

## Case Report

# Compound odontoma associated with a dentigerous cyst

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**Abstract** Dentigerous cysts (DCs) and odontomas are lesions of odontogenic origin relatively common in the oral cavity with a rare recurrence after treatment. The purpose of this study is to present a case of association of odontoma with DC in the mandible of a young patient and to describe all cases published in English literature to date. A 16-year-old male patient presented with a well-defined unilocular radiolucent lesion associated with impacted mandibular right canine. Mesially, there was another lesion, mixed with small radiopaque structures, similar to dental structures surrounded by a radiolucent halo. The diagnosis of DC associated with compound odontoma was made. This case report associated with the literature review of other reports highlights that the association of DC with odontoma is rare and further studies are needed to investigate the probable etiology of this association.

**Keywords:** Dentigerous cyst, jaw diseases, odontoma

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## INTRODUCTION

Dentigerous cysts (DCs) and odontomas are lesions of odontogenic origin relatively common in the oral cavity with a rare recurrence after treatment.<sup>[1,2]</sup> DC is an odontogenic developmental cyst with uncertain etiopathogenesis probably due to the accumulation of fluid between the reduced epithelium of the enamel organ and the dental crown. It is the second-most common cyst of the oral cavity mainly associated with impacted third molar.<sup>[2,3]</sup> Odontoma is a tumor-like lesion or hamartoma of mixed epithelial and mesenchymal origin composed of hard and soft dental tissues. It is classified into complex and compound, and its etiopathogenesis is not yet clear having local genetic mutation or trauma as the possible causes.<sup>[2-5]</sup> Odontomas are often surrounded by fibrous capsules and

rarely associated with a DC. Hence, the enamel organ from the odontoma could contribute to the growth of this cyst of development.<sup>[6]</sup> The purpose of this study is to present a case of association of odontoma with DC in the mandible of a young patient and to describe all cases published in English literature to date.

## CASE REPORT

A 16-year-old male patient, white, was referred to the oral diagnosis service by an orthodontist due to radiographic findings during treatment planning. On clinical examination, the patient revealed no systemic disease or traumatic history. On extraoral examination, the patient revealed no alterations, and intraoral examination showed the presence of deciduous right lower canine.

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On radiographic examination, a well-defined unilocular radiolucent lesion, associated with the crown of an impacted lower right canine, was noted. In addition, there was another lesion, mixed with small radiopaque structures, similar to dental structures surrounded by a radiolucent halo, located mesially to the first lesion [Figure 1a and b].

The clinicoradiographic diagnosis was compound odontoma associated with a pericoronal follicle tooth. The patient underwent surgery to remove the lesions, receiving the pre- and postoperative guidelines, in addition to the postoperative medication prescriptions and daily mouthwash with 0.12% chlorhexidine gluconate solution for 7 days. The surgery was conducted under conscious sedation of the patient.

The lesions were removed, and the tooth #43 was extracted, which was closely related to the lesion, which made it impossible to pull the element. The procedures of edge regularization, final cleaning with saline, and repositioning of the flap and suture with 5-0 nylon thread were performed.

On microscopic examination, a fragment of fibrous connective tissue partially covered by stratified squamous epithelial tissue exhibiting the loosely arranged suprabasal layer was observed. The dense connective tissue was permeated by discrete chronic inflammatory infiltrate, where focal areas of pericoronal follicle-like epithelium were observed [Figure 2a and b].

Fragments of pulp-like tissue surrounded by dentinoid tissue were observed [Figure 3]. After clinical, radiographic, and microscopic examinations, the diagnosis of DC associated with compound odontoma was made. The patient continues to be followed up for visualization of new bone formation, and informed consent was obtained for the report.

## DISCUSSION

A literature review was carried out for clinical cases in which DCs and odontomas were associated [Table 1]. Twelve cases were found, excluding those that were not published in Anglo-Saxon literature in addition to those cases that cannot be accessed due to the old year of publication, totaling 18 reported cases.<sup>[1,3-13]</sup> It is observed that, although isolated lesions are frequent, their association is poorly reported. A review of 81 cases of odontomas was performed, and this association was observed in only 9% of the cases.<sup>[14]</sup> The association is uncommon and can be a challenge only through clinicoradiographic information. The diagnosis of these



