

Adenomatoid Odontogenic Tumor. Report of a Case

Paola Peserico-Dal Farra^{1*}, Antonio Gagliardi-Lugo², Edgar Cabrera³ and Mariana Villarroel-Dorrego⁴

¹Resident of the Third Year of the Postgraduate Course in Oral and Maxillofacial Surgery of the Gran Mariscal University of Ayacucho in the General Hospital of the West Dr. José Gregorio Hernández, Caracas, Venezuela

²Specialist in Oral and Maxillofacial Surgery of the Postgraduate Course of Oral and Maxillofacial Surgery of the Gran Mariscal University of Ayacucho in the General Hospital of the West Dr. José Gregorio Hernández, Caracas, Venezuela

³Specialist and Deputy of the Postgraduate Course in Oral and Maxillofacial Surgery at the Gran Mariscal de Ayacucho University, Chief of Oral and Maxillofacial Surgery, General Hospital of the West Dr. José Gregorio Hernández, Caracas, Venezuela

⁴Specialist in Pathological Anatomy and Assistant of the Postgraduate Course in Oral and Maxillofacial Surgery at the Gran Mariscal de Ayacucho University, Chief of Oral and Maxillofacial Surgery, General Hospital of the West Dr. José Gregorio Hernández, Caracas, Venezuela

***Corresponding Author:** Paola Peserico-Dal Farra, Resident of the Third Year of the Postgraduate Course in Oral and Maxillofacial Surgery of the Gran Mariscal University of Ayacucho in the General Hospital of the West Dr. José Gregorio Hernández, Caracas, Venezuela.

Received: October 26, 2017; **Published:** December 05, 2017

Abstract

The adenomatoid odontogenic tumor (TOA) is considered by the World Health Organization as a benign tumor of odontogenic epithelium, with mature fibrous stroma, without ectomesenchyme, which represents between 2 - 7% of all benign tumors. It is characterized by slow but progressive growth of predilection by the anterior region of the maxilla and associated with a retained tooth.

Objectives: To report surgical management and follow-up of enucleation of TOA plus immediate reconstruction.

Materials and Methods: A 19-year-old female patient with progressive left ventricle enlargement who, after an incisional biopsy, was diagnosed with maxillary TOA, which is why enucleation and immediate reconstruction are performed.

Results: Postoperative controls are performed at 7th days and 4th months, showing satisfactory evolution. Conclusions: Enucleation plus the immediate reconstruction of these lesions represents a safe treatment alternative due to the low relapse rate.

Keywords: Degloving; Tumor; Odontogenic; Adenomatoid

Introduction

The adenomatoid odontogenic tumor (TOA) consists of a neoplastic lesion of odontogenic epithelium embedded in mature connective tissue characterized by slow but progressive growth [1,2]. The World Health Organization (WHO) classifies it into benign tumor lesions of odontogenic epithelium with mature fibrous stroma without ectomesenchyme [1]. Dreibladt was first described in the year 1907 as a variant of the ameloblastoma, naming it pseudo-adenoma-adamantine, later in the year 1915 Harbitz called it cystic adamantinoma. In 1948 Stafne considered it as a variant of ameloblastoma and finally in 1969 Philipsen establishes it as an individual entity, independent of the ameloblastoma, giving it the name of adenomatoid odontogenic tumor, denomination that was adopted in the first edition of the classifications of tumors odontogenic diseases in 1971 and ratified in the classifications of 1992 and 2005 [3].

It is known as the two thirds tumor, since it occurs most frequently between the second and third decade of life, 2/3 of the cases present in the anterior zone of the jaw, 2/3 occurs in the female sex, and 2/3 is associated with an impacted tooth, especially a canine [1,4-6].

The treatment of choice for this type of lesion consists of the conservative treatment through enucleation of the lesion, since it is a benign, low-aggressive, encapsulated lesion with low recurrence [1,3,6].

The objective of the present investigation is to report the case of enucleation of TOA in the left maxillary region with immediate reconstruction and to demonstrate by means of a series of controls performed, the nonrecurrence of the lesion and the preservation of facial fullness.

