

Odontogenic Keratocyst Masquerading as a Dentigerous Cyst in the Maxilla: A Case Report of an Unusual Presentation

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ABSTRACT

We encountered a case of an odontogenic keratocyst (OKC) that radiographically mimicked a dentigerous cyst and was associated with an impacted maxillary third molar. The common OKC location is in the mandible, with a lateral radiolucency. The attachment of the radiolucent mass to the coronal surface of the tooth in a maxillary molar is clinically highly unusual. The finding was pathologically confirmed after an excisional biopsy. The clinical and radiographic presentation is different than the typical description of OKC.

Keywords: Dentigerous cyst, odontogenic keratocyst, unusual, rare

INTRODUCTION

Odontogenic keratocyst (OKC) is officially known as a true benign tumor, and it is aggressive in nature. First described by Philipsen in 1956 as "odontogenic keratocyst," it was renamed by the World Health Organization (WHO) in 2005 as the keratocystic odontogenic tumor (1, 2). The entity was reverted back to be classified as a cyst in the 2017 WHO classification for the lack of evidence of being a tumor. It constitutes approximately 4%-16.5% of all cysts of the jaws (3). This aggressive cystic lesion has a high predilection for recurrence, higher than other odontogenic cysts (4). Many of the studies have shown that OKC occurs more in the mandible than in maxilla, with the posterior mandible being the commonest location according to published literature. The usual presentation is in the mandible and with a lateral radiolucency, and the entity spreads mesiodistally and has minimal buccolateral swelling (5, 6). This paper highlights a case of OKC occurring in the posterior maxilla in association with an impacted maxillary third molar, with a significant swelling in the maxillary vestibule. The radiological investigation showed coronal attachment of the radiolucent mass, typical of a dentigerous cyst.

CASE PRESENTATION

A 24-year-old male patient reported to our department with a chief complaint of pus discharge from the upper-right back region of the jaw lasting for 1 year. He reported a history of pain and pus discharge in the upper-right back region of the jaw lasting for 1 year. The patient visited a local dental surgeon, where he underwent extraction with respect to the upper-right second molar. Unfortunately, the associated symptoms persisted as mild

dull aching pain, which was intermittent nature. The patient had no significant medical or surgical history.

During the visit to our institute, the patient was subjected to radiological examination, by conducting a cone beam computed tomogram (Figure 1-4). The investigation showed an impacted maxillary molar on the right side, with a significant envelope of a hazy mass attached to the coronal section of the impacted tooth. The radiologist gave a provisional diagnosis as dentigerous cyst since the mass was attached to the coronal head of the impacted tooth.

Written informed consent was obtained from the patient, and he was operated under general anesthesia. The lesion and the tooth were removed in completely. The hazy mass seen in the radiograph was a thick curdled pus fluid. The entire specimen was sent for a histopathological examination.

