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# Case report

# Retention of upper first molar caused by complex odontoma. A case report



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#### ABSTRACT

Odontomas are benign tumors composed of ectomesenchyme and odontogenic epithelium with or without dental hard tissue formation. Histologically, they are divided into complex and compound odontomas, the latter being the most common type. In general, they can be found between the roots of erupted teeth or between deciduous and definitive teeth, causing, in many cases, eruptive disorders or alterations in the dental position. Their growth is slow and asymptomatic, and they are usually diagnosed by chance in radiological exams, in which they are observed as a well-defined radiopacity with a radiolucent halo. The treatment of choice is surgical removal to prevent associated complications. We present the case of a 12-year-old male with a lesion in the left upper quadrant that was preventing the eruption of tooth 2.6. The lesion was surgically removed, and its subsequent histopathological analysis revealed a complex odontoma diagnosis. (Rev Port Estomatol Med Dent Cir Maxilofac. 2019;60(1):27-31)

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#### Retenção do primeiro molar causado por odontoma complexo. Caso clínico

#### RESUMO

Odontomas são tumores benignos compostos de ectomesênquima e epitélio odontogénico, com ou sem formação de tecido duro dentário. Histologicamente eles são divididos em odontomas complexos e compostos, sendo este último o tipo mais comum. Em geral, elas podem ser encontradas entre as raízes dos dentes irrompidos ou entre os dentes decíduos e definitivos, causando, em muitos casos, distúrbios eruptivos ou alterações na posição dentária. Seu crescimento é lento e assintomático, e geralmente são diagnosticados por acaso pelo exame radiológico, no qual são observados como uma radiopacidade bem definida com

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**Palavras-chave:** Criança Tumores odontogénicos

Odontoma Cirurgia Oral um halo radiolucente. O tratamento de escolha é a remoção cirúrgica para evitar complicações. Apresentamos o caso de um paciente do sexo masculino de 12 anos que apresentou lesão no quadrante superior esquerdo, impedindo a erupção do dente 2,6. A remoção cirúrgica da lesão foi realizada e sua análise histopatológica posterior revelou um diagnóstico de complexo odontoma. (Rev Port Estomatol Med Dent Cir Maxilofac. 2019;60(1):27-31)

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#### Introduction

Odontogenic tumors have an incidence of 0.002-0.1%<sup>1,2</sup> and odontomas are the most common one,<sup>3</sup> besides being the most frequent odontogenic neoplasm.<sup>2</sup> These tumors are composed of ectomesenchyme and odontogenic epithelium with or without dental hard tissue formation.<sup>4</sup> They are also called mixed tumors.<sup>4</sup> Some authors consider them hamartomas, due to their unlimited, autonomous and progressive growth, structural alterations and the prevalent development of one or more components.<sup>4,5</sup>

Histologically, the World Health Organization (WHO) differentiates between compound odontoma and complex odontoma.<sup>1-3,6,7</sup> The compound type is approximately twice as common as the complex type.<sup>1</sup>

- Complex odontomas: They form amorphous and disordered masses of odontogenic tissue that appear most frequently in the lateral region of the jaw.<sup>4,5,8</sup>
- Compound odontomas: They form a conglomeration of small, well-formed structures similar to teeth called denticles. They usually appear in the intercanine area in the maxilla.<sup>4,5,8</sup>

Clinically, odontomas can be central (intraosseous), peripheral (extraosseous) and erupted (especially in the compound type).<sup>5,8</sup> In general, they can be found between the roots of erupted teeth or between deciduous and definitive teeth, causing, in many cases, eruptive disorders or alterations in the dental position.<sup>4,8</sup> Their growth is slow and asymptomatic, and they are usually diagnosed by chance in routine radiographic exams.<sup>4,8</sup>

Radiologically, odontomas appear as a well-defined radiopacity with a radiolucent halo.<sup>1,6,8</sup> Complex odontomas present as non-specific, disorganized, irregular, unique or multiple radiopacities, which may be similar to osteolysis.<sup>3</sup> On the other hand, compound odontomas have an irregular radiopacity that corresponds to the denticles.<sup>3,8</sup>

