

ORTHOKERATINIZED ODONTOGENIC CYST- A RARE CASE

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INTRODUCTION

Odontogenic Keratocyst (OKC) and Orthokeratinized Odontogenic Cyst (OOC) have variable clinical biological and histopathological behaviour. Philipsen in 1956 separated seven jaw cysts as a distinct entity and termed as odontogenic keratocyst. Out of these, 2 showed an orthokeratinized pattern¹.

OKC is known for its potentially aggressive biologic behaviour, its significant recurrent rate (42.6%) and occasional occurrence as a component of basal cell nevus-bifid rib syndrome. Typical OKC is histologically characterized by a uniform epithelial lining with a well developed basal cell layer and luminal surface of corrugated parakeratin².

OOC usually presents as a solitary cyst in a dentigerous cyst relationship in posterior mandible. It exhibits male predilection from 2nd to 5th decade and typically less aggressive and low recurrence rate (2.2%) than the conventional type of OKC. Histopathologically, OOC has a thin epithelial lining with a luminal surface of orthokeratin, basal cell layer containing flattened squamous or cuboidal cells, and well developed granular cell³.

These findings suggest the importance of distinguishing between OKC and OOC. Following is a case report of an OOC.

CASE REPORT

A 20 year old female visited the Manipal College of Dental Sciences, Mangalore with a chief

complaint of pain on the right side of jaw since 1 day. The pain was dull aching and continuous in nature. There were no aggravating and relieving factors, no difficulty in opening the mouth. There was no history of any trauma. No relevant medical history was reported. This was her first visit to a dentist. Clinical examination revealed a healthy well nourished female with an extra oral solitary swelling on the right body of the mandible, oval in shape, approx. 3×2 cms in size, with normal overlying skin. On palpation, the swelling was tender, no rise in temperature. It was hard in consistency and was immobile.

TMJ examination revealed bilateral smooth well coordinated symmetrical movements. On lymph node examination, right submandibular group of lymph nodes were palpable which was firm, fixed and tender.

On intraoral examination of hard tissues, 37 was carious and crowding on the mandibular anteriors and a retained deciduous 83 which had grade II mobility. There was a diffuse swelling in relation to 46, 47 region. The overlying mucosa and gingiva were pale. On palpation, the swelling was approx 3×2 cms in size and roughly oval in shape, firm in consistency with pitting and cracking was felt suggestive of perforation of buccal cortical plate. Vitality test was positive for all these teeth (heat test).

Radiographic examination included OPG which revealed well defined multilocular radiolucency between 46 and 47 with cortical bone expansion

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extending upto the inferior border of the mandible leading to thinning of the bone. There was distal displacement of the roots of 47 without the root resorption. Lamina Dura of all the roots were intact.

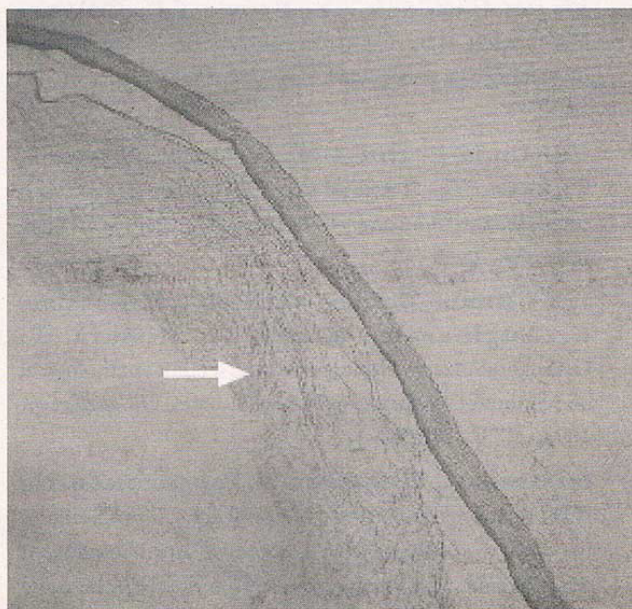


Orthopantomograph showing multilocular radiolucency extending upto the inferior border of the mandible.

