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Huge calcifying epithelial odontogenic tumor of the mandible and management with a teeth preserving surgical approach: a case report

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Abstract

Background Calcifying epithelial odontogenic tumor is a rare benign tumor that predominantly occurs in posterior sites of the mandible in adults.

Case presentation This case report describes a case of calcifying epithelial odontogenic tumor in a 37-year-old Iranian female with the chief complaint of swelling in the anterior region of the mandible and tooth displacement. This paper summarized the pathological findings, radiographical features and clinical management of the patient. Clinical examinations revealed a bony hard swelling ~ 3 cm × 4 cm in size with tenderness in the chin area. Radiography revealed an expansile mixed radiolucent and radiopaque lesion with cortical borders. A minimally aggressive surgical approach is used to remove the lesion.

Conclusion This case report outlines the successful outcomes of this surgical approach and the positive outcome associated with the restoration of lost functions of displaced mobile teeth in an arranged and nonmobile way after treatment.

Keywords Pindborg tumor, Calcifying epithelial odontogenic tumor, Odontogenic tumor

Introduction

Danish pathologist Jens Pindborg first described calcifying epithelial odontogenic tumors (CEOT)in 1955. CEOT is a benign epithelial odontogenic tumor without ectomesenchymal components. This tumor is a slow-growing and expansile neoplasm with locally aggressive and invasive growth [1].

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CEOTs present as intraosseous lesions in most clinical cases. Extraosseous variants also occur in less than 5% of cases [2]. The peripheral type exhibits painless swelling [3]. This tumor is seen more frequently in the mandible than in the maxilla [4]. In ~ 50% of cases, CEOT is associated with a tooth, erupted or unerupted [1].

Radiologically, central CEOT is a well-defined, unilocular or multilocular radiolucency. Calcified materials are usually found in intraosseous regions, such as scattered fleck calcifications [2]. A literature review of 339 cases revealed that ~ 40% of peripheral CEOTs result in underlying bone resorption, and 50% of central CEOTs result in perforated cortical bone [5].

The central types usually affect premolar and molar areas, whereas the peripheral types are usually located

in anterior regions. CEOT usually occurs equally in both genders. The patients affected by this tumor ranged in age from 8 to 92 years, with a mean age of 40 years. This tumor is found in different populations, but it has a slight predilection for white people [2].

Case presentation

In accordance with the case report (CARE) guidelines [6], this report describes a patient with calcifying epithelial odontogenic tumor by histological and radiographical examinations. This patient was cured by a tooth preserving surgical approach in the Department of Oral and Maxillofacial Surgery at Taleghani General Hospital.

Chief complaints

The patient, a 37-year-old Iranian female, was referred to the Department of Oral and Maxillofacial Surgery at Taleghani General Hospital with a chief complaint of tenderness in the anterior region of the mandible and tooth displacement in the swelling region.

Patient information

The patient's past medical history was unremarkable but patient mentioned a facial trauma about 20 years ago. The patient also had penicillin antibiotic allergy. Patient report a slow-growing mass for ~ 1 year. The familial history was unremarkable.

Clinical finding

Extraoral examinations revealed a bony hard swelling ~ 3 cm × 4 cm in size with tenderness in the chin area.

The patient reported lower lip hypoesthesia along the right and left mental nerve branches. The lymph nodes were normal with no tenderness or swelling.

Intraorally, there was an expansile lesion with bicortical growth and displacement of teeth from tooth 25 to tooth 22 (Fig. 1).

This swelling was ~ 3 cm × 4 cm in size on the buccal side and ~ 2 cm × 3 cm on the lingual side of the anterior mandible region. Tenderness and a hard bony consistency on palpation were also noted. The oral mucosa was normal and intact in color and texture. The mandibular central incisors and right mandibular lateral incisor were mobile in grade one, and the left mandibular lateral incisor was mobile in grade two.

The inferior border of the mandible was swollen and tender in the lesion area.

Fistula and pus discharge were not evident in intra- and extra-oral examinations.

The patient had no symptoms of dysphagia and dyspnea.

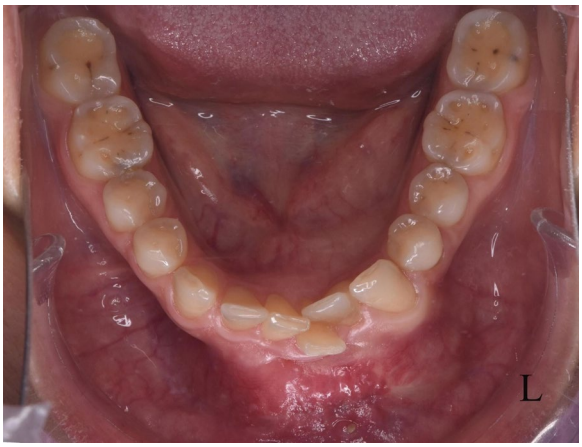

Fig. 1 Intra-oral view: bicortical swelling and displaced teeth

Vitality tests were performed by cold and electrical pulpal tests, and the results showed that the teeth adjacent to this lesion were vital.

