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WHEN AN ODONTOGENIC KERATOCYST MASQUERADES AS A DENTIGEROUS CYST: A CASE REPORT

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ABSTRACT

Background: The Odontogenic Keratocyst (OKC) is a locally aggressive developmental cyst known for its high recurrence rate and its ability to mimic other odontogenic lesions. When an OKC presents as a unilocular radiolucency associated with the crown of an impacted tooth, it frequently leads to a misdiagnosis of a Dentigerous Cyst (DC), particularly in pediatric patients where DCs are more prevalent.**Case Description:** A 12-year-old male presented with a one-month history of a painless, slowly progressing swelling in the left anterior maxilla, resulting in facial asymmetry and an obliterated nasolabial fold. Clinical examination revealed a 3x3 cm, bony-hard, tender swelling associated with an over-retained deciduous lateral incisor (62) and a clinically missing permanent canine (23). Radiographic evaluation (OPG) demonstrated a well-defined, unilocular, oval radiolucency (2.7 x 2.5 cm) extending from the mesial of tooth 21 to 24. The radiolucency enveloped the crown of the superiorly displaced tooth 23 at the cemento-enamel junction (CEJ), mimicking a classic dentigerous relationship. However, histopathological analysis revealed a cystic cavity lined by a corrugated, uniformly thick para-keratinized stratified squamous epithelium with a palisaded basal layer and satellite cysts within the fibrocellular capsule, confirming the diagnosis of an OKC.**Conclusion:** This case underscores the diagnostic challenges posed by OKCs in the maxillary anterior region. In a 12-year-old patient, the clinical and radiographic features of an impacted canine and a pericoronal radiolucency strongly favour a dentigerous cyst; however, the aggressive nature of an OKC necessitates a different surgical approach and long-term surveillance. This report emphasizes that histopathology remains the gold standard for diagnosis and that clinicians must maintain a high index of suspicion for "mimickers" to prevent undertreatment and subsequent recurrence.

KEYWORDS

Odontogenic Keratocyst, Dentigerous Cyst, Maxillary Sinus, Pediatric Oral Radiology, Impacted Canine, Diagnostic Mimicry.

INTRODUCTION

The Odontogenic Keratocyst (OKC) remains one of the most intriguing and clinically challenging entities in oral and maxillofacial pathology. Originally described by Philipsen in 1956, it was briefly reclassified by the World Health Organization (WHO) as a "Keratocystic Odontogenic Tumor" in 2005 due to its aggressive growth and high recurrence rate, before being returned to the "cyst" category in 2017 [1, 8]. This diagnostic "identity crisis" mirrors the clinical challenge we face: an OKC often behaves more like a neoplasm than a simple cyst.

Clinically, the OKC is a "great mimicker." While most cases appear in the posterior mandible, its presence in the maxilla—especially in pediatric patients—is rare and often misleading [3]. When an OKC develops in a pericoronal position, it can perfectly replicate the radiographic presentation of a Dentigerous Cyst (DC) by attaching to the cemento-enamel junction (CEJ) of an impacted tooth [4, 14]. For a young patient, a misdiagnosis at this stage is more than a clerical error; it dictates the surgical approach. A simple enucleation suitable for a DC may prove insufficient for an OKC, which frequently harbours satellite cysts in its wall, leading to a high probability of recurrence [10,15].

This case report details a significant diagnostic dilemma in a 12-year-old male. We describe a large, expansile maxillary lesion that clinically and radiographically pointed toward a dentigerous cyst associated with an impacted canine, but was ultimately revealed to be an Odontogenic Keratocyst upon histopathological examination. By highlighting this "wolf in sheep's clothing" presentation, we aim to underscore the vital role of advanced imaging and microscopic analysis in guiding the management of pediatric jaw lesions [6].

Case Presentation

A 12-year-old male patient presented with a one-month history of a painless, insidious swelling in the left anterior maxillary region. The swelling had gradually progressed, resulting in facial asymmetry and difficulty in mastication. Extra-oral examination revealed a **3 x 3 cm**, well-defined, dome-shaped, and bony-hard swelling extending from the left ala of the nose to the outer canthus of the eye. The nasolabial fold was obliterated, and the area was tender and warm upon palpation, suggesting secondary infection.

Intra-orally, a 1 x 1 cm(approximately) swelling was noted in the labial vestibule, extending from the over-retained 62 to the 63 region. Clinical findings included an over-retained 62, 63 with physiologic mobility(62), a missing permanent 22, 23, 24, and a palpable eruption bulge in the 22 region.[figure 1]

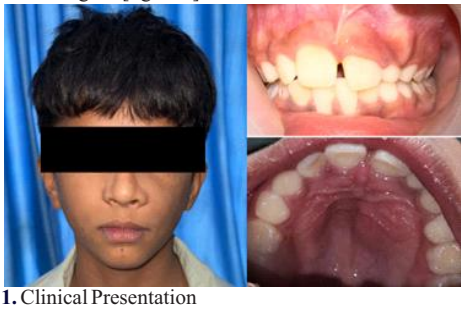

Figure 1. Clinical Presentation

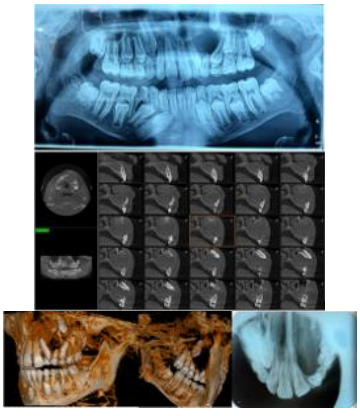

Figure 2. radiographic examination

An orthopantomogram (OPG) revealed a solitary, unilocular, oval radiolucency with thin corticated borders, measuring approximately 2.7 x 2.5 cm. The lesion extended mesiodistally from the 21 to the 64 region and superoinferiorly from the alveolar crest to the nasal floor. Notably, the radiolucency enveloped the crown of the superiorly displaced 23 at the cemento-enamel junction (CEJ), a classic radiographic feature of a dentigerous cyst. CBCT revealed unilocular cystic lesion with evident expansion of cortical plate bucco-palatally in region with anterior maxilla on left side.[figure 2]

