Eruption of an impacted canine in an adenomatid odontogenic tumor treated with combined orthodontic and surgical therapy

Emire Aybuke Erdur,[a] Zehra Ileri,[b] Ceyhan Ugurluoglu,[c] Mustafa Cakir,[d] and Dogan Dolanmaz[e]
Konya and Manisa, Turkey

An adenomatoid odontogenic tumor is an uncommon asymptomatic lesion that is often misdiagnosed as a dentigerous cyst. It originates from the odontogenic epithelium. Enucleation and curettage is the usual treatment of choice. Marsupialization may be attempted instead of extraction of the impacted tooth, since it provides an opportunity for tooth eruption. This case report is the first to report on the eruption of an impacted canine in an adenomatoid odontogenic tumor treated with combined orthodontics and marsupialization. The impacted canine erupted uneventfully, with no evidence of recurrence 3 years after the treatment. (Am J Orthod Dentofacial Orthop 2016;149:923-7)

An adenomatoid odontogenic tumor is a relatively rare benign lesion.[1] Although many terms have been used to describe this disease, the World Health Organization named it "adenomatoid odontogenic tumor" in 1971.[2] It is most frequently observed in the anterior maxilla, followed by the anterior mandible.[3] Although there are some reported differences, the female-to-male ratio is about 2:1 in all age groups and for all variants. Generally, the disease occurs in the second decade of life.[2,4] Clinically, an adenomatoid odontogenic tumor resembles a dentigerous cyst or an ameloblastoma. But unlike an ameloblastoma, an adenomatoid odontogenic tumor is a benign, painless, noninvasive, slow-growing tumor that does not infiltrate bone.[5] It is completely radiolucent and mimics a dentigerous cyst in growth pattern and appearance. However, it often appears to envelop both the crown and roots of a tooth, unlike a dentigerous cyst, which does not envelop the roots.[6] A histopathologic examination is the most important assessment in the differential diagnosis.

Because of the slow growth and asymptomatic nature of the lesion, patients are usually diagnosed incidentally. An adenomatoid odontogenic tumor is generally smaller than 3 cm, with a solid or cystic structure, but rarely can grow to a large size.[7] Three types of adenomatoid odontogenic tumors have been described—follicular, extrafollicular, and peripheral—with the follicular type being the most common.[8,9]

Surgical enucleation and curettage is the standard treatment of choice because of the encapsulated, benign, noninvasive, and nonaggressive characteristics of the lesion.[10] During this procedure, the impacted tooth is usually excised with the lesion. The reported results for almost all adenomatoid odontogenic tumors treated with enucleation are good, and recurrence is rare. This article presents a patient with an impacted permanent mandibular canine, which was diagnosed as an adenomatoid odontogenic tumor after histopathologic examination, and was treated with marsupialization.

CASE REPORT

A 12-year-old girl consulted the Department of Orthodontics at Selcuk University in Turkey with the chief complaint of an unesthetic dental appearance. Clinically, she had a well-balanced facial profile and Class II molar and canine relationships on both sides, with maxillary

[a]Specialist dentist (orthodontist), Department of Orthodontics, Faculty of Dentistry, Selcuk University, Konya, Turkey.
[b]Assistant professor, Department of Orthodontics, Faculty of Dentistry, Selcuk University, Konya, Turkey.
[c]Assistant professor, Department of Pathology, Faculty of Medicine, Selcuk University, Konya, Turkey.
[d]Specialist dentist (maxillofacial surgeon), Department of Oral and Maxillofacial Surgery, Manisa Oral and Dental State Hospital, Manisa, Turkey.
[e]Professor, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Selcuk University, Konya, Turkey
All authors have completed and submitted the ICMJE Form for Disclosure of Potential Conflicts of Interest, and none were reported.
Address correspondence to: Emire Aybuke Erdur, Faculty of Dentistry, Department of Orthodontics, Selcuk University, Konya, Turkey 42075; e-mail, dtaybuke@gmail.com.
Submitted, June 2015; revised and accepted, February 2016.
0889-5406/$36.00

http://dx.doi.org/10.1016/j.ajodo.2016.02.011

Fig 1. Pretreatment facial and intraoral photographs.

anterior crowding, increased overbite, and mandibular dental midline deviation (Fig 1). She had a mandibular left deciduous canine and painless swelling in the mandibular left vestibule (Fig 2, *A*). Cephalometric and panoramic radiographs and dental casts were obtained. Radiologically, her permanent canine was an impacted, well-circumscribed radiolucency that looked like a cystic lesion that started from the middle of the root and surrounded the crown completely with a well-defined corticated border. There was migration of the neighboring teeth. The swelling extended from the left lateral incisor to the left premolar both buccally and lingually. On palpation, its consistency was firm. Based on the clinical and radiographic findings, an adenomatoid odontogenic tumor or a dentigerous cyst was considered the primary diagnosis.

