

## CALCIFYING ODONTOGENIC CYST - A CASE REPORT

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### ABSTRACT

The calcifying odontogenic cyst (cocc) is a developmental odontogenic cyst, which was first categorized as a distinct entity by Gorlin in 1962. It is an unusual and unique lesion, which may show characteristics of both a solid neoplasm and a cyst. It usually occurs as an intraosseous lesion but may occasionally occur as an extraosseous or peripheral variant. It shows a nearly equal distribution between the maxilla and mandible and is commonly seen anterior to the first molar. The clinical and radiographic features of this lesion are not pathognomonic, and it is characterized by its histological diversity, with the most characteristic feature being the presence of a variable number of ghost cells within the epithelial component. Treatment of choice for COC is surgical enucleation which is done in a conservative manner and recurrences are unlikely. This report describes a case of COC of a 26-year-old female in association with swelling on the left side of the maxilla since 2 years.

**KEY WORDS:** Calcifying odontogenic cyst, Ghost cell, Calcifying odontogenic tumor

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## INTRODUCTION

Calcifying odontogenic cyst (COC) an uncommon benign odontogenic lesion was first categorized as a distinct entity by Gorlin et al. in 1962<sup>1</sup> and was named after him since then<sup>2</sup>. Although named and defined as a cyst, there is no agreement in the literature regarding its classification as a cyst or a neoplasm, as some examples of COC shows areas suggestive of neoplasia<sup>3</sup>. In 1992, WHO classified COC as a neoplasm rather than a cyst, but confirmed that most of the cases are nonneoplastic<sup>4</sup>. In view of this duality, many different terminologies have been applied to cystic and solid COC variants, but COC is still the preferred term<sup>5</sup>. It represents 2% of all odontogenic pathological changes in the jaw. The COC usually arises intraosseously, but it may also occur extraosseously, with about equal frequency in the mandible and maxilla (1:1). The age of the patients may range from 5 to 92 years, with peak incidence in the second and sixth decade of life.<sup>6</sup> The cyst occur with equal frequency in both genders. Buchner reported that in Asians there was predilection for the maxilla, whereas in whites there was 62% predisposition for mandible<sup>7</sup>. COC is usually asymptomatic and may be an incidental radiographic finding. Radiographically, the lesion appears as a unilocular or multilocular well defined radiolucency that may contain small irregular calcified bodies of varying sizes and it may be associated with an odontome or an unerupted tooth<sup>8</sup>.

## CASE REPORT

A 26 years female reported to the department of oral and maxillofacial surgery, ucms college of Dental Surgery with a Chief complain of painless swelling on left side of maxilla since 2 years. The swelling was small initially which gradually increased to a size of 3cm\*2.5cm\*2.1cm. It was well confined, hard, nonfluctant, non tender and was not associated with any discharge. Examination of CN V and CN VII was normal. Overlying skin was normal and there was no obliteration of nasolabial fold. There was no relevant medical and family history. No history of parafunctional or deleterious habits present.



Fig1: Extraoral view



Fig 2: Intraoral view

Extraoral examination in fig.1 revealed an asymptomatic swelling extending from left side of nasolabial fold to the

zygoma which was hard and non tender on palpation. There was no obvious change in skin colour and texture present.

Intraorally in fig.2 there was a diffuse swelling extending from 21 to 25. Mucosa was normal in color, consistency, hard and non tender on palpation. There was no mobility or tenderness to palpation with any tooth. No sign of caries, pulp pathosis or periodontitis.



Fig: 3 CT scan reveals unilocular radiolucent lesion with radiopaque foci

CT scan, fig.3 revealed a well defined unilocular radiolucent lesion with radiopaque foci extending from 22 to 25 with obvious expansion and thinning of buccal cortical plate. No sign of root resorption, displacement or impacted tooth and bony perforation were revealed. Radiological differential diagnosis of adenomatoid odontogenic tumor, ossifying fibroma, complex odontome, ameloblastic fibroodontome, intraosseous CEOC, CEOT were made.

