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Case Series

Calcifying Epithelial Odontogenic Tumour (Pindborg Tumour): Report of Two Clinical Cases with A Rare Maxillary Localization

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Abstract

A calcifying epithelial odontogenic tumour (CEOT) or Pindborg tumour is a rare benign eithelial odontogenic neoplasm that was first described by Jens Jorgen Pindborg in 1955. It accounts for less than 1% of all odontogenic tumors [1]. This benign odontogenic tumour is slow growing and expansile. The age of discovery varies between the third and fifth decades, and the M/F sex ratio is balanced. Involvement of the mandibular molar region is more frequent.Clinically, the tumor may evolve silently, and discovery is often fortuitous or the result of swollen tooth movements.Radiologically, it may be homogeneous or heterogeneous, with or without calcifications. The origin of the tumor remains debated, and treatment is surgical, involving simple enucleation.

The aim of this work is to report two cases of Pindborg tumor with a rare maxillary localisation and to discuss its clinical and radiological signs.

Keywords: Pindborg Tumour; Calcifying Epithelial Odontogenic Tumor; Odontogenic Epithelial Tumor

Introduction

A calcifying epithelial odontogenic tumour (CEOT) or Pindborg tumour is a rare benign epithelial odontogenic neoplasm that was first described by Jens Jorgen Pindborg in 1955. It accounts for less than 1% of all odontogenic tumors [1]. This benign odontogenic tumour is slow growing and expansile.

These tumors affect a wide range of ages, but are most common between the ages of 40 and 60. No gender preference has been reported. The majority of CEOTs are intraosseous (96%), with the mandible being more affected than the maxilla [3].

According to the latest WHO classification (2022), three subtypes have been described as clear cell CEOT, cystic/microcystic CEOT, and non-calcified/Langerhans cell-enriched CEOT [2]. The origin of CEOT is still debated. It may originate from the oral epithelium, reduced enamel epithelium, intermediate layers, or remnants of primitive dental ridges [1,12].

The aim of this work is to report two rare cases of the maxillary localization of Pindborg tumor and to discuss its clinical and radiological signs.

Case 1

A 28-year-old female patient, with no particular medical history, was referred to the Department of Oral surgery for diagnosis and treatment of a nasolabial swelling, as well as dental displacements.

The patient reported that the history of the disease goes back to 2 years ago with the appearance of a painless swelling progressively increasing in size, associated with dental displacement and mobility.

On exobuccal examination, facial symmetry was not respected by the presence of a right nasolabial swelling without adenopathy. The bi-commissural line was oblique down the right side with filling of the right nasolabial fold.Palpation revealed a well-limited, painless, non-depressible mass. (Figure 1).

On endobuccal examination, dental hygiene was defective with halitosis, the gingival mucosa was reddish in places, sessile, budding with the impression of adjacent teeth occlusally (Figure 2).

we also noted

- Egression of the 11 12 13 14
- Mobility of the 13 14
- Negative pulp vitality test on 14
- 26 dilapidated

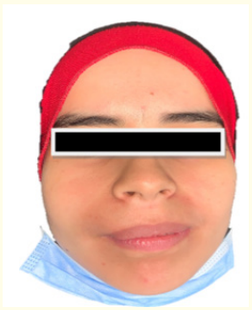

Figure 1: Extra oral swelling in the right nasolabial region.

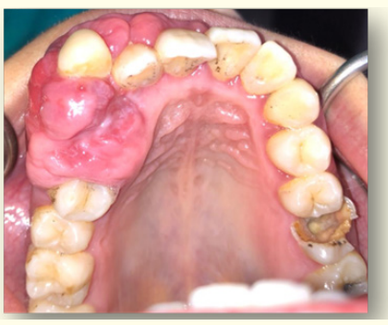

Figure 2: Intraoral picture of the lesion.

Palpation was painless with no bleeding, soft in occlusal and firm vestibularly.

Orthopantamogram (OPG) revealed voluminous mixed image with sharp borders,which was present between 11 and 15. 11, 12 and 13 were displaced apically. Moreover, the panoramic radiograph revealed radicular resorption in the apical third of teeth 12 and 14, along with desmodontal enlargement of teeth 11 and 21 (Figure 3). However, three-dimensional imaging showcased a blown-out appearance and thinning of the vestibular cortex (Figure 4).

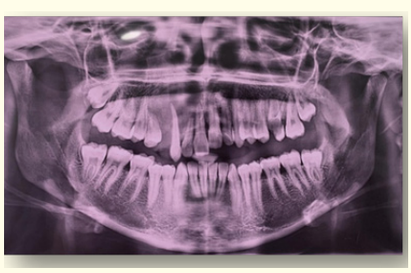

Figure 3: Orthopantamogram (OPG) showing mixed image with sharp borders.

