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CASE REPORT

Squamous cell carcinoma of the base of tongue with initial presentation of cystic metastasis in contralateral cervical lymph node

Mei Yonemoto [a], Hiroshi Yusa [a], Kenji Yamagata [a], Shuichi Fujita [b], Akira Yamaguchi [c], Hiroshi Yoshida [a,*]

[a] *Department of Oral and Maxillofacial Surgery, Institute of Clinical Medicine, University of Tsukuba, 1-1-1 Tennoudai, Tsukuba, Ibaraki 305-8575, Japan*
[b] *Division of Oral Pathology and Bone Metabolism, Department of Developmental and Reconstructive Medicine, Nagasaki University Graduate School of Biomedical Sciences, 1-7-1 Sakamoto, Nagasaki 852-8588, Japan*
[c] *Section of Oral Pathology, Department of Oral Restitution, Graduate School of Tokyo Medical and Dental University, 1-5-45 Yushima, Bunkyo, Tokyo 113-8549, Japan*

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Summary A case of squamous cell carcinoma of the base of tongue presenting initially as a cystic metastasis in the contralateral neck is described. The patient complained of a painless mass in the left neck, which was removed and histologically diagnosed as cystic malignancy. One year and seven months after the removal, a squamous cell carcinoma of the right tongue base and cystic metastases of the right neck were discovered. The histologic similarity among these three lesions strongly indicated the cystic metastases in the bilateral neck were from the squamous cell carcinoma of the tongue base.

Introduction

Most cervical lymph node metastases from carcinomas of the oral and maxillofacial region are solid, but carcinomas arising in Waldeyer's ring, which includes the tonsil, base of tongue, and nasopharynx, often appear as a cystic metastatic mass in the

* Corresponding author. Tel./fax: +81 29 853 3050.
E-mail address: hyoshida@md.tsukuba.ac.jp (H. Yoshida).

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upper neck.[1–5] A cystic squamous cell carcinoma (SCC) in the upper neck is histologically indistinguishable from branchial cleft cyst carcinoma (BCCC),[6–10] making the differential diagnosis difficult in the case of a clinically occult primary site of the SCC.[1,6,9,11] However, diagnoses of BCCC using the criteria of Martin et al.[1] are so rare that the very existence of BCCC has recently been questioned. Therefore most cystic SCCs in the neck can safely be diagnosed as probable metastases from Waldeyer's ring.[1,3,4] Although bilateral neck cystic metastases are occasionally reported,[1,3] many are unilateral, and most are ipsilateral to the primary lesion.[5,9,11,12]

This report presents a case of SCC of the base of the tongue with bilateral cystic metastases in the neck. The neck metastasis that was contralateral to the primary lesion was the initial presentation, and the primary site and ipsilateral neck metastasis were discovered one year seven months after the initial cystic malignancy of the neck was removed.

Case report

A 62-year-old man was referred because of a painless and gradually enlarging mass on the left neck that had persisted for two months. The mass was soft, well-defined, 3 cm in diameter, located anterior to the sternocleidomastoid muscle, and was not adherent to the surface skin. The patient had no history of previous malignancy or of neck swelling as a child. The oral examination was normal. Magnetic resonance (MR) images depicted a well-defined round mass with high signal intensity that was anterior to the sternocleidomastoid muscle and caudal to the submandibular gland, and involved the jugular digastric lymph nodes. The mass also had an internal septum and a low signal-intensity area. The right neck appeared normal (Fig. 1(A)). A biopsy by ultrasound-guided fine needle aspiration collected transparent, serous, yellow fluid from the mass. Cytology of the fluid was negative for malignancy. Chest radiograms were negative. The lesion was clinically diagnosed as a branchial cleft cyst.

The mass was removed surgically. It was easily dissected from the surrounding soft tissues and small adjacent lymph nodes were removed simultaneously. Histologic examination revealed that the lesion was surrounded by a thick fibrous capsule, and contained cystic spaces lined by stratified squamous epithelium exhibiting apparent cellular atypia and many mitotic figures. The lining epithelium protruded into the cystic space and grew downward into the underlying lymphoid elements. Some of the lining epithelium resembled normal

