Review Article

Ameloblastomatous Calcifying Odontogenic Cyst: A Rare Entity

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Abstract

Introduction: Ameloblastomatous calcifying odontogenic cyst (COC) is an extremely rare histopathologic variant of COC, an odontogenic cyst of the jaws. It needs to be differentiated from closely associated variant ameloblastoma ex COC that is entitled to a more aggressive form of surgical management. **Aim:** The aim of this paper is to present a case of ameloblastomatous COC of the right mandibular angle region with review of literature describing this rare entity. The paper also highlights the requirement of including this lesion in differential diagnosis of various jaw lesions. **Materials and Methods:** Google search, Wikipedia, ScienceDirect, MEDLINE, the Cochrane library, and PubMed were used extensively to search and collect all reported cases of ameloblastomatous COC using keywords such as ameloblastomatous COC, COC, maxilla, mandible, and gorlin cyst ameloblastic proliferation. **Results:** To the best of our knowledge, a total of twenty one cases of ameloblastomatous COC have been reported in the literature in the maxilla-mandibular region and we represent the twenty second case in a 20-year-old female patient along with the review. **Conclusion:** Ameloblastomatous COC – a subtype of COC needs to be differentiated from true ameloblastoma arising from COC as it warrants a conservative form of surgical management unlike other neoplastic variants of COC. Owing to scarcity of data of this lesion in literature, more reporting of such cases is required to shed light on its behavior.

Keywords: Ameloblastomatous calcifying odontogenic cyst, calcifying odontogenic cyst, ghost cell, gorlin cyst ameloblastic proliferation, mandible, maxilla

Introduction

Since Gorlin's first discussion of calcifying epithelial odontogenic cyst in 1962,[1] the World Health Organization (WHO) in 2005 had classified calcifying odontogenic cyst (COC) in the group of odontogenic tumors having odontogenic epithelium with odontogenic ectomesenchyme with/without dental hard tissue formation which now in 2017 has been reclassified in the group of odontogenic cysts. It is well known that the lesion is often associated with the tumors of odontogenic origin such as odontoma, ameloblastoma, and adenomatoid odontogenic tumor.[2] Although the association of ameloblastoma with this lesion is important, only a few such cases with synchronous representation of more than one type have been reported so far. WHO classification by Kramer and Pindborg used the term COC and described its cystic or neoplastic variants in the jaw in 1992.[3] Even majority of authors have categorized it under two basic groups of cysts and tumors. Due to its variable histology, clinical behavior and association with dysregulated β-catenin signaling, there is confusion till date whether or not it is a reactive or developmental or neoplastic entity.[4] The cystic variant of COC is characterized by a unicystic lesion associated with or without odontoma and are in majority. They may also show ameloblastomatous proliferative activity intraluminally or intramurally.[4] The neoplastic variants of COC which show a solid growth pattern consisting of ameloblastoma such as strands and islands of odontogenic epithelium infiltrating into mature fibrous connective tissue, are further subclassified into ameloblastoma arising from COC (ameloblastoma ex COC) and odontogenic ghost cell tumors.[4] Malignant transformation of COC has been reported.[5]

Ameloblastomatous COC resembles unicystic ameloblastoma except for ghost cells and calcifications within the proliferative epithelium and the fact that it occurs only intraosseously.

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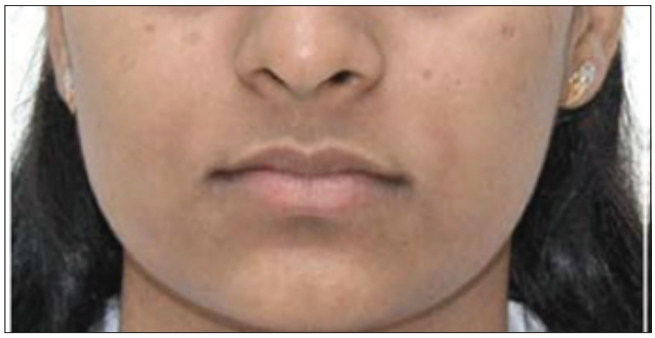