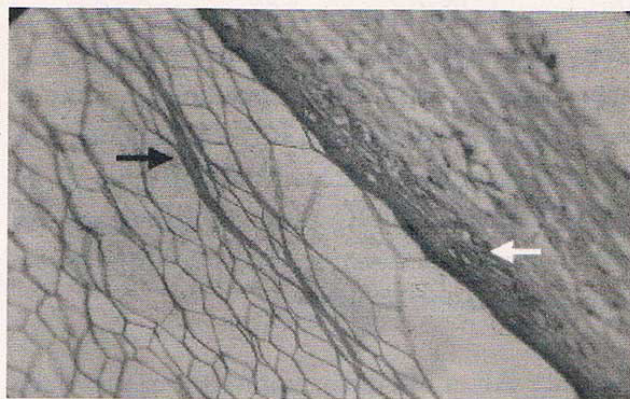
Fine needle aspiration yielded a straw coloured fluid which was sent for cytological examination. Cytology report revealed RBCs, a very sparse mixed inflammatory cells mainly lymphocytes and eosinophils. Few superficial epithelial squames were also present.

Hence based on all these findings provisional diagnosis of cystic ameloblastoma was made.

Differential diagnosis of OKC, Glandular odontogenic cyst, lateral periodontal cyst was given.



Low power photomicrograph of orthokeratinization (white arrow). Epithelial lining is thin and irregular without rete ridge proliferation (H&E: original magnification X10)



High power photomicrograph of a orthokeratinized lining epithelium showing thick granular cell layers (white arrow) present beneath the orthokeratin (black arrow). Basal cell layer does not have typical histologic features of OKC (H&E: original magnification X40)

Incisional biopsy was done in the Department of OMFS and was subjected to histopathological examination.

On histopathological examination, the epithelium was 6-8 layers thick with an orthokeratotic surface. Keratohyaline granules were prominent in the superficial epithelial layer adjacent to the orthokeratin. Basal cell layer consisted of flattened cuboidal cells, with little tendency to palisade and flattened interface between the epithelium and the connective tissue. Connective tissue wall consisted of dense bundle of collagen fibres and mild chronic inflammatory cell infiltrate was present.

Due to the thinning of the inferior border of the mandible, it was then resected along with the lesion.

DISCUSSION AND REVIEW OF LITERATURE

OOC occurs predominantly in young adult male with an M:F ratio of 2:1. Lesion is twice as frequently in the mandible than the maxilla with a tendency to involve the posterior areas of the jaw. About two third of OOC are encountered in a lesion that appears clinically and radiographically to represent a dentigerous cyst. Sometimes it may be multilocular which may mimic an ameloblastoma. Owing to the rare occurrence of OOC, detailed studies are scarce and only brief notes on differences from OKC are available in the literature^{4,9}.

Wright et al (1981) reported 60 cases of OOC and compared with OKC and found that the OOC appears to be a distinct clinicopathologic entity. It was suggested that this cyst be called odontogenic keratocyst, orthokeratinized variant in order to avoid confusion. OOC shows a limited growth potential, appears to have a lower recurrence rate than parakeratinized variant¹.

Hancock AF et al (1986) reported a case of long standing radiolucency caused by OOC which was interpreted and treated as an endodontic lesion. He emphasized on the importance of radiographic recall as a part of endodontic therapy and the necessity of histological examination for patients with long standing lesions not subsiding even after endodontic treatment⁵.

Vahahula E et al (1993) termed OOC under the odontogenic jaw cyst and found most cases were seen in slightly younger patients than those of OKC. 75% of patients were below 27 years and peak frequency was in 3rd decade. He also noted slight male predilection and found the cyst to be less aggressive than OKC. It was concluded that OOC are heterogeneous group showing diversity in presentation and majority represent dentigerous cyst with orthokeratosis or may be central dermoid/epidermoid cyst. He strongly suggested that the orthokeratinized OKC are another possible type of odontogenic jaw cyst⁶.

Li TJ et al (1998) reported 15 cases of OOC and compared histological and immunocytochemical features with that of OKC and suggested that OOC is clinicopathologically separate from other types of odontogenic cysts and may thus constitute a distinct clinical entity⁷.

Iamaroon et al (2004) compared the proliferation index of the epithelial cells between OKC, OOC, dentigerous cyst and ameloblastoma. He concluded that OKC should be regarded as a benign tumour rather than simply an odontogenic cyst and OOC as a non aggressive cystic lesion⁸.

CONCLUSION

It is very important to distinguish between the OKC and OCC clinically, histopathologically and by immunohistochemical technique. Based on the previous studies, the findings suggest that the OCC is clinicopathologically and biologically different from OKC and thus should be regarded as a distinct clinical entity. If proper clinical, radiographic, and histopathological examinations are done OOC can be distinguished from OKC and unnecessary extensive surgical intervention can be avoided.

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