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Management of Adenomatoid Odontogenic Tumor in a Pediatric Patient with Preservation of an Associated Impacted Tooth: A Combined Surgical and Orthodontic Approach

Saumya Taneja[1*], Anuj Jain[2]

1. Department of Pedodontics and Preventive Dentistry, Inderprastha Dental College, Ghaziabad, U.P, India.
2. Department of Oral and Maxillofacial Surgery, Dr.H.S.R.S.M. Dental College, Hingoli, Maharashtra, India.

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**** Corresponding author:***
Department of Pedodontics and Preventive Dentistry, Inderprastha Dental College, Ghaziabad, U.P, India.
Email: drsaumyataneja@gmail.com

A B S T R A C T

The adenomatoid odontogenic tumor (AOT) is a rare benign neoplasm that is typically treated through surgical enucleation and curettage. Any impacted tooth associated with the tumor are also extracted during the procedure. We present a case of AOT encompassing an impacted left maxillary central incisor in a 13-year-old male. The patient underwent routine treatment, but the tooth was orthodontically extruded using traction. Within two years, the incisor successfully attained its functional position in the arch, without any signs of recurrence. Preserving an impacted tooth associated with AOT is not common, and only a few cases have been reported in the literature. It is important to manage AOT conservatively and to save the associated tooth for both functional and aesthetic purposes. Long-term follow-up is crucial to monitor any potential recurrence. In summary, our case highlights the successful orthodontic management and preservation of an impacted tooth in a teenager with AOT.

Keywords: Odontogenic Tumors; Tooth, Impacted; Oral Surgical Procedures; Orthodontic Extrusion

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INTRODUCTION

Adenomatoid Odontogenic Tumor (AOT) is a rare benign odontogenic neoplasm originating from the dental lamina or its remnants, representing about 3% of all odontogenic tumors [1]. Initially, AOT was considered to be a type of ameloblastoma; however, it was later recognized as a separate entity by Steensland in 1905 [2]. Various terminologies such as ameloblastic ameloblastoma, adamantinoma, adeno-ameloblastoma, teratomatous odontoma and epithelioma adamantinum had been used to describe this lesion until 1969 when Philipsen and Birn coined the term adenomatoid odontogenic tumor. In 1971, the World Health Organization (WHO) adopted this terminology [3].

AOT is more common in the second decade of life with rare occurrence in children and individuals above 30 [4]. It shows a female preponderance, with a male to female ratio of around 1:2 [5]. The most common site of occurrence of AOT is the anterior maxilla followed by the mandible. It has been observed in posterior regions as well, but seldom beyond the premolars [6].

There are three clinical variants of AOT, including intraosseous follicular, intraosseous extra-follicular and extraosseous peripheral. The most prevalent form is the follicular type which is associated with an impacted tooth

and accounts for 70% of the cases. Extra-follicular AOT occurs between erupted teeth and constitutes 25% of the cases, whereas 5% of tumors are of the peripheral variety present in the gingiva. [7] AOT is usually a small (<3 cm), painless, slow growing tumor but occasionally can have an aggressive nature, unusual increase in size, and extension into intracranial spaces [8].

Radiographically, AOT appears as a unilocular radiolucency with a distinct radiopaque border, mostly associated with an impacted tooth. Multiple small radiopaque foci or calcification may also be seen in some cases. Management of this odontogenic tumor is surgical enucleation or curettage along with extraction of the associated impacted tooth. [9] We hereby present a young male diagnosed with AOT associated with an impacted maxillary central incisor and discuss its surgical management.

CASE REPORT

A 13-year-old male patient reported with a painless swelling in the upper front region of the jaw since 15 days. Extraoral examination revealed a diffuse, firm, non-fluctuant and non-reducible swelling in the supralabial region with normal overlying skin. There was no sensory or motor deficit. Intraoral examination revealed a painless swelling in the left maxillary anterior region extending from the midline to the left maxillary canine. The buccal cortical bone showed expansion and the left maxillary central incisor was missing (Figure 1A).

An orthopantomogram was taken which depicted a well-defined unilocular, rarefying lesion with a well-corticated border completely surrounding the crown and middle third of the root of the impacted left maxillary central incisor (Figure 1B). There was slight displacement and resorption of the adjacent lateral incisor. Based on the clinical and radiographic examination, a provisional diagnosis of AOT was made.