The treatment of choice is surgical exeresis to prevent associated complications and subsequent histopathological study. When odontomas are associated with unerupted teeth, the tooth must be preserved while waiting for spontaneous eruption or orthodontic traction. A follow-up should be carried out to evaluate the affected tooth.<sup>4,8</sup>

The histopathological analysis is performed with an optical microscope by hematoxylin-eosin staining. Autofluorescence analysis with a laser confocal microscope has also been performed in some cases.<sup>4</sup> In this analysis, enamel, dentin, and, sometimes, cement and normal-looking pulp, can be found.<sup>1,3</sup> However, the are not organized due to the disordered expression and localization of extracellular matrix molecules in the dental mesenchyme.<sup>3</sup>

#### **Case report**

A twelve-year-old male patient visited the dentist for an orthodontic assessment. In the intraoral clinical examination, teeth 2.5 and 2.6 were absent in the arch, while tooth 6.5 was still present. The panoramic radiograph showed a radiopaque lesion with a circular appearance, which was causing the retention of tooth 2.6. (Figure 1). The patient did not report any symptoms, and no relevant background was found in his medical record.

Complementary cone beam computed tomography (CBCT) was prescribed to quantify the size and exact location of the finding (Figure 2).

The treatment plan decided was to perform surgical removal of the lesion (compatible with odontoma) in the second quadrant, as well as the extraction of tooth 6.5, and wait for the spontaneous eruption of teeth 2.5 and 2.6 (Figures 3 and 4). The possibility of orthodontic traction if the spontaneous eruption did not occur was considered. The lesion was enucleated and sent for anatomopathological study (Figure 5). The result of the analysis determined that it was a complex odontoma with the presence of disorganized dental hard tissues (enamel, tubular dentine, pulp tissue and cement) and odontogenic epithelial remnants related to the enamel matrix.



Figure 1. Initial panoramic radiograph. A radiopaque image related to tooth 2.6 can be observed.



Figure 2. A) Three-dimensional reconstruction in CBCT. B) Sagittal sections of the CBCT. From sections 22 to 27, the image compatible with complex odontoma is observed.



Figure 3. Surgical removal of the lesion.



Figure 5. Image of the odontoma after its extirpation.



Figure 4. Vision of teeth 2.5, 2.6 and 2.7 once the odontoma was removed.



Figure 6. Panoramic radiograph nine months after surgery. Tooth 2.6 is completely erupted.



Two months after the surgery, tooth 2.5 had erupted, and there had been no change in the position of tooth 2.6. So, it was decided to extract tooth 2.5 for orthodontic reasons, and the same was done with teeth 1.5 and 4.5 (3.5 was absent due to agenesis), and wait for the spontaneous eruption of tooth 2.6.

Nine months after the surgery, tooth 2.6 had erupted completely (Figures 6 and 7). The patient is currently under orthodontic treatment in order to place tooth 2.6 properly in the arch.

#### **Discussion and conclusions**

The etiology of the odontoma is not well defined, but it has been attributed to various pathological conditions, such as trauma, infections, hereditary anomalies (Gardner syndrome, Herman syndrome and basal cell nevus syndrome), odontoblastic hyperactivity and alterations in the genetic components responsible for the control of dental development.<sup>1,3,6</sup> A case of multiple complex odontomas associated with Otodental Syndrome, an autosomal dominant disease characterized by globodontia and high-frequency hearing loss, was recently published.<sup>9</sup>

Odontomas act similar to impacted teeth and, therefore, often cause alterations in tooth eruption (impaction or delayed eruption, retention of primary teeth and anomalies in tooth position, tilt or displacement of adjacent teeth).<sup>1,2</sup> Although most eruptive alterations occur in permanent dentition, the problem can be identified in early mixed dentition.<sup>1</sup> There is an association between eruptive delay and the presence of an odontoma.<sup>2</sup> Many retrospective studies obtained demographic and clinical data on patients who presented odontomas, analyzing the influence of active treatment in the dentition and in the treatment of impacted teeth.<sup>1-3,8,10</sup>

