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Follicular Keratocyst Masquedering as Dentigerous Cyst- A Report of Two Cases

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Authors' contributions

This work was carried out in collaboration between all authors. Author JVRS wrote the draft of the manuscript. Author AS managed the literature searches. Author FB designed the figures, managed literature searches and contributed to the correction of the draft. Author DD provided the case and supervised the work. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Odontogenic keratocyst is one of the most aggressive odontogenic cysts owing to its relatively high recurrence rate and its tendency to invade adjacent tissues. Odontogenic keratocyst associated with unerupted tooth, surrounding the crown with the cyst lining attached to the neck of the tooth in a true dentigerous relationship, are termed as Follicular keratocyst. Follicular keratocyst accounts for 25-40% of all Odontogenic keratocyst's. The aim behind bringing these cases is to help the clinician in modifying the final treatment plan after the final diagnosis, as Odontogenic keratocyst's are potentially aggressive lesions with high recurrence rates in contrast to Dentigerous cysts.

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Keywords: Follicular keratocyst; odontogenic keratocyst; dentigerous cyst.

1. INTRODUCTION

The odontogenic keratocyst is a distinctive form of developmental odontogenic cyst that deserves special consideration because of its specific histopathologic features and clinical behavior [1].

The term "odontogenic keratocyst" was introduced by Philipsen in 1956, while Pindborg and Hansen in 1963, described the essential features of the cyst [2]. Radiographically odontogenic keratocyst can be of different varieties- follicular, envelopmental, replacement, extraneous and collateral [3].

The follicular type of odontogenic keratocyst may be defined as a "cyst with typical histology of an odontogenic keratocyst which surrounds the crown and is attached to the neck of an unerupted tooth". Radiographically it mimic's a dentigerous cyst [2].

2. CASE REPORT 1

A 29 yr old male patient reported to the department of oral diagnosis and radiology with a chief complaint of swelling on the lower left side of the face with difficulty in mouth opening since three weeks. Patient also gave a history of severe pain which relieved on taking medication.

Extraorally a solitary diffuse swelling was seen in the middle third of the face on the left side, extending anteriorly from the parasymphyseal region to about 1 cm away from the angle of the mandible. Superiorly the swelling was extending along the ala-tragus line and inferiorly about 2 cms below the inferior border of the mandible (Fig. 1). Mouth opening was about 12 mm.

Case 1

Fig. 1. Profile

Intraorally swelling was seen distal to 37, which was firm in consistency and tender on palpation. Teeth on the affected side were healthy and 38 was clinically missing.

Radiographically, OPG showed a unilocular radiolucency associated with impacted 38 with well defined borders and extending into the ramus of the mandible upto the coronoid process with 38 being pushed to the inferior border of the mandible (Fig. 2).

Fig. 2. Panaromic radiograph

A provisional diagnosis of Dentigerous Cyst was considered. Complete enucleation and curettage of the lesion followed by primary closure of the wound was carried out. The excised tissue was sent to our department for histopathological examination. On macroscopic examination, tissue was attached to the neck of the tooth (Fig. 3).

Fig. 3. Gross specimen

On microscopic examination, lumen with cystic lining having a parakeratinised stratified squamous epithelium with tall columnar basal cells was evident (Figs. 4, 5). An area of non keratinised epithelium with gradually transition into keratinised epithelium was seen (Fig. 7). Some areas showed separation between epithelium and connective tissue (Fig. 6). Connective tissue was fibrous and showed areas of inflammation and abundant vascularity. A final diagnosis of Follicular keratocyst was given based on these features.

