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Case Report

Adenomatoid Odontogenic Tumor. Case Report

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Abstract

The adenomatoid odontogenic tumor (AOT) occurs in low frequency and originates from epithelial remnants, specifically from the dental lamina, and shows characteristics similar to a wide variety of pathologies. It is a benign non-invasive lesion that can be managed with conservative surgical treatment such as enucleation and curettage. Therefore, it is important to recognize it and differentiate it from other tumors that required more aggressive surgical interventions for treatment.

We present an unusual symptomatic case that affects the posterior region of the mandible in a 25-year-old woman with clinical, radiographic and histopathological characteristics. Clinical examination showed labial and lingual bony expansion in the body part of the mandible on the left side. Radiographic examination revealed a lesion with radiolucent characteristics. Not related to any retained tooth. The diagnosis was obtained after a confirmatory intraosseous biopsy with histopathological examination and was diagnosed with (TOA). Tumor enucleation was performed with excellent functional and aesthetic results.

Key words: *Adenomatoid odontogenic tumor (AOT), cyst benign oral tumors, enucleation, surgical treatment, histopathologist.*

Introduction

Maxillofacial surgeons encounter a large number of patients in their practices on a daily basis; each case presents a unique challenge for the specialty—particularly when dealing with tumors and cysts of the jaws. These conditions demand great dexterity and experience, largely due to the inherent difficulty of establishing a differential diagnosis given their striking clinical similarities. This challenge is especially pronounced when these lesions manifest as radiolucent images, as this radiographic appearance is shared by a wide spectrum of lesions. In this context, the pathologist plays a pivotal role in establishing the definitive diagnosis, upon which the subsequent management and treatment of these conditions depend (1,2,3,4).

Odontogenic tumors (OTs) constitute a heterogeneous group of rare lesions affecting the bony tissues of the maxilla and mandible. They originate from the mesenchymal and/or epithelial components involved in odontogenesis (tooth formation).

OTs are lesions that exhibit a wide variety of clinical manifestations and histopathological characteristics, ranging in nature from hamartomas to malignant neoplasms (1,2,5,6,7,8).

The Adenomatoid Odontogenic Tumor (AOT) was first described over 100 years ago by Harbitz, who termed it a "cystic adamantoma." Since then, it has been referred to by numerous names; however, in 1969, the World Health Organization (WHO) officially adopted the term "Adenomatoid Odontogenic Tumor," as proposed by Philipsen and Birn (3).

The AOT is the fourth most common type of odontogenic tumor, accounting for 2–7% of all cases (5,6,9,10,11). It arises from odontogenic epithelium, suggesting that its origin may lie within the enamel organ, reduced enamel epithelium, the rests of Malassez, or the dental lamina remnants—the latter being the most widely accepted theory (1,2,7). Consequently, the classification of this pathology—specifically whether it should be regarded as a hamartoma or a true neoplasm—remains a subject of considerable controversy. It is a slow-growing benign neoplasm, although many authors categorize it as a hamartoma given its characteristics (8, 10, 11, 12, 13, 14, 15). It presents as an extra- and intraoral enlargement that is asymptomatic and non-invasive, and which has frequently been misdiagnosed as an odontogenic cyst.

Some authors refer to the AOT as the "two-thirds tumor" because two-thirds of cases present in the anterior maxillary region, occur in women, and are associated with an impacted tooth—of which two-thirds are canines; furthermore, two-thirds of the intraosseous-type lesions exhibit radiopacities within the lesion itself. Dental resorption is rare; however, cortical expansion and displacement of adjacent teeth are regularly observed (3,4,8,9,10). Three types are distinguished based on location: follicular and extrafollicular—which are differentiated radiographically—and peripheral, which is observed only clinically (3,4,5,10,11). Differential diagnosis from other pathologies, such as ameloblastoma and keratocyst, is crucial due to the aggressive treatment these conditions require. Conservative treatment is indicated for the AOT given its low recurrence rate (2,5,6,10,13,14,15).

The objective of this article is to describe the pre-, intra-, and postoperative management of a female patient with a mandibular Adenomatoid Odontogenic Tumor, treated at Princess Marina Hospital in the city of Gaborone, Republic of Botswana.