Figure 1: Preoperative front facial profile

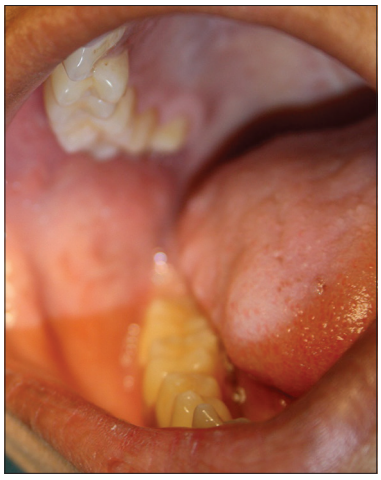

Figure 3: Preoperative intraoral view

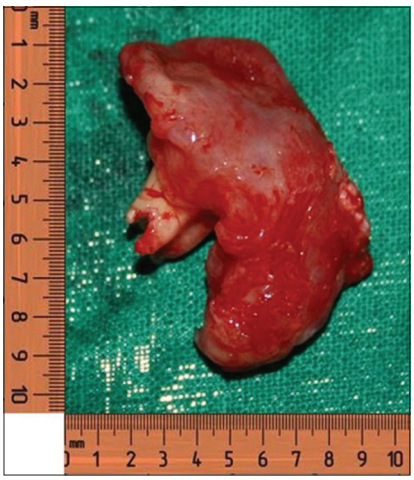

Figure 5: Excised lesion

Newer guidelines by the WHO have mentioned it in the group of cysts. The purpose of this paper is to present a case of ameloblastomatous COC along with a systemic review in the scientific literature.

Case Report

An asymptomatic 20-year-old female visited our institute for treatment of her malaligned teeth [Figure 1]. Her

