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Calcifying Epithelial Odontogenic Tumour of the Maxilla: A Case Report with Respect to Immunohistochemical Findings

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Abstract. *Calcifying epithelial odontogenic tumour (Pindborg tumour) is a rare benign neoplasm with a poorly understood histogenesis. This report describes a single case of a maxillary calcifying epithelial odontogenic tumour including immunohistochemical analysis. The vast majority of tumour cells were positive for CK5/6 and p63. Furthermore, basal cells also displayed moderate reaction for vimentin and strong membranous positivity for podoplanin. Interestingly, the tumour invaded the dental pulp of the partially disintegrated tooth root. While the tumour showed focal connection with the superficial gingival epithelium and revealed intercellular bridges, the findings of this case study seem to support the suggestion of an epithelial origin of a calcifying epithelial odontogenic tumour derived from the dental lamina.*

In 1955 Jens, J. Pindborg described an epithelial tumour of odontogenic origin with characteristic regions of calcifications (1, 2). This tumour was accepted as an entity by the World Health Organization (WHO) classification of odontogenic tumours in 1971. For the classification of odontogenic tumours, the WHO applies the method of specifying a tumour entity according to the evidence for derivation from odontogenic epithelia and/or odontogenic ectomesenchyme. Although it has been classified as a benign tumour of odontogenic origin, its true histogenesis remains undetermined. The tumour has been denominated as "calcifying epithelial odontogenic tumour" (CEOT) and had long been described under the eponym 'Pindborg tumour' (3, 4).

CEOT (ICD-O code 9340/0) is an invasively growing benign neoplasm characterized by the formation of homogenous eosinophilic material. These structures are amyloid-like proteins (5-7), possibly remnants of intermediate filaments (8-12), that might calcify during the time that elapses for tumour growth and create the characteristic extracellular ossification (3).

The predominant findings in CEOT are a slowly growing, painless mass in the alveolar process and adjacent soft tissues (6). Nevertheless, a malignant transformation of CEOT is possible and has occasionally been reported (13-15).

The CEOT is a tumour arising most frequently inside the bone and has been found to be associated with retained teeth in approximately 50% (6). The lower jaw is about twice as frequently affected than the maxilla (3, 6, 16). The molar region is affected predominantly (3, 6, 16), followed by the premolar region. Other regions of the jaws are substantially less frequently affected (6, 17, 18). The extraosseous variant is exceedingly rare, preferentially found in anterior parts of the oral cavity (18-20). Most patients are between 20 to 40 years of age at the time of diagnosis. This report adds some clinical and morphological findings to the literature on CEOT.

Case Report

The medical history of the patient's therapy has been detailed elsewhere (21). In brief, prior to the first consultation in the outpatient clinic (University Medical Center Hamburg-Eppendorf), a 49-year-old man had noticed a painless loosening of his maxillary frontal teeth and an increasing swelling of the anterior maxillary alveolar process and palatinal mucosa for approximately six months. An orthopantomogram was acquired after the first investigation and the osteolysis was tentatively assessed as a cyst. Physical investigation revealed an extensive, firm and resilient swelling of the anterior maxilla. This region was indolent on digital pressure. Initial investigation showed an elevation of the nasal base and the teeth reacted rapidly on cold stimulus and were markedly loosened.

Radiological examination. In an orthopantomogram a homogeneous, sharply demarcated translucency was projected on the anterior maxilla. The bony parts of the nasal floor appeared disintegrated and elevated. The maxillary sinuses, the