Case Presentation

A 19-year-old female patient, with no previous medical history, who reported onset of current illness for a year or so, presenting progressive increase in volume, not painful at the expense of left hemicara, so she goes to our service for evaluation. The clinical examination shows facial asymmetry at the expense of the left hemicara, presenting an increase in normothermic, normothermic, non-painful, and indurated volume in the nasogeniano region that moves the left nasal cavity to the contralateral side with effacement of the nasogeniano sulcus, preserved mouth opening, effacement of the Quadrant II Lobby Fund (Figure 1). Imaging evaluation shows a hypodense image with defined borders, with a unilocular aspect associated with the anterior region of the jaw, ranging from the alveolar region to the left orbit, with anterolateral expansion of the maxillary sinus, and a hyperdensal image compatible with third molar included in the lesion (Figure 2).

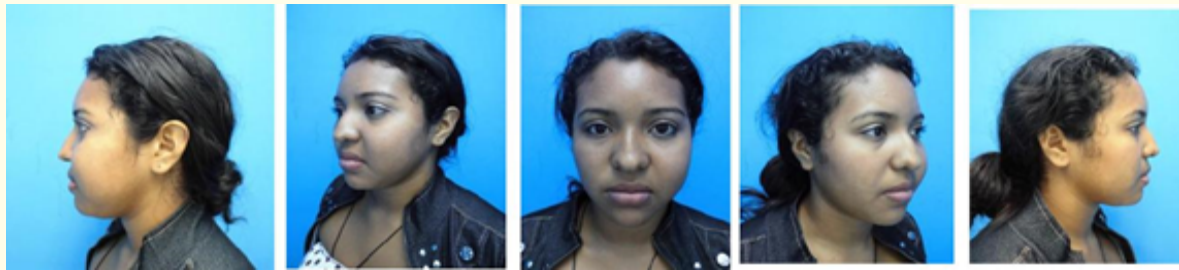


Figure 1: Preoperative clinical examination.

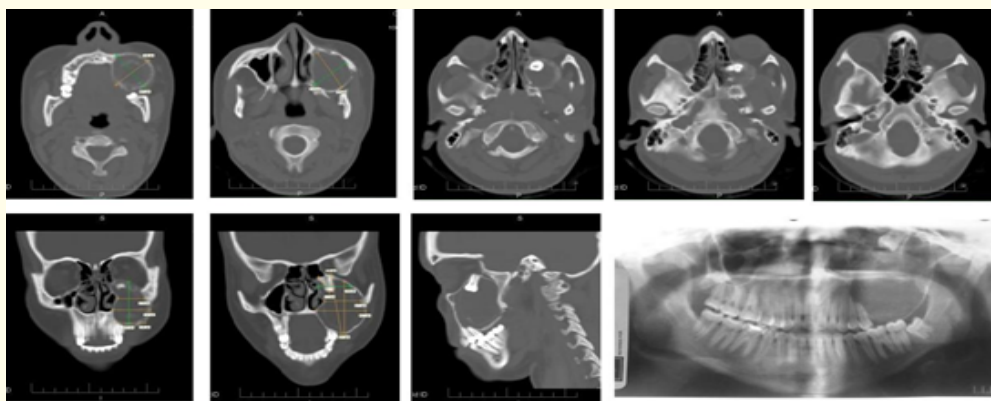


Figure 2: Preoperative imaging test.

It is performed under local anesthesia initially aspiration puncture, obtaining as a result clear citrus liquid that does not coagulate, and then through a triangular flap is performed incisional biopsy, followed by the placement of passive drainage type penrose. After the histopathological analysis, adenomatoid odontogenic tumor is obtained as a result, and the enucleation of the lesion is decided (Figure 3). The patient is taken to operative table, under general anesthesia, maxillary degloving is performed, exposing the lesion in left hemimaxilar region, exodoncia of dental units 2.3.2.4.2.5, (Figure 4) the tumor lesion marking and enucleation of the tumor (Figure 5 and 6) are performed without compromise of the orbit floor; pre-machined titanium mesh system 2.0 facial completeness, fixed to nasomaxillary and maxillary zygomatic buttocks with screws of the 2.0 system, and antrostomy and placement of gauze is carried out in order to avoid the collapse of the middle meatus (Figure 7 and 8). The patient is extubated without any complications.