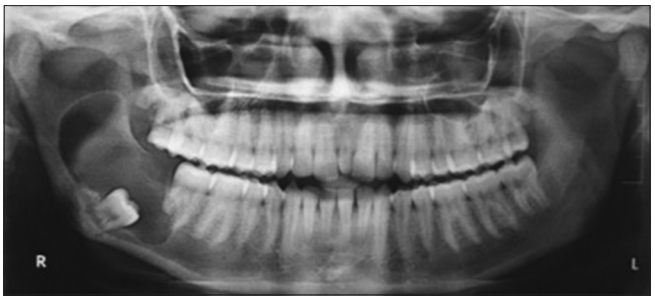

Figure 2: Preoperative orthopantomogram

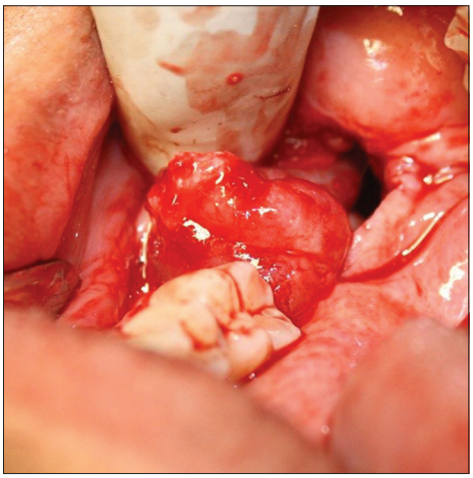

Figure 4: Intraoperative intraoral view

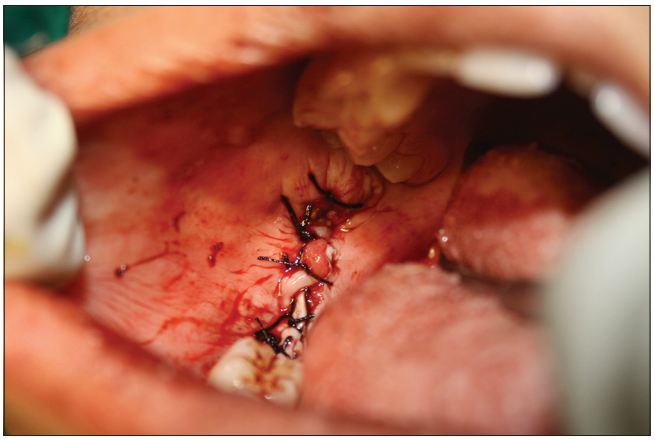

Figure 6: Immediate postoperative intraoral view

orthopantomogram revealed a unilocular, well-defined radiolucency extending from the right mandibular second molar to condylar and coronoid process involving the entire ramus. Root resorption of mandibular right second molar was present. The lesion had mandibular right third molar located inferiorly toward the lower border of the mandible [Figure 2]. Calcifications were not evident in the radiograph. There was no limitation in temporomandibular joint movements or sensory disturbances in lower lip. The oral examination was also within normal limits [Figure 3]. Mandibular right second molar was nonvital. A differential diagnosis of cystic ameloblastoma,

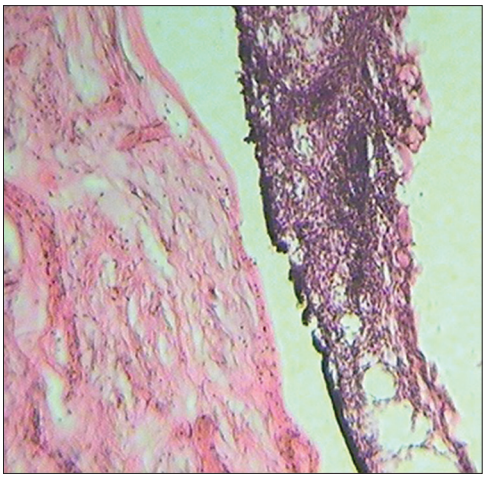

Figure 7: Microscopic view of H and E slide (×40)

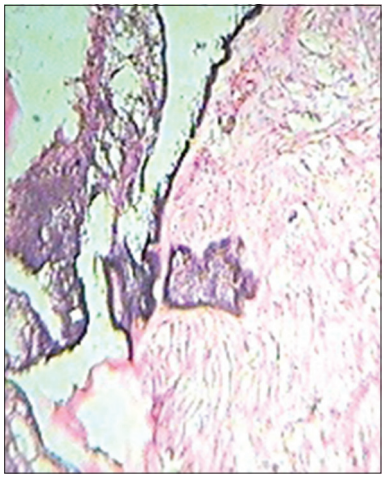

Figure 8: Microscopic view of H and E slide (×10)

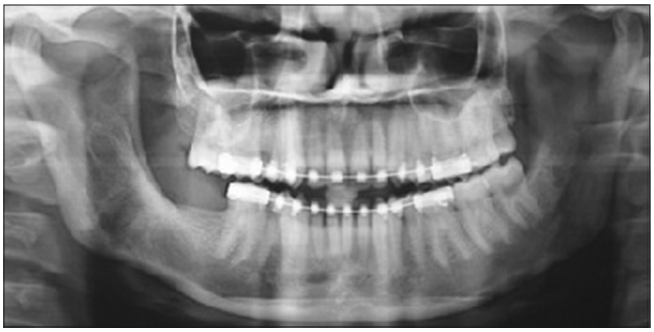

Figure 9: Postoperative orthopantomogram after 6 months

dentigerous cyst, and odontogenic keratocyst was made. The lesion was enucleated with removal of second and third molars through intraoral approach followed by chemical cauterization using carnoy's solution [Figure 4]. The excised lesion [Figure 5] was sent to Department of Oral Pathology in the same institute for histopathological examination. Closure was performed using silk sutures [Figure 6]. Histopathological report revealed thin odontogenic epithelial lining in some region and luminal proliferation in some region. Tall columnar ameloblast like basal cells were present with hyperchromatic nuclei. Above the basal layer, cells were loosely arranged resembling stellate reticulum. The presence of ghost cell in the cystic lining was noted. Underlying connective tissue showed dentinoid-like material with mature collagen fiber bundles. Ameloblastic follicles and odontogenic islands were present in some regions along with presence of nonkeratinized stratified squamous epithelium [Figure 7 and 8]. These features were confirmatory for ameloblastomatous COC. The bone healing is satisfactory [Figure 9] and there has been no recurrence of the lesion since the past 6 years. Written consent and approval for publication were obtained from the patient.