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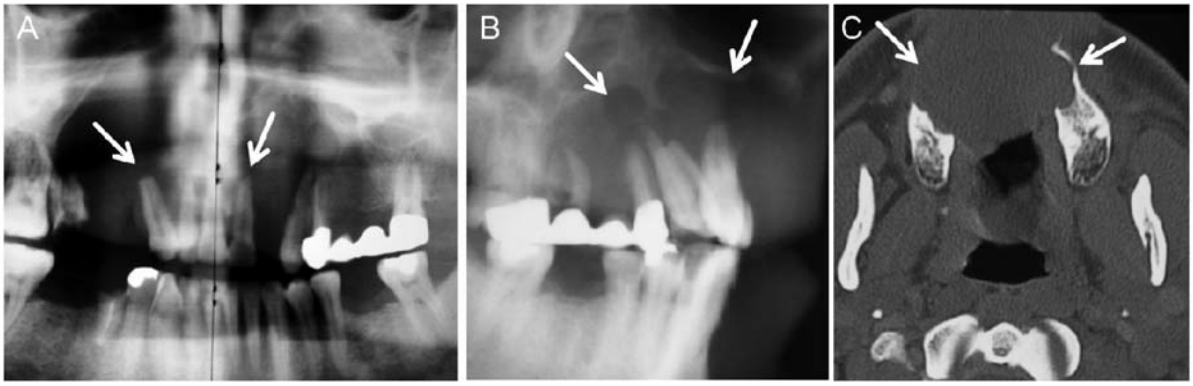

Figure 1. *Calcifying epithelial odontogenic tumour: radiographic findings. (A) Detail of orthopantomography of the patient at the time of admission: a roundish osteolytic lesion is depicted in the anterior maxilla. The apices of some frontal roots (no. 13, 21) seem to be partially disintegrated (arrows). The lesion extends to the maxillary sinus. The midfacial bones in contiguity to the lesion show a sclerotic line that is almost continuously expressed. (B) Detail of lateral skull view showing the osteolytic lesion of the anterior maxilla. In particular, the alveolar process is extensively destroyed (arrows). The disintegrated apices of the teeth appear to have no osseous embedding. Round satellite lesions are seen in the cranial portion of the lesion. (C) Detail of an axial CT scan showing a wide, expansively growing osseous lesion with loss of osseous margins in the frontal maxilla (arrows).*

