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Expert Opinion Article

Unmasking The True Pathology: Differentiating Between OKC And Follicular Ameloblastoma

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Abstract
Diagnosing cystic lesions in the oral cavity can be tricky due to overlapping features. This case report tells the story of a 34-year-old man who came to our clinic with a painful swelling in his left jaw. Initially, it looked like an odontogenic keratocyst (OKC) based on x-rays and clinical signs. However, a detailed histopathological exam revealed it was actually a follicular ameloblastoma. This case highlights the importance of thorough diagnostic tests and teamwork among healthcare providers to get the right diagnosis and treatment. It also emphasizes the need for long-term follow-up because both OKC and Follicular ameloblastoma (FA) can come back after treatment.

Keywords: odontogenic keratocyst, Follicular ameloblastoma, jaw cyst, histopathology, diagnosis, recurrence, multidisciplinary approach.

INTRODUCTION

Diagnosing cystic lesions in the oral cavity can be quite challenging due to overlapping clinical, radiographic, and histopathological features among different pathologies. Two entities that often cause diagnostic confusion are odontogenic keratocyst (OKC) and Follicular ameloblastoma (FA). Both are derived from odontogenic epithelium and primarily affect the jawbones, especially the mandible. They share several clinical and radiographic characteristics, making it difficult to differentiate them, which can influence patient management and prognosis.

Odontogenic keratocysts, previously known as keratocystic odontogenic tumors, are aggressive, have a high recurrence rate, and can cause significant local bone destruction. They are often associated with the posterior mandible and can grow to a considerable size before symptoms appear, usually presenting as a painless swelling. OKCs are sometimes discovered incidentally on routine radiographs. Radiographically, they appear as well-defined, unilocular or multilocular radiolucencies with smooth, corticated borders, which can be mistaken for other cystic or neoplastic lesions. The prevalence ratio in the upper jaw versus the lower jaw is 1:7 for dentigerous type and 1:4.7 for non-dentigerous type.[1]

Ameloblastomas are common benign neoplasms that frequently arise in the molar and ramus regions of the mandible. The global incidence of ameloblastoma is about 0.5 cases per million people, with 10– 15% of cases occurring in the pediatric population, reaching up to 25% in Africa and Asia.[2] In 2005, the World Health Organization classified ameloblastomas into solid/multicystic, extraosseous/peripheral, desmoplastic, and unicystic types. Solid/multicystic ameloblastomas were further divided into follicular and plexiform types.[3] Follicular ameloblastomas, a variant of ameloblastoma, tend to present as a single cystic cavity. They are most commonly seen in younger patients and are usually located in the posterior mandible, similar to OKCs. Clinically, FAs may manifest as asymptomatic swellings, often remaining undetected until they are discovered during routine dental exams or have grown considerably. Radiographically, FAs present as well-demarcated radiolucencies, frequently multilocular, mimicking the appearance of odontogenic keratocysts.

The overlapping clinical presentations of OKCs and FAs often include asymptomatic jaw expansion, delayed tooth eruption, and displacement or resorption of adjacent teeth. Both conditions may have a slow-growing, progressive nature, leading to jawbone expansion and facial asymmetry. Given their similar presentations, thorough diagnostic work, including clinical examination, imaging, and histopathological evaluation, is crucial for accurate differentiation.

Despite these similarities, the underlying pathology and management strategies for OKCs and FAs differ significantly. OKCs are treated primarily through surgical excision, with a focus on minimizing recurrence, often employing methods such as enucleation, curettage, or more radical approaches depending on the lesion's extent. In contrast, the management of FAs may range from conservative approaches like enucleation and marsupialization to more aggressive resections, depending on the histopathological subtype and the tumor's extent.

In this case report, we explore a diagnostic conundrum where the clinical and radiographic findings pointed towards both an odontogenic keratocyst and a follicular ameloblastoma, making the differential diagnosis particularly challenging. This case underscores the importance of a multidisciplinary approach in evaluating and managing jaw lesions, incorporating clinical insights, advanced imaging techniques, and definitive histopathological analysis to ensure accurate diagnosis and appropriate treatment planning.