The patient was subjected to surgery under local anesthesia with a treatment plan of enucleation and curettage of the lesion and possible preservation of the tooth. Under all aseptic precautions of antibiotic coverage, infraorbital nerve block was given using 2% lidocaine hydrochloride with 1:80,000 adrenaline. After confirmation of anesthesia, crestal incision was given and a full thickness mucoperiosteal flap was elevated. Necrotic fragile bone around the fenestration defect was removed. The cyst content included a clear straw-colored fluid. The cyst lining was gently teased out from the bone cavity and was sent for histopathological examination (Figure 2).

The central incisor was retained to allow its eruption into the oral cavity. Hydroxyapetite crystals (Sybograf-T, Eucare, Chennai, Tamil Nadu, India) along with platelet rich fibrin were placed and condensed into the bony defect to facilitate healing and osseogenesis.

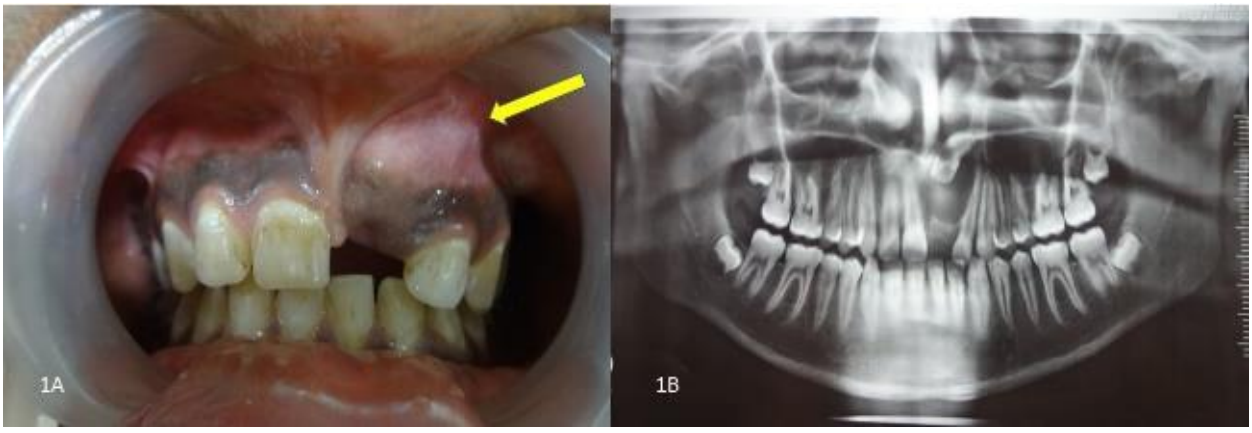

Fig. 1. Clinical findings of the patient. **A)** Intraoral photograph revealing swelling in the maxillary left central incisor region. **B)** Orthopantomogram showing a well-defined radiolucent area surrounding the impacted left maxillary central incisor

Fig. 2. Exposed tooth in bony cavity after removal of cystic lining.

The flap was repositioned and sutured using 3-0 silk (Ethicon Mersilk, Hyderabad, Telangana, India). The patient was recalled after 7 days for suture removal and was kept on a regular monthly follow up.

Histopathological examination of the sample revealed a well circumscribed proliferation of duct-like epithelium in rosette like aggregates with sparse areas of eosinophilic material in the center confirming the diagnosis of AOT (Figure 3).

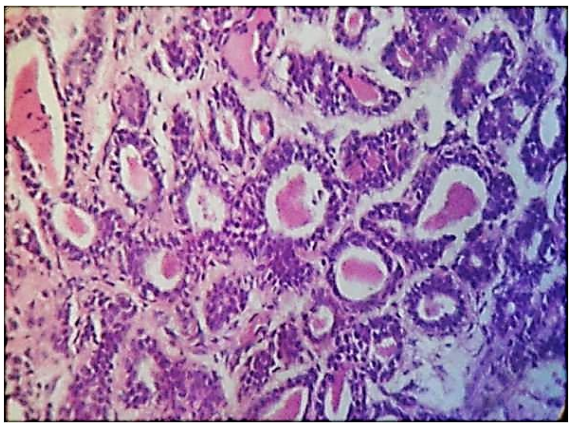

Fig. 3. Representative histopathological image of the enucleated adenomatoid odontogenic tumor (hematoxylin & eosin staining, magnification ×100)

At 3 months postoperatively, clinical and radiographic examination showed satisfactory healing. However, no signs of tooth eruption were present (Figure 4). Hence it was decided to orthodontically extrude the tooth by traction as three months is considered a critical period for beginning an additional treatment to stimulate tooth eruption.