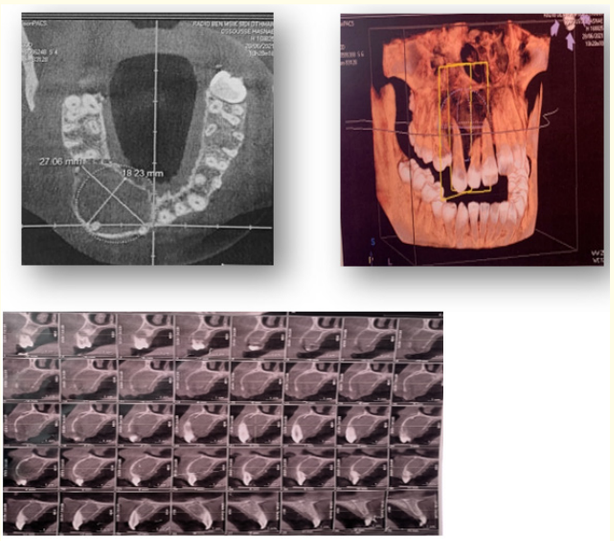

Figure 4: The 3D imaging showing blowing and thinning of the vestibular cortex.

As diagnosis we had thought of an ossifying fibroma.

The management was surgical .After a sedative preparation of the young patient, we proceeded to the removal of the lesion as well as the extraction of the 13 and 14 under local anaesthesia with sending of the operative part (Oval-shaped, measuring 3 cm in diameter, enclosed by an irregular fibrous membrane, and exhibiting a firm consistency reminiscent of a complete tumor) to the laboratory for an anatomopathological examination. (Figure 5) The latter was in favor of calcifying epithelial odontogenic tumour. The patient was seen 3 months later, and after a year, and was free of recurrence (Figure 6).

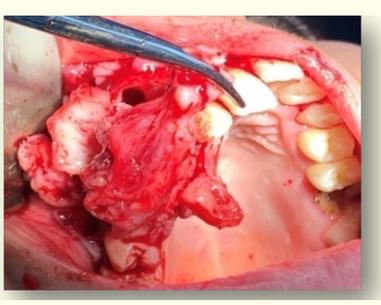

Figure 5: Intraoperative photo of the lesion's exerisis.

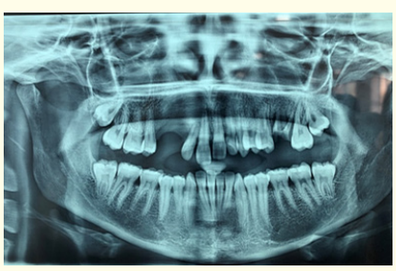

Figure 6: Follow-up radiograph after 3 months.

Case 2

A 18-year-old female patient, without any notable medical history, has been referred by her orthodontist for diagnosis and treatment of a left maxillary swelling that has been evolving since childhood.this swelling was painless.

During the extraoral examination, we observed labial incompetence, superior proalveolus, and the effacement of the left nasogenian groove. (Figure 7).

During the intraoral examination, we observed a well-defined swelling in the left maxilla covered by a mucosa of healthy appearance. It extends from the mesial aspect of the 22 to the distal aspect of the 24, and vertically, it reaches beyond the mucogingival line.

Upon palpation, there is a depressible vestibular filling adjacent to the 21 and 22, with the 22 exhibiting rotation. (Figure 8).

Orthopantamogram (OPG) showed a well-defined bilobed mixed anterior maxillary image, delineated by a radiopaque border adjacent to the 63, displacing the root of the 22 and extending up to the nasal fossa, with the presence of a radiopaque image of dental density. (Figure 9).

Given these clinical and radiological findings, we have considered the possibility of an odontoma.

The management was surgical .After local anesthesia, a full-thickness mucoperiosteal flap was created, allowing visualization of the externalization and bulging of the outer cortical plate. After osteotomy, calcified yellowish elements was identified (Figure 10).

After the complete excision of the lesion, the surgical specimen, consisting of a cystic wall and disorganized dental elements, was sent to the laboratory for histopathological examination. The latter Confirmed the diagnosis of of calcifying epithelial odontogenic tumour.

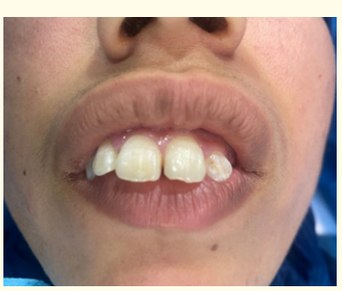

Figure 7: Extraoral view revealing the obliteration of the left nasolabial fold.

