Case Report

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Dentistry Section

Peripheral Calcifying Cystic Odontogenic Tumour - A Rare Case Report

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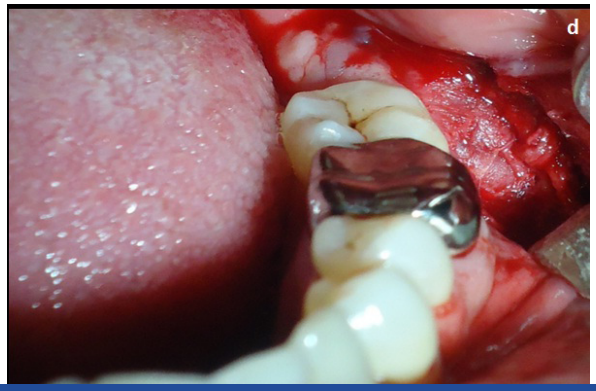
ABSTRACT

Odontogenic lesions are derived from remnants of the components of the developing tooth germ. The calcifying cystic odontogenic tumour (CCOT) is a benign cystic neoplasm of odontogenic origin that is characterized by ameloblastoma-like epithelial cells and ghost cells. Most peripheral CCOTs are located in the anterior gingiva of the mandible or maxilla. This is a rare case report of CCOT. The rare feature in our case was its peripheral nature of existence and its location in the left buccal vestibule and retromolar region. Based on the radiological, cytological and histopathological findings the lesion was surgically excised.

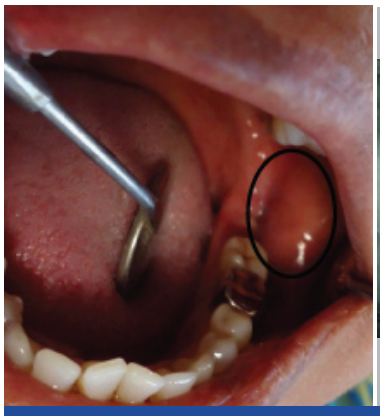
Keywords: Ghost cells, Mallory stain, Peripheral lesion, Van Gieson stain

