ISSN: 0975-833X

Available online at http://www. journalcra. com

International Journal of Current Research
Vol. 9, Issue, 11, pp.60457-60460, November, 2017

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

CASE REPORT

SIMPLE CYSTIC VARIANT OF CALCIFYING CYSTIC ODONTOGENIC TUMOR (CCOT) ASSOCIATED WITH AN IMPACTED CANINE: A RARE PATHOLOGICAL ENTITY

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ARTICLE INFO

Article History:
Received 11th August, 2017
Received in revised form
06th September, 2017
Accepted 21st October, 2017
Published online 30th November, 2017

Key words:
Calcifying Cystic Odontogenic Tumor, (CCOT), Calcifying Odontogenic Cyst(COC), Odontome; Multilocular, Ghost cells.

ABSTRACT

Calcifying Cystic Odontogenic Tumor (CCOT) was first described by Gorlin(1962) as Calcifying Odontogenic Cyst(COC)and accounts for only 1% of jaw cysts reported. Because of its histopathological diversity, there has always been controversy about its nature as a cyst or neoplasm. WHO (2005) has categorized this lesion from cystic - COC to neoplastic - CCOT. It has been found to be associated with various pathologies such as odontoma, ameloblastoma, ameloblastic fibroma. This paper presents clinicopathological, radiographic & histopathological features of a case of CCOT highlighting its histopathological subtypes.

Citation: Anchal Srivastava, Bhavthankar J.D., Abdul Ahad Khan, Deepak Passi, Vikram Ahuja and Lokesh Chandra. 2017. "Simple cystic variant of Calcifying Cystic Odontogenic Tumor (CCOT) asssociated with an impacted canine: A rare pathological entity", *International Journal of Current Research*, 9, (10), xxxxxxxxxx.

INTRODUCTION

Calcifying cystic odontogenic tumor is a rare developmental pathological entity which is derived from the odontogenic epithelium. It was first described by Gorlin *et al*, in 1962 as Calcifying odontogenic cyst or Gorlin cyst (Gorlin, 1962). World Health Organization (WHO) in 2005, classified this lesion as odontogenic tumor and named calcifying cystic odontogenic tumor (CCOT), due to the neoplastic nature of the cystic lesion (Praetorius, 2005). Meanwhile, the solid pattern of COC was designated as dentinogenic ghost cell tumor (DGCT) (Praetorius, 2005). CCOT is usually associated with odontogenic tumors like odontome, ameloblastoma, calcifying epithelial odontogenic tumor, odontogenic keratocyst, adenomatoid odontogenic tumor (Hirshberg, 1994). CCOT is a rare benign odontogenic tumor, accounting for 1.6% of all central odontogenic tumors. The lesion has a wide age distribution, varying from 5 – 92 years but most cases are diagnosed in the second and the third decades of life without gender predilection (Neville *et al*., 2002). Clinically, CCOT usually presents as a painless slow-growing swelling commonly present in anterior region of the jaws.

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Maxilla and the mandible are equally affected. Radiographically, presents as well defined unilocular or multilocular radiolucency containing different amounts and shapes of radiopacity. About one-third of cases, CCOT is associated with an impacted tooth, specially canine (Rajendran, 2012). A case is presented here in which the lesion occurred in mandible associated with an impacted permanent canine.

Case report

A 18-year-old female presented to the department of Oral pathology and microbiology with the chief complaint of swelling in the right lower jaw since 6 months. The swelling started gradually and increased to attain the present size. Initially it was painless and gradually became tender to touch. Her past medical history and dental history were non-contributory. Extraoral examination revealed mild facial deformity. Intraorally, roughly oval swelling in region of 42 to 46 obliterating buccal vestibule was seen. Mucosa over the swelling was normal along with presence of a retained deciduous canine (Figure 1). Aspiration was performed which yielded dark reddish brown coloured aspirate. Panoramic radiography revealed well defined radiolucency associated with impacted canine showing flecks of calcification in periapical area of deciduous canine.

