Calcifying Epithelial Odontogenic Cyst Associated with Complex Odontome of Maxilla

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Abstract The term calcifying odontogenic cyst was first introduced by Gorlin in 1962 (Eshghyar et al., *Acta Med Iran* 44(1):59–62, 2006). The lesion is unusual in that it has some features of a cyst but also has many characteristics of a solid neoplasm (Cysts and tumors of odontogenic origin: textbook of oral pathology, 2006). It is classified into two types—Type I—the cystic variant, Type II—solid tumor variant. This case report present Type I B—odontome producing intraosseous calcifying odontogenic cyst and Type I C—with ameloblastomatous proliferation in a 19 years old male patient in the right maxillary quadrant. The lesion involved an unerupted permanent maxillary central incisor, which was displaced to the infraorbital ridge of right side and the radiograph revealed a calcified mass in the periapical region of right incisor that was later recognized histopathologically as complex composite odontoma. The lesion was removed surgically. This case report emphasizes on the presence of this Type I B and C lesion and the need to keep them in follow up.

Keywords Odontome · Neoplasm · Incisor · Cytoplasm

Introduction

Calcifying odontogenic cyst an uncommon benign odontogenic lesion was first categorized as a distinct entity by Gorlin et al. in 1962 [1] and was named after him since then [2]. 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 [3]. In 1992, WHO classified COC as a neoplasm rather than a cyst, but confirmed that most of the cases are non-neoplastic [4]. In view of this duality, many different terminologies have been applied to cystic and solid COC variants, but COC is still the preferred term [5].

It represent 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 [6]. 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 [7]. 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 [8].

Case Report

A 19 year old boy complained of painless swelling in the right bicuspid and molar region of maxilla since 6 months. Extraorally the lesion did not produce any obvious swelling

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Fig. 1 Intraoral picture showing expansion of buccal cortex (2) and odontome (1)

Fig. 2 Intraoral picture showing the odontome exposed

and can be felt only on palpation. Intraoral examination revealed a firm, uniform enlargement of the buccal cortex of right maxillary quadrant extending from the central incisor up to the second molar of maxilla Fig. 1. The overlying mucosa was normal. The deciduous right central incisor was still present and the permanent central incisor was missing. The detailed intraoral examination revealed no mobility or tenderness to palpation with any tooth. There was also no sign of caries, pulp pathosis or periodontitis. Patient complained of heaviness and numbness in right infraorbital region since 2 months.

However, in the central incisor region on right side a firm, stony hard and irregular mass can be palpated Fig. 2.

Fig. 3 CT scan reveals the boundaries of the cyst with impacted Central incisor (1) and radio-opaque mass, i.e. the odontome (2)

Fig. 4 Coronal section in CT scans showing the superior, inferior, anterior and posterior boundaries of the cyst

A CT scan and panoramic views revealed a well defined unilocular radioluscent lesion involving the entire right side of maxilla circumscribing the entire quadrant and reaching up to the infraorbital ridge of right side, anteriorly the lesion extended up to the lateral incisor and posteriorly up to the pterygoid plates Figs. 3 and 4. A radio-opaque irregular mass seen present in the periapical region of deciduous central incisor reaching up to the level of floor of the nose. An impacted central incisor was visible in the right infraorbital region. Displacement of lateral incisor was seen. Root resorption was not present with any teeth. Radiological differential diagnosis included adenomatoid odontogenic tumor, intraosseous COC, cystic odontome, ossifying fibroma, ameloblastic fibro-odontome and

Fig. 5 Picture showing the cyst lining, odontome and impacted teeth after removal

calcifying epithelial odontogenic tumor. CEOT is most commonly found in the posterior region of the mandible, and the margin of this lesion are often scalloped. Ameloblastic fibro-odontome is usually encountered in children of 10 years of age, on average. Although ossifying fibroma has many features in common with case but is more often seen in females.

The teeth involved in the lesion received endodontic treatment before the surgical excision of the lesion and during surgery the impacted permanent central incisor was also removed. The lesion was removed conservatively by first removing the odontome which was enlarged and the enucleation of the cyst lining was done. Enucleation was done from anterior region from where the odontome was removed and the expanded buccal cortical plate was compressed and preserved Fig. 5. Healing occurred uneventfully Fig. 6.

