Case report

Lateral Odontogenic Keratocyst Clinically Diagnosed as a Dentigerous Cyst (A case report and literature review)

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ABSTRACT

Odontogenic keratocyst (OKC) has been an area of considerable research over the last decades owing to its unique behavior, debated origin, distinctive tendency to recur, and argued nature. In 2005, WHO has adopted the designation of keratocystic odontogenic tumor (KCOT) because of its aggressive nature and tendency to recur, so, it has been long been considered as a benign jaw neoplasm rather than a cyst, though the last classification has returned the OKC back to cyst category. It is not uncommon for OKC to be clinically and radiographically identical to dentigerous cyst which makes the initial diagnosis rather confusing; however, in our reported case another interesting and an unusual feature was the arrival of an intact cyst attached to the neck of an impacted tooth which has further drawn the attention toward the dentigerous cyst, particularly the lateral type. The final diagnosis was made following the microscopic examination of the surgical specimen which was almost convincing and consistent with OKC. The clinical, radiological and histological features of this pathological entity along with brief relevant studies have been discussed.

Key words: odontogenic keratocyst, odontogenic tumor, dentigerous cyst

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INTRODUCTION

Odontogenic keratocyst (OKC) is a developmental odontogenic cyst which has an epithelial origin. It was first identified in 1876 and further characterized by Phillipsen in 1956 [1].

Compared to other odontogenic cysts; the odontogenic keratocysts (OKCs) may exhibit tumor like behavior; hence the other name is keratocystic odontogenic tumor (KCOT). The tumor-like nature of the OKC is manifested in the aggressive clinical course of some cysts, the significantly high recurrence rate, and its association with nevoid basal cell carcinoma syndrome (NBCCS) [2].

Importantly, some OKCs may be misdiagnosed as dentigerous cysts especially those arising in dentigerous relationship, hence microscopic examination must be carried out to get accurate diagnosis [3].

This article presents a silently growing OKC that was initially diagnosed as a dentigerous cyst based on its radiographic appearance, though OKC was considered as a differential diagnosis; even though, the whole cyst tissue was received intact in the laboratory which is rare for the OKC because of its thin and folded wall.

CASE REPORT

A 25 years old female Libyan patient attended the department of oral pathology, medicine, diagnosis and radiology, faculty of dentistry- Benghazi university. The patient was concerning about a missing wisdom tooth, she had no pain, swelling, discharge, or any other symptoms. On examination the patient was looking well, and had no extra oral swelling. Intraoral examination revealed missing right and left wisdom teeth with no swelling or bulging being detected.

Macroscopic features:

The cyst was received intact along with the impacted third molar tooth, attached to its cervical region just like a lateral dentigerous cyst (Figure 2). An abundant yellowish cheesy material was released from the specimen upon cutting of the cystic mass.

Histological examination:

Hematoxylin and eosin stained histological sections demonstrated a thin (about six to eight cell) layer, rather uniform lining of para-keratinized stratified squamous epithelium with corrugated surface, the basal cell layer is well defined showing palisading columnar cells with hyperchromatic nuclei. Cyst wall is made up of fibrous connective tissue with scattered fibroblasts, and moderately infiltrated by chronic inflammatory cells. Cyst wall is loosely attached to the lining epithelium with foci of detachment being detected along the basal cell layer. The cyst lumen contains abundant keratin and cellular debris. (Figures 1-3).

Radiographic findings:

Orthopantomograph (OPG) revealed an elliptical, unilocular radiolucency in the right mandibular third molar region closely related and attached to the neck of the mandibular third molar, with no evidence of perforation or expansion of the cortical plate. (Figure 4).

Differential diagnosis

Based on the clinical, radiographic, and macroscopic findings, the lesion was initially diagnosed as a dentigerous cyst; though OKC has also been suggested. However, the histopathological features were almost conclusive, and consistent with those of OKC.

Outcome and follow up

The Patient was asymptomatic following three years of treatment. Because of the high recurrence rate, patient should be reviewed at least after five years.

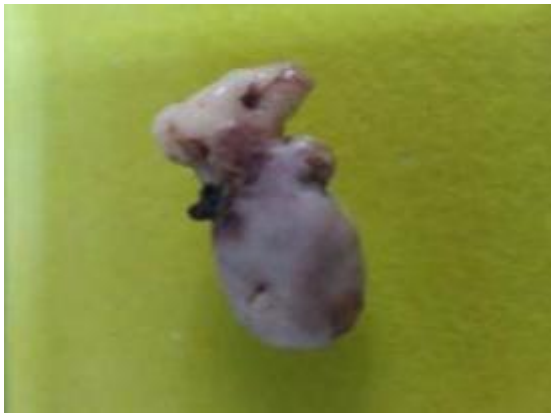

Figure 1: intact cyst wall attached to an impacted molar tooth

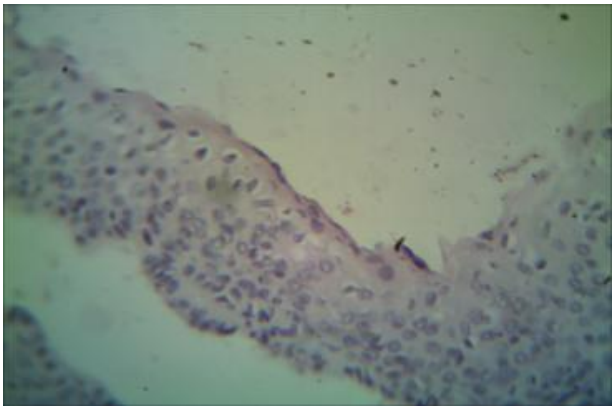

Figure 1: parakeratininzed stratified squamous epthelial lining with currugated surface (40×10)