Similarities Between Odontogenic Keratocyst and Follicular Ameloblastoma:

1. **Clinical Presentation**:

- Location: Both OKCs and FAs predominantly occur in the posterior mandible but can also be found in the maxilla. They often present as painless swellings or asymptomatic jaw expansions.
- Age of Onset: Both lesions commonly present in young adults, though FAs tend to affect a slightly younger demographic. The most common age of presentation of ameloblastoma is 30-60 years, with a slight male preponderance, and the most common site is the mandible.[4]
- Symptoms: Initial stages of both pathologies are typically asymptomatic, leading to delayed diagnosis until they become large enough to cause noticeable jaw expansion, discomfort, or interfere with tooth eruption.

2. **Radiographic Features**:

- Radiolucency: Both lesions appear as well-defined radiolucent areas on radiographs. They can be unilocular or multilocular, though FAs are more often unilocular.
- Borders: The borders of the lesions are usually smooth and corticated, making it difficult to distinguish between the two based on imaging alone.
- Impact on Surrounding Structures: Both lesions can cause displacement or resorption of adjacent teeth and expansion of the cortical bone. The presence of a radiolucent lesion with these effects is a common finding in both OKCs and FAs. [5]

3. **Histopathological Considerations**:

- Although histopathological analysis is essential for definitive diagnosis, initial biopsy specimens may not always distinguish between OKCs and FAs due to overlapping microscopic features. Both lesions exhibit cystic spaces lined by odontogenic epithelium, necessitating careful examination for distinguishing characteristics.

Given these overlapping features, clinicians must exercise a high degree of suspicion and employ comprehensive diagnostic evaluations to differentiate between odontogenic keratocyst and follicular ameloblastoma accurately. This differentiation is critical as it dictates the appropriate therapeutic approach and informs the prognosis for the patient.

CASE REPORT

A 34-year-old male(Figure 1) presented to the Department of Oral and Maxillofacial Surgery at Awadh Dental College and Hospital in East Singhbhum, with a chief complaint of pain and swelling on the left side of his mandible, persisting for four weeks. The swelling had gradually increased in size and was associated with a dull, throbbing pain. Upon extra-oral examination, a draining sinus tract was noted, with purulent discharge suggestive of an underlying infection. The area was tender on palpation. Intra- oral examination revealed significant asymmetry and erythematous swelling over the mandibular region. An orthopantomogram (OPG)(Figure 2) displayed a large unilocular radiolucency in the left mandible, extending from the first premolar to the mandibular angle, with an impacted tooth within the lesion. The well-corticated borders and displacement of adjacent teeth raised suspicions of a cystic nature.

Figure 1

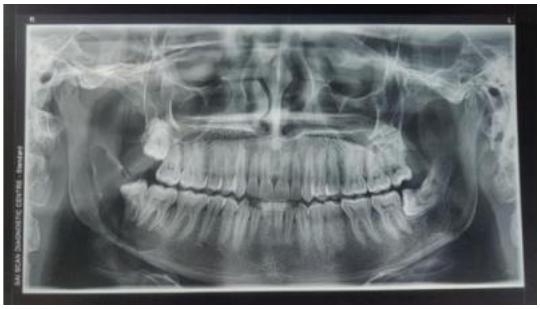

Figure 2

Aspiration of the lesion yielded pus, indicative of an infected cystic cavity. Correlating the clinical and radiographic features, a provisional diagnosis of odontogenic keratocyst (OKC) was made. Subsequently, the patient underwent a complete excisional biopsy under general anesthesia. The lesion, along with the impacted tooth, was enucleated, and the specimen was sent for histopathological analysis(Figure 3).

Figure 3

Histopathological examination revealed a cystic lumen lined by parakeratinized stratified squamous epithelium with a corrugated superficial layer, characteristics consistent with OKC. However, further analysis showed features of a follicular ameloblastoma (Figure 4). This included islands of ameloblastic epithelium with peripheral palisading and reverse nuclear polarity, and a central stellate reticulum-like area, indicative of follicular ameloblastoma rather than an odontogenic keratocyst.