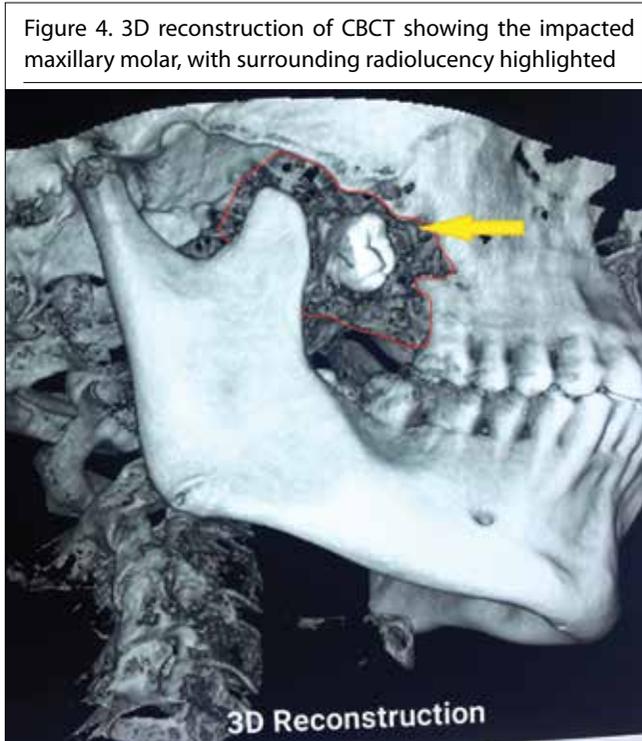
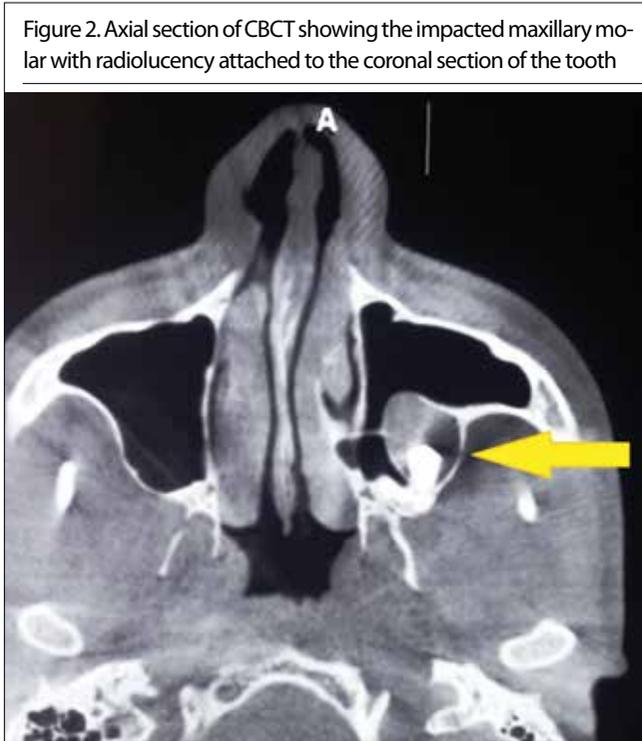
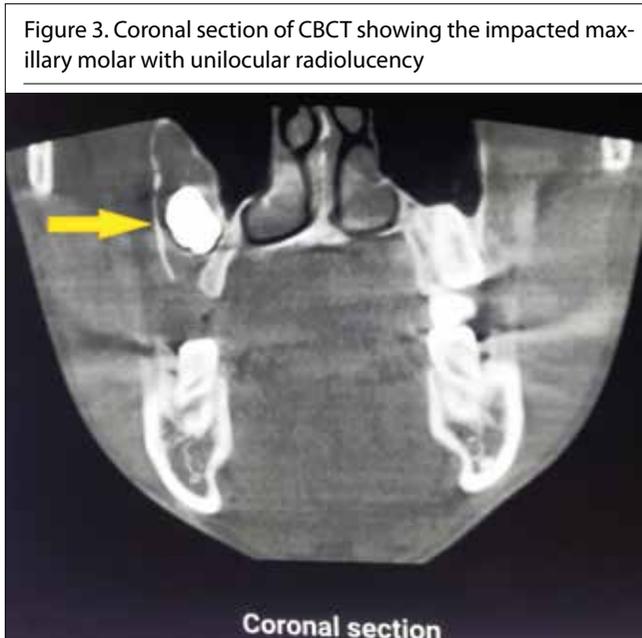
The histopathological report in our case described the lesion as follows: The H and E sections of the submitted specimen showed a cystic odontogenic epithelial lining and an underlying connective tissue capsule. The odontogenic epithelial lining was the stratified squamous parakeratinized type, showing the areas of minimal corrugation (Figure 5, 6). The epithelium is six- to seven-cell-layers thick with basal cells showing the palisaded arrangement of the nuclei and nuclear hyperchromatism. In a few areas, the lining epithelium can be seen separating from the connective tissue. The underlying connective tissue capsule shows bundles of collagen fibers, fibroblasts along with chronic inflammatory cell infiltrate consisting of lymphocyte, and plasma cells,

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blood capillaries, and extravasated RBC's. Daughter-cyst-like areas can also be seen in the connective tissue capsule, thus giving an impression of the lesion to be suggestive of the keratocystic odontogenic tumor (Figure 7-9).