Fig. 4. Orthopantomogram at 3 months depicting successful healing but no signs of eruption of the central incisor

A second surgery was carried out where a full thickness mucoperiosteal flap was raised under local anesthesia and a bracket was bonded to the impacted tooth (Figure 5).

Fig. 5. Bonded bracket and secured ligature wire on the impacted exposed tooth to carry out orthodontic extrusion

A ligature wire was secured to the bracket for orthodontic extrusion and the flap was sutured back in position. Light continuous orthodontic forces were applied and the patient was recalled every month for activation.

The extrusion of the impacted tooth continued for approximately two years, during which there was no recurrence of the tumor. At the end of two-year period, the incisor became well-aligned in its functional position in the maxillary dental arch and the patient was satisfied with the treatment outcome, which was both esthetic and functional (Figure 6).

Fig. 6. Extruded and well aligned left maxillary central incisor at the end of the 2-year treatment period

DISCUSSION

AOT is a rare benign neoplasm originating from the odontogenic epithelium, which occurs in young patients in the anterior part of the maxilla. It is most commonly associated with an impacted maxillary canine. [5] AOT is also known as the "tumor of 2/3" because 2/3 of cases appear in the maxilla, 2/3 arise in young females, and 2/3 are found in association with maxillary canines and unerupted teeth. However, our case involved an impacted central incisor of a male child.

Differential diagnosis of AOT includes dentigerous cyst, odontogenic keratocyst, calcifying odontogenic cyst or calcifying odontogenic tumor. Painless swelling and missing permanent teeth are the most common symptoms of this tumor. Therefore, clinically AOT can be misdiagnosed as a dentigerous cyst or ameloblastoma. AOT usually envelopes the crown and root of an impacted tooth, whereas dentigerous cyst just surrounds the crown. [10] Histopathological examination remains the key to accurate diagnosis. The present case displayed typical features of a cystic cavity lined by proliferative odontogenic epithelium with palisaded basal cells, polarized columnar cells and inverted nuclei characteristic of AOT [11].

Management of AOT is surgical enucleation and curettage with extraction of the associated tooth in the majority of cases. Variations exist depending on the size and progression of the lesion. In such cases extensive surgical procedures are adopted for management of the tumor [12].

In our case, surgical enucleation and curettage of the lesion was performed and the involved tooth was salvaged by extruding it through orthodontic traction ensuring that the patient was functionally and esthetically satisfied.

After extensive search of the English literature, we came across very few cases where impacted teeth associated with AOTs were preserved and orthodontically managed [13-15]. Bonardi et al. [14] presented a case where AOT caused impaction of a mandibular deciduous canine. The tooth was extracted along with the lesion, releasing the path of eruption for the permanent tooth but no orthodontic intervention was performed. The other two cases were related to AOT involving the maxillary canine where the tooth was orthodontically extruded and aligned [13,15]. It is recommended that in cases where permanent teeth are associated with a lesion, preservation should only be considered in cases where there is still potential for eruption. The degree of root formation is also an important consideration, meaning that in order to prevent tooth dislodgement, the root of the involved tooth should be completely formed. Other factors to consider include the angulation or position of the tooth, the age of the patient, the individual's esthetic expectations, the likelihood of tumor recurrence, and the availability of enough space for tooth eruption[15]. In the current case all these issues were taken into account before deciding to retain the tooth. It is noteworthy that the treatment procedure and tooth preservation performed in the present case can't be generalized to all the cases of AOT encompassing a permanent tooth in a child.

