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Case Report

Calcifying Epithelial Odontogenic Tumor (Pindborg's Tumor)

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Abstract

Calcifying epithelial odontogenic tumor (CEOT), also known as Pindborg's tumor, is a rare benign odontogenic tumor of locally aggressive behavior. The most frequent location is the mandibular premolar and molar area; less frequently, the lesion is found in the maxilla, typically in the fourth to fifth decades. It usually starts as a painless swelling and is often concurrent with an impacted tooth. A case of CEOT in a 21-year-old male showed up in the right body of the mandible and ramus region was described. Clinical, radiological, histopathological features and treatment were discussed. This tumor was managed by surgical removal and reconstruct of the mandible using a reconstruction plate. The case was followed up for 3 months postoperatively.

Keywords: Mandible, odontogenic tumors, Pindborg's tumor, reconstruction plate

Introduction

The calcifying epithelial odontogenic tumor (CEOT) is a rare benign tumor of the jaws. It was first described by Pindborg in 1955 who reported three benign odontogenic tumor arising from the mandible;[1] since then, nearly, two hundred cases have been reported for CEOT as a benign odontogenic tumor of epithelial origin that accounts for approximately 1% of all odontogenic tumors.[2] The origin of this neoplasm is not clearly known, although it is generally accepted to be derived from oral epithelium, reduced enamel epithelium, stratum intermedium, or dental lamina remnants.[2-4] The mean age of the occurrence at the time of diagnosis is 40–41 years, and there is no significant difference in the occurrence between genders.[5] Most cases involve the posterior part of mandible; there have been few reported maxillary cases.[6] CEOT is also a painless slow-growing tumor which causes jaw expansion.[7] The differential diagnosis for Pindborg's tumor includes calcifying odontogenic cyst, ameloblastic fibro odentoma, and adenomatoid odontogenic tumor.

The current case report describes an extraordinary type of CEOT regarding the patient's age and the extension and the size of the tumor.

Case Report

A 21-year-old male with a complaint of facial asymmetry was presented at the Department of Maxillofacial Surgery in the Faculty of Dentistry at Damascus University, with 10 years history of nonfluctuant painless mandibular right side swelling. The extraoral inspection revealed an oval-shaped swelling, presented over the right angle of the body of the mandible. The skin over the swelling was slightly stretched with no secondary changes. The swelling was fixed to the underlying structures. It was not associated with discharge and numbness. No associated signs and symptoms were present or lymphadenopathy. Intraoral examination revealed a diffused swelling present in the lower right posterior angle and body region extending anteriorly up to the buccal gingival sulcus of tooth #41 causing obliteration of lingual and buccal vestibule. On palpation, it was not tender. The swelling pushes the tongue distally away from the midline.

The mucosa overlying the lesion was intact and teeth in the vicinity showed no mobility except the tooth number 46 which

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showed a significant movement grade III. No discoloration, no tenderness and responded positively to the vitality tests.

His medical and dental history was non-contributory. General examination revealed a moderately built and nourished individual of normal gait with mild back bowing. Vital signs within the normal range. No localized rise in temperature was noticed [Figures 1-3].

Radiographical investigations

On a panoramic radiograph, a multilocular radiolucency with a sclerotic border involving the right body and ascending ramus to the level of the inferior mandibular foramen with a "honey comb" appearance at the site between 46 and 47, at a size of 1 cm × 1 cm was noticed. A multilocular radiolucent appearance was also shown [Figure 4].

Cone-beam computed tomography (CBCT) radiograph showed that the mass was pushing both the hyoid bone and laryngopharynx distally [Figures 5-8].

Histopathological examination

A biopsy was taken from the affected site. It was shown that the tumor consisted of discrete islands, sheets, or strands of polyhedral epithelial cells (with abundant eosinophilic cytoplasm) in a fibrous stroma [Figure 9].

Within these epithelial sheets, large areas of homogeneous amyloid-like were found, which in turn, contained developed calcifications of concentric laminated structures.

Surgical technique and treatment

After general anesthesia using nasal intubation, a linear incision was drawn on the skin. And because of the missing of the lower border of the mandible, this line was put approximately 2 cm below. The incision line was about 11 cm long, extended from the proposed mandibular angle to the midline of the chin.