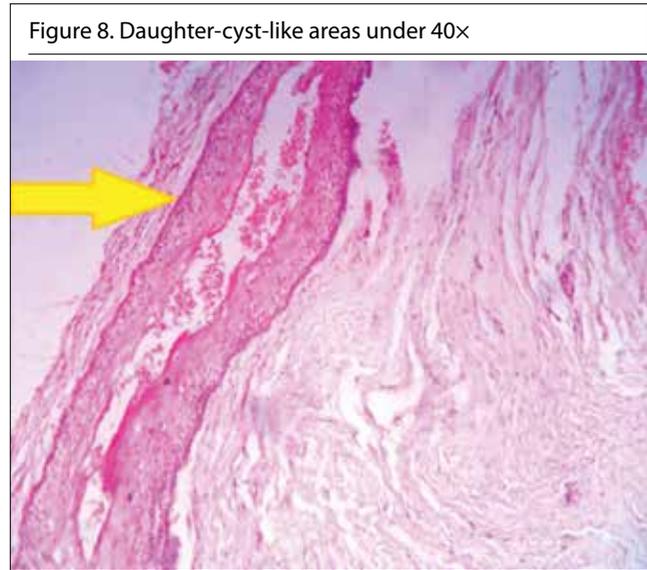
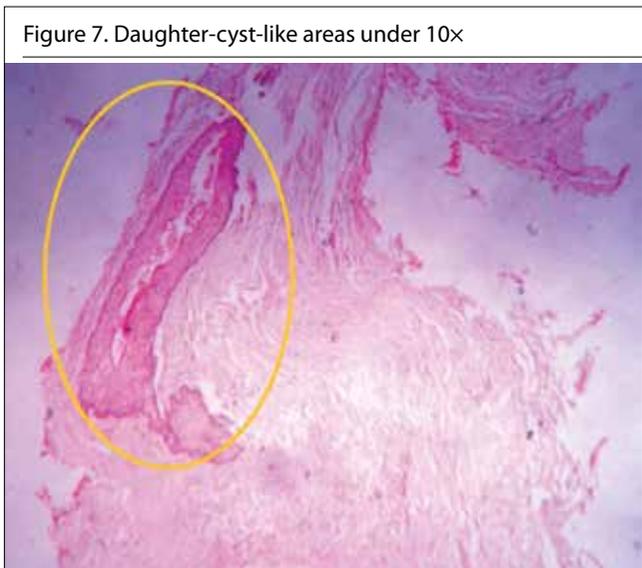
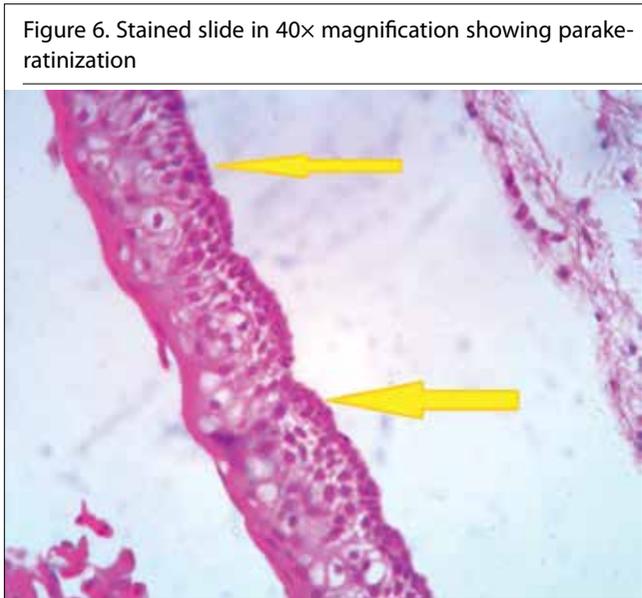
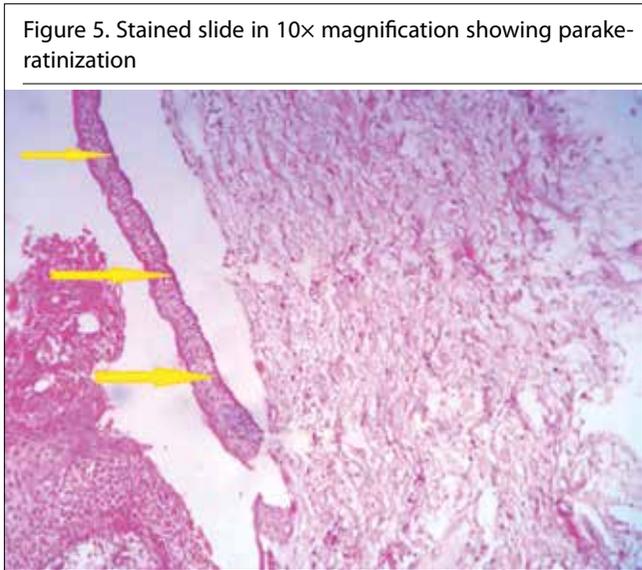
Healing was uneventful with no untoward complications noted in the past 1 year of regular follow-ups.

