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Case Report: An Ameloblastic fibro-odontoma in the posterior maxilla

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ABSTRACT

Ameloblastic fibro-odontoma is a rare mixed odontogenic tumor, contains variable amounts of dental hard tissues. It is a controversial lesion. Previously, it was considered as an individual tumor, but recently, the World Health Organization (WHO) has defined it as a developing odontoma. It has histologic features of both ameloblastic fibroma and complex odontoma.

Ameloblastic fibro-odontoma occurs more frequently in the posterior mandible of children and young adults.

This case report presents an extensive developing odontoma or Ameloblastic fibro-odontoma in a 4-year-old boy, positioned in the posterior maxilla; that is a rare region for its occurrence. Facial asymmetry and intraoral swelling were apparent. There was also excessive displacement of permanent first and second molar teeth of the upper jaw to the floor of the orbit. Surgical removal of the lesion and the associated teeth was indicated for treatment. No recurrence was observed after 2 years of follow-up.

Management of an expansile ameloblastic fibro-odontoma with maxillary sinus involvement is challenging. Cone beam computed tomograghy (CBCT) is helpful for more detailed investigation.

Keywords:
Odontoma
Maxilla
Maxillary Neoplasms
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Introduction

Ameloblastic fibro-odontoma(AFO) is a rare mixed odontogenic tumor.1 Histologically, AFO has the same features of the ameloblastic fibroma(AF), but this tumor comprises hard dental tissues, additionally.(2)Clinically, it usually appears as an expansile, painless swelling in the affected region and causes facial asymmetry.(1-3) The lesion is usually contributed with unerupted permanent teeth.(4)The most common region for AFO's occurrence is the posterior mandible1,3 and the posterior region of the maxilla is affected in seldom cases.Radiograpically, AFO is commonly unilacular radiolucency with variable size of radiopaque foci.(2)Based on the latest definition of the World Health Organization (WHO) in 2017, AFO defined as a developing odontoma and is categorized under odontoma. (5)The purpose of this study is to report an extensive developing odontoma in a 4-year-old boy. It was uncommonly located at the posterior maxilla. Opposed to the most of reported cases, AFO in the current patient was locally invasive with invagination into the maxillary sinus and excessive tooth displacement.

Case presentation

In December 2019, a 4-year-old boy was referred to an oral and maxillofacial surgeon's office. His parents complained of swelling in the right posterior maxilla. Extraoral examination showed slight facial asymmetry. The skin color was normal (Figure 1).

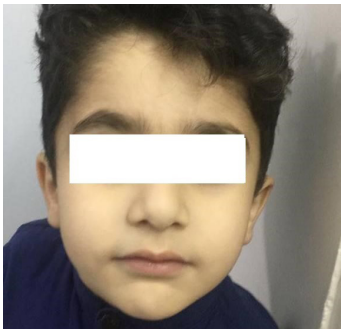

Figure 1. The extraoral photograph shows slight facial asymmetry.

Intraoral examination showed the swelling of the right maxillary alveolar ridge, mainly palatally with traumatized mucosa (Figure 2).

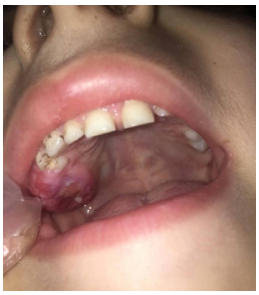

Figure 2. The intraoral photograph reveals traumatized alveolar mucosa in the right posterior maxilla with buccal and palatal expansion. Tooth #3 is unerupted compared to the left side.

The parents mentioned the extraction of loosened tooth #A, recently. The onset of the lesion was unknown, and the parents did not report any previous trauma. A panoramic radiograph was ordered, which showed a mixed radiolucent and radiopaque lesion. There were multiple tooth-like structures within a well-defined radiolucent lesion located at the posterior right region of the maxilla (Figure 3).

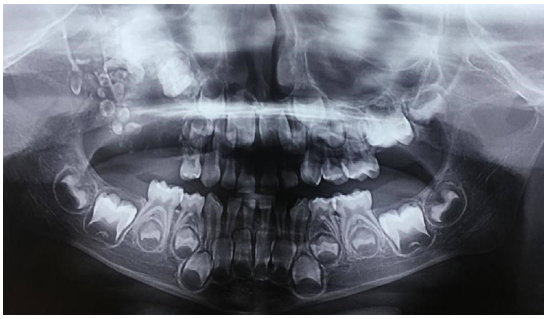

Figure 3. The panoramic radiograph shows multiple tooth-like structures in the right posterior maxilla. Superiorly displaced tooth #3 to the floor of the right orbit is observable. Teeth #2 is not detectable on this radiograph.

