

Epiphora and nasal blockage – An unusual presentation of Pindborg tumor: A case report and literature review

ABSTRACT

Odontogenic tumors are silent ailments which can affect any individual. One such lesion is a calcifying epithelial odontogenic tumor (CEOT). It is a rare lesion with a locally aggressive nature. A 33-year-old male presented with nasal blockage and continuous flow of tears from the left eye for the past 7–8 months. Clinical examination revealed slightly protruded left eyeball and altered level of eyeballs with continuous watery discharge. Hess chart confirmed normal eye movements. Intraoral findings were not significant. The final diagnosis of CEOT was established based on the histopathological aspects. CEOT is mostly found in the third to fifth decade of life without gender predilection. Presenting symptoms and signs comprise painless expansile mass, although there are reports associated with pain, nasal obstruction, epistaxis, and proptosis. In this article, we would like to present a case of CEOT with epiphora and nasal blockage as the main and only presentation, an uncommon finding. Furthermore, discussing and posing a question of an adequate period of follow-up required to negate the presence of recurrence. One must stay vigilant enough not only to attest a singular symptom to the commonest ailment related to it but also to explore the possibility of the less known. Furthermore, we need to further research in depth to establish a certain duration after which the likeliness of recurrence is to the minimum.

Keywords: Calcifying epithelial odontogenic tumor, epiphora, maxilla, nasal blockage, pathology, Pindborg tumor

INTRODUCTION

A calcifying epithelial odontogenic tumor (CEOT) is an uncommon entity comprising 1% of all odontogenic tumors. Its behavior is synchronous to that of ameloblastoma, like being locally expansile, can reoccur, and does not metastasize. This tumor is usually seen in patients in their third to fifth decade, with no gender predilection. Lesions are most commonly found in the molar region of the mandible, followed by premolar region, except for Langerhans cell variant. Large CEOTs may require either marginal or segmental resection, but as it is less aggressive than ameloblastoma, small lesions can be treated more conservatively.^[1] An overall recurrence rate of roughly 10% has been reported.^[2] Because of its slow growth, a follow-up period of 5 years has been suggested.^[3]

CASE REPORT

A 33-year-old male patient presented to the oral and maxillofacial surgery department complaining of nasal blockage and continuous flow of tears from the left eye

for the past 7–8 months. Clinical examination revealed slightly protruded left eyeball and altered level of eyeballs with continuous watery discharge [Figure 1]. Complete ophthalmologic evaluation of the patient was done. A Hess chart confirmed normal ocular movement with no neurologic defect. A common cause of epiphora is tear duct obstruction which is, in turn, caused by allergies, infection or inflammation, trauma, excessive eye drop usage, and history of radiation. All these reasons were ruled out by subtractive history taking. It arose the suspicion of obstruction due to a


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growth within maxilla. The patient also complained regarding nasal blockage which raised suspicion of underlying tumor. Further history revealed that the patient had undergone a previous surgery for growth in the maxillary sinus. At present, there was no complaint of paraesthesia. Intraoral findings were insignificant. The lymph node examination was unremarkable. The patient did not have any comorbidities or destructive habits.

Six and a half years ago, the patient had presented with a swelling concerning the left posterior maxillary arch. Examination heeded the presence of a firm nontender growth in the left maxilla involving posterior alveolar mucosa and vestibule. The patient had a history of extraction of 26, 27, and 28 with thinning of the medial and superior walls of the maxillary sinus, showing calcification and tooth within it. The excised lesion with tooth 28 was diagnosed as CEOT.

The histopathological examination of the previous tumor showed discrete islands, strands, and sheets of polyhedral epithelial cells in fibrous stroma with distinct intercellular bridges. Large areas of amorphous, eosinophilic, hyalinized extracellular material with areas of calcifications were observed. Liesegang rings were found too.

Computed tomography scan of the present illness [Figure 2] stated the presence of ill-defined soft-tissue lesion with diffuse areas of dense calcifications with an expansion of sinus and erosion of its anterior, medial, posterior, and superior wall, extending medially into the left nasal cavity causing pressure on the septum. Attenuation of the left lamina papyracea was also evident. The lesion was extending into the infratemporal fossa and superiorly into the orbit and inferiorly caused erosion of maxillary alveolar ridge. Incisional biopsy affirmed our suspicion of recurrence.

Conservative surgical resection was planned under general anesthesia, and a modified Weber–Fergusson’s approach was used. Intraoperatively, a well-demarcated lesion from the adjacent tissues was noted [Figure 3].

On histopathological examination, tumor mass showed that sheets of epithelial cells with mild nuclear pleomorphism and enlargement associated with hyperchromasia were observed. Plenty of hyaline areas admixed with foci of calcification were noted. The final diagnosis made was CEOT [Figure 4].

Postoperative recovery was inconsequential. Epiphora was controlled overtly during recovery phase. No adverse eye signs were noted. The patient is asymptomatic to date [Figure 5].

