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TREATMENT AND HISTOPATHOLOGICAL ANALYSIS OF COMPLEX ODONTOMA: CASE REPORT

TRATAMENTO E ANÁLISE HISTOPATOLÓGICA DE ODONTOMA COMPLEXO: RELATO DE CASO

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ABSTRACT

Odontoma is a benign calcifying odontogenic tumor of unknown etiology, although local trauma, genetic factors, and chronic inflammation may be related. They are classified into compound and complex types, depending on the morphological, radiographic and histological characteristics. Clinically they hardly show signs or symptoms, and, when present, are related to delayed tooth eruption, cortical bone expansion and tooth displacement. Although radiographic exams are complementary, most cases of odontomas reported in the literature are found on routine radiographs. The diagnostic hypothesis is confirmed by histopathological examination that shows evidence of the presence of enamel, dentin, cementum and pulp tissue arranged in the form of denticles (compound) or a disorganized mass (complex). The treatment consists of surgical excision of the lesion. The aim of this article was to report a clinical case and histopathological analysis of a complex odontoma associated with an unerupted tooth, located on the left side in the posterior region of the maxilla, discovered by routine radiographic examination of a 57-year-old male patient. The treatment was surgical excision of the lesion, removal of the unerupted tooth and histopathological examination, which confirmed the diagnosis of complex odontoma. The case was followed-up clinically and radiographically for one year and showed no recurrence.
Keywords: Odontogenic Tumors. Odontoma. Tooth Unerupted.

RESUMO

Odontoma é um tumor odontogênico benigno calcificante, de etiologia desconhecida, embora traumas locais, fatores genéticos e inflamação crônica possam estar relacionados. Eles se classificam em composto e complexo, dependendo da característica morfológica, radiográfica e histológica. Clinicamente dificilmente apresentam sinais ou sintomas, e, quando presentes estão relacionados ao atraso da erupção dentária, expansão da cortical óssea e deslocamento dentário. Apesar dos exames radiográficos serem complementares, a maioria dos casos de odontomas relatados na literatura é encontrada em radiografias de rotina. A hipótese diagnóstica é confirmada pelo exame histopatológico que evidencia a presença de esmalte, dentina, cemento e tecido pulpar dispostos na forma de dentículos (composto) ou uma massa desorganizada (complexo). O tratamento consiste na excisão cirúrgica da lesão. O objetivo deste trabalho é relatar um caso clínico e análise histopatológica de um odontoma complexo associado à um dente incluso, localizado na região maxilar posterior esquerda, descoberto por um exame radiográfico de rotina de um paciente de 57 anos e do gênero masculino. O tratamento realizado foi a excisão cirúrgica da lesão, remoção do dente incluso e exame histopatológico, que confirmou a hipótese diagnóstica de odontoma complexo. O caso foi acompanhado clínico e radiograficamente por um ano e não apresentou recidiva.
Palavras-chave: Dente não erupcionado. Odontoma. Tumores odontogênicos.

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INTRODUCTION

Odontogenic tumors are rare findings in Dentistry. In the literature, their incidence is considered low, and ranges from 0.002% to 0.1% (TROELTZSCH *et al.*, 2012). Odontomas are commonly found in the oral cavity (ABDUL *et al.*, 2014; BEREKET *et al.*, 2015; SANTOS *et al.*, 2016; ISOLA *et al.*, 2017) and account for 22% of odontogenic tumors found (ABDUL *et al.*, 2014).

