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Case report

Calcifying Epithelial Odontogenic Tumor of the Maxilla (Pindborg Tumor)

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A B S T R A C T

Calcifying epithelial odontogenic tumor (CEOT), or the Pindborg tumor, is very rare neoplasm, which accounts up to 1% of all odontogenic tumors. These tumors involve mandible almost twice as common as the maxillary bone, mostly in the premolar and molar region and present at first with local swelling. There is no gender predilection and the tumor usually appears between 2nd and 6th decade of life. We report the case of a 36-year-old male patient with a Pindborg tumor in the maxillary region on the right side, also involving the adjacent maxillary sinus, with destroying of the local anatomical structures. Complete surgical excision of the tumor has been performed and four years after surgical treatment, there is no sign of recurrence.

Key words: *calcifying epithelial odontogenic tumor (Pindborg tumor), maxilla*

Introduction

Calcifying epithelial odontogenic tumor (CEOT) also known as Pindborg tumor is very rare, locally invasive neoplasm, characterized by amyloid-like material that may become calcified[1]. It accounts for less than 1% of all odontogenic tumors, without gender predilection and usually clinically presents as slow-growing, painless mass, most often envolving premolar and molar region[2–4]. The most common and prominent clinical symptom of this tumor is swelling with slowly growing mass causing mechanical effects[1]. The mandible is affected twice as often as maxilla[3], with about two thirds beeing found in the posterior parts of the mandible, most often in connection with the unerrupted mandibular third molar[4]. Patients between 2nd and 6th decade, with a mean age of 40 years are usually affected[2]. The vast majority of cases are intraosseous, with approximately 6% arising in extraosseous locations[2]. On radiological images the tumor usually shows a multilocular, or less common, an unilocular radiolucency, sometimes with calcified structures inside the lesion, most often around the crown of the impacted (unerupted) tooth[4], up to 60% of cases[3]. The third mandibular molar is usually involved in such cases[4]. The two most common appearances of CEOT on radiological images are of pericoronal lucency and of lucent areas with diffuse opacities[5]. According to Kaplan et al., their research based on 67 cases of CEOT reveals that the mixed radiolucent-radiopaque pattern was the most frequent one with 65%, followed by 32% of radiolucent cases and just 3% of radiopaque cases[6]. In the same study, the unilocular type was more frequent in the maxilla than in the mandible[6].

The clear cell variant of CEOT is mainly composed of clear cells, which can be often very similar to other clear cell tumors. This variant is often associated with recurrence, together with incompletely excised cases[3].

Case Report

A 36-year-old Caucasian male patient was admitted to our Department of Maxillofacial Surgery because of painless swelling, which lasted over 3 months in the region of hard palate and buccal aspect of maxilla on the right side. All radiographic findings, including AP cranial radiography, panoramic radiography and computed tomography, showed a tumorous mass of the right maxillary sinus destroying and resorbing the bone of the lateral wall of the nasal cavity with disrupted structure of the ipsilateral upper jaw, affecting the adjacent teeth and intact lower floor of the orbital space (Figure 1a, b and c). The last two loose molar teeth (17 and 18) were extracted

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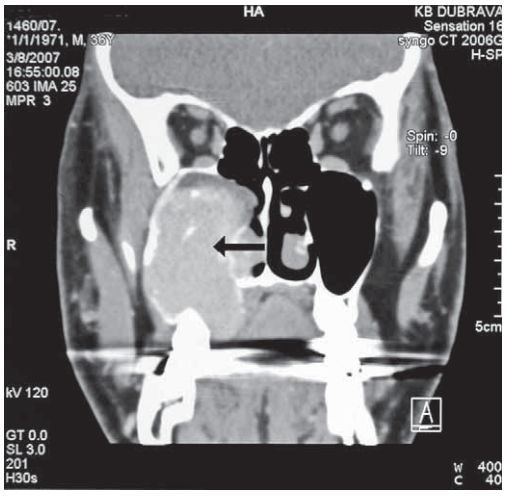

Fig. 1a. CT-scan showing tumor mass in the right maxillar sinus (frontal view).