DISCUSSION

Dentigerous cysts are odontogenic cysts that are associated with the crowns of unerupted teeth. These cysts are termed

dentigerous, which means "containing tooth," and this is the character description of the cyst. A characteristic feature of a dentigerous cyst is its appearance: The cyst typically surrounds the crown of an unerupted tooth, expands the follicle, and is attached to the cemento-enamel junction of the unerupted tooth (7).

Dentigerous cysts most frequently involve unerupted upper and lower third molars and are generally seen in patients aged between 10 and 30 years. Males are more affected by an incidence rate of 1.6:1 (8). The cysts are usually solitary (9). The exact



pathogenesis of these cysts is unknown; however, it is assumed that they develop due to the accumulation of fluid between the reduced enamel epithelium and tooth crown (10-11). These cysts are generally without symptoms and exist for many years without being discovered.

Dentigerous cyst occurs radiographically as a well-defined unilocular radiolucency, often with a sclerotic border. As the epithelial lining is derived from the reduced enamel epithelium, in radiographs, the radiolucent shadow characteristically surrounds the crown of the tooth. A large dentigerous cyst may sometimes resemble a multilocular process (12).

Radiographically, dentigerous cyst can be described as;

1. The central variety, in which the tooth crown is enclosed by the radiolucency, and the crown protrudes into the cystic lumen;
2. The lateral variety in which the cyst occurs laterally along the tooth root, thus partially surrounding the crown; and

3. The circumferential variety exists when the cyst not only surrounds the crown but also extends down along the root surface, thus giving the impression of the tooth within the cyst (13).

The above-mentioned features clinically and radiological are well known to represent a dentigerous cyst. A histological feature seen in the dentigerous cyst lining is usually a fibrous wall with two- to four-layer thickness of non-keratinized stratified squamous epithelial lining, and it consists of the myxoid tissue, odontogenic remnants, and occasionally, the connective tissue wall resembles dental papillae and shows mild chronic inflammatory cell infiltrate with mild vascularity and areas of hemorrhage (13).

The typical radiographic features of OKC are unilocular, multi-locular, or multiple well-circumscribed radiolucent lesions surrounded by a thin radiopaque border with a smooth or loculated periphery. The lumen is frequently densely filled with keratin, causing the image to show a hazy appearance (14). An unerupted tooth is involved in the lesion in 25%-40% of OKC cases; in such instances, the radiographic features suggest the diagnosis of a dentigerous cyst. In these cases, the cyst has presumably arisen from a dental laminal cyst in the vicinity of an unerupted tooth and has grown to envelop the unerupted tooth. Altini and Cohen (15) introduced the term follicular primordial cyst (follicular keratocyst) for this group of lesions, this lesion was typically keratocyst on histology, but on macroscopic examination, it is seen surrounding the crown of the tooth and is firmly attached to the neck.

Histological examination accounts for the definitive diagnosis. The epithelial lining in the keratocyst is highly characteristic with its features being unchanged even in different specimens. Five- to eight-cell-layer thick, regular keratinized stratified squamous epithelium without rete ridges lines the cyst. The form of keratinization is usually parakeratotic (80%-90%), but it is sometimes orthokeratotic. The keratin formation amounts to no more than a thin eosinophilic layer of parakeratin in the parakeratotic variant.

The squamous epithelium has a clearly defined, palisaded layer of tall basal cells. The cells superficial to the basal layer are polyhedral and often exhibit intracellular edema.

Although the treatment of both the dentigerous cyst and OKC are excision, protocol calls for aggressive resection followed by chemical cauterization in cases of OKC owing to its high incidence rate. Extensive resection and use of the Carnoy's solution are considered the normal modality for the OKC treatment.

CONCLUSION

Differential diagnoses of radiolucencies that occur in the maxilla and mandible cover a broad spectrum of cysts and tumors of odontogenic and non-odontogenic origin. An accurate diagnosis is based on the distinctive clinical, radiographic, and histopathologic aspects, but variations from these typical features also exist, and the clinician should be aware of the possibility of these variations in diagnosis and treatment of the radiolucent

lesion occurring in the mandible and maxilla (15). Although the patient was treated as a case of dentigerous cyst with no routine protocol for an OKC treatment not being followed, the patient was placed on regular follow-ups after the histopathological diagnosis as a precaution for further recurrence and repercussions. Regular radiographic monitoring was also done to establish the same.

This paper is an attempt to highlight the many possible clinical outcomes of a non-suspecting lesion and the need for clinicians to be aware of these masquerading lesions and to anticipate future complications if any.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

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