



Extensive Adenomatoid Odontogenic Tumor of the Maxilla: A Case Report of Conservative Surgical Excision and Orthodontic Alignment of Impacted Canine

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Abstract

The present report describe the surgical therapy, clinical course, orthodontic treatment and morphological characteristics of an adenomatoid odontogenic tumor in the maxilla of an 11-year-old patient. The cystic tumor filled the maxillary sinus and involved a tooth. Marsupialization was accompanied by partial enucleation and applied traction to the affected tooth by a fixed orthodontic appliance. Healing was uneventful and no local recurrence was observed during a 1-year period of follow-up control.

Key words: Adenomatoid odontogenic tumor, Impacted tooth

Introduction

Adenomatoidodontogenic tumor (AOT) is a slow growing lesion, constituting only 2.2% to 7.1% of all odontogenic tumors and 0.1% of jaw tumors, with a predilection for the anterior maxilla of young females in the second decade of life[1-4]. Patients are usually referred to dentists with complaints of impacted teeth within the lesion, usually a canine. The lesion may be associated with the coronal portion of the tooth or extend to the root[1-4].

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. The histogenesis of AOT is controversial.

Some authors describe it as a true benign, non-aggressive, non-invasive neoplasm, while others conceptualize it as a developmental hamartomatous odontogenic growth[1-6].

Generally, AOT is treated by surgical removal. However, large tumors that involve serious loss of bone and thin the bone dangerously are often treated by insertion of a drain or by marsupialization. Marsupialization decreases intracystic pressure and promotes shrinkage of the cyst as well as bone fill[7-9].

The author report a case using surgical and orthodontic approaches to preserve the impacted canine due to extensive AOT.

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Case Report

An 11-year-old boy presented with a swelling of the right side of his face for one month. The patient's medical history was unremarkable. Physical examination showed a painless and circumscribed swelling of the right maxillary, but skin color was normal. Panoramic radiograph demonstrated a well-defined unilocular radiolucent lesion at the apices of the roots of the right maxillary from the lateral incisor to the first molar. Due to the expansive growth there was deviation of the roots of right maxillary lateralis, first and second premolar (Fig. 1A), and of the roots of right mandibular canine and first premolar (Fig. 1B). The clinical impression was benign odontogenic tumor. Based on these features, we considered a differential diagnosis of odontogenic cysts and tumors.

1. Surgical approaches

We took a preventive approach to preserve the developing canine. Therefore, we planned marsupialization of the lesion through the extracted socket of deciduous canine to create a window allowing continuous drainage of the tumor content. We treated the patient by marsupialization under local anesthesia, extracting the deciduous canine

affected by the tumor. We made a pouch-like opening within the tumor. We then enlarged the extraction socket to form a bony window. Care was taken to avoid disturbing the developing tooth buds. During the marsupialization procedure, we took a biopsy sample to confirm the histopathologic diagnosis. Finally, we inserted a vaseline gauze into the cyst cavity to keep it open and to achieve hemostasis.

Antibiotics were administered for seven days to prevent postoperative infection. After one week, we inserted an obturator using prosthetic putty. The putty was replaced every week. A space maintainer was used to maintain the spacing from the adjacent teeth. Panoramic radiographs were taken at three month intervals until the tumor-associated teeth erupted.

After six months, there was further occlusal movement of the developing tooth with reduced radiolucency. The root formation of maxillary canine had continued, and bone formation was evident in the tumor (Fig. 2).

Follow-up examination revealed occlusal movement of the developing tooth bud, but no apparent reduction in

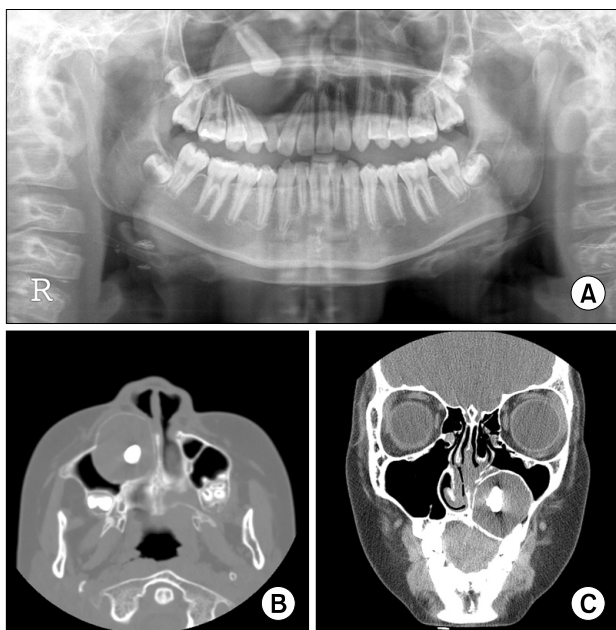


Fig. 1. Initial panoramic radiograph showing a radiolucent lesion associated with a developing left maxillary canine (A), initial paranasal sinus computed tomography axial view (B), coronal view (C).



