CASE REPORT

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Simultaneous lesions of complex odontoma associated with dentigerous cyst: case report and critical review in CBCT

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ABSTRACT

Objectives: To our knowledge, there is limited evidence reporting cystic lesions associated with odontoma. Therefore, this case report aims to describe the clinical, histological, and particularly the characteristic cone beam computed tomography (CBCT) diagnostic features .

Case Report: A 20-year-old male patient presented with a complaint of a lump in the upper right jaw. There was no history of systemic disease. A panoramic radiograph showed a solid radio-opaque mass surrounded by an osteolytic lesion with a radio-opaque margin causing impaction of the maxillary canine. Given the limitations of panoramic radiography in assessing lesion extent, a CBCT examination was performed. Multiplanar reconstruction revealed a well-defined unilocular hyperdense lesion with a hypodense encapsulation and corticated margin, measuring 31.5 x 32 mm, expanding the buccal cortical plate and pushing the upper right canine tooth into the maxillary sinus cavity. The extensive expansion of the lesion required histopathological analysis to confirm the final diagnosis. The histopathological examination concluded a dentigerous cyst.

Conclusion: Several reports in the literature discuss the development of cysts and tumors in the jaw, but some cases are very difficult to identify. The variability of lesions can complicate pattern identification, leading to misinterpretation of conventional radiographs, thus requiring additional CBCT evaluation.

Keywords: *Complex odontoma, dentigerous cyst, cone beam computed tomography (CBCT)*
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INTRODUCTION

In 1866, Paul Broca was the first to identify "odontoma" as a tumor caused by the enlargement of all dental tissues. Developmental abnormalities in the function of ameloblasts and odontoblasts, which are produced by fully differentiated epithelial and mesenchymal cells, result in odontoma. As a small fraction of all odontogenic tumors, odontomas vary from 23% to 77%.[1,2] Odontomas are classified as compound odontomas, which appear as normal dental tissues arranged in an orderly pattern and resemble multiple small tooth-like structures called odontoids or denticles, and complex odontomas, which consist of an irregular mass of calcified tissue with little or no resemblance to normal teeth. Complex odontomas have an incidence of 40%–68% according to the literature. As one of the most common odontogenic tumors encountered in clinical practice, complex odontomas pose unique diagnostic challenges and therapeutic considerations that require a deeper understanding of their pathogenesis, clinical features, and management strategies.[3,4]

On the other hand, dentigerous cysts appear as developmental cysts associated with the crown of unerupted teeth, often surrounding the crown and causing impaction. Formed from remnants of the dental formation apparatus, dentigerous cysts appear as fluid-filled cavities surrounding the crown of unerupted or partially erupted teeth. Although each lesion exhibits distinct histopathological features and clinical behavior, the simultaneous occurrence of complex odontomas and dentigerous cysts in the oral cavity is relatively rare, with only a few cases documented in the literature.[5]

Given the challenges in diagnosing and managing these dual lesions, the integration of advanced imaging modalities such as Cone Beam Computed Tomography (CBCT) is of paramount importance. CBCT provides high-resolution three-dimensional visualization of dental and maxillofacial structures, offering unparalleled insights into the morphology, extent, and relationship of lesions to adjacent anatomical structures.[6] By utilizing CBCT capabilities, clinicians can achieve accurate preoperative assessments, aid in treatment planning, and improve surgical outcomes in cases involving complex odontomas and dentigerous cysts.

This article presents a detailed case report of a patient diagnosed with both a complex odontoma

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and a dentigerous cyst simultaneously, emphasizing clinical and radiographic findings, case management, and postoperative outcomes. Additionally, we critically review the existing literature, discussing diagnostic challenges, differential diagnosis, and the role of CBCT in enhancing diagnostic accuracy and therapeutic efficacy in similar cases.

CASE REPORT

A 20-year-old male patient came to the Unhas dental and oral hospital with a complaint of a lump in the upper right jaw that had been present for a year. Occasionally, there was pain in the area of the lump. There was no history of trauma or extraction of permanent teeth, and no history of systemic disease.

Intraoral examination revealed an enlargement in the vestibular and palatal regions of teeth 11, 12, 13, 14, and 15, measuring 4.5 x 3.5 x 2 cm. The consistency was soft, crepitation was present only in the vestibular area, there was no pain on palpation, and the color of the soft tissue in the lesion area was redder than the surrounding tissue. Tooth 13 was unerupted, there were caries in teeth 36, 46, 11, 21, and 53, root remnants of teeth 16 and 24, an edentulous region of tooth 12, and poor oral hygiene (Figure 1). Upon extraoral examination, there was facial asymmetry, with the right third of the face showing expansion and the mouth opening normally (Figure 2).