Case Presentation

A 25-year-old Black female patient—a housewife with a history of good health—presented to the Maxillofacial Surgery clinic at Princess Marina Hospital in Gaborone, Republic of Botswana. She was attended to by Cuban medical staff after being referred from a primary dental care facility in the city of Lobatse due to a swelling in the region of the left mandibular body. This swelling had previously been treated with antibiotics based on a suspicion of odontogenic facial cellulitis, as the first and second molars in that area exhibited Grade IV mobility. The patient reports that she began to notice swelling in the mandibular region of

her face in 2021; this swelling grew slowly until it eventually caused expansion of the buccal cortical bone (Figure 1). She states that she received care at the dental clinic within her local health district until a lateral radiograph was ordered (Figure 3A); this revealed a radiolucent area situated between the left lower first and second molars, leading to her referral to our service for specialized care.

A physical examination and a new imaging study (Computed Tomography) were performed (Figure 2), revealing a well-defined, rounded radiolucent pattern with a radiopaque cortex. Furthermore, separation of the roots of both molars was observed, without any evidence of root resorption. It was decided to perform an incisional biopsy under local anesthesia (Figure 3B, preoperative view), which confirmed the diagnosis of an Adenomatoid Odontogenic Tumor with no signs of malignancy. Complementary Examinations

Complementary laboratory tests were ordered (Hemoglobin: 15.5 g/dL; Leukocytes: 6.2 × 10^3/µL; Platelets: 245 × 10^3/µL; Blood Type: O Positive; Rapid HIV Test [ELISA]; Blood Glucose: 3.55 mmol/L; COVID-19 Test: Negative [PCR]).

The patient was taken to the operating room, where, under nasotracheal general anesthesia, enucleation of the lesion was performed, along with the extraction of the mobile molars associated with the tumor (Figure 3).

The surgical specimen was sent to the Department of Anatomical Pathology at Princess Marina Hospital.

The postoperative histopathological examination results were consistent with the preoperative findings.

Treatment with analgesics, antibiotics, and anti-inflammatory medications was administered

The patient remained hospitalized for three days; sutures were removed one week later. Subsequent monthly follow-up visits were conducted for six months. At the one-year postoperative mark, the patient was discharged, having achieved a full recovery; she was instructed to return to our service immediately should any signs of recurrence appear.

