

A Unique Case of Keratocystic Odontogenic Tumor Mimicking Malignancy

Mylanahalli Doddarangiah Prakash, Borlingegowda Viswanatha*, Glen Edwin D'Souza

Otorhinolaryngology department, Bangalore medical college & research institute, Bangalore, India

Abstract Philipsen in 1956 [1] was the first person to describe odontogenic keratocyst (OKC). It is now designated by the World Health Organization (WHO) as a keratocystic odontogenic tumor (KCOT) and is defined as “a benign uni or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior” [2]. WHO recommends the term keratocystic odontogenic tumour as it better reflects its neoplastic nature [2]. They have an annual incidence of 3.5-5 per million population [3]. They usually but not always present as large multilocular cysts, which drives the patient to the hospital. Here we present a case of keratocystic odontogenic tumor in a 28 years old male patient who presented to us with a large hard painful swelling over the right cheek, with clinical features suggestive of malignancy of the maxillary sinus.

Keywords Keratocysticodontogenic Tumor (KOCT)

1. Introduction

Odontogenic cysts develop from redundant epithelium left behind in the jaws as a result of tooth development [4], but keratocystic odontogenic tumors are lined by parakeratinized and stratified epithelial lining whose basal layer has a neat arrangement of palisaded columnar and cuboidal cells, on top of which are several layers of squamous epithelium. This lining has a high mitotic rate and rarely can become dysplastic and develop into squamous cell carcinoma [5]. It isn't unheard of for patients to wait till the pain supersedes the physical unsightliness in our part of the country for seeking medical attention. Here we present a case of a large keratocystic odontogenic tumor which occupied the whole of the right maxillary sinus mimicking malignancy of the area and brought the patient to us when he started experiencing pain in the area.

2. Case Report

A 28 year old male presented with a history of swelling in the right cheek since 6 months. Initially it was 0.5X0.5 cms in size and it gradually increased to the present size over a period of 6 months. Patient came to the hospital after pain started in his right cheek. It was associated with unilateral headache and difficulty in chewing on the right side. There

was no history of previous irradiation or surgery. No significant dental health issues were reported by the patient.



Figure 1. Showing the patient with gross swelling at the right cheek

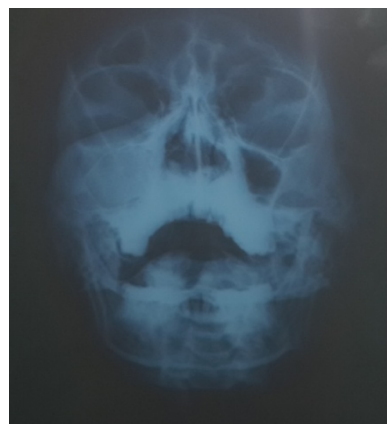


Figure 2. Radiograph of paranasal sinuses [Water's view] showing opacity in the right maxillary sinus

* Corresponding author:

drbviswanatha@yahoo.co.in (Borlingegowda Viswanatha)

Published online at <http://journal.sapub.org/otolaryn>

Copyright © 2014 Scientific & Academic Publishing. All Rights Reserved

On clinical examination a fixed hard oval mass of about 6X6 cms was present over the maxillary sinus region [Figure 1]. The skin over the mass was regular and it had a smooth surface. The mass was tender on deep palpation. The nasal examination and endoscopy was within normal limits. Oral cavity and oropharynx examination was within normal limits. There was no cervical lymphadenopathy.

X-ray paranasal sinus (Water's view) showed a well defined heterogenous predominantly hypersclerotic lesion with solid component, multiple air foci in the maxillary sinus [Figure 2].



Figure 3. CT scan image showing mass in the right maxillary sinus and erosion of the lateral wall

CT scan of nose and paranasal sinuses showed a mass in the right maxillary sinus which was causing thinning of the anterior, posterior and lateral wall of right maxillary sinus extending antero-inferiorly and laterally causing mass effects and break in the cortex of anterolateral wall of maxillary sinus and the zygomatic process. Posterolateral wall of maxillary sinus was breached and anterior border of temporalis muscle had been abutted. Superomedially it was limited by medial wall of right maxillary sinus. Inferomedially it extended up to perpendicular plate of palate eroding the hard palate on right side and right maxillary alveolar process of central, lateral incisors and canine and their roots. Calcifications and septations were present in the lesion [Figure 3 & 4]. Right maxillary osteomeatal complex was compromised. Radiological findings were suggestive of keratocystic odontogenic tumor was made.

The patient was informed about the diagnosis and given possible management options. After receiving the patients consent for surgery, and finding him to be surgically fit, he was posted for surgery under general anesthesia. Once anaesthetized, under Caldwell-Luc's approach, the mass was removed in piece meal fashion. Parts of the surgery required meticulous drilling [Figure 4].