Materials and Methods

We methodically searched Google Scholar, Wikipedia, MEDLINE, the Cochrane library, and ScienceDirect using keywords as described previously to review all the content regarding ameloblastomatous COC. We even checked for the references of the articles we found in these sites and thoroughly studied them. In total, we found 21 cases of ameloblastomatous COC and have included ours as the twenty-second case. Hong *et al*.[4] in his study presented eleven cases of ameloblastomatous COC but did not reveal regarding the clinical features, bone resorption, and associated impacted teeth. Hence, we have not included his work in our review. The clinical and radiographic findings of these cases are shown in Table 1. The tooth numbering system used in this article is Zsigmondy palmEr notation system.

Discussion

Since the first description of eleven cases of COC in 1962 by Gorlin,[1] the lesion became recognized as a distinct entity and is considered as an analog of cutaneous calcifying epithelioma of Malherbe. It is an uncommon lesion and accounts from 1% to 2% of all odontogenic jaw cysts.[2] In 1971, WHO defined COC as "a nonneoplastic cystic lesion, in which the epithelial lining shows a well-defined basal layer of columnar cells, an overlying layer that is often many cell layers thick that may resemble stellate reticulum and masses of ghost cells that may be in the epithelial cyst lining or in the fibrous capsule. The ghost cells may become calcified. Next, to the basal layer of the epithelium, dysplastic dentin may be laid down."[14] Derived from odontogenic epithelium, COC is usually intraosseous having about 70% of cases and rarely extraosseous accounting for <25% of cases.[15] It has been reported to be found between the first and ninth decades of life, with a highest incidence occurring in the sixth and seventh decades and tends to appear with equal frequency among males and females. It is predominantly found in the anterior region.[16]

Various names given by researchers to COC over the past years are as given in Table 2.[2,17] It has been accepted that there are two variants of COC: cystic and neoplastic. Due to this, dualistic nature various classifications have been proposed by different clinicians.

Table 1: The previous cases of ameloblastomatous calcifying odontogenic cyst

Reference	Age and sex	Presentation	Bone involved	Size of lesion	Root resorption	Cortical plate	Radiology	Recurrence	Histologic diagnosis	Impacted teeth
Aithal *et al*.[6]	28 years, female	Tender swelling in lower left posterior region	Mandibular left posterior region	2.5 cm × 2 cm	Absent	Buccal plate expanded	Multilocular radiolucency from 33 to 37 region	Absent	Ameloblastomatous COC	Absent
Iida *et al*.[7]	17 years, male	Tender right facial swelling	Mandibular right posterior region	DNF	Absent	Buccolingual expansion	Multilocular radiolucency from 47 to right ramus and coronoid region	Absent	Ameloblastomatous COC	Present
Kamboj and Juneja[8]	58 years, female	Tender right facial swelling	Mandibular left posterior	DNF	Absent	Buccolingual expansion	Multilocular radiolucency-43 to condyle-coronoid region	DNF	Ameloblastomatous COC	Absent
Ledesma-Montes *et al*.[9]	Details of cases by Ledesma et al in Table 3									
Nosrati and Seyedmajidi[10]	22 years, male	Nontender swelling-right facial region	Mandibular right posterior region	DNF	Present in mandibular right first and second molar	Buccolingual expansion	Unilocular radiolucency from 46 to 48 region	Absent	Ameloblastomatous COC	Present
Gupta and Gupta[11]	65 years, male	Tender swelling in lower left jaw	Mandibular left posterior region	4 cm × 5 cm	Absent	Buccal bone perforated	Bilocular radiolucency 37 to 38 region	Absent	Multicystic ameloblastomatous COC	Absent
Singh *et al*.[12]	24 years, female	Left facial swelling	Mandibular left anterior region	5 cm × 3 cm	Absent	Buccolingual expansion	Unilocular radiolucency from 35 to 48 region	Absent	Ameloblastomatous calcifying ghost cell odontogenic cyst	Present
Tamanna *et al*.[13]	21 years, male	Tender swelling on lower left jaw	Mandibular left posterior region	DNF	Present in mandibular left second molar region	Buccolingual expansion	Unilocular radiolucency in left ramus region	Present	Ameloblastomatous COC	Present
Present case	20 years, female	Asymptomatic	Mandibular right posterior region	8 cm × 8 cm	Present in mandibular right second molar region	Absent	Unilocular radiolucency from 37 to condyle-coronoid region	Absent	Ameloblastomatous COC	Present

