Adenomatoid Odontogenic Tumor With Fibro-osseous Reaction in the Surrounding Tissue

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Abstract: Adenomatoid odontogenic tumor (AOT) is a relatively rare odontogenic tumor that is exclusively odontogenic epithelium in origin. We present a rare case of an AOT in a patient with fibro-osseous reaction in the surrounding tissue. A 22-year-old woman complained of gradual swelling of the right maxillary for 1 month. Radiography showed a well-defined radiolucent lesion with root resorption of the involved teeth. The biopsy revealed a primarily cystic lesion surrounded by a solid portion. Microscopically, the cystic part mainly consisted of epithelial cells organized in solid nodules, whorls, and rosettes, typically characteristic of AOT. But the surrounding solid portion showed cellular fibroconnective tissue stroma with prominent calcified spherules corresponding to ossicles and cementicles, characteristic of ossifying fibroma. The presence of a prominent fibro-osseous reaction in our case is unique. To our knowledge, these findings have not been observed in the previous reports of AOT. It could well represent a cellular cystic wall with metaplastic ossification, rather than a benign fibro-osseous neoplasm such as ossifying fibroma. The tumor had no recurrence after local resection at 5-year follow-up.

Key Words: Adenomatoid odontogenic tumor, fibro-osseous reaction, ossifying fibroma

Adenomatoid odontogenic tumor (AOT) is composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth.[1] Adenomatoid odontogenic tumors hybrid with other odontogenic tumors or several variant histopathologic subtypes were presented in some reports.[2–6] The overlapping clinical and histopathologic features of these hybrid tumors or subtypes have led to diagnostic dilemma and confusion. Our patient was a 22-year-old woman thought to have a benign odontogenic tumor. In this report, we describe the histopathology of this rare AOT with feature of ossifying fibroma (OF) and discuss the priority of diagnosis of AOT with fibro-osseous reaction to diagnosis of AOT with OF. We also reviewed the reported variant histological patterns of AOT. To our knowledge, this is the first case of AOT developing together with fibro-osseous reaction described in the literature.

CLINICAL REPORT

A 22-year-old girl presented in June 2007 with a swelling of the right side of her face for 1 month. The patient's medical history was noncontributory. Physical examination showed a painless and circumscribed swelling of the right maxillary, but skin color was normal. Panoramic radiograph demonstrated a well-defined radiolucent lesion at the apices of the roots of the right maxillary from the lateral incisor to the second molar. It is unilocular. There were root resorptions in the involved teeth (Fig. 1). The clinical impression was benign odontogenic tumor. The tumor was treated by local resection, and excisional biopsy was performed. Postoperative follow-up was performed annually, and the tumor had no recurrence until June 2012 (5-year follow-up).

The biopsy specimens were fixed in 10% buffered formalin, dehydrated through graded concentrations of ethanol, and subsequently embedded in paraffin wax. Serial 4-mm-thick sections of the tumor were processed for hematoxylin-eosin staining.

Grossly, the size of the mass was 4.5 × 2.5 × 2.5 cm. Transversely, the biopsy specimen was primarily a cystic lesion surrounded by a solid portion (Fig. 2). What is interesting is the correspondence between gross and microscopic findings. Microscopically, the cystic portion had an ameloblastoma-like odontogenic epithelium. There were deposits of dentinoid material adjacent to the epithelial lining (Fig. 3A). In most of the area, the tumor wall mainly consisted of cells organized in solid nodules, whorls, and rosettes (Fig. 3B). Small ductlike structures lined with cuboidal to low columnar cells were present throughout the tumor. From the above histological findings, we can diagnosis it as AOT. In addition, the surrounding solid part showed cellular fibroconnective tissue stroma with prominent calcified spherules corresponding to ossicles and cementicles (Fig. 4), characteristic of OF.

DISCUSSION

Philipsen and Birn[7] first introduced the term AOT in 1969. It appears in 3 clinicotopographic variants: follicular, extrafollicular, and peripheral.[8] The age distribution with a very tall peak in the second decade makes the AOT unique among odontogenic tumors. Asians have marked female predominance compared with non-Asians. The present case was an extrafollicular intraosseous variant and accorded with population characteristics. This occurrence of AOT developing together with an OF-like lesion was considered very unusual.

