



## Orthokeratinized odontogenic cyst arising from dentigerous cyst: A case report

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### ABSTRACT

One specific type of keratinized odontogenic cysts is orthokeratinized odontogenic cyst which is different from odontogenic keratocyst in clinical and pathological features completely. Also orthokeratinized odontogenic cyst can mimic typical characteristic clinicopathologic features of some other developmental odontogenic lesions such as dentigerous cyst and the odontogenic keratocyst, there is no previous report based on transforming of dentigerous cyst to orthokeratinized odontogenic cyst. We described a case of orthokeratinized odontogenic cyst originated from a dentigerous cyst in a 30-year-old female discovering accidentally in routine radiography.

**Keywords:** Dentigerous cyst; Odontogenic cyst; Orthokeratinized odontogenic cyst.

### Introduction

The orthokeratinized odontogenic cyst (OOC) is a relatively rare developmental odontogenic cyst arising from the cell rests of the dental lamina [1]. Orthokeratinised odontogenic cyst comprises approximately 17% of all keratinising jaw cysts [2]. Odontogenic keratocysts with orthokeratosis were also defined as orthokeratinized variant of odontogenic keratocysts by Wright in 1981, but in 1998 Lie et. al. suggested OOC as a different terminology [3]. In this article we present a case of orthokeratinized odontogenic cyst in relation to an impact-

ed mandibular third molar originated from a dentigerous cyst. The purpose of this article is to present the first case of OOC associated with dentigerous cyst in a thirty-year-old woman.

### Case Report

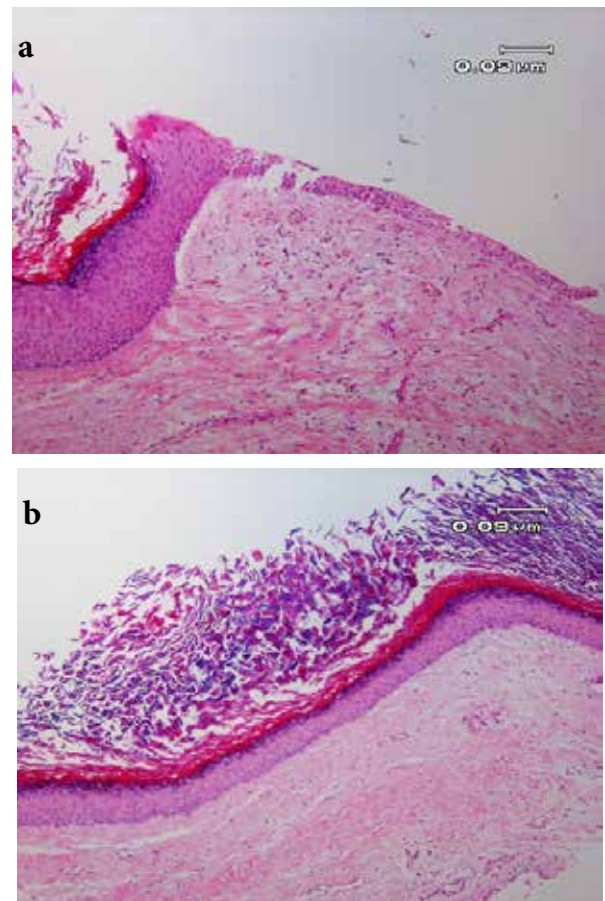
A 30-year-old woman presented with an impacted third molar in the posterior side of mandible surrounded by a radioluscent lesion found accidentally in the panoramic radiography. With regard to lesion's radiographic

features, there was a well-circumscribed, unilocular radiolucency surrounding the crown of an impacted third molar tooth in the posterior region of the lower jaw, with the most probable differential diagnosis of dentigerous cyst (Fig 1a). The tooth was extracted with the associated cyst completely. The gross of the lesion consisted of two pieces of cystic tissue with irregular internal surface and maximum thickness of 0.2cm in cyst wall and a white, non-transparent, cheese-like tissue which mimicking condensed keratin (Fig 1b). Microscopic sections revealed a cystic lesion lined by thin non-keratinized epithelial lining was evident in many areas, mimicking the epithelial lining of dentigerous cyst which continued with transforming to keratinizing epithelium (Fig 2a). Also orthokeratinized odontogenic epithelium with extensive layers of keratin in lumen and prominent granular cell layer (Fig 2b). Numerous areas of daughter cyst buddings were evident in epithelium and connective tissue interface. Thick fibrotic connective tissue wall revealed sheets of cholesterol clefts and numerous giant cells, diffuse chronic inflammatory cells, Russell bodies, diffuse hemorrhage, hemosiderin deposition and nerve bundles. With regard to histological findings and attachment of the cyst to the CEJ of the tooth, and also considering that the cyst showed characteristics of both a dentigerous cyst and an orthokeratinized odontogenic cyst at the same time, it could be concluded that this cyst had been a dentigerous cyst which was transforming to an orthokeratinized odontogenic cyst. So the diagnosis of “orthokeratinized odontogenic cyst arising from a dentigerous cyst” was reported.

a



**Figure 1.** a) Radiographic view shows a pericoronal cystic lesion around the crown of third impacted mandibular molar. b) Macroscopic view of the excised lesion with the associated tooth. The middle one revealed soft cheesy material resembling keratin.