COC=Calcifying odontogenic cyst; DNF=Data not found

Table 2: Various names for calcifying odontogenic cyst[2,23]

Name of researcher	Year	Name given for COC
Gorlin *et al*.	1962	COC
Gold	1963	Keratinizing COC
Bhaskar	1965	Keratinizing ameloblastoma
Fejerskov and Krogh	1972	Calcifying ghost cell odontogenic tumor
Freedman *et al*.	1975	Cystic calcifying odontogenic tumor
Praetorius *et al*.	1981	Dentinogenic ghost cell tumor
Ellis and Shmookler	1986	Epithelial odontogenic ghost cell tumor
Colmenero *et al*.	1990	Odontogenic ghost cell tumor
Shear	1994	Odontogenic ghost cell ameloblastoma
Wirshberg *et al*.	1994	Odontocalcifying odontogenic tumor
Toida *et al*.	1998	Calcifying ghost cell odontogenic cyst
WHO	2005	Calcifying cystic odontogenic tumor

COC=Calcifying odontogenic cyst; WHO=World Health Organization

First classification is proposed by Praetorius *et al*.,[18] in 1981:

Type 1: Cystic type:
a. Simple unicystic type
b. Odontoma-producing type
c. Ameloblastomatous proliferating type.

Type 2: Neoplastic type: Dentinogenic ghost cell tumor (DGCT).

Classification by Hong *et al*.,[4] in 1991.

Type 1: Cystic
a. Nonproliferative
b. Proliferative
c. Ameloblastomatous
d. Associated with odontoma.

Type 2: Neoplastic:
a. Ameloblastoma ex COC
b. Peripheral epithelial odontogenic ghost cell tumor
c. Central epithelial odontogenic ghost cell tumor.

Classification of COC proposed by Buchner:[5]
A. Peripheral (extraosseous) COC
    1. Cystic variant
    2. Neoplastic (solid) variant.
B. Central (intraosseous) COC
    1. Cystic variant
        a. Simple (unicystic or multicystic)
        b. Associated with an odontoma
        c. Associated with odontogenic tumors (other than odontoma)
        d. Other variants (such as clear cell variant, pigmented variant).
    2. Neoplastic (solid) variant: known as DGCT or epithelial odontogenic ghost cell tumor (EOGCT)
    3. Malignant COC.

Dualistic classification of COC by Toida[2] in 1998:
1. Cyst: Calcifying ghost cell odontogenic cyst (CGCOC)
2. Neoplasm:
    A. Benign: Calcifying ghost cell odontogenic tumor (CGCOT)
        a. Cystic variant: Cystic CGCOT
        b. Solid variant: Solid CGCOT.
    B. Malignant: Malignant CGCOT
3. Combined lesion: Each of the categories described above (CGCOC, CGCOT and malignant CGCOT) associated with the following lesions
    a. Odontoma
    b. Ameloblastoma
    c. Other odontogenic lesions.

Proterious classified COC into cysts and neoplasms after staining them using Goldner staining, masson's trichrome and van geison connective tissue staining.[18] The COC presents a wide range of histologic features. The lesion is occasionally associated with odontomas and other odontogenlc tumors such as ameloblastoma, ameloblastic fibroma, and ameloblastic fibroodontoma.[19]