In consultation with the Department of Oral and Maxillofacial Surgery at our university, we attempted to allow the tooth to erupt with marsupialization. The mandibular deciduous left canine was extracted under an inferior alveolar nerve block. Then, a mucoperiosteal flap was elevated, the bone window was extended, and the upper part of the lesion was excised from the bone window border. A biopsy sample was taken and sent for histopathologic examination. After the wound was irrigated with saline solution, the crown of the impacted canine was observed in the cavity. The cavity was filled with antibiotic gauze, and then the flap was sutured in the primary position. The patient was recalled to irrigate the cavity with saline solution and to replace the antibiotic gauze in the socket every 2 days. After 1 week, an impression model was obtained to design an obturator for a cavity, which the patient wore for nearly 4 months. Her parents were taught to clean the cavity every day with saline solution and an injector.

The histopathologic examination of the biopsy showed an adenomatoid odontogenic tumor. Well-circumscribed proliferation of the duct-like epithelium, which had a rosette formation with calcification, was observed in the center. Columnar epithelial cells with

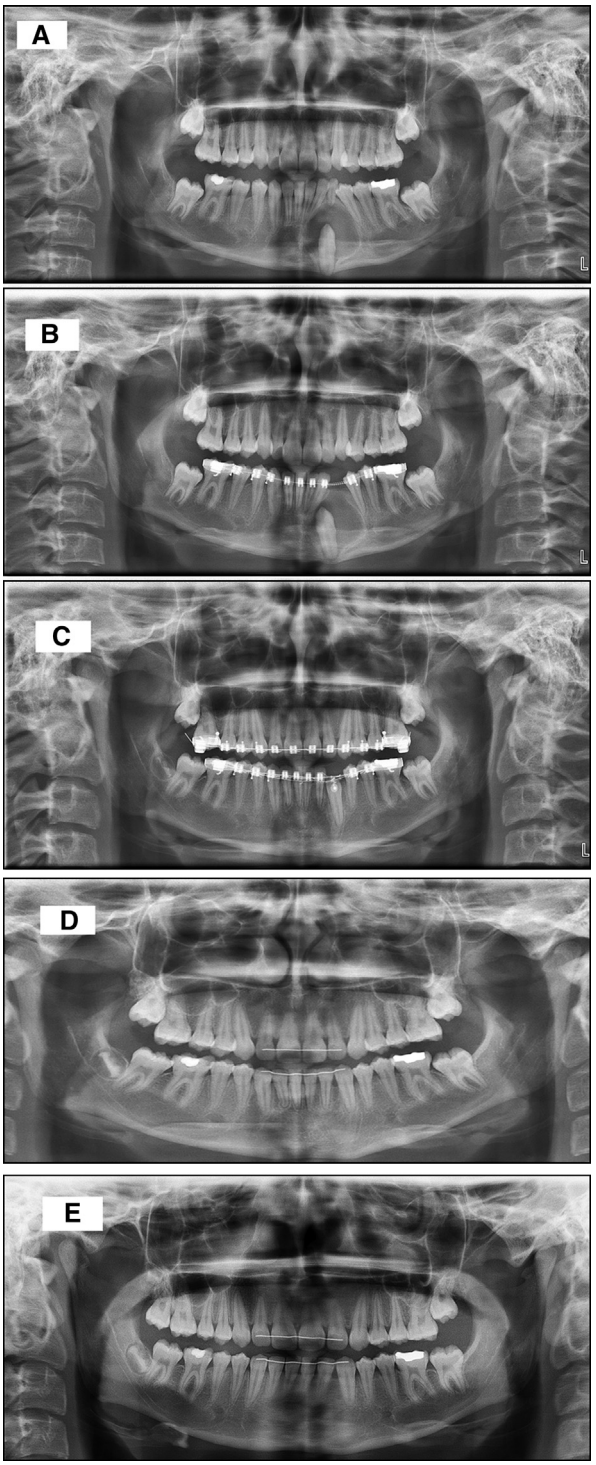

Fig 2. Panoramic radiographs: **A,** pretreatment; **B,** after surgery; **C,** 6 months after orthodontic button was bonded; **D,** posttreatment; **E,** 3 years after the surgery.

basal nuclei and clear cytoplasm resembled preameloblasts. Eosinophilic fibrillar material was present between the tumor cells and in the duct-like structures.