Diagnosis assessment

Panoramic radiography revealed an expansile mixed radiolucent and radiopaque lesion with cortical borders in the anterior area of the mandible that extended from the right mandibular canine to the left mandibular second premolar teeth. This lytic lesion extended toward the inferior border of the mandible, but the border was intact. Radiopaque odontoma-like materials were observed in the central part of the lesion. Cone beam computed tomography (CBCT) revealed expansion and perforation of the buccal and lingual plates. This lesion measured 42.6 mm to 40 mm and extended from the right mandibular canine to the left mandibular second premolar. The apical root resorption of teeth was evident in teeth 26, 25, 24, 23, 22, and 21 (Fig. 2).

An incisional biopsy was performed under local anesthesia with 2% lidocaine and 1/100,000% epinephrine.

The specimens were fixed in 10% formalin. Microscopic examination of hematoxylin and eosin-stained samples confirmed that the tumor was a benign odontogenic tumor.

Sections showed pieces of benign odontogenic neoplasm composed of a few islands and strands of inactive odontogenic epithelium with juxta-epithelial hyalinization in fibrous connective tissue. Mild inflammatory cells and some hemorrhages were also present. Hard tissue sections showed normal lamellar bone adjacent to the fibrous connective tissue. Muscle fibers and nerve bundles were evident.

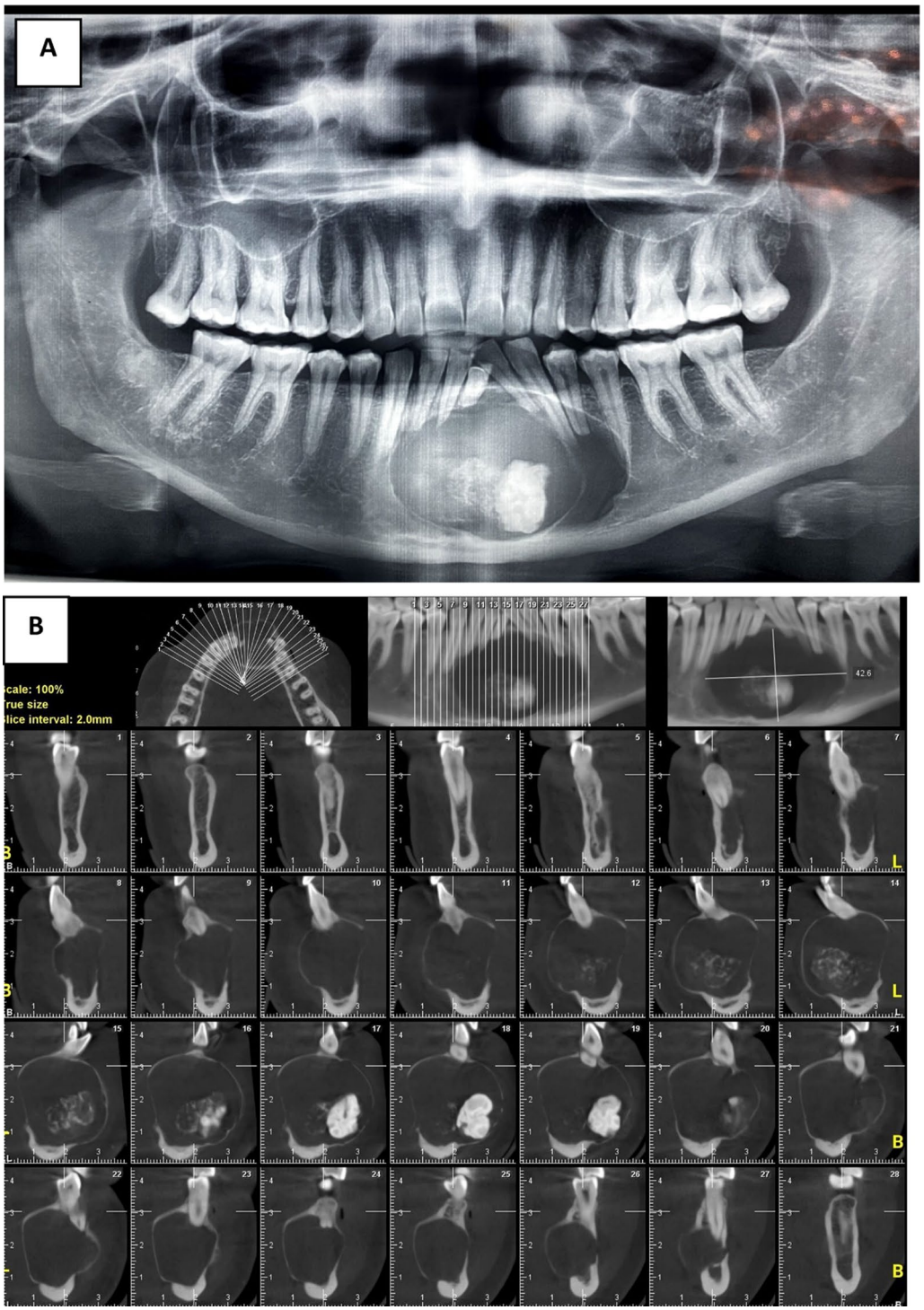

Fig. 2 **A**–**C** Panoramic and CBCT modalities showed an expansile mixed radiolucent and radiopaque lesion with cortically borders

