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Pindborg Tumor : A Surgical Case of Calcifying Epithelial Odontogenic Tumor Combined with Minute Amount of Adenomatoid Odontogenic Tumor Components

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Clinicopathological changes of calcifying epithelial odontogenic tumor (CEOT, Pindborg tumor) of a male case were described. A 67-year-old male who had tolerated his right buccal swelling over forty years noticed recently that the swelling turned painful. Clinical examination at Yatsushiro General Hospital revealed a tumor growing inside the right maxillary bone. On histopathological examinations of the tissues excised surgically by scraping out, the tumor was diagnosed as a Pindborg tumor. However, since the tumor histology contained a minute amount of adenomatoid odontogenic tumor (AOT), it meant a combined CEOT and AOT when expressed in detail. General and specific histologic features including above hybrid patterns, and the situation on literature of the present tumor are discussed.

Key words : Pindborg Tumor, Pathology

Introduction

Calcifying epithelial odontogenic tumor (CEOT, Pindborg tumor) is a rare odontogenic neoplasm first proposed by Pindborg[1,2] as a distinct entity in 1955, and subsequently in 1958 in detail. Franklin and Pindborg[4] reviewed the world literature including 113 reported cases of this tumor in 1976, and then, Pindborg and associates[21] reviewed again in 1991 on the literature after 1976 adding new report of a case. Also in Japan, reports of CEOT have been accumulated especially in recent years.[5,7,8,10,13,15–20] Thus, the total number of the cases reported is still increasing in the world.

The present report is that of a Pindborg tumor which proliferated chiefly inside the maxillary bone. Histology of the tumor mostly fulfilled the histologic criteria of CEOT. In addition, the detailed examination revealed that the tumor contained minute amount of adenomatoid odontogenic tumor (AOT) components. This paper is a description of the histopathological aspect of the tumor lesions examined by means of microscopic observations in the main.

Clinical History

A 67-year-old male noticed first his right buccal swelling over forty years ago. Although the swelling had enhanced very slowly during a long period, he had never consulted a doctor for medical treatment. However, since the swelling has become painful recently, he visited Rhinolaryngology Clinic of Yatsushiro General Hospital on August 23, 1993. On X-ray examination, malignant tumor of the upper jaw was suspected, and then, CT tomogram was taken for the exact

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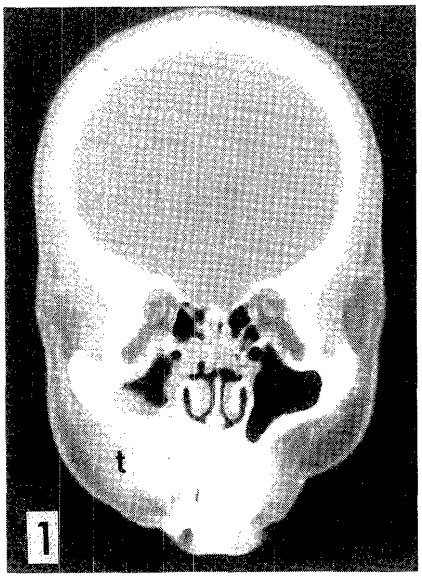


Fig. 1 CT tomogram of the patient's head and face involving the maxillary lesion. The right maxillary sinus is narrowed by compression of the tumor proliferating expansively inside the bone (t).

diagnosis on August 30. Tumor growth inside the right maxillary bone was confirmed, and the surgical excision by scraping out was performed on October 1. Since the excision of the tumor had been incomplete, reoperation was carried out on March 1, 1994. But, complete removal of the entire tumor seemed to be impossible at the second operation because of its complicated location. Radiotherapy has been employed thereafter against still remaining tumor tissues to this day.