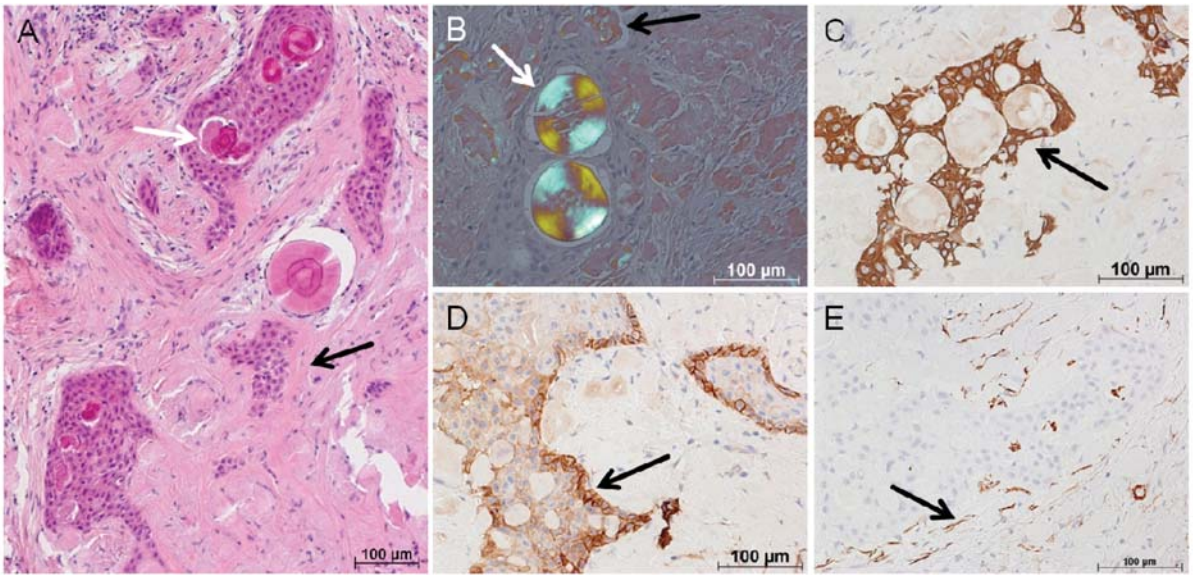

Figure 2. *Calcifying epithelial odontogenic tumour: histopathological findings. (A) Low power view showing islands of neoplastic polyhedral epithelia and fibroblastic stroma. Focal mineralisations (white arrow) are apparent within epithelial nests. Furthermore, homogenous eosinophilic material (black arrow) can be seen in both the stroma and within the epithelial structure (staining: haematoxylin-eosin, original magnification ×50). (B) Under polarised light, eosinophilic material (black arrow) and psammomatous bodies (white arrow) show a characteristic red colour, with apple-green birefringence (white arrow) (stain: Congo red, viewing under polarised light, original magnification: ×100). Using immunohistochemistry, tumour cells show positive reaction for CK5/6 (arrow) (C), basal cells also show membranous expression of podoplanin (arrow) (D), and the reaction for α-SMA highlights scattered positive stromal cells (arrow) (E); immunohistochemistry, original magnification: ×100.*

anterior nasal spine and the adjacent soft tissues also appeared to be affected. The roots of teeth no. 16, 13-21, 23 and 24 were lying in the area of radiological translucency of the maxilla and showed some disintegration of the apices (Figures 1A, B).

In a computed tomography (CT) scan, a solid tumour of 4×4×4 cm^3 volume was depicted, extending from the nasal cavity into the hard palate and both maxillary sinuses. The nasal septum was deviated and had been partially destroyed. The neoplasm showed a displacing growth pattern. However, it was not possible to make a distinction between the tumour and anterior vestibule. No structures isointense to bone were seen inside the lesion (Figure 1C).

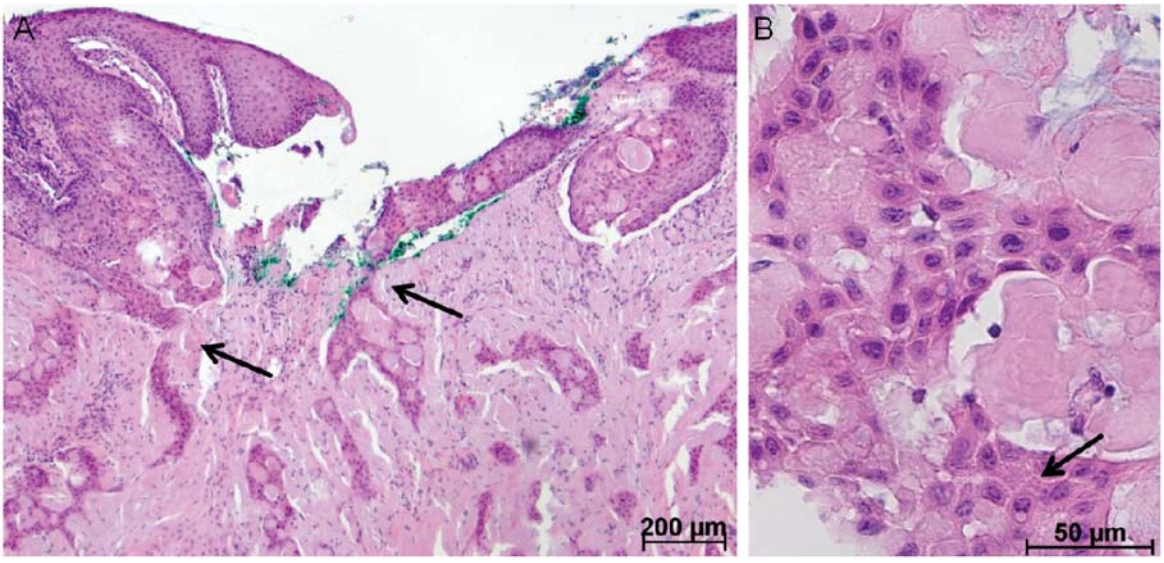

Figure 3. *Calcifying epithelial odontogenic tumour (proximity of the tumour to the superficial squamous epithelium). (A) The tumour displays focal connection (arrow) with the superficial squamous epithelium of the gingiva (arrows) (stain: haematoxylin-eosin, original magnification: ×50). (B) Under high power magnification, the epithelial tumour cells appear to be polyhedral with centrally located oval nuclei and eosinophilic cytoplasm. Between the tumour cells, focal intercellular bridges and desmosomes (arrow) are apparent (stain: haematoxylin-eosin, original magnification: ×400).*

