Odontogenic carcinoma with dentinoid

DANNY R. SAWYER, ALAGUMBA L. NWOKU, ADEYEMI MOSADOMI AND A. T. KEKERE-EKUN

Department of Oral and General Pathology Loyola University of Chicago School of Dentistry, and Departments of Oral and Maxillofacial Surgery and Oral Pathology and Biology, College of Medicine University of Lagos

Abstract - An unusual case of odontogenic carcinoma with dentinoid is presented and discussed. This case is the first reported to show this peculiar histologic picture. The lesion recurred following what was deemed adequate surgical excision from both the clinical and histologic viewpoints. Adequate therapy and follow-up for such lesions is thus stressed.

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Malignant neoplasms related to the odontogenic apparatus are rare. For example if epidermoid carcinomas arising from odontogenic cysts are considered, Gardner in a review of the literature from 1889 through 1967 found 25 cases which he felt were acceptable. Another 9 cases were reported in the English literature from 1968 through 1981. Elzay in his review of primary intraosseous carcinomas (PIOC), which excluded cases of central carcinomas arising in odontogenic cysts, found 12 acceptable cases. Likewise, the other odontogenic carcinomas such as the malignant ameloblastoma and the odontogenic sarcomas are quite rare.

The purpose of this report is to present a case of odontogenic carcinoma which has a very unusual histologic picture because of the presence of dentinoid associated with the neoplastic epithelium.

Case report

On 27 January 1981, a 14-year-old Nigerian male presented at the Dental Outpatient Clinic of Lagos University Teaching Hospital with the complaint of swelling and pain of the right maxilla of 3 months duration.

An intraoral examination revealed a fluctuant, non-tender swelling of the right maxillary molar region of about 3 cm in diameter. The overlying oral mucous membrane was of normal color and not ulcerated. With the exception of the third molar, all the maxillary teeth on the involved side were present clinically. The teeth were non-mobile and there was no tenderness upon percussion. The oral hygiene was described as fair. A right submandibular lymph node was palpable, non-tender and non-fixed.

Upon roentgenographic examination, a well-defined cystic lesion was noted in the
right maxilla. The lesion was radiolucent with patchy areas of increased radiodensity. The cyst-like lesion was felt to be associated with the unerupted third molar. The lesion was aspirated and a blood-stained "cystic" fluid was obtained, the volume of which was not recorded. A diagnosis of odontogenic cyst, dentigerous type, was made based on clinical and radiologic findings.

On 6 February, 1981, the patient was taken to surgery. Under general anesthesia, a buccal mucoperiosteal flap was raised and a window made in the buccal plate of the maxilla. The cystic lesion, which was associated with an unerupted tooth, was enucleated. The cavity was irrigated and then closed with black silk sutures.

The excised tissue mass, minus any associated tooth, was submitted to the Department of Oral Pathology and Biology for examination. On gross examination, the soft tissue mass consisted of a well-defined, well-encapsulated, thick-walled cystic lesion containing a definite lumen. The cystic lumen was approximately one-third the size of the lesion as a whole. The cystic contents consisted of a yellowish-brown fluid, and semi-solid material. Histologic sections showed a thick fibrous connective tissue wall deep inside of which there was a thick, almost papilliferous neoplastic mass of epithelium lining a cystic cavity (Fig. 1). In some areas the epithelium consisted of tall columnar cells at the periphery with some evidence of palisading and pleomorphic internal epithelial cells with both the peripheral and internal epithelial cells showing a high mitotic index (Fig. 2). Associated with the epithelial cells was a dentin-like (dentinoid) collagenous matrix. In some areas, a few cells appeared trapped in this matrix and in others, structures which appear to be ill-formed tubules were observed. The diagnosis was atypical malignant (epithelial) odontogenic cyst noting the presence of the dentin-like material. An attached note queried a malignant Gorlin cyst, Pindborg tumor or related lesion and reported to the surgeons that because the lesion was unusual, sections would be sent to other centers for their opinions.

The patient was followed in the outpatient clinic, briefly showing some resolution of the residual swelling, before being lost to follow-up. As the lesion was of interest to the Department of Oral Pathology and Biology, numerous attempts were made to contact the patient for a follow-up examination. Finally, in January 1982 the patient re-appeared at the outpatient clinic. Because of massive swelling extending from the lat-

Fig. 1. Photomicrograph showing the well encapsulated cystic lesion. (H&E; 30×).

Fig. 2. Photomicrograph demonstrating the neoplastic epithelium in association with dentinoid. (H&E; 160×).
eral incisor to the maxillary tuberosity area and the suspected recurrence of the lesion, the patient was readmitted to the ward on 21 January 1982. By this time, the premolars and right maxillary first and second molars on the involved side had become mobile although not tender. There was no palatal swelling and the oral mucosa was of normal color and not ulcerated. The radiologist's report noted that there was extension into the maxillary antrum.

Under general anesthesia, a buccal mucoperiosteal flap was raised. The teeth in the area of the lesion were extracted and a marginal resection of the right maxilla was performed including bone about 1 cm beyond the boundary of the lesion. Soft tissues were mobilized from the palate and cheek to effect primary closure of the wound.

For 18 months following this second surgical procedure there has been no recurrence of the lesion.

The surgical specimen was sent to the histopathology laboratory for examination. Histologically, the lesion appeared similar to the previously submitted biopsy with the exception being the lesional mass contained no cystic lumen.

Discussion

As the lesion appeared quite unusual on histologic section and did not seem to fit well into any of the specific entities described in odontogenic cyst and tumor classifications, sections were sent to 5 internationally known diagnostic pathologists and/or centers including the AFIP. The diagnoses suggested included: “consistent with atypical (? malignant) odontogenic epithelial neoplasm,” “malignant epithelial tumor of maxilla (? variant of Gorlin tumor, ? other odontogenic lesion),” “odontogenic carcinoma with dentinoid,” “atypical malignant odontogenic tumor probably a form of ameloblastoma,” and “malignant odontogenic cyst with dentin formation.”

The authors as well as each of the consultants who reviewed this case felt that the collagenous eosinophilic matrix seen in association with the odontogenic epithelium qualifies as dentinoid, if not atypical or dysplastic dentin. In accordance with the classification of dysplastic forms of dentin by Gardner & Farquhar, this collagenous matrix is considered dentinoid.

In the WHO classification of odontogenic carcinomas, this lesion would be placed in the category of “other carcinomas arising from odontogenic epithelium including those arising from odontogenic cysts.”

References


Address:
Danny R. Sawyer
Loyola University School of Dentistry
2160 South First Avenue
Maywood, IL 60153
USA