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CASE REPORT

Primary intraosseous odontogenic carcinoma with osteoid/dentinoid formation

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Primary intraosseous carcinoma/odontogenic carcinomas are extremely rare malignant odontogenic tumours that are thought to arise from residues of odontogenic epithelium. An unusual case of primary intraosseous carcinoma arising *de novo* in a previously unreported site of posterior maxilla is described. The tumour was characterized by sheets of pleomorphic round-to-ovoid cells and marked osteoid/dentinoid formation with foci of globular mineralization.

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Keywords: mineralization; odontogenic carcinoma; osteoid/dentinoid; PIOC *de novo*; posterior maxilla

Introduction

An 18-year-old female patient presented with a gradually progressive painful swelling on the right side of the face. The swelling was of 6 months duration.

On examination, the swelling was bony hard in consistency. Intraoral swelling was found on the gingival aspects of maxillary right first and second premolars, resulting in obliteration of the buccal vestibule. The teeth were uninvolved by caries and were in good periodontal condition. The overlying mucosa was intact.

Radiograph showed a well-defined radiolucency in relation to the teeth with multiple small radiopaque foci. Resorption of the roots of the teeth was also observed. A provisional diagnosis of odontogenic cyst was made. Past medical history was not significant. A detailed physical examination revealed no significant findings.

Fine needle aspiration cytology performed from the intraosseous mass yielded blood-tinged straw-coloured fluid. Cytopathology showed sheets of minimally pleomorphic and hyperchromatic round-to-ovoid cells. The features were suggestive of an epithelial lesion of odontogenic origin.

The 'cyst' was enucleated. The patient is being followed up regularly. One year and 4 months have elapsed and there has been no evidence of recurrence. Post-operative radiograph taken after 1 year showed normal bone healing (Fig. 1). It appears that the lesion will continue to behave in a non-aggressive manner.

The surgical specimen resembled a cyst filled with granular material of sandy consistency, which became powdery on digital pressure (Fig. 2).

Microscopically, sheets of pleomorphic round-to-ovoid cells, arranged in dense aggregates with vesicular and hyperchromatic nuclei, were seen. The cytoplasm was minimal and nucleoli were small (Fig. 3) Several mitotic figures were also evident. The tumour cells appeared to invade the surrounding fibrous tissue. A fibrous capsule, which, in areas, showed invasion by tumour cells, was observed (Fig. 4)

No cystic lining was evident even after studying multiple tissue samples. Notable finding in our case was presence of multiple, homogeneous, eosinophilic areas with occasional cell inclusions, present adjacent to and within sheets of tumour cells. Rosettes of cells often surrounded the material (Fig. 3). Mineralization, often in globular pattern, was noted in some of these eosinophilic secretory masses (Fig. 5)

Special stains were performed to characterize the eosinophilic material and were found to be positive for Collagen (Van Gieson and Masson's Trichome) and negative for keratin (modified Mallory's) and amyloid (Congo red). Von Kossa confirmed the areas of mineralization. Polarized microscopy revealed yellow to orange red fibres arranged in a cross-hatched and lamellar pattern, suggestive of mature collagen fibres.

The material was thus suggestive of either dentinoid/osteoid. Immunohistochemically, the round-to-ovoid cells stained positive with Cytokeratin (Dako, USA 3515), confirming their epithelial origin (Fig. 6)

A correlation of histomorphologic features in conjunction with the clinical and radiologic features was made and they were most consistent with a diagnosis of odontogenic carcinoma.