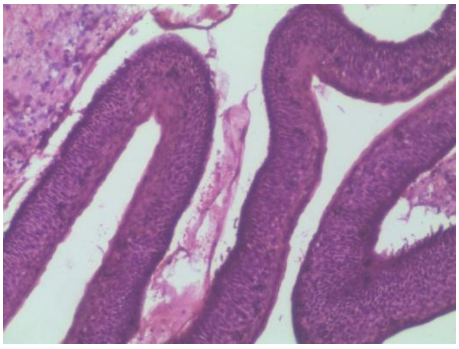

Fig. 4. Cystic lumen showing the typical corrugated epithelium in OKC

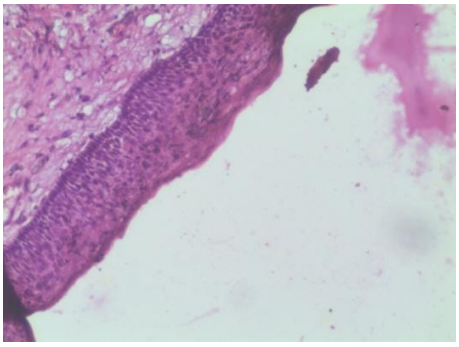

Fig. 5. Cystic lining showing parakeratinised stratified squamous epithelium with tall columnar basal cells

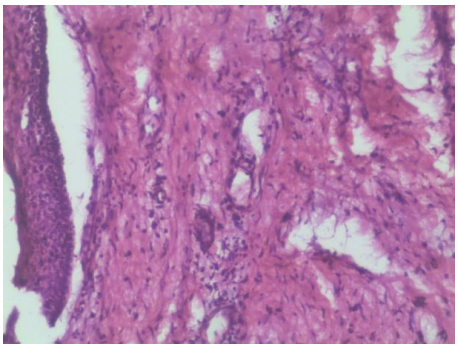

Fig. 6. Separation between cystic lining and capsule

3. CASE REPORT 2

An 11 yr old female patient reported to the department of oral diagnosis and radiology with a complaint of swelling in the middle one third of the face on the right side since 1 month. Patient gave history of an asymptomatic swelling which gradually increased to the present size.

Extraorally, a diffuse swelling measuring about 4 cm x 3 cm extending superiorly to about 3 cm below the infraorbital margin, anteriorly upto the ala of the nose, inferiorly upto the corner of the mouth was evident. Skin over the swelling appeared normal (Fig. 8). On palpation inspectory findings were confirmed. Swelling was non tender, non compressible, bony hard in consistency with no local rise of temperature.

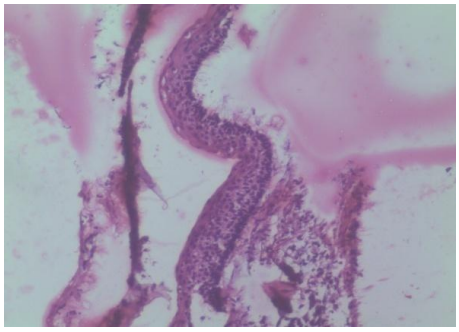

Fig. 7. Transitional area of keratinised epithelium with a non keratinised focal area

Case 2

Fig. 8. Profile

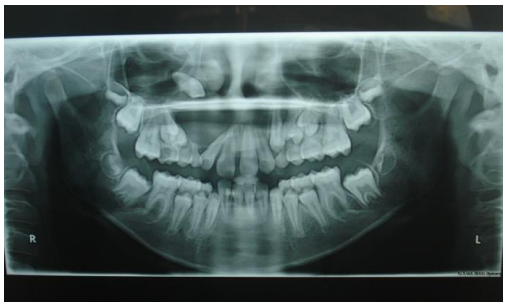

Fig. 9. Panoromic radiograph

Intraorally swelling was seen to be extending from 54 to 11 region. Vestibular obliteration,