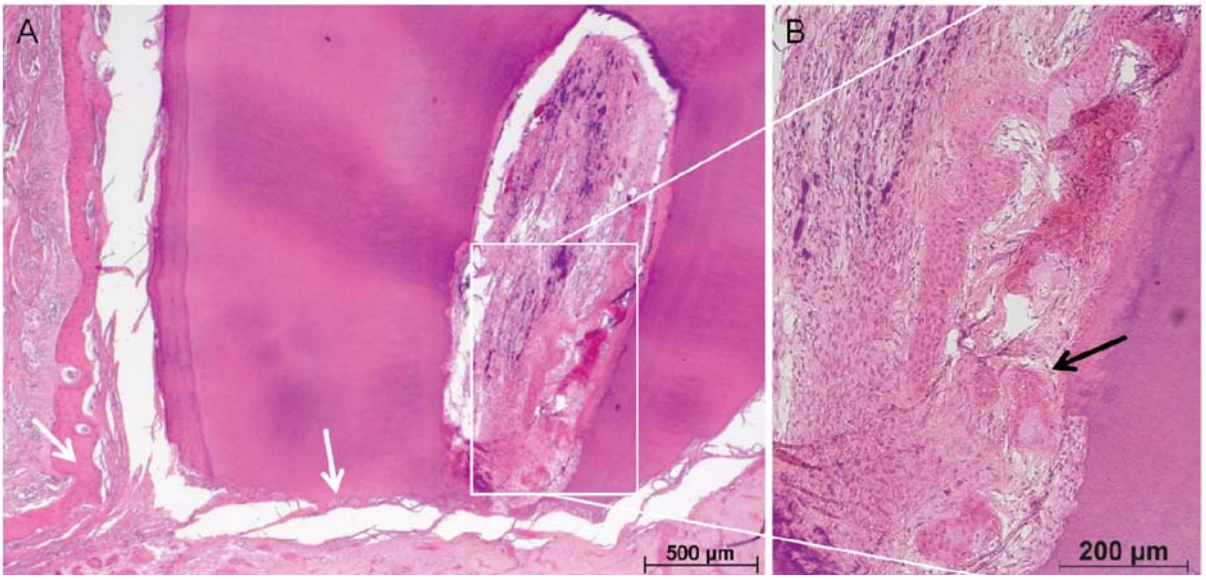

Figure 4. *Calcifying epithelial odontogenic tumour (focal tumour invasion of a dental pulp adjacent to the partial disintegrated tooth root). (A) A deep portion of a vital tooth root had disintegrated adjacent to the tumour tissue (white arrow). On the left, the alveolar bone is apparent (stain: haematoxylin-eosin, original magnification: ×25). (B) Under high-power magnification, the lower portion of the dental pulp can also be seen to be infiltrated by the tumour (black arrow) (stain: haematoxylin-eosin, original magnification: ×100).*

Surgical therapy. Under general anaesthetic, a vestibular incision of the mucosa was performed and the tumour was detached from the covering soft tissues. The tumour was resected *in toto* keeping a safe distance between the line of resection and the estimated border of the lesion. The cavity was temporarily filled with gauze and re-epithelialised rapidly. Oral rehabilitation of the patient was achieved with an obturator prosthesis. No local recurrence was observed during a five-year follow-up period after which the patient was lost to follow-up.

Histopathology. Intraoperatively, a 2×1×0.8 cm native biopsy specimen was first excised for frozen section diagnosis. Based on the microscopic finding of band- and nest-like arranged tumour cells embedded in a broad acellular homogenous matrix, a tentative diagnosis was suspected of benign odontogenic tumour, most probably CEOT. Subsequently, partial anterior maxillary excision of a complete neoplastic lesion was performed. The surgical specimen was fixed in formalin immediately after the excision.

