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Case Report

Low Malignant Intraductal Carcinoma on the Hard Palate: a Variant of Salivary Duct Carcinoma?

Y. Tatemoto, A. Ohno and T. Osaki

Department of Oral Surgery, Kochi Medical School, Kohasu, Oko-cho, Nankoku-city, Kochi 783, Japan

A rare, minor salivary gland tumour of the hard palate in a middle-aged woman was presented. The small (1.0 × 0.5 cm in diameter) hemispherical tumour was well circumscribed with a fine papillomatous surface. Histopathologically, tumour cells with eosinophilic cytoplasm and a large nucleus were single-strand cuboidal and columnar cells, which showed intraductal growth exhibiting a cribriform pattern. The histological features were distinct from adenoid cystic carcinoma and polymorphous low-grade adenocarcinoma because the tumour lacked the neurotropic infiltration, cord-like proliferation and targetoid arrangement. The tumour could not be identified as a typical salivary-duct carcinoma because Roman bridging, papillary projection, and severe cell atypia were not found. Tumour cells were negative for PAS, Alcian blue, mucicarmine, p53, c-erbB-2, CEA, S-100 protein, α-smooth muscle actin, lactoferrin or vimentin. About 5% of the tumour cells were positive for proliferating cell nuclear antigen. Taking these factors into account, together with the clinical features, the name low malignant intraductal carcinoma seems appropriate. Copyright © 1996 Elsevier Science Ltd

Keywords: salivary duct carcinoma, hard palate, low-grade malignancy

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INTRODUCTION

Salivary gland tumours manifest various histological patterns, and the histological complexity makes classification of these tumours difficult [1, 2]. The variations appear to be based on the multi-differential potentiality of salivary-gland cells and their stem cells [3–5].

There are three types of salivary tumours, adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma and salivary duct carcinoma (SDC) which exhibit cribriform patterns [2]. Some histological differences among these carcinomas have been documented [6–15]. In adenoid cystic carcinoma, a ductal structure composed of double layered cells and prominent neurotropic invasion are characteristic [2, 6–8]. Polymorphous low-grade adenocarcinoma characteristically exhibits lobular or solid growth, sometimes with a peripheral palisade of columnar cells, and the tumour shows cord-like proliferation with trabecular, fascicular and also targetoid arrangement (concentric whorls) around nerve fibres and blood vessels [2, 8–10]. Furthermore, Roman bridging and central necrosis are well known as characteristics of SDC [2, 11–14]. However, as shown by Delgado *et al.* [15], SDC has a broad clinicopathological spectrum, and there are many tumours which do not exhibit the typical histological characteristics. Recently, we experienced a small salivary gland tumour on the hard palate which showed a cribriform pattern. The tumour lacked the typical characteristics of SDC. The findings in this case are described and tumours exhibiting cribriform patterns are briefly discussed.

Correspondence to T. Osaki.
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CASE REPORT

A 58-year-old woman was referred for examination and treatment of a small mass on the palate. The patient had visited a dental office about 2 weeks earlier asking for a new denture of the upper jaw, and the small tumour was detected.

The tumour, measuring 1.0 × 0.5 cm in diameter, was located on the right side of the hard palate. The tumour was hemispherical, elastic, hard and well circumscribed. The surface was fine papillomatous, and not ulcerated. X-ray examination revealed no abnormality of the bone.

Under a diagnosis of benign salivary gland tumour, the tumour was resected with a safe surgical margin. The wound healed well without any sequelae during 30 months following extirpation.

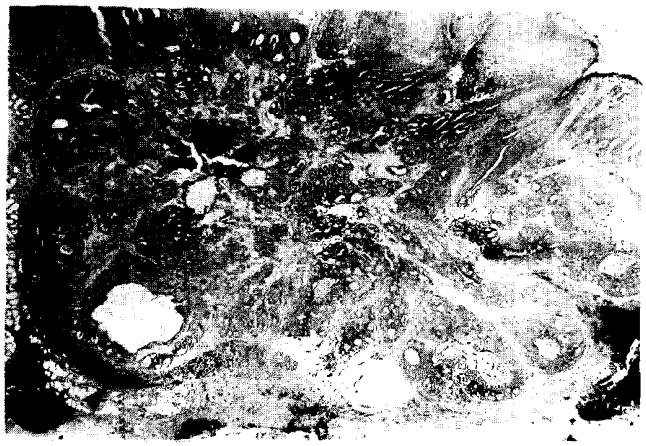

Fig. 1. Many tumour nests are formed exhibiting a duct-like appearance under the protruding epithelium. Focal lymphocyte infiltration (arrow heads) is observed, but no clear fibrous encapsulation or diffuse tumour cell infiltration is visible (H-E, original magnification: ×8.0).