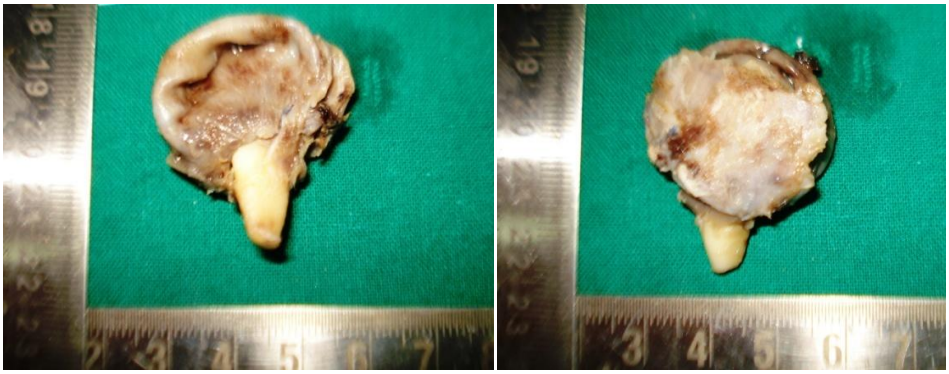

Fig. 10(a,b). Gross speciemen

buccal cortical expansion along with decortications was seen extending from 11 to 16 region.

Radiographically, OPG showed increased follicular space along with well defined radiolucency with sclerotic border surrounding 13 at the CEJ. Root of 12 was tilted mesially (Fig. 9).

A provisional diagnosis of Dentigerous Cyst was considered. Complete enucleation and curettage of the lesion followed by primary closure of the wound was done. The excised tissue was sent to our department for histopathological examination. On macroscopic examination, tissue was attached to the neck of the tooth (Fig. 10a, b).

On microscopic examination, cystic lining having a thin, stratified squamous keratinized epithelium of 6-8 layers thick with loss of rete ridges was seen. Palisading tall columnar cells were seen at the basal portion of the epithelium (Fig. 11). An area of non keratinized epithelium with gradually transition into keratinised epithelium was seen (Fig. 13). The separation between the cystic lining and the capsule could be well appreciated (Fig. 12). Few daughter cysts were also evident. A final diagnosis of Follicular keratocyst was given based on these features.

4. DISCUSSION

OKC's are thought arise from the remnants of dental lamina or the basal layer of the surface epithelium [4,5]. Peak incidence is in 2nd to 3rd decade, male predominance, more commonly seen in mandible than maxilla. Most common location for occurrence is body and ramus of the mandible, in the maxilla it is in the 3rd molar area followed by cuspid region [4].

Radiographically seen as unilocular or multilocular radiolucency with smooth, corticated borders that is often associated with an impacted tooth [6]. Scalloping of the border is a frequent finding [2].

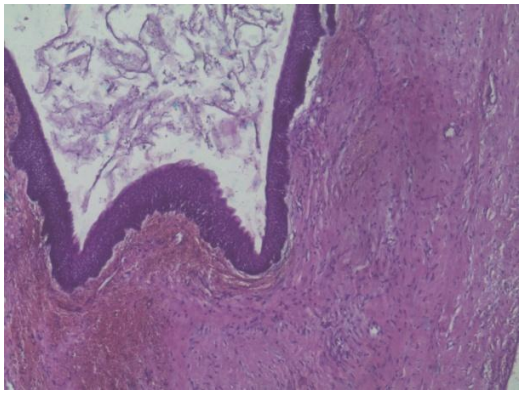

Fig. 11. Cystic lumen showing the typical corrugated epithelium with multiple keratin flakes