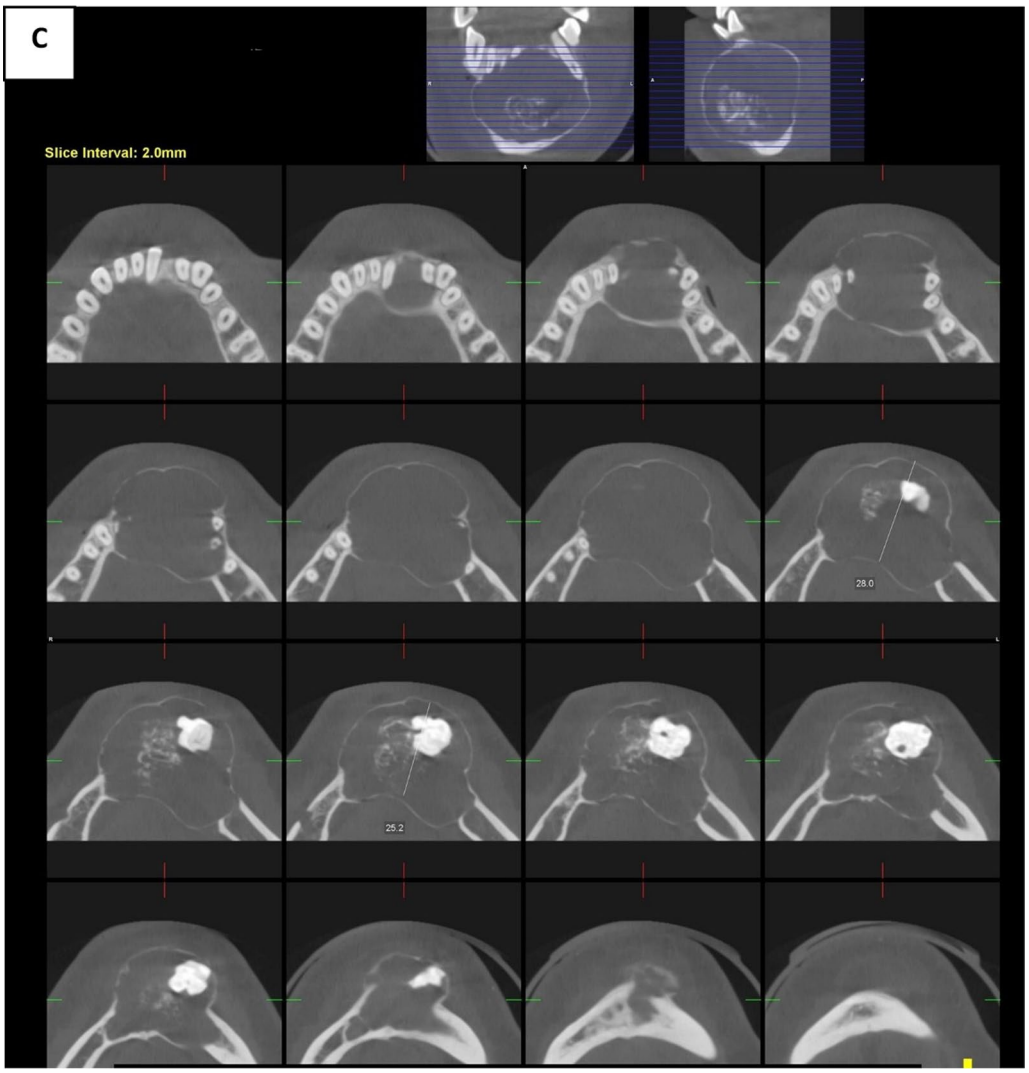

Fig. 2 continued

An aspiration of 1.5 cc of red fluid was conducted using an 18-gauge needle. The smear showed many red blood cells (RBCs) and scattered inflammatory cells in an eosinophilic background material.

Therapeutic intervention

The patient underwent surgery to remove the lesion under general anesthesia.

A sulcular incision with releasing incisions was made from the right first molar to the left first molar. After subperiosteal dissection, perforation of the buccal plate was present. To access the lesion, the remaining thin cortical bone was removed by a green stick fracture. The lesion was separated from the bone walls and ultimately completely removed (Fig. 3). Curettage of the bone adjacent to the tumor was conducted to reduce the risk of recurrence. The defect was simultaneously augmented with the remaining intact buccal cortex bone. The defect site was filled with Gelfoam (Pfizer company, USA) and covered with resorbable membrane (Fig. 4), and the incision lines were sutured watertight by vicryl 4–0.

The tumors were stored in 10% formalin for histopathological analysis.

Follow up and outcomes

The surgery was uneventful, and the wound healed without complications.

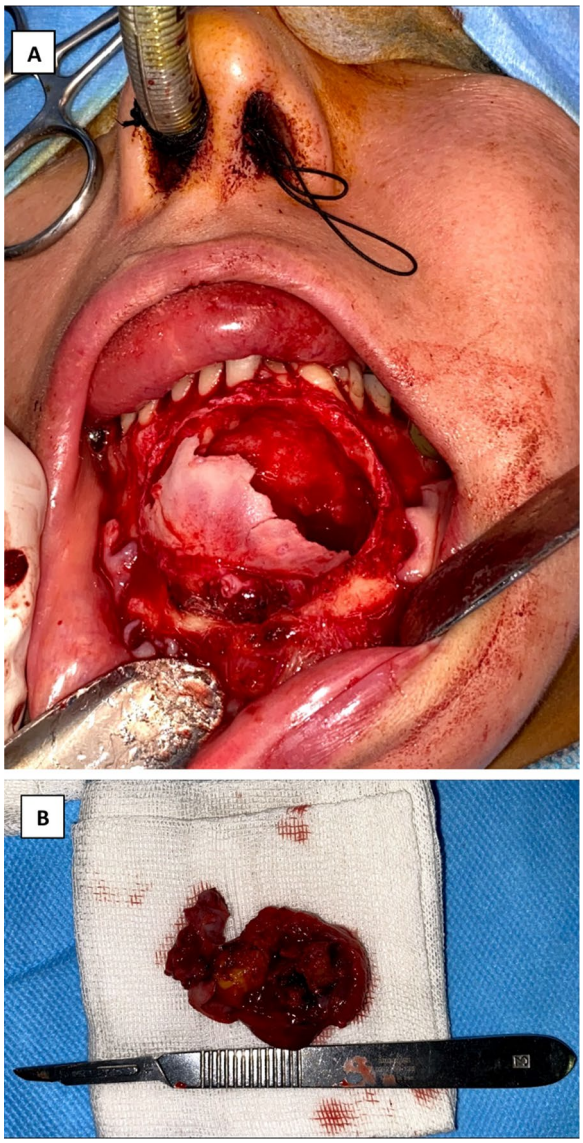

Fig. 3 **A** The perforated cortical bone is seen. **B** the lesion after complete excision

Microscopic examination revealed an odontogenic neoplasm of islands, strands, and sheets of polyhedral epithelial cells in a fibrous stroma. The cellular outlines of epithelial cells were distinct. Multiple concentric Liesegang ring calcifications were observed. Nests of odontogenic epithelium in fibrous connective tissue and disorganized dentin and cementum were also evident. Scattered chronic inflammatory cell infiltration, muscle fiber sections, nerve bundles, adipose tissue, extracted RBCs, reactive bone, and hemorrhage were also evident. There was no evidence of malignancy (Figs. 5, 6).

Clinical, radiographical, and histopathological findings suggested a diagnosis of a combined odontogenic tumor that consisted of an epithelial odontogenic tumor and an odontoma.

During the 6-month follow-up, the displaced teeth of the anterior mandible were arranged as before without any additional treatment (Fig. 7).

The follow-up panoramic radiograph showed new bone formation in the peripheral wall of the defect. CBCT revealed a radiolucency, ~ 13 mm × 17 mm in size on the lingual side of the mandible, which indicates a lack of bone formation at the time of follow-up (Fig. 8).

Vitality tests by cold and electrical pulpal tests at the 6-month follow-up showed that the teeth involving adjacent lesions were vital.

No recurrence was observed in the first 12 months of follow-up.

Discussion

CEOT is a benign odontogenic tumor that accounts for ~ 1% of all odontogenic tumors. This tumor usually affects adults, with a peak of occurrence between the ages of 35 and 60 years. The subject patient was within this range.

Approximately 50% of the case reports of CEOT are associated with a tooth impacted and/or displaced by the tumor.

CEOT commonly occurs in the posterior jaw with associated cortex discontinuity.

The recurrence rate of CEOT is reported to be ~ 10–15%. Malignant transformation is very rare [7]. The tissue origin of CEOT remains controversial. Pindborg suggested that it originated from the retracted enamel epithelium of the embedded teeth. However, the tumor cells are similar to those in the middle layer of the enamel organ. Hence, most scholars believe it comes from the middle layer cells of the enamel organ [3].