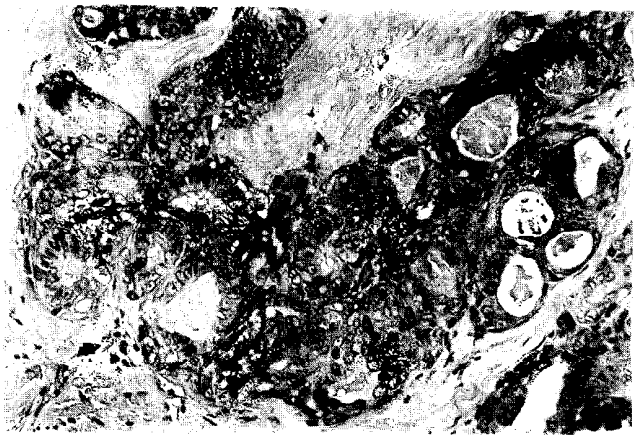

Fig. 3. A high-power view of the tumour nests (×250). Single strand cuboidal and columnar cells with relatively clear and eosinophilic cytoplasm and a large nucleus are seen. In the ductal spaces, eosinophilic substance is contained generally. Hyalinisation and a few lymphoid cell infiltration are also visible.

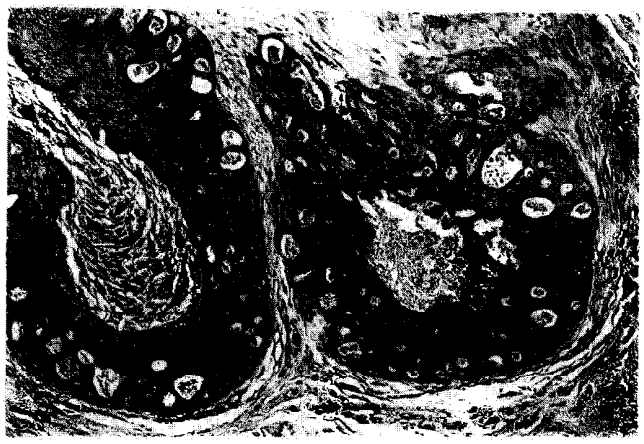

Fig. 2. A magnified view (×100). Each tumour nest exhibits a cribriform pattern, and necrotic cells are observed in the ductal space (arrow).

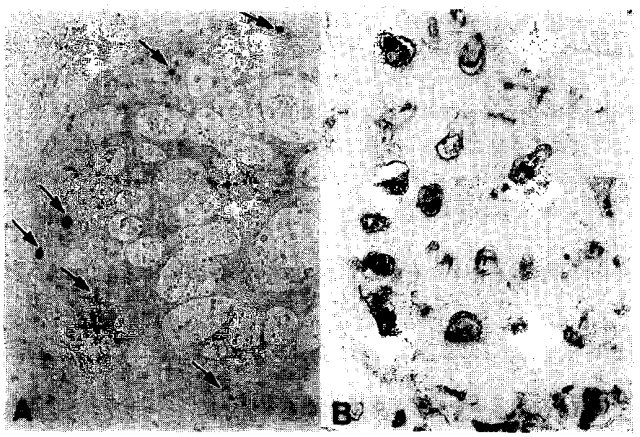

Fig. 4. Immunostaining for PCNA (A) and EMA (B). PCNA-positive cells (arrows) are scattered in the tumour tissue showing a cribriform pattern, and EMA is localised in the luminal surfaces. ×150, ABC method.

