Squamous Cell Carcinoma Arising from Thyroglossal Duct Remnants: Report of a Case and Results of Immunohistochemical Studies

YOSHIHISA HAMA, AKIRA SUGENOYA, SHINYA KOBAYASHI, NOBUO ITOH, and JUN AMANO
Departments of 1 Surgery and 2 Pathology, Shinshu University School of Medicine, 3-1-1 Asahi, Matsumoto 390, Japan

Abstract: We report on the case of a 57-year-old male found to have squamous cell carcinoma (SCC) arising from thyroglossal duct remnants. The patient presented with an asymptomatic tumor in his anterior neck which was immovable on palpation. Aspiration biopsy cytology revealed class V malignancy with many atypical clusters and marked keratinization. After preoperative radiation, a radical operation employing Sistrunk's procedure with bilateral neck dissection was performed. Histopathological examination confirmed a diagnosis of moderately differentiated SCC, but revealed ciliated columnar epithelium in the walls of the cyst without a normal layer of squamous cells. Furthermore, immunohistochemical studies demonstrated the tumor to be negative for thyroglobulin staining, but positive for cytokeratin and carcinoembryonic antigen. These histopathological findings proved attributable to squamous metaplasia occurring in the ciliated columnar epithelium of the thyroglossal duct. Thus, SCC might originate in the metaplastic portion of the thyroglossal duct remnants. Although the prognosis associated with SCC in the thyroglossal duct is not as optimistic as that associated with papillary carcinoma, no evidence of recurrence has been observed in this patient in the 7 years since his operation. This suggests the effectiveness of our therapeutic approach for this unusual disease.

Key Words: thyroglossal duct remnants, squamous cell carcinoma, metaplasia

Introduction

Thyroglossal duct remnants represent the most common congenital anomaly, characterized by a swelling mostly centered in the anterior neck. These remnants rarely become malignant, although there have been about 100 reported cases of such malignancies, the majority of which were papillary thyroid adenocarcinoma. Only nine cases of squamous cell carcinoma (SCC) arising from thyroglossal duct remnants have been documented; however, no detailed histopathological description regarding the precise origin of the SCC in such cases has been published. We report herein an extremely rare case of SCC arising from thyroglossal duct remnants, the definitive diagnosis of which was proven by immunohistochemical techniques. The various characteristic findings of diagnostic imaging techniques are also described.

Case Report

The patient was a 57-year-old male who first noticed an asymptomatic tumor in his anterior neck in 1988. As the tumor was initially suspected to be a purulent mass, he was treated with antibiotics by his family physician; however, the tumor did not shrink, and he was therefore referred to our department. He had noted no change in voice or any difficulty in swallowing. He had no previous history of any serious illness, operations, or hospitalizations.

On admission, an immovable tumor was palpated in the anterior neck at the level of the hyoid bone, which was 4.2 cm x 3.2 cm in size, nodular, firm, and ill-defined, but not tender. The thyroid gland and pathological cervical lymph nodes were not palpable (Fig. 1). A chest X-ray was unremarkable, but X-rays of the soft tissues of the neck showed microcalcified deposits confined to the tumor. Ultrasonography revealed a tumor, 3.1 cm x 3.6 cm in size, with a solid pattern, not extending to the thyroid gland. The hypoechoic lesion showed echogenic dots inside the tumor. No other specific findings were observed. A 123I scintigram demonstrated a normal thyroid gland in the normal site with no ectopic thyroid tissue, and a 201Tl scintigram showed no abnormal accumulation in either the early or late
phases. However, a $^{67}$Ga scintigram revealed an apparent positive accumulation confined to the tumor. Both computed tomography (CT) and magnetic resonance imaging (MRI) showed the tumor to be located in front of the epiglottis, involving the hyoid bone. The border of the adjacent muscles was preserved and multilocular cysts were visible in the tumor. Laryngoscopic examination showed the epiglottis to be displaced to the left by the tumor, but the vocal cord was intact. Thyroid function was euthyroid. Serum SCC antigen (SCCA) and carcinoembryonic antigen (CEA) were slightly elevated at 1.8 ng/ml and 4.1 ng/ml, respectively, the normal levels being <1.5 ng/ml and <2.0 ng/ml, respectively. The aspiration biopsy cytology revealed class V malignancy, the cytological analysis showing many separate atypical cells with bizarre nuclei. A significant number of altered cells, almost rectangular in shape, were also found, which were classified as keratinized epidermoid carcinoma cells.

External radiation was applied to the neck preoperatively to a total dose of 40 Gy for about 1 month to prevent local invasion and reduce the size of the tumor. However, the tumor was not diminished by this radiotherapy, and 2 weeks later a radical operation employing Sistrunk's procedure with bilateral neck dissection was performed.

**Operative Findings**

A well-encapsulated mass was found above the hyoid bone, extending to the thyroid cartilage. The many cavities in the tumor contained a small amount of necrotic debris. The mass was adhered to part of the strap muscles, which were also resected. A frozen section of the surgical margin revealed no cancerous invasion.

**Gross Findings of the Specimen**

The resected specimen was an ellipse-shaped mass, 4.3 cm × 2.5 cm in size. Multilocular cystic lesions were observed on the cut surface. The cystic wall was yellowish-white in color and had a rough surface. A malignant lesion was revealed on the tip of the posterior aspect of the hyoid bone where the cyst had been formed. It extended beyond the walls of the cyst and invaded the surrounding connective, adipose, and muscular tissues (Fig. 2).

**Histopathological Findings**

The cancerous part of the tumor was shaped in a somewhat rectangular fashion resulting from the disorganized multiplication of cancerous cells in a fairly large-sized mass. Degeneration and necrosis of the cancerous cells, as well as the formation of granulation tissues, were observed in the foremost invaded areas. These findings might have been the result of radiotherapy. The inner surface of the cyst was covered with a thick layer of cancerous cells (Fig. 3). Some areas were found to be covered with ciliated columnar epithelium,