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Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology

Journal homepage: www.joooo.org



Case Report

An odontogenic keratocyst: Correlating histopathological and clinicoradiographic features avoids a diagnostic dilemma

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ARTICLE INFO

Article history:

Received 28-02-2022

Accepted 05-03-2022

Available online 25-03-2022

Keywords:

Histopathologic dilemma

OKC

Infected cyst

ABSTRACT

Odontogenic lesions are often seen as challenging diagnostic lesions. Odontogenic keratocyst (OKC) is a developmental odontogenic cyst, characterized by unique and distinct histopathologic characteristics and an aggressive biologic behavior. Inflammatory changes in the cystic lining epithelium may result in diagnostic dilemma histologically. In Such cases one should consider clinicoradiographic features for better diagnosis. As such lesions has high recurrence rate it is necessary to perform a periodic radiographic monitoring. This article integrate the necessary clinical and radiographic findings required to address the odontogenic keratocyst.

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1. Introduction

The potential for histopathological confusion between odontogenic cysts has long been known. Distinguishing between various odontogenic cyst and tumor is essential because contrast in treatment is stark.¹ As epithelial and mesenchymal tissues remaining after completion of dentition are subjected to influence of inflammation, largely as sequelae of dental caries and periodontitis. These lesions are common only infrequently cause diagnostic confusion. Other lesions, such as odontogenic cysts and odontogenic tumours are rarer having varied clinical, radiological, histopathological features can lead to uncertainty in diagnosis. This can be ameliorated by careful attention to appropriate clinicoradiographic information.²

Odontogenic keratocyst (OKC) is a developmental odontogenic cyst, characterized by unique and distinct histopathologic characteristics and an aggressive biologic behavior. The frequency of OKC has been reported to vary from 3% to 11% of odontogenic cysts. The origin of OKC

is not clear, it is from dental lamina remnants, basal cells of overlying epithelium or reduced enamel epithelium of dental follicle, detected accidentally during radiographic imaging, as a unilocular or multilocular radiolucency with sclerotic border. It is usually asymptomatic, but may associated with pain and swelling.³⁻⁵

This article highlight the importance of clinicoradiographic findings for differentiating OKC when diagnostic dilemma arise histologically.

2. Case Report

A 17 years old male patient reported with chief complaint of pain and swelling on lower right back jaw region since 1 month. Before 1 month, he felt intermittent, throbbing pain in lower back jaw region which was relieved on taking medication. Again, he noticed swelling on lower front and back jaw region which was gradually increasing. Patient had past history of trauma before 2 years, and he got fracture irt 41 with pain which relieved on taking medication. No significant past medical, family history.

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Fig. 1: Shows well defined extraoral swelling in lower front jaw region and intraoral findings shows vestibular obliteration and fractured tooth irt 41

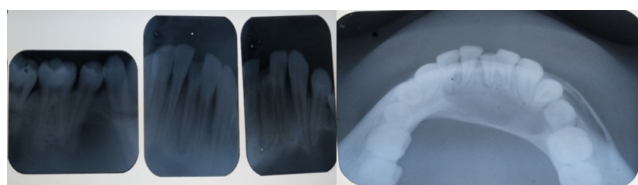


Fig. 2: Intraoral radiographs shows unilocular radiolucency extending from 32 to distal root of 46 with well-defined periphery and widening of PDL space, root resorption was seen irt 44,45,46. Distal displacement irt 45. Diffuse radiolucency with loss of lamina dura irt apical region of fractured 41 suggestive of periapical abscess irt 41 and OKC of body of mandible.



