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Cystadenocarcinoma of the salivary glands with potential lymph node metastasis

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ABSTRACT

Cystadenocarcinoma derived from the salivary gland in the head and neck region is an extremely rare malignancy. Therefore, the clinicopathological characteristics is not well understood. The purpose of this study was to clarify the clinicopathological characteristics, and present a total of four patients who were treated at the National Cancer Center Hospital East during the period between 1995 and 2012. The patients were all male, with ages ranging from 47 to 74 years old. The primary sites were the parotid glands, submandibular gland and minor salivary gland of the tongue. Within the follow-up period from 19 to 54 months, lymph node metastases were observed in three of the four patients. However, all the patients were salvaged by additional resection, including neck dissection, and remain alive to date without distant metastases. Cystadenocarcinoma is classified as a low-grade histological subtype of salivary gland tumors. Although the tumor has the potential to produce lymph node metastases, as shown in our patients, it is generally an indolent tumor with a good prognosis as compared with high-grade subtypes. This study suggests that a long-term follow up paying close attention to lymph node metastases is necessary for cystadenocarcinoma.

1. Introduction

Cystadenocarcinoma of the head and neck region is an extremely rare malignancy described by World Health Organization (WHO) in 2005 [1]. The estimated incidence of cystadenocarcinoma in this region is only 2% of all salivary glands malignancies [2]. The WHO classification of head and neck tumors defines cystadenocarcinoma as characterized by predominantly cystic growth that often exhibits intraluminal papillary growth, but which is lacking in any additional specific pathological features that characterize the other types of salivary gland carcinomas [2]. Although the clinicopathological characteristics of cystadenocarcinoma of the salivary glands have been described in the past [1–4], the progression of cases presenting with lymph node metastasis is little understood. The purpose of this study is to show the clinicopathological characteristics of cystadenocarcinoma with lymph node metastasis through the presentation of our cases.

2. Case report

A total of 4 patients with cystadenocarcinoma received treatment at our institution. The primary site of the tumor was the parotid gland in two cases, the submandibular gland in one case, and the minor salivary gland of the tongue in one case. The clinicopathological characteristics of all cases are shown in Table 1.

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Table 1
The clinicopathological features of the cases experienced in our institution.

Case	1	2	3	4
Age	69	66	74	47
Sex	M	M	M	M
Localization	Submandibular gland	Tongue	Parotid gland	Parotid gland
FNA	Class 3	Not done	Class 2	Class 2
Operation	Sialoadenectomy + ND (Rt. I)	Partial glossectomy + ND (Rt. I–III, Lt. I–IV)	Superficial parotidectomy	Superficial parotidectomy
TNM	pT2N2bM0	pT1N2bM0	pT3N0M0	pT2N0M0
Invasive micro papillary component	(+)	(−)	(−)	(+)
Recurrence	(+): Neck node (Rt. level III)	(+): Neck node (Lt. Level I)	(−)	(+): Local and neck node (superficial and intra-parotid node)
Treatment for recurrence	Surgery: ND (Rt. II–IV)	Surgery: ND (Lt. I)		Surgery
Prognosis	NED 19 months	NED 33 months	NED 48 months	NED 54 months

ND: neck dissection; NED: no evidence of disease.

2.1. Case no. 1

A 69-year-old male had noticed a painless mass in the right submandibular gland for a period of 3 months. MRI identified a well-circumscribed, solid, round mass of 25 mm in diameter in the submandibular gland. The tumor had a clear capsule structure and showed both hyper and hypo-intensity part in T2-weighted images (Fig. 1A). No lymph node metastasis was detected in CT or MR images. Fine-needle aspiration (FNA) resulted in a class 3 diagnosis because of lack of cellular atypia with high grade. In consideration of low-grade salivary gland carcinoma, submandibular sialoadenectomy with submandibular dissection (level I) were performed. Microscopically, the tumor revealed multi-cystic lumens filled up with solid parts, which were occupied by the papillary structures of the tumor cells (Fig. 2A). The nuclei were uniformly bland, and mitotic figures were rare (Fig. 2B). The tumor was encapsulated with a fibrous capsule, and showed the expansive growth. Some micro-papillary nests of tumor cells were observed in the stroma and salivary gland around the tumor with some stromal

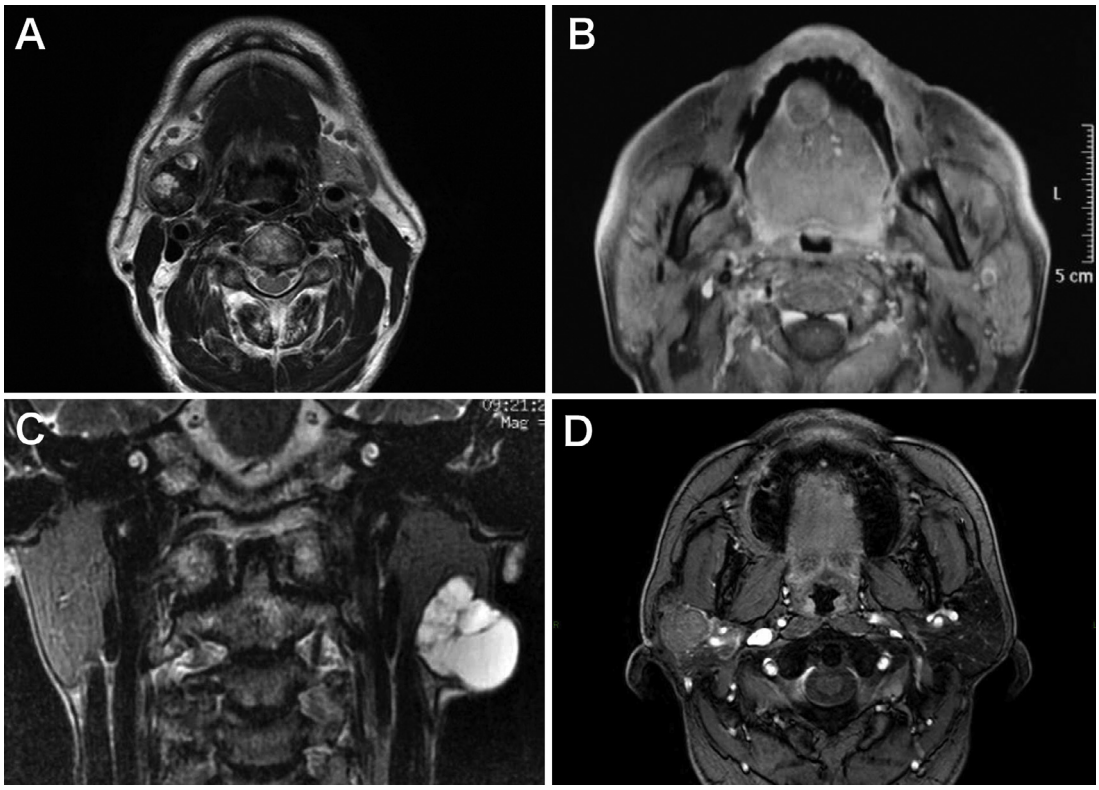

Fig. 1. (A) An MR axial T2-weighted image showing the tumor in the right side of the submandibular gland (Case no. 1). (B) A post-gadolinium axial T1-weighted image showing the tumor in the ventral tongue (Case no. 2). (C) A coronal T2-weighted image showing the tumor had a multicystic form with clear borders, and the inside of the tumor showed a hyperintense signal (Case no. 3). (D) A post-gadolinium axial T1-weighted image showing relatively uniform contrast throughout the tumor. No clear permeation to the normal parotid gland tissue is apparent (Case no. 4).