Paul Broca, in 1866, used the term odontoma to describe this tumor for the first time (ABDUL *et al.*, 2014; SANTOS *et al.*, 2016). Due to the abnormal and disorganized development of the epithelial and mesenchymal cells (ABDUL *et al.*, 2014; ISOLA *et al.*, 2017), that give rise to the enamel, dentin, cement and pulp tissue, odontomas are considered developmental anomalies of the hamartoma type (KÄMMERER *et al.*, 2016; ISOLA *et al.*, 2017) and not true neoplasms (BOFFANO *et al.*, 2012; ABDUL *et al.*, 2014; KUDVA *et al.*, 2016; AKERZOUL *et al.*, 2017; MALTAGLIATI *et al.*, 2020). They are also denominated benign tumors of odontogenic origin (MURPHY *et al.*, 2014; AKERZOUL *et al.*, 2017; SPEIGHT; TAKATA, 2018; ELSAYED *et al.*, 2020).

According to Isola *et al*. (2017) their etiology has not yet been clearly established. Among the possible causes are the association with local trauma, inflammatory and infectious processes, remnants of the dental lamina or hereditary anomalies, such as for example, Gardner's syndrome and Hermann's syndrome (BOFFANO *et al.*, 2012; ABDUL *et al.*, 2014; KÄMMERER *et al.*, 2016; PREOTEASA; PREOTEASA, 2018; LEVI-DUQUE *et al.*, 2019).

The odontoma generally affects the permanent teeth more than the primary teeth (ISOLA *et al.*, 2017). They are intraosseous lesions more frequently found in the anterior region of the maxilla (compound) and posterior region of the mandible (complex), however, in the literature there has been a report of lesions localized in gingival soft tissues (ISOLA *et al.*, 2017) or eruptions (MURPHY *et al.*, 2014; KUDVA *et al.*, 2016). They are commonly localized between the roots of erupted teeth or between the primary and permanent dentition (BEREKET *et al.*, 2015). The first and second decade of life are the mean age when they are diagnosed (BOFFANO *et al.*, 2012; MURPHY *et al.*, 2014; SANTOS *et al.*, 2016; LEVI-DUQUE *et al.*, 2019).

Clinically, they are painless, slow growing and in the majority of cases they are found in routine radiographic exams (KÄMMERER *et al.*, 2016; ISOLA *et al.*, 2017; PREOTEASA; PREOTEASA, 2018; LEVI-DUQUE *et al.*, 2019). However, the retention of primary or permanent teeth, cortical bone expansion and displacement of teeth are the most common signs that may lead to diagnosis (SANTOS *et al.*, 2016; PREOTEASA; PREOTEASA, 2018). Other rarer symptoms include dormancy of the lip, swelling in the area affected, pain, infection and eruption (BEREKET *et al.*, 2015; SANTOS *et al.*, 2016). When no sign or symptom appears, the odontomas may remain include in the bone for long periods (BEREKET *et al.*, 2015).

Based on the histological and radiographic characteristics, odontomas are classified as compound or complex (BOFFANO *et al.*, 2012; ABDUL *et al.*, 2014; ISOLA *et al.*, 2017; LEVI-DUQUE *et al.*, 2019; ELSAYED *et al.*, 2020). Radiographically, the compound type is shown as solitary or multiple small structures, similar to teeth that are not completely equal to a normal tooth, but have and ordered pattern of dental structures, enamel, dentin, cement and pulp. Whereas the complex type, is characterized as an asymmetrical radiopaque mass and no tooth-shaped structure is observed. Both types are delimited by a narrow, defined, radiolucent margin (ABDUL *et al.*, 2014; LEVI-DUQUE *et al.*, 2019).

The treatment generally indicated is surgical excision of the lesion and confirmation of the diagnostic hypothesis by means of histopathological exam (MURPHY *et al.*, 2014; KÄMMERER *et al.*, 2016). Early removal generally avoids growth of the odontoma, but should the lesion be in the initial stage, with an incomplete level of mineralization, there could be recurrence due to partial removal (KÄMMERER *et al.*, 2016).

Therefore, the aim of this article was to report the treatment of a complex odontoma in the posterior region of the left side of the maxilla, associated with an impacted third molar, and its histopathological analysis.

MATERIAL AND METHODS

The present study was approved by the Research Ethics Committee in Report No.4.681.410.

