

Hybrid lesion formed by a complex odontoma and dentigerous cyst: radiographic and histomorphological findings. A case report.

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Abstract: Hybrid lesions of the oral cavity are infrequent and share characteristics with a number of other pathologies. Both odontomas and dentigerous cysts are of odontogenic origin, but their simultaneous occurrence is rare and scarce. Clinical and radiographic examinations are not conclusive, making their identification difficult, while histopathological studies can reveal their defining characteristics. The aim of this report was to describe the radiographic and histomorphological findings of a hybrid lesion formed by a complex odontoma and a dentigerous cyst, affecting the mandible of a 22-year-old man, from Cartagena, Colombia, who had no relevant medical history, and no symptoms or discomfort in the affected area.

Keywords: odontoma; dentigerous cyst; third molar.

INTRODUCTION.

Hybrid lesions are rare tumor entities with histopathological elements from different lesions.¹ Qizhang *et al.*, reported a case of a complex odontoma with dentigerous cyst affecting the maxilla. In the histological study it was described as a combination of the two different patterns within the same lesion;² Norifumi *et al.*, reported a case of a calcifying cystic odontogenic tumor with an odontoma in a pediatric patient, these two tumors appeared inside a single lesion, with a heterogeneous radiological appearance,³ confirming the relationship of odontomas with cysts and with tumors of odontogenic origin.

Odontomas are benign tumors of odontogenic origin. They are composed of epithelial and mesenchymal cells; their etiology is associated with epithelial remains of the epithelial radicular sheath, odontoblastic hyperactivity and genetic factors.⁴

Histologically, compound odontomas are tumors formed by multiple structures similar to small teeth, consisting of enamel, dentin, cement and pulp, arranged in an ordered pattern, partially surrounded by a fibrous tissue capsule. Multiple and well differentiated⁵ radiopaque zones of various sizes can be observed in radiographs. On the other hand, complex odontomas consist of amorphous conglomerations of odontogenic tissue arranged in a disordered pattern, making them difficult to discern. Cords of odontogenic epithelium can be found in the periphery. Radiographic images shows a radiopaque amorphous and diffuse mass with a radiolucent halo; they are treated with surgery, followed by confirmatory histological studies.⁶

The dentigerous cyst is a structure that encloses the crown of an unerupted tooth, and its cavity is lined with epithelium. It is derived from the reduced enamel epithelium; a thin wall of connective tissue can be observed in histological analysis, with a layer of stratified squamous epithelium that covers the lumen of the cyst, the surface of the epithelium is usually covered with a thin layer of parakeratin and orthokeratin; the cyst cavity contains a watery thin yellowish liquid, sometimes with traces of blood. Radiographic examination reveals a unilocular radiolucent image well defined by sclerotic borders related

to the crown of an unerupted tooth and is mostly linked to retained third molars.⁷

It is important to report hybrid lesions formed by odontomas and dentigerous cysts, in order to contribute to a better understanding of their clinical, radiographic and biological behavior.

The aim of this report was to describe the radiographic and histomorphological findings of a hybrid lesion formed by a complex odontoma and dentigerous cyst, located in the right mandibular body. The CARE guidelines for a case report were followed.

Figure 1. Panoramic radiograph with radiolucent area in the lower part of the mandible with a well-defined radiopaque border, and radiopaque area in the upper part with defined borders.



Figure 2. A. Computed tomography scan showing a lesion involving the mandibular canal. B. Cross-section tomography with expansion of the internal cortex.



Figure 3. A. 10x, odontogenic lesion with deposits of enamel and dentin, without organized morphology, without pulp cavity, surrounded by a cystic area lined with cuboidal epithelium without atypia. B. 40x, irregular dense connective tissue with concentric calcifications.

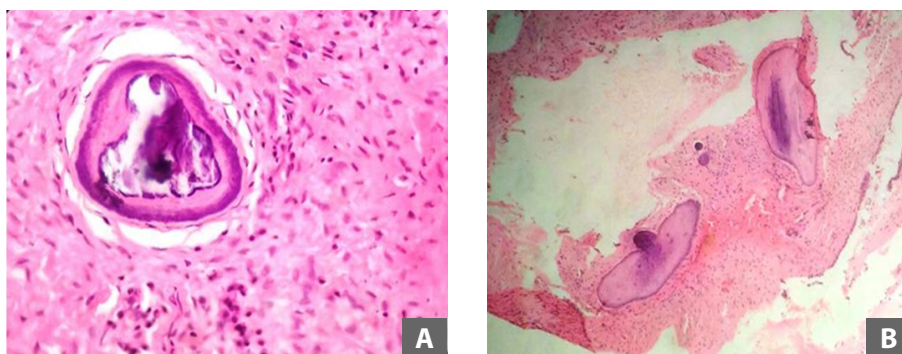


Figure 4. Radiography at three months post-treatment shows a radiolucent area with well-defined borders, with trace areas of variable radiopacity, and gray areas inside.



CASE.

A 22-year-old male patient from Cartagena, Colombia was referred by the orthodontic unit to the stomatology and oral surgery service of Corporación Universitaria Rafael Núñez, Colombia, presenting on a radiographic image a radiopaque zone on the posterior area of the right mandibular body, compatible with a cyst. In the anamnesis the patient does not present systemic alterations, nor symptoms or discomfort related to the affected area.

During intraoral examination, mucosa and gingival tissue were observed in the area with normal characteristics, mild expansion of the internal cortex and absence of pain on palpation. No pathologies were found during the clinical examination of other organs and systems, as well as no relevant medical family history.

In the panoramic radiograph, two areas were observed in the posterior area of the right mandibular body, a radiolucent zone in the lower part with a well-defined radiopaque edge, approximately 1.7cm in diameter, involving the mandibular canal, without affecting the distal root of tooth 47; the other area consisted of a radiopaque image in the upper part, with defined borders, approximately 1.1cm in diameter.

The crown of tooth 48 was observed inside the lesion, and a radiolucent area 5mm in diameter in the center. The computed tomography (CT) scan showed involvement of the mandibular canal; expansion of the internal cortex was observed in the cross section (Figure 1, Figure 2A and Figure 2B), with radiographic impression of odontoma and dentigerous cyst forming the lesion. Surgical therapy was decided as treatment.

The surgical procedure was performed under local

anesthesia, with a truncal anesthetic technique used, linear and scalloped incision was performed, flap detachment, and osteotomy to expose the tumor and cystic lesion. First the complex odontoma, and then the cystic membrane in all its extension were extracted; followed by bone curettage of the surgical bed, hemostasis and stitches. Postoperative recommendations, pharmacological therapy and clinical control visits were prescribed.

Tissue samples were sent in 10% formalin for histopathological study in two containers; the pathology department reported receiving a 1x1cm tooth and membrane tissue corresponding to the cystic cavity wall. The specimen was processed in a tissue processor, after decalcification of the dental structure, followed by embedding in paraffin wax and the posterior cutting of histological sections 3 microns thick with a microtome. Hematoxylin and eosin staining was performed.

The resulting anatomo-pathological report described an odontogenic lesion, characterized by a proliferation of well-defined tissue structures consisting of deposits of enamel and dentin, surrounded by fibro-connective tissue. An amorphous structure was identified, without a pulp cavity and with a cystic area covered with cuboidal epithelium without atypical cells. The findings corresponded to a complex odontoma with cystic changes related to a dentigerous cyst. (Figure 3A and Figure 3B)

Good healing of the soft tissues was observed eight days after the procedure. Three months after the surgical procedure, a control radiograph was obtained that showed a radiolucent area with well-defined borders, with trace areas of variable radiopacity, and gray areas within, indicating appropriate bone healing. (Figure 4)

DISCUSSION.