Patient was advised to go for routine blood investigation, serology, serum calcium, serum phosphorus, alkaline phosphatase which were in normal range. Incisional biopsy was performed under local anesthesia and histopathologic diagnosis of calcifying odontogenic cyst was made. Patient was planned for surgical enucleation under general anesthesia for definitive treatment.

Surgical enucleation of a lesion was done conservatively in fig.4 and specimen in fig.5 was sent for the histopathological examination. Defect was packed with ribbon gauze impregnated with neomycin initially. On subsequent visit patient was kept on iodoform dressing and was changed periodically until the defect was filled by granulation tissue. Healing was uneventful at 6<sup>th</sup> month period in fig.6 and is still under close follow up.



Fig4: Enucleation of the lesion



Fig 5: Specimen for biopsy



Fig 6: Postoperative 6 week

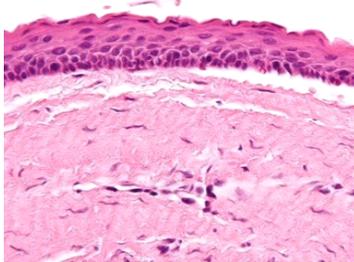


Fig 7: Histopathological section showing the cyst lining (Haematoxylin and eosin stain 10X)

Final histopathological report in fig.7 revealed fibrous capsule with a lining of odontogenic epithelium. The epithelium shows columnar to cuboidal basal cells with overlying loosely arranged epithelial cells with few resembling stellate reticulum like cells, varying number of ghost cells are evident with few undergoing calcifications. Features are suggestive of calcifying epithelial odontogenic cyst.

## DISCUSSION

Since 1971, calcifying odontogenic cyst has been described by the World Health Organization as a non-neoplastic cystic lesion in which the epithelium lining shows a well-defined basal layer that is often many cells thick and that may resemble the stellate reticulum of an enamel organ, and masses of ghost epithelium cells that may be in the epithelial cyst lining or in the fibrous capsule.<sup>9</sup> The ghost cells may become calcified and dysplastic dentin may be laid down next to the basal cell layer of the epithelium. It may be associated with complex odontoma or with tissue resembling an ameloblastic fibro-odontoma.

As the number of reports increased, it was proposed that CEOC was indeed a heterogenous group of entities, with distinct histopathologic findings that included solid tumor. Praetorius<sup>10</sup> proposed a subclassification for the heterogenous group of calcifying odontogenic cysts, in which the cystic lesions were separated from the neoplasms (solid lesions). These researchers further divided the cystic entity into three types, the simple unicystic type, the unicystic-odontoma-producing type, and the ameloblastomatous proliferating type. Calcifying odontogenic cyst may be otherwise describe as developmental odontogenic lesion that has its cutaneous counterpart in Malherbe's calcifying epithelioma<sup>11</sup>

The case report is in agreement with the literature finding that calcifying odontogenic cysts occur predominantly as an intraosseous lesion. Other authors have reported only 13 to 21 percent of the cysts to be peripheral (extraosseous) lesions<sup>12</sup>. The radiographic findings of this case (unilocular radiolucency with a radiopaque mass and well circumscribed borders) are encountered in the majority of odontoma producing intraosseous calcifying odontogenic cysts<sup>13</sup>. This variant was more prevalent in females, with a mean age at discovery of 16 yrs and the most cases were located in the maxilla. The findings of our case are in accordance with the above mentioned observations. In spite of the low frequency of this lesion and the fact that most cases are surgically removed and heal uneventfully, there must be a close follow up because there have been reports of association with carcinoma, adenomatoid tumor and ameloblastoma<sup>14</sup>.

## CONCLUSION

The COC, is an uncommon lesions which may show characteristics of both cyst and a solid neoplasm. Its clinical and radiographic features may mimic other odontogenic cysts / tumors and a definitive diagnosis can only be made histologically.