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ORIGINAL RESEARCH

Thirty Years Clinicopathological Study of 60 Calcifying Cystic Odontogenic Tumors in Iranian Population

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ABSTRACT

Aim: To reevaluate all cases previously diagnosed as calcifying odontogenic cyst (COC) from the archives of the Department of Oral Pathology at Mashhad Faculty of Dentistry from 1978 to 2008.

Materials and methods: Clinical histories and microscopic slides of all cases diagnosed as COC were reviewed and analyzed statistically. The information about patients such as age, sex, clinical features, radiographic view and other necessary data was gathered from biopsy files.

Results: The majority of cases occurred in mandible and four cases were peripherally. Thirteen cases were associated with other odontogenic tumors and 11 cases were odontoma type. In two cases, recurrence was reported after surgical treatment which underwent malignant transformation.

Conclusion: Calcifying cystic odontogenic tumor (CCOT) in Iranian population occurs in younger age group with slightly male predilection and more mandibular, unicystic and simple type.

Clinical significance: It is better to address the noncystic CCOT lesions with their specific terms in order to make histopathological characteristics clear.

Keywords: Odontogenic cyst, Odontogenic tumor, Longitudinal study, Calcifying cystic odontogenic tumor.

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INTRODUCTION

Any improvement in the field of oral and maxillofacial pathology helps in better diagnosis of disease and successful treatments. An important part of this field is odontogenic lesions.[1] Since these lesions appear only in oral cavity, diagnosing them is of high importance for dentists. Among these, calcifying odontogenic cyst (COC) was first described by Gorlin et al[2] with these characteristics: Varieties in histological characteristics and its similarities with other odontogenic lesions have made the diagnosis difficult.

Nonspecific clinical and radiographic aspects of this lesion have caused its ignorance in oral differential diagnosis.[2,3] This lesion presents with some characteristics of cyst and some symptoms of solid neoplasm; and so, WHO classification of odontogenic tumors renamed it as calcifying cystic odontogenic tumor (CCOT) in 2005.[4,5]

The aim of this study was to evaluate the epidemiological and histopathological aspects of CCOT and its clinicopathological characteristics.

MATERIALS AND METHODS

In this retrospective study, all CCOT cases at Department of Oral and Maxillofacial Pathology at Mashhad Faculty of Dentistry with diagnosis of calcifying odontogenic cysts for the last 30 years were detected. This research was exempted by the university review board.

The microscopic sections related to all cases of CCOT were considered again and pathologic diagnosis of them was confirmed blindly by two oral pathologists. All of the sections had been prepared by H&E staining. The information about patients, such as age, sex, clinical features, radiographic view, and other necessary data was gathered from biopsy files and filled in forms. The data were analyzed by using SPSS v.11 and nonparametric chi-square test.

RESULTS

Among 7707 patients, about 1403 odontogenic lesions, 195 odontogenic tumors, 1208 odontogenic cysts and 60 CCOT with the age range of 6 to 80 (mean 23) were detected.

According to later classification, prevalence rates for odontogenic lesions was 18% (1403/7707 cases), for CCOT among all of the lesions was 0.8% (60/7707 cases), for CCOT among odontogenic lesions was 4.3% (60/1403) and for CCOT among odontogenic cysts was 4.6% (60/1208).

CCOT was the most common in the second decade of life in both sexes. The prevalence of CCOT decreases with age.

Although the average age of affected men was 5 years more than affected women, there was not any significant difference between the average ages of these two groups. In 60 cases with CCOT, 37 cases were male and 23 female ($p = 0.07$). The most common sign observed was swelling (41 cases) and also eight cases were accompanied with pain. Other cases were diagnosed accidentally through radiography.

The most common radiographic view was unilocular radiolucency (reported in 35 cases) among which only three cases were with multilocular radiolucent view. Another radiographic view was the mixed type (radiolucent/radiopaque) observed in 11 cases, especially in those with odontoma.

In 15 cases, the lesion was accompanied with impacted tooth and five cases (all in mandible) had radiographic view of external and internal cortexes destruction. The roots divergence of adjacent teeth was seen in four cases.

In one of the peripheral lesions of CCOT, depression of bone was seen in radiography. In other cases because of unavailable radiographic view or report of the patient, the radiographic view of lesion could not be studied.

77.5% of studied CCOT lesions occurred in mandible, and there was a significant relationship between CCOT and mandibular location ($p < 0.001$).

