Case Report

An unusual case of recurrent jaw swelling

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Abstract

Calcifying odontogenic cyst was first reported in 1962 by Gorlin. At that time it was classified as cyst related to odontogenic apparatus, but it was renamed as calcifying cystic odontogenic tumour(CCOT) by WHO in 2005 due to histological complexity, morphological diversity and aggressive proliferation. We report a case of Benign Odontogenic Cyst, presenting as a recurrent painless jaw swelling over Right angle of mandible, in a 70year old Bengali male, for which he was operated twice, with recurrence after each time. Right Hemimandibulectomy was done and specimen was sent for Histopathological examination. Histopathological sections showed an unilocular benign odontogenic cyst lined by a thin layer of basal squamous epithelium with cholesterol and haemosiderin deposit, with a possibility of radicular or paradental cyst.

Keywords: Benign odontogenic cyst, Odontogenic keratocyst, Calcifying odontogenic cyst.

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INTRODUCTION

The jaws are host to a wide variety of cysts and neoplasms, due in large part to the tissues involved in tooth formation. Odontogenic cysts that can be problematic because of recurrence and/or aggressive growth include odontogenic keratocyst (OKC), calcifying odontogenic cyst, and the recently described glandular odontogenic cyst. Traditional histopathology continues to be the mainstay for the diagnosis of these lesions as immunohistochemistry and molecular techniques have had, as yet, little impact in this area. Li and YU classified these lesions into three groups cyst, benign and malignant tumours. In more recent edition a new classification is suggested by Praetorius[6].

Table 1

A. Group 1 'Simple' cysts
1. Calcifying odontogenic cyst (COC)
B. Group 2 Cysts associated with odonotogenic hamartomas or benign neoplasms: calcifying cystic odontogenic tumours (CCOT).
The following combinations have been published:
1. CCOT associated with an odontome
2. CCOT associated with adenomatioid odontogenic tumor
3.CCOT associated with ameloblastoma
4. CCOT associated with ameloblastic fibrom a
5. CCOT associated with ameloblastic fibro-odontoma
6. CCOT associated with odonto-ameloblastoma
7. CCOT associated with odontogenic myxofibroma
C. Group 3 Solid benign odontogenic neoplasms with similar cell morphology to that in the COC, and with dentinoid formation
1. Dentionegic ghost cell tumor
D. Group 4. Malingnent odontogenic neoplasms with feature similar to those of the dentinogenic ghost cell tumour
1. Ghost cell odontogenic carcinoma

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Table 2: Current Classification of Jaw Cysts

Odontogenic	Non-odontogenic
Inflammatory	Nasopalatine cyst
Periapical cyst	
Developmental	Pseudocyst
Dentigerous cyst	Traumatic bony cyst
Lateral periodontal cyst	Static bony cyst
Odontogenic keratocyst	Haematopoetic bone marrow cyst
Calcifying odontogenic cyst	
Glandular odontogenic cyst	

CASE REPORT

A 70year old male patient presented to the surgical ward of NRS Medical College and Hospital with a recurrent painless jaw swelling over Right angle of mandible in December 2011. He was operated on the swelling twice (in 1976 and 1985), with recurrence after each time. The subject was a known hypertensive taking Amlodipine, Losartan and Hydrochlorthiazide. Local examination showed a tense, cystic, non-tender swelling of approximately 4cm x 3cm size, over right angle of mandible, fixed to underlying bone. Scar of previous surgery was visible over right angle of mandible. There was no cervical lymphadenopathy. Pre-operative FNAC from the current cystic lesion showed anucleated squamous inflammatory cells and few epithelial cells with nuclear atypia. His OPG showed Bony cyst on Right lower mandibular region. He underwent a MRI scan of neck which showed a well defined expansile predominantly cystic SOL of Right Mandibular Ramus at its postero-lateral aspect compressing the right parotid gland (both deep and superficial part) and extending upto the level of right submandibular gland, containing solid component at the supero-medial aspect of the lesion. Marrow hyperintensity was noted within the expanded mandible. On exploration on 15th of December, 2011, a cystic swelling was found to arise from posterior aspect of Right hemimandible extending up to 2.5cm lateral to symphysis menti, with spillage of cheesy material from the cystic cavity. Right Hemimandibulectomy was done and specimen was sent to pathologist for Histopathological examination. Mucosa and muscles were closed in layers using 3-0 Vicryl and skin closed with staplers. Patient was kept on Naso-gastric feeding for 5days post-operative. Post-operative recovery was uneventful. On follow-up at one month, patient had minimal cosmetic deformity and functional loss. Histopathological sections showed an unilocular benign odontogenic cyst lined by a thin layer of basal squamous epithelium which was denuded in places and associated with a prominent histiocyte rich inflammatory reaction. Prominent cholesterol and haemosiderin deposit was seen.