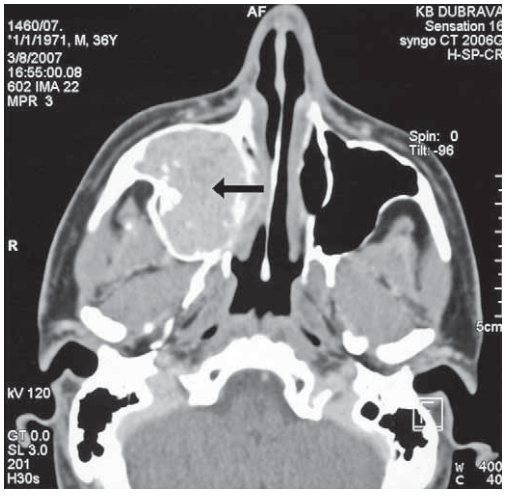

Fig. 1b. CT-scan showing tumor mass in the right maxillar sinus (horizontal view).

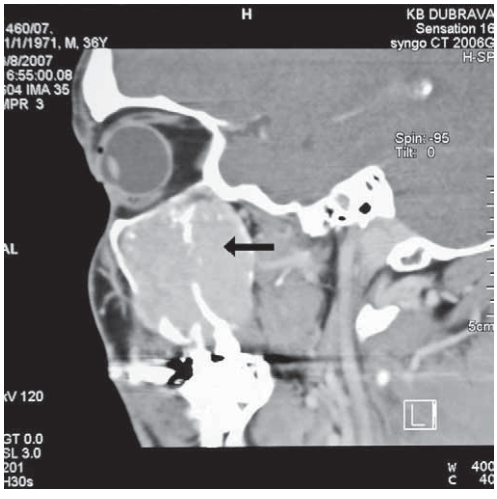

Fig. 1c. CT-scan showing tumor mass in the right maxillar sinus (sagital view).

and the biopsy was performed which confirmed the diagnosis of calcifying epithelial odontogenic tumor. Surgical treatment included transoral enucleation of the tumorous mass with curretage. The tumorous tissue was very soft and vasculated. After tamponade of the sinus with iodoform band, the same was removed 5 days after surgery and the stiches were removed 12 days after operation. So far, four years after surgical removal, there is no sign of recurrence, both clinically and radiologically.

The tumor volume was about 15 cm^3 and histologically it was composed of small islands and sheets of polygonal cells with abundant eosinophilic cytoplasm, clear cell borders and noticeable intercellular bridges (Figure 2a, b and c). Nuclei were pleomorphic, without mitotic figures. Eosinophilic, homogeneous hyaline material, within and around tumor cells, was proven to be amyloid-like, because when stained with Congo red, demonstrated the classic bright green birefringence viewed with polarized light under microscope (Figure 2d). This amyloid-like material was often centrally calcified in the form of Liesegang concentric laminar rings. The entire lesion showed typical cribriform appearance with islands of tumor cells enclosing the previous described hyaline, amyloid-like material.

Discussion and Conclusion

In 1958 Pindborg described these relatively rare lesions[7]. There are around 200 cases described in the literature to the present. Long-term follow up is essential because there is a recurrence risk if the tumor was incompletely resected and in particular with the clear cell variant[2], which is locally more aggressive. The clear cell variant carries the least favourable prognosis[8]. The recurrence rate of 15% has been reported in this variant, with highest frequency beeing in the lesions treated with curretage[4]. According to the current literature, the prognosis appears to be good, although a high mitotic activity tends to be connected with malignant behaviour[1].

Malignant transformation and metastatic spread to cervical lymph nodes has also been described in the literature, however in only few cases to the present[9–12]. Kawano et al. described a case of CEOT of the mandible with malignant transformation and development of metastases of the lung after repeated local recurrence[10]. These recurrent tumors had malignant features, like increased nuclear pleomorphism, increased mitotic activity together with increased proliferative activity measured with MIB-1 and vascular invasion[12]. Chemotherapy with platinum derivatives has been performed for the pulmonary metastases in this case, which showed very good response, suggesting use of chemotherapeutic agents in such rare cases[12]. On the other hand, one has to ask himself about malignant transformation in such rare cases and if it was pure CEOT in the first place, considering the rare occurence of benign cases.

