[症例報告]Carcinoma of the Thyroglossal Duct Cyst : A Case Report and a Brief Review of Literatures

Ryukyu Med. J, 7(1): 45~50, 1984.

Carcinoma of the Thyroglossal Duct Cyst: A Case Report and a Brief Review of Literatures

Shinichiro Koga, Yoshihiro Muto, Akira Satomi
Osamu Hidaka and Yoshiyuki Sho

The First Department of Surgery, School of Medicine,
University of the Ryukyus

Key words : thyroglossal duct cyst, papillary carcinoma, surgical treatment

Abstract

Carcinoma of the thyroglossal duct cyst in a 38-year-old male patient is presented herein reviewing literatures. The cystic mass, 2 × 2.5 × 3 cm. in size, was located in the midline of the neck below the hyoid bone and was easily excised without any troubles, since there were no invasion into surrounding tissues and lymph node metastases. Microscopically, foci of papillary carcinoma were scattered on the inner surface of the posterior wall of the cystic mass. The patient has been well ten months after surgery.

Introduction

Benign thyroglossal duct remnants (cysts, fistulae and masses) are occasionally encountered in the clinical practice at the large hospital, but carcinoma arising from the thyroglossal duct remnant is a rare entity. Following the first reported case in 1915, about 100 cases of carcinoma originating in the thyroglossal duct remnant have been documented in Europe and America[1,2], and several instances in Japan have been reported in literatures.

In this paper, a case with carcinoma of the thyroglossal duct cyst surgically treated by us at the Ryukyu University Hospital is presented reviewing literatures on incidence, histologic characteristics and rationale for the treatment of this malignant lesion.

Case Report

A 38-year-old man was admitted at the Ryukyu University Hospital on September 6, 1983 with a four-year history of a slow evolution of a cystic mass in the midline of the neck. There were no symptoms associated with the mass.

A cystic mass, about 2.5cm, in size, was palpable in the midline of the neck just below the hyoid bone. The thyroid gland was found to be essentially normal in

palpation with no abnormality of thyroid scintigram. There was no cervical lymphadenopathy. Ultrasonography revealed a cystic lesion with hyperechogenic areas in the mass, suggestive of calcification of the cyst wall.

It was diagnostic of a benign thyroglossal duct cyst preoperatively, and simple excision of the cystic mass was carried out on September 20. The cystic mass was located in the midline of the neck just below the hyoid bone and attached to the hyoid bone by a fibrous stalk. The mass was easily removed and found to be not associated with the thyroid gland. The regional lymph nodes were not enlarged.

The excised cystic mass was 2 × 2.5 × 3 cm. in size. The outer surface of the mass was smooth and it contained yellow-brown and turbid fluid. However, a thick and shaggy lesion on the inner surface of the posterior wall, which was four times as thick as the anterior wall, was found.

Histologically, the epithelial lining on the inner surface of the cyst wall was entirely desquamated. The cyst wall was almost composed of fibrous to hyalinized connective tissue, however scattered foci of papillary carcinoma identical to carcinoma of the thyroid gland were evident on the shaggy posterior wall. Calcification was also scattered on the inner surface of the wall (Fig. 1). The tumor cells were cuboid with slight cellular and nuclear pleomorphism. The each tumor cell was arranged in a layer and hyalinized connective tissues were projected into the lumen of the cyst (Fig. 2). Areas of irregular calcification were found in the connective tissue stalk.

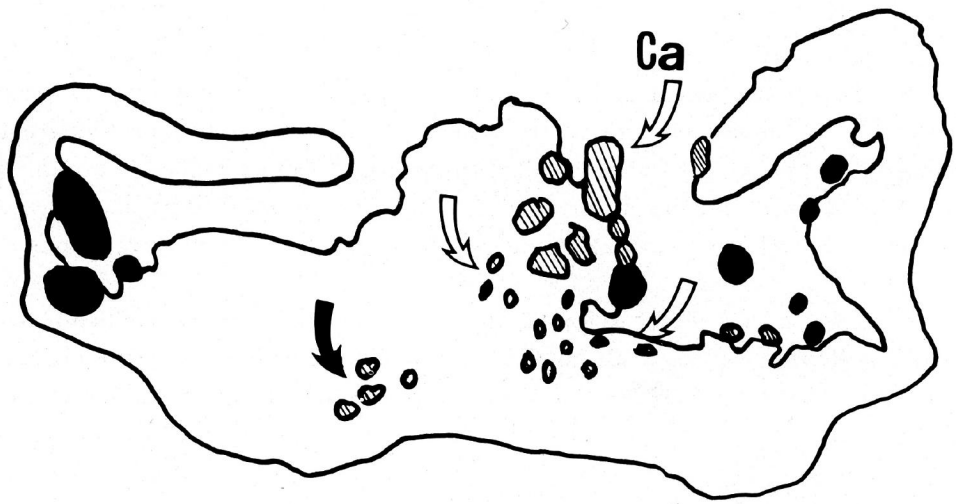

Fig. 1. Schematic illustration of cross section of the specimen
Papillary carcinoma (striped) shows infiltration into the cyst wall (white arrows) and is noted within the confines of the cyst wall. Calcification (black) and normal thyroid tissue (black arrow) are present.

