CALCIFYING ODONTOGENIC CYST: AN ANALYSIS OF THIRTY-SIX CASES

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Abstract- The term calcifying odontogenic cyst (COC) was first introduced by Gorlin in 1962. COC occurs mainly as an intra-osseous lesion in mandible or maxilla but the peripheral variation of COC has also been reported. The confusion about COC nature as cyst or tumor has not been resolved and a vast diversity has been shown in clinicopathologic aspects of COC. The purpose of this study was the review and analysis of COC variables such as age, gender and location and reclassification of histopathologic features of cases. The study was a retrospective review, cross sectional and case series one. The records of 36 patients were obtained and the clinical characteristics such as age, gender and location of COC were registered. The histopathologic features of cases were reviewed and reclassified. The data showed that 38.3% of cases occurred in the fifth decade and beyond; 67.6% were in males and 32.4% in females. Considering location, 51.5% of COCs were found in mandible and 48.5% in maxilla; 65.6% were located in posterior part of jaws. Of the 36 cases diagnosed as COC, 32 (88.8%) had cystic features and 4 (11.11%) were tumoral. For better determination of histopathologic and biologic features of COC, more investigations are needed.

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Key words: Calcifying odontogenic cyst; developmental odontogenic cysts; odontogenic tumors

INTRODUCTION

The odontogenic cysts comprise extensive part of jaw lesions. These cysts are true cysts with a well-delineated epithelial lining. The epithelial lining originates from odontogenic epithelium. The odontogenic cysts are divided into two main types: inflammatory and developmental (1).

The term calcifying odontogenic cyst (COC) was first introduced by Gorlin and his colleagues in 1962. The COC is a developmental odontogenic cyst with diverse origin as either a cyst or a neoplasm.

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The World Health Organization (WHO) has classified COC as a neoplasm (1). Because of this diversity a "dualistic" concept is proposed. This concept regards COC as a cyst and neoplasm (2).

The histologic variation of COC has led to different terminologies such as calcifying ghost cell odontogenic tumor (Fejerskov and Krogh, 1972), dentinogenic ghost cell tumor (Preatorius *et al.*, 1981), epithelial odontogenic ghost cell tumor (Ellis and shmookler, 1986), and Odontogenic ghost cell tumor (Colmenero *et al.*, 1990) (1-3). At first, this cyst was classified as a) simple, b) associated with odontoma, and c) associated with odontogenic tumors (other than odontoma) (2). Because of the diversity of histopathologic features of COC, Praetorius *et al.* proposed a new classification in 1981. In this classification COC was divided into two types. Type 1 or cystic type includes a) simple

unicystic type, b) odontoma producing type, and c) ameloblastomatous proliferating type. Type 2 or neoplastic type includes dentinogenic ghost cell tumor (2, 3). In order to eliminate the problems of previous classifications, Toida proposed a simple and basic classification based on "dualistic" concept (Table 1) (2).

COC mainly occurs as an intra-osseous lesion but the peripheral (extra-osseous) lesion has also been reported. The extra-osseous COC is a localized sessile or pedunculated gingival mass. This type has not distinctive clinical features (1). The intra-osseous COC usually presents as a uni or multilocular radiolucency with irregular or tooth-like densities. Sometimes the root resorption or divergence of adjacent teeth is reported (1). The mean age of patients has been reported as 33 years and ranged from infancy to elderly. Other factors such as gender and location seem to have equal frequency (1, 4).

Regarding the diversity of clinicopathologic features of COC and rarity of information, we attempted to review and analyze the COC cases in our department. The purpose of this study was to analyze the variables such as age, gender and location of COC cases and re-classification of cases.

MATERIALS AND METHODS

The study was a retrospective, cross-sectional and case-series one. The source of materials was the histopathologic records of patients in Oral and Maxillofacial Pathology Department of Dental Faculty of Tehran university of Medical sciences from 1967 to 2004. All specimens diagnosed as COC or Gorlin cyst were retrieved from archive and evaluated.

Table 1. The new dualistic classification of so-called COC by Toida

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, malignant CGCOT) associated with the following lesions:
      α. odontoma
      β. ameloblastoma
      γ. Other odontogenic lesions

The clinical informations such as age, gender and location of COC lesions were registered on data forms. The locations of lesions were classified into two anatomic regions: posterior and anterior. The patients' ages were distributed by one decade. In the next step, the histopathologic sections of COC specimens were reviewed by two oral pathologists (authors) and re-classified.

The obtained data were analyzed with SPSS software.

RESULTS

From 1967 to 2004, a total of 36 cases were registered. Incidence was 23.5% in first and second decades, 23.5% in second and third decades, 14.7% in third to fifth decades and 38.3% in fifth decade and beyond. The highest incidence was in the fifth decade; 67.6% of patients were males and 32.4% were females.

Considering location, 51.5% were located in mandible and 48.5% in maxilla. The most common locations of lesions were in posterior regions of both jaws. In the maxilla, 35.7% of cases were in the anterior and 64.3% in the posterior regions. In the mandible, 35.3% of lesions were registered in the anterior and 64.7% in the posterior regions.

Of 36 cases diagnosed as COC, 32 (88.8%) had cystic and 4 (11.11%) tumoral (solid) features.

The cystic group had epithelial lining with variable thickness. The basal layer was cuboidal and had one layer thickness. Above this part, the cells similar to stallate reticulum of dental organ were prominent. The isolated or groups of ghost cells were seen between the cells of these two layers. Mineralized particle and dentinoid formation were also noticed in some parts of COC.