According to many authors,<sup>1,3,10</sup> odontomas are located more frequently in the region of the second and third mandibular molars. One study that analyzed 69 cases found 11 (55%) complex odontomas in the mandible and 9 (45%) in the maxilla, of which 9 were in the second and third molar area in the jaw.<sup>8</sup> Odontomas may also appear in the posterior maxilla.<sup>1</sup> Moreover, one study indicated that the most common area for the development of odontomas is the anterior maxilla, where the central incisors and the upper canines are more frequently involved, while the first and second lower molars seemed to be the least affected.<sup>2</sup> In that study, twice as many cases were located in the maxilla.<sup>2</sup>

Regarding gender, the rate of compound odontomas is distributed equally between men and women, according to most authors.  $^{\rm 1-4}$ 

In one study, 45 patients with odontoma (29 complex and 16 compound) were analyzed, and the average age of the patients with complex odontoma was  $15.7 \pm 2.4$  years old while that of patients with compound odontoma was  $16.6 \pm 2.1$  years old. Those authors indicated that the odontoma affected much more frequently the permanent dentition than the deciduous dentition.<sup>2</sup> Several authors state that the odontoma occurs most frequently in children and adolescents, between the first and second decades of life.<sup>1-4,10</sup> In the case presented, a 12-year-old male had a complex odontoma that affected his permanent dentition.

Some authors indicate that, although most of the odontomas are asymptomatic,<sup>2-4,8,10</sup> in some cases, swelling, pain, suppuration and bone expansion have been observed.<sup>2</sup> The latter is more frequent in complex odontomas.<sup>1</sup> Complex forms are less common and are more often associated with unerupted teeth as, according to the literature, 50% of complex odontomas may be associated with unerupted teeth.<sup>2,8</sup> In our case, the odontoma was asymptomatic, and the only sign was the unerupted tooth 2.6.

Regarding treatment, there is consensus on the need to remove the lesion, but there is no general agreement on the best management approach for the associated impacted teeth.<sup>1</sup> It is considered that teeth can develop normally if the obstruction is eliminated within the normal period of eruption,<sup>10</sup> and it seems reasonable to preserve neighboring teeth as long as the lesion can be surgically separated, and the teeth do not show resorption. However, it is still questionable whether the teeth directly affected by the lesion could be preserved.<sup>3</sup>

In one study,<sup>2</sup> 29 teeth associated with 45 odontomas analyzed were completely aligned through the orthodontic-surgical approach, and only four teeth erupted spontaneously after surgery during the 15 years of follow-up. 32 of the 45 odontomas were in the vicinity of at least one tooth (21 in the incisal area). A total of 12 teeth were extracted (8 complex and 4 compound odontomas) and, of the preserved teeth, 33 were displaced and retained.

If an odontoma is near or in direct contact with a tooth or is associated with other complications, early removal of the slow-growing lesion is recommended to avoid serious orthodontic alteration and distortion of the jaws, among other complications.<sup>3</sup>

The odontoma is the most frequent odontogenic tumor. In spite of its benign nature, it can cause numerous alterations, including eruptive disorders and displacement of teeth. The complex odontoma is less frequent than the compound type and is more frequently related to dental retentions. This case shows the preventive value of panoramic radiography as a basic diagnostic tool at early ages. Regarding the affected tooth, orthodontic traction for its repositioning in the arch is most probably required.



#### **Ethical disclosures**

**Protection of human and animal subjects.** The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

**Confidentiality of data.** The authors declare that they have followed the protocols of their work center on the publication of patient data.

**Right to privacy and informed consent.** The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

#### **Conflict of interest**

The authors have no conflicts of interest to declare.

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