Fig. 2. Panoramic view showing huge reduction in the radiolucent lesion with further occlusal movement of the tooth and continuation of root formation (6 months postoperative).

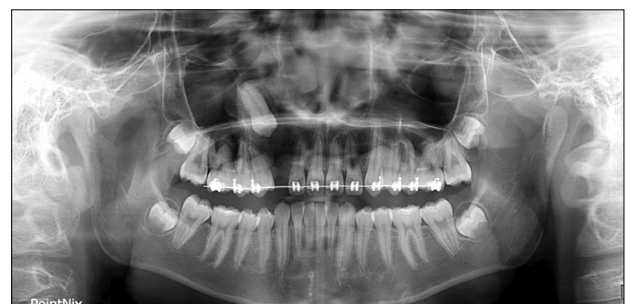


Fig. 3. Panoramic view showing reduced bony defect and further occlusal movement of the tooth as well as continuation of root formation (Bonding of orthodontic button at the time of partial enucleation and curettage).

the radiolucency. So we performed enucleation and curettage, exempting the impacted canine, 12 months after marsupialization and decompression (Fig. 3).

2. Orthodontic approach

Six months after marsupialization, a multibracket appliance was placed in the maxillary arches, and to reinforce anchorage, a Nance appliance was placed. We created space in the canine location. A small orthodontic button, threaded with soft twisted ligature wire of 0.012-inch gauge, was bonded during the partial enucleation and curettage. The ligature was immediately engaged by the Nance appliance. We did follow-up activation every month thereafter. After 20 months of traction, the canine erupted and orthodontic treatment was finished after 37 months. Although we did orthodontic treatment only at the maxillary arch, Class I occlusion was achieved with an acceptable interincisal relationship. The root of the impacted canine

showed normal morphology without root deformation (Fig. 2), but there was decalcification on the labial surface of canine. An electric pulp test was normal (25/60). Optimal intercuspation of teeth was achieved without recurrence (Fig. 4). We continued to observe the patient, and the tumor did not recur as of one year postoperatively (Fig. 5).



Fig. 4. Final panoramic view.

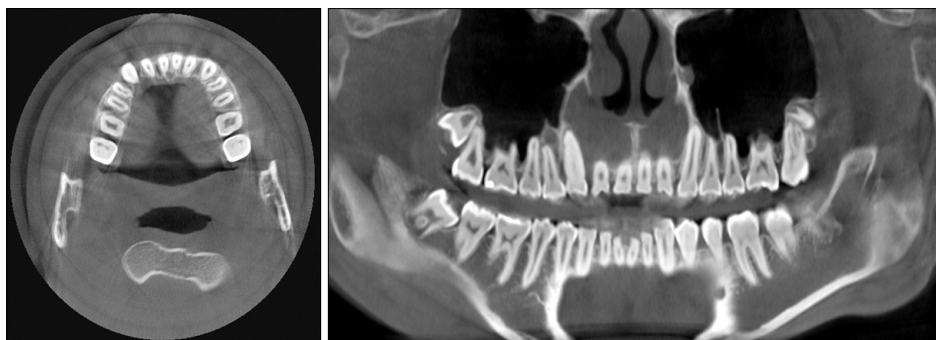


Fig. 5. One year after orthodontic treatment. Cone beam computed tomography view showing successive eruption of maxillary left canine with complete ossification of the bony defect.