Figura 1 A & B. Preoperatorio extra e intraoral

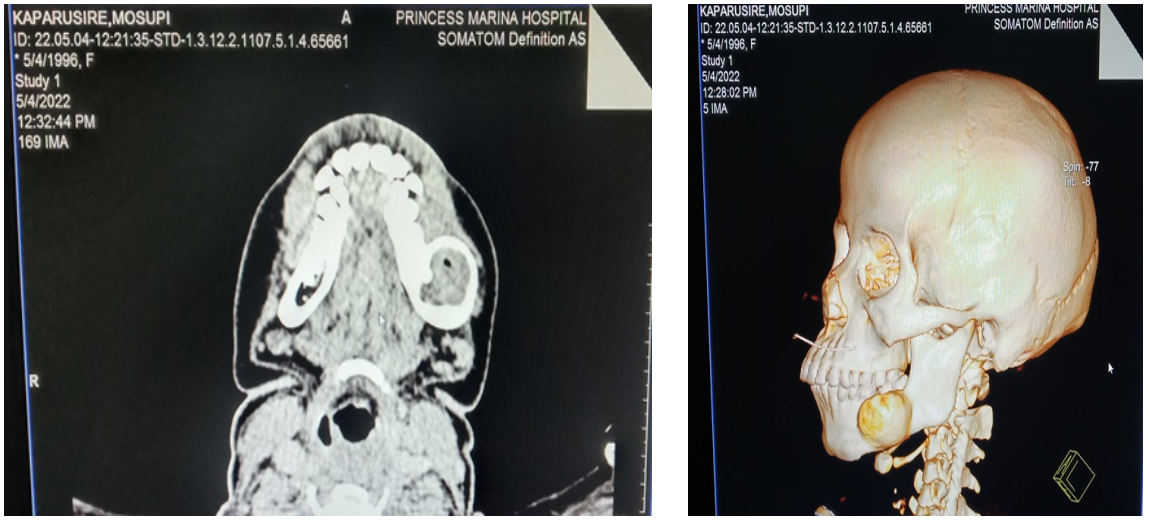

Figure 2 A & B. Imágenes obtenidas en la Tomografia

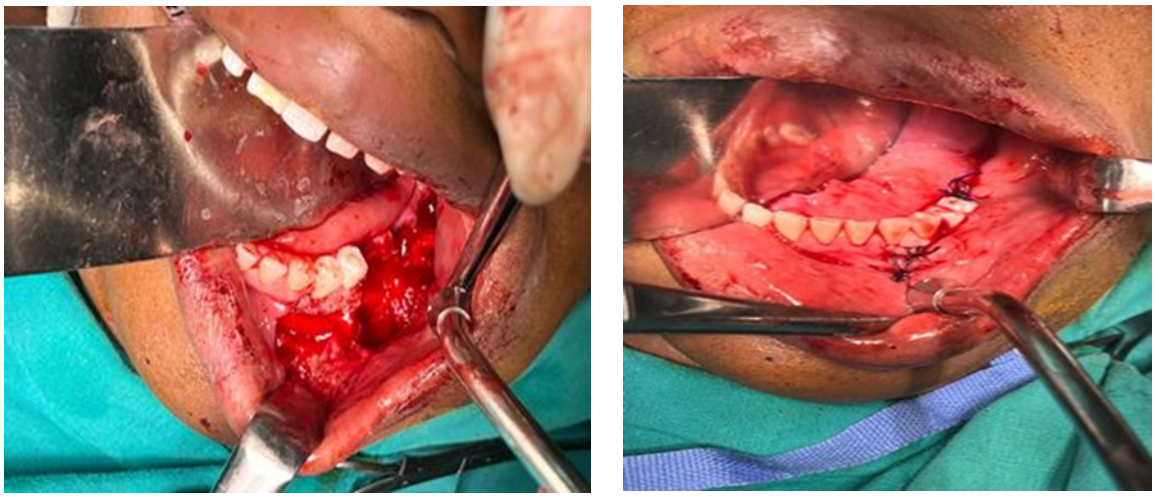

Figura 3. Tratamiento quirurgico (enucleacion).

Histopathology

Microscopically, several grayish-brown tissue fragments were examined, measuring a combined 3.5 × 2.12 cm. They revealed solid areas of spindle-shaped epithelial cells arranged in a swirling pattern, as well as scattered ductal structures composed of circumferentially arranged columnar cells containing amorphous eosinophilic material (Figure 4 A, B).

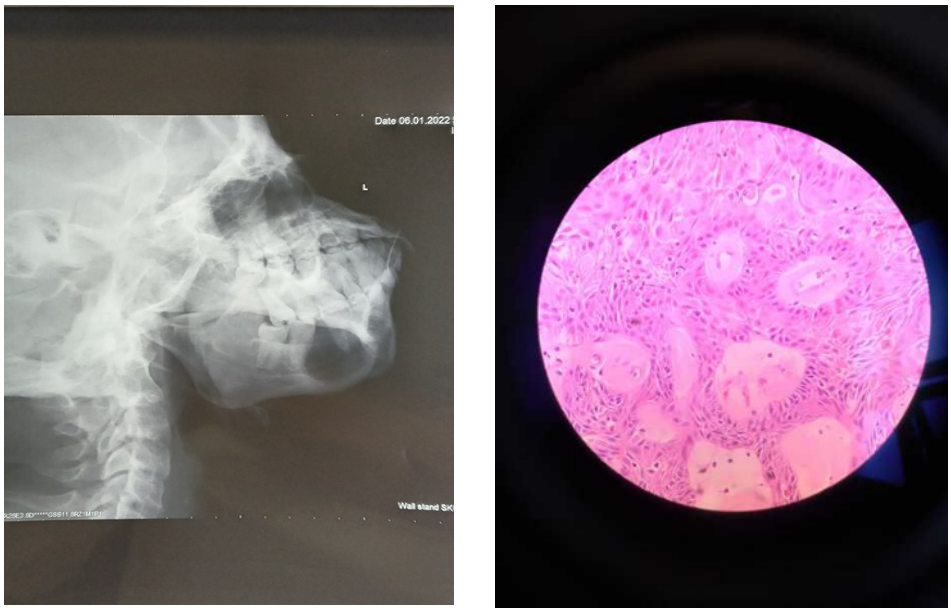

Figure 4. Estudios radiografico (A) e histopatologicos.(B).

Discussion

The AOT is a highly variable entity; it has not been possible to definitively determine whether it constitutes a true neoplasm or a hamartoma, while other authors even regard it as an odontogenic cyst. Ahire et al. (3) argue that the AOT should not be classified as a cyst, and its true cystic nature remains questionable. Clarifying this point would require further studies on its histogenesis—research that could potentially lead to a change in nomenclature from AOT to Adenomatoid Odontogenic Cyst (AOC), given that, in terms of its origin, it is not a true cyst. If the AOT were indeed a true cyst, one would expect to encounter peripheral and extrafollicular cystic variants; however, only one such case has ever been reported—specifically, by Dwivedi (7). This entity exhibits irregular behavior, presenting a mixture of characteristics typically associated with other distinct entities at the clinical, radiographic, and histopathological levels; consequently, a thorough preoperative evaluation is of paramount importance, as the choice of therapeutic approach depends directly upon it.