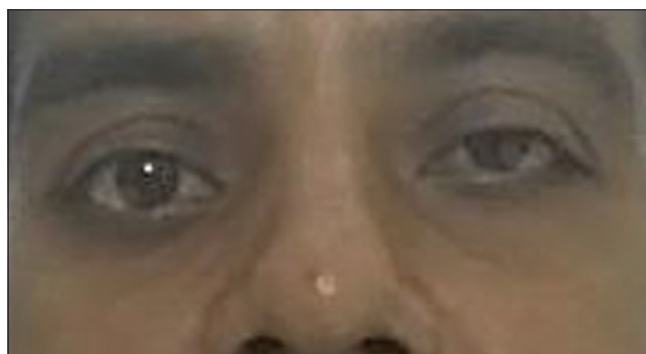


Figure 1: Altered eye levels

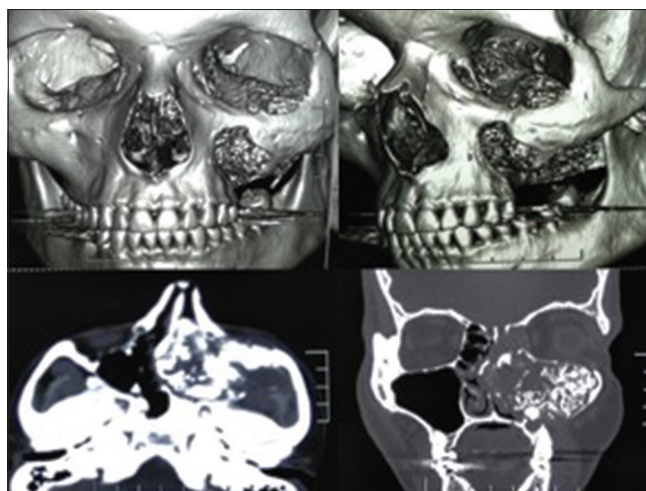


Figure 2: Computed tomography scan with three-dimensional reconstruction and axial and coronal views



Figure 3: Surgical procedure

One-year postsurgery endoscopic sinus examination was done to rule out recurrence and came out to be negative.

DISCUSSION

The CEOT is additionally referred to as Pindborg tumor and is considered a rare pathological entity representing <2.5% of all odontogenic tumors. CEOT is a benign neoplasm

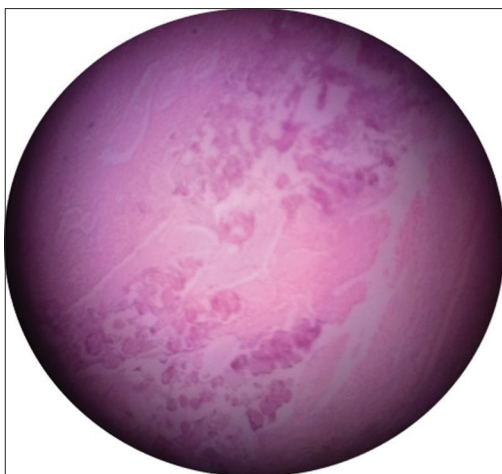


Figure 4: H and E-stained specimen slide



Figure 5: Postoperative image after 1 year

with painless, slow-growing expansion of the jaws. The tumor is usually treated surgically. Devising an effective treatment plan, especially in destructive cases of the upper jaw, becomes necessary as recurrence is not rare with such lesions. The lesions in the posterior maxilla represent <25% of all CEOTs.^[4] CEOT is associated with a tooth, erupted or unerupted, in 48% of cases. Radiographically, a mixed radiolucent and radiopaque lesion, unilocular or multilocular, is most characteristic.^[1] Radiographic features are similar to an ameloblastoma, dentigerous cyst, or other odontogenic tumors. Although typically benign, CEOT tends to invade local structures and has a potential for recurrence.^[5]

Franklin and Pindborg reviewed 113 cases. They mentioned that Pindborg tumor usually presents between 30 and 50 years of age with no gender predilection. Ninety-five percent are intraosseous lesions. Prevalence in the molar region to the premolar region is 3:1. Prevalence according to jaws is mandible to maxilla 2:1. The extraosseous cases (5%) described appearing to have a predilection for the anterior region. Although Pindborg tumors are well described in the mandible, descriptions of lesions involving the maxilla are rare.^[6,7]

The typical presenting symptom is painless asymptomatic expansile mass, although there are reports associated with pain, nasal obstruction, epistaxis, and proptosis. The additional and unusual symptom associated with this case was epiphora, which we did not find mentioned anywhere in the literature. Moreover, it was the main complaint of the patient with nasal blockage as a second and none of the other aforementioned entities.