Some authors have described COC as calcifying cystic odontogenic tumor (CCOT) as they have more heterogeneous components than envisaged. In 2005, WHO in their classification of odontogenic tumors, classified COC neoplastic lesions into GCOT which comprised of CCOT, DGCT, and GCOC. In this guideline, CCOT is defined as a benign cystic neoplasm of odontogenic origin, characterized by ameloblastoma-like epithelium with ghost cells that may be calcified.[20] In a detailed multicentre review of ghost cell lesions, Ledesma-Montes *et al*.[9] showed that over 85% of ghost cell lesions are simple cysts which may be alone (in 65% of cases) or associated with odontomas. Ameloblastomatous proliferations were shown by very few and comprised of only three patients (2.46%) while 5% of lesions were solid and could be regarded as true neoplastic DGCTs which also agreed with the study by Hong *et al*.[4]

Thus, a good evidence established that CEOT should be regarded as cysts, which arise alone or in association with other lesions, especially odontomas. Recently, in the "WHO classification of head and neck tumors" in 2017, the consensus was researched, and reclassification of CCOT was agreed on to revert back to the original terminology and classify cyst as COC due to its nonneoplastic clinical behavior[21] and the neoplasm as DGCT.[22] CCOT was listed as a synonym in this latest edition. However, these fluctuations reflect the lack of precise knowledge of the pathogenesis as well as overlapping definition of tumors and cysts.[23]

One of the variants of COC, ameloblastomatous type as described by Hong *et al*.,[4] and Ledesma-Montes *et al*.,[9] is what our article is about along with its review. On microscopic examination, ameloblastomatous COC resembles unicystic ameloblastoma except for the ghost cells and calcifications within the proliferative epithelium. Ameloblastomatous COC only occurs intraosseously. This subtype of COC needs to be differentiated from the ameloblastoma arising in COC as the former requires conservative form of management while the later demands for aggressive measures for its treatment. In contrast to ameloblastoma ex COC, dystrophic calcifications and ghost cells are within the proliferative epithelium, lacking histopathologic criteria as suggested by Vickers and Gorlin[24] and is confined to the cystic lumen. Ameloblastoma ex COC designates an ameloblastoma arising from the cyst lining epithelium of COC.[8] Ameloblastoma ex COC occurs intraosseously, appearing as cyst-like, radiolucent lesions. Whether this tumor is potentially as destructive as typical ameloblastoma and has the same propensity for recurrence is unknown. Buchner[5] suggested that if the COC was associated with an ameloblastoma, its behavior and prognosis would be same as that of ameloblastoma, not COC. There have been numerous reports of ameloblastoma arising from odontogenic cysts, including radicular cyst, dentigerous cyst, primordial cyst, and residual cyst.[4]

From the genetic point of view, missense mutations in CTNNB1 which encodes beta-catenin were found in 91% of the cases studied by Yukimori *et al*.,[23] and this activation of beta-catenin abolishes the phosphorylation sites Asp32, Ser33, or Ser37. Mutations in CTNNB1 are the major driver mutations of CCOT and that CCOT is a genetic analog of pilomatrixoma and adamantinomatous craniopharyngioma in the odontogenic tissue.

On reviewing the literature, we found the reporting of twenty-one cases out of which Hong *et al*.,[4] reviewed ninety-two cases of COCs which were divided into 85.9% cases of cysts and 14.1% cases of neoplasms. From these ninety-two cases, eleven cases were of ameloblastomatous COC which were characterized by ameloblastoma-like, cyst-lining epithelium with ghost cells and calcifications. Nine

of eleven cases occurred in mandible and two in maxilla. In his article, he did not give details such as bone resorption, radiolucency, or recurrence rate. Thus, we have not included his research in our review.

From our review of eleven cases, the age of occurrence ranges from 15 to 65 years with the mean age of occurrence being 34.09 years. Female predilection is concurrent with the literature. It was found that in all the cases, the site of lesion was mandible. Clinical presentation varies from being asymptomatic to painful swelling with or without bony expansion. From the cases reviewed, five patients (45.45% of cases) had mild pain present over the site of lesion. Seven patients (63.63% of cases) had swelling present while only one patient was asymptomatic. Details regarding pain and swelling were not mentioned in review by Ledesma-Montes *et al*.,[9] regarding the three patients.