After excising the layers in the region one by one and after the facial vein and artery were cut and roped, the whole tumor mass was exposed anteriorly and posteriorly. There was a yellow, pure liquid coming out from the mandibular mass, in addition to the profuse bleeding from it with a thin bony cortex surrounding the whole tumor mass. The lingual side of the tumor was dissected easily and gently from the underlying structures. An anterior osteotomy was made distally to the left canine and another one superiorly at the level of the mandibular foramen. After the mass was removed, an expansion of the medial cortex of the ramus above the posterior

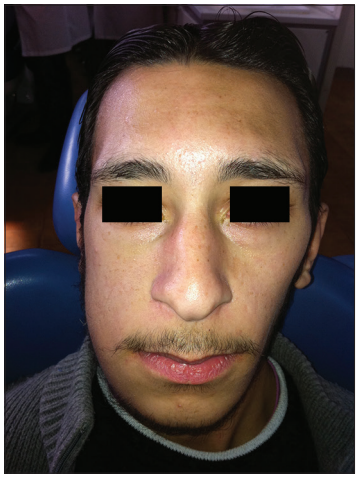

Figure 1: Extraoral frontal view of the lesion

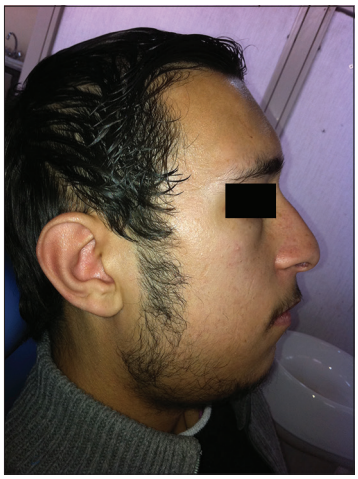

Figure 2: Extraoral lateral view of the lesion

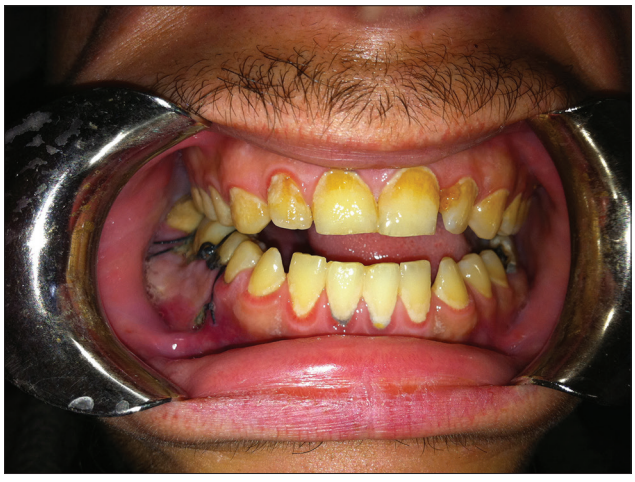

Figure 3: Intraoral view of the lesion

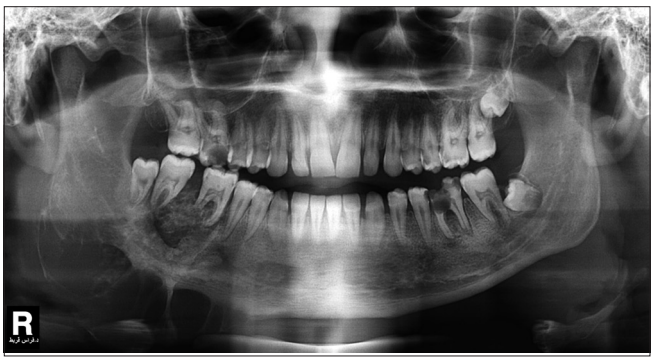

Figure 4: Panoramic radiograph revealed expansion and thinning of the cortex of the right lower border of the mandibular body and posterior border of the right ramus

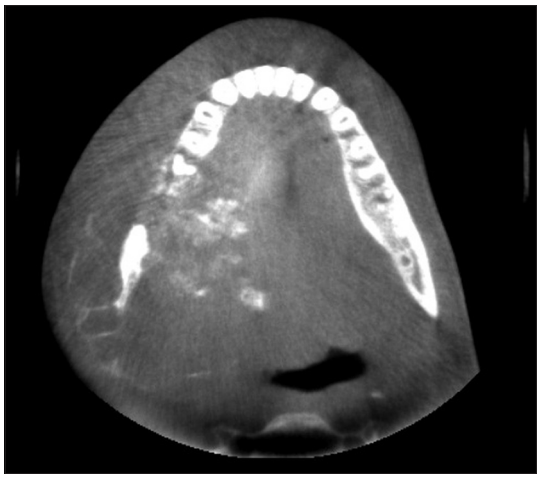

Figure 5: Axial CBCT view of the mandible that shows the extent of the lesion mesially