Resorption of roots of permanent first and second premolar along with mesial root of 1st molar was also seen. CBCT revealed presence of a large unilocular radiolucency with the expansion of well-corticated buccal and lingual cortical plates with the dimensions of lesion (18 X 30) mm. The lesion had enclosed tooth 43 in its entirety and displaced it mesially beneath the roots of contralateral central and lateral incisors (Figure 2). On the basis of history and clinical examination, provisional diagnosis was given as benign odontogenic tumor of the jaw. Incisional biopsy revealed cystic lumen lined by non-keratinized stratified squamous epithelium demonstrating basal layer of tall columnar cells with palisaded nuclei suggestive of ameloblast like cells. Overlying these cells were present loosely arranged stellate reticulum like cells. There was presence of numerous small to large, homogenous eosinophilic cells with pyknotic nuclei suggestive of "ghost cells". Among them some of the ghost cells stained with hematoxylin were suggestive of undergoing calcification. Underlying connective tissue demonstrated various endothelial lined blood vessels engorged with RBC's and there was presence of a mixed inflammatory cell infiltrate, predominantly lymphocytes. Focal areas demonstrated cholesterol crystals along with the presence of foreign body type of giant cells (Figure 3). Concluding the case history, clinical and radiographic examination along with the histopathologic findings a final diagnosis of simple cystic, type of calcifying cystic odontogenic tumor was made. It was surgically enucleated along with the impacted canine, under general anesthesia with nasal intubation. No evidence of recurrence was noted clinically or radiologically after 1years of follow up.

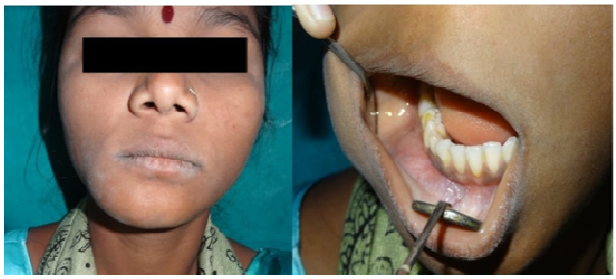

Figure 1. Extraoral view showing mild facial deformity and intraoral view showing vestibular obliteration

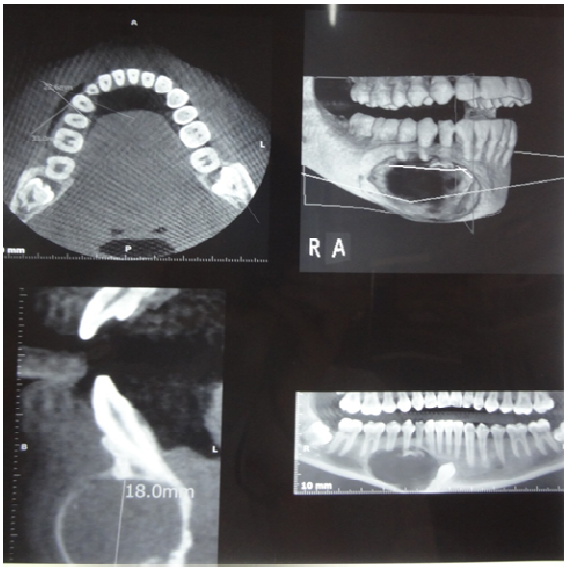

Figure 2. Radiographic view of lesion showing large unilocular radiolucency associated with expansion of cortical plates

