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Recurrent Odontogenic Keratocyst: A Rare Clinical Presentation

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***Abstract**: OdontogenicKeratocyst (OKC) aggressive cystic lesion arising from cell rest of dental lamina. It occurs as mutilocular or unilocular radiolucency mainly associated with mandible posterior region. It possess unique histopathological and clinical features, has high recurrence rate and aggressive behavior. Here we discuss a recurrent case of OKC of non- syndromic nature with high infilterating nature and aggressive behavior.*

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I. Introduction

The term 'odontogenickeratocyst' first introduced byPhilipsen (1956). In the earlier literature, the OKC was described as a cholesteatoma (Hauer, 1926; Kostecka, 1929).Later on, Pindborg and Hansen (1963) suggested that the term 'keratocyst' was used to describe any jaw cyst in which keratin was formed to a large extent. Later on due to controversial clinical behavior several other terms were suggested as, 'keratocysticodontogenictumour' (Philipsen, 2005), and 'keratinising cystic odontogenictumour' (Reichart and Philipsen, 2004)[1]The World Health Organization has classified odontogenickeratocysts as dysgnathic cysts in 1971, and in 2005 categorized the parakeratotic type of odontogenickeratocysts as benign tumors, and proposed the termkeratocysticodontogenic tumors (KCOT) and odontogenickeratocysts with orthokeratotic type of keratosis[2].Keratocysticodontogenictumours (KOTs) arebenign cystic epithelial neoplasm that affect the jaw bones and arise from the dental lamina or its remnants and extensions of basalcells from the overlying oral epithelium[1,3].

Among the jaw cysts, the frequency of OKC in different studies ranges from 3.2 to 11.25%[1]. Multiple OKCs usually occur along with nevoid basal cell carcinoma syndrome (NBCCS), also called Gorlin-Goltz syndrome or Ehler-Danlos syndrome, or orofacial digital syndrome[4].

II. Case Report

A 50 years male patient had reported to the department with the complaint of continuous pus discharge from lower right back region of jaw since 8 months. Patient gives history of treatment for KCOT 22 years back. Patient was also carrying a biopsy report done 1 year back from same region with diagnosis of KCOT. On clinical examination, there was mild diffuse right facial swelling over right mandibular molar region. The swelling was non- fluctuant on palpation. An intraoral fistula was observed in right mandibular buccal vestibule in region of 48. It was oval shaped, reddish pink colored and smooth margins. On palpation, it was non- tender, thickened around opening of fistula, it was fixed to underlying structure and pus discharge was present.

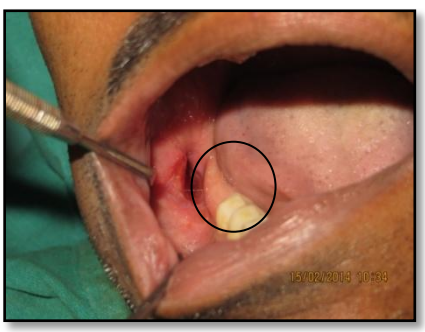


Fig.1 clinical presentation of intraoral fistula.

A panoramic radiograph revealed non uniform thickness of lower border of mandible with irregular localized increase thickness towards lower side suggestive of bony expansion. Loss of continuity of upper

border of mandible at mandibular angle region indicative of perforation of bone. Awell defined solitary localized mixed radiolucency involving ramus, angle of mandible and body of mandible till mesial root surface of 45. Periphery is scalloped, well defined, corticated margins in ramus region, small area of sclerotic margin associated with 45, and areas of blending borders in anterio-superior region of ramus. Internal structure gives fine septal appearance.

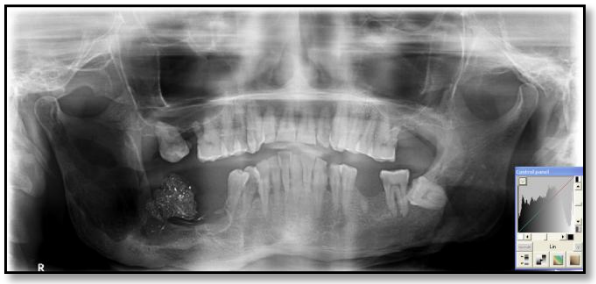
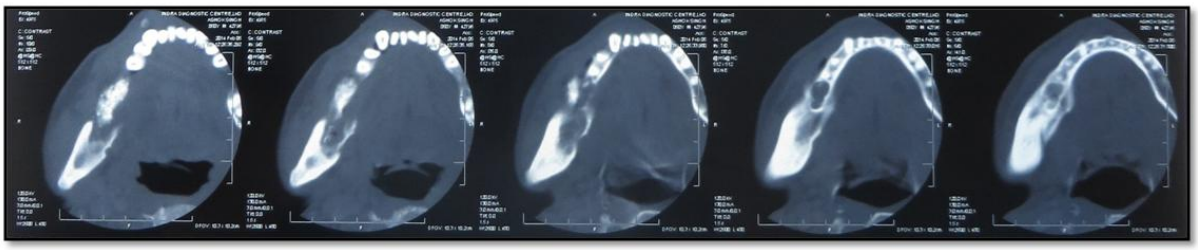


Fig. 2 panoramic radiographs showing a well- defined radiolucency on right mandibular region.

Considering the clinical findings a provisional diagnosis of secondarily infected odontogenic cyst was given. Microscopic examination revealed parakeratinized stratified squamous epithelium of variable thickness with areas of focal proliferation. Connective tissue composed of` inflammatory cells and daughter cell, suggestive of keratinizing cystic odontogenic tumor (secondarily infected).