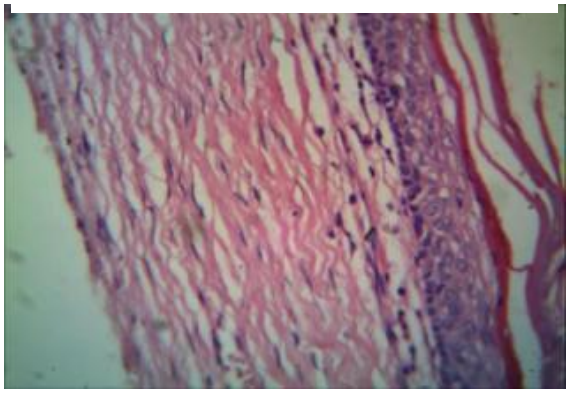

Figure 2: Figure 3: fibrous cyst wall with moderate inflammatory infiltration (40×10)

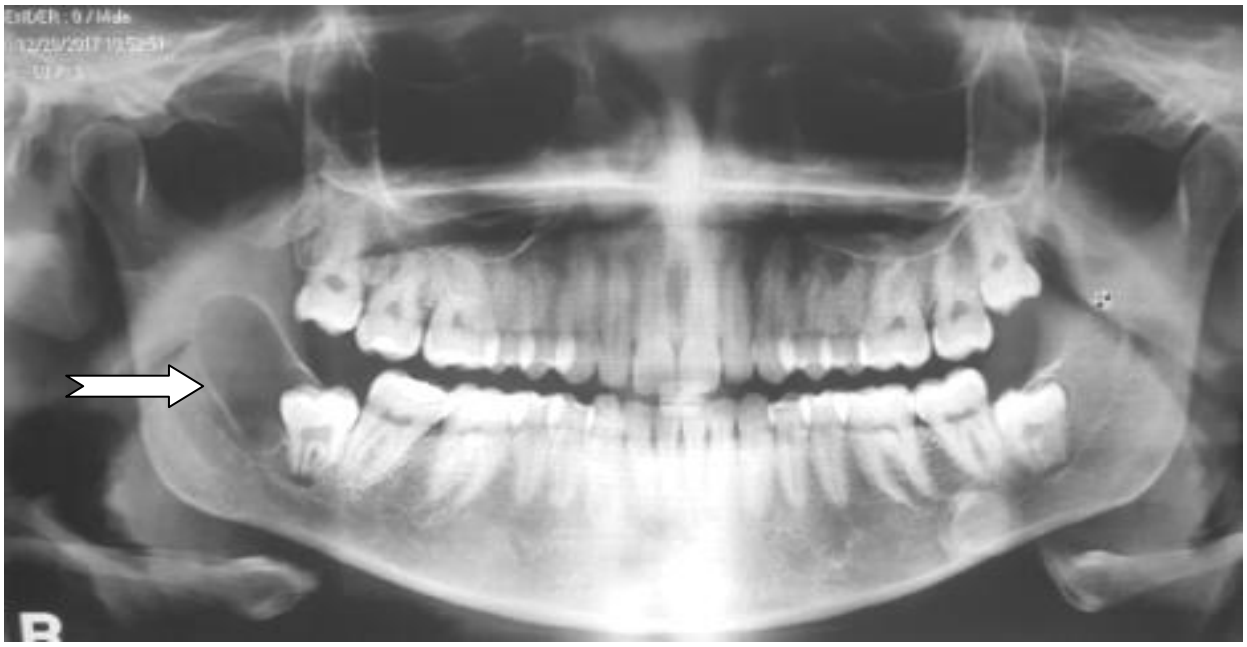

Figure 4: Orthopantamograph showing unilocular radiolucent lesion attached to the third molar tooth extending to the ramus of mandible

DISCUSSION

The odontogenic keratocyst (OKC) has been an area of considerable research by many authors for long time owing to its disputed nature, and aggressive behavior. Many are arguing that the OKC should be classified as a benign neoplasm rather than a cyst [3], which was lastly confirmed by Ahlfors and his colleagues in (1984) who proposed that OKC should be considered as a benign cystic tumor [4]. This notion was further confirmed in 2005 where WHO moved the OKC from cyst list to tumor category and gave the designation of keratocystic odontogenic tumor (KCOT) instead of odontogenic keratocyst (OKC) [5]. However; the updated WHO classification of odontogenic tumors in (2017) has moved the OKC from tumor category back to cyst category due to lack of sufficient evidence [6,7]. The term 'odontogenic keratocyst' was first invented by Philipsen (1956), and has been defined as *"a benign developmental odontogenic tumor with many distinguishing clinical and histologic features*". Among them are: a potential for locally destructive behavior, a relatively high recurrence rate, and designation as a consistent finding in the nevoid basal cell carcinoma syndrome, or Gorlin Goltz syndrome [8].