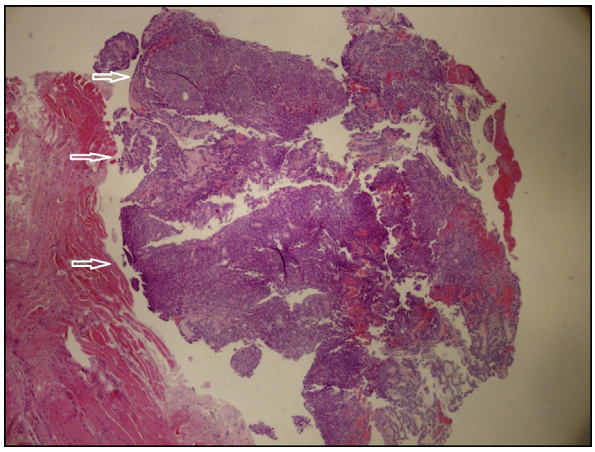

Fig 3. Histopathologic appearence of well-circumscribed proliferation of duct-like epithelium (*arrows*).

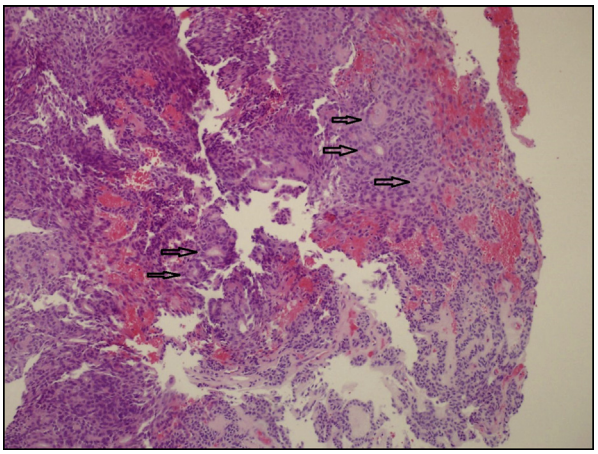

Fig 4. Histopathologic appearence of columnar epithelial cells with basal nuclei and clear cytoplasm resembling preameloblasts (*arrows*). Eosinophilic fibrillar material was present between the tumor cells and in the duct-like structures.

Rare mitoses, no necrosis, and no epithelial atypia were detected (Figs 3 and 4).

The patient was recalled to the Department of Orthodontics once a month. As the tooth continued to erupt, the obturator was trimmed. Nearly 2 months later, the mandibular arch was bonded to open sufficient space because the impacted canine had erupted toward the lateral incisor root (Fig 2, *B*). Six months later, an orthodontic button was bonded to the canine to provide traction into the oral cavity. One month later, the maxillary arch was bonded, and the teeth were leveled. The impacted canine was examined with panoramic radiographs every 3 months to evaluate the level of eruption (Fig 2, *C*). The mandibular left canine erupted without

Fig 5. Posttreatment facial and intraoral photographs.

root resorption (Fig 2, *D*). After the fixed orthodontic treatment was finished, a functional and esthetic dentition was obtained (Fig 5). There was no evidence of tumor recurrence and no evidence of root resorption in the neighboring teeth 3 years after the operation (Figs 2, *E*, and 6).

DISCUSSION

An adenomatoid odontogenic tumor is an uncommon benign lesion that originates from the odontogenic epithelium. These constitute 3% of all odontogenic tumors and 0.1% of all jaw tumors.[11] The most common symptoms of adenomatoid odontogenic tumors are painless swelling and delay in the eruption of the permanent teeth. Furthermore, the lesion can cause a slight displacement of impacted and adjacent teeth, and it is usually misdiagnosed clinically as a cyst.[7] Sometimes, more than 1 tooth is affected, and the adenomatoid odontogenic tumor can reach a sufficiently large size

Fig 6. Posttreatment periapical radiographs.

to cause cortical bone expansion, tooth displacement, and swelling that can cause facial asymmetry.[12,13]

Radiographically, the adenomatoid odontogenic tumor usually surrounds an impacted tooth, enveloping the crown as well as the root, and is seen as a corticated radiolucency with small radiopacities.[1,7] Regezi et al[9] stated that the radiolucent area around the impacted tooth at the level of the root is a distinct finding of an adenomatoid odontogenic tumor. In 39 patients with adenomatoid odontogenic tumors, Leon et al[14] observed a unilocular radiolucency with well-defined borders associated with the tooth crown. Calcification and cystic areas were found in more than half of the patients. The authors also mentioned that the clinical and radiologic findings of the adenomatoid odontogenic tumor mimicked those of a dentigerous cyst. In our patient, the well-circumscribed radiolucent area around the impacted tooth was at the midlevel of the root and surrounded the crown completely with a well-defined corticated border. Initially, our differential diagnosis was an adenomatoid odontogenic tumor or a dentigerous cyst.

