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Peripheral Keratocystic Odontogenic Tumor in the Anterior Mandible, A Clinical Case Report

Ali Forouzanfar[1]

[1]Associate Professor, Department of Periodontics, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.

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ABSTRACT

Keratocystic odontogenic tumour formerly known as Odontogenic keratocyst (OKC) is a developmental uni- or multi-cystic intraosseous tumor originating from the cell rests of dental lamina. It is known as a rare and benign but locally aggressive tumor with a high recurrence rate that can occur anywhere in the jaw but usually is seen at the posterior part of the mandible. This article represents an unusual case of OKC that has occurred as an inter-radicular lesion between roots of lateral incisor and canine of the mandible with gingival bulging and discharge.

1. Introduction

Odontogenic keratocyst was first identified in 1876. It was described as a developmental odontogenic cyst by Phillipsen in 1956.[1] Considering the neoplastic nature of the lesion, in 2005 World Health Organization (WHO) renamed the lesion to keratocystic odontogenic tumour (KCOT) and defined it as a benign uni- or multicystic, intraosseous tumor that originates from the cell rests of dental lamina which has the ability of keratinization, proliferation, and infiltration of connective tissue during teeth development.
Study of J Henley et al indicated that majority of KOCs harbour chromosomal abnormalities.[2] In 2017 however,WHO reclassified okc into cystic lesions. OKC may be found at any age but usually occurs in second and third decades of life. Since the radiographic appearance of the lesion varies from a small unilocular radiolucency to a large multilocular radiolucency, it can be easily mistaken as a periapical or periodontal lesion.[3]
Although, odontogenic keratocyst can occur in any part of the jaw, the majority of cases occur in the mandible, most commonly in the posterior body and ramus (near 50 percent of all cases). Small lesions are usually asymptomatic and may be found in radiographic examination, but larger cysts can cause pain, swelling or discharge. Odontogenic keratocyst has been taken into consideration because of its special characteristics such as aggressive behavior, central antero-posterior medullary bone growth, and high recurrence rate. Histopathologically the odontogenic keratocyst shows uniformly thin parakeratinised stratified squamous epithelium with six to eight cell layers.[4] The basal epithelial layer is composed of a palisaded layer of cuboidal or columnar epithelial cells, which are often hyperchromatic. The cystic lumen may contain transudate of serum or keratin debris. Radiolucencies may be unilocular (80 percent of cases) or multilocular.[5] Since the radiographic findings of odontogenic keratocyst are not exclusive, the diagnosis is based on the microscopic histopathologic features of the lesion. Differential diagnosis of odontogenic keratocyst includes radicular cyst, residual cyst, dentigerous cyst and lateral periodontal cyst[6]. This article represents an unusual case of inter-radicular odontogenic keratocyst of the anterior mandible.

2. Case Presentation

A 50-year-old woman was referred to the Department of Periodontology and Implant Dentistry at Mashhad University of Medical Science with the chief complaint of gingival swelling in the anterior left part of the mandible. Her past medical history and general physical examinations were unremarkable. The swelling had gradually increased during past 3 months and was discovered to be fluctuant in palpation. The patient reported no pain or discomfort in the swollen gingival area but a slight mobility had observed for the neighboring teeth. During gingival examination of teeth number 21 and 22 the periodontal probe penetrated through the gingival sulcus for several millimeters and yellowish pus-like discharge came out instead of bleeding on probing. Radiographic examination revealed a well-defined cyst-like radiolucent lesion between the roots of mandibular canine and first premolar. Diverted roots of adjacent teeth but no root resorption was obvious.