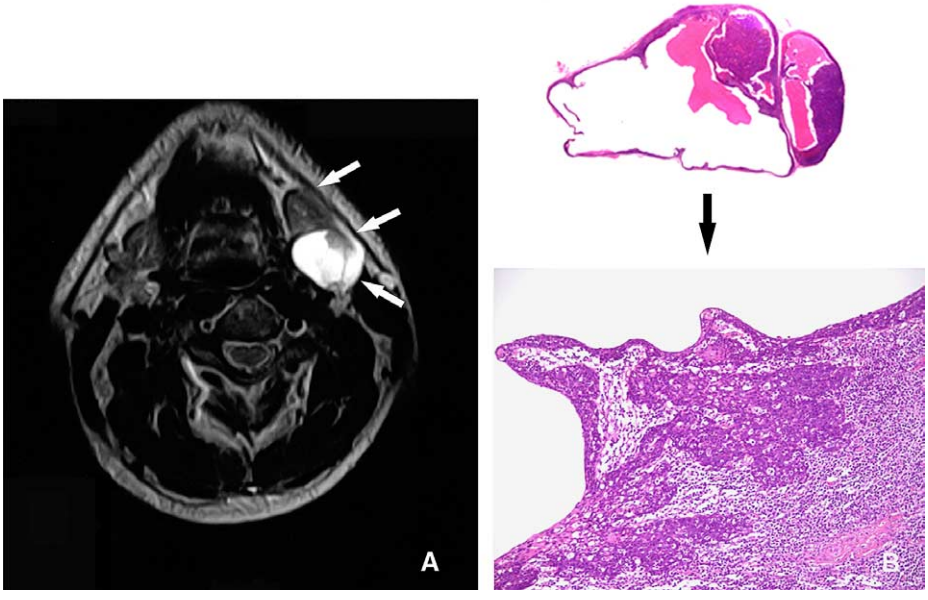

Figure 1 (A) T2-weighted magnetic resonance image of the left neck mass. A well-defined round mass with high signal intensity that had an internal septum and a low signal intensity area was found anterior to the sternocleidomastoid muscle and caudal to the submandibular gland; it involved the jugular digastric lymph nodes. (B) Histologic features of a lymph node from the left neck mass. The lesion contained large cystic spaces lined by stratified squamous epithelium showing cellular atypia. Note that the lining epithelium protruded into the cystic space and grew downward into the underlying lymphoid elements (H&E, ×10).

squamous epithelium that transitioned into the epithelium that exhibited prominent cellular atypia (Fig. 1(B)). All the small lymph nodes were solid and negative for cancer. These findings strongly suggested the lesion was either a cystic metastasis from an unknown primary SCC or a BCCC.

Neither endoscopy with ipsilateral tonsillectomy and directed biopsies of Waldeyer's ring nor additional treatments, such as radiotherapy, neck dissection, or a combination, were performed. Because there was a high probability of metastasis, a search for the occult primary lesion was carried out by examining the nasopharynx, tonsils, tongue base, upper aerodigestive tract, and lungs at monthly follow-up visits during the next year. However, no primary site was discovered, and a bimonthly examination restricted to Waldeyer's ring was instituted.

One year and seven months after the removal of the neck mass, the patient suddenly visited complaining of slight bleeding from the right lingual tonsil. The biopsy specimen of the right tongue

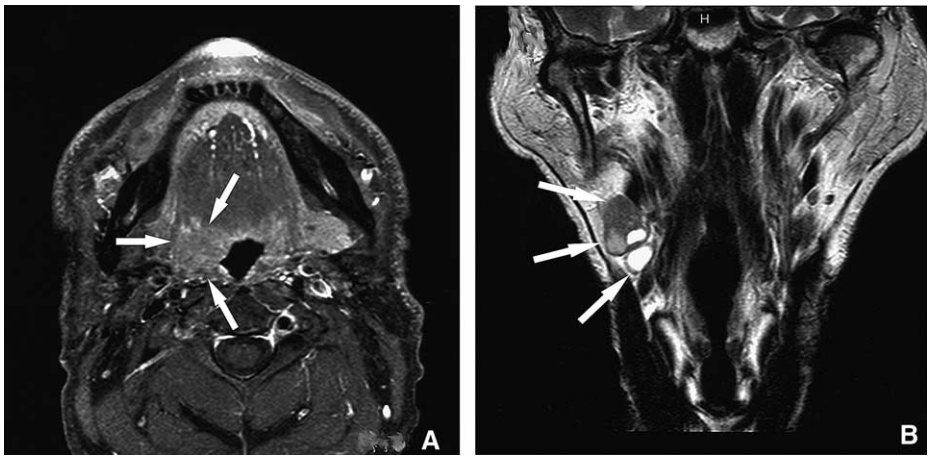

Figure 2 MR images of the tongue base and right neck. (A) T1-weighted sequence depicts a mass with low signal intensity at the right tongue base. (B) T2-weighted sequence shows the cystic lesions in the jugular digastric lymph nodes.