Most of lesions were extensive and included more than one dental region. Because of unavailable radiographic view of some patients and lack of exact explanation of lesion boundaries, more accurate anatomical investigation was impossible. About 94% of lesions were central but peripheral type is more prevalent in women.

In the 60 COC cases studied, 36 cases were simple cystic type, 11 cases were odontoma producing type, eight were ameloblastomatous proliferating type and 5 were neoplastic type. There was only one malignant case after the recurrence of a benign CCOT lesion.

There were two recurrent cases. The first one was a patient referred for the first time at the age of 15 with a painless swelling in the mental region. In the radiographic view, a radiolucent lesion extended from the left mandibular central incisor to the second molar of the same side and expansions of both external and internal cortexes were observed.

Biopsy was performed and the patient was operated regarding the pathologic diagnosis of 'COC type IA'. Three years later, through the control radiography of this patient, an extensive radiolucent lesion from the mandibular canine to the left second molar was detected. The patient was reoperated and the reported diagnosis of simple type COC was confirmed. Six years later at the age of 24, the patient came back with a radiolucent lesion and a painless swelling at the previous site. After biopsy for the 3rd time, the pathologic diagnosis of this lesion was reported to be 'COC IC' with ameloblastomatous proliferating type and then it was treated as an ameloblastoma without recurrence for more than 10 years.

The second case with recurrence was a 9-year-old boy referred with a radiolucency at the site of right mandibular second molar (the second molar was embedded) for the first time. According to the clinical diagnosis of ameloblastoma, biopsy was performed. The pathologic diagnosis of the lesion was ghost cell odontogenic tumor or COC type II. The lesion was resected and a bone graft was applied. Five years later, the lesion recurred with signs of malignancy and malignant epithelial ghost cell odontogenic tumor was diagnosed.

Among our four peripheral cases, two were simple unicystic and the others were odontoma producing type.

DISCUSSION

Since the time that COC was described by Gorlin for the first time, research attempts toward it showed up,[2,4] but there is a few completely studies for compression about this lesion unfortunately.

Over the past 50 years, our knowledge about CCOT including the classification and behavior has been evolving. With increase in time and other COC cases, researchers concluded that COC has heterogeneous property. This lesion has described several times from 1971 by WHO.[5-8] Praetorius et al[8] revised classification in 1992 by WHO again and in 2005 the WHO published their latest classification scheme and accepted it as a tumor. This study confirms some of the findings in other studies, although it is in contrast with many others.

CCOT happens more at the second decade of life and it was approved in the present study as 45% of the cases were at this age group.[9] But more mandibular and men involvement are in contrast to another studies.[7,9,10] Nevertheless, there was no meaningful statistically relationship between sex and either age or clinical manifestation, except anatomical position in our study. Although the prevalence of the CCOT among odontogenic lesions was 4.3% in our study, it described as rare to less common lesion (Table 1).[11-17]

Likewise other studies indicating that CCOT may be asymptomatic and may be diagnosed accidentally,[18] 11 cases in our study were asymptomatic and diagnosed through routine radiography.

The radiographic feature of CCOT is well-defined unilocular radiolucency with distinct boundaries which was confirmed in our study, and scattered radiopacities may be seen that it was observed in 11 cases in our study.[19]

Although most studies declared no sex predilection and equal distribution of CCOT in both jaws,[9,10] but we found it more in males and mandibular involvement.

Researchers believe that central lesions are more frequent and 15 to 21% of the CCOT cases are peripheral,

Table 1: Prevalence rate of CCOT in odontogenic tumors

Author/ year	*Country*	*CCOT prevalence*
Mosadomi et al (1975)[11]	Africa (Cameroonian, Nigerian)	3.4%
Mosqueda-Taylor et al (1997)[12]	Mexico	6.8%
Ladeinde et al (2005)[13]	Nigeria	5.3%
Fernandes et al (2005)[14]	Brazil	3.5%
Adebayo et al (2005)[15]	Nigeria	2.5%
Buchner et al (2006)[16]	USA	1.6%
Saghravanian et al (2010)[17]	Iran	0%
Present work	Iran	4.3%

although in our study, 6.6% of the cases were peripheral.[9,19] Follow-up of the surgical treatment after enucleation shows different recurrence rate [20] but in this study it was 3%.

CONCLUSION

CCOT is a lesion with heterogeneous clinicopathologic characteristics and in the typical histological properties COC is the best nomination for that.

CLINICAL SIGNIFICANCE

It is better to address the noncystic CCOT lesions with their specific terms in order to make histopathological characteristics clear.

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