CEOTs present either intraosseous or extraosseous lesions. It more frequently occurs in the posterior sites of the mandible and is often associated with an unerupted tooth. This tumor manifests as painless and slow-growing

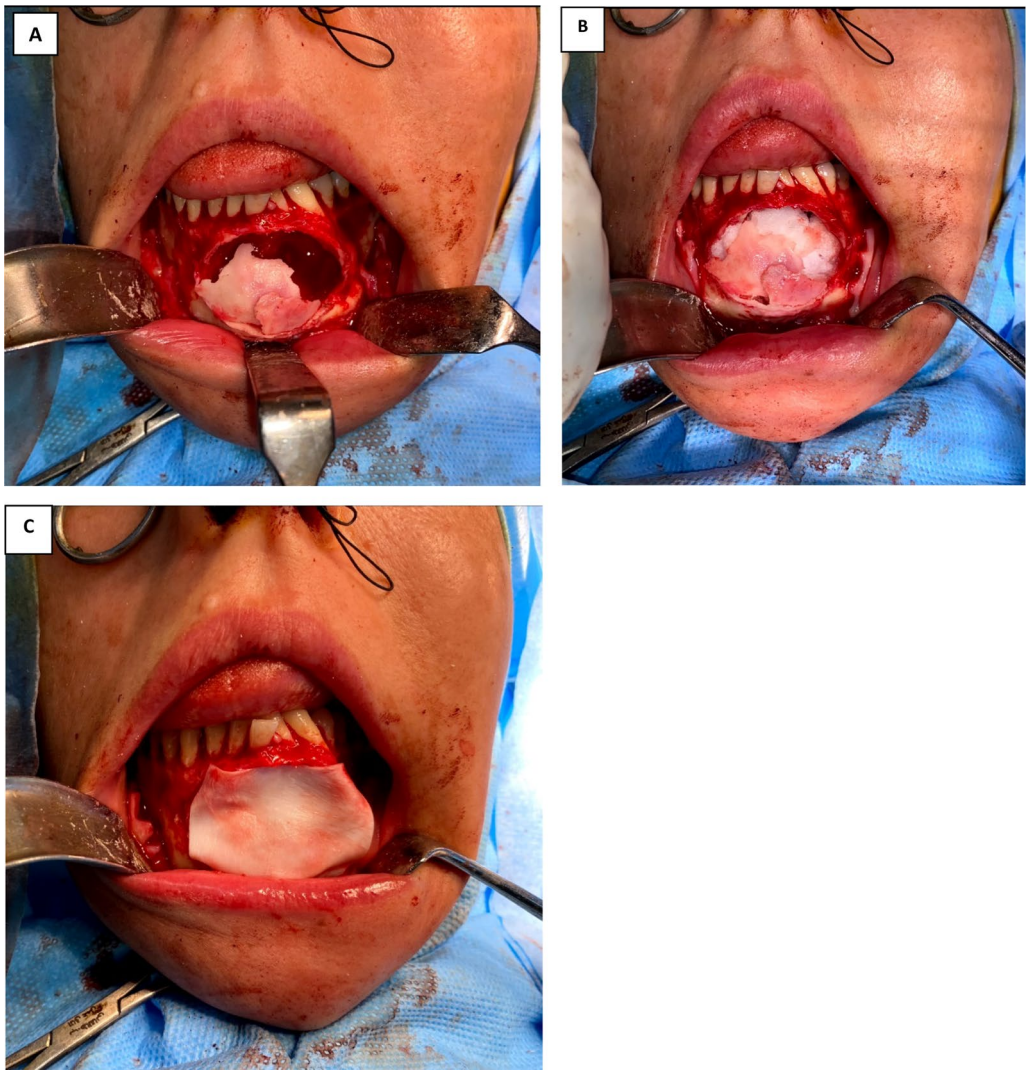

Fig. 4 The defect was simultaneously augmented with the remaining intact buccal cortex bone (**A**). The defect site was filled with Gelfoam (**B**) and covered with resorbable membrane (**C**)

swelling. The patient in this case study experienced practically painless swelling for ~ 1 year.

Radiographically, these lesions may have different features according to age. The mature lesions that display unilocular or multilocular radiolucency with radiopaque material are referred to as driven snow. The immature lesions are more radiolucent. Radiographically, the borders of tumors are usually well circumscribed but may have irregular borders [8].

There are several jaw lesions with similar features to CEOT in radiographic views.

The most frequent jaw masses that may be appear as multilocular lesions are ameloblastoma and odontogenic keratocysts (OKC).

Ameloblastoma is a benign odontogenic tumor with a locally aggressive growth and strong tendency for recurrence [9]. CEOT is thought to exhibit less aggressive growth pattern comparison to ameloblastoma [3].