Cystic lesions are a common finding in the daily practice of oral health care professionals and maxillofacial radiologists. Cases of simultaneous pathologies are rare, especially of a complex odontoma related to a dentigerous cyst. An important consideration is that the diagnosis based on the radiological appearance of these lesions is a challenge for the dentist, as these lesions do not possess many defining clinical and radiographic characteristics.⁸

Several authors report the presence of simultaneous pathologies within the same lesion, such as Preeti *et al.*, Alkmim *et al.*, Norifumi *et al.*,³ who described cases of compound odontoma lesions with calcifying odontogenic cysts. While Qizhang *et al.*,² Astekar *et al.*,¹¹ reported cases of a complex odontoma with a dentigerous cyst, confirming the relationship between odontomas and cysts with tumors of odontogenic origin.^{9,10} Such relationship was evidenced in the current report of a hybrid lesion formed by a complex odontoma and a dentigerous cyst. Based on these findings, a change in the current understanding of the progression of the odontoma as a pathology is suggested, since although it is a benign lesion, when it is associated with other tumors and odontogenic cysts, it can be more aggressive. Due to their large size, odontomas can affect neighboring structures such as the roots of teeth and the mandibular canal, among others.

Dagrus *et al.*,⁸ reported a case of a dentigerous cyst arising from a large complex odontoma, located in the right posterior region of the jaw. They reported there were no clinical signs of inflammation, but there was the absence of molars and mild pain in the affected area. Their findings agree with the present report describing the case of a complex odontoma associated with a dentigerous cyst in the posterior area of the right mandibular body with no clinical alterations observed during the intraoral examination. However, there is disagreement regarding the description of symptoms, as in the present case, the patient did not refer any pain. These differences could be explained by the size of the lesion and the degree of involvement of the mandibular canal.

Simultaneous lesions are rare and the diagnosis based on the radiographic appearance poses a great challenge.

For this reason it is important to carry out a radiographic study with periapical, panoramic and CT images. These help in identifying radiographic characteristics of the lesion and contribute to the understanding of a correlation to radiographic findings. Sales *et al.*,¹² reported the case of an odontoma associated with a dentigerous cyst in the maxillary sinus, emphasizing the importance of the radiographic study. They reported that this type of lesion does not present clear and conclusive characteristics, and as such it is important to include radiographic panoramic images, CT and three-dimensional reconstructions in the examination.¹² In the current case, the panoramic image showed the well-defined borders of the lesion, with a density similar to the calcified dental tissue within. However, an additional radiographic evaluation with CT was required to determine the extension and the characteristics of the lesion, as the two-dimensional limitations of the panoramic radiograph did not allow the complete visualization of the lesion. CT images are necessary not only for the evaluation of the lesion itself, but also for the examination of the neighboring anatomical structures, for identification of associated pathologies, and to aid in the appropriate treatment plan.

From a histopathological point of view, dentigerous cysts may arise from the fibrous lining of the connective tissue of the odontoma, but they are very rare and could lead to an erroneous diagnosis. Reporting these hybrid lesions is important in order to contribute to a better understanding of their biology. Astekar *et al.*,¹¹ reported a case of complex odontoma associated with a dentigerous cyst, emphasizing histopathological features for diagnosis. They described the presence of mature dental tissue, enamel, dentin and cement arranged in a disordered pattern, small amount of stroma support, fibrous connective tissue that represented a fibrous capsule, compatible with a dentigerous cyst with complex odontoma.¹¹ Their description agrees with the current clinical case, in which the histopathological study describes an odontogenic lesion, with proliferation of well-defined tissue structures consisting of deposits of enamel and dentin, surrounded by dense irregular connective tissue, an amorphous structure, without pulp cavity and a cystic area lined with cuboidal epithelium without atypia, corresponding to a complex odontoma and dentigerous cyst.

CONCLUSION.

The hybrid lesion formed by complex odontoma and dentigerous cyst is rare. The histomorphological study is crucial for the diagnosis, revealing cellular characteristics that contribute to the identification of

the two pathologies.

Radiological findings alone are inconclusive and could lead to erroneous interpretations based only on conventional images, so an additional CT examination is highly recommended.

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