Cone-beam computed tomography (CBCT) was suggested for more investigation of any impingement of the maxillary sinus. The lesion was extended to the lateral wall of the nasal cavity, medially, and the floor of orbit, superiorly. A CBCT examination showed that teeth #2 and #3 (universal numbering system) were displaced superiorly to the inferior rim of the orbit. Expansion and thinning of the posterior border of the maxillary sinus were evident in axial views (Figure 4).

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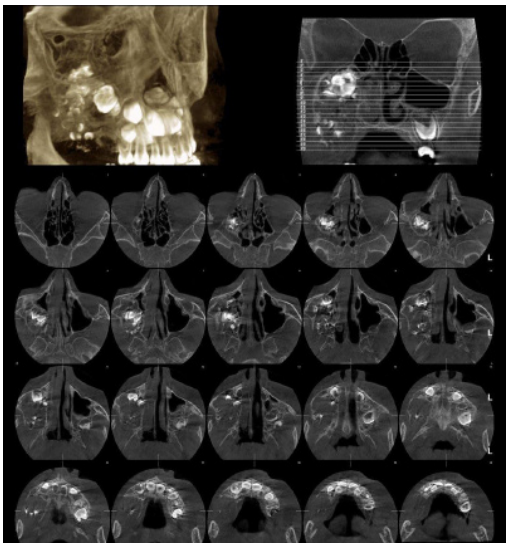
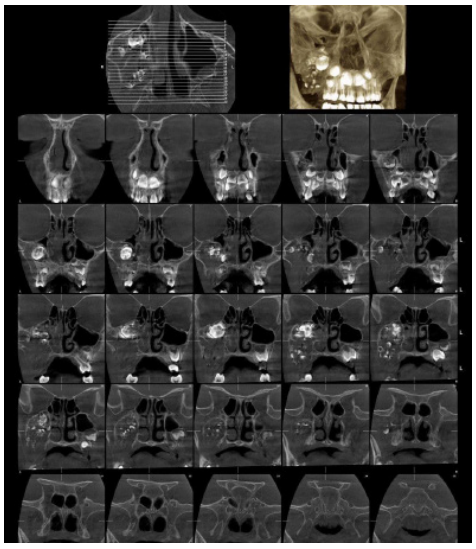
Figure 4. Axial CBCT images show multiple radiopaque tooth-like structures in the right maxillary sinus. Expansion and thinning of posterolateral and anterior walls of the maxillary sinus are detectable.



Also, there was expansion and thinning in the anterior wall of the maxillary sinus. Coronal views of CBCT revealed expansion in the buccal and palatal cortexes of the alveolar ridge (Figure 5).

Figure 5. Coronal CBCT images show medial and lateral expansion of the right alveolar ridge and maxillary sinus. Teeth #2 and #3 are displaced superiorly into the floor of the right orbit. The ostium of the right maxillary sinus is obstructed. There is also right nasal cavity involvement.



The right maxillary sinus ostium was obstructed, and involvement of the right nasal cavity was detected. These features suggested an extensive developing odontoma or ameloblastic fibro-odontoma (AFO) as diagnosis.

Treatment

After taking consent from the parents, surgery was carried out under general anesthesia. An incision from tooth C was made (Figure 6.a). A full mucoperiosteal flap was elevated buccally. There were many small tooth-like structures (Figure 6.b), most of which were irregular in shape. The whole lesion was enucleated along with tooth-like structures (Figure 6.c). Teeth #2 and #3 were extracted because they could not erupt normally. Careful curettage was carried out after lesion removal, followed by irrigation with 0.9% saline solution. The removed buccal fat pad of the right side was used for the closure of the oroantral communication and defect filling (Figure 6.d). Bone graft was not used because of the patient's financial problem. After hemostasis, the flap was sutured back to its normal position. The excised tissue was sent for histopathological evaluation.

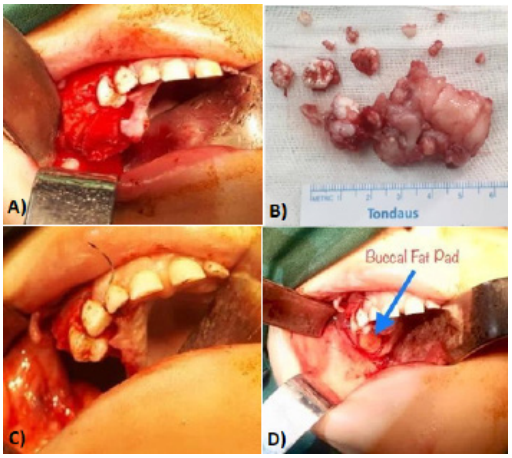


Figure 6. Surgical procedure. An excision was made from tooth C (A), and the entire lesion with tooth-like structures was removed. In addition, unerupted teeth #2, and #3 were extracted (B), careful curettage and irrigation with 0.9% saline was carried out (C). The removed buccal fat pad of the right side was used for closing the oroantral communication (D)

Pathology

Different sections of the received specimen revealed dental hard tissues within ectomesenchymal tissue. Small islands of odontogenic epithelium(ameloblastic) were observed. No evidences of malignant transformation were detected(Figure 7). Based on these features, the diagnosis of AFO was confirmed.