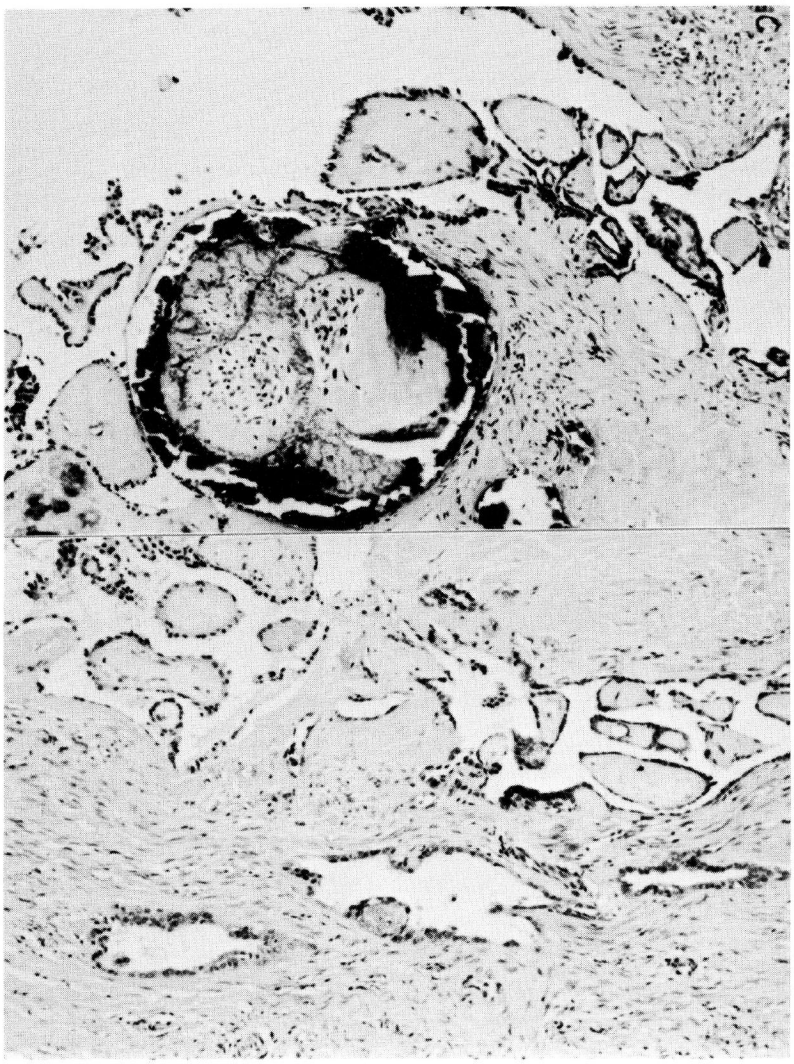

Fig. 2. Microscopic findings of papillary carcinoma
Papillary projections of carcinoma is noted. The stalk is thickened, hyalinized and partially calcified (top). Invasion of carcinoma in follicular pattern into the cyst wall is demonstrated (bottom) (HE, ×100).

Invasive follicular carcinoma was also evident in the cyst wall. The tumor cells showed more cellularity and pleomorphism. A few normal thyroid tissues with colloid were present in the cyst wall (Fig. 3).

The postoperative course was uneventful and the patient has been well ten months after operation.