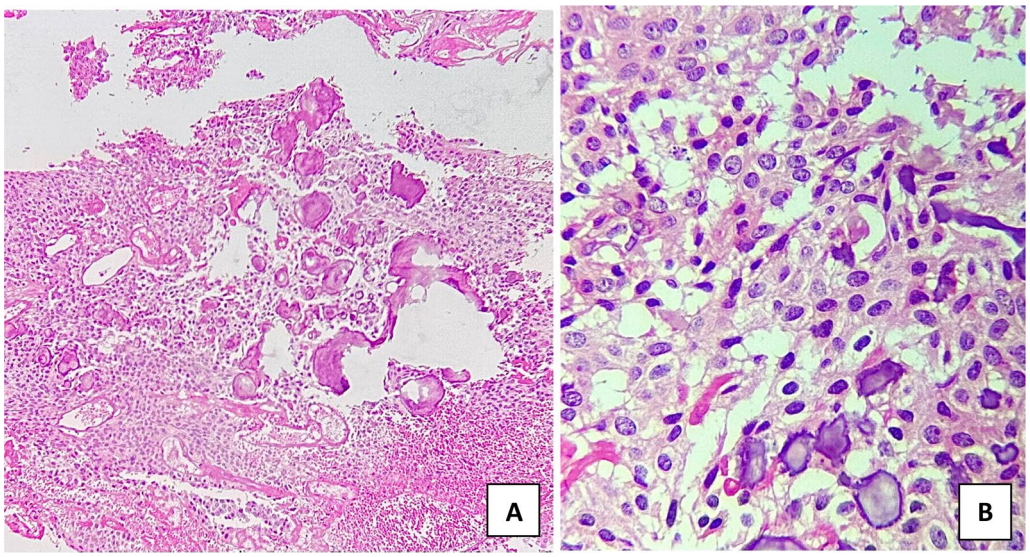

Fig. 5 **A**, **B** Photomicrograph showing an odontogenic neoplasm of sheets of polyhedral epithelial cells with abundant eosinophilic cytoplasm and inter cellular bridges. Multiple concentric calcifications (Liesegang ring) are seen [hematoxylin and eosin (H & E) staining, × 100, × 400, **A**, **B**]

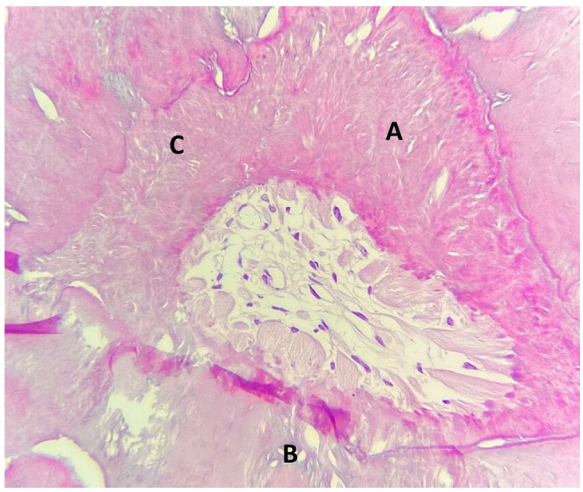

Fig. 6 Photomicrograph showing irregular tubular dentin (**A**), pre dentin (**B**), and incremental line(**C**) (H & E staining, × 400)

World health organization categorized OKS as a neoplasm due to destructive growth pattern and high frequent recurrences. One study reported the recurrence rate of OKC 9% to 60% according to the surgical approach [10] while Chrcanovic et al., reported the recurrence rate of CEOT of 12.6% [5].

In the patient, the lesion had mixed radiolucency and radiopacity. The radiopaque material appeared as a complex odontoma on the central side of the lesion. Radiographical examination revealed that the border of the lesion was well defined and that apical root resorption was considerable.

Hybrid odontogenic tumors consist of two or more odontogenic tumors that occur at the same site. The most common of these tumors are calcifying odontogenic cysts associated with odontoma, followed by central odontogenic fibroma with central giant cell granuloma, adenomatoid odontogenic tumors (AOT) with calcifying epithelial odontogenic tumor, AOT with dentigerous cyst (DC), and DC with odontoma.

Radiographical and histopathological examinations confirmed that the tumor in our study consisted of a CEOT and odontoma. The recurrence of hybrid tumors after nonaggressive surgical approaches is uncommon, and the prognosis of hybrid tumors is good [11].

CEOT leads to the displacement of adjacent teeth, which is often the first clinical sign. Despite the displacement, there is no evidence of tooth loosening [12]. In this patient, the anterior mandibular teeth were displaced and