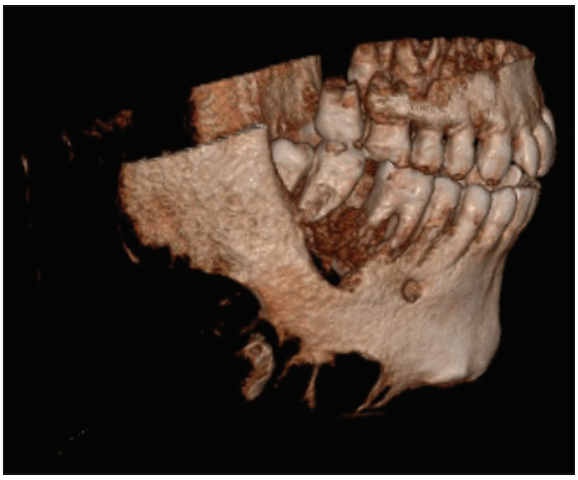

Figure 7: A Lateral and Posterior 3D view of CBCT of the mandible that shows the extent of the lesion's border inferiorly and mesially.

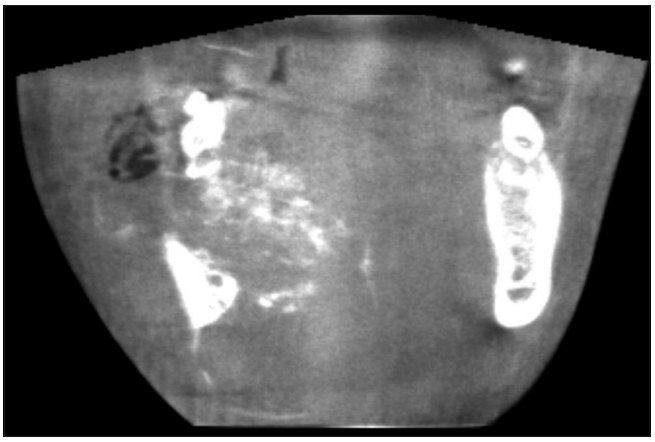

Figure 6: Anterior Posterior CBCT view of the mandible that shows the extent of the lesion mesially

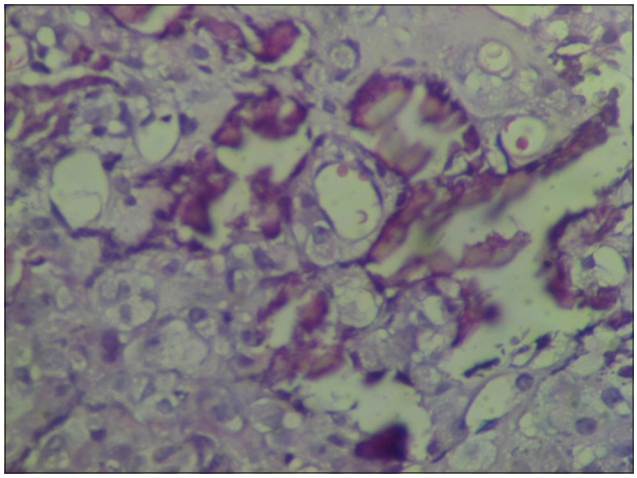

Figure 8: Discrete islands, sheets or strands of polyhedral epithelial cells (with abundant eosinophilic cytoplasm) in a fibrous stroma

osteotomy cut level was noticed. That demanded an additional excision to the coronal process, and the condylar neck was left sound [Figures 10 and 11].

After that, we have adapted the reconstruction plate carefully and fixed it in position securely.

Three months later, the patient was asked to visit the department for frequent recall in which he showed normal anatomical features of the face without any complaints. Unfortunately, the follow-up pictures were lost due to the war situation in the country.

Discussion

Pindborg's tumor is a rare, benign, but locally aggressive odontogenic tumor, which accounts for <1% of all odontogenic tumors.[2] Most investigators believe that the tumor cells originate from the reduced enamel epithelium, but today they believe origin from the stratum intermedium as cellular morphology is similar to tumor cells[8] and they agree that the central type is usually located in the premolar and molar regions with a mandibular to maxillary ratio of 2:1.[9]

In some cases, the lesion becomes multilocular with a honeycomb pattern. In others, multiple radio-opacities are seen within the radiolucent area, giving rise to the term "driven snow appearance."

In our case, the tumor reached large size that is not common in most Pindborg's tumors and that made the tumor mass push both the hyoid bone and the laryngopharynx, distally, and extending to the coronal process. However, we could not find in literature a case reporting such size and location.