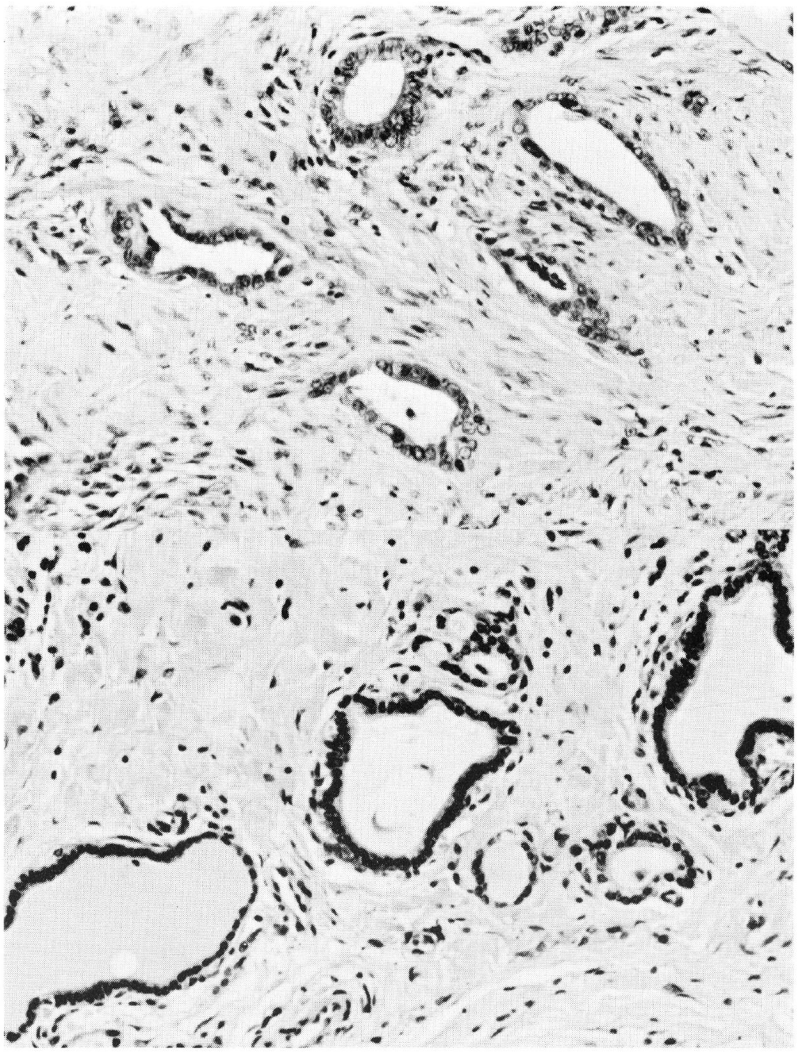

Fig. 3. Microscopic findings of carcinoma and normal thyroid gland
Invasive carcinoma is of follicular type with cellularity and pleomorphism (top). The normal thyroid follicles are filled with colloid (bottom) (HE, × 200).

Discussion

Surveying literatures[3)-7)] in Japan, up to March 1984, 6 cases of carcinoma arising from the thyroglossal duct remnant, including our case, were acceptable for clinical and pathological analysis.

Patients were seen from 11 to 66 years with an average of 35 years and frequently in the third decade. Although several cases in children under 10 years of age have been

documented in Europe and America[2], none of cases under 10 years have been reported in Japan[3)-7)]. In Japanese cases, the disease was uncommon in men, only one of six being male patient, while no distinct predominance of female was demonstrated in Europe and America. All of the patients were asymptomatic.

The mass was located in the midline of the neck just below the hyoid bone in the majority of cases, but in one instance the mass was present above the hyoid bone. The size of the tumors ranged from 2 to 6 ㎝. with an average of 3.6 ㎝. The duration of the history of slow evolution of the mass was one month to 4 years before operation in Japanese cases, while it was longer than one year, in some cases up to 15 years, in Europe and America[1)2)].

The lesion was usually cystic and was filled with yellow to brownish fluid, except for a case in which it was solid. The gross feature of the lesion was closely similar to the appearance of benign thyroglossal duct cyst. Therefore, it is usually difficult to make a definite diagnosis of carcinoma of the thyroglossal duct cyst. In the majority of cases, however, small nodules or papillary projections from the cyst wall were noted by careful examination of the specimen.

Microscopically, the type of carcinoma was papillary carcinoma in all cases in Japan, while adenocarcinoma, squamous cell carcinoma and anaplastic carcinoma were rarely found in Europe and America[1)2)8)9)].

In regard with the diagnostic criteriae for carcinoma of the thyroglossal duct remnant, the following conditions have been proposed[8)]; 1) no other carcinomas, 2) presence of carcinoma in the location of the thyroglossal duct remnants, 3) demonstration of normal thyroid tissue in the cyst wall not in continuity with the thyroid gland, and 4) evidence pf carcinoma invasion into the surrounding tissue. In our instance, carcinoma was confined in the cyst wall in which the normal thyroid tissues and invasive foci of carcinoma into the outer layer of the cyst wall were evident. Accordingly, our case fulfilled these criteriae.

The histological diagnosis of the thyroglossal duct remnant depends upon the demonstration of both thyroglossal duct epithelium and thyroid tissue in the cyst wall. There are two epithelial components present in the thyroglossal remenants. Therefore, the origin of thyroglossal duct carcinoma may be two sources; the one arising from the duct epithelium (cuboid to columnar, squamous), the other from the thyroid tissue in the cyst wall. In the former the histologic type is adenocarcinoma or squamous cell carcinoma, in the latter papillary or follicular carcinoma identical to carcinomas of the thyroid gland. The most common type of carcinoma was papillary or follicular carcinoma originating in the thyroid tissue in the cyst wall. Although Nussbaum et al.[9)] porposