Case Report

Mucoepidermoid carcinoma arising in Warthin’s tumor of the parotid gland

Shin-ichi Yamada, Takemitsu Matsuo, Shuichi Fujita, Kazutaka Suyama, Akira Yamaguchi and Akio Mizuno

Divisions of Oral and Maxillofacial Surgery and Oral Pathology and Bone Metabolism, Department of Developmental and Reconstructive Medicine, Course of Medical and Dental Sciences, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan

Malignant transformation of Warthin’s tumor is extremely rare, although it is the second most common benign tumor of the parotid gland. We describe our experience of mucoepidermoid carcinoma arising in Warthin’s tumor of the parotid gland in a 64-year-old man. He had a swelling in the left parotid-masseteric region. The removed tumor was well encapsulated and histopathologically comprised Warthin’s tumor and low-grade mucoepidermoid carcinoma. The mucoepidermoid carcinoma infiltrated lymphoid stroma of the Warthin’s tumor, but capsular invasion is not found. Considering the clinical course and physical examination, the lesion was diagnosed as mucoepidermoid carcinoma arising in Warthin’s tumor because its features filled the criteria of malignant transformation of Warthin’s tumor. There has been no recurrence or distant metastasis for 19 months. Histological change from Warthin’s tumor to mucoepidermoid carcinoma may be implicated in squamous or goblet cell metaplasia of epithelial cells. It is considered that the neoplastic cells of Warthin’s tumor acquire malignant genotypes simultaneously with this dual differentiation.

Key words: malignant transformation, mucoepidermoid carcinoma, parotid gland, Warthin’s tumor

Warthin’s tumor, which is known as papillary cystadenoma lymphomatosum or adenolymphoma, is most frequently seen in the parotid gland and is the second most common benign tumor next to pleomorphic adenoma. There have been reports that Warthin’s tumor involves other neoplasms synchronously or metachronously. However, malignant transformation of Warthin’s tumor and carcinoma arising in Warthin’s tumor are extremely rare. To date, there have been 29 reports of carcinoma arising in Warthin’s tumor of the parotid gland. This report describes a case of mucoepidermoid carcinoma arising in Warthin’s tumor of the left parotid gland.

CLINICAL SUMMARY

A 64-year-old man presented to our clinic with a slowly enlarging mass, of which he was unaware, in the left parotid-masseteric region and xerostomia. At the time of the first visit, the patient’s medical history was uneventful except for hypertension 28 years ago, and hepatitis B infection 26 years ago.

Physical examination revealed a 40 × 30 × 15 mm, relatively well-defined, elastic hard and tender mass in the parotid-masseteric region of the left parotid gland. The patient did not know when the swelling began. The skin over the growth was intact and movable. The facial nerve was not involved. Intraoral examination showed no abnormality. A computed tomography scan revealed a 35 × 25 × 20 mm encapsulated tumor with a relatively clear margin and soft tissue density in the region (Fig. 1). Clinically, a diagnosis of benign tumor was made. Enucleation was carried out under general anesthesia. During the operation, the tumor was easily separated from the adjacent tissues. The patient made an uneventful recovery with good facial nerve function and was discharged 23 days postoperatively. He is alive with no evidence of recurrent disease 19 months after surgery.

PATHOLOGICAL FINDINGS

A cut surface of the extirpated specimen showed a totally encapsulated, grayish-white mass consisting of multilocular...
structures. Microscopic examination demonstrated that the tumor contained two types of neoplasm (Fig. 2). One type occupied half of the tumor, and showed the features of Warthin’s tumor (i.e. papillary protrusions into cystic lumens lined by double-layered columnar cells with oncocytic appearance). Under the epithelium, lymphoid stroma including lymphoid follicles was noted. Most of the columnar epithelial cells exhibited oncocytic metaplasia (Fig. 3), and sparse goblet cell metaplasia was noticed. The other type revealed small solid nests of epidermoid cells and intermediate cells, and cystic structures lined by mucous and intermediate cells (Fig. 4). Some solid nests comprised intermediate and mucous cells invaded the lymphoid stroma (Fig. 5). Histologically, this type was consistent with low-grade mucoepidermoid carcinoma. The mucoepidermoid carcinoma was nodular in focus, and measured 20 × 14 mm. No infiltration in the capsule was found. The histopathological diagnosis was mucoepidermoid carcinoma arising in Warthin’s tumor.

**DISCUSSION**

Warthin’s tumor is a well-defined salivary gland neoplasm that occurs preferentially in the parotid gland. A variable mixture of lymphoid stroma and double-layered oncocytic columnar epithelium characterizes Warthin’s tumor. Malignant transformation of Warthin’s tumor and carcinoma arising in Warthin’s tumor are extremely rare. Until now, there have
been 29 cases of different types of carcinoma arising in Warthin's tumor.\textsuperscript{1,8} Squamous cell carcinoma\textsuperscript{7–9} and mucoepidermoid carcinoma\textsuperscript{1,6,10,11} were reported as common types of carcinoma arising within Warthin's tumor. Other types of carcinoma arising in Warthin's tumor are oncocytic carcinoma,\textsuperscript{12} undifferentiated carcinoma\textsuperscript{13} and adenocarcinoma not otherwise classified.\textsuperscript{14}

The diagnosis of malignant transformation of Warthin's tumor is based on distinct criteria: (i) the presence of a pre-existing benign Warthin's tumor; (ii) the presence of transitional zones from benign oncocytic to frankly malignant epithelia; (iii) the infiltrating growth in the surrounding lymphoid tissue; and (iv) the exclusion of metastases to the lymphoid stromal component of a primary extrasalivary tumor.\textsuperscript{10} In the present case, the patient did not know when the swelling of the parotid gland occurred. Warthin's tumor may have existed for a long time because of the painless and slow growth characteristics of the tumor. The Warthin's tumor and mucoepidermoid carcinoma were totally encapsulated, and some nests of mucoepidermoid carcinoma had invaded the lymphoid stroma of Warthin's tumor. We were unable to identify the histological figures of the malignant transformation of Warthin's tumor, because it was difficult to distinguish between goblet cells of Warthin's tumor and mucous cells of mucoepidermoid carcinoma. However, we think that there were some malignant transformative points from Warthin's tumor to mucoepidermoid carcinoma. The differential diagnosis of mucoepidermoid carcinoma arising in Warthin's tumor includes metastatic carcinoma to Warthin's tumor.\textsuperscript{1} In our case, the metastasis to Warthin's tumor from a previous or synchronous malignant neoplasm involving a different anatomical site was excluded by the clinical history and physical examination. Therefore, the mucoepidermoid carcinoma was thought to arise in Warthin's tumor.

The surgical management of mucoepidermoid carcinoma arising in Warthin's tumor is total or subtotal parotidectomy with a safe margin. The results of the treatment were relatively good without evidence of recurrent disease. The prognosis of patients with parotid gland mucoepidermoid carcinoma is considered to be largely dependent on tumor stage, tumor grade and the adequacy of the resection margins. The good clinical outcome of patients with mucoepidermoid carcinoma involving Warthin's tumor is probably due to these factors.\textsuperscript{1}

REFERENCES

1 Williamson JD, Simmons BH, el-Naggar A, Medeiros LJ. Mucoepidermoid carcinoma involving Warthin tumor. A report