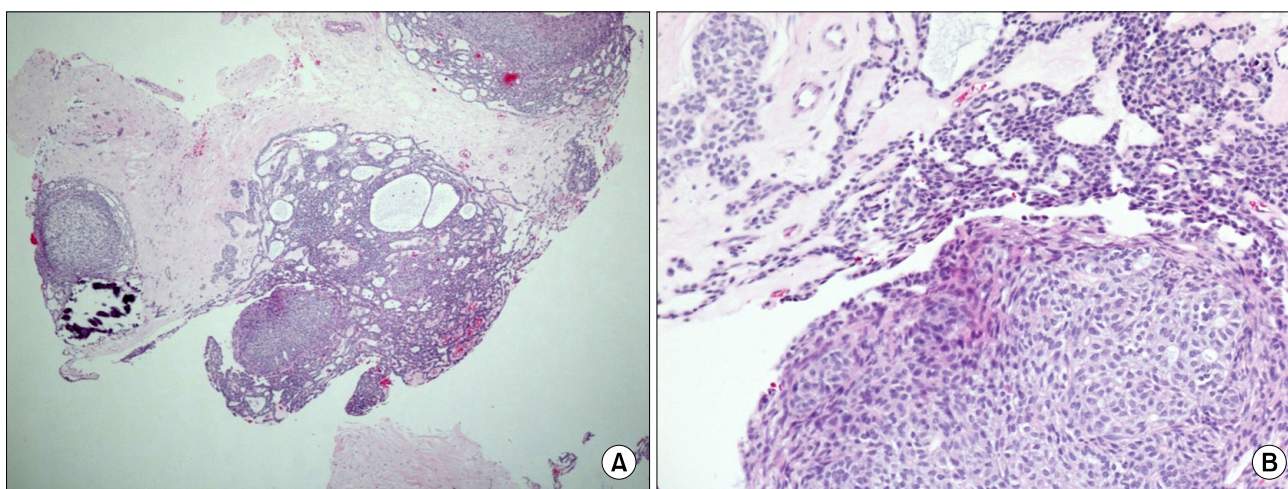


Fig. 6. (A) Tumor with fibrous connective tissue capsule. Nodular aggregates of cells and Duct-like structures (H&E, $\times 40$). (B) Gland-like spaces are surrounded by cuboidal to columnar cells and homogenous zone of hyaline materials and frequent calcified deposits (H&E, $\times 200$).

3. Histologic results

Microscopic examination revealed an extremely vascular encapsulated lesion showing multivariate patterns of cellular arrangements ranging from sheets of polygonal cells arranged in ductal patterns, rosettes to solid sheets of cells. In the center of these ducts, eosinophilic amyloid-like material was also seen. The solid lobular masses showed numerous spindle to columnar hyperchromatic cells with interspersed deposits of eosinophilic hyaline-like material. The above features were consistent with the diagnosis of AOT (Fig. 6A).

At low magnification, the most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue (Fig. 6B).

Discussion

Current convention classifies AOTs into three main types: follicular (or pericoronal), extrafollicular (or extracoronal), and peripheral (or extraosseous/gingival). In a recent retrospective study, 70.8% were of follicular type, as was the present case[1-4]. Radiographically, this lesion usually surrounds an unerupted tooth and is seen as a corticate radiolucency with small radiopacities, but there are cases where the lesion has no radiopaque component, as in our case, and in such a case, a dentigerous cyst is the preferred differential diagnosis. However, an AOT often appears to envelop the crown as well as the root as shown in the picture of this case[1-6].

When deciding treatment methods, the age of patient, anatomical structures, the region of lesion and the size of cyst should be considered. Only a few exceptionally rare cases are reported of recurrence of this tumor. Surgical enucleation or curettage is the usual treatment modality for AOT associated with impacted teeth and/or complete removal of the tooth buds. Marsupialization, decompression, and the Partsch operation is a treatment in which a pouch-like surgical window on the wall of the cyst is incised, the contents of the cyst evacuated, and the continuity between the cyst and oral cavity, maxillary sinus, or nasal cavity is maintained. This technique is usually used for large cysts, because it is more conservative. After

marsupialization, the reduction of tumor size should be carefully monitored, and if needed, immediate surgical removal should be performed[7-11].

Conservative surgical enucleation is the treatment modality of choice. Usually, the tumors do not exceed 1 to 3 cm in greatest diameter, and while usually asymptomatic, may be associated with cortical expansion as in the present case. To the best of our knowledge, this case of large AOT with the sequential surgical approaches and the preservation of impacted tooth is rare in the published literature[1-12].

We performed the orthodontic treatment to bring the involved tooth into the arch for eruption. Traction on an impacted tooth with an immature root often causes root resorption, root deformation, and pulpitis[13]. In the present case, the root length of the impacted canine was less than three-fifth that of a normal root, and the root apex was open. Observation of the impacted tooth is necessary after creating a space for tooth spontaneous eruption. We applied the orthodontic button bonding and orthodontic traction after tooth did not erupt further. Therefore, careful observation and orthodontic treatment during the optimal period is necessary in a growing patient with an impacted tooth and tumor.

It should be emphasized that careful diagnostic procedures and adequate interpretation of clinical and radiographic findings will lead to a correct diagnosis. To preserve the involved tooth, the tumor size and the position of tooth should be carefully considered. The patient should be checked periodically for recurrences over time. Further validation of recurrence of AOT and prognosis of saved tooth needs large clinical studies.

Acknowledgements

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