Panoramic examination results showed a radio-opaque calcified lesion surrounded by a radiolucent lesion with sclerotic borders in the anterior maxilla region. The lesion involved the floor of the nasal cavity and the lateral wall of the right maxillary sinus, causing displacement of tooth 13 and pushing the roots of teeth 11 and 14 (Figure 3).

Due to limitations in assessing the extent of the lesion on the panoramic radiograph, the patient was referred for CBCT imaging. CBCT examination results showed a lesion in the anterior maxilla in the periapical region of the persistent tooth 53; a well-defined unilocular hyperdense lesion with an irregular shape resembling a calcified mass and encapsulated by soft tissue (hypodense) with corticated margins measuring 35 x 41.5 mm (coronal view), 31.5 x 32 mm (sagittal view), and 35 x 37 mm (axial view). The lesion caused expansion of the buccal cortical plate in the right maxilla, elevation of the right nasal cavity floor, and obstruction of the eruption of teeth 12 and 13 (impaction), with tooth 13 pushed into the right maxillary sinus in a vertical position and tooth 12 pushed to the apex of tooth 11 in a horizontal position (crown facing labially and root in the palate) (Figure 4).

Figure 1. Intraoral view showing enlargement in the vestibular and palatal regions of teeth 11, 12, 13, 14, and 15

Figure 2. Extraoral examination showed facial asymmetry with enlargement in the right third of the face.

Figure 3. Panoramic examination showed a radio-opaque calcified lesion surrounded by a radiolucent lesion with sclerotic borders in the right maxilla region, causing displacement of the upper right canine tooth, pushing the roots of the central incisor and first premolar

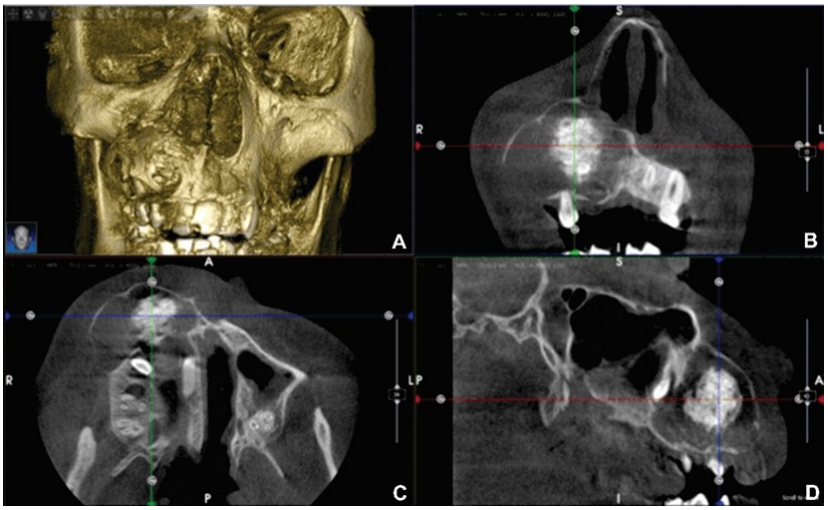

Figure 4. A well-defined irregular hyperdense lesion was seen in the right anterior region of the maxilla surrounded by a hypodense capsule with corticated margins. 3D Reconstruction View (A), Coronal View (B), Axial View (C), Sagittal View (D).

Aspiration tests revealed yellow fluid. The patient underwent an incisional biopsy under general anesthesia, and anatomical pathology results with microscopic examination showed tissue with a cystic structure lined with stratified squamous epithelium with connective tissue stroma, including scattered inflammatory lymphocytes and lymphocyte extravasation. Tissue analysis concluded a dentigerous cyst.

