Case Report

A rare nonsyndromic case of adenomatoid odontogenic tumor associated with multiple impacted supernumerary teeth

ABSTRACT

Adenomatoid odontogenic tumour (AOT) is a rare benign odontogenic tumour characterized by a progressively slow growing pattern and symptomless behavior. The differential diagnosis between AOT and other odontogenic tumours, such as ameloblastoma, should be well conducted in order to avoid extensive ablative surgery. The present case report is of a 27 year old female who reported with a chief complaint of mild intermittent pain and a swelling which was gradually increasing in size, on the left mid-facial region since 6 months. Radiographic Investigations revealed a round uni-locular radiolucent image of an intra-osseous lesion with a well defined border in the left maxilla. The tumor was encapsulated and was attached with 2 impacted supernumerary teeth which were fused and shaped as maxillary premolars. The images also showed multiple impacted supernumerary teeth in the maxilla and mandible. The clinical and radiographic diagnostic hypothesis of Adenomatoid odontogenic cyst and a differential diagnosis of Dentigerous Cyst was given. Surgical enucleation of the lesion was done under General Anaethesia. The histological sections were consistent with AOT.

Keywords: Adenomatoid odontogenic tumor, benign tumor, supernumerary teeth

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) was initially known as "adenoameloblastoma" or "ameloblastic adenomatoid tumor," due to the nascent concept of it being a variant of ameloblastoma. In due course, its odontogenic neoplastic characteristics were identified and recognized by Stafne in 1948.[1-3] AOT is quite rare and is reported to account for 2%–7% of all the odontogenic tumors.[4] The revised WHO classification defined it to be a tumor which is composed of odontogenic epithelium depicting a variety of histo-architectural patterns, embedded in a mature connective tissue and is characterized by slow but progressive growth.[5] In the oral cavity, any teeth which exceed from the normal dental formula is termed as supernumerary tooth. It can be erupted or unerupted and the phenomenon is termed as hyperdontia. It can be single or multiple and can involve either single or both the jaws.[6] In the present case report, we present a case of nonsyndromic multiple impacted supernumerary teeth associated with adenomatoid odontogenic tumor.

CASE REPORT

A 27-year-old nursing female came to the Department of Oral and Maxillofacial Surgery, Kalinga Institute of Dental Sciences, with a chief complaint of mild intermittent pain and

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a swelling which was gradually increasing in size on the left mid-facial region for 6 months. The patient had undergone a C-section delivery 3 months back. There was no other relevant medical or family history of the patient. On extraoral examination, there was a facial asymmetry with a solitary diffuse swelling on the left side near the nasolabial fold of size approximately 1 cm × 1 cm [Figure 1]. The swelling was hard and nontender, and there was no associated regional lymphadenopathy. Intraorally, there was a diffuse swelling of size approximately 3 cm × 2 cm present in the left side maxilla extending from the lateral incisors to the first premolar, obliterating the buccal vestibule [Figure 2]. There were 30 permanent teeth present with the absence of the upper right and lower left third molars. The patient was advised to undergo orthopantomogram and cone-beam computed tomography (CBCT) imaging, which revealed a round unilocular radiolucent image of an intraosseous lesion with a well-defined border in the left maxilla extending from the periapical region of the lateral incisor to the mesial root of the first molar [Figure 3]. The lesion had engulfed a radio-opaque mass, which was supposedly an impacted tooth located within. The images also showed multiple impacted supernumerary teeth in the maxilla and mandible [Figures 3-5]. Surgical excision was planned and performed under general anesthesia, in which the affected area was approached through a vestibular incision. The tumor was encapsulated and was attached with two impacted supernumerary teeth, which were fused and shaped as the maxillary premolars [Figure 6]. The tumor was removed and sent for histopathological examination. The patient was healthy postoperatively without any specific complaints.