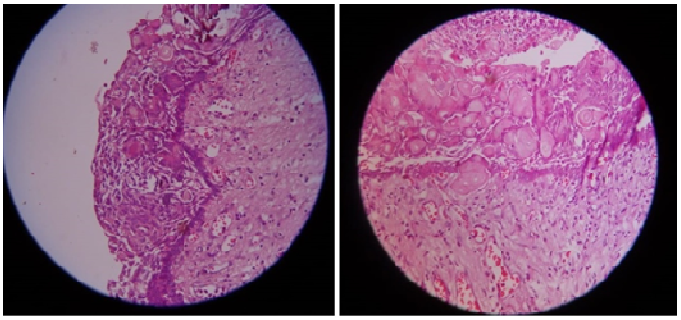

Figure 3. Histological view of lesion showing ghost cells

DISCUSSION

Owing to the diverse behavior of this lesion various types of descriptive terminologies have been allotted to address Calcifying cystic odontogenic tumor (CCOT). Initially it was Gorlin who addressed it as Gorlin's cyst in 1962. Gorlin also suggested that COC might be the oral analogue of dermal calcifying epithelioma of Malherbe (Gorlin *et al*., 1962). The first author to publish COC was Rywkind and later on he termed it as cholesteatoma of jaw. In 1992, World Health Organization (WHO) coined the term COC and referred it as odontogenic cyst of the jaws that resembles a tumor and was categorized under benign odontogenic tumors. CCOC was generally considered as a cyst, but many researchers choose to address it as a neoplasm, which led to further subcategorize the lesion into cystic and neoplastic variants. In 2005, WHO referred the cyst as CCOT and DGCT representing the cystic and neoplastic variants respectively. According to recent classification suggested by International Collaborative study on ghost cell odontogenic tumors for CCOT (2008) CCOT is classified as (Ledesma *et al*., 2008)

Type I	Simple cystic
Type II	Odontoma associated
Type III	Ameloblastomatous proliferating
Type IV	Associated with odontogenic tumors other than odontoma

The present case falls in the first category i.e. Type I – Simple cystic type.

The majority of the CCOT arise centrally in bone, but several peripheral (extraosseous) variants have been reported. The age of occurrence has been reported to vary from 3 to 80 years, with a definite peak incidence found in the second decade without any gender predilection. Most frequently it has been reported to occur in the incisor and canine region, with approximately equal frequency of 1:1 within the maxilla and mandible. According to Buchner, there was predisposition for the maxilla in Asians; whereas, there was 62% predilection for mandible in whites. Usually they appear as well-delineated swellings having smooth surface with a pink to reddish hue (Marx, 2003). They are usually asymptomatic unless secondarily infected. Radiographically, it is well-defined unilocular or multilocular radiolucent lesions having calcified bodies of different sizes and is associated with an odontome or an unerupted tooth. Root resorption of the associated teeth has also been observed in about 75-77 % of the cases. Thinning of the cortex might be seen as a result of expansion, along with soft central region yielding on pressure and the periphery hard on palpation.

Marx *et al*. in 2003 discussed three patterns of radiopacity with this tumor; first, salt and pepper pattern of flecks, second, fluffy cloudlike pattern throughout, and third, a crescent shaped pattern on one side of the radiolucency. Current case had small crescent shaped radiopacity in peripheral part of radiolucency close to periapical region of retained deciduous tooth. On aspiration the lesion usually yields a viscous, yellow granular fluid, but in present case it was serous and blood tinged. Extra-osseous variants of CCOTs may reveal a shallow depression in the bone along with occasional displacement of adjacent teeth (Shear, 2007). The most characteristic histopathological feature of CCOT is the ghost cells. They are characterized by eosinophilic, aberrant epithelial cells with intact outline but without nuclei. The exact process of formation of ghost cells is still controversial. It presents as coagulative necrosis and or aberrant proliferative keratinization of odontogenic epithelium (Gold, 1963; Günhan, 1993). Ghost cells might derive from the apoptotic process of odontogenic cells (Kim *et al*., 2000). Some theories shows that there is ischemic process, resulting into squamous metaplasia and further pregression of calcification process (Sedano, 1975). Other investigators suggested that ghost cells may represent the product of abortive enamel matrix in odontogenic epithelium (Mori *et al*., 1991; Takata *et al*., 2000). In some cases masses of ghost cells may coalesce together to form large sheets of amorphous material. However, these ghost cells are not exclusively seen in CCOT, but are also seen in other odontogenic tumors like odontoma, ameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma, dentinogenic ghost cell tumor and ghost cell odontogenic carcinoma. In the present case, the lesion appeared as a well-circumscribed, unilocular radiolucency with a crescent shaped small radiopaque fleck in the peripheral region. Age of the patient also lies in the most prevalent second decade. In majority of CCOT cases, feature such as impacted tooth most often a canine is associated which is in accordance with the current case along with the presence of root resorption.

Conclusion

Calcifying cystic odontogenic tumour may mimic numerous odontogenic and non odontogenic lesions, making diagnosis difficult. Histopathological findings and analysis are must for the diagnosis of the lesion. The present case manifested classical histopathological and clinical findings of simple cystic CCOT and ghost cells were very distinctly appreciated. However, the biological behavior of the different variants of CCOT can't be determined because of rarity of lesion.

Source of Support: Nil

Conflict of Interest: None Declared

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