The origin of OKC has been an area of great debate for many years until 1967 where Soskolne and Shear provided an evidence supporting the origin of OKC from primordial odontogenic epithelium, particularly the remnant of dental lamina [9]. Moreover; a second source of the epithelial lining of OKC was proven to be the basal cell extensions of the overlying epithelium [10].

The odontogenic keratocyst accounts for 4–12% of all odontogenic cysts, and occurs over a wide age range with a peak of incidence in the second and third decades of life, and with slight male predilection [1]. Rare cases were reported as early as the first decade, and as late as the ninth decade of life [11]. The lesion could be discovered anywhere in the jaw; however, about (65–83%) were found in the mandible, particularly the molar region and the ascending ramus [8].

The clinical signs and symptoms of OKC involves: pain, swelling, teeth displacement, and occasionally paresthesia; however, in many cases cysts were discovered accidently on routine radiographic examination, or may go undetected until they reach huge size involving the whole ramus or the maxillary sinus. This is due to the tendency of OKC to grow and expand antero-posteriorly through the bone marrow [1].

Considerable research has been conducted to explore the unique microscopic features of OKC accounting for its aggressiveness and tumor-like behavior. The capsule of OKC is invariably thin and fragile, and almost collapsed during surgical enucle-

ation, that is why such lesions are more likely to recur. The lining epithelium is either ortho or parakeratinized with characteristic morpho and functional differentiation of epithelial cells from the basal cell layer to the surface corneal cell layer. This feature is not found in the other jaw cysts, even when keratinization occurs [1]. Another distinguishing feature proven to account for the possibility of OKC to recur was the presence of small micro-cysts (satellite cysts) in their capsules [7].

Furthermore, an agreement has been made about the significant association between nevoid basal cell syndrome and OKC (about 75%), particularly the Para keratinized type [2].

Further evidence provided by molecular studies concluded that there is increased expression of tumor markers; mainly, PCNA and Ki 67, as well as, mutation of NBCCS gene, and PTCH gene especially in syndrome associated OKCs. All of these findings provided supportive evidence that the OKC might be considered a benign tumor and should be treated accordingly [1]. Even though, some researchers were discreet regarding considering the OKC as a benign neoplasm though they acknowledge the previously mentioned theory. Actually, many oral pathologists agreed that the evidence was not convincing, especially those related to PATCH gene mutation, as mutation has been demonstrated also in non-neoplastic lesions particularly; dentigerous cyst. So the new WHO classification (2017) reverted back to the original and the widely accepted classification of OKC under cyst category [7].

Many OKCs arise in dentigerous relationship making the diagnosis rather confusing, so carful and comprehensive investigations should be conducted as the treatment and the behavior of the two lesions are quite different. Dentigerous cysts are not aggressive and simple enucleation is considered the treatment of choice and recurrence is rare. On the other hand, OKCs are aggressive, may invade the adjacent structure, and are more likely to recur [11]. In 2013, Chaudhary and his colleagues reported a case of OKS which was initially suspected as a dentigerous cyst due to the unusual clinical and radiographic presentations, but the histopathological examination was consisting with OKC [12]. Likewise, in our current study, dentigerous cyst was the first diagnosis which has been made according to the radiographic appearance; the cyst appeared as a unilocular radiolucency associated with crown of an impacted tooth, which is a common feature for the dentigerous cyst [1]. The attention was further drawn toward the dentigerous cyst when the cyst lining along with an impacted tooth was received intact in the laboratory which is rare for the OKC due to its thin scalloped capsule [2].

However, the microscopic examination was almost convincing and conclusive; the cyst lining was made up of keratinized stratified squamous epithelium with corrugated pattern, supported by thin fibrous capsule which is consistent with OKC. Moderate inflammatory infiltrates were detected both in the epithelium and the connective tissue capsule; this raises the question whether the inflammatory infiltrate has altered some of the features of the lesion enabling its complete enucleation without fragmentation. This proposal was made according to the widely accepted theory that the unique characteristic features of the OKC could be greatly altered by the presence of inflammation in its capsule or epithelial lining [13].

In conclusion, OKCs may impede the eruption of related teeth resulting in a radiographic appearance of dentigerous cyst; such lesions are usually misdiagnosed as dentigerous cysts. Our reported case was an OKC attached to an impacted tooth resembling a dentigerous cyst. So, carful and comprehensive examination should be carried out; this will involve thorough clinical and radiographic examination, as well as, carful microscopic evaluation of the surgical specimen. Thus, an appropriate diagnosis and subsequently a proper treatment plane will be established.

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