Figure 3: Incisional biopsy.



Figure 4: Exodontia UD 2,3,24,25.

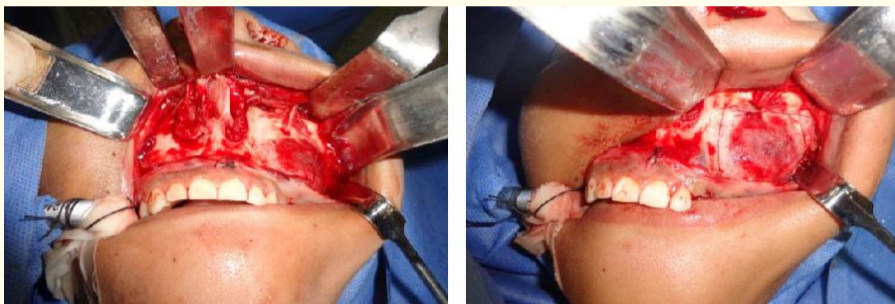


Figure 5: Degloving maxillary and tumor delimitation.

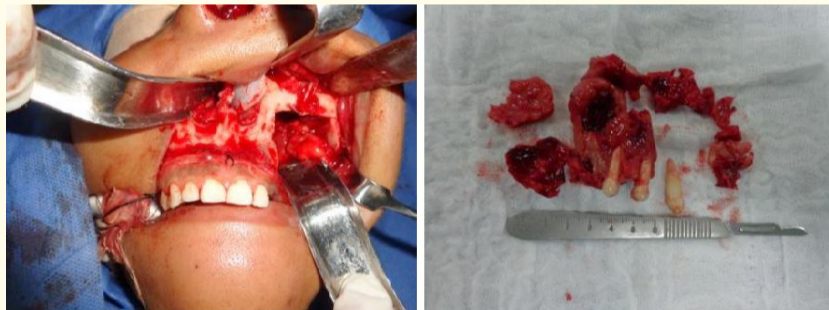


Figure 6: Tumor Enucleation.



Figure 7: Middle meatus antrostomy.



Figure 8: Reconstruction of the anterolateral maxillary sinus wall.

Postoperatorio control is performed at the week presenting mild volume at the expense of left hemicara, paraesthesia of infraorbital region, preserved permeability both nasal passages, preserved buccal opening and suture points in vestibule fund of quadrant II in position, without expense nor dehiscence. Images are evaluated as orthopantomography and computed tomography, where radiopaque and hyperdensal images are respectively compatible with osteosynthesis material in position (Figure 9). In the fourth postoperative month, a new control was performed, showing interpupillary line centered, preserved interpupillary distance, preserved ocular movements, no binocular diplopia, pronounced nasogenian furrows, infraorbital paraesthesia, preserved oral opening, normochromic and hydrated mucosa associated with quadrant II, permeable nostrils (Figure 10).

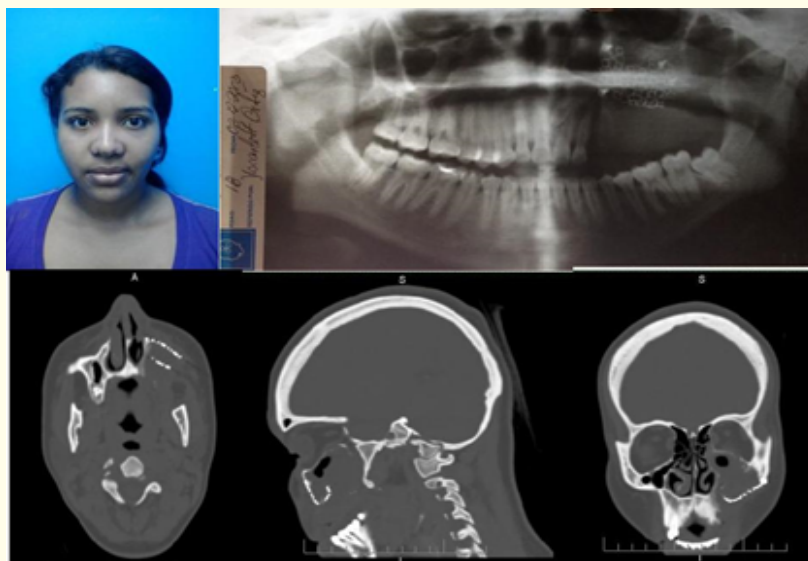


Figure 9: PO Control 1 week.