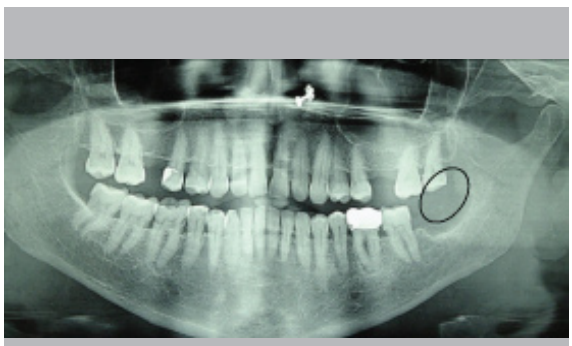
CASE REPORT

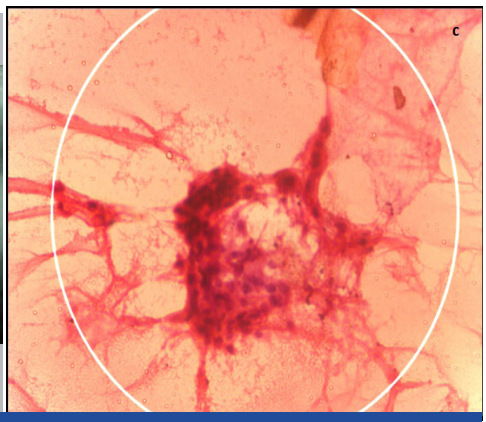
An 59-year-old female patient reported to outpatient department of Vidya Shikshan Prasarak Mandal's Dental College and Research Centre (VSPM's DCRC) Nagpur, India with a chief complaint of a swelling in left posterior region of lower jaw since 8 months. The swelling was nodular, sessile, ovoid in shape with diffuse margins, soft to firm on palpation, painless and of size 3x3 cm approx. covered by a normal oral mucosa. The swelling was located in the buccal vestibule involving buccal mucosa in relation with second molar [Table/Fig-1a]. There was no evidence of any associated dental infection or periodontal defect however the patient underwent extraction of mandibular left third molar three months back in order to relieve cheek bite. Past medical history was not contributory. In panoramic and lateral oblique radiographs, a well defined radiolucency was seen in third molar region extending upto ascending ramus of mandible [Table/Fig-1b]. After obtaining informed consent from the patient, aspiration from lower left buccal vestibule in the retromolar region was done. 3.5 ml of straw coloured slightly turbid fluid was obtained and exfoliative cytology was performed which revealed chronic inflammatory cell infiltrate [Table/Fig-1c]. After aspiration of fluid, size of the swelling was slightly reduced. The history of extraction of mandibular left third molar and clinical features made us to arrive at a provisional diagnosis of residual cyst. The lesion was enucleated along with curettage under local anesthesia and subjected to histopathological study. During cyst enucleation, procedure, peripheral nature of the lesion could be noticed. Osseous erosion of cortical bone was seen in relation to mandibular left third molar [Table/Fig-1d]. The histological examination revealed a cystic lumen lined by stratified squamous parakeratinised epithelium with associated fibrovascular connective tissue capsule. The epithelial lining of 3-4 cells thickness was seen exhibiting globules of eosinophilic structure suggestive of ghost cells [Table/Fig-2a-c]. Ghost cells were very distinctly appreciated by the special stains like Van Gieson and Mallory [Table/Fig-3a-c]. Ghost cells appeared yellow coloured in Van Gieson stain and orange coloured in Mallory stain. So, based on the clinical findings, radiological and histopathological investigations final diagnosis of



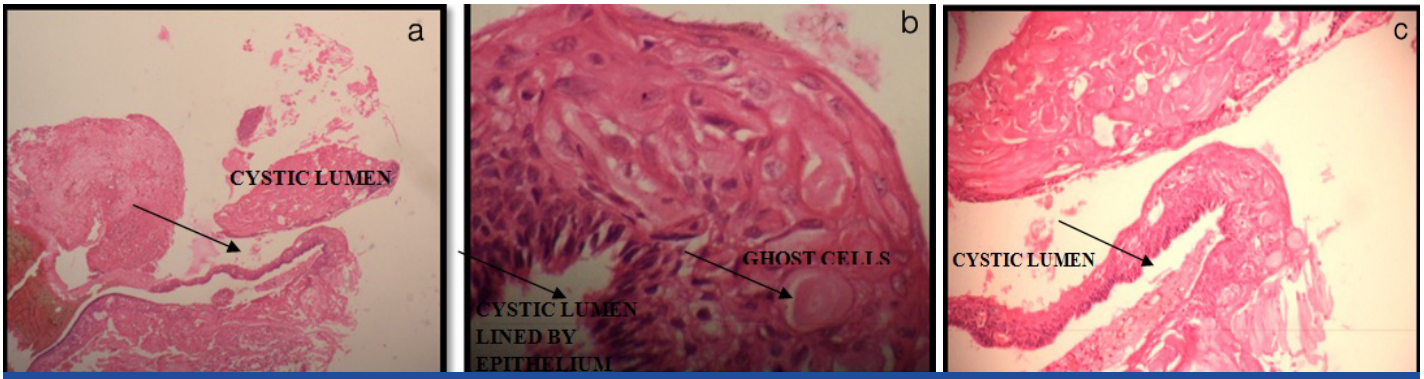
[Table/Fig-1d]: Photograph showing cystic lining after reflecting mucoperisteal flap