Figure 1: Clinical photograph showing swelling of the left paranasal area

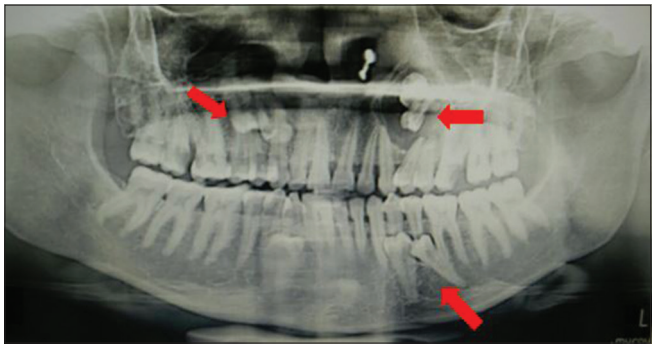

Figure 3: Orthopantomogram showing the well-circumscribed radiolucent lesion with distinct, radiopaque margins, and the multiple impacted supernumerary teeth

Histopathological findings

The section of the given specimen showed a connective tissue component composed of densely packed collagen fiber bundles, arranged parallel resembling a capsule. The capsule enclosed a cystic space. The inner wall of the cystic space was lined by an odontogenic epithelial lining [Figure 7]. The epithelial lining was proliferative in nature at the one end and is composed of spindle-shaped epithelial cells. These epithelial cells were also forming duct-like structures. The

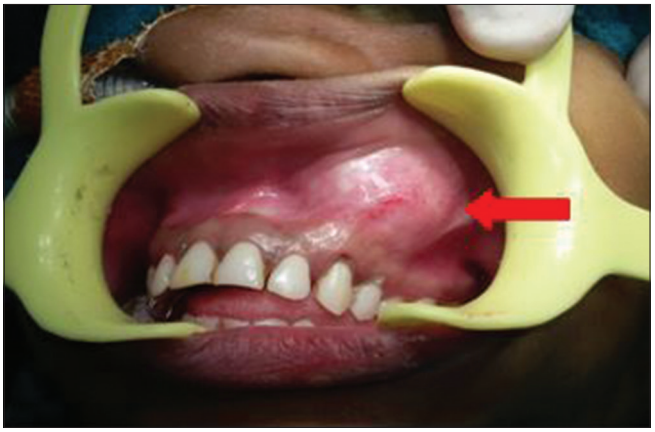

Figure 2: Intraoral photograph showing the swelling of the left maxillary vestibular area

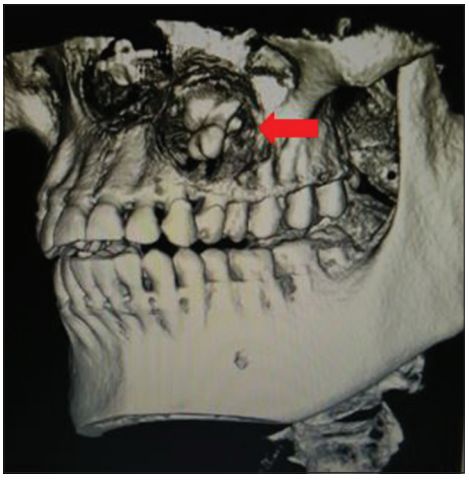

Figure 4: Three-dimensional cone-beam computed tomography image showing the lesion and the associated impacted teeth

Figure 5: Computed tomography scan showing circumscribed radiolucency with thin radiopaque margin encompassing a supernumerary tooth and the multiple impacted supernumerary teeth in both the jaws

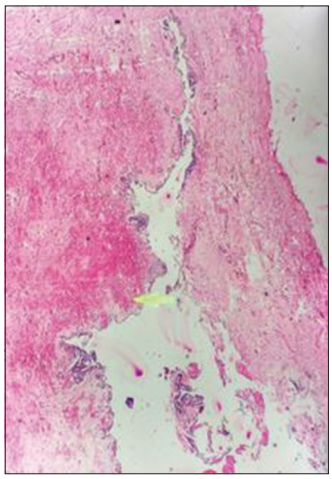

Figure 7: A connective tissue capsule lined by an odontogenic epithelial lining which is proliferating at one area

peripheral cells of the ductal pattern showed a palisading structure and reversal of polarity [Figure 8]. Some areas showed calcification inside the epithelial island. Based on these above histological findings, a final diagnosis of AOT was given.