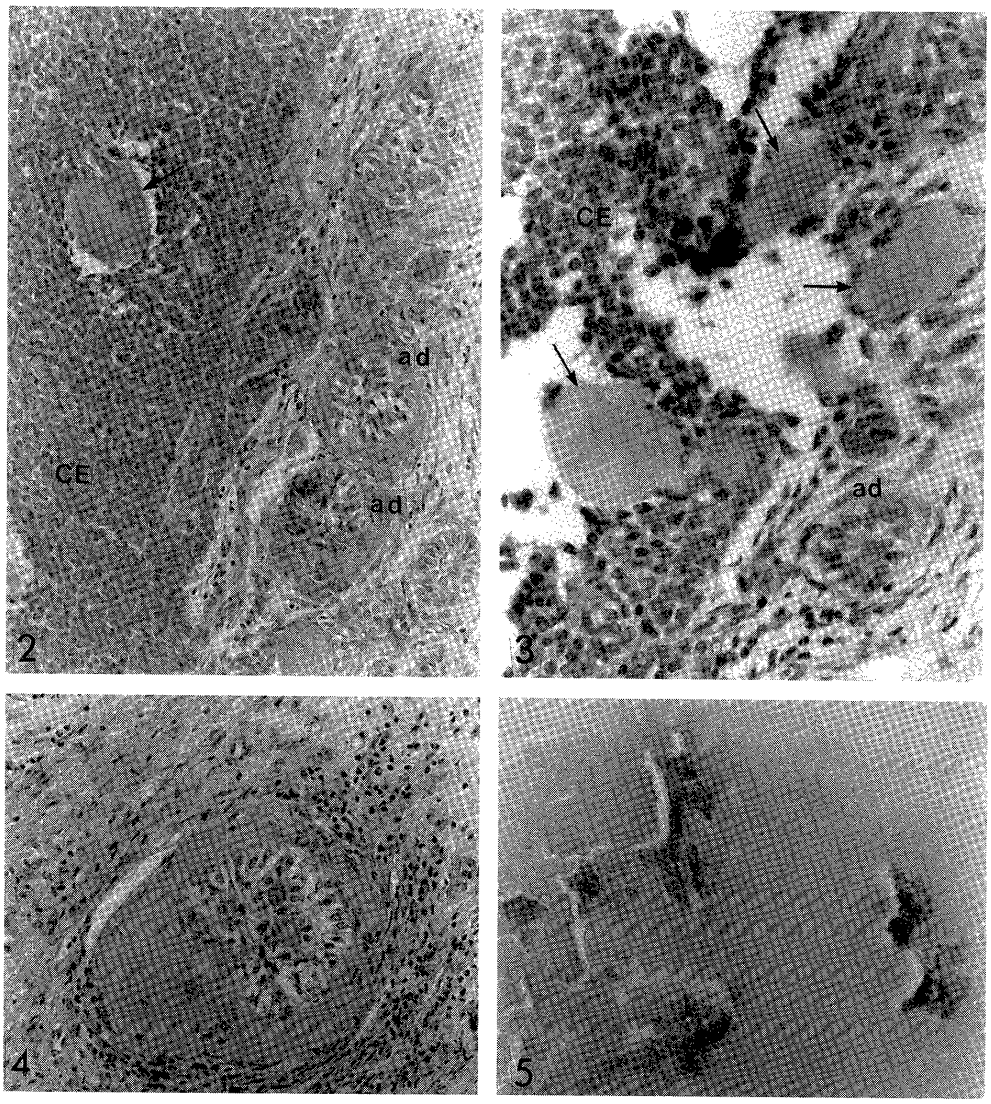
Histology of the tumor examined in this study was derived from the tissues obtained at the operations.

Histological Findings

Since the tumor of the present case grew inside the maxillary bone rather than in maxillary sinus, tissues of the tumor mass were scraped out as many piecemeal fragments in the process of surgical excision. So that, it was impossible to make a panoramic survey on wider tumorous lesions under the microscope. However, since the fundamental histopathologic features were maintained in these fragmented tissues, the authors could obatin sufficient informations in respect of microscopic details of the neoplastic properties.