Histopathological Report

Microscopically, haematoxylin and eosinophilic stained soft tissue section shows lesional tissue composed of cystic lumen lined with thin odontogenic epithelium which is 2–3 cell layer thick and parakeratinized at some places. The basal cell layer is prominent. The cells in the superficial layer are tall columnar ameloblast like cells with round to oval hyperchromatic nuclei showing palisading and reversal of polarity. Interspersed within this epithelium are numerous ghost cells showing eosinophilic cytoplasm and blurred cellular outline and nuclei in various stages of degeneration Fig. 7. Broader areas of epithelium shows stellate reticulum like cells. The surrounding connective tissue capsule is

Fig. 6 OPG taken 3 months post-operatively showing acceptable bone formation

Fig. 7 Histopathological section showing the cyst lining (1) and the Ghost cell (2) (Haematoxylin and eosin stain at 10×)

composed of loosely arranged collagen fiber bundle, fibroblasts and blood vessel. Interspersed within these collagen fiber bundles is dentinoid like material at few places. Several inactive odontogenic islands are also evident few of them show tall columnar ameloblast like cells with round to oval nuclei showing hyperchromatism, palisading and reversal of polarity Fig. 8. At few places, areas of calcification are also evident.

H&E stained decalcified section shows mature irregular tubular dentin cut in longitudinal and cross sections in moderately cellular connective tissue stroma. This dentin shows clefts or enamel spaces. Pulp like connective tissue is evident at some places Fig. 9. The connective tissue stroma

Fig. 8 Histopathological section showing the ameloblastic proliferation (2) and the Ghost cell (1) (Haematoxylin and eosin stain at 10×)

Fig. 9 Histopathological section of odontome showing structures similar to stained section of tooth (Haematoxylin and eosin stain at 10×)

consists of thin endothelial lined blood vessels with RBCs within it. Extravasated RBCs are also evident at some places.

Discussion

CEOC first described by Gorlin in 1962, was later in 1971 described by WHO as a non-neoplastic cystic lesion in which the epithelial 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

epithelial cells that may be in the epithelial cyst lining or in the fibrous capsule [5].

As reports in literature increased, CEOC was classified as heterogenous group of entities with distinct histopathological finding that included solid tumor.

Preatorius proposed a subclassification for the heterogenous group of lesion, in which the cystic lesion were separated from the neoplasm (solid lesion), and divided into three classes [9].

Type I—Cystic lesion

- A. Unicystic
- B. Odontome producing
- C. Ameloblastomous proliferating type

Type II—Solid neoplasm

The new dualistic classification of so-called COC by Toida [1]:

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 radio-opaque mass observed in the panoramic radiograph of this case, later diagnosed as complex composite odontome are considered to be present in 24–50% of reported cases. These cases were classified as a subtype of COC under the term Odontoma-producing type or as a cystic variant associated with odontoma.

Hirschberg et al. [10] proposed that this variant should be classified as a separate entity for which they suggested the name Odontocalcifying odontogenic cyst. They observed that this variant was more prevalent in females, with a mean age at discovery of 16 years and that most of the cases were found in maxilla.

In our case the lesion was found in maxilla in a male patient of age 19 years.

Hirschberg et al. [10] claimed that separation of this variant from the heterogenous group of COC could lead to the better understanding of its pathogenesis.

Although Gorlin and his coworker suggested the calcifying odontogenic cyst as the oral counterpart of the

cutaneous calcifying epithelioma of Malherbe (pilomatrixoma), it is more likely to be the oral counterpart of the intracranial craniopharyngioma due to immunologic similarity [2].

In this regard, our case may indicate that this variant may be more common in young adult, but it may also be discovered in older person so that a different pathogenesis might not be associated with this variant.

In spite of the low frequency of this lesion, and the fact that most cases are surgically removed and heal uneventfully, however, in this case with ameloblastomatous proliferation a close follow up is needed and also there have been reports of association with carcinoma, adenomatoid odontogenic tumor and ameloblastoma [11].

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