Nearly 81.81% of the lesions were in the mandibular posterior region, 9.09% were in the mandibular anterior region while 9.09% were in the mandible wherein the region was not specified. This showed propensity of the lesion toward ramus-body area. Radiographically, ameloblastomatous COC presents itself as a radiolucent lesion which may be multilocular or unilocular with well-defined borders. Nearly 54.5% of the cases represented unilocular radiographic picture while 36.3% cases showed a multilocular radiographic picture. Root resorption was present in three cases (27.27%). Buccolingual plate expansion was noted in seven cases (63.6%). Impacted teeth were present in 54.54% of cases. Most probable differential diagnosis for this lesion includes dentigerous cyst, ameloblastoma, odontogenic keratocyst, and COC.

Tajima *et al*.[25] in his article presented a case of a 35-year-old male patient with well demarcated cystic lesion in mandibular symphysis region. Radiographically, radiolucency extended between mandibular premolar regions bilaterally with root resorption of the anterior teeth. Lesion was surgically removed, and histopathologic report gave ameloblastoma ex COC as the final diagnosis. There was no recurrence after a follow-up of 5 years.

Aithal *et al*.[6] reported a case in a 28-year-old female with a hard, nontender swelling in the left posterior region of mandible. Radiograph showed a multilocular radiolucency from canine to second molar. Microscopic examination revealed ghost cells in cystic epithelium and juxtaepithelial hyalinization. Odontogenic epithelium formed rosettes and acanthomatous ameloblastic islands were also seen in the connective tissue lining of the cyst. It was surgically excised and there was no recurrence at 2 years follow-up. Final diagnosis of ameloblastomatous COC was made.

Iida *et al*.[7] presented a case in a 17-year-old male with a tender bony swelling in the right mandibular body. Radiographs revealed a well-defined multilocular lesion in the right side of mandible in the ramus region with buccolingual expansion with displacement of lower second molar below the first molar. The tumor was treated with a more conservative approach of enucleation followed by ostectomy with no recurrence even after 13 years. Final diagnosis of ameloblastomatous COC was given.

Iida *et al*.[26] again concluded that the impaction and displacement of teeth in cases affecting the posterior jaw segment can be indicative of the development of COC. The presence of impaction or displacement of teeth for COCs occurring in the posterior segments of the jaw and absence of this finding for anteriorly placed lesions may be indicative of the timing of COC development.

Kamboj and Juneja[8] reported a case in a 58-year-old female with a painful swelling in right side of mandible. Radiographically, multilocular lesion from canine to posterior ramal region with buccolingual expansion was evident. Hemimandibulectomy was performed in this case.

Ledesma-Montes *et al*.[9] reviewed the clinical-pathological features of 122 CCOT, DGCT and GCOC cases from 14 institutions in seven countries of three different continents and concluded that of all the cases, Ameloblastomatous proliferating type of CCOT accounted for only three cases (2.46%) indicating the lesion to be very rare. He described the lesion having intraluminal or capsular plexiform growths similar to those seen in the plexiform variant of cystic Ameloblastoma. The rest of the details regarding the three cases is mentioned in Table 3.

Nosrati and Seyedmajidi[10] operated a 22-year-old male patient having tender swelling in mandibular right molar region. Panoramic radiography revealed a well-defined unilocular

Table 3: Review of cases by Ledesma-Montes *et al*.[9]

Reference	Age and sex	Presentation	Bone involved	Size of lesion	Root resorption	Cortical plate	Radiology	Recurrence	Histologic diagnosis	Impacted teeth
Ledesma-Montes *et al*.[9]	50 years; male	DNF	Mandibular posterior region	2.1 cm	Absent	Buccal plate expanded	Unilocular, radiolucent	Absent	Ameloblastomatous COC	Absent
Ledesma-Montes *et al*.[9]	55 years, female	DNF	Mandibular posterior region	6 cm	Absent	Buccolingual expansion	Well-defined, unilocular, radiolucent	Absent	Ameloblastomatous COC	Absent
Ledesma-Montes *et al*.[9]	15 years, female	DNF	DNF	DNF	Absent	DNF	DNF	Absent	Ameloblastomatous COC	Present

COC=Calcifying odontogenic cyst; DNF=Data not found

radiolucent lesion extending from mandibular right first molar region to third molar region. Surgical enucleation of the lesion was done along with removal of first and second molar. The concerned oral and maxillofacial pathologist diagnosed it as ameloblastomatous COC.