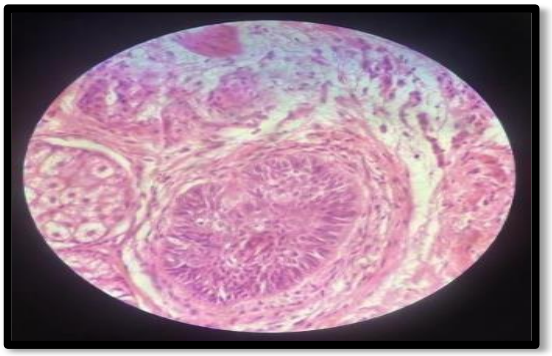

Figure 4

Following surgery, the patient's recovery was uneventful. He was monitored closely over several weeks with periodic clinical evaluations and radiographs. Follow-up radiographs demonstrated progressive bone regeneration at the surgical site, and the patient reported complete resolution of symptoms with no evidence of recurrence.

This case highlights the diagnostic challenges posed by the overlapping features of odontogenic keratocysts and follicular ameloblastomas. Despite initial clinical and radiographic findings suggestive of an OKC, definitive diagnosis required detailed histopathological analysis, which identified the lesion as a follicular ameloblastoma. This distinction is crucial, as it dictates different treatment protocols and has significant implications for prognosis. The case underscores the necessity of a comprehensive approach to diagnosis and management, including meticulous histopathological evaluation, to ensure accurate diagnosis and optimal patient outcomes

A study by Sujata et al. found that ameloblastoma, despite being benign, is aggressive and has a high recurrence rate. A 21-year retrospective study of 92 patients in a tertiary care hospital found that 90% had mandibular involvement, with the most common site being the Body and Angle and Ramus region. The most common histological subtype was plexiform ameloblastoma. The study concluded that adequate surgical planning and long-term follow-up can reduce recurrence rates.

Discussion

This case report highlights the challenges in diagnosing odontogenic keratocysts (OKC) and follicular ameloblastomas (FA), further complicated by a secondary infection leading to a sinus tract formation.

OKCs and UAs commonly affect the lower jaw, making up around 10-15% of all cystic lesions. UA, which is typically found in younger individuals with an average age around 23 years, tends to localize in the back part of the mandible and often remains asymptomatic until it grows significantly. [6]

Clinical and radiographic differentiation between OKCs and FAs are challenging due to their similar presentations.[7] In this case, a large unilocular radiolucent area in the left lower jaw extending from the first premolar to the mandibular angle, containing an impacted tooth, raised suspensions of a cystic lesion. Both entities manifest as well-defined radiolucent areas on imaging, complicating the diagnosis based solely on radiographic features.

Histopathological examination is crucial for an accurate diagnosis. Initially diagnosed as an OKC based on clinical and radiographic findings, further analysis revealed a cystic tumor lined with parakeratinized stratified squamous epithelium and a corrugated superficial layer, which is consistent with OKC. However, additional features such as islands of ameloblastic epithelium with peripheral palisading and reverse nuclear polarity suggested a diagnosis of follicular ameloblastoma.[8]

Classification and management of these lesions involve nuanced considerations. The World Health Organization's reclassification of OKC as a benign neoplasm highlights its potential for aggressive behavior, contrasting with the varied nature of ameloblastomas, which are categorized into solid, multicystic, unicystic, and peripheral types. Treatment strategies range from radical surgical approaches like "en bloc" resection for smaller lesions to conservative measures such as enucleation and curettage for larger, non-invasive mandibular lesions.[9]

Research provide insights into understanding these lesions. Studies indicate the prevalence of hybrid Desmoplastic/Follicular ameloblastomas in the mandible and emphasizes the importance of meticulous surgical planning to mitigate recurrence rates.[10]

Long-term follow-up is critical due to the high recurrence rates of both OKCs and ameloblastomas. Monitoring should include yearly evaluations during the initial five years post-treatment, transitioning to biennial check-ups for at least 25 years to detect late recurrences and ensure optimal patient management.

Conclusion

The overlap in clinical and radiological features

between odontogenic keratocysts and Follicular ameloblastomas presents a diagnostic challenge that requires a thorough histopathological examination for accurate differentiation. Effective management necessitates tailored treatment approaches and vigilant long-term follow-up to address the high recurrence rates associated with these lesions.

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