Macroscopically, the specimen from the partial anterior maxillary resection with three teeth displayed a tumoural mass with a diameter of 4 cm. The lesion was cut to parallel thin slices by means of a water-cooled, diamond-coated band saw and embedded in paraffin wax. Prior to embedding, the tooth tissue was decalcified using formic acid. Histopathological sections were stained with haematoxylin-eosin, elastica van Gieson and Congo red staining methods. The slides were also analysed under polarised light. For immunohistochemical analysis, calretinin (clone: 18-0211, Zymed, San Francisco, USA, dilution: 1:100), CK5/6 (D5/16B4, Dako, Glostrup, Denmark, 1:100), CK20 (Ks20.8, Novocastra, Newcastle upon Tyne/UK, 1:50), CK19 (RCK, M0888, Dako, 1:50), CK18 (DC10, M7010, Dako, 1:25), CK7 (OV-TL 12/30, M7018, Dako 1:50), CD117 (c-kit, A4502, Dako, 1:400), p63 (A4A, M7247, Dako, 1:800), p53 (DO-1, Oncogene, Cambridge, MA, USA, 1:2000), Ki67 (MIB-1, M7240, Dako, 1400), vimentin (V9, M0725, Dako, 1:1000), CEA (II-7, M7072, Dako, 1:100), EMA (E29, M0613, Dako, 1:50), and a podoplanin antibody (D2-40, Signet, Dedham, MA, USA, 1:40), were used in sections of the paraffin-embedded, formalin-fixed material. The immunohistochemical analysis was validated through positive and negative controls (by omitting the primary antibody).

Microscopically, the tumour displayed solid epithelial islands within fibroblastic stroma with multifocal acellular eosinophilic material (Figure 2A). The latter displayed characteristic apple-green birefringence under polarised light, when stained with Congo red (Figure 2B). Moreover, scattered psammomatous inclusions and focal calcifications were apparent. Tumour cells were polyhedral and showed no nuclear atypia. Immunohistochemically, the majority of the tumour cells were positive for CK5/6 (Figure 2C) and p63. Interestingly, the vast majority of the basal epithelia showed strong membranous positivity for podoplanin (Figure 2D). Within the tumour stroma, several cells were strongly α-SMA-positive (Figure 2E). The proliferative activity of tumour cells was inconspicuous (Ki-67<2%). Other immunohistochemical reactions gave negative results. The tumour grew expansively and showed focal connection with the basal layers of the gingival epithelium (Figure 3A). Under higher power, intercellular bridges were apparent between the tumour cells (Figure 3B). Thorough histopathological investigation revealed that the tumour had invaded the alveolar bone (Figure 4A) and dental pulp of tooth no. 13 (Figure 4B). The resection margins were not invaded by the tumour.

Discussion

The reports on CEOT have predominantly been based on case analyses. Reviews were provided by Pindborg and others (22-29). Franklin and Pindborg (6) collected the data of 35 patients with maxillary CEOT out of a total of 113 cases in 1976. It is noteworthy that two of these 35 patients had extraosseous manifestations. Although the molar region was predominantly affected (17 cases, 48.5%), four CEOT developed in the anterior maxilla. Tumour resection with safety distance allowed a one-stage therapy in the majority of patients. However, local recurrences were rarely reported and should be expected following curettage or incomplete resection of the tumour.