Sonone *et al*.[27] presented a case of a 23-year-old female patient who had a bony hard swelling on right mid-facial region in relation to maxillary right central incisor to first premolar region for which enucleation of the lesion was carried out and the histopathologic report revealed it as CGCOC.

Gupta and Gupta[11] reported a case in a 65-year-old male with a tender swelling in left mandibular region. Radiograph showed a well-defined bilocular lesion extending from mandibular left canine to third molar region. The treatment constituted of surgical excision with normal margins. A diagnosis of multicystic ameloblastomatous COC was made in the histopathologic report.

Singh *et al*.,[12] reported a case of 24-year-old female patient with a chief complaint of swelling in mandibular left anterior region since 6 months in canine-premolar region. Radiographic examination showed solitary well-defined radiolucency with sclerotic margins extending from mandibular right premolar region to the left third molar region. The lesion was enucleated and a definitive diagnosis of ameloblastomatous CGCOC given. Follow-up was done for 2 years with no signs of recurrence.

Sidana *et al*.[28] reported a case in 2013 of a 23-year-old male patient with painless swelling in mandibular left molar region since a month which gradually increased with time. Radiographically, a well-defined radiolucent lesion extending from left mandibular second premolar to ramus of the mandible anteroposteriorly was seen. Resorption of distal root of left mandibular first molar and both the roots of left mandibular second molar were seen along with impaction of mandibular left third molar. Histological diagnosis of CCOT was made. Lesion was enucleated. Lower left second premolar, first and second molars, and impacted third molar were extracted. No recurrence was noted in the further follow-ups.

Desai *et al*.[29] in his article presented a case of a 5-year-old boy with a painless, slow-growing swelling in mandibular left posterior region of the face. Radiographic examination revealed 3 cm × 2 cm ill-defined unilocular radiolucency around the crown of a developing permanent left second molar. Enucleation of the lesion was carried out with extraction of developing crown of permanent mandibular left second molar and the permanent mandibular left first molar. No recurrence noted in the follow-up period and the histopathologic diagnosis was CCOT.

Tamanna *et al*.[13] reported a case of a 21-year-old male patient with a tender swelling in the mandibular left back tooth region. Radiographically, unilocular radiolucency in the left ramus of mandible involving impacted mandibular left third molar along with resorption of mandibular left second molar. Marsupialization was done and the histopathologic features were suggestive of COC. The patient came with a recurrence a year later. This time microscopic examination revealed well-defined cystic lining with ghost cells and calcifications and diagnosis of COC with ameloblastomatous proliferation was made. The lesion was surgically removed and thereafter, no recurrence was noted in a year of follow-up.

In our case, wide surgical excision of the lesion with normal margins followed by chemical cauterization of Carnoy's solution was done. There has been no recurrence during these 6 years of follow-up period.

Among these, ameloblastomas may be most important in terms of its histopathology and management. So far, very few cases have been documented on ameloblastomatous COC. However, there have been a considerable number of reports of ameloblastoma arising from a variety of odontogenic cysts.[30] Therefore, it is not surprising that the cyst-lining epithelium of COC can be transformed into a true ameloblastoma that may fulfil the criteria of Vickers and Gorlin.[24] Whether to have a conservative or a radical approach for management of this lesion has been an issue of debate.

Several authors have suggested that if COC is associated with ameloblastoma, its behavior and progression is not of COC and hence should be treated more aggressively. Our case did not show any recurrence after its thorough excision with normal margins.

Conclusion

Ameloblastomatous COC, a cystic variant of COC occurs in the mandible with a higher propensity toward the posterior region. The lesion is intraosseous and generally presents as a tender swelling clinically with a radiographic appearance of unilocular or multilocular lesion. However, in our case, though the lesion involved an extensive posterior mandibular region, it was asymptomatic. As conservative treatment in the form of enucleation has proven to be effective, it needs to be differentiated from ameloblastoma ex COC since it requires more aggressive form of treatment.

Acknowledgment

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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