The tumor histology revealed solid, parenchymatous mass instead of cystic configulation. The tissues were composed basically of epithelial parenchyma and vasculofibrous stroma. The first type epithelial cell nests consisted of broad or narrow sheets of large polyhedral cells arranging with epithelial continuity and some nuclear pleomorphism (Figs. 2 & 3). However, the intercellular bridges between the cells were difficult to recognize in this case. A distinctive feature relating to these epithelial cell sheets was the presence of an eosinophilic amorphous and homogeneous substance within cytoplasms of some of the tumor cells. This eosinophilic substance could be seen also extracellularly arround the cells as amorphous and acellular globules (Figs. 2 & 3). In addition, above amorphous substance grew into large deposits of irregular shape probably by accumulation, and was found here and there in the vasculofibrous stroma independently of the cell sheets (Fig. 8).

These eosinophilic deposits showed histochemically variable Congo red-positive and PAS-positive properties (Figs. 7 & 8). Especially, positive results on polalized light microscopic findings for Congo red-stained sections and on fluolescent microscopic findings for thioflavin T-processed sections suggested strongly that the deposits were of amyloid-like nature. On the other hand, calcifications were only focally detectable on the amorphous materials (Fig. 5). Non-calcifying variant of CEOT is reported also in literature.[6] Among the immunohistochemical techniques applied for staining the tumor cells, only cytokeratin antigen stained intensely on the tumor



Figs. 2 & 3 Epithelial cell sheets consisting of numerous polyhedral cells are there (CE) as the main tumor components. Note the round or oval eosinophilic amorphous materials among the tumor cells (arrows). Near the polyhedral cell sheets, a different type of epithelial cells with adenomatoid or glandular fashions can be seen (ad). hematoxylin-eosin (H. E.) stain. direct magnification 200x (Fig. 2) 400x (Fig. 3).

Fig. 4 One of the adenomatoid, glandular structures. Eosinophilic droplets are discernible within the glandular lumen. The epithelial cells are of columnar shape and of clear cytoplasms. H. E. 200x

Fig. 5 Irregular and spotty calcification in the center of a large homogeneous deposit. H. E. 400x

cell-sheets and single tumor cells (Fig. 6). But, trials for other antigens such as EMA, vimentin and S-100 protein gave all negative or unreliable results.

The second type epithelial elements were the presence of cell nests of adenomatoid or glandular fashion consisting of columnar epithelial cells (Figs. 2, 3 & 4). The polyhedral cell sheets described above and these adenomatoid cell nests appeared occasionally in close locations to each other. In some adenomatoid nests, eosinophilic droplets could be observed within the glandular lumens (Fig. 4). However, no transitional appearance between the polyhedral cell sheets and the adenomatoid