Kaplan *et al*. (30) analysed the radiologic profile of CEOT. They confirmed the numerical predominance of mandibular CEOT (74%) and the molar region as the main site of affection (69%). CEOT appeared predominantly as unilocular lesions (58%). This phenotype was seen preferentially in the maxilla and appeared similar to ameloblastoma. Indeed, the maxillary ameloblastoma is the most frequently unilocular lesion (31). A distinct osseous border of the lesion was seen in only 20% and was not identifiable in 59% of the cases. However, a diffuse invasion into the adjacent structures was rarely seen (21%). The blurry outline of the tumour increased markedly with a maximum diameter exceeding 3 cm. This radiographic feature was substantiated in the current case. Kaplan *et al*. (30) emphasised the finding of disintegrated roots in CEOT to be rare (4%) compared to ameloblastoma (81%) and recommended this radiological finding as a valuable tool to establish the differential diagnosis. However, in the present case, root resorptions and even invasion of the dental pulp by CEOT was evident. These findings confirm that the cause of disintegration of dental roots needs to be clarified and does not predict the histological diagnosis (28). Furthermore, Cross *et al*. (22) performed a comparison of CT and magnetic resonance imaging (MRI) scans in a case of an extensive CEOT. They reported that CT scans visualised the distinct borders of the tumour and also the fine trabecular network inside the lesion that appeared isointense to the bone. Moreover, numerous isolated splitters were seen that were also isointense to the bone. The covering cortical bone appeared to be extremely thinned but not osteolytic. In MRI scans, no cystic lesions were detectable. This finding suggested that a distinction between CEOT and jaw cysts or ameloblastoma may be made. Confusion of CEOT with odontogenic cysts is possible, based on their similar radiographic appearance (6).

The histological aspects of CEOT are manifold (18, 23, 24). Polygonal epithelial cells with sharp cellular boundaries and wide intercellular bridging are cord- or nest-like arranged inside a fibrous stroma that may show degenerative alterations. Tumour cells are characteristically pleomorphic and may form giant cells, *i.e.* multinucleated cells with prominent nuclei. Differential diagnosis of a clear cell variant of CEOT from the bright cell odontogenic tumour may be difficult (25-28). Inside the tumour cell nests, lay rounded acidophilic-forming homogeneous masses that are often calcified and show Liesegang rings (16). The calcification might not be found in selected cases (29, 30). Fluorescence staining of the homogeneous material lying between the tumour cells with thioflavin T showed an amyloid-like reaction pattern (5). Recent immunohistochemical studies suggested that the amyloid-like deposits may represent remnants of epithelial intermediate filaments (7-9, 11, 32). This theory was in accordance with earlier speculations relating the amyloid of CEOT with the amelogenesis (6, 33). In the present case, immunohistochemical analysis demonstrated the reactivity of tumour cells for basal epithelial markers. Interestingly, the basal cells of the tumour proliferations expressed podoplanin in a manner that was observed in some other benign odontogenic lesions (34). The function of podoplanin in odontogenic epithelial cells is presently unknown, but it was also detected in tooth germ tissue of different developmental stages (34), cystic odontogenic lesions (34), and odontogenic tumours (34-36). The present case demonstrated myofibroblastic differentiated stromal cells which were focally entrapped within the epithelial nests. Several α-SMA-positive cells were also apparent in the vicinity of the basal epithelial cells, but not intraepithelial. Therefore, using light microscopy, the ultrastructural recognisable duality of the epithelial component of CEOT described by El Labban *et al*. (37) was not confirmed. The differential diagnosis of CEOT is the adenomatoid odontogenic tumour (38). In fact, transitions from adenomatoid odontogenic tumour to CEOT were reported (3, 39). Other odontogenic tumours may show a similar radiographic appearance (40), in particular, those forming radiopaque material. The formation of amyloid-like extracellular matrix in ameloblastic fibro-odontoma (41) may cause problems in deciding the diagnosis (6).

CEOT represents a very rare tumour. The histogenesis of CEOT is currently uncertain. First, Pindborg (1) suggested its origin from the reduced enamel organ of an unerupted tooth. Then Chaudhry *et al*. (42) emphasised the finding that the tumour cells exhibit morphological characteristics of squamous epithelium. Recently, Reichart and Philipsen (28) suggested the origin of CEOT in epithelial remnants of the dental lamina complex. The present findings of the focal connection of the tumour nests to the superficial gingival epithelium and of intercellular bridges seem to further support the latter suggestion.

Conclusion

This report described the case of a CEOT, located in the anterior maxilla. While the tumour showed focal connection to the superficial gingival epithelium and revealed intercellular bridges, the findings seem to support the suggestion of an epithelial origin of CEOT, derived from the dental lamina.

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