 24 region. It measured approximately 3 cm × 2.5 cm × 1.5 cm. It was of same color as that of adjacent mucosa. The surface was smooth [Figure - 1]. These findings were in accordance with previous case reports. [5],[9]

Peripheral lesions rarely are detectable radiographically but there may be slight erosion of the underlying alveolar bone cortex. One reported case demonstrated central and peripheral involvement; it could not be determined whether the bilobed lesion was primarily a gingival lesion that had eroded into the underlying alveolar bone or if a superficial, primarily intraosseous lesion had expanded out into the overlying gingiva. [11] In the present case, IOPA radiograph revealed faint circumscribed radiolucency with radiopaque outline in 22, 23 and 24 region with no bone involvement [Figure - 2].

AOTs macroscopically appear as a soft, roughly spherical mass with a distinct fibrous capsule. [12] Upon gross sectioning, the cut surface shows a variegated appearance with small areas of hemorrhage in a grayish-white tissue. Small or large cystic spaces may be present and these may contain yellowish gelatinous material or blood-stained fluid. In some cases, the tumour may be almost entirely cystic. A tooth or teeth may be embedded in the tumour or attached to it. [13]

There is little variation in the histopathology of a peripheral AOT compared with an intraosseous AOT-proliferation of odontogenic epithelium with variable-sized duct-like structures and solid nodules of cuboidal or columnar cells that are encapsulated by thin connective tissue, with a vascular stroma and diffuse calcifications- although there is some evidence to suggest that extraosseous lesions are unencapsulated more frequently. There is no evidence to suggest that these peripheral neoplasms arise from surface epithelium. [10] In the present case, the tumour was well encapsulated [Figure - 3].

The treatment recommended for all AOTs is enucleation or excision. [5] As reported in the literature, [10] in the present case, the lesional tissue was also easily separated from the overlying mucosa. There have been no reports of recurrences, with the exception of one case reported by Takigami, in a 49-year-old female patient with a number of recurrences of a maxillary AOT. [5] The very few peripheral AOT cases have been reported to draw definite conclusions on biologic behavior. [10]

A case of peripheral (extraosseous) adenomatoid odontogenic tumour in a 19-year-old female patient is reported. Though it is a rare site, it should be added to the differential diagnoses of soft tissue lesions of the gingivae. Excision of the lesion is the recommended treatment. This report adds to the available literature of 18 previously reported cases of peripheral AOT. [3]

References

1. Philipsen HP, Reichart PA (1998): Adenomatoid odontogenic tumour: Facts and figures oral Oncol, 35: 125-31.
2. Manor Y, Mardinger O, Katz J, Taicher S, Hirshberg A (2004): Peripheral odontogenic tumours-

differential diagnosis in gingival lesions, Int J Oral Maxillofac surg, 33: 268-73.
3. Buchner A, Merrell PW, Carpenter WM (2006): Relative frequency of peripheral odontogenic tumours: A study of 45 new cases and comparison with studies from the literature, J Oral Pathol Med, 35: 385-91.
4. Manor Y, Merdinger O, Katz J, Taicher S (1999): Unusual peripheral odontogenic tumours in the differential diagnosis of gingival swellings, J Clin Periodontol, 26: 806-9.
5. Kearns GJ and Smith R (1996): Adenomatoid odontogenic tumour: An unusual cause of gingival swelling in a 3-year-old patient, Br Dent J, 181: 380-2.
6. Buchner A and Sciubba JJ (1987): Peripheral odontogenic tumours: A review, Oral Surg Oral Med Oral Pathol, 63: 688-97.
7. Philipsen HP, Samman N, Ormiston IW, Wu PC, Peichert PA (1992): Variants of the adenomatoid odontogenic tumour with a note on tumour origin, J Oral Pathol Med, 21: 348-52.
8. Waldron CA (2002): Odontogenic cysts and tumours. In: Neville BW, Damm DD, Allen CM, Bouquot JE (Eds.) Oral and Maxillofacial Pathology, 2 nd ed. Saunders, Philadelphia, Pages: 589-642.
9. Yazdi I and Nowparast B (1974): Extraosseous adenomatoid odontogenic tumour with special reference to the probability of the basal-cell layer of oral epithelium as a potential source of origin, Oral Surg Oral Med Oral Pathol, 37: 249-56.
10. Curran AE (2004): Peripheral odontogenic tumours, Oral Maxillofac Surg Clin N Am, 16: 399-408.
11. Rick GM (2004): Adenomatoid odontogenic tumour, Oral Maxillofac Surg Clin N Am, 16: 333-54.
12. Rajendran R (2006): Cysts and tumours of odontogenic origin. In: Rajendran R and Sivapathasundharam B (Eds.), Shafer's textbook of oral pathology, 5 th ed. Elsevier, Delhi. Pages: 357-432.
13. Lucas RB (1984): Pathology of tumours of the oral tissues, 4 th ed. Churchill Livingstone, Edinburgh, Pages: 61-6.

Figures

[Figure - 1], [Figure - 2], [Figure - 3], [Figure - 4], [Figure - 5], [Figure - 6], [Figure - 7]