The patient in our case was at the age of 21 and that agrees with Philipsen and Reichart[10] and both Franklin and Pindborg.[1,11]

The lesion was not associated with unerupted tooth, and that is, which did not agree with the features previously described by Philipsen and Reichart.[10]

No malignant deformities or metastasis were shown in our case which did not agree with the features previously described by Veness *et al*.[12]

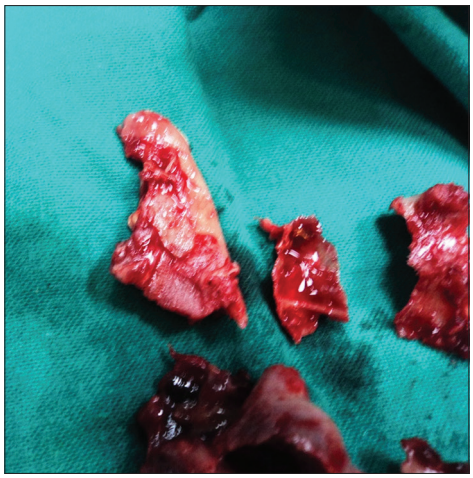

Figure 9: Multiple excised pieces of the lesion from the ramus area

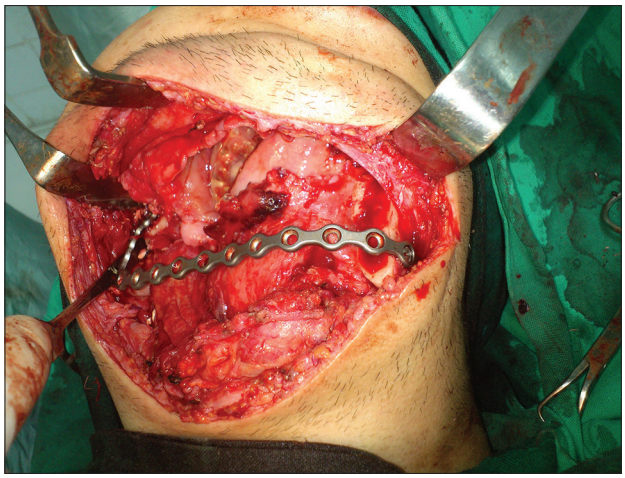

Figure 10: A view during the sufgical operation shows the titanum reconsruction plate adapted and fixed in its proper place.

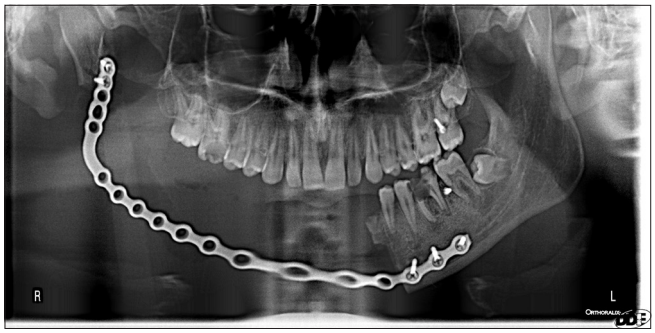

Figure 11: Post operation panoramic radiograph shows the reconstruction plate fixed in its place.

The tumor in our case affected the premolars' area of mandible, which agrees with Cicconetti *et al*.[13] in their clinical case description.

The CBCT was not sufficient to insure the extent of the lesion in the ramus, and it showed a perforation areas in the bone cortex surrounding the lesion. However, surgically, the whole lesion was capsulated securely.

The weird thing that appeared during surgery was that the lesion was extremely bleeding from the distal face.

Because of the wrong appearance of the lesion radiographically by Panoramic view and the insufficient view of the ramus in CBCT, the assumed cutting line superiorly was not true and made us do additional excision to the ramus and keeping only the condylar neck which seemed to be sound.

However, in case of gross deformity of the tumor, we suggest using a rubber barrier with powerful surgical suction. We also suggest a careful surgical manipulation on such tumors to avoid the tumor cells spillage into surrounding tissues.

Conclusion

After tumor excision, we faced a lot of difficulties in adapting the reconstruction plate due to the missing contour of the mandible. Hence, we recommend to make a tunnel through the tumor mass and adapt the reconstruction plate before incision.

For diagnosis and treatment plan construction for CEOT, it is not enough to depend on radiographical diagnosis by means of CBCT alone, because it does not always help giving the true border and extension.

Declaration of patient consent

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

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Nil.

Conflicts of interest

There are no conflicts of interest.

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