The patient, a 57-year-old white male, sought dental attendance with the chief complaint of a fractured complete maxillary denture. During anamnesis, the patient reported that he was depressive and has been a smoker and alcohol consumer for 30 years. In the extraoral exam, no change was found. In the intraoral exam, two changes were observed in the right maxillary alveolar ridge.

One of these was a plaque, close to the canine bossa, with a whole surface, of firm consistency, rough texture, measuring approximately 2 mm in diameter, with the diagnostic hypothesis of leukoplasia or hyperkeratosis. The second change, a papule, measuring approximately 1.5 mm in diameter, pale pink color, whole surface, firm consistency, rough texture suggestive of papilloma, granuloma or inflammatory fibrous hyperplasia. Both lesions were stained with toluidine blue. The result of the exam was positive, with focal marking. Afterwards, a biopsy was performed, and the histopathological exam confirmed hyperkeratosis.

The panoramic radiographic exam showed evidence of a radiopaque mass, closed to the crown of tooth 28, which was impacted (Figure 1). Cone beam computed tomography exam of the left maxillary alveolar ridge, allowed a well-defined, hyperdense lesion measuring approximately 10.5 mm x 8.6 mm to be identified on the crown of tooth 28, in the vertical position, in contact with the maxillary sinus (Figure 2). The diagnostic hypothesis was odontoma.

Figure 1 – Initial panoramic radiographic image.

Source: The authors.

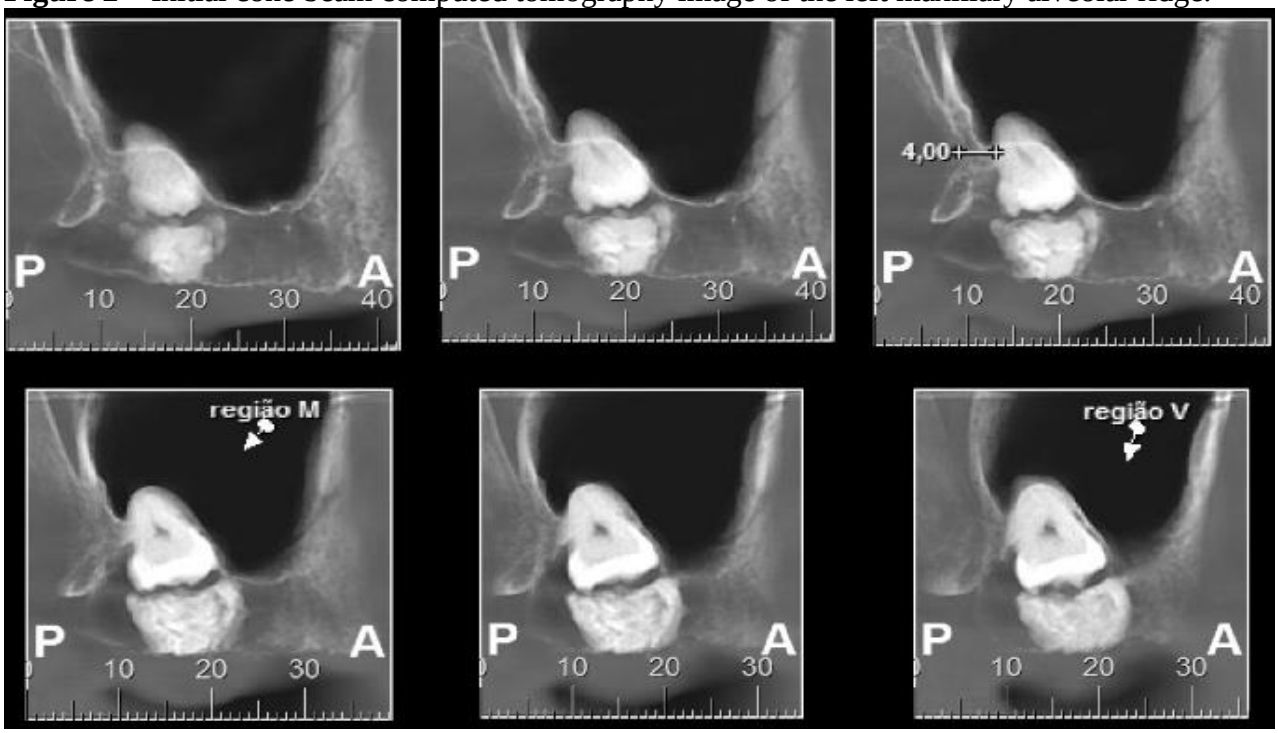
The treatment plan proposed was extraction of tooth 28 and excisional biopsy of the lesion. Prior to surgery, complementary hematological exams were requested. These showed conditions within normality for performing surgery.