There are case reports (1, 4, 5, 6, 10) demonstrating, at the histopathological level, a mixture of features characteristic of both an odontoma and an AOT. The authors conducted a systematic review of all published cases containing components of both AOT and dental tissue, raising the question of whether these represent a single entity or two distinct lesions. This inquiry stems from the fact that the AOT is typically characterized by its occurrence in the anterior maxillary region—usually involving a canine tooth—whereas these mixed lesions containing dental tissue elements tend to present in the posterior mandibular region. In the review presented by this author, only 1 out of 11 reported cases was located in the maxilla, while the remainder were

found in the mandible (3, 4, 6, 9)—a distribution pattern that deviates from the typical presentation of an AOT. The TOA bears a strong resemblance to a keratocyst and a unicystic ameloblastoma—particularly if the TOA is situated in a pericoronal location. This was, in fact, our initial impression regarding our patient during her first consultation; however, we consistently maintained that the lesion was neither associated with the crown of an impacted tooth nor had it caused root resorption of adjacent teeth—two highly useful data points for differential diagnosis. Both of these lesions are typically diagnosed between the second and third decades of life—similar to the TOA—yet they occur more commonly in the posterior region of the mandible (5, 6, 10, 12, 15), in contrast to the TOA. Another lesion that must be differentiated from the TOA is the calcifying epithelial odontogenic tumor (CEOT); this lesion is also found in the anterior region of the jaws, is associated with an unerupted tooth, may contain radiopacities, and is likewise diagnosed during the second decade of life. Consequently, a variety of similarities exist between the two, making their differentiation challenging (2, 7, 10). The TOA may also resemble an ameloblastic fibro-odontoma; while both lesions are primarily located in the posterior mandible, the fibro-odontoma is seen predominantly in children around the age of 10 (6, 7, 9). Given its benign behavior, slow growth, and clear demarcation—as well as its low propensity for recurrence—the elective treatment is simple enucleation and curettage. However, in exceptional cases involving a large tumor size or a risk of bone fracture, partial *en bloc* resection of the mandible or anterior maxilla has been indicated (2, 5, 6, 7, 11, 14, 15). Furthermore, the use of lyophilized bone and tissue regeneration guides has been recommended in cases where surgical removal has left a large bone cavity exposed (6, 13). Rarely does this tumor exhibit aggressive behavior, as described by Shaikh (13) in a report detailing a case involving extensive destruction of the mandibular bone, which necessitated a wide resection of the mandible. The patient treated at our facility experienced an excellent postoperative course, confirming that enucleation is an effective treatment modality for this type of tumor. Espinosa (2), Camara (5), Edur (8), Costa (14), and Marin (15) report cases very similar to ours, in which tumor enucleation proved highly effective, with no subsequent recurrences.

Structurally, the TOA is a well-circumscribed neoplastic lesion derived from odontogenic epithelium. It typically manifests—at least in the follicular variant, such as the one presented here—surrounding the crowns of unerupted anterior teeth in young patients. Its histological configuration consists of epithelium arranged in swirling nests, alongside areas exhibiting ductal or glandular patterns, interspersed with occasional spherical calcifications (1, 2, 4, 6, 10). These findings correspond to the observations made in our patient and proved highly instrumental in guiding her clinical management.

It is crucial to remain cognizant of the various clinical presentations of this tumor and to establish an accurate differential diagnosis; failure to do so—given the clinical similarities this lesion shares with other pathologies—may lead to inappropriate treatment. The work of the anatomical pathology specialist plays a

crucial role in determining the course of action to be taken regarding this tumor—as was the case in our instance, where a detailed and meticulous histopathological study was performed both pre- and postoperatively.

Conclusion

It is concluded that a comprehensive preoperative physical, imaging, and histopathological examination is indispensable to achieve satisfactory results in the surgical treatment of this type of tumor, given its strong resemblance to other neoplastic lesions. The patient achieved an excellent aesthetic and functional outcome—reflecting her high degree of satisfaction—which serves to underscore the professionalism of Cuban medical personnel on the international stage.

Conflicts of Interest.

The authors declare no conflicts of interest.

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