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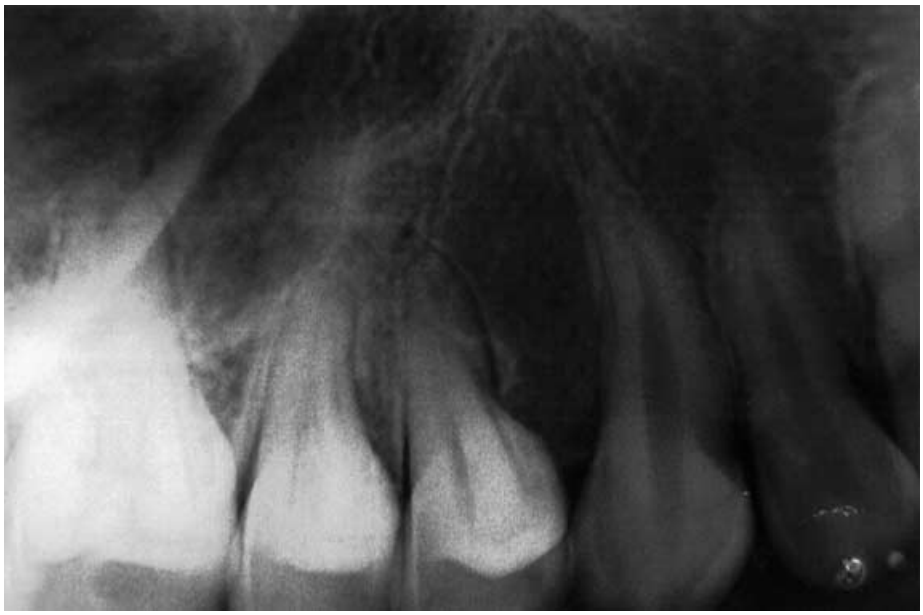

Figure 1 Post-operative radiograph shows normal healing of bone at the surgical site and resorbed roots of 14 and 15.

Comments

Primary intraosseous tumours are extremely rare tumours which occur only in jaw bones (1). As they arise from residues of odontogenic epithelium, they are termed as odontogenic carcinoma (1).

The case reported belongs to primary intraosseous carcinoma Type iii, i.e. arising *de novo* and of the non-keratinizing type so far, of which 35 cases have been reported (1). (Medline search keywords: 'primary intraosseous carcinoma' and 'odontogenic carcinoma').

Most of these tumours occur in the posterior mandible and few occur in the anterior maxilla, but no case appears to be reported in posterior maxilla (1). This case belongs to the lower end of the observed age group (4–81 years; 1) and is against the male predilection (M:F = 3 : 1; 2).

A detailed search for primary was carried out and a possibility of metastasis was ruled out by performing relevant clinical examination, chest radiograph and ultrasound examination of various organs. Clinical presentation resembled a cyst. Aspiration yielded a straw-coloured fluid as in a previously reported case (2).

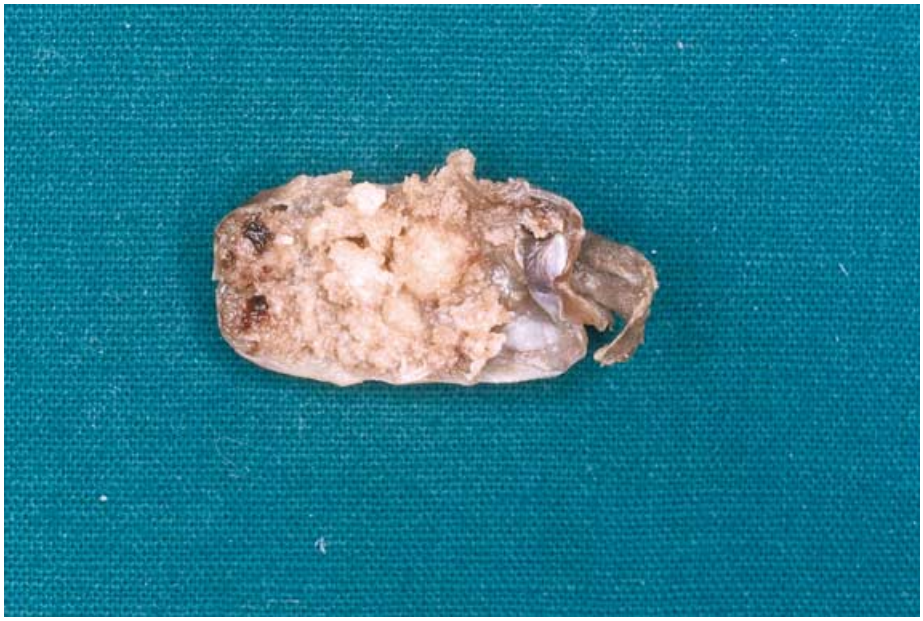

Figure 2 Surgical specimen showing well-defined mass with nodules of granular material.