Figure 10: PO Control 4th month.

Discussion

The TOA presents many controversies regarding its specific etiology, there is consensus that it is odontogenic but not which tissue exactly, it has been proposed that it develops from the enamel organ, hard plate, reduced enamel epithelium, star reticulum, embryonic structures of the dental germ or its remains [4]. The latest revision of odontogenic tumors of the World Health Organization indicates that this lesion has a predilection for the female sex in a range of 1.9 on the male sex, the age of onset varies between 3 and 82 years, being the majority of appearance of cases before the third decade of life [1] epidemiology that is repeated in other epidemiological reports worldwide [2,3].

In relation to its clinical characteristics, it is an asymptomatic lesion, which presents a predilection for the maxilla in relation to the mandible [1,2,5], produces expansion of the bony tables, dental displacement, rarely exceeds 1.5 cm although it can reach a larger dimension [1,2,5]. Radiographically, it is evidenced as a radiolucent unilocular image with defined borders, which may or may not be associated with a retained tooth (usually canines or premolars), sometimes radiopaque areas may be present [5]. All these characteristics are presented in a similar way both in the maxilla and in the rare mandibular cases [5]. In our case all these characteristics mentioned above agree except that our injury was associated with a third retained molar and not a canine.

Philipsen and others performed a classification in three variants according to their clinical and radiographic characteristics [6].

Central or Intraosseous: present in 97% of cases [3].

- Follicular: If the tumor is associated with the crown of a retained tooth. It is usually associated with a retained canine (70%).
- Extrafollicular: If the tumor is not related to the crown of a retained tooth.
- Peripheral or Extraosseous: It is located in soft tissues, both in the gingiva and in the alveolar mucosa of edentulous areas, is present in 2 - 3% of cases [3].

Histopathologically, it presents as odontogenic epithelium of columnar or cuboidal appearance forming a nest with minimal stroma [1]. Epithelial cells organized in sheets and concentric masses form in their central part structures similar to the ducts of the salivary glands, with an amorphous eosinophilic material [1]. Another pattern is the formation of nodules composed of eosinophilic squamous polyhedral cells, the nucleus being slightly degenerate, with a calcified amyloid content [1,6,7].

Growth is slow and its little relapse is related to the low reactivity to immunostaining for the Ki67 antigen. They also present marked positivity for AE-1-3 associated with a universal nuclear reactivity for p53 antigen, being then classified as a tumor with low proliferative capacity and with inductive mesenchymal changes with production of amyloid material [8].

Differential diagnoses can be established for dentigerous cyst, calcifying epithelial odontogenic tumor, keratinizing cystic odontogenic tumor, unicystic ameloblastoma, ameloblastic fibroma, ameloblastic fibrodentinoma, globulomaxillary cyst [2,3,5].

The treatment plan of choice for authors such as Reyes, Sánchez and Vera is conservative by performing enucleation, due to the low recurrence of the lesion [2,6,8]. Reported cases of relapse are thought to be due to incomplete removal of the tumor during the initial surgical procedure [2]. Although the bone defect resulting from enucleation may be left without further treatment, authors such as Briones, et al. have reported epithelial migration compromising bone regeneration, which is why the use of combined frozen bone dry therapy for guided tissue regeneration has been suggested immediately following the removal of the TOA [9].

In the presented case, it was decided the enucleation of the lesion and the immediate reconstruction by means of the use of 2.0 system titanium mesh fixed to the nasomaxillary and zygomatic maxillary buttresses, in order to return the facial fullness and to avoid the collapse of the tissues soft, presenting satisfactory results in the post-operative 1-year delay. The reconstruction is recommended immediately to the resection of the defect.