Tomographic image of right side of mandible showing extend of the lesion.

III. Discussion

Large odontogenic cysts of extraosseous and intraosseousorigin may perforate and produce draining sinuses onto oral mucosa. Cysts which are generally secondarily infected produce this kind of lesions. Generally large dentigerous cyst and keratocysts are the culprits in most of the cases.[5]Keratocysticodontogenictumours (KCOTs) are an independent clinical entity with a typical microscopic picture, clinical growth and biological behaviour.Odontogenickeratocyst (OKC) is a benign but locally aggressive developmental odontogenic cyst [6]. It is derived from the remnants of the dental lamina with a biologic behavior similar to a benign neoplasm[7,8]. They occur most commonly in the mandible, especially in the posterior body and ramus regions[8,9].In mandible, majority of cysts occur in ramus- third molar area, followed by first and second molar and then the anterior mandible. In maxilla, the most common site is third molar area followed by cuspidregion[10]. Age of occurrence ranges from 1st to 9th decade, but peak frequency is been reported in the 2nd and 3rd decade[4]. It shows bimodal age distribution, in non syndromic patients shows first peak at 15- 45 years and second peak at 55- 65 year and in patients with the syndrome, there was a single peak at 10–30[1,6]. It shows higher predilection for males than in females with the ratio 1.28 to 1[11]. Clinical presentation can be of pain, swelling or discharge or it can be completely asymptomatic.In case of infected cyst there is usually pain or swelling of the involved area before sinus formation. Pain ceases when the periosteum and mucosa are perforated, and a purulent discharge ensues. If the sinus is small, the drainage may continue as a chronic case. If the sinus is large, the infection regresses because of the excellent drainage established, and the cyst may disappear completely[5]. Usually these remain dormant until they attain large size and cause considerable destruction. Bony expansion is evident in later stage because OKC tends to extend in the medullary cavity in anterioposterior direction[3,4]. It grows by extension rather than by expansion. The extension here is due to reasons like fingerlike projections from the cyst wall into the marrow spaces, and enlarges slowly but relentlessly along the path of least resistance[6].The particular tendency to rapid growth is due to higher activity of the epithelial cells of the cyst lining stimulating osteolytic activity of prostaglandin substances in the cell population of the cyst lining and higher accumulation of hyperkeratotic scales in the lumen of the cyst with resulting greater difference in hydrostatic pressure[4].

Forssell (1980) suggested that maxillary OKC are more likely to get infected even smaller in size than compared to mandibular cyst and hence can be easily diagnosed at early stage[1].Whereas in our case, it was long

standing mandibular cyst which was infected.It has been well documented that the OKC has aparticular tendency to recur after surgical treatment[4]. The possible cause of recurrence has been suggested as; firstly presence of satellite cyst[4] that is usually retained even after enucleation, secondly cystic lining of OKC are very thin and fragile that act as huddle for removal of cyst in single section. Thirdly scalloped margins of cyst which is the characteristic feature results in increased incidence of remnant of cystic lining after enucleation, and can results in recurrence[1,8]. Voorsmit*et al*. (1981) believed that a recurrent OKC may develop in three different ways: by incomplete removal of the original cyst lining; by the retention of daughter cysts, from microcysts or epithelial islands in the wall of the original cyst; or by the development of new OKCs from epithelial off-shoots of the basal layer of the oral epithelium[1].

Aggressive nature of OKC results in penetration of cortical bone and involvement of surrounding soft tissues (Emerson et al., 1972; partridge and towers, 1987) also OKC may develop as a result of hamartomatous growth from basal cell of oral mucosa particularly in third molar and ascending ramus region, so it is been recommended that while surgical removal of cyst, the overlying mucosa should be surgically excised to prevent possible recurrence[1, 10].

The histopathological picture of OKCs/KCOTs is a characteristic thin epithelial layer, composed of from 8 to 10 cell layers.Uniform cyst lining, hyperchromatic and palisaded basal cells, wavy parakeratin production and a flat interface between the epithelium and connective tissue wall[12]. In the direction of the cyst lumen there is parakeratosis with a focal zone created of orthokeratins. These classic microscopic features are often completely lost when the cyst is inflamed[13]. Sometimes there is invasion of the basal cell layer into the region of surrounding connective tissue and the formation of satellite micro-cysts. The fibrous walls of the cells may be relative thin and usually without inflammatory cell infiltrates. Other variant describedas orthokeratinisedtype with a prominent granular layer lying immediatelyunder the thin surface layer.Clinically, the lesion with parakeratotic type of keratosis is characterized by aggressive growth and the increased tendency to recur following surgical procedures[2] same parakeratotic nature with aggressive nature was present in our case.Recently new variants described are peripheral OKC[1,4,13]& solid variant of OKC[4].

Radiographically, OKCs present as welldefined radiolucent lesions which maybe unilocular or multilocular, with smooth cortical or, more often, scalloped margins.Tsukamoto et al in 2002 reported that scalloping is most of the cases associated with mandibular 3rd molar compared to KCOT not associated with teeth[10]. In mandible, epicenter lies superior to inferior alveolar nerve canal[10]. Root resorption and paraesthesia are common phenomena in these relatively aggressive cystic lesions[4] likewise as seen in our case with root resorption associated with 45.

In our case which was of non syndromic type shows recurrence of lesion during 2nd peak of life that is in 50's, long standing lesion has given a different clinical presentation than cyst with completely asymptomatic intraoral fistula.

IV. Conclusion

Keratocyst of recurred variety can be quiet aggressive with more extensive involvement and a very diverse clinical presentation as in our case.

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