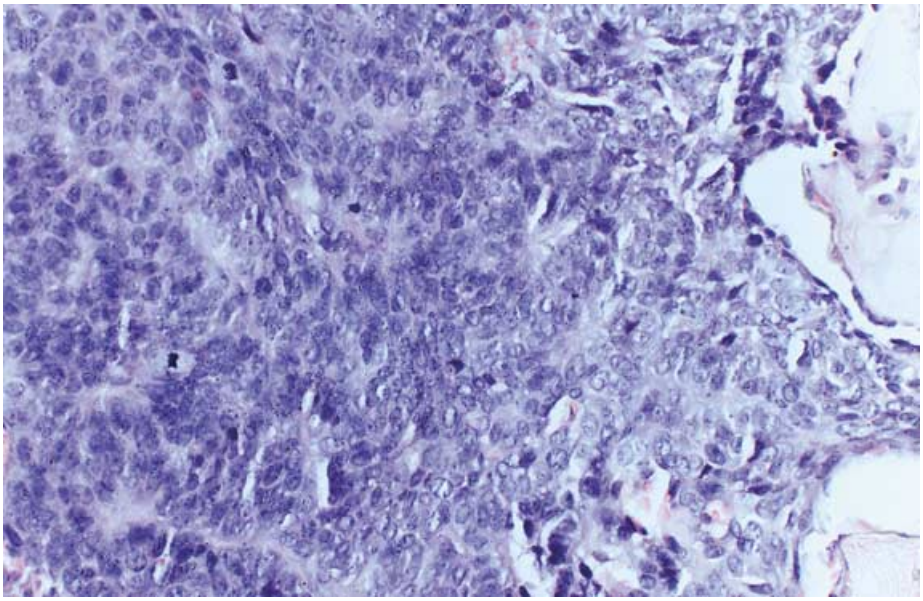

Figure 3 Shows rosettes of round and ovoid cells with hyperchromatic and vesicular nuclei surrounding the eosinophilic material. Few mitotic figures are also evident.

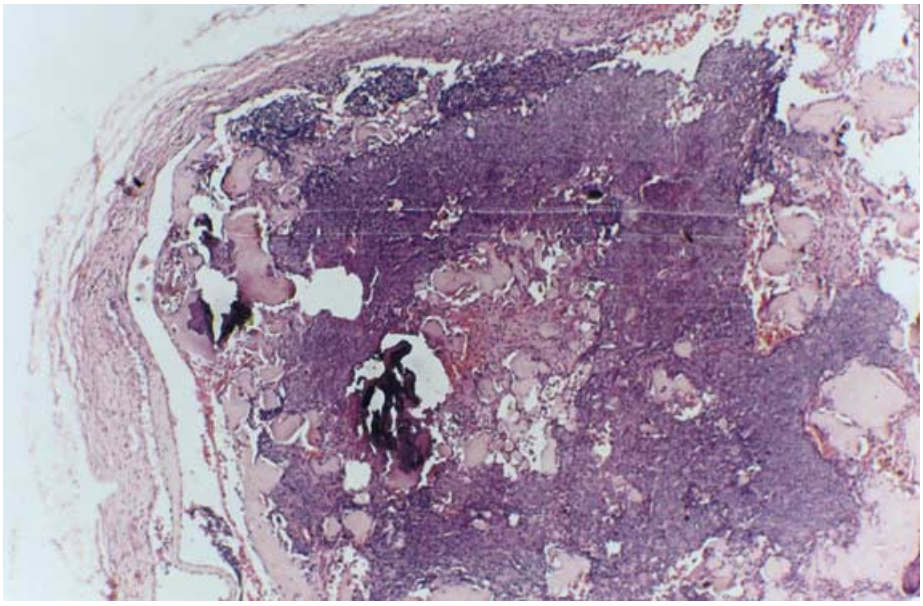

Figure 4 Fibrous capsule is seen with tumour cells interspersed with eosinophilic masses.

The usual histopathological features of odontogenic carcinoma usually resemble squamous cell carcinoma of non-keratinizing type with low-to-high mitotic activity (1). Peripheral palisading with alveolar/plexiform pattern was reported in few cases (3). But, in this case only, sheets of pleomorphic round or ovoid cells with few mitotic figures were seen.