Figure 4. Intraoperative photograph showing right maxillary sinus after removal of the mass

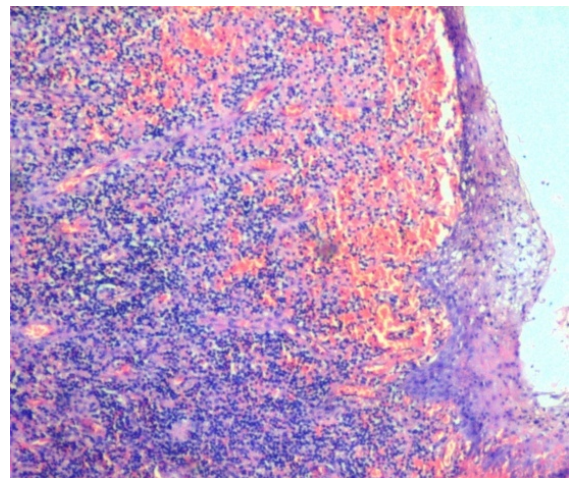


Figure 5. Photomicrograph showing epithelial lining with parakeratinised cells, keratin debris and satellite cysts

Histopathological examination confirmed that the diagnosis of keratocystic odontogenic tumour [Figure 5]. Patient was followed up 6 months after the operation and is asymptomatic.

3. Discussion

Keratocystic odontogenic tumors are low grade neoplasm supported by abnormal tumor suppressor genes [6]. Keratocystic odontogenic tumors have a peak incidence between the second and fourth decade of life. Usually they are asymptomatic and are found incidentally. They can be associated with erupted, unerupted or non tooth bearing areas of the jaw. They are commonly found in the mandible and the maxilla. They sometimes masquerade as large dentigerous cysts. Rarely do they reabsorb tooth roots and this makes them resemble neoplasm. Malignant transformation into squamous cell carcinoma is rarely reported at 0.12% [5, 6, 8].

Treatment options include marsupilisation, enucleation with or without usage of Carnoy's solution which is 1 g of ferric chloride (FeCl₃) dissolved in 24 mL of absolute

alcohol, 12 ml of chloroform, 4 ml of glacial acetic acid. These methods show a recurrence rate between 17% to 56%. Ghali and Connor report 0% recurrence after complete removal after radical treatment, but this may result in damage to inferior orbicular nerve, inferior alveolar nerve or tooth root. In addition reconstruction surgery may be required [9, 10].

4. Conclusions

Keratocystic odontogenic tumors are low grade neoplasm which sometimes mimic malignancy. They may require surgical management when symptomatic. Recurrence rates being high are a matter of concern for the patient and the surgeon and complete removal is essential to prevent recurrence.

REFERENCES

- [1] Philipsen HP. Om keratocystedr (Kolesteratomer) and kaeberne. *Tandlaegebladet* 1956; 60:963-71.
- [2] Barnes L, Eveson JW, Reichart P, Sidransky D, editors. Pathology and genetics of head and neck tumours. Lyon: IARC Press; 2005. WHO classification of tumours series.
- [3] Rachanis CC, Shear M. Age standardized incidence rates of primordial cyst (keratocyst) on the Witwatersrand. *Community dentistry and oral epidemiology* 1978;6:296-29.
- [4] Cantwel CP, Obyrne J, Eustace S. Current trends in treatment of osteoid osteoma with an emphasis on radio frequency ablation. *European radiology* 2004;14:607-617.
- [5] Gurk MM, Cassoni A, Pitkin L, Scott Brown textbook of Otolaryngology and Head and Neck Surgery, 7th Edition Volume 2, Chapter 148, pp1924-1925.
- [6] Shear M. The aggressive nature of the odontogenic keratocyst: is it a benign cystic neoplasm? Part 2. Proliferation and genetic studies. *Oral Oncology* 2002; (38) 4; 323-331.
- [7] Barreto DC, Küstner EC. New considerations about the diagnosis of odontogenic keratocyst. *Medicina Oral* 2001; 6(5); 350-357.
- [8] Agaram NP, Collins BM, Barnes L. Molecular analysis to demonstrate that odontogenic keratocysts are neoplastic. *Archives of Pathology and Laboratory Medicine* 2004;128(3); 313-317.
- [9] Timosca GC, Cotutiu C, Gavrilita L. Malignant transformation of odontogenic cysts. *Revue de Stomatologie et de Chirurgie Maxillo-Faciale* 1995;(96) 2;88-95.
- [10] Maurette PE, Jorge J, Moraes MD. Conservative treatment protocol of odontogenic keratocyst: a preliminary study. *Journal of Oral and Maxillofacial Surgery* 2006; 64(3);379-383.