Fig. 3: OPG showing A) Lesion at 1st visit B) Regression of lesion after decompression c) Follow up after 1 year of excision of lesion

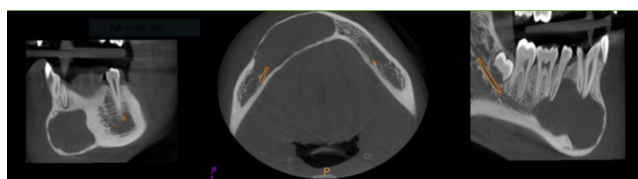


Fig. 4: 3D CBCT showing unilocular hypodense lesion irt 32,31,41,42,43,44,45,46. Displacement of root of premolar distally with mild resorption of apex of 43. There was buccal plate perforation and thinning of buccal cortex with minimal expansion suggestive of periapical abscess irt 41 and OKC of body of mandible.

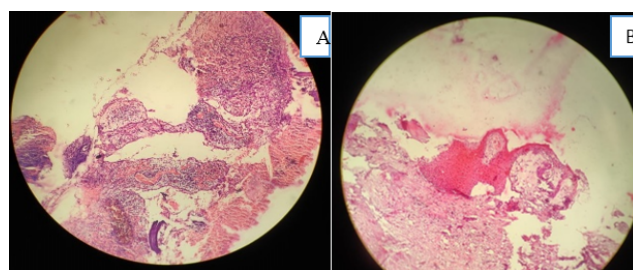


Fig. 5: A) Proliferating non keratinized stratified squamous epithelium, connective tissue capsule is severely infiltrated by acute and chronic inflammatory cells B) Proliferating non keratinized stratified squamous epithelium forming arcades into connective tissue capsule in which severe chronic inflammatory cell infiltration

On extraoral examination, there was single, diffuse swelling of approximately 3x5 cm size present in anterior mandible extending mediolaterally from left parasymphysis to right side approximately 2 cm anterior to angle of mandible, superior-inferiorly from lower third of face to 1 cm below inferior border of mandible with normal overlying skin. (Figure 1) On palpation swelling was non-warm, non tender, and soft to cystic consistency. Right submandibular lymph nodes were enlarged, palpable, not tender, not fixed.

On intraoral examination, 41 shows fracture upto cervical third of crown. Obliteration of buccal vestibule in canine- premolar region with normal overlying mucosa. On palpation, there was well-defined swelling which shows buccal expansion extending from 32-46 without egg shell crackling. (Figure 1) Grade I mobility with tenderness on percussion irt 41-46. All teeth in lower right quadrant and incisors in left quadrant were non-vital. Based on clinical examination provisional diagnosis of chronic periapical abscess with acute exacerbation irt 41 and differential diagnosis of radicular cyst, lateral periodontal cyst (LPC), OKC were considered.

Conventional imaging showed unilocular radiolucency of approximately 6x3 cm size extending from 32 to distal root of 46 with well defined corticated periphery. Internal structure was completely radiolucent with resorption of root apex irt 43,44. Loss of lamina dura irt apical region of 44-46 with bodily displacement of 45. Thinning of inferior border of mandible extending from 32-46. Diffuse periapical radiolucency was noted in periapical region of 41. 3D Cone beam computed tomography (CBCT) revealing unilocular hypodense lesion present in right body of mandible with thinning and perforation of buccal cortex and minimal expansion. Fine needle aspiration cytology showed negative aspiration. Provisional diagnosis of OKC of right body of mandible and periapical abscess irt 41 was considered. Radicular cyst, lateral periodontal cyst and ameloblastoma were kept as differential diagnosis and referred to oral surgery.

Decompression and incisional biopsy of lesion was performed with extraction irt 41,42,45 with placement of draining tube and specimen was sent for histopathological evaluation which showed proliferating non keratinized stratified squamous lining epithelium, severely infiltrated connective tissue by acute and chronic inflammatory cells, plenty of dilated, engorged blood vessels and extravasated RBCs favouring Infected cyst.(Figure 5 A) After 7 months follow up, lesion size was reduced and enuculation of lesion with extraction of 43,44 was performed with placement of carnoy's solution. Specimen showed proliferating non keratinized stratified squamous epithelium forming arcades into connective tissue capsule with severe chronic inflammatory cell infiltration suggestive of Residual cyst.(Figure 5 B) Patient was kept for follow up, no recurrence found.