DISCUSSION

Impaction of teeth is not quite a rare incidence, and its etiological factors can vary from rotated teeth, lack of space for permanent dentition, early loss of deciduous teeth to lack of eruptive forces, or genetic factors. On the other hand, causes of the presence of the supernumerary teeth are actually unknown, and the presence of multiple impacted supernumerary teeth is quite rare.[7,8] Although there are many documented syndromes of which supernumerary teeth are one of the features, such as cleidocranial dysostosis, Gardner's syndrome, Fabry–Anderson's syndrome, Ehlers–Danlos syndrome, and trichorhinophalangeal syndrome, our patient was nonsyndromic and also had no

Figure 6: The whole of the excised lesion with the associated impacted teeth

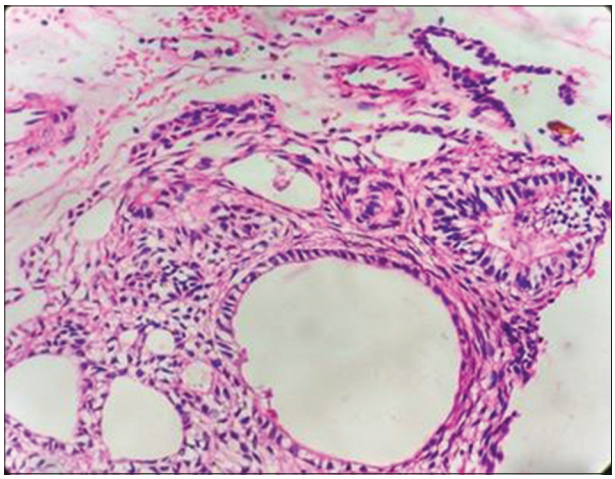

Figure 8: The peripheral cells of the ductal pattern showing palisading arrangement

such genetic link. Impacted teeth have a large tendency to be associated with dentigerous cyst, thus we went ahead with the treatment plan of surgical excision for our patient, with a differential diagnosis of dentigerous cyst and adenomatoid odontogenic cyst (AOT) based on the radiological findings. There are three basic variants of AOT: (1) follicular, (2) extrafollicular, and (3) peripheral. Follicular variant is mostly associated with an impacted tooth and displays a central intraosseous lesion. Extrafollicular type also presents with an intraosseous lesion but is unrelated to any unerupted tooth and mostly is seen between the roots of erupted teeth. Peripheral AOTs are extremely rare entities.[3] The tumor is generally seen in the second decade of life, showing a maxillary and female predominance.[9] The dentigerous cyst is distinguished from the AOT because it encloses only the coronal portion of an impacted tooth. In contrast, the AOT usually surrounds both the coronal and radicular aspects of the involved tooth. Orthopantomograms are the most widely used imaging modality used to locate supernumerary teeth, impacted teeth, and other pathologies of the jaws such as AOT and dentigerous cysts, but with the advent of three-dimensional imaging options such as CBCT, it has become the diagnostic tool of choice. The shortcomings in the panoramic radiographs such as differentiating the supernumerary teeth and the associated osseous structures,

as they overlap each other, are averted in CBCT, thus making it radiograph of choice.[10]

In the present case, CBCT helped in locating the lesion as well as in confirming the presence of impacted teeth associated with the tumor. It also helped in determining the extent of the tumor.

The patient was reluctant to get other impacted supernumerary teeth extracted as they were asymptomatic, thus no further treatment was done for it.

Patient consent

Relevant consent was obtained from the patient.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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