CONCLUSION

In cases where AOT is associated with an impacted permanent tooth all efforts should be made to use the most conservative approach to manage the tumor and simultaneously preserve the tooth. This strategy is essential to ensure optimal esthetics and functional rehabilitation of the patient. Additionally, it is recommended to maintain regular follow-ups to monitor any potential recurrence.

CONFLICT OF INTEREST STATEMENT

None declared

REFERENCES

1. Reddy Kundoor VK, Maloth KN, Guguloth NN, Kesidi S. Extrafollicular Adenomatoid Odontogenic Tumor: An Unusual Case Presentation. J Dent (Shiraz). 2016 Dec;17(4):370-374.
2. Adisa AO, Lawal AO, Effiom OA, Soyele OO, Omitola OG, Olawuyi A, et al. A retrospective review of 61 cases of adenomatoid odontogenic tumour seen in five tertiary health facilities in Nigeria. Pan Afr Med J. 2016 May 31;24:102.
3. Philipsen HP, Birn H. The adenomatoid odontogenic tumour. Ameloblastic adenomatoid tumour or adeno-ameloblastoma. Acta Pathol Microbiol Scand. 1969;75(3):375-98.
4. de Matos FR, Nonaka CF, Pinto LP, de Souza LB, de Almeida Freitas R. Adenomatoid odontogenic tumor: retrospective study of 15 cases with emphasis on histopathologic features. Head Neck Pathol. 2012 Dec;6(4):430-7.
5. Sadasivan A, Ramesh R, Kurien NM. Peripheral Adenomatoid Odontogenic Tumor - A Rare Cause of Gingival Enlargement: A Case Report with CBCT Findings. Clin Cosmet Investig Dent. 2020 Jul 21;12:297-304.
6. Kalia V, Kalra G, Kaushal N, Sharma V, Vermani M. Maxillary adenomatoid odontogenic tumor associated with a premolar. Ann Maxillofac Surg. 2015 Jan-Jun;5(1):119-22.
7. More CB, Das S, Gupta S, Bhavsar K. Mandibular adenomatoid odontogenic tumor: Radiographic and pathologic correlation. J Nat Sci Biol Med. 2013 Jul;4(2):457-62.
8. Narayanan VS, Naidu G, Ragavendra R, Mhaske-Jedhe S, Haldar M. Adenomatoid odontogenic tumor of the mandible with unusual radiographic features: A case report. Imaging Sci Dent. 2013 Jun;43(2):111-5.
9. Assael LA. Surgical management of odontogenic cysts and tumors. In: Peterson LJ, Indresano TA, Marciani RD, Roser SM, editors. *Principles of oral and maxillofacial surgery.* Lippincott; Philadelphia: 1992. pp. 685–8.
10. Rashmi G, Santosh G, Harshavardhan S, Praveen KM. Adenomatoid odontogenic tumour. Indian J Dent Adv. 2009 Oct 1;1(1):67-72.
11. Assao A, Freitas DL, Oliveira DT. A rare case report of an adenomatoid odontogenic tumor associated with odontoma in the maxilla. Int J Surg Case Rep. 2017;39:245-249.
12. Kamble A, Shimpi MR, Dash JK, Sahoo PK, Chaudhary S, Doiphode M. Adenomatoid Odontogenic Tumor of the Maxilla in a 13-year-old Patient: A Rare Case Report with a Review of Literature. Int J Clin Pediatr Dent. 2021 Jul-Aug;14(4):596-600
13. Erdur EA, Ileri Z, Ugurluoglu C, Cakir M, Dolanmaz D. Eruption of an impacted canine in an adenomatid odontogenic tumor treated with combined orthodontic and surgical therapy. Am J Orthod Dentofacial Orthop. 2016 Jun;149(6):923-7.
14. Bonardi JP, da Costa FH, Matheus RA, Ito FA, Pereira-Stabile CL. Rare presentation of adenomatoid odontogenic tumor in a pediatric patient: a case report. Oral Maxillofac Surg. 2016 Jun;20(2):215-7.
15. Moon JW. Extensive Adenomatoid Odontogenic Tumor of the Maxilla: A Case Report of Conservative Surgical Excision and Orthodontic Alignment of Impacted Canine. Maxillofac Plast Reconstr Surg. 2014 Jul;36(4):173-7.