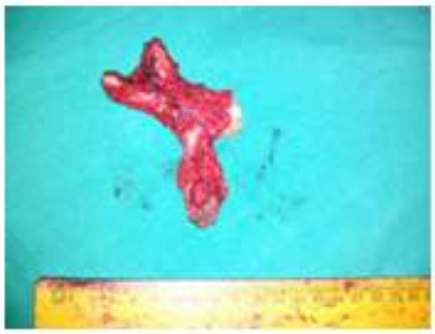

Figure 1: Specimen

Figure 2: Intraoperative picture

DISCUSSION

Many benign jaw tumors and several cysts, of both odontogenic and nonodontogenic origin, can exhibit a biologically aggressive course and can be diagnostically difficult. Odontogenic cysts of diagnostic significance are Odontogenic Keratocyst (OKC), Calcifying Odontogenic cyst (COC) and Glandular Odontogenic cyst[1]. The odontogenic keratocyst (OKC) is a commonly encountered developmental cyst of considerable potential for aggressive clinical behavior and recurrence[2] Also, it may be a component of the nevoid–basal cell carcinoma (Gorlin) syndrome. OKC occurs anywhere in the jaws and in any position. It may be superimposed over the apices of tooth roots or adjacent to the crowns of impacted teeth. Radiographically, it appears as a well-defined lucency and is often multilocular. OKCs represent 5–15% of all odontogenic cysts. The recurrence rate for solitary OKC is 10–30%. Approximately 5% of

OKC patients have multiple jaw cysts (and no syndrome), and their recurrence rate is greater than that for solitary lesions. Microscopically, the epithelial lining exhibits a characteristic thickness of 6–10 cell layers. The epithelium shows basal palisading and a thin refractile parakeratinized lining layer. Separation of the epithelium from the thin and uninflamed supporting fibrous wall is often seen. Budding of the basal layer and "daughter cyst" formation are frequent findings. If the cyst wall becomes secondarily inflamed, hyperplasia ensues and the characteristic microscopic pattern disappears. The epithelial proliferation rate in the OKC is relatively high, especially in the case of those that are syndrome associated[3]. The nevoid–basal cell carcinoma syndrome is inherited as an autosomal dominant trait that consists principally of multiple odontogenic keratocysts, multiple basal cell carcinomas, skeletal anomalies, and cranial calcifications. Syndrome associated OKCs have the highest recurrence rate and represent approximately 5% of all OKC patients[1,4]. Calcifying odontogenic cyst (COC) is a developmental cyst that may exhibit occasional aggressive/recurrent behavior. This is particularly true of an occasionally encountered solid variant that is regarded as a neoplasm and termed odontogenic ghost cell tumor. A very rare malignant variety of odontogenic ghost cell tumor has been reported as odontogenic ghost cell carcinoma. COC shows a predilection for females and the maxilla. It occasionally is seen in the gingiva. It may be unilocular or multilocular and may show areas of opacification because of the partial calcification of keratinized lining cells. The distinctive microscopic feature of this lesion, be it cystic or solid, is "ghost cell" keratinization of the epithelial lining. The keratin may undergo dystrophic calcification and may incite a foreign-body reaction in the cyst wall, giving it features similar to the pilomatrixoma of skin. Ghost cells alone are not diagnostic, as they may occasionally be seen in other odontogenic tumors, such as ameloblastomas and odontomas. Glandular Odontogenic Cyst (Sialo-Odontogenic Cyst) is a rare and recently described developmental jaw cyst that may superficially mimic a central muco-epidermoid carcinoma. It is seen in adults in any jaw site, although anterior regions are favored. This multilocular cyst is lined by nonkeratinized epithelium with focal thickenings composed of mucous cells in a pseudoglandular pattern. This lesion has shown local aggressiveness and has recurrence potential[5]. Dentigerous cysts that exhibit occasional mucous goblet cells in their linings are not believed to be related to the glandular odontogenic cyst. The glandular odontogenic cyst should not be confused with the more exuberant intraluminal lobular proliferation of adenomatoid odontogenic tumor. Radiographically, they are clearly-delineated and appear as unilocular or multi- locular radiolucencies, with calcifications of variable density noted in one-third to one-half of cases[8]. Enucleation is the treatment of choice for most in- traosseous CCOTs and few recurrences have been re- ported in the literature[7].

CONCLUSION

With the histopathological picture, it was difficult to subtype the lesion in our case and there was a possibility of the lesion being a radicular or a paradental cyst. Nevertheless, the pathologists' impression was of the lesion being a benign odontogenic cyst of the mandible. CCOT is an uncommon entity. Although rare, calcify- ing epithelial odontogenic tumor and adenomatoid odon- togenic tumor should be included in the differential diagnosis of jaw lesions. The chief danger is the losing of permanent teeth[9].

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