Following surgical intervention, the specimen was submitted for histopathological analysis. The cystic cavity was filled with keratin flakes and lined by a uniformly thick, corrugated parakeratinized stratified squamous epithelium (6–8 layers thick). The basal layer exhibited characteristic palisading of hyperchromatic nuclei (picket-fence appearance). The fibrocellular connective tissue wall contained isolated strands of odontogenic epithelial rests, confirming the diagnosis of an Odontogenic Keratocyst (OKC).

DISCUSSION

Odontogenic keratocyst (OKC) is a developmental cyst that arises from remnants of the dental lamina. Although it is classified as a cyst, it often exhibits aggressive biological behaviour and a tendency to recur, which makes it clinically significant. The lesion has been widely studied because of its unique histopathological features and growth pattern within the jaws.[3,11]

OKC most frequently affects the posterior mandible, particularly in individuals during the second and third decades of life. Involvement of the maxilla is comparatively less common, and lesions in this region may present diagnostic difficulties. One notable characteristic of OKC is its ability to grow anteroposteriorly within the cancellous bone with minimal expansion of the cortical plates, which may allow the lesion to reach a considerable size before clinical detection.[1,14]

The Diagnostic Trap of "Dentigerous" Presentation

The primary challenge in this case was the radiographic "dentigerous" presentation. Statistically, the **dentigerous cyst (DC)** is the most common developmental cyst in children, usually associated with an impacted maxillary canine.[4] When a radiolucency appears to attach to the cemento-enamel junction (CEJ) of an unerupted tooth, as seen in our patient's OPG, a diagnosis of DC is almost always the first consideration.[14]

However, literature suggests that an OKC can mimic this relationship in two ways:

1. The Follicular Variant: The OKC originates from the dental lamina within the follicle before the tooth erupts.

2. Envelopment: An extra-follicular OKC expands and eventually envelops the crown of a nearby unerupted tooth.[2,6]

Radiographically, OKC usually appears as a well-defined radiolucency with corticated margins, which may be either unilocular or multilocular. In certain situations, particularly when the lesion is associated with an impacted tooth, its radiographic appearance may closely resemble that of a dentigerous cyst. Dentigerous cysts typically present as radiolucencies surrounding the crown of an unerupted tooth and attached at the cementoenamel junction. Because of this similarity, distinguishing between these two lesions based solely on radiographic findings can be challenging.[1,5]

In the present case, the lesion appeared as a well-defined radiolucency enveloping the crown of an impacted maxillary canine, which initially suggested a dentigerous cyst. Similar diagnostic dilemmas have been reported in previous studies where odontogenic keratocysts mimicked dentigerous cysts due to overlapping clinical and radiographic features.[2,4,6] This highlights the importance of further diagnostic evaluation when managing cystic lesions associated with impacted teeth. Histopathological examination plays a crucial role in establishing the final diagnosis. OKC typically demonstrates a thin parakeratinized stratified squamous epithelial lining with a corrugated surface and palisaded basal cells, and the cystic lumen often contains keratin debris. The connective tissue wall may also show odontogenic epithelial remnants and mild inflammatory infiltrate.[3,9] The microscopic features observed in the present case were consistent with these classical findings, confirming the diagnosis of odontogenic keratocyst.

Management of OKC generally involves surgical enucleation, marsupialization, or more extensive procedures depending on the size and location of the lesion. In a 12-year-old, the management of such a lesion requires a delicate balance. While aggressive treatment like peripheral ostectomy or the application of Carnoy's solution is recommended to reduce recurrence, clinicians must be mindful of the developing dentition and the growth centers of the maxilla.[13, 15] Even with appropriate treatment, the possibility of recurrence remains a concern; therefore, long-term follow-up is recommended.

This case emphasizes that odontogenic keratocysts can closely mimic dentigerous cysts in their clinical and radiographic presentation, particularly when associated with impacted teeth. Careful correlation of clinical findings, imaging, and histopathological examination is essential to reach an accurate diagnosis and ensure appropriate management.

CONCLUSION

Odontogenic keratocyst can sometimes present with clinical and radiographic features that closely resemble other odontogenic cysts, particularly dentigerous cysts when associated with impacted teeth. Because of this overlap, it may be difficult to establish an accurate diagnosis based only on clinical examination and radiographic findings.

The present case highlights the importance of histopathological evaluation in confirming the final diagnosis. Identifying odontogenic keratocyst correctly is essential because of its potential for aggressive growth and recurrence, which may influence treatment decisions and the need for long-term follow-up.

Therefore, a combined approach involving clinical assessment, radiographic interpretation, and histopathological analysis is crucial for accurate diagnosis and appropriate management of such lesions.

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