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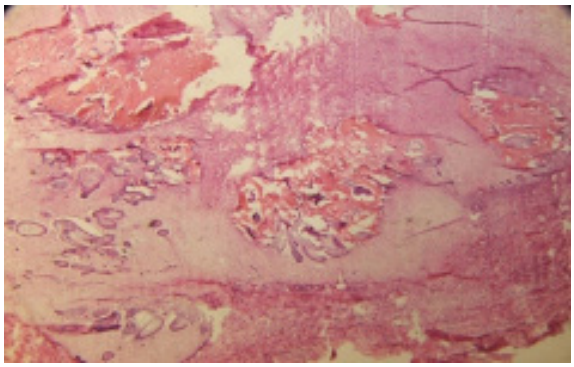

Figure 7. histological examination shows dentin formation and small islands of odontogenic epithelium (ameloblastic) in a loose mesenchymal tissue.

Discussion

AFO is a rare mixed odontogenic tumor.1 Hooker described it as an individual tumor for the first time in 1972.(6)

Histologically, it refers to an AF contains variable amounts of dental hard tissues.3

Radiograpically, AF is a well-defined unilacular or multilacular radiolucent lesion.(2)

Previous literatures stated there are two types of AF. First is the neoplastic type; that does not mature and remains as a radiolucent lesion. Second type considers as the hamartous one, matures to form a complex odontoma.(3)

The WHO in 2005 defined AFO and odontoma as individual lesions,7 but it has considered AFO as a developing odontoma and has categorized it under odontoma since 2017. This classification is based on a hypothesis stating the AFO finally transforms into the odontoma, either complex or compound.8

Cahn and Blum in 1952 stated the theory of sequential maturation, in which AF, ameloblastic fibrodentinoma, AFO and comlex odontoma are different maturation states of a same lesion. The AF is the least and odontoma is the most differentiated state of a lesion maturation spectrum. There is no consensus between authors yet.(3)

AFOs usually finds in the first and second decades of life,2,9 as the current reported case.

Clinically, AFO can manifest as an asymptomatic slow growing lesion, but they usually cause painless swelling and failure of tooth eruption.(1-4,10) Facial asymmetry, intraoral swelling at the posterior upper jaw, and breathing disturbances were the clinical features of the current reported case.

Radiographically, AFO is a radiolucent lesion with radiopaque foci of dental hard tissues. The most common region for its occurrence is the posterior mandible. Maxilla has been involved in rare cases, such as the current reported patient.

AFO can manifest with variable features on radiographs. Most of detected lesions have mixed radiolucent-radiopaque appearance. Just a few completely radiolucent AFOs are reported.(2,10)

Extensive AFO as the presented case, can cause more complications, like cortical expansion, feeding difficulties, breathing problems owing to nasal obstruction, and facial asymmetry. Proptosis due to the lesion expansion and destruction of the floor of the orbit is reported.(3)

Commonly, treatment of the AFO is conservative enucleation and the involved teeth should be removed. There are some disagreements about committal of tooth removal. Teeth which are within the lesion should be removed but the impacted teeth close to the lesion could be preserved.

AFO is an encapsulated lesion and there is a little local invasion,10 but in the current case, AFO was invaginated to the maxillary sinus and obstructed its ostium. Thinning of cortical plates was detectable. Teeth #2 and #3 were displaced to the floor of the orbit. In these extensive cases, there is more complications with surgery. In the current case, preserving the contours of the alveolar process and maxillary sinus integrity was challenging.

The recurrence of the AFO is rare and is contributed with inadequate surgical removal.(9) A literature review revealed no recurrent cases approximately 24 months after the surgery.(11) Malignant transformation occurs rarely and seems to be more in adults.(3)

Conclusion

AFO is a rare benign tumor with variable radiographic features. In extensive lesions, CBCT can be useful for more detailed investigation. Surgical excision is the treat-

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ment of choice after histopathological diagnosis, but probably there are more surgical complications with these extensive lesions.

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Authors'contributions

Zahra Jahanshahiafshar: Conceptualization, Methodology, Writing - Review & Editing

Mina Yazdizadeh: Resources, Investigation, Visualization **Ramin Forooghi:** Data curation, Writing - Original Draft **Maryam Johari:** Project administration, Supervision, Funding acquisition

Conflict of Interests

The authors declare no conflict of interest.

Ethical declarations

Not applicable

Financial support

None

Availability of data and material

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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