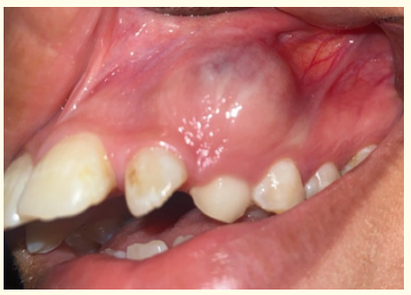

Figure 8: Preoperative view of the lesion.

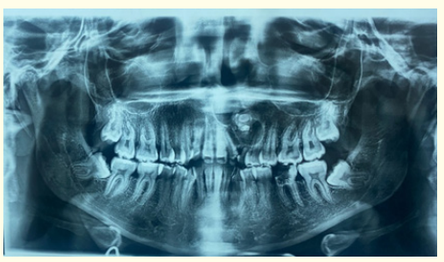

Figure 9: Preoperative orthopantamogram showing a well-defined bilobed mixed anterior maxillary lesion.

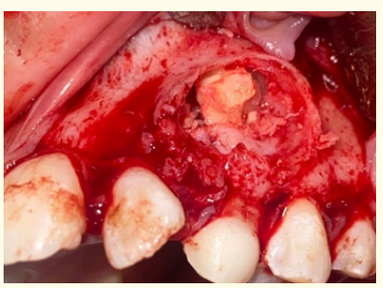

Figure 10: The surgical approach to the lesion shows calcified yellowish elements.

Discussion

The literature refers to CEOT,as a rare slowly growing neoplasm . According to the current statistics, CEOT has no sex predilection. Most patients of central and peripheral type CEOT are above 40 and 35 years of ag [5]. The first reported case, the patient was 28 years old, while in the second case, the patient was 18 years old.

Approximately 90% of CEOT cases are intraosseous, and the mandible is the most affected site in these patients. Maxillary tumors are observed in 25% to 41% of cases and generally occur in the posterior region. Peripheral CEOT accounts for about 10% of cases and has a predilection for the anterior gingiva [3,4,7,8]. The present reports adds to the small number of CEOT cases located in the maxilla since most cases occur in the posterior mandible.

The central type of CEOT is usually asymptomatic for longer periods and a bony hard swelling is evident only in the advanced stages of the tumor. Other signs can be accompanied, in particular the mobility and dental displacement [5]. In the first case of central CEOT patient has swelling for the last 2 years with mobility and dental displacements. In the second clinical case, the patient experienced a left swelling that had been evolving since childhood. The swelling was painless and associated with rotation of the tooth 22.

Kaplan., *et al*. reported that root resorption occurred in only 4% of 67 cases. Whereas, in solid ameloblastoma root resorption is a more common feature, this might help in differentiating it from CEOT [10]. The first clinical case also showed root resorption of the apical third of 12 and 14.

On the radiograph, central CEOTs appear as unilocular or multilocular, well to ill-defined, corticated or partly corticated radiolucent lesions.[7] Lesions of the maxilla are majorly unilocular. In the mandible, the tumor is frequently associated with the impacted mandibular molars, in which mandibular second molar is more common as compared to the first and third molars. In the present cases, the lesion was in the maxilla unilocular for the first case and bilobed for the second case, both with well-defined borders [5,8-11].

The treatment for CEOT is surgical excision, but treatment plans vary from patient to patient, ranging from simple excision to mandibular hemisectomy/mandibular resection [1].

Generally, treatment begins with surgical removal of the tumor. Most authors agree that the resection should include a bony safety margin that is considered clinically and radiologically sound because the lesion is not encapsulated [2].

CEOT has been characterized histologically by the presence of three components - polyhedral, often pleomorphic epithelial cells with prominent intercellular bridges, amorphous amyloid-like deposits and calcifications. For Krolls and Pindborg, the presence of calcifications in CEOT may influence prognosis. Lower calcification indicates a higher probability of recurrence of poorly differentiated tumors. It was also found that similar histopathologic features were present in our cases with calcification, indicating a well-differentiated tumor and a low propensity for recurrence [1].

There is very low incidence of transformation of CEOTs into malignant tumors and few such cases have been reported in the literature and may rarely show metastasis.

To prevent the recurrence of the treated cases of CEOT periodic follow up is necessary. Five years minimum follow up period is necessary for treated patients [5].

In the recent literature, mutations in tumor suppressor genes (PTEN, CDKN2A, PTCH1) and oncogenes (JAK3, MET) are also detected in CEOT [12]. Patients with CEOT are therefore followed up every 3 months. Patients are also monitored and regularly evaluated for recurrence [5].

Conclusion

Calcifying epithelial odontogenic is a rare locally invasive neoplasm. Its diagnosis is guided by radiology, but is difficult before the appearance of calcifications. The final diagnosis is based on histology .A long-term follow-up must be undertaken.

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