Letters to the editor*

Adenomatoid odontogenic tumor: An outdated report

We read with some surprise a clinical report by Erdur et al[1] in the June 2016 issue of the *AJO-DO*. The authors presented a textbook case of adenomatoid odontogenic tumor (AOT), claiming that "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." Since this therapy for AOT is both well documented and well established, it is difficult to accept this statement. The authors appeared to be unaware of the many previous reports documenting active orthodontic therapy for AOT after surgical exposure, a representative sample of which are presented chronologically in our reference list.[2-7] Although these English-language papers would have appeared in most computerized literature searches, oddly, all seem to have escaped the attention of Erdur et al.

The authors also concluded that marsupialization has never been used as a treatment option for an AOT, and further prospective studies with more patients are needed before recommending marsupialization as a treatment option for AOT. It has long been recognized that subtotal excision (marsupialization) can be successfully applied for the treatment of AOT.[8] In the previously reported cases, this conservative procedure resulted in complete bone healing and facilitated spontaneous eruption of the impacted tooth or teeth.[9-13] The third series of the Armed Forces Institute of Pathology atlas written 15 years ago stated that "in appropriate circumstances, it may be possible to preserve the involved tooth."[14] This view has been reiterated in more contemporary standard texts.[8,15,16] Of additional interest was a report of reimplantation of a developing AOT-related tooth,[17] although this is not a preferred treatment.[8] According to that report, no tumor recurrence was evident 4 years after surgery, and the involved mandibular first molar continued to erupt spontaneously, showing completion of root formation and reaching the occlusal plane.[17] It is now 50 years since Philipsen and Birn[2] first reported a case of AOT treated by marsupialization and orthodontic treatment. This type of combined therapy has since become popularized[3-7] and is now accepted worldwide.[18-20] The present single case report should merely serve to confirm and validate the above studies, one of which, previously unrecognized by the authors, was published with a comparable title in the *AJO-DO*.[4]

In summary, the steady stream of reports on orthodontic-guided eruption of the tooth or teeth associated with AOT conclusively indicates that the treatment modality described by Erdur et al[1] is nothing new and in fact is rather standard. Furthermore, the term AOT was first coined by Philipsen and Birn[2] in 1969 and not by the World Health Organization in 1971.[20] It is always prudent to avoid claims of first description if authors are not sufficiently familiar with the work of experts in the field. In addition to our surprise that the authors' survey of the AOT literature was insufficient, we are puzzled that reviewers for this reputable journal who accepted the article for publication were unaware that the reported case was unremarkable and the authors' claim quite without merit.

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Authors' response

Thank you for giving us the chance to respond to Ide and colleagues' comments about our article, "Eruption of an impacted canine in an adenomatoid odontogenic tumor treated with combined orthodontic and surgical therapy." These comments on our research made us search for more data and further clarify some aspects of our research.

Most of the articles on adenomatoid odontogenic tumor (AOT) are case reports, and research studies are fewer. Although there is no consensus for the treatment of AOT, the most common treatment option is enucleation and curettage.[1-7] We read the articles that Ide et al cited in their letter, and we have seen that in these documented orthodontic treatments for AOT, the authors preferred total excision of the tumor and preserved the teeth.[2-12] Ide et al also mentioned a report of subtotal excision (marsupialization) with eruption of the teeth.[13] The subtotal excision they mentioned is not true marsupialization. In textbooks, the marsupialization process is described as creating a surgical window in the wall of the cyst, draining the contents, and maintaining continuity between the cyst and the oral cavity.[14] The remaining cystic epithelium is left in situ. This procedure decreases cystic pressure and promotes contraction of the cyst and bone fill.

The marsupialization that we wanted to describe as a treatment option was different.[13] We opened a bone window on the upper side of the tumor. The upper part of the lesion was excised from the bone window border, the inside of the lesion was irrigated, and after recognizing the crown of the teeth, surrounding area was filled with antibiotic gauze. The socket was irrigated with saline solution, and the antibiotic gauze was replaced every other day for 1 week. An obturator was inserted in the window of the lesion, and irrigation was done for 4 months. This technique is usually used for benign cystic lesions.[15]

Although our claim that this case report is the first to report on the eruption of an impacted canine in an AOT treated with combined orthodontics and marsupialization might be controversial, we believe it is not wrong. We think that the marsupialization technique that is used for benign cystic lesions with protection of the involved teeth can be a treatment option for AOT.

Various terms have been used before to describe this tumor, including adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma admentinum, and teratmatous odontoma.[16] In 1971, the World Health Organization made the classification of odontogenic tumors and adopted "AOT" as the term for this tumor, which was coined before.[2]

As a result, our success may pave the way for marsupialization for the management of AOT with orthodontic treatment. Further clinical studies are necessary to validate its clinical indications for the management of AOT treatment.

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