Figure 1: (a and b) Radiographic examination

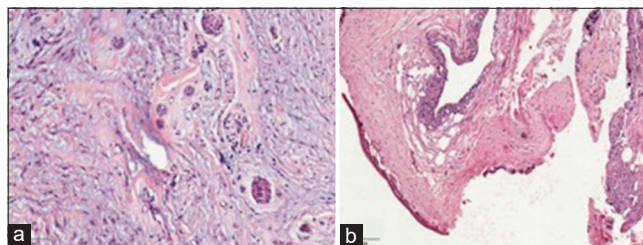


Figure 2: (a and b) Microscopic examination of the soft tissue

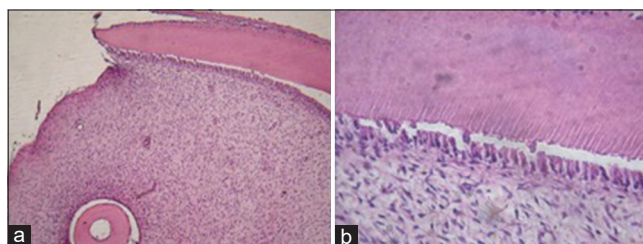


Figure 3: Microscopic examination of the hard tissue

associated lesions is also important because of the risk of severe bone changes.<sup>[1,3,4]</sup>

The mean age of the reported cases was 19.91 years, ranging 3–58 years. The age of our patient was very close to the average. In the literature survey, it was observed that most of the cases (67%) were males, as the case reported here.<sup>[1,3,4,6,8,10,11,13]</sup> Most cases (58%) occurred associated with impacted teeth as found in the case reported.<sup>[1,4,5,9-12]</sup> Only two cases have been reported without compound odontoma, unlike the present case.<sup>[5,11]</sup> Four cases occurred in the anterior region as reported in the present case, and only one case was associated with lower canine.<sup>[4,5,9,11]</sup> The treatment for both lesions is surgical removal with a good prognosis.<sup>[11]</sup>

Compound odontomas can be observed radiographically as multiple micro-teeth. In the case reported, radiopaque structures with radiolucent halo were observed with a capsule of odontogenic epithelium demonstrating the association of lesions.<sup>[2,11]</sup> Thus, the microscopic examination was fundamental for the confirmation of the diagnosis of associated lesions. In addition, the odontoma capsule could be diagnosed as dental follicle due to some focal areas of odontogenic epithelium.<sup>[14]</sup>

**Table 1: Literature review of odontomas associated with dentigerous cyst**

Author	Gender	Age	Location	Classification	Tooth
Astekar <i>et al.</i> , 2014 <sup>[3]</sup>	Male	17	Maxilla (left)	Complex	No association
Bansal <i>et al.</i> , 2014 <sup>[5]</sup>	Female	13	Maxilla (right)	Compound	#11
Biocic <i>et al.</i> , 2010 <sup>[7]</sup>	-	10	Mandible (right)	Complex	No association
Dagrus <i>et al.</i> , 2016 <sup>[1]</sup>	Male	21	Mandible (right)	Complex	#47
Jayachandran <i>et al.</i> , 2016 <sup>[8]</sup>	Male	16	Mandible (right)	Complex	No association
Jayam <i>et al.</i> , 2014 <sup>[9]</sup>	Female	11	Maxilla (anterior)	Complex	#11
Jeyaraj and Murali Mohan, 2008 <sup>[10]</sup>	Male	58	Mandible (right)	Complex	#44
Moraes <i>et al.</i> , 2016 <sup>[11]</sup>	Male	12	Mandible (right)	Compound	#43
Motokawa <i>et al.</i> , 1990 <sup>[12]</sup>	Female	3	Maxilla (left)	Complex	#65
Sales and Cavalcanti, 2009 <sup>[13]</sup>	Male	31	Maxilla (posterior)	Complex	No association
De Visscher <i>et al.</i> , 1982 <sup>[6]</sup>	Male	17	Maxilla (left)	Complex	No association
Wanjari <i>et al.</i> , 2011 <sup>[4]</sup>	Male	30	Maxilla (anterior)	Complex	#22
Current case	Male	16	Mandible (anterior)	Compound	#43

Odontoma could be confused with the diagnosis of ameloblastic fibro-odontoma due to the presence of soft tissues consisting of epithelial strands that form knots. These have a peripheral rim of columnar cells similar to the inner enamel epithelium that embraces a loosely arranged spindle-shaped epithelium identical to stellate reticulum. The epithelial strands are permeated by a myxoid cell-rich stroma with stellate-shaped fibroblasts having cytoplasmic extensions resembling embryonic tooth pulp and parts of hard tissue such as enamel and dentin. However, the new WHO classification (2017) classified the lesion as an odontoma in formation.<sup>[2]</sup> Several mixed lesions could be considered as differential diagnoses of complex odontoma; however, in this case, with the presence of multiple denticles, the clinicoradiographic diagnosis of compound odontoma was made.<sup>[2,7]</sup>

One of the theories of DC formation associated with odontoma is the presence of the enamel organ that is associated with the formation of both lesions, being formerly called cystic odontomas.<sup>[6,11,12]</sup> The authors also reported the hypothesis of a secondary trauma associated with an already-present lesion; however, a history of trauma was not reported by the patient.<sup>[11]</sup>

This clinical case report associated with the literature review of other reports highlights that the association of the DC with odontoma is rare and hence, further studies are needed to investigate the probable etiology of this association.

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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#### Conflicts of interest

There are no conflicts of interest.

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