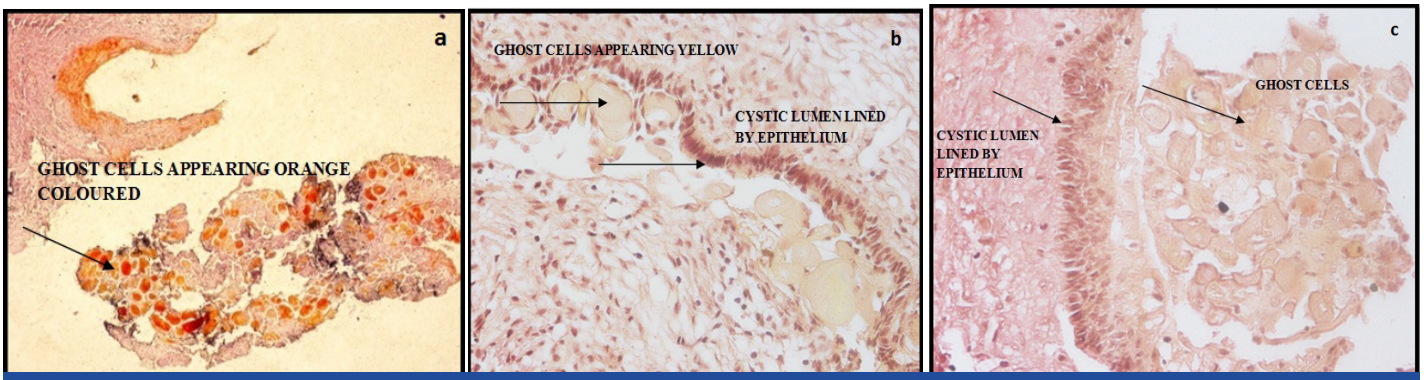




[Table/Fig-1a]: Clinical photograph showing swelling in cheek mucosa adjacent to lower left second molar **[Table/Fig-1b]:** Orthopantomogram showing bony erosion in left retromolar region with well corticated margins **[Table/Fig-1c]:** Fine needle aspiration cytology. H&E Stain (100X): Cytological smear showing chronic inflammatory cell infiltrate



[Table/Fig-2a]: H&E Stain, Scanner view (4X): Photomicrograph showing cystic lumen lined by odontogenic epithelium **[Table/Fig-2b]:** H&E Stain, High power view (40X): Photomicrograph showing cystic lumen lined by odontogenic epithelium with basal cells that are cuboidal to columnar in shape showing hyperchromatism. Numerous ghost cells are seen in the cystic epithelium of variable sizes with nuclei in different stages of degeneration **[Table/Fig-2c]:** H&E Stain, Low power view (10X): Photomicrograph showing cystic lumen lined by odontogenic epithelium



[Table/Fig-3a]: Special stain, Scanner view (4x): Ghost cells appear orange coloured in Mallory stain. **[Table/Fig-3b]:** Special stain, High power view (40X): Ghost cells appear yellow coloured in Van Gieson stain **[Table/Fig-3c]:** Special stain, High power views (40X): Ghost cells appear yellow coloured in Van Geison stain

peripheral CCOT was made. Following removal of the lesion, the healing process was uneventful.

DISCUSSION

The calcifying cystic odontogenic tumour (CCOT) was first described by Gorlin et al., (1962,1964) who were impressed by significant presence of so called 'ghost cells' and its histological resemblance to the cutaneous calcifying epithelioma of Malherbe [1]. There was a report of four cases by Gold, who chose a similar, term for the lesion, namely "keratinizing and calcifying odontogenic cyst" [2]. In 2005, the World Health Organization (WHO) designated Gorlin's cyst as a tumour and described it as belonging to a group of related neoplasm, including the benign cystic-type (CCOT) [3]. A calcifying cystic odontogenic tumour (CCOT) is an extremely rare benign cystic neoplasm that is characterized by an ameloblastoma-like epithelium and ghost cells that have the potential to undergo calcification [3]. There is a wide age range from 1 to 82 years with a first peak in the second decade and the second peak in the sixth/seventh decade. CCOT presents both intraosseous (central) and extraosseous (peripheral) locations. The intraosseous CCOT is a unilocular or multilocular destructive radiolucent lesion that may contain irregular calcifications [4]. The peripheral CCOT represents less than 25% of all CCOTs [5].