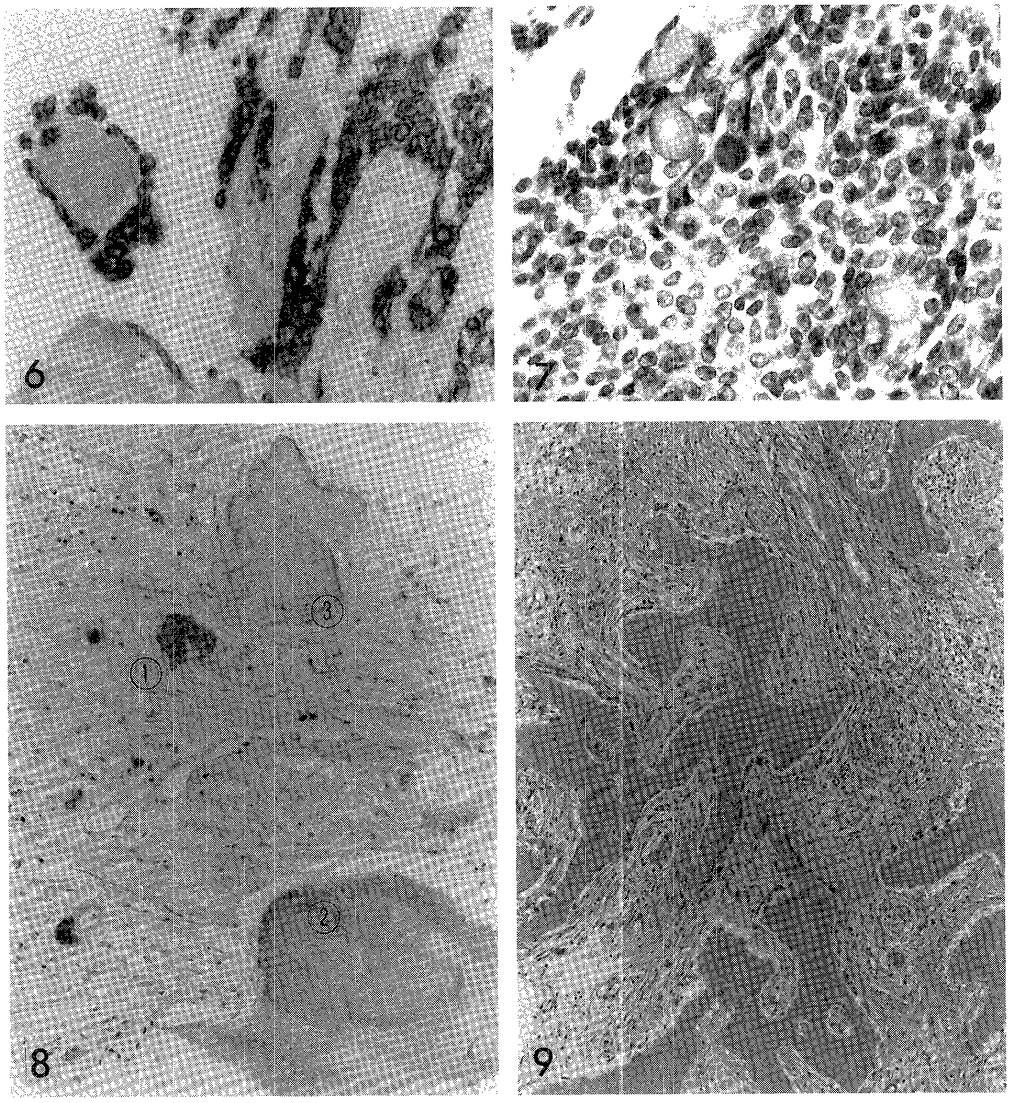


Fig. 6 Immunohistochemistry for cytokeratin antigen. The polyhedral cells stained intensely in a fashion rimmed at cell border.

Fig. 7 Congo red-stain for oval homogeneous materials among the polyhedral cells. The deposit substances are well stained in amyloid-like appearances. 400x

Fig. 8 Homogeneous materials are variably stained with PAS reaction. As is seen in this micrograph, they stain intensely positive (1), slightly positive (2) and completely negative (3). 200x

Fig. 9 Reactively hyperplastic trabeculae of the maxillary bone. The trabeculae were rimmed by osteoblastic cell arrangements. H. E. 200x

cell nests was discernible on the microscopic fields.

From whole histological results mentioned above, we understand that this tumor is of calcifying epithelial odontogenic tumor (Pindborg tumor) as the main constituents, but containing small amount of adenomatoid odontogenic tumor elements.

The vasculofibrous stroma extended widely supporting the epithelial cell groups. It contained, as described, deposits of the amorphous material large or small, being scattered in random fashions. Trabeculae of the maxillary bone that had been scraped out together with the neoplastic tissues at operation appeared much hyperplastic than in normal, accompanying osteoblastic cell-arrangement at the rims of each trabecle (Fig. 9).

Discussion

Calcifying epitherial odontogenic tumor (CEOT, Pindborg tumor) proposed first by Pindborg[1] in 1955 has been widely accepted as a distinct entity tumor of odontogenic origin, with accumulation of many reported cases.

In 1976, Franklin and Pindborg[4] listed the criteria for histological diagnosis of CEOT, that is, (1) sheets of polyhedral cells, (2) nuclear pleomorphism, (3) amyloid material, (4) calcification of amyloid material, (5) well-defined cell borders, (6) intercellular bridges. With regard to the case presented here, the authors believe that the histology of our case can mostly fulfil above criteria except the last two items. However, we presume that the unclearness of cell borders and intercellular bridges might be attributable to poor preservation of the tissues. Because cell borders of the tumor cells could become clearly recongnizable on cytokeratin-stained tissue sections (Fig. 6). Among these criterion items, characteristic histology of (1) to (4), especially the presence of amyloid-like material confirmed by Congo red and thioflavin T would be the final decider for diagnosis of CEOT in the present case.