Although the eosinophilic material showed staining reactions suggestive of osteoid/dentinoid, owing to its close association to the odontogenic epithelial cells, it could be considered to be dentinoid. Sawyer et al. (4) made a similar observation. A rarity in this series is the finding of marked osseous metaplasia (5). This case is reported for its unusual clinical presentation and rare histological features.

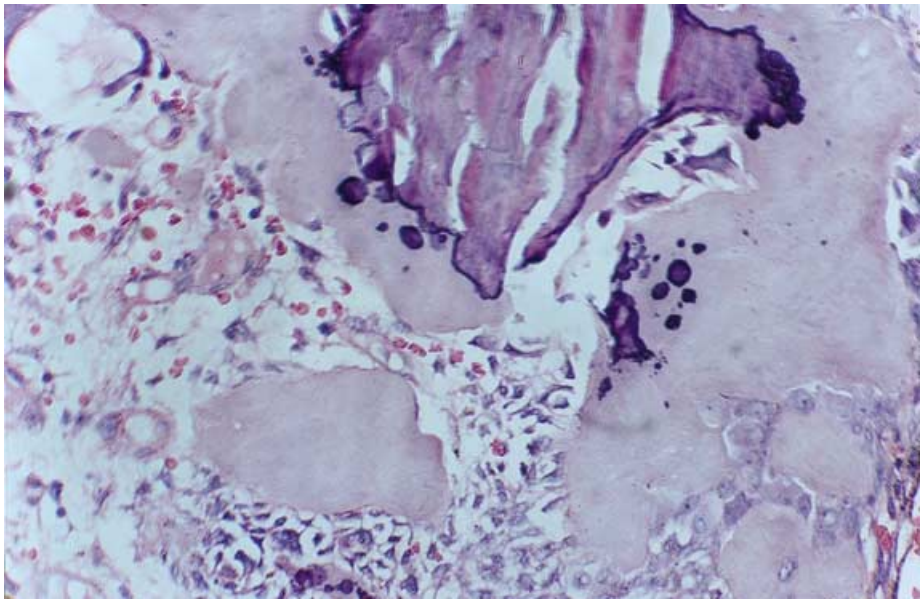

Figure 5 Abundant amount of osteoid/dentinoid undergoing globular mineralization in close proximity to the tumour cells.

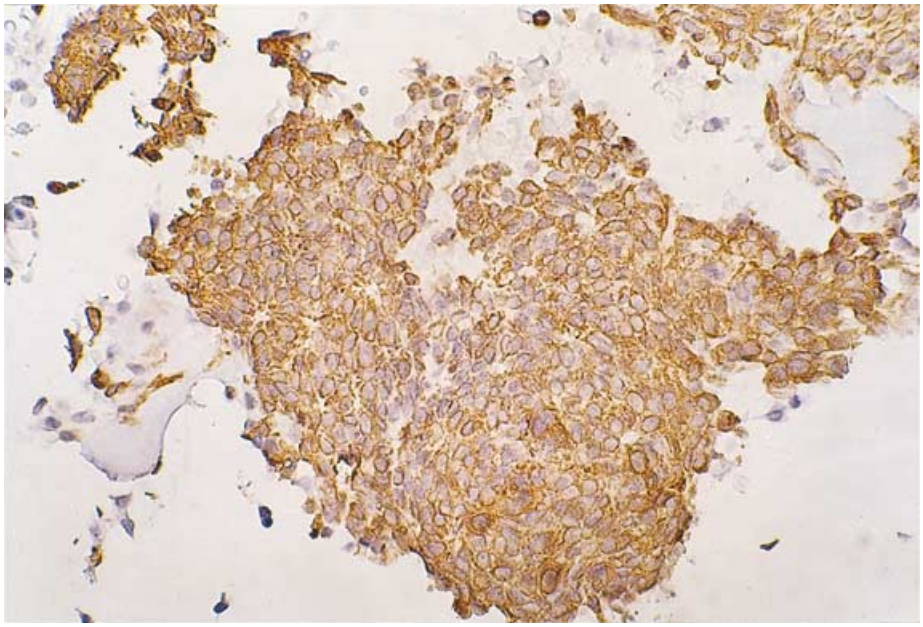

Figure 6 Tumour cells showing immunohistochemical positivity to cytokeratin.

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