The management of a dentigerous cyst includes enucleation and marsupialization,[15] whereas an adenomatoid odontogenic tumor is treated with enucleation and curettage. An adenomatoid odontogenic tumor does not need extensive and aggressive surgery because it has a benign, nonaggressive growth pattern, and its capsule is easily separated from the surrounding bone. The reported recurrence rate is low.[8] Because of the similar characteristics of these lesions, the patient's esthetic expectations and age, a favorable position for orthodontic movement, sufficient space for eruption of the tooth, and the low recurrence rate of adenomatoid odontogenic tumors, we chose marsupialization for treatment.

To the best of our knowledge, marsupialization has never been used as a treatment option for an adenomatoid odontogenic tumor. Marsupialization is a more conservative treatment modality than enucleation and is considered first-line therapy, especially in the initial treatment of benign cystic lesions during the mixed dentition period. This permits decompression of the cyst, reducing the extent of the bone defect, and has the advantage of promoting the eruption of a cyst-associated tooth.[15] Orthodontic traction is often performed on the impacted tooth after marsupialization to guide the impacted tooth. Depending on the size of the lesion, the position of the tooth, and the available space, the tooth can erupt spontaneously.

Marsupialization was selected for treatment after consulting with the patient. This conservative technique enabled eruption of the impacted canine with orthodontic treatment. In this way, the functional and esthetic features were ensured. At the 3-year follow-up, there had been no recurrence of the adenomatoid odontogenic tumor.

CONCLUSIONS

This case shows that marsupialization can be an effective option for the treatment of an adenomatoid odontogenic tumor, and it allowed for eruption of the impacted tooth. Further prospective studies with more patients are needed before recommending marsupialization as a treatment option for adenomatoid odontogenic tumor.

REFERENCES

1. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: a case report. J Oral Maxillofac Surg 2000;58:1161-4.
2. Philipsen HP, Reichart PA. Adenomatoid odontogenic tumour: facts and figures. Oral Oncol 1999;35:125-31.
3. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumor of the mandible: review of the literature and report of a rare case. Head Face Med 2005;1:3.
4. Courtney RM, Kerr DA. The odontogenic adenomatoid tumor. A comprehensive study of twenty new cases. Oral Surg Oral Med Oral Pathol 1975;39:424-35.
5. Garg D, Palaskar S, Shetty VP, Bhushan A. Adenomatoid odontogenic tumor—hamartoma or true neoplasm: a case report. J Oral Sci 2009;51:155-9.
6. Rashmi G, Santosh G, Harshavardhan S, Praveen KM. Adenomatoid odontogenic tumour. Indian J Dent Adv 2009;1:67-71.
7. John JB, John RR. Adenomatoid odontogenic tumor associated with dentigerous cyst in posterior maxilla: a case report and review of literature. J Oral Maxillofac Pathol 2010;14:59-62.
8. Philipsen HP, Reichart PA, Zhang KH, Nijai AH, Yu QX. Adenomatoid odontogenic tumor: biologic profile based on 499 cases. J Oral Pathol Med 1991;20:149-58.
9. Regezi JA, Kerr DA, Courtney RM. Odontogenic tumors: analysis of 706 cases. J Oral Surg 1978;36:771-8.
10. Assael L. Surgical management of odontogenic cysts and tumors. In: Peterson LJ, editor. Principles of oral and maxillafacial surgery. Philadelphia: Lippincott; 1992. p. 698-701.
11. Gold L. Odontogenic tumors: surgical pathology and management. In: Fonseca RJ, editor. Oral and maxillofacial surgery. Philadelphia: W. B. Saunders; 2000. p. 328-30.
12. Manjunatha BS, Mahajan A, Mody BM, Shah V. Adenomatoid odontogenic tumor (AOT) arising from a dentigerous cyst: literature review and report of a case. J Maxillofac Oral Surg 2015;14:393-7.
13. Baskaran P, Misra S, Kumar MS, Mithra R. Adenomatoid odontogenic tumor–a report of two cases with histopathology correlation. J Clin Imaging Sci 2011;1:64.
14. Leon JE, Mata GM, Fregnani ER, Carlos-Bregni R, de Almeida OP, Mosqueda-Taylor A, et al. Clinicopathological and immunohistochemical study of 39 cases of adenomatoid odontogenic tumour: a multicentric study. Oral Oncol 2005;41:835-42.
15. Miyawaki S, Hyomoto M, Tsubouchi J, Kirita T, Sugimura M. Eruption speed and rate of angulation change of a cyst-associated mandibular second premolar after marsupialization of a dentigerous cyst. Am J Orthod Dentofacial Orthop 1999;116:578-84.