Odontogenic tumours that are derived from the epithelial, mesenchymal, or epithelial/mesenchymal remnants of the components of the developing tooth germ are relatively uncommon lesions. They are found in the mandible and maxilla and must be considered in differential diagnoses of lesions involving these sites. They can be classified by location as peripheral or central lesions [6]. Peripheral odontogenic lesions are rare, exhibiting the histologic features of their central counterpart but occur only in the soft tissue covering the tooth-bearing portion of the maxilla and mandible [7]. CCOTs are believed to be derived from odontogenic epithelial remnants within the gingiva or within the mandible or maxilla. The presence of ghost cells, the characteristic microscopic feature of CCOT, may also be seen in other lesions, including ameloblastomas, odontomas, adenomatoid odontogenic tumours, ameloblastic fibroodontomas, and ameloblastic fibromas [7]. Although ghost cell formation can be seen in several odontogenic and nonodontogenic lesions e.g. central giant cell granuloma, peripheral giant cell granuloma, cherubism, paget's disease, fibrous displasia, it is characteristic of CCOT [8]. CCOT is divided into two different entities, peripheral and central, on the basis of clinicopathologic features. The peripheral epithelial tumour resembles the peripheral ameloblastoma except for ghost cell formations in the central portion of neoplastic epithelial clusters and juxtaepithelial dentinoid. CCOTs occurred predominantly as nodular swellings on the edentulous alveolar mucosa of denture wearers, a feature that implicates trauma or irritation as aetiologic factors [8]. In this case aetiological factor was trauma i.e. cheek bite, for which the patient had undergone extraction of third molar. Radiographically the extra-osseous lesion shows localized superficial alveolar bone resorption, or saucer-shaped radiolucencies [1] as also seen in our case.

Buchner et al., noted that peripheral CCOT tend to occur more often in mandibular incisor/canine and premolar areas and also occurred more often in females (66.6%) than in males (33.3%) [7]. Resende et al., in their review of 44 well-defined cases of peripheral CCOT [9], also found a slight predilection for females and the anterior region. However, they found a similar distribution throughout the maxillary (40.9%) and mandible (47.7%) regions. In their study, the mean age at the time of diagnosis was 49.4 years. Here, we are reporting a case of CCOT in retromolar region in vestibule extending into the cheek.

The nature of ghost cells is not clearly known. In literature, it has been demonstrated as positive expression of 'amelogenin protein' in the cytoplasm, suggesting that the epithelium lining of CCOT might show ameloblastic differentiation in ghost cells [10]. Many investigators have made efforts to clarify the nature of ghost cells by

employing special histochemical methods and using transmission electron microscopy, scanning electron microscopy. SP Hong et al., all believed that ghost cells represent normal or abnormal keratinization [8]. According to them, the ghost cells represented different stages of normal and aberrant keratin formation and that they were derived from the metaplastic transformation of odontogenic epithelium. Other investigators suggested or implied that ghost cells may represent the product of abortive enamel matrix in odontogenic epithelium. However, the morphology of ghost cells seems different from that of enamel matrix [8]. Due to the non-aggressive behaviour of peripheral calcifying cystic odontogenic tumour conservative treatment like enucleation or local resection is appropriate [3]. The lack of recurrence depends on the degree of completion of the excision.

CONCLUSION

Calcifying cystic odontogenic tumour may mimic numerous odontogenic and non odontogenic lesions, making diagnosis difficult. Histological findings have a crucial role in diagnosis of the lesion. So, all biopsy material should be sent for histological examination. Our case represent classical histological and clinical findings of Peripheral CCOT and ghost cells were very distinctly appreciated by the special stains like Van Gieson and Mallory.

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