Initially, in the surgical procedure intraoral antisepsis was performed with a mouth wash of 0.12% Chlorhexidine digluconate (PerioMax), for one minute, and extraorally, with 2% (Vic Pharma) followed by placement of the surgical fields. For anesthesia of the posterior superior alveolar and major palatine nerves, 2 tubes of 2% Mepivacaine withe epinephrine 1:100.000 (DFL) were used. In sequence, a scalpel blade no.15 (Solidor) was used to perform a mucoperiosteal incision in the region of the left superior posterior alveolar ridge and a relaxant incision in the vestibular and vital regions, with a view of obtaining improved visualization of the operating field.

Mucoperiosteal displacement was performed with a Mold displacer. Because tooth 28 was impacted, osteotomy was performed with the use of a straight handpiece and spherical carbide bur 702 (KG Sorensen), at low speed and irrigation with 0,9% sterile saline solution (Equiplex).

The lesion (Figure 3) and tooth (Figure 4) were removed with the aid of an Aplexo 303 and Potts lever (Quinelato). In sequence, the alveolus was submitted to curettage and regularization of the bone edges. Synthesis was performed by means of simple stitches (Figure 5), using silk thread Ethicon). The patient was medicated with 500 mg Amoxicillin, 600 mg Ibuprofen, 500 mg Paracetamol, for internal use. For external use, by means of mouthwash, 0.12% Chlorhexidine digluconate was prescribed.

Figure 2 – Initial cone beam computed tomography image of the left maxillary alveolar ridge.