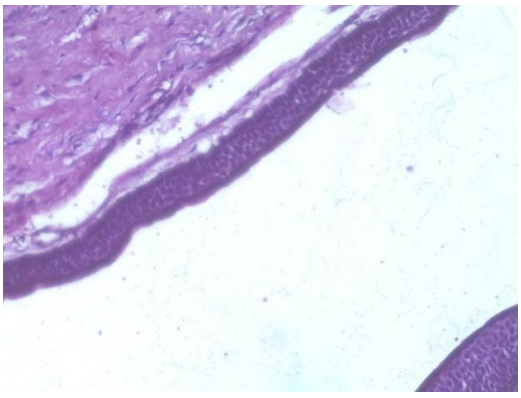

Fig. 12. Separation between cystic lining and capsule

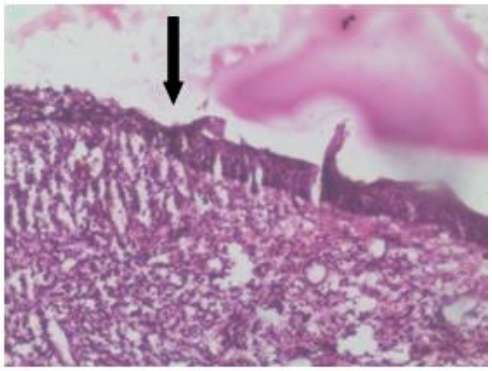

Fig. 13. Transitional area of the epithelium

Histopathologically, the cystic lining is stratified squamous epithelium, parakeratinized or orthokeratinized, [5-8] cell layers thick without rete ridges. The parakeratotic layers have a corrugated surface. There is a well defined, often palisaded basal layer consisting of columnar or cuboidal cells or mixture of both giving a "picket fence" or "tombstone appearance", which was also seen in the present cases [2]. The cells superficial to basal layer are polyhedral and often exhibit intracellular oedema. Mitotic figures are evident in basal and suprabasal layers [2].

Odontogenic keratocysts are often associated with unerupted tooth. Superimposition may result in a dentigerous cyst appearance radiographically [3]. There is no direct connection between the cyst lining and the follicle of the unerupted tooth [2,7]. Recently a case of primodial odontogenic tumor was reported which was associated with un erupted third molar. Primodial odontogenic tumor can also be considered as a radiological differential diagnosis in the present case [8].

It was suggested by Browne that an enlarging keratocyst may involve the follicle of an unerupted tooth and fuses with the reduced enamel epithelium. This concept was developed by Altini and Cohen who have introduced the term "follicular keratocyst" for these groups of lesions in 1982 [2,7]. They studied 17 cases in which the cyst lining was typically odontogenic keratocyst on histological examination but which on macroscopic examination had completely surrounded the crown of the tooth and had been firmly attached to the neck. That portion of the lining immediately adjacent to the neck of the tooth consists of reduced enamel epithelium whereas the rest of the cyst is lined by typical primordial cystic epithelium [2,9].

Altini and Cohen postulated that follicular keratocysts are extrafollicular in origin and might arise following eruption of a tooth into a pre-existing keratocyst cavity in the same way as a tooth erupted into the oral cavity [2,7]. They also suggested that between the epithelium attached to the neck of the tooth and the typical OKC epithelium, a short segment of non keratinized stratified squamous epithelium is seen [2].

OKC can be treated with enucleation and curettage or in advanced cases with segmental resection with no recurrences seen in 3 year follow-up [5]. When considering removal of a keratocyst, however, it is important to keep in mind to eliminate all the possible vital cells left behind in the defect to prevent recurrence. Whether they are from the original lining or derived from microcysts in the wall, they are bound to be located rather superficially in the defect. For this reason a mild, not deeply penetrating, cauterizing agent such as Carnoy's solution should be used [10,11].

In an immunohistochemical study done by Kim DK et al in 2003, it was revealed that the staining pattern and intensity for Ki-67 was same for both the follicular (associated with impacted tooth) and extrafollicular variant of OKC. Thereby suggesting that the aggressiveness of the follicular OKC is similar to the extrafollicular one and should be attended with the same therapeutic approach in order to prevent reccurence [12].

In an immunohistochemical study done by Seifi et al in 2008 to compare the immunoreactivity in different layers and linings of both epitheliums of the follicular cyst and odontogenic keratacyst using BCL-2 anti-apoptotic marker, suggested that the overexpression of BCL-2 in basal layer of odontogenic keratocyst can be considered as a useful marker to differentiate it from follicular cyst [13].

5. CONCLUSION

In the present two cases, the clinical, radiographic and gross findings were favouring the diagnosis to be that of Dentigerous Cyst, however histopathological examination showed predominant OKC lining with a transitional focal area of non keratinised to keratinised epithelium,

guiding to a diagnosis of Follicular Odontogenic Keratocyst.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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