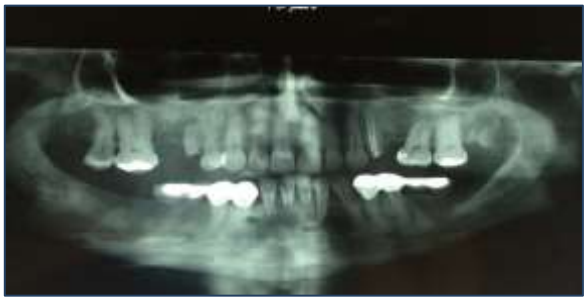

Figure 1-radiographic appearance of the lesion

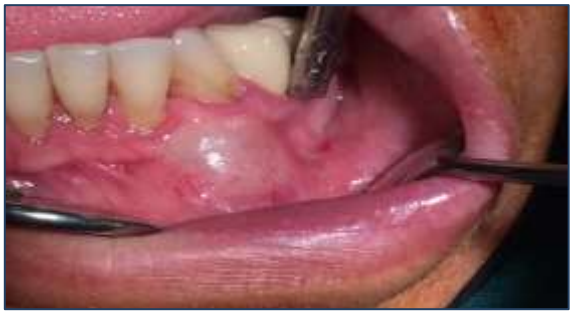

Figure 2-clinical appearance of swelling

On the basis of clinical and radiographic findings, provisional diagnosis such as radicular cyst, residual cyst and lateral periodontal cyst were suggested. Under local anesthesia (Persocaine E, Lidocaine HCl 2% + epinephrine 1/80,000; Daru Pakhsh Pharmaceutical Mfg. Co, Tehran, Iran) surgical enucleating and complete removing of the lesion was done, the surrounding bone scraped with a curette and tissue specimens were sent for histopathological examination. Microscopic evaluation revealed a thin, friable wall with stratified squamous lining epithelium. The surface of the cyst showed parakeratotic epithelial cells, which exhibited a corrugated appearance and flat connective tissue. The basal cell layer was hyperchromatic with palisaded pattern and cuboidal epithelial cells. There were a few inflammatory cells in the thin fibrous wall. Based on the clinical, radiological, and histopathological findings a final diagnosis of odontogenic keratocyst was given. In 3 month follow-up postoperatively the patient was completely asymptomatic.

Figure 3-surgical enucleating of the lesion

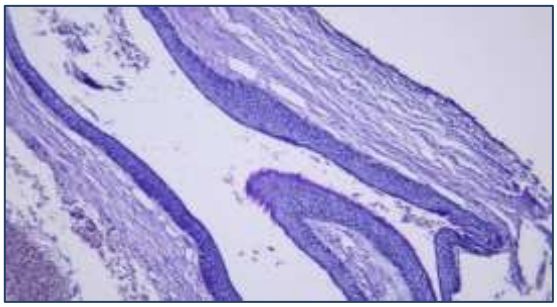

Figure 4-pathological view conclusive of OKC diagnosis

3. Discussion

Philipsen reported the OKC for the first time in 1956 and the lesion was described as a cyst with a thin fibrous capsule and a lining of keratinised stratified squamous epithelium by the World Health Organization (WHO) in 1990. In 2005 it was reclassified as keratocystic odontogenic tumor by WHO because of its aggressive behavior, histopathological characteristics and its high recurrence. Since 2017 it has been considered as a cystic lesion but many aspects of its behavior and etiology is still unclear. This lesion is believed to be arisen from cell rests of dental lamina, which has the ability for keratinization, proliferation, and infiltration of connective tissue during odontogenesis process. OKC has a tendency toward anterior-posterior growth causing extension and little expansion so it usually remains invisible until it is identified during routine dental examinations or expands to an extend which could be detected by the patient. A similar clinical situation was observed in the case we reported. OKC is usually expected to be found at posterior part of the jaws but mostly in the mandible. Based on the previous case reports it has been detected at any part of the jaws or even at the mandibular ramus. This study reported the lesion in the anterior part of the mandible which is not a usual place for occur. Various surgical approaches have been proposed for the treatment of OKC. They include curettage, enucleation, marsupialization, and resection. Considering the surgical difficulties in removing the thin capsular portion of the cyst or presence of the daughter cysts, OKC have a high recurrence rate of 62%.[8- 12] This case was surgically treated by complete enucleating of the cyst and curettage of the surrounding area.

4. Conclusion

OKC represents a unique developmental odontogenic cyst that requires special consideration because of its locally aggressive behaviour and remarkable histopathological features. The case we reported is one of the examples that the clinical appearance and the radiological findings are not distinctive enough to give an accurate and immediate diagnosis of any cyst happening in the jaws.

It should not be diagnosed just on the symptoms, age, sex and the location of the lesion. It is mandatory to have a proper histopathological evaluation and a long-term postoperative follow-up after treatment.

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