**Figure 2.** Microscopic view of the lesion. a) Transforming area of non-keratinized epithelial lining of dentigerous cyst to orthokeratinized epithelial lining of orthokeratinized odontogenic cyst. b) Desquamation of keratin layers in orthokeratinized odontogenic cyst. Note prominent granular layer.

## Discussion

Odontogenic cysts are the most common cysts, encountered in dental practice and comprise a major aspect in oral pathology. They are considered as developmental in origin, because it is believed that they start to develop, when one of the rests of odontogenic cells such as the rests of Malassez, rests of Serres or the enamel organ gets activated and proliferates inside the bone or gingiva [4]. Dentigerous cysts (DCs) are one of the most common types of cysts which occur in the jaw. This cyst accounts for approximately 24% of all true cysts of the jaws, and their frequency in the general population has been estimated at 1.44 cysts for every 100 unerupted teeth [5,6]. In most of the cases, the diagnosis of a DC is straight forward; because of typical radiographic appearance as a radiolucent cystic lesion surrounding the crown of an impacted tooth [7]. But even radiographically, a 'typical' DC can be diagnosed as something else, e.g. a hyperplastic dental follicle [8]. The histological diagnoses of these lesions are therefore critical [9].

Orthokeratinized odontogenic cyst (OOC) is rare developmental odontogenic cyst originating from the cell rests of Serre. OOC has been designated as a discrete clinicopathological entity by the World Health Organization based on its peculiar clinicoradiological and pathologic characteristics [10]. OOC accounts for about 11% of all jaw cysts and is about 5.2% to 16.8% of all cases which were previously diagnosed as OKC [11]. In a recent review it has been shown that females were more frequently affected than males (ratio 2.59:1) [12]. Respectably, in the case presented here, the patient was a young woman. However, Li et al. have shown greater preference for males [13]. DC is said to be more frequent in males; but our case didn't fulfil this criteria. Both DC and OOC are mostly commonly seen in the molar – ramus region of mandible and usually associated with an unerupted tooth. This characteristic is also consistent with our case.

OOC is usually asymptomatic and slow growing, and occasionally associated with pain [14]. DC is usually asymptomatic, unless infected, and slow-growing. The case we presented also had no clinical signs or symptoms. Radiographically OOC appears as a well-defined, unilocular or multilocular radiolucency that is occasionally concordant with an unerupted tooth or root, as observed in this case [14]. Furthermore, a typical DC presents as an asymptomatic, unilocular radiolucency, which encloses the crown of an unerupted or an impacted tooth [8]. This is also consistent with

the our case. Histopathologically, the cystic cavity of OOC is lined by 4-9 layers of orthokeratinized stratified squamous epithelium with a prominent granulosum layer [14]. DC reveals a fluid-filled cyst lined by a thin, non-distinctive, non-keratinized epithelium. The epithelial lining consists of 2 to 6 layers of cuboidal epithelial cells, and the epithelium-connective tissue interface is flat [15]. The histopathological findings of our case were compatible with both OOC and DC.

Having all of the mentioned characteristics of these two cysts in mind, and considering these fact that DC is the most common odontogenic cyst with transformation potential to other odontogenic and non-odontogenic lesions, such as ameloblastoma, squamous cell carcinoma, and mucoepidermoid carcinoma [16]. it could be concluded that the cyst had been a dentigerous one but it had converted to OOC partially. Thus, we could see a dentigerous epithelium along with an orthokeratinized epithelium in the cyst. Based on our best knowledge, it is the first report of DC transforming to OOC, and on the other hand, the first collesion of these two odontogenic cysts.

Enucleation with curettage is the treatment of choice for OOCs and DCs. Only 0-4 % of OOCs showed recurrence [17,18]. This recurrence rate is completely different than the 30% or more recurrence rate associated with odontogenic keratocyst [18]. Although some studies believe that dentigerous cysts do not recur after complete excision [15,19], there are also studies implying that dentigerous cyst had 27.3% recurrence rate [20].

## Conflicts of Interest

The authors of this manuscript declare that they have no conflicts of interest.

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