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Case Report

A case of adenomatoid odontogenic tumor-like tumor, unusual benign odontogenic tumor in the maxilla

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ABSTRACT

Adenomatoid odontogenic tumors (AOTs) are rare, benign odontogenic tumors characterized by a progressively slow growing pattern and asymptomatic behavior. The most common presentation is a cystic mass involving an unerupted tooth (especially canine), and the usual site is the anterior maxillary region. These tumors are histopathologically thought to arise from the odontogenic epithelium with or without inductive changes in the connective tissue. We herein report a rare case of AOT-like tumor arising in the first premolar region to the first molar region of the maxilla. A 33-year-old male was referred to our hospital for further evaluation of a round radiolucent lesion of the maxilla. After performing a biopsy, which confirmed the diagnosis of AOT, surgical excision was performed under general anesthesia. The tumor was encapsulated and relatively large (approximately 30 mm in maximal diameter) for an AOT. Furthermore, an unusual finding of the root resorption of adjacent teeth was observed. The histopathological examination showed duct-like structures composed of regularly single- or double-layered cuboidal cells; however, there were no duct-like structures composed of columnar epithelial cells characteristic of AOT. On the other hand, the existence of melanocytes, ghost cells, and CK19-positive cells suggests that our case was a benign odontogenic tumor. Taking all findings into account, we diagnosed this patient with an AOT-like, benign odontogenic tumor. The patient's postoperative course was uneventful, and no signs of recurrence have been found 2 years after the operation.

1. Introduction

An adenomatoid odontogenic tumor (AOT) is a rare, benign epithelial odontogenic tumor which accounts for 2–7% of all odontogenic tumors [1,2]. The recently revised WHO classification of odontogenic tumors [3] defines an AOT as a tumor composed of odontogenic epithelium in a variety of histo-architectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth. Typically, these lesions arise in the lateral incisor/canine region of the maxilla, where they produce swelling. Only in very rare cases is the lesion found distal to the premolar area [4,5]. We herein report an AOT-like tumor that presented highly unusual clinicopathological findings.

2. Case report

In June 2008, a 33-year-old male was referred to our hospital for further evaluation due to a gradually increasing swelling in the gum and a cyst-like radiolucent lesion of the left maxilla that had been detected 5 months before. There was neither pain nor paresthesia. The patient had no past or familial history of tumors or cysts in the oral region. There was no regional lymphadenopathy. An intraoral examination revealed a rather well-circumscribed swelling of the left maxilla extending from the first premolar region to the first molar region (Fig. 1). The lesion presented elastic-like softness due to disruption of the underlying bone, and the overlying mucosa was normal in color.

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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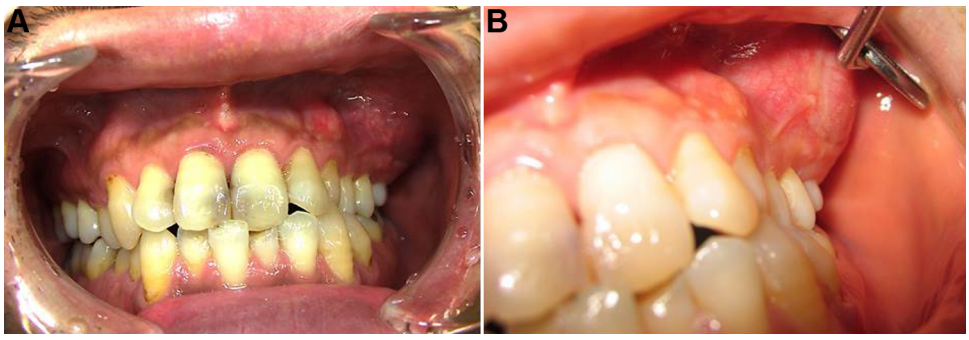

Fig. 1. Intraoral photographs taken at the initial examination. A rather well-circumscribed swelling was seen in the left maxilla extending from the first premolar region to the first molar region. (A) Frontal image; (B) magnified image.

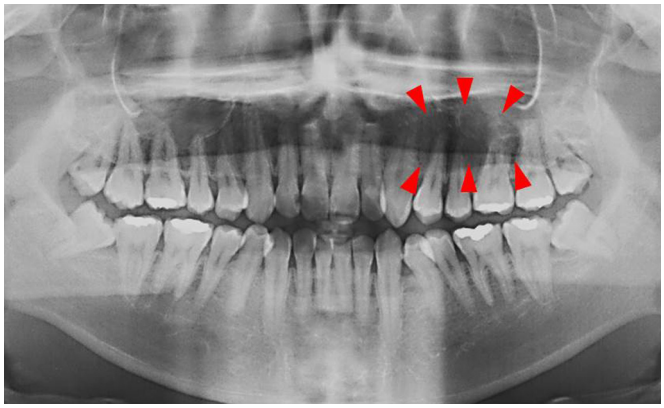

Fig. 2. A panoramic radiograph taken at the initial examination. A panoramic radiograph demonstrated a round radiolucent area of an intraosseous lesion, with a well-defined border (arrowheads) in the left maxilla extending from the first premolar region to the first molar region.