Aside from above basic criteria, the present case proposed clinically and pathologically some problems to be considered. The patient had been aware of the right buccal swelling over forty years prior to the current visit. Although the exact time of occurrence of the present tumor remained unknown, such a long-standing swelling made us presume some oncogenic relationships with the current tumor growth. With reference to the fact that the age predilection of occurrence of Pindborg tumor is in middle ages of about 40 years of age,[11,21-23] the tumor of the present case seemed to have arisen at rather older age. But it is also the fact that there had been long period unexamined in this patient before diagnosis. Literature describes no sex predilection in Pindborg tumor.

Major case reports in the past described that the occurrence of CEOT had been associated with the impacted teeth. Unfortunately, our case has not been confirmed on the presence or absence of an impacted tooth. Regarding the locations of occurrence of CEOT, Franklin and Pindborg[4] indicated, on the review of 113 cases, a predilection in the mandible over the maxilla. However, many reported cases are there in which the tumor originated in the maxilla.[7,10,14-16,20,22] Already in 1982, Kaji et al.[7] counted 12 reports of CEOT in Japan in which the tumor origin in the upper jaw had been confirmed.

As stated before, we believe that the histology of our patient's neoplasm can mostly fulfil Franklin and Pindborg's criteria for histological diagnosis of CEOT, though lacking the formation of Liesegang's ring with abundant calcifications on the eosinophilic homogeneous deposits. On the other hand, the second type of epithelial proliferation consisting of adenomatoid or gladular

configulation suggests the coexistence CEOT and adenomatoid odontogenic tumor (AOT) in the same tumor. Accordingly, the histology of this tumor would imply a combined CEOT and AOT. However, CEOT as the major neoplastic components much exceeded AOT in whole amount of the tumor lesion. Also in literature, the combined type tumor of CEOT and AOT has been reported as uncommon cases by Damm et al.[9] (1983), Bingham et al.[12] (1986). And Hicks et al.[23] summarized in 1993 this hybrid type neoplasm from clinicopathological viewpoint. Whereas they explained that in these hybrid cases AOT primarily exceeds CEOT, our present case manifested a reverse condition. Also age at detection differed from younger age they stated. Therefore, our case tended obviously to reveal the general conditions of CEOT. Bingham et al.[12] introduced the respective criteria of AOT and CEOT, referring to those by Courtney & Kerr (for AOT) and Franklin & Pindborg (for CEOT). However, since the tumor of the present case included only small amount of AOT components within the major CEOT proliferation, we could not obtain sufficient informations to refer to the AOT criteria. Diagnosis of combined AOT depended chiefly on the histologic configulation of adenomatoid or glandular fashion and of the presence of eosinophilic droplets inside the glandular lumen. Other special findings to fulfil the criterion items such as swirling spindle cells or latticework epithelial proliferation etc. were difficult to find out. In conclusion, we recongnize that this case is fundamentally a calcifying epithelial odontogenic tumor (CEOT, Pindborg tumor), but including insufficient adenomatoid odontogenic tumor (AOT) elements.

As is seen in the literature, most patients with Pindborg tumor visit dental department or clinic. Because the majority of CEOT proliferate in oral regions, and the tumor is associated frequently with impacted teeth or odontogenic cysts.[3] We consider that CEOT proliferating as solid tumor in the midst of the maxillary bone as this case would belong to an unusual and rare occasion. Cameron et al.[22] made a report of CEOT arising within the maxillary sinus as an extremely rare case. In our own case, although the right maxillary sinus was narrowed by compression of the tumor mass, it maintained original radiolucency on CT tomogram being exempted from tumor invasion.

CEOT has been accepted as basically benign tumor, though rare existence of CEOT exhibiting some malignant characters was suggested in the literature.[11] Recurrence has also been recongnized to be low in frequency. However, the present case recurred once after the first excision, and the possibility of further recurrence still remains. Because, the difficulty of complete removal of the tumor is the problem in this case.

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