CASE REPORT

A case report of a hybrid odontogenic tumour: Ameloblastoma and adenomatoid odontogenic tumour in calcifying cystic odontogenic tumour

Weiping Zhang, Yu Chen *, Ning Geng, Dongmei Bao, Mingzhong Yang

Department of Oral Pathology, West China College of Stomatology, Sichuan University, No. 14, 3-section, Renminnan Road, Chengdu 610041, PR China

Received 23 May 2006; received in revised form 27 June 2006; accepted 7 July 2006

Summary A hybrid odontogenic tumour comprising three distinct lesions is extremely rare. We presented a hybrid odontogenic tumour composed of a calcifying cystic odontogenic tumour (CCOT), a solid multicystic ameloblastoma (A-S/M) and an adenomatoid odontogenic tumour (AOT). This tumour was observed in the anterior area of the mandible of a 64-year-old Chinese woman. Masses of ghost epithelial cells with the characteristics of CCOT were seen in the lining of the cyst. The odontogenic epithelia with the features of A-S/M and AOT were also observed.

KEYWORDS Ameloblastoma; Adenomatoid odontogenic tumour; Calcifying odontogenic cyst

Introduction

In the oral maxillofacial region, calcifying cystic odontogenic tumour (CCOT), solid multicystic ameloblastoma (A-S/M) and adenomatoid odontogenic tumour (AOT) are well-recognised whereas hybrid odontogenic tumours have been rarely reported.1–17 The previously reported hybrid odontogenic tumours usually comprised two types of odontogenic tumours. However, no hybrid odontogenic tumour with the definite components of CCOT, A-S/M and AOT simultaneously in one lesion has been well-described in the English literatures so far.

Case report

On July 19, 2001, a 64-year-old woman was referred to the West China College of Stomatology, Chengdu, China, for a painless enlarging mass in the anterior of the mandible region with a history of 16 months duration. The patient stated that the lesion had grown gradually in the past year. The mass enlarged and paraesthesia appeared in the lower lip left side 20 days prior to the hospital visit. Physical examination revealed an obvious mass in the anterior of the mandible, with the size of about 3·3·5·1 cm³. Part of the lesion was felt to fluctuate. Panoramic radiograph revealed a large intraosseous cystic lesion with multiple radiopaque clusters and a well-demarcated border in the anterior of the mandible. The cyst extended from the left first molar region to the right second premolar region. An
unerupted left lateral incisor was also detected in the cystic lesion (Fig. 1). A biopsy prior to surgery was done, and the histopathologic diagnosis was the CCOT. Under general anesthesia, an excision of the lesion was performed on the mandible left side via the interoral approach and the segmental resection of the mandible was performed from the left first molar region to the right first molar region. The right seventh and eighth ribs were taken to repair the mandibular defect. The histopathologic diagnosis after the surgery is a hybrid odontogenic tumour characteristic of CCOT, A-S/M and AOT. No recurrence occurred during the post-surgical follow-up of three years.

Histological examination revealed a hybrid odontogenic tumour, showing the histopathological characteristics of CCOT, A-S/M and AOT, which accounted for about 60%, 25% and 15% of the lesion, respectively. Nests of ghost epithelial cells were observed in the lining of ameloblastomatous epithelium or in the fibrous capsule (Fig. 2a), suggesting the characteristics of CCOT. Deposits of scattered dentinoid or calcified materials were also noted. Furthermore, follicular islands of odontogenic epithelium were detected within a fibrous stroma. The basal cells of some islands of odontogenic epithelium were columnar and hyperchromatic, and were lined up in a Palisade fashion with reversed polarity (Fig. 2b). The central cells were sometimes loosely arranged and resembled the satellite reticulum of the enamel organ, which fulfilled the ameloblastic histological criteria. Variably sized solid nodules of cuboidal or columnar cells of odontogenic epithelia in the form of rosettes and duct-like structures were also observed in the connective tissue capsule of the cyst (Fig. 2c). In addition to forming ducts, the cuboidal or columnar cells formed convoluted cords in a complicated pattern that often exhibited invaginations. Based on its diverse histopathological features, the pathological diagnosis of a hybrid odontogenic tumour composed of CCOT, A-S/M and AOT was made.

Discussion

Calcifying odontogenic cyst (COC) is an uncommon odontogenic lesion, which was first described by Gorlin et al in 1962. COC has been classified as calcifying cystic odontogenic tumour (CCOT) according to the most recently established histopathological criteria of the World Health

Figure 2  (a) Ghost cells of CCOT: pale, eosinophilic, and swollen ghost cells lost their nuclei but they showed a faint outline of the cellular and the nuclear membrane. (b) Follicular islands of odontogenic epithelium within a fibrous stroma, the central cells were sometimes loosely arranged and resembled the satellite reticulum of the enamel organ, basal cell is hyperchromatism, vacuolisation and nuclear polarisation.-component of A-S/M. (c) Variable sizes of solid modules of cuboidal or columnar cells of odontogenic epithelia in the form of rosettes and duct-like structures in the connective tissue capsule of the cyst - component of AOT (HE stain, original magnification 200×).
Organization (WHO). The microscopic feature of CCOT is characterised by the lining of a well-defined ameloblastic epithelium with variable amounts of ghost cells. Several arguments have been proposed regarding the nature of ghost cells. Some studies suggested that the ghost cells might present the product of abortive enamel matrix in odontogenic epithelium because enamel proteins were immunohistochemically expressed in the ghost cells. However, other studies indicated that the ghost cells represented different stages of normal and aberrant keratin formation or the metaplastic transformation and coagulative necrosis of the odontogenic epithelium.

The components of other odontogenic tumours are often observed in CCOT, and the most common one is odontoma, ameloblastic fibroma (AF), ameloblastic fibro-odontoma (AFO), odontoameloblastoma (OA), odontogenic myxofibroma (OM), and odontogenic ghost cell carcinoma (OGCC) with some structures like those of A-S/M and AOT.

The biological mechanism causing such a unique mixture has not been well-known up to now. The transformation from one lesion to another seems to be a possible pathogenic mechanism. However, it is widely considered that the development of CCOT which possesses the features of other odontogenic tumours results in the development of these tumours secondarily, rather than that these tumours are themselves secondary phenomena of a pre-existing odontogenic tumour. Odontogenic tumours originate from odontogenic epithelium, which has the potentiality for diverse differentiations and mesenchymal induction. In this case, the proliferation of strands of odontogenic epithelium from the lining of the cyst resembled primary ectomesenchymal induction of the dental lamina, and the associated condensation of cells within the stroma also resembled primary ectomesenchymal induction of the dental lamina. Abrams and Howell pointed out that the combined occurrence of CCOT with another odontogenic lesion could be expected because of the multipotentiality of the odontogenic epithelium, which results from the processes of differentiation and degeneration of the odontogenic epithelium. Praetorius et al. believed that what appeared to be an associated odontogenic tumour was simply an integral part of the entire lesion developing from the wall of CCOT. In addition, Tanaka et al. reported a case of CCOT, which recurred one year after surgery with the element of malignant odontogenic tumour. It appeared that the lesion might represent different directions that the tumour may take, depending on the initial inductive stimulus, the degree of "odontogenesis" prior to the application of the stimulus, and individual human variability. Therefore, we speculate that A-S/M and AOT in the lesion might have been induced by the epithelium of CCOT in the present case.

Acknowledgements
The case report is funded by Sichuan University. We thank Professor Zhixiu He (Department of oral pathology, West China College of Stomatology, Sichuan University) and Dr Weihua Zhang (Department of civil engineering, Hongkong University of science and technology) for their valuable advice on the article writing.

References