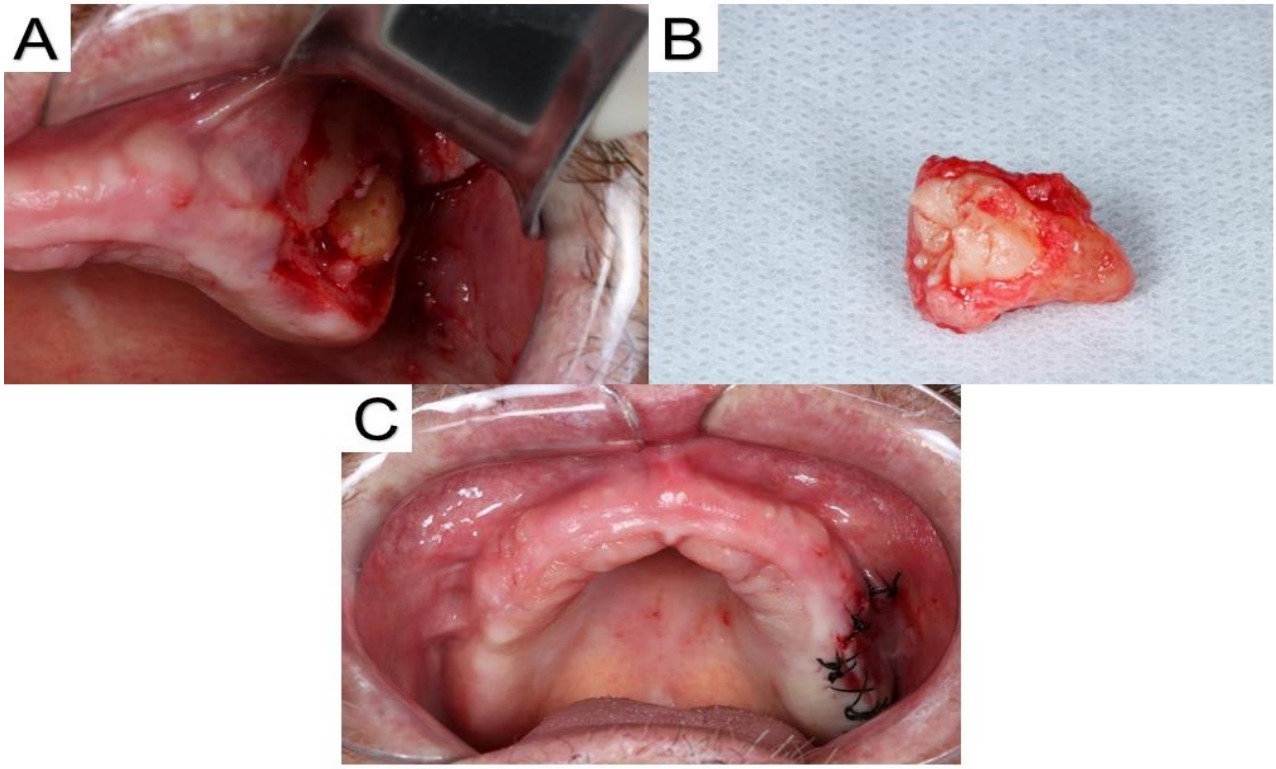


Source: The authors.

The result of the histopathological exam allowed identification of irregular arrangements if dentin and enamel, enamel matrix, cement and connective tissue similar to that of pulp, enveloped by a fibrous capsule, and absence of ameloblastic epithelium and signs of malignancy (Figures 6 and 7), confirming the diagnostic hypothesis of complex odontoma.

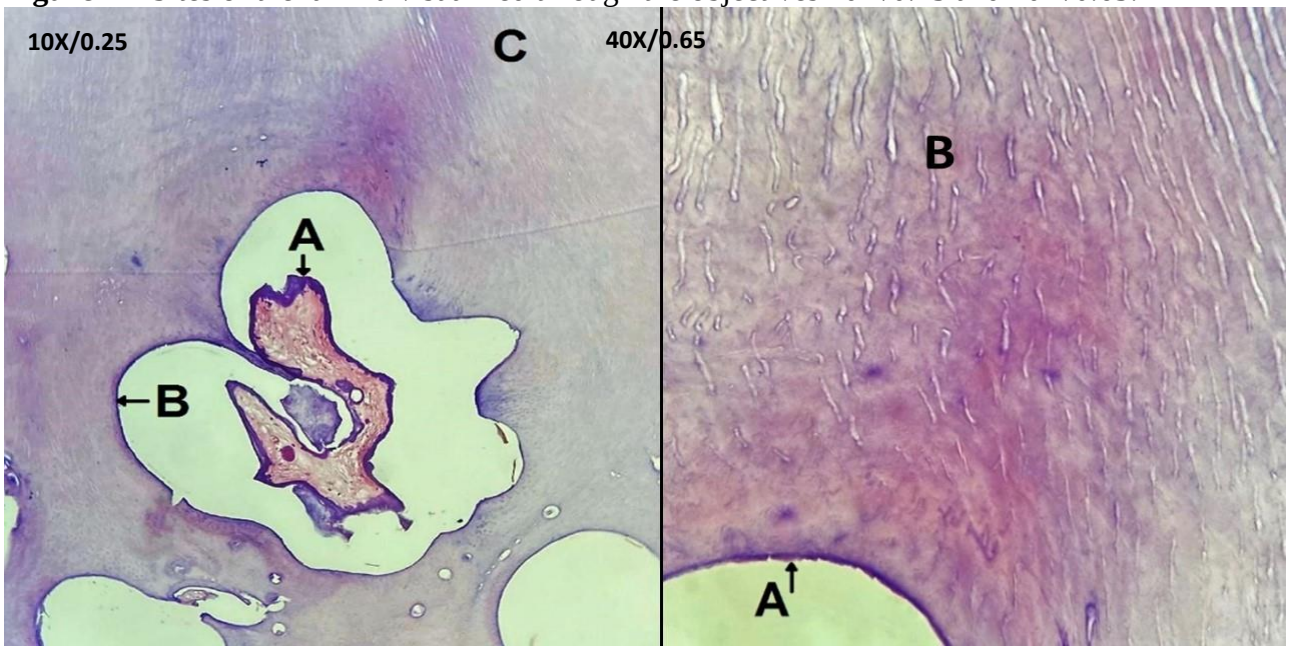
The patient returned for follow-up and removal of the stitches, after one week. After cicatrization, he was referred to the prosthesis clinic of the institution, for fabrication of a new complete maxillary dental prosthesis. The case was followed-up clinically and radiographically for one year. In the tomographic exam at 12 months, good cicatrization of the bone tissue was observed (Figure 8).

