CASE REPORT

Polymorphous low-grade adenocarcinoma of submandibular gland origin

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Summary Polymorphous low-grade adenocarcinoma (PLGA) occurs almost exclusively in minor salivary glands, and there are only a few reports of major salivary gland origin. We herein report a case of de novo PLGA arising in a submandibular gland and discuss preoperative findings of magnetic resonance imaging. © 2005 Elsevier Ltd. All rights reserved.

Introduction

Polymorphous low-grade adenocarcinoma (PLGA) of the salivary glands was first described in 1984 and this tumor is now a well-recognized entity in minor salivary glands, particularly of the palate. PLGA can be summarized as cytological uniformity, morphological diversity and a low metastatic potential. Since PLGA arising in major salivary gland is considered extremely rare, its characteristics have not been well defined. We describe a case of PLGA arising in a submandibular gland.

Case report

A 67-year-old Japanese woman was referred to the Oral Surgery Clinic of Hokkaido University Hospital in March 2003, with a symptom of swelling in the right submandibular region. She had first noticed the swelling approximately 3 years earlier and it had gradually enlarged thereafter. On examination, a firm and non-tender mass measuring...
2.5 × 2 cm in size was palpable in the submandibular region. Neither lymphadenopathy nor enlargement of any other salivary glands was present. CT showed a well-defined mass with irregular enhancement in the submandibular gland. Magnetic resonance imaging (MRI) demonstrated that the mass had a low signal on T1-, a low to isosignal on T2-weighted images, and hardly any enhancement on a Gd-DTPA image (Fig. 1). 201TilCl showed the absence of uptake both in early and delayed phases. The clinical diagnosis was a suspicion of chronic sclerosing sialoadenitis. Since the intraoperative biopsy specimen yielded a provisional diagnosis of pleomorphic adenoma, a total submandibular glandectomy was performed. Histopathologic examination revealed various morphological patterns such as solid, granular, cribriform, tubular, trabecular and cystic growth patterns (Fig. 2). The tumor invaded the parenchyma of residual salivary gland and surrounding connective tissues.

Figure 1  (A) Coronal T1-weighted MRI shows a well-defined mass of low signal intensity in the right submandibular gland (arrowheads). (B) The lesions have a heterogeneous mass of lower signal intensity than surrounding normal submandibular gland on the T2-weighted image (arrowheads).

Figure 2  Variable morphological patterns of solid (A), cribriform (B), tubular (C), trabecular (D) growth patterns (hematoxylin and eosin, original magnification × 100).
Perineural invasion was identified with a targetoid growth pattern. The tumor was surrounded by dense collagenized stroma that displayed hyalinized and occasionally myxoid degeneration. Immunohistochemical studies showed positive staining for cytokeratins, vimentin and S-100 protein. The pathologic diagnosis was polymorphous low-grade adenocarcinoma (PLGA), and the patient underwent adjuvant radiation therapy (total 55 Gy). She remains alive without any evidence of disease at 1 year 10 months after the operation.

**Discussion**

PLGA is considered to have low-grade malignant potential in that regional lymph node metastasis is relatively uncommon, and distant metastasis is rare. Since PLGA occurs almost exclusively in minor salivary glands, and its origin in a major salivary gland is considered extremely rare, its characteristics have not been well defined. In a review of the literature, Nagao et al. reported that PLGAs arising in major salivary glands have clinicopathological and immunohistochemical features similar to those of PLGAs originating in minor salivary glands. In general, definitive diagnosis of PLGA of an excised tumor is relatively easy because of the unusual histopathological and immunohistochemical findings. On the other hand, it is virtually impossible with frozen sections of small biopsy specimens. In fact, the provisional diagnosis in our case was pleomorphic adenoma. Therefore, preoperative differential diagnosis is quite important in submandibular gland lesions such as benign or malignant neoplasms and inflammation. In the present case, preoperative MRI findings showed features of chronic sclerosing sialadenitis rather than a benign salivary gland tumor. A mass in the submandibular gland demonstrated a low signal intensity on T2-weighted images, and hardly any enhancement on Gd-DTPA images. These findings usually suggest fibrous or scar tissues, which are identical with chronic sclerosing sialadenitis. In benign salivary gland tumors such as pleomorphic adenoma, high signal intensity is shown on T2-weighted images. The possibility of PLGA should be considered when MRI of the submandibular gland shows low signal intensity on T2-weighted images and no contrast enhancement, although it is extremely rare.

**References**