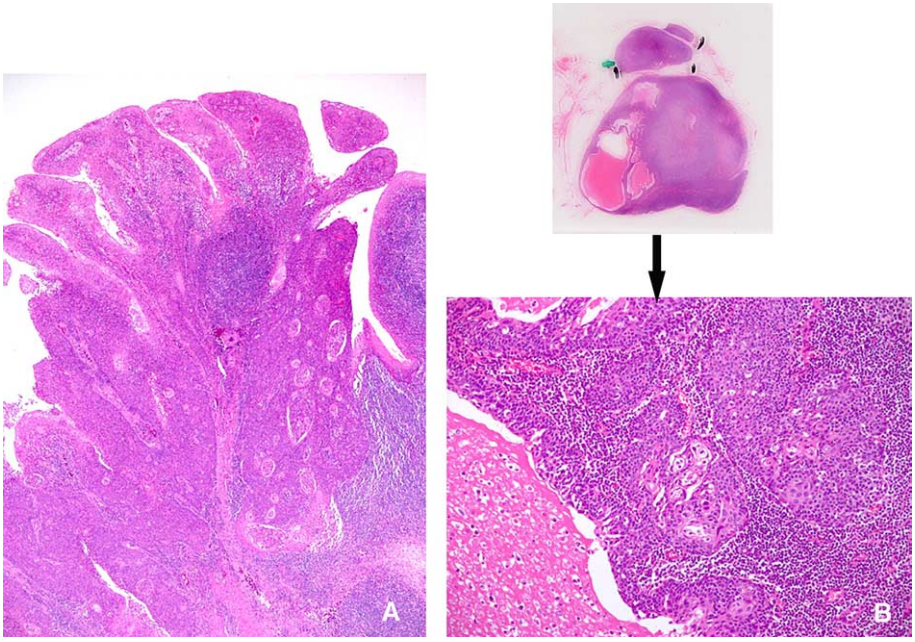

Figure 3 (A) Histologic features of the base of the tongue. The tumor shows papillary growth. SCC invaded the subepithelial layers (H&E, ×10). (B) Histologic features of the right neck lymph node. The lymph node contained cystic spaces lined by stratified squamous epithelium similar to those observed in the left neck lymph node (H&E, ×60).

base revealed SCC. MR images depicted a mass occupying the right tongue base and cystic lesions in the right neck (Fig. 2(A) and (B)). Considering the high possibility of cystic neck metastases from cancer of the base of the tongue, resection of the tongue base, right radical neck dissection, and immediate reconstruction with a forearm free flap were performed. Histologic examination of the resected specimen demonstrated that the tumor of the tongue base was SCC with papillary architecture. The tumor cells showed cellular atypia and invasion into the subepithelial layers (Fig. 3(A)). The cystic lesions were in lymph nodes and were diagnosed histologically as SCC. This tissue was histologically similar to that of the tongue base and the lymph node metastasis removed previously from the left neck (Fig. 3(B)). Thus, a diagnosis of bilateral cystic neck metastases from SCC of the tongue base was made. All the lymph nodes removed in the radical neck dissection, except the cystic ones, were negative for cancer. The patient has remained free from local and regional recurrence, and from remote metastasis for the one year and nine months since the second surgery.

Discussion

The differential diagnosis of a cystic cervical lymph node metastasis of SCC and a BCCC is very difficult in the case of an occult primary site of the SCC. However, extremely few cases that satisfy the diagnostic criteria proposed by Martin et al.[1] for BCCC have been reported,[1,2,6,8–10] and the great majority of cases reported as cystic SCCs in the lymph node of the upper neck have ultimately been shown to originate in Waldeyer's ring.[1,3,4] Although the average time to discovery of the primary is reported to be 12.4 months, most primaries are found within six months of the initial presentation of the cystic mass.[3] The current case had bilateral metastases, and the disease initially occurred contralateral to the primary site. Although unusual, given that lymph nodes in Waldeyer's ring form a circle at the transition between head and neck,[13] the initial presentation as a mass in the contralateral neck is not particularly curious.

Cystic SCCs of the neck with an occult primary lesion have been successfully treated with radiotherapy, radical neck dissection, or a combination.[1–3,5,6] However, since many of these cystic metastases are solitary[2,3] and the initial surgical excision of the neck mass probably removes all the metastatic disease,[3] the current case was followed-up periodically without additional treatment, in an attempt to find a clinically occult primary lesion.

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