Figure 3 – Trans- and postoperative images.



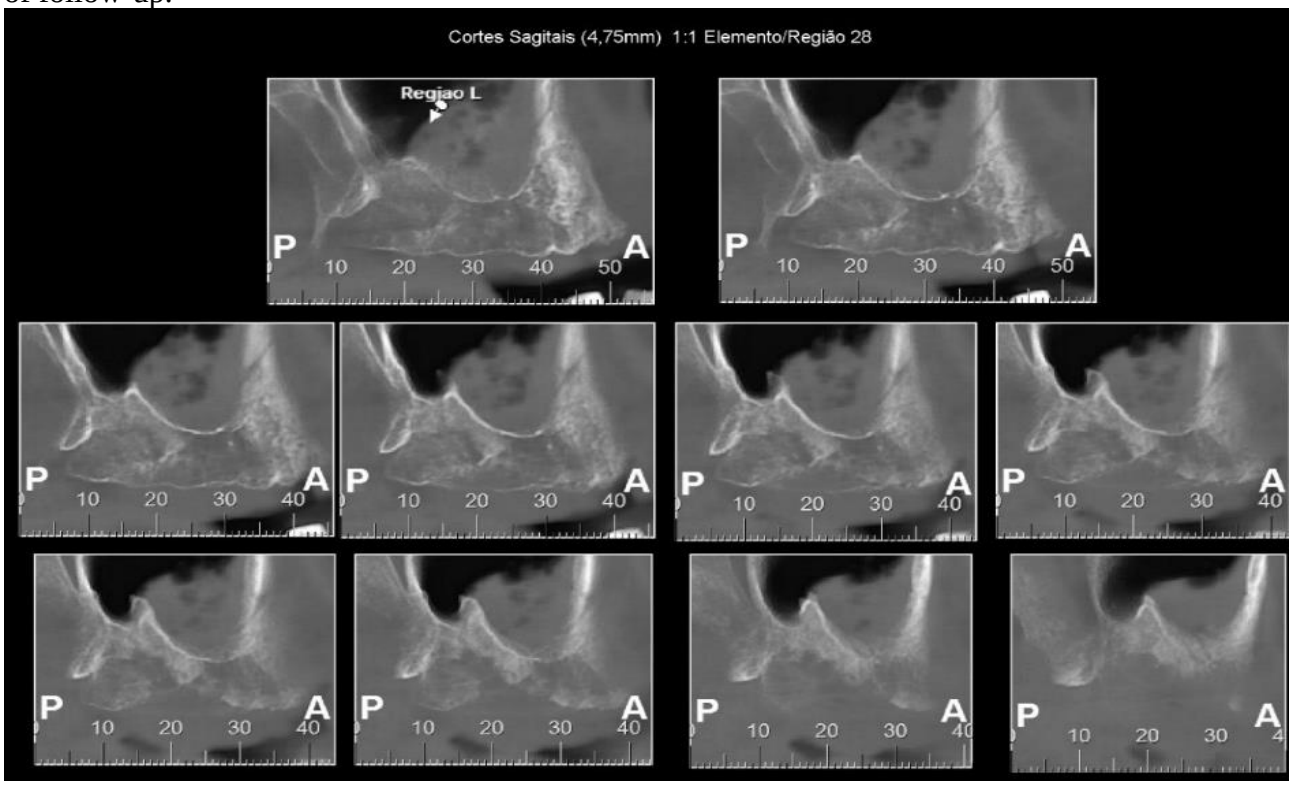
Notes: A: Surgical exposure of the tumor mass **B:** Tooth 28 after removal **C:** Suture.
Source: The authors.