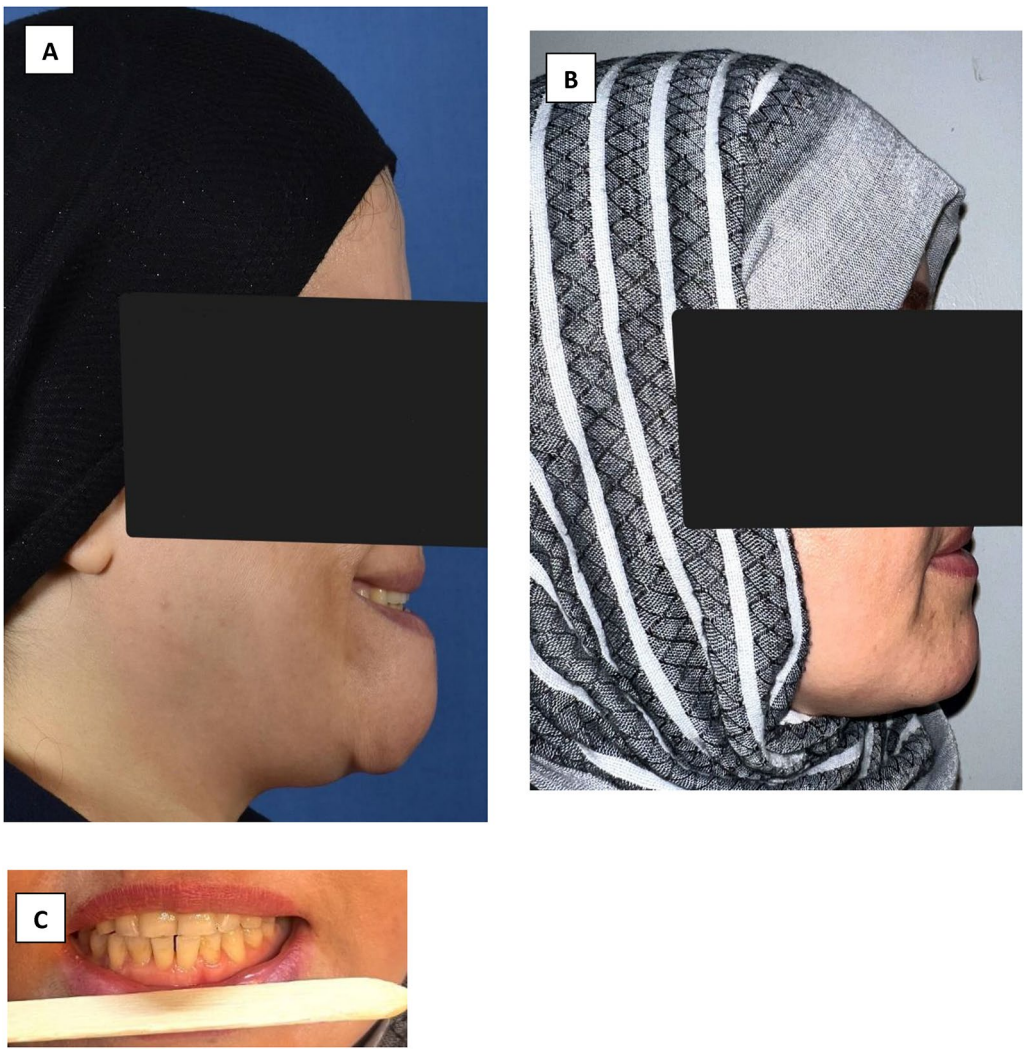

Fig. 7 **A** Pre operative view. **B** and **C** Follow up examination after 6 months. Notice the arranged teeth in **C**

mobile, but the vitality tests were positive. The patient was scheduled for strict follow-up. After 6 months, the displaced teeth were arranged naturally without additional treatment, such as root canal therapy or orthodontic treatment. The treatment plan for CEOT ranges from enucleation and curettage to radical and extensive resection. The appropriate surgical approach is based on the clinical manifestations and histopathological features [13]. Patient age and tumor location are other considerations for a suitable surgical plan [12].

Specifically, CEOT exhibits distinctive morphological features, such as clusters of floating tumor cells or clumps of cells. The histological manifestations of "classic" CEOT include masses of polyhedral epithelial cells with clear cell borders, nuclear pleomorphism, and minimal mitotic activity. Mitoses and nuclear pleomorphisms are highly uncommon. The matrix typically appears to be myxoid or myxoid with calcified islands, sometimes showing concentric or sandy calcifications. Amyloid deposition is another hallmark of CEOT, although the origin of this homogeneous material remains debated [14].

An ultrastructural study of CEOT by Page et al. revealed that amyloid is a protein product of enamel organs that is distinct from endocrine-related or systemic amyloid. Congo red staining demonstrated green