A panoramic radiograph and computerized tomography (CT) revealed a round radiolucent image of an intraosseous lesion with a well-defined border in the left maxilla extending from the first premolar region to the first molar region (Figs. 2 and 3). The roots of the second premolar and first molar were resorbed (Fig. 3B), but both teeth were vital. No embedded tooth was found.

An incisional biopsy was performed under local anesthesia. A histopathological examination revealed a diagnosis of AOT with no evidence of malignancy. Thereafter, surgical excision was performed under general anesthesia (Fig. 4A). During the surgery, the maxillary first molar in the affected area was extracted. The tumor was encapsulated and approximately 30 mm in maximal diameter (Fig. 4B). The patient's postoperative course was uneventful, and no signs of recurrence have so far been observed 2 years after the operation (Fig. 5).

3. Histopathological findings

The excised lesion was primarily composed of proliferative epithelial cells with a hyalinized stroma that formed a variety of duct-like structures of various sizes (Fig. 6A). These duct-like structures were composed of regularly single- or double-layered cuboidal cells; however, there were no duct-like structures composed of columnar epithelial cells characteristic of AOT (Fig. 6B and C). The luminal side of the duct-like structures was mostly lined with thin layers exhibiting a degree of hyalinization with exudate inside (Fig. 6B and C). Some pigmented cells, likely melanocytes, were seen in the cell-rich areas of the stroma (Fig. 6D), as well as in the parenchymal tissues of the tumor. In addition, masses composed of ghost cells were often observed in the parenchymal tissue of the tumor (Fig. 6E). The tumor cells were immunohistochemically positive for CK19 (Fig. 6F). Based on the above histopathological findings, the patient was diagnosed with an AOT-like, benign odontogenic tumor.

4. Discussion

The AOTs can be subdivided into three clinico-topographic variants: the follicular type, associated with an unerupted tooth; extrafollicular type, not associated with an unerupted tooth and superimposed upon the roots of erupted permanent teeth; and the peripheral type [4–6]. The follicular and extrafollicular variants are both intrabony and central tumors and account for approximately 96% of all AOTs, of which 71% are of the follicular type [4]. Our case was topographically classified as the extrafollicular type. Concerning the localization, the AOTs most frequently occur intraosseously, with a preference for the anterior maxilla. Based on these classifications and the incidence of the different types, the present case therefore seemed to be a relatively rare case of central AOT, if it was indeed an AOT.

The radiographic appearance of an extrafollicular AOT is reported to present as a well-defined unilocular radiolucent lesion [3], and the present case also showed such an appearance. On the other hand, when the lesions are located between teeth, divergence of roots may be seen, but root resorption is rare [7,8]. To the best of our knowledge, only four cases of AOT with root resorption have so far been reported [7,9]. In the present case, however, an irregular type of root resorption was observed on the second premolar and first molar. Since the size of the intraosseous AOTs generally varies from 1 to 3 cm in diameter [10], the root resorption may be an unusual finding observed in the relatively larger AOTs, if the tumor was an AOT.

Histopathologically, the tissue specimen of the excised lesion showed a variety of duct-like structures of various sizes formed by proliferative tumor cells. In the low-power field, since the duct-like structures appeared to be cribriform structures, a salivary gland tumor should be considered in the differential diagnosis. On the other hand, the immunohistochemical analysis revealed CK7, CK14, and CK19 to be positive and vimentin to be partially positive, whereas α-SMA and S-100 were negative (data not shown, except for those for CK19). These results suggest that this case did not involve a salivary gland tumor. Furthermore, the histopathological findings revealed no duct-like structures composed of columnar epithelial cells characteristic of AOT, thus contributing to the final diagnosis of an AOT-like tumor. However, since the luminal side of the duct-like structures was mostly lined with thin layers exhibiting a degree of hyalinization, these structures were considered to be identical to stromal cysts. Including the reports from Japan, the existence of melanocytes (pigmented variant) is often observed in odontogenic tumors [11–13]. In addition, many ghost cells were