Figure 4 – Sites of the lamina visualized through the objectives 10X/0.25 and 40X/0.65.



Notes: A (connective tissue, enveloped by a fibrous capsule), B (thin layer of enamel) and C (dentin of dentinal tubules).
Source: The authors.

Figure 8 - Cone beam computed tomography image of the left maxillary alveolar ridge at one year of follow-up.



Source: The authors.

RESULTS AND DISCUSSION

The study of Costa *et al*. (2012), who evaluated the cases of odontogenic tumors in Brazil from 1997 to 2007, showed that 94.5% were benign and 5.5% were malignant. The keratocystic odontogenic tumor (32.3%) was the lesion most frequently found, followed by the ameloblastoma (29.8%) and odontoma (18;4%).

The posterior region of the mandible, between second and third molars, (ABDUL *et al.*, 2014; PIPPI, 2014; ISOLA *et al.*, 2017) and the anterior region of the maxilla, between the incisor and canine, are the sites of greatest predilection of the complex odontoma and of the compound odontoma, respectively (BOFFANO *et al.*, 2012; PIPPI, 2014; KÄMMERER *et al.*, 2016). The differential in this clinical case was that the complex odontoma was localized in the posterior region of the maxilla. Nevertheless, according to Sekerci *et al*. (2015) the localization of these lesions is variable, due to regional and cultural diversity.

In the literature, it may be observed that the predilection for gender is not consensual among the authors (PIPPI, 2014; LEVI-DUQUE *et al.*, 2019). There are studies, such as that of Boffano *et al*. (2012), who associated predilection with the male gender and the study of Bereket *et al*. (2015) who associated it with the female gender.

For Kudva *et al*. (2016), odontomas may be seen in different stages of development. In the first stage, radiolucence is shown, due to lack of calcification. In the second, a partial calcification, and in the third stage, radiopaque masses surrounded by radiolucent areas are seen, histologically corresponding to connective tissue. Knowing these different stages is important, because there are rarely times when this lesion is recurrent after surgical excision. These cases may occur when the lesion is removed in an initial stage and all the cells were not yet completely differentiated. Whereas, postponing its removal is not recommended because the lesion may cause bone resorptions (LEVI-DUQUE *et al.*, 2019). In the patient in this case, in spite of the clinical evidence of tooth losses, the diagnosis and treatment were performed at a late stage but did not generate bone resorptions.

Due to their slow growth, odontomas manifest few clinical signs and are generally seen in complementary exams when they are of a considerable size or show indications of mineralization (BEREKET *et al.,* 2015; KÄMMERER *et al.,* 2016; PREOTEASA, PREOTEASA, 2018). Kemmerer *et al*. (2015) concluded that 6 % of the odontomas were found in tomographic exams, 16.8% were a coincidental finding, 6.6% due to late eruption of permanent teeth, 1.8% were found due to signs and symptoms of pain and tumefaction. Levi-Duque *et al*. (2019) concluded that 63.3% of the cases were found in routine radiographic exams, 30% due to the lack of permanent tooth eruption, 5% due to infectious symptomatology and only 1.7% as a consequence of increase in volume. Therefore, it is consensual in the literature that in the majority of cases the odontomas are found in complementary exams, such as routine radiographs (KUDVA *et al.,* 2016; MALTAGLIATI *et al.,* 2020).