The cystic variation had three features: a) luminal type in which proliferation of described cells was limited to luminal surface of cyst, b) intraluminal type: the multiple proliferation of cystic (odontogenic) epithelium was seen into lumen, c) hybrid type in which the above changes were seen in fibrous connective tissue and epithelial lining of cyst. Four of 32 COC were associated with complex odontoma and 3 cases with ameloblastom-like proliferation.

The tumoral (or solid) type showed a compact proliferation of strands or islands of odontogenic epithelium resembling ameloblastoma like proliferation (with peripheral palisading cuboidal cells and stellate reticulum-like cells) and scattered ghost cells in stroma. No cystic changes were seen.

DISCUSSION

In 1971, COC was described as a non-neoplastic cystic lesion, but in 1992 the WHO classified this lesion with odontogenic tumors (2). Many investigators had tried to resolve this confusion. By employing clinical and histopathological features, Buchner *et al.* reviewed 17 cases of central COC and analyzed their clinical and histomorphologic features. The data revealed that central COCs were usually diagnosed in the second decade of life with an equal distribution between maxilla and mandible (4). According to Shamaskin *et al.*, central COCs occur more commonly than peripheral lesions by a 3:1 ratio and they are usually diagnosed in the second decade of life, while the peripheral ones are usually noted after 50 (5).

Buchner *et al.* reviewed 45 cases of peripheral COC and found that most of the lesions were located in the maxillary and mandibular gingival or alveolar mucosa anterior to the region of the first molar (6). In another study, Buchner analyzed the clinical, radiographic and histomorphologic features of the 215 cases of central COC reported in the literature. He reported that of 215 cases, 51.6% were in the maxilla and 48.4% in mandible; 65% of the lesions were located in the incisor-canine area of maxilla and mandible. Of 215 cases, 51.2% were females and 48.8% were males. The mean age of patients was 30.3 years. The highest incidence was in the second (40%) and third (18%) decades (7).

Based on analyzing 21 cases of COC, Li and Yu reported that the age of 16 cases of cystic COC peaked at the second decade. The maxilla was affected (69%) more than mandible. The 4 cases in the tumoral group occurred in the mandible (8). Nagao *et al.* has reported that in a survey of 23 cases of COC in Japanese literature, the involvement of male and female was equal. The mean age of patient's was 21. The maxillary lesions were three folds more than mandible (9).

In present study, COCs were more frequent in men (67.6%), in mandible (51.5%), and in posterior region of both jaws (65.6%). The highest incidence was seen in fifth decade (38.3%).

Without considering some similarities, there were considerable differences between the previous studies and present one. Based on our results, occurrence of COC was higher in men, mandible and fifth decade. On the other hand, other studies have reported more cases in females, maxilla and second decade. With consideration of sampling method, we attributed these differences to the numbers of cases, the period of investigation, entering different classifications and consideration or elimination of peripheral and central subtypes of COC.

Since the first introduction of COC by Gorlin *et al.* in 1962, there has been diversity and confusion in the relationship between cystic and solid subtypes of COC. These subtypes had a similar cellular and histomorphologic characteristics. For overcoming this problem, different histopathologic classifications have been proposed by authors. In the second step, we reviewed the COC pathologic sections. Of the 36 COC, 32 (88.8%) had cystic and 4 (11.11%) solid features. In cystic variant, three morphologic changes were seen. These changes included luminal, intraluminal and hybrid. The solid type composed of strands and islands of ameloblastomatous proliferation of odontogenic epithelium and scattered ghost cells. Hong *et al.* reviewed histopathologic features of 92 cases of COC. The cases were divided into 79 (85.9%) cysts and 13 (14.1%) neoplasms. The cysts had four variants: 1) non proliferative (35 cases), 2) proliferative (17 cases), 3) ameloblastomatous (11 cases), and 4) COC associated with odontoma (16 cases). The neoplasms occurred as three variants: 1) ameloblastoma ex COC (two cases), 2) peripheral epithelial odontogenic ghost cell tumor (8 cases) and 3) central epithelial odontogenic ghost cell tumor (3 cases) (10). Praetorious *et al.* studied 16 cases of COC. On the basis of their findings, this series contained two entities, a cyst and a neoplasm. The cyst occurs as three variants: 1) a simple unilocular cyst with mural

proliferations and no or sparse amounts of dentinoid, 2) a unilocular cyst with compound or complex odontoma, and 3) a unilocular cyst with luminal and mural ameloblastoma-like proliferation. The neoplastic variants consist of ameloblastoma like strands and islands of odontogenic epithelium and varying amounts of ghost cells (3).

By studying the 21 intraosseous COCs, Li and Yu suggested that the term COC should be used for unicystic lesions with or without an associated odontoma and other related lesions identified as benign or malignant tumor should be termed separately (8).

For characterizing the histologically and immunohistochemically of COC, Yoshida *et al.* studied 16 cases of COC with cytokeratin 19 and bcl-2. They concluded that COCS with various histological features have neoplastic potential (11).

In present study, two variants of COC were observed. One of these variants was cystic (luminal, intraluminal and hybrid) and the other one had solid pattern. In agreement with described studies, it seems that the so-called COC has two features of cyst and neoplasm. In spite of several dualistic classifications of COC, confusion about terminology and nature of solid (neoplastic) variant still exists. In summary, this study showed that COC is more frequently occurs in males, in mandible and posterior anatomic region of both jaws.

The highest incidence was seen in the fifth decade.

We suggest that for better determination of histopathologic and biologic nature of COC, more investigations based on histomorphological and immunohistochemical studies are needed.

Conflicts of Interests

We have no conflicts of interest.

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