In the present case, the part of AOT was very typical. However, the diagnosis of OF might be controversial. Admittedly, induction of hyaline, dysplastic dentinoid material, or calcified osteodentin has often been found in AOT. But the connective tissue stroma of AOT is generally very loosely structured and contains thin-walled congested

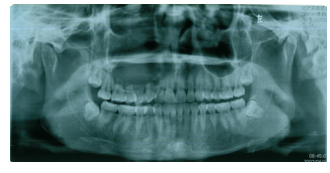

FIGURE 1. Panoramic radiograph reveals a circumscribed intrabony unicystic lesion absorbing the root apices from the right lateral incisor to the second molar.

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Received August 8, 2012.
Accepted for publication October 11, 2012.
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The authors report no conflicts of interest.

ISSN: 1049-2275
DOI: 10.1097/SCS.0b013e3182799005

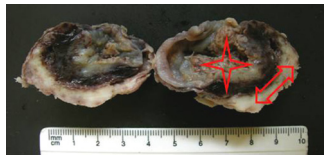

FIGURE 2. Gross examination shows that the excised lesion was primarily cystic as the red stars show, but there was a solid portion in the surrounding as the long arrows suggest.

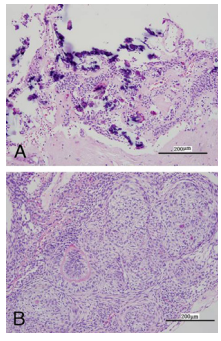

FIGURE 3. A, Photomicrograph of excisional biopsy specimen shows a cystic lesion lined by an ameloblastoma-like odontogenic epithelium with deposits of dentinoid material. B, The epithelial wall mainly consisted of cells organized in solid nodules, whorls, and rosettes.

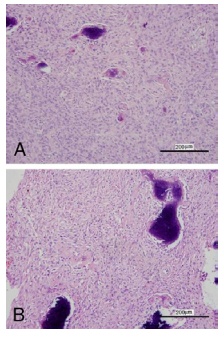

FIGURE 4. A and B, Fibro-rich mesenchymal tissue admixed with numerous islands of calcified ossicles and cementicles, resembling an OF.

vessels characteristically showing marked degenerative changes of the endothelial lining, vessel wall, and perivascular connective tissue. This case showed cellular fibroconnective tissue stroma with prominent calcified spherules corresponding to ossicles and cementicles, characteristic of OF. What is interesting in this case is that the section of OF-like lesion encompassed the section of cystic AOT as a shell. The biologic mechanism causing such a unique combination is not readily apparent. Given the complexity of the inductive effects of odontogenic epithelium on cellular mesenchyme, it is not surprising that this clinical report is unique.

Hybrid odontogenic neoplasms are rare lesions displaying 2 or more histopathologic patterns.[9] Nonaka et al[3] presented a case of AOT associated with a dentigerous cyst affecting the left maxillary region in a 13-year-old girl. Philipsen and Birn[7] reported that occurrence of areas of calcified epithelial odontogenic tumor–like tissue in an otherwise "classic" AOT should be considered a normal feature within the continuous histomorphologic spectrum of AOT. Therefore, calcified epithelial odontogenic tumor–like areas found in AOTs should be considered a histological variant of AOT, as should areas of AOTs mimicking calcifying ghost cell odontogenic cysts,[10] developing odontomas, or other odontogenic tumors or hamartomas.[4,5]

Several scholars have suggested that proliferating odontogenic epithelial might induce the adjacent mesenchymal tissue to develop features of other odontogenic tumors[9] and recommended enucleation of these lesions with close follow-up, including clinical and radiographic examinations. The reason why we prefer to call it a fibro-osseous reaction of AOT instead of a collision of 2 separate lesions is that we believe this case, on the whole, had the same biological behavior and prognosis as AOT. Local resection is an appropriate treatment. It is more likely that this was a single neoplastic process manifesting 2 distinct types of odontogenic lesions. It could well be that the long-standing nature of this particular lesion accounted for a benign fibro-osseous reaction in the surrounding tissue, rather than being representative of a benign neoplasm.

In summary, the present investigation described a case of AOT with fibro-osseous reaction, highlighting the importance of the histopathologic examination in the cystic lesions of the maxillary bones. Moreover, we stress the conservative surgical intervention as a treatment of option for this kind of AOT.

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