Computed tomography is not indicated as the complementary exam of choice and must be used in specific cases in which there are higher risks and complications in the treatment, thereby reducing exposure to radiation (TROELTZSCH *et al.,* 2012). However, in this case the tomographic exam was necessary, due to proximity of the tumor to the maxillary sinus.

Among the possible differential diagnoses for complex odontoma are the ossifying cement fibroma, osteoíde odontoma, ameloblastic fibro-odontoma, cementoblastoma and the ameloblastic fibroma (MURPHY *et al.,* 2014; KUDVA *et al.,* 2016; ISOLA *et al.,* 2017).

Ossifying cement fibromas are differentiated from odontomas because they are more radiopaque and are frequently involved with unerupted molars. Osteoid osteomas have the characteristic of being small, oval, radiolucent and surrounded by sclerotic bone. Ameloblastic fibro-odontomas normally show greater radiolucence than the odontoma, but because they contain soft tissue components, they may resemble an odontoma in the stage of development. The cementoblastoma manifests as a well-defined radiopaque mass, adhered to the root of the tooth and surrounded by a radiolucent border (KUDVA *et al.,* 2016). The ameloblastic fibroma is a separate entity and presents as a radiolucent lesion (MURPHY *et al.,* 2014).

The gold standard treatment for the odontoma is the excisional biopsy (ABDUL *et al.,* 2014; REDDY *et al.,* 2014; BEREKET *et al.,* 2015; KÄMMERER *et al.,* 2016; ISOLA *et al.,* 2017; MALTAGLIATI *et al.,* 2020) and histopathological exam for confirmation of the diagnostic hypothesis (KÄMMERER *et al.,* 2016). Generally, small odontomas are removed without difficulty, and the larger types may need removal of large quantities of bone structure (BOFFANO *et al.,* 2012). The teeth involved are seldom damaged in the process of excision of the odontoma because in the large majority of cases there is a bone sept separating the lesion from the other structure (ABDUL *et al.,* 2014). Enucleation of the odontoma will result in a better prognosis for the impacted teeth because the risk of delay in eruption is greater with late diagnosis (ISOLA *et al.,* 2017). As is the case with any tumor, early diagnosis favors a more conservative treatment (SANTOS *et al.,* 2016).

Levi-Duque *et al*. (2019) observed that all the patients treated with enucleation had no recurrence within a year of follow-up, as occurred with the patient of this case report. However, in many cases, success depends on the eruption of the tooth involved because retained teeth associated with odontomas must be preserved while awaiting spontaneous eruption, or, when necessary, they must be submitted to orthodontic traction (BEREKET *et al.,* 2015; PREOTEASA; PREOTEASA, 2018). Another therapeutic option reported for odontoma is Er: YAG laser because it has a wavelength consistent with the treatment of hard tissues (MALTAGLIATI *et al.,* 2020).

When the odontoma is close to important anatomic structures, good planning of the case is necessary. In this report, the tooth impacted by the odontoma was in close contact with the maxillary sinus, increasing the risks of perforation during enucleation. In specific cases, in which computed tomography will contribute to the treatment, its indication is extremely important. In addition to allowing greater safety, CT will help with planning and performing a treatment with fewer risks in cases that involve important anatomic regions.

CONCLUSION

The treatment performed in the case here reported was surgical excision of the lesion, followed by removal of the impacted tooth and histological exam. This confirmed the diagnostic hypothesis of complex odontoma. The case was followed-up clinically and radiographically for one year and showed no recurrence.

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