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Namrata N Patil, Abhishek Singh Nayyar, Vijay Wadhwan

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Case Series: Herein, we have discussed a case series of four cases of follicular adenomatoid odontogenic tumors in the maxilla, illustrating the diverse clinical, microscopic and biological features of the tumor.

Conclusion: Several updates and data available have grouped adenomatoid odontogenic tumor into different areas of classifications of odontogenic group of tumors. Histological evidences have been put forth by several authors by denoting the histogenesis of each cell type that an inductive influence process does indeed occur in adenomatoid odontogenic tumor. Through this case series we have emphasized the importance of the relation between the dental follicle and the tumor tissue in the origin and histogenesis of this vital odontogenic lesion.
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Keywords: Adenomatoid Odontogenic tumor (AOT), Hamartomatous, Neoplasm, Epithelial origin, Odontogenic, Histogenesis

INTRODUCTION

The unique hamartomatous benign odontogenic tumor distinctively recognized as adenomatoid odontogenic tumor (AOT) located centrally or peripherally in the jaws is not unknown. A large number of literatures prominently describe this lesion based on its clinical, radiological, histological and epidemiological characteristics [1]. A variety of terms have been used to portray this lesion of which the adenoameloblastoma was in common use for many years since the tumor was considered a histological variant of ameloblastoma. An AOT known today has been first described by Dreybladt in 1907. Philipsen and Birn in 1969 introduced the now generally accepted and very popularly used nomenclature of AOT. Some authors reflect AOTs as being truly benign, non-aggressive non-invasive neoplasms, whereas others see them as a developmental hamartomatous odontogenic growths [2]. Adenomatoid odontogenic tumors constitute about 2.2–7.1% of all odontogenic tumors, and the escalating number of reports in literature on AOT indicates that the tumor develops more often than formerly expected [3, 4]. It has been theorized that the complex system of
the dental laminae or its remnants is the likely origin of the AOT mimicking a peri-apical radiolucent lesion most commonly in the maxillary incisor area [5].

This report presents several unique presentations of AOT with their diversely occurring radiographic and histological connotations and provides an overview about the diagnostic aspects of this tumor including current controversies in its pathogenesis and management.

CASE SERIES

Case 1: A 16-year-old girl was presented with complaints of swelling in the upper right front tooth region of one month duration. Extra-oral examination revealed a diffuse firm non-tender expansion in the right maxillary anterior region. Intra- orally the right maxillary lateral incisor was clinically missing and a hyperemic swelling in the upper labial vestibule was observed. Radiological examination revealed the tumor extending to the close proximity of the right maxillary sinus and the impacted permanent maxillary lateral incisor with a unilocular radiolucent area around the tooth. An occlusal projection radiograph revealed a unilocular cystic lesion measuring 1x1.5 cm near the periapical region of 11 (Figure 1). There was slight erosion of the palatal bone. Vitality test was positive for the teeth in adjoining areas. Based on the clinical and radiographic findings, a provisional diagnosis of dentigerous cyst in relation to impacted maxillary right lateral incisor was given and a differential diagnosis of a benign odontogenic tumor was suspected. Radiographic differential diagnoses included adenomatoid odontogenic tumor and calcifying odontogenic tumor/cyst with impacted maxillary right lateral incisor. Macroscopic findings of the extirpated tumor revealed a variegated, dark-red and grayish-white, roughly granular wall with impacted tooth along with some areas in the lateral side of it appearing like a thin membrane (Figure 2). Microscopic examination revealed the typical pattern of AOT. The odontogenic cells constituted of whorled nests of epithelium together with areas of glandular or ductal pattern (Figure 3). At a few places the cystic cavity was lined by the thin layered stratified squamous epithelium. The solid part of the tumor was composed of sheets of epithelial cells with scanty connective tissue stroma. Characteristic duct like structures comprised a central lumen lined by tall columnar cells in a single layer scattered throughout the tumor (Figure 4). The lumina enclosed fibrillar or homogenous, eosinophilic material attached to the free-margin of the columnar cell in a cuticle-like fashion. In other places, the epithelial cells formed convoluted or whorl-like structures consisting of double strands of columnar cells without central lumen. Other remaining cells were stellate or spindle-shaped, with clear cytoplasm and oval nuclei, forming solid sheets among the structures of columnar cells. The spindle-shaped cells in looser areas gave an appearance resembling stellate reticulum. Eosinophilic, non-calcified, droplets like materials were also recognized within the epithelial sheets. The periphery of the tumor comprised of strands of epithelium one or two cell in thickness forming a trabecular or cribriform configuration.