3. Discussion

The term 'odontogenic keratocyst' was introduced by Philipsen in 1956. According to WHO classification (1971-1992), there were two subtypes- parakeratinized and orthokeratinized. In 2005, parakeratinized type classified as "keratocystic odontogenic tumor" (KCOT) and orthokeratinized type as an orthokeratinized odontogenic cyst. According to WHO classification 2017, KCOTs were again reclassified as OKC.³⁻⁶ The peak incidence is in second and third decade with male predilection. Mandible is frequently affected than maxilla in ramus third molar area, followed by 1st-2nd molar, anterior mandible. Radiographically, present as well-defined unilocular or multilocular radiolucency extending in antero-posterior direction with smooth, corticated margins. In present case clinicoradiographic findings are as mentioned in literature.^{3,4,7,8} Recurrence rate between 13% and 60%, no recurrence was found in present case.

Radicular cysts are periapical lesions associated with nonvital teeth. Radiographically, present as well-demarcated periapical radiolucency. LPC is a non-inflammatory cyst that arises in close proximity of roots of vital teeth present as an asymptomatic gingival swelling most frequently in the mandibular canine-premolar regions. Radiographically present as well circumscribed ovoid or round radiolucency on lateral surface of root. Ameloblastoma is a multilocular cystic lesion. It may be unilocular characteristically producing jaw expansion, thinning of the cortical plate.^{3,4,8,9} In present case, clinically minimal expansion with large unilocular radiolucency growing in mediolateral direction in anterior body of mandible with thin corticated margin favors diagnosis of OKC.

Histologically, OKC consist of stratified squamous epithelium which has highly characteristic-'picket fence' appearance produces orthokeratin, parakeratin, or both.³⁻⁶ Inflammation masks diagnostic features of

tissue suggested to be responsible for transformation of keratinized to non-keratinized epithelium.¹⁰ In present case, no definitive type of lining epithelium was seen histologically creating diagnostic confusion. Diagnosis of OKC is largely based on histological examination. However, Inflammation in fibrous wall results in significant changes in histological features. Proliferation of epithelial cells and loss of parakeratosis and palisaded basal layer result in appearance of nonspecific inflamed odontogenic cyst. If these changes affect larger parts of cyst it may be very difficult to establish definitive diagnosis. This might be a reason for why definitive diagnosis of odontogenic cyst with inflammation cannot offered. Chen et al. reported 3.4% error in diagnosis of lesions such as OKC due to masking inflammation. In a series of 112 OKCs, M. Andric et al. noticed inflammation of fibrous wall as much as 76% of cases. They also suggested biopsy sample taken during decompression and marsupplization are not representative.^{6,10,11} These factors possibly can be reason of diagnostic dilemma in our case. In such cases we should consider clinicoradiographic findings with histological findings to derive definitive diagnosis. Proper evaluation of clinicoradiographic findings are important because significantly different treatments required for each and higher possibility of recurrence in OKC demands longer follow-up.

4. Conclusion

"Tissue Pathways for Head and Neck Pathology" issued by the Royal College of Pathologists states clinical information and preferably radiographic examination is required for accurate diagnosis" of cysts of jaws.¹ Histopathologically inflammation has puzzling effect on epithelial lining, it can obscure characteristic histologic appearance of lesion. In such cases entire lesion requires clinicoradiographic evaluation to reach a definitive diagnosis.

5. Source of Funding

None.

6. Conflict of Interest

None.

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Cite this article: J Parikh S, Patel H, S Shah J. An odontogenic keratocyst: Correlating histopathological and clinicoradiographic features avoids a diagnostic dilemma. *J Oral Med, Oral Surg, Oral Pathol, Oral Radiol* 2022;8(1):45-48.