MICROSCOPIC FINDINGS

Many large and small tumour nests proliferated in the fibrous connective tissue (Fig. 1). In the tumour nests, a ductal structure exhibiting a cribriform pattern was prominent. Eosinophilic substances were contained in the ductal spaces, and slightly necrotic tumour cells were visible in some places (Fig. 2). However, neither Roman bridging nor papillary projection of the tumour cells was observed, nevertheless so-called central necrosis was found in some areas. The tumour cells had eosinophilic cytoplasm with slightly clear, large nuclei. Hyalinisation and focal lymphocyte infiltration were scattered in the stroma (Fig. 3). There was no encapsulation of the tumour, tumour invasion into the surrounding tissues or targetoid arrangement of the tumour cells around nerves or blood vessels. Histochemically, tumour cells were fully negative for PAS, Alcian blue or mucicarmine. In addition, none of p53, c-erbB-2, carcinoembryonic antigen (CEA), S-100 protein, α-smooth muscle actin, vimentin or lactoferrin were detected immunohistochemically in tumour cells. However, about 5% of the tumour cells were positive for proliferating cell nuclear antigen (PCNA) and epithelial membrane antigen (EMA) was observed in the luminal surface of ducts (Fig. 4).

DISCUSSION

Various classification systems for salivary gland tumours have been tried, and recently a new classification was proposed [2]. These efforts manifest the difficulty of classifying salivary gland tumours. Complicated histopathological variation and rare occurrence combine to cause difficulties in classification [1–3].

According to the WHO classification [2], there are three types of tumour which exhibit cribriform pattern: adenoid cystic carcinoma [6], polymorphous low-grade adenocarcinoma [9] and SDC [11]. In the present case, the cribriform

pattern was prominent. However a ductal structure was formed by monolayered columnar cells. Together with the lack of neurotropic infiltration, the present tumour seems clearly different from adenoid cystic carcinoma.

It was reported that polymorphous low-grade adenocarcinoma, which usually occurs in the minor salivary glands, exhibits a variety of characteristic microscopic appearances such as trabecular, lobular, papillary and fascicular growth patterns and a targetoid arrangement around nerve fibres and blood vessels [8–10]. However, these features were lacking in the present case.

SDC, which is most prevalent in the parotid gland, is highly malignant [2]. SDC displays a duct-like structure, and manifests cribriform pattern and Roman bridging. Necrosis of the central region of the proliferating tumour nests frequently occurs [2, 11–14]. The histopathological characteristics of the present tumour seem to be more similar to those of SDC than to those of the other two types of carcinoma. However, the present tumour did not exhibit any of the typical histological features of SDC, that is, Roman bridging and infiltrative proliferation as well as severe tumour cell atypia. Therefore, controversy concerning the final diagnosis of the present tumour as SDC seems to remain. There is a possibility that this palatal tumour is metastatic from the breast. A variety of tumours, such as adenoid cystic carcinoma, SDC and other ductal carcinomas, arise from the mammary glands. In fact, in some fields, the present tumour resembles comedocarcinoma of the breast. However, no breast tumour has been detected.

Specific clinical and histological characteristics of SDC do not seem to be confirmed yet. In the literature, there are 12 SDC cases which originated from the minor salivary gland [15–24] and one showed good clinical sequence [15–21, 23]. SDCs with severe cell atypia were found in the major salivary gland [11–14]. Based on the investigation of Delgado *et al.* [15], SDC shows a broader clinicopathological spectrum than previously considered. They proposed that SDC may arise in a pleomorphic adenoma and the proportion of intraductal and extraductal growth is of prognostic significance. SDC arising from the minor salivary gland may remain in its intraductal phase and has features that overlap papillary cystadenocarcinoma and papillary-cystic acinic cell carcinoma [15]. Acinic cell carcinoma, however, exhibits basophilic cytoplasm, and cystadenocarcinoma is characterised by papillary endocystic projections with narrow fibrous cores. In conclusion, the position of minor SDC with low malignancy has yet to be established in the classification of salivary gland tumours. As described above, the present tumour lacked the highly malignant characteristics of SDC. Therefore, low malignant intraductal carcinoma seems to be the most suitable name for the present case.

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