According to the literature, our case is one of rare CEOT-s described in the maxillary sinus[13–17], adding this way to the series of reported CEOT-s. A case of CEOT

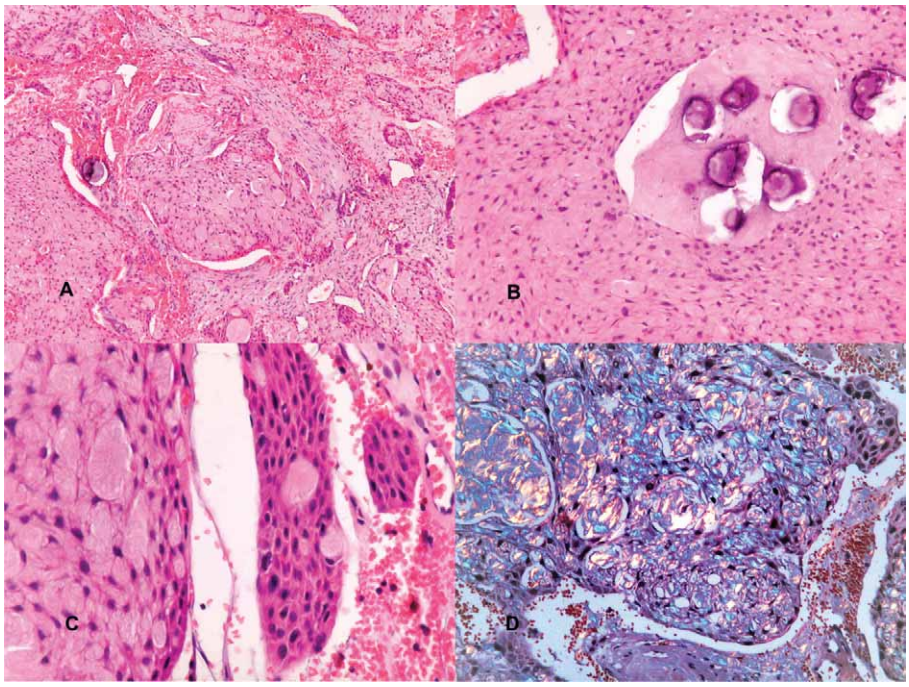

Fig. 2. Hemalaun-eosin stain. A – 100x magnification, B – 200x magnification, C – 400x magnification, D – polarized light (200x).

with intracranial extension has also been reported[14] and sometimes it can even present itself with abnormal eye signs[11]. A case of CEOT, which presented itself with nasal obstruction as the first presenting symptom was described by Bousdras VA et al.[15].

Differential diagnosis includes adenomatoid odontogenous tumor and other odontogenous tumors, especially the group with radioopaque particles[18]. The sqamous cell carcinoma should also be considered because of cytological pleomorphism and intercellular bridges[2]. The clear cell variant of CEOT can be mistaken for clear cell odontogenic carcinoma, some metastatic carcinoma, like renal cell carcinoma, and some salivary gland tumors like mucoepidermoid carcinoma and acinic cell carcinoma[2].

The method of treatment varies and depends mostly on the size and anatomic location of the lesion. Franklin and Pindborg advise to remove a rim of normal tissue as safe margins together with complete local resection[19]. Enucleation with curretage is not recomended because of local recidive possibility[19]. They advise against a radical surgical approach of wide resection such as hemimaxillectomy which used to be performed in the past[19]. However, sometimes it's impossible to avoid enucleation with curretage, like in our case, because of previously mentioned anatomic location of the tumor. In such cases surgical treatment depends very much on tumor size and its localization.

As previously mentioned, this case is one of the rare occurencies of CEOT in the maxillary sinus, which are documented in the literature. In this way we would like to contribute the series of this tumor occuring in the maxillar region.

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KALCIFICIRAJUĆI EPITELIJALNI ODONTOGENI TUMOR

S A Ž E T A K

Kalcificirajući eitelijalni odontogeni tumor ili Pindborgov tumor je vrlo rijetka novotvorina, koja čini oko 10% svih odontogenih tumora. Oni zahvaćaju mandibulu gotovo dva puta češće nego maksilu., najčešće u području premolara i molara, a prvo se manifestiraju kao ograničena oteklina. Tumor se javlja obično između drugog i šestog desetljeća života. Mi prezentiramo bolesnika straog 36 godina sa Pindborgovim tumorom desne maksilarne regije, koji zahvaća maksilarni sinus sa destrukcijom okolnih anatomskih struktura. Učinjena je kompletna kirurška ekscizija i nakon četiri godine i dalje nema znakova bolesti.