DISCUSSION

Hybrid tumors are much rarer compared to solitary tumors. Odontomas exhibit the proliferation of odontogenic epithelium and ectomesenchymal tissue associated with dental structures. These lesions occur in only about 3% of odontogenic tumors and typically occur in patients under 20 years of age.[7]

Dentigerous cysts are odontogenic cysts associated with the cementoenamel junction of unerupted teeth. Dentigerous cysts are usually asymptomatic and are often identified during routine radiographic examinations. Sometimes, dentigerous cysts can be associated with supernumerary teeth or odontomas.[8]

Since concurrent lesions are uncommon, diagnosing them might be difficult. Odontomas are the most prevalent odontogenic tumor of the jaws among tumoral disorders. Situations where ameloblastomas originated from dentigerous cysts are well-known examples of cystic metamorphosis or development from the tumoural capsule. Otherwise, despite observations in the literature, dentigerous cysts originating from odontomas are highly uncommon and may result in misdiagnosis.[9]

Cases of simultaneous lesions are rare, and diagnosis based solely on radiographic imaging presents a challenge for dentists due to the large size of the lesion and characteristics that can mimic fibro-osseous lesions such as fibrous dysplasia and ossifying fibroma.[10] An inaccurate diagnosis can result in serious damage to maxillofacial structures during treatment.

We also conducted a literature review to clarify and confirm various CBCT radiographic features of simultaneous lesions that are relatively large. Given the rarity of such cases, we focused on literature published over the past fifteen years with homogeneous cases to help characterize the radiographic appearance of simultaneous lesions in the jaw using CBCT imaging (Table 1).

Kwang et al. and Secran et al. reported findings of simultaneous lesions for the category of compound odontoma with characteristic multilobular cysts with fairly clear boundaries. However, it should be noted that the diagnosis of compound odontoma is determined by the presence of a radio-opaque mass resembling dental denticles, in contrast to complex odontoma, which presents as a more homogeneous irregular radio-opaque mass.[16]

Other findings were reported by Josip et al.[12] and Mahnaz et al.,[14] where the location of simultaneous lesions was in the mandibular region. A characteristic sign found was the presence of a calcified mass surrounded by a radiolucent expanding lesion that can usually destroy the cortical plate. Another feature that can be used is the measurement of the density of the mass compared to the density of the teeth, which will increase the diagnostic accuracy for odontoma lesions.

Brazao-Silva et al.[15] demonstrated that the lesion is easily visible using three-dimensional reconstruction in CBCT examination, showing a solid radio-opaque mass resembling enamel with an irregular surface associated with the impaction of tooth upper maxillary canine. As in this case, a large lesion was found related to an odontoma in the right anterior maxilla, which interfered with the eruption of the permanent canine. CBCT examination was very useful in this case to identify the internal structure of the lesion. Calcification

Table 1. Related published literatures of homogenous cases

Study	Age, sex	The lesion accompanying the dentigerous cyst	Localization	CT Findings
Kwang S K et al.,[11] Korea, 2019	56 year, male	Compound odontoma	Right infraorbital	Well-defined, thin-walled, non-enhancing, multilobulated cystic, size (3.4 x 2.2 x 3 cm) at regio dextra of infraorbital.
Josip Biocic et al.,[12] Croatia, 2010	10 year	Complex Odontoma	Right mandible	Calcified mass, widened surrounding radiolucent zone, tooth bud, dan a thinned lingual cortex.
Sercan K et al.,[13] Turkiye, 2018	53 year, male	Compound Odontoma	Right mandibular body	The multilobular structure of the cyst and a radiopaque area surrounded by a thin radiolucent border
Mahnaz Sheikhi et al.,[14] Iran, 2016	26 year, female	Complex Odontoma	Left mandible	Pericoronal radiolocencies around the impacted teeth, with well -defined sclerotic borders that displaced the teeth apically. There was a well-defined mixed radiolucent- radio-opaque lesion in the position of the first molar, and CBCT densitometry of radioopacities foci of the lesion showed a density near dentin and cancellous bone
Brazao-Silva MT et al.,[15] Brazil, 2022	21 Year, male	Compound-complex Odontoma	Left maxillary anterior	• Three-dimensional reconstruction of the lesion by computed tomography, showing a solid mass with a density similar to that of dental enamel, with an irregular surface, in proximity to the cystic area and impacted tooth • Sagittal sections by computed tomography show a wide hypodense area and coronal part of the impacted tooth 23.

within the lesion in the anterior region poses a risk to several vital structures such as the nasal cavity and maxillary sinus cavity, analyzing CBCT radiographs important for the characterization and diagnosis of simultaneous lesions suspected to be a dentigerous cyst associated with an odontoma, which cannot be shown by conventional radiography.[17]

CONCLUSION

Several reports in the literature discuss the development of cysts and tumors in the jaw, but some cases are very difficult to identify. The variability of lesions can complicate pattern identification, leading to misinterpretation of conventional radiographs, thus requiring additional CBCT evaluation.

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FOOTNOTES

All authors have no potential conflict of interest to declare for this article. Informed consent was obtained from the patient for being included in this case report.

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