Keratocystic Odontogenic Tumor Involving Coronoid Process and Condyle

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Abstract

Keratocystic odontogenic tumor (KCOT) is a peculiar entity affecting the jaw bones. It is a benign intraosseous neoplasm of jaws that shows a very high recurrence rate. It is locally aggressive and is lined by keratinized stratified squamous epithelium. The most important feature that separates KCOTs and other odontogenic cysts and tumors is the presence of microcysts also called as daughter cysts. Daughter cysts are the main source of recurrence, and they complicate the treatment plan to a great extent. KCOT is commonly seen in the posterior mandible and ramus area and rarely the condyle. In this paper, we present the case of a 25 year old female patient with KCOT involving the condyle and coronoid process along with relevant review of literature. This patient was subjected to marsupialisation, and is followed up once in 3 months.

Key Words: Keratocystic odontogenic tumor, odontogenic keratocyst, multilocular osteolytic lesion, mandibular ramus

Introduction

Originally, KCOT was referred as Odontogenic keratocyst (OKC). It was first described by Hans Phillipsen in 1956 and its characteristic features were elaborated by Pindborg and Hansen in 1963 [1,2]. The term OKC was changed to ‘Keratocystic Odontogenic tumor’ by the World health organisation (WHO) in 2005. This change in nomenclature was mainly based on its potential for high recurrence, genetics and its neoplastic behaviour [3]. KCOT is a benign uni or multicystic neoplasm with characteristic features of parakeratinised lining with stratified squamous epithelium. This tumor which was once considered as an odontogenic cyst arises from the remnant cell rests of the dental lamina. In this article, a thorough literature review along with a case discussion is presented.

Case Report

A 25 year old female patient reported to the department of Oral medicine with the complaint of pain in the lower back tooth region since 3 months and a swelling on the same region (left lower 1/3rd of face) since 1 month. The patient was asymptomatic 3 months back, but later she developed pain which was intermittent, dull and localized. Pain aggravated on chewing food and relieved on taking antibiotics. According to the patient, the swelling was initially small but slowly increased and reached the present size. Her past dental, medical and family history was insignificant. On physical examination, she was healthy and had no constitutional symptoms.

Extra oral examination revealed facial asymmetry due to a diffuse swelling in the lower 1/3rd of the face (Figure 1). The swelling was approximately 8 cm x 7 cm in its greatest dimension, oval in shape, extending anteriorly from ala of nose upto the tragus of ear; superior inferiorly it extended from the infra-orbital margin to a point 1 cm below the lower border of the mandible. The skin over the swelling appeared normal. The swelling had ill-defined margins and was tender with mild local rise of temperature. Lymph node examination revealed a palpable, movable and tender left submandibular lymph node.

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Figure 1. Extraoral photograph showing asymmetry due to a swelling on the left side of face.

Figure 2. Intraoral picture showing partially erupted 38.

Intraoral examination revealed a partially erupted 38 associated with pericoronitis (Figure 2). Surprisingly, there was no obliteration of the buccal vestibule in the region of the swelling. Based on history and clinical findings a provisional diagnosis of pericoronal abscess in relation to 38 was thought.
However, the origin of facial swelling without vestibular obliteration couldn’t be explained. The presence of a partially erupted 38 suggested other pathology such as dentigerous cyst, but to investigate these radiographs were necessary. Intra oral periapical radiograph of lower left posterior teeth showed 36, 37, 38 associated with a well-defined multilocular radiolucent lesion extending from mesial root of 37 upto ramus of mandible. The lesion was surrounded by a sclerotic border.

Root resorption was noticed in relation to the distal root of 38 with widening of PDL space and loss of lamina dura. Mandibular occlusal view shows an small radiolucency on the buccal aspect of 37 and 38. There is no expansion of buccal and lingual cortical plates (Figure 3).

An orthopantomogram (OPG) demonstrated a well-defined multilocular radiolucent lesion of size 5 cm x 4 cm involving mandibular posterior body, ramus, coronoid and condylar process with scalloped margins. The lesion extends from the mesial root of 37 upto the condyle and coronoid process and is surrounded by a thin radiopaque margin. There is a pathological fracture seen at the lower border of mandible (Figure 4). Computed Tomography (CT) revealed a well-defined large osteolytic lesion measuring approximately 8 cm x 7 cm (Figure 5). CT 3D Reconstructive images show a multilocular osteolytic lesion from 37 with destruction of ramus, condyle and coronoid process (Figure 6). Based on radiographic impression a differential diagnosis of KCOT, Ameloblastoma and its variants were considered.

On aspiration blood tinged cheesy material was obtained. Incisional biopsy was also performed and histopathological features suggested a cystic lesion lined with parakeratinized stratified squamous epithelium. It had wavy inner margins and there was basal nuclear palisading. It is filled with lamellar anucleated keratinous material and daughter cysts located near the connective tissue adjacent to the lining epithelium (Figure 7).

Based on histology, a final diagnosis of ‘Keratocystic Odontogenic Tumor’ was considered. Due to the wide extensions of the lesion, relation with anatomical structures and age, marsupialisation was chosen as the line of treatment. Extraction of 37 and 38 with marsupialisation was followed by regular open dressing. A follow up after 3 months revealed no evidence of recurrence (Figure 8).
Figure 6. CT 3D Reconstructed images show a multilocular lesion.

Figure 7. Palisaded basal cell layer with a stratified squamous epithelium.

Figure 8. Postoperative panoramic radiograph taken 3 months after surgery.

Discussion

The most significant features of KCOT include its aggressiveness, infiltrative behaviour, and high recurrence rate. Based on its character, as mentioned earlier, OKC was re-designated by WHO as KCOT [3]. KCOTs account to 11% of all jaw cysts [4]. It is clinically benign and is locally destructive. They can occur at single or multiple sites. The
KCOTs are normally asymptomatic and are incidental findings on dental radiographs. In the present case there was complete loss of condylar architecture on the left side along with root resorption. Although KCOTs exhibit a locally aggressive behaviour and spread via narrow spaces, the involvement of condyle and coronoid process is rarely reported. This may be due the high bone density and deficient narrow spaces in these regions. Because of its variable clinical picture and radiological features they are often misdiagnosed and diagnosed late leading to a more aggressive treatment plan. A careful rehabilitation is hence necessary and minimizing morbidity is the primary objective in the management of KCOT. Long term and close follow up is mandatory to identify recurrence. Newer approaches like targeted therapy can be tried in the future to control the associated high recurrence.

References