Case 2: A 13-year-old boy was presented with pain and swelling in upper left back tooth region of the jaw since 15 days. The pain was continuous and dull in nature. Intra-orally buccal and palatal extension of swelling was seen extending from left central incisor to first premolar area. Deciduous lateral incisor and canine in the region were retained. A provisional diagnosis of dentigerous cyst was suggested which was followed by differential diagnoses of AOT, infected radicular cyst with the anterior teeth, central giant cell granuloma and ameloblastoma. Radiographic examination revealed a unicuspic radiolucent lesion encroaching the left maxillary sinus with impacted maxillary left lateral incisor and canine. A panoramic radiograph revealed a large, well circumscribed radiolucency extending throughout the anterior maxilla from the left canine to first premolar region (Figure 5). The lesion produced an expansion extending into the alveolar processes, disrupting the usual orientation of the anterior teeth. Pathology test results were in accordance with the clinical diagnosis of lesion compatible with AOT. Surgical removal by complete enucleation of the lesion and associated unerupted teeth and associated lining was achieved in toto under general anesthesia. Postoperative endodontic therapy and prosthetic rehabilitation were planned further. The excised specimen revealed crown of tooth enclosed within tumor mass and suggested the tumor in reality a follicular cyst with intra-cystic epithelial tumor proliferation or follicular cyst epithelium which may give rise to the tumor (Figure 6). The canine was also extirpated and thin fragmented tissue was seen attached to the cervical portion of the crown. Histopathology indicated typical findings of follicular AOT derived from odontogenic epithelium, which establishes itself around the corona of unerupted, anterior teeth in young patients and constituted of whorled nests of epithelium with areas of glandular or ductal patterns intermixed with occasional spherical calcifications.

Case 3: A 17-year-old boy without any clinical history observed slow growing firm painless mass in the right upper front tooth region extending from the right maxillary lateral incisor to the mesial aspect of right first premolar since three months (Figure 7). Intra-orally retained deciduous canine was observed with normal eruption of the lateral incisor and premolars. Intra-oral periapical radiograph found an impacted permanent canine within a single unilocular radiolucent lesion. A provisional diagnosis of a dentigerous cyst in relation to impacted maxillary canine was arrived at. On local excision, the mass revealed the tumor enclosing the tooth. Microscopy indicated a distinctive histomorphology of AOT in addition to ductular or glandular patterns. The cuboidal or columnar cells formed convoluted cords or bodies in complicated patterns showing invaginations.
Case 4: A 12-year-old boy was diagnosed with gradually increasing extra-oral painless swelling on left facial region since two months. Intra-orally extension on the palate was seen extending from the central incisor...
area towards the first permanent molar. Panoramic view revealed impacted permanent canine with a unilocular radiolucency and well demarcated distinct sclerotic margin. Root resorption with premolars and second molar was distinctively visible. Local excision of the mass in toto revealed tumor enclosing the tooth within with gritty and fragile lining of the cystic cavity. Hematoxylin and eosin stained sections indicated cystic lumen lined by highly proliferative odontogenic epithelium arranged in the form of whorls, solid nests, rosettes and ductal patterns with scanty intervening stromal tissue (Figure 8). The epithelial cells in nest and rosette pattern were polyhedral in shape with round prominent nuclei with two to three layers of spindle shaped cells forming stream like pattern, center of nodules showed eosinophilic amorphous or flocculent material within the lumen. The ducts like structures were lined by tall columnar epithelial cells with elongated oval nuclei which were seen polarized away from luminal surface. Calcified masses scattered among the cellular pattern were significantly observed.

**DISCUSSION**

This case series illustrates the clinico-pathological variants of the follicular variety of AOT which is often pre-operatively interpreted as a follicular odontogenic cyst with the association of an unerupted or impacted tooth. AOT is frequently misdiagnosed as other odontogenic cysts and tumors on radiographic examination [5, 6]. Seventy-seven percent of follicular type AOT’s are initially diagnosed as dentigerous cysts [7]. This entity is most commonly encountered in young patients, especially in the second decade of life and is exceptional in the older age groups [8]. Its common occurrence in the maxilla, presentation as a slow growing asymptomatic lesion and its frequent discovery during routine radiography makes it amongst the preferred presumptions in the differential diagnosis of anterior maxillary tumors. Radiographically, the intra-bony variants comprise follicular and extra-follicular types. The most common and widely accepted clinico-topographic variants include (i) follicular (ii) extra-follicular and (iii) peripheral; all of which possess analogous histological findings [9]. The follicular type shows a well-defined unilocular round or ovoid radiolucency associated with the crown and often part of the root of an unerupted tooth thus mimicking a
Odontogenic tumors including the so-called adenomatoid odontogenic tumors (AOTs) comprise a heterogeneous group of lesions that ranges from hamartomas to benign and malignant neoplasms of variable aggressiveness. This case series shows how the lack of uniform criteria employed for their proper identification as well as the histomorphologic similitude found among some of them which behaves in different way, and the scantiness of proper methods to determine their precise origin makes necessary to recognize that at present, in spite of having more or less strict diagnostic criteria which have been internationally accepted, there is a need to continue developing research in the epidemiological, clinico-pathological, morpho-physiological and therapeutic fields in this area of oral and maxillofacial pathology and medicine.

CONCLUSION

Odontogenic tumors including the so-called adenomatoid odontogenic tumors (AOTs) comprise a heterogeneous group of lesions that ranges from hamartomas to benign and malignant neoplasms of variable aggressiveness. This case series shows how the lack of uniform criteria employed for their proper identification as well as the histomorphologic similitude found among some of them which behaves in different way, and the scantiness of proper methods to determine their precise origin makes necessary to recognize that at present, in spite of having more or less strict diagnostic criteria which have been internationally accepted, there is a need to continue developing research in the epidemiological, clinico-pathological, morpho-physiological and therapeutic fields in this area of oral and maxillofacial pathology and medicine.

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Author Contributions

Namrata N Patil – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published.
Abhishek Singh Nayyar – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Vijay Wadhwan – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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