Epiphora implies overflowing of tears due to impairment of lacrimal drainage. It indicates an unbalance between

tear production and tear loss. Causes can be anatomic or functional. Nasolacrimal duct obstruction is caused by allergies, infection or inflammation, trauma, excessive eye drop usage, tumor, or history of radiation. In our case, mechanical obstruction of lacrimal apparatus due to pressure exerted by tumor mass led to difficulty in normal drainage of tears produced which is normally drained in the nasal cavity in the inferior meatus through nasolacrimal duct connected to the lacrimal sac situated at the lacrimal fossa of the lacrimal bone. Furthermore, nasal stuffiness was a result of pressure over the septum by the tumor.

The patient had a previous history of such lesion, so he reported to the maxillofacial surgery department, but in instances where such lesion manifests only as epiphora with no other clinically significant symptom, one is likely to entertain the thought of visiting experts of other specialties as, for example, an ophthalmologist. Hence, getting to the core of a greater disease through means of investigating an unusual symptom becomes crucial.

Literature review

An electronic search was conducted without time restriction in March 2020 from the following databases: PubMed/Medline, Science Direct, Cochrane, and Google Scholar. The terms used for the search were: calcifying epithelial odontogenic tumor or Pindborg tumor.^[4,7-17] Inclusion criteria comprise tumor involving naso-orbito-maxillary complex, patients with adequate follow-up periods, and cases where presenting signs and symptoms are mentioned. Exclusion criteria included CEOT at other sites than posterior maxilla, cases where presenting signs and symptoms are not mentioned or described vaguely, and cases with irregular/no follow-ups. The titles, abstracts, and full reports (when required) of all reports identified through the electronic searches were read independently by the authors.

Table 1: Presenting symptoms of the patient with maxillary calcifying epithelial odontogenic tumor

Reference	Age/sex	Main complaint	Surgery	Follow-up period	Recurrence
Gon 1964	35/female	Nasal stuffiness and swelling over the right antrum and proptosis	Conservative	1 year	Absent
Stimson <i>et al.</i> , 1967	35/male	Intraoral growth	Hemimaxillectomy	15 years	Absent
Ai-Ru <i>et al.</i> , 1982	28/female	Swelling of the jaw	Hemimaxillectomy	12 years	Absent
Lee <i>et al.</i> , 1992	27/female	Progressive enlargement of the malar region	Intraoral hemimaxillectomy	7 years	Absent
Patino <i>et al.</i> , 2005	67/female	Intraoral growth	Conservative	4 years	Absent
	24/female	Intraoral growth	Partial maxillectomy	4 years	Absent
Gopalakrishna <i>et al.</i> , 2006	15/Female	Cystic swelling over the maxilla	Enucleation	1 year	Absent
Bridle <i>et al.</i> , 2006	30/Female	Right eye higher in level than the left eye	Enucleation and curettage	18 months	Absent
Sehgizhadeh <i>et al.</i> , 2007	51/male	Incidental finding on a routine radiograph	Enucleation	17 years	Present
Channappa <i>et al.</i> , 2011	30/male	Swelling over the left side of the face	Enucleation	2 years	Absent
Foroughi <i>et al.</i> , 2015	34/female	Intraoral swelling	Hemimaxillectomy	9 years	Present
Gruber <i>et al.</i> , 2019	26/male	Swelling over the right side of the face	Enucleation	5 years	Absent
De Arruda <i>et al.</i> , 2020	24/female	Pain over the left posterior tooth region	Enucleation	15 years	Present
Present case	33/male	Epiphora and nasal blockage	Conservative	8 years	Present

Twelve articles were obtained which fit our criteria [Table 1]. Data of 14 patients were noted. The male: female ratio found was 1:1.8. Of 14 cases, only 2 cases (14.28%) had presenting symptoms of nasal stuffiness and proptosis of the eye, whereas 9 cases (64.3%) had a chief complaint of swelling. One case (7.14%) was an accidental finding on routine radiograph. Pain was the first symptom in only one case (7.14%). Epiphora was observed in only one case (7.14%), that is, our case. In 35.7%, maxillectomy was performed. The follow-up period ranged from 18 months to 17 years. In 28.6% of cases, recurrence was observed.

In 2020, De Arruda *et al.* published the case of a 45-year-old female with CEOT of the left maxilla. First diagnosed in 2004, the patient was operated and recurrence was noted in 2009. The patient refused extensive surgery, and later, in 2014, malignant transformation of the lesion was noted and she was treated accordingly.^[17] Hence, malignant transformation of such lesions is also possible, but it is rare. However, recurrence and aggressiveness of lesion is also dependent on histologic type.^[18-20]

Hence, we infer that following the set protocols for treatment and maintaining regular follow-up can result in betterment of the patient. Having said that, further studies to decide adequate duration of follow-up are encouraged.

Ethical approval

Ethical approval was not required and patient identifying knowledge was not presented in this report. Ethical approval is waived.

Consent

The written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review on request.

Declaration of patient consent

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

Author's contribution

All authors contributed significantly and in agreement with the content of the article. All authors have read and approved the final version of the manuscript.

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Conflicts of interest

There are no conflicts of interest.

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