Depending on the location and size of the lesion, an intraoral or extraoral approach may be indicated. In the present case, the tumor involved the alveolar ridge, maxillary antrum and even the most posterior portion of the orbital cone. The AO-CFM recommends the Weber-Ferguson approach for the resection of tumors that affect the maxilla and present an extension up to the infraorbital nerve [10]. The degloving of the middle third is a little used approach, developed and popularized in America by Dr. Maniglia in 1986 in its publication in an otolaryngology journal, is based on the combination of approaches for cheiloplasty and rhinoplasty, since then it has been demonstrated its versatility for mid-trauma type of naso-orbito-etmoidales, of zygomatics [11], resection of benign and malignant tumors of the middle third [11,12]. Its main advantages are the low rate of complications, easy preservation of vascular and nervous structures and the leaving no visible skin scars [11,12]. Being a benign lesion and based on recurrence rate, it was decided to use this approach. The main complications associated with this approach are nasal valve stenosis, nasal pyramidal deformity and temporal paraesthesia [11,12]. Stenosis of the nasal valve is easily avoided by performing a good suturing technique. In the present case, a series of 3M® Micropore-type adhesive strips were used to prevent nasal pyramidal deformity, which allowed the nasal shape and colloquial spots to be maintained with 5-0 polypropylene to prevent collapses of the nasal tip [12,13]. The patient's extraoral appearance was satisfactory in the mid and late postoperative periods, as she did not present external scars or deformities that could impede socio-cultural development. He is currently planning to place zygomatic implants for his oral rehabilitation.

Statement of Financing and Conflicts of Interest

The aforementioned authors, we give faith not to present any conflict of interest of financial or personal origin with other persons or organizations, the required any external funding.

Bibliography

1. Barnes L, *et al.* "World Health Classification of Tumors. Pathology and Genetics. Head and Neck Tumors". Lyon: International Agency for Research on Cancer (2005): 304-305.
2. Reyes S and Romero S. "Tumor Odontogénico Adenomatoide-Reporte de un Caso y Revisión de la Literatura". *Acta Odontológica Venezolana* 43.3 (2004).
3. Duque F, *et al.* "Characterization of the Odontogenic Adenomatoid Tumor in a Colombian Population". *Revista Archivo Médico de Camagüey* 17.2 (2013): 187-200.
4. Escalante M and Rebolledo M. "Concepto Actual, Diagnóstico y Tratamiento del Tumor Odontogénico Adenomatoide. Reporte de un Caso". *Revista Científica Salud Uninorte* 28.3 (2012): 432-437.
5. Yoris O, *et al.* "Tumor Odontogénico Adenomatoide con Características Inusuales. Reporte de un Caso". *Ciencias Odontológicas* 7.2 (2010): 163-168.
6. Sánchez E, *et al.* "Tumor Odontogénico Adenomatoide en Región de Mandíbula". *Revista Cubana de Estomatología* 47.4 (2010): 447-454.
7. Regezi J and Sciubba J. "Patología Bucal. Correlación Clinicopatológica. Tercera Edición". Mexico: McGraw-Hill Interamericana (1999): 340-343.
8. Vera Sempere F, *et al.* "Tumor Odontogénico Adenomatoide Folicular: Estudio Inmunohistoquímico". *Medicina Oral Patología Oral y Cirugía Bucal* 11 (2006): 305-308.
9. Briones D, *et al.* "Tumor Odontogénico Adenomatoide. Reporte de Caso y Revisión Bibliográfica". *Revista Dental de Chile* 96.2 (2005): 14-16.
10. Ehrenfeld M, *et al.* "Principles of Internal Fixation of the Craniomaxillofacial Skeleton". Americas 1st Edition. USES (2012).
11. Har-El G. "Medial maxillectomy via midfacial degloving approach". *Operative Techniques in Otolaryngology - Head and Neck Surgery* 10.2 (1999): 82-86.
12. A Baumann and R Ewers. "Midfacial degloving: an alternative approach for traumatic corrections in the midface". *International Journal of Oral and Maxillofacial Surgery* 30.4 (2001): 272-277.
13. Mulliken J. "Bilateral cleft lip". *Clinics in Plastic Surgery* 31.2 (2004): 209-220.

Volume 16 Issue 3 December 2017

© All rights reserved by Paola Peserico-Dal Farra., *et al.*