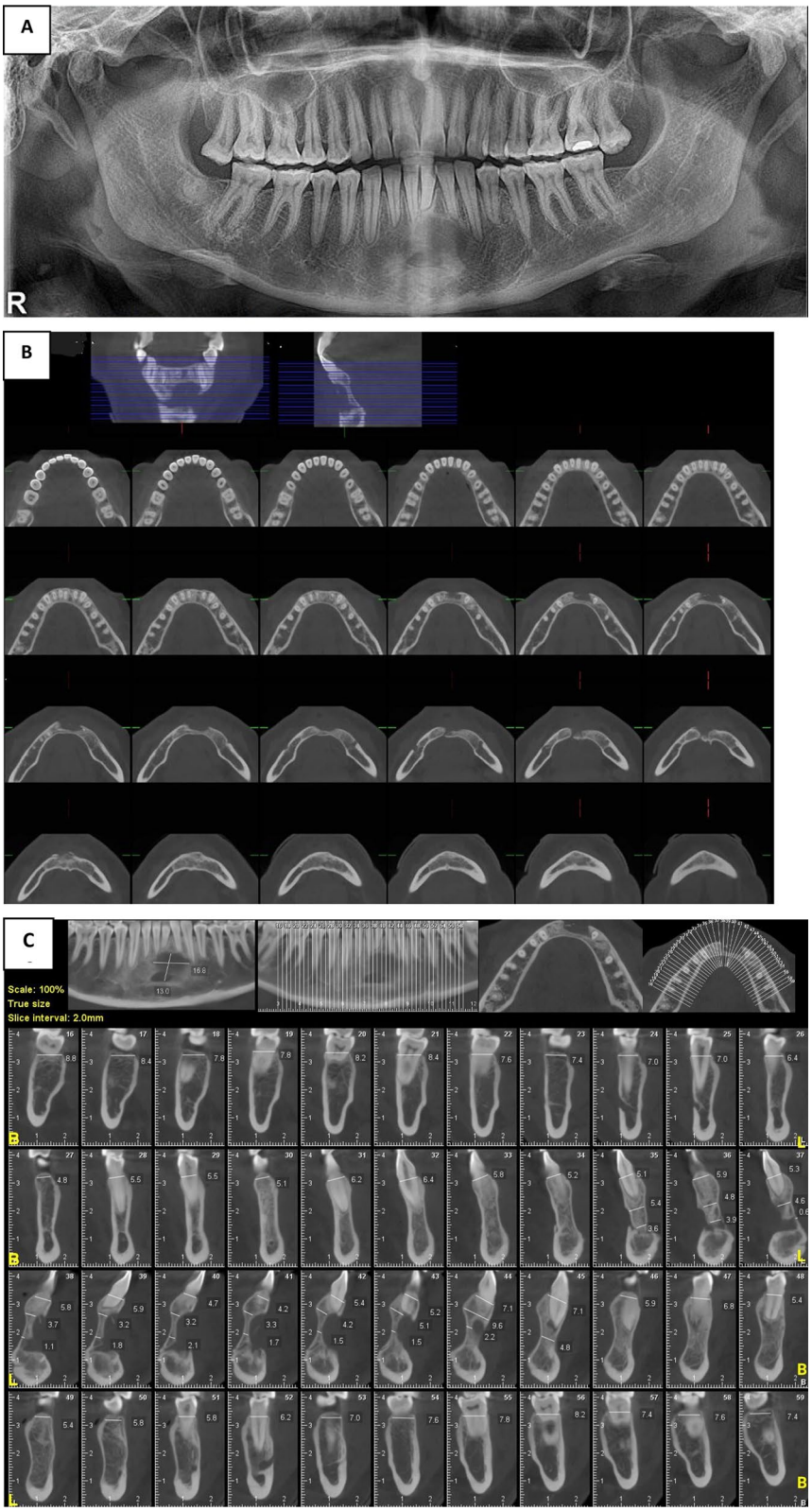

Fig. 8 **A**–**C** Postoperative radiographic evaluations. Notice the bone formation

birefringence of amyloid deposits in CEOT, aiding in its differentiation from other lesions [15].

Furthermore, El-Labban et al. proposed that the amyloid protein in CEOT is the result of breakdown of the gel-like substance secreted by tumor epithelial cells [16].

According to the WHO 2022 study, the classification has undergone significant changes and is divided into the following categories:

- clear cell CEOT
- cystic/microcystic CEOT
- noncalcifying/Langerhans cell-rich CEOT [17].

In this patient, the inferior border of the mandible was intact. Enucleation with curettage was chosen as the optional treatment plan. After excision of the lesion, the remaining bone of the buccal cortex was used to reconstruct the defect.

Conclusion

This case demonstrated the use of a less aggressive surgical approach for treating CEOT. By this surgical approach, without any tooth extraction, the teeth were functional and vital in the oral cavity without any mobility.

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Author contributions
Conception: Fereydoun Pourdanesh. Design of the work: Fereydoun Pourdanesh, Ardeshir Khorsand, Maryam Armanfar. Analysis, or interpretation of data: Fereydoun Pourdanesh, Maryam Armanfar, Fatemeh Mashhadiabbas, Sanaz Gholami, Maryam Mohammadalizade. Writing: original draft: Maryam Armanfar. Writing—revising and editing: Maryam Armanfar, Ardeshir Khorsand, Maryam Mohammadalizadeh.

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Availability of data and materials
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Declarations

Ethics approval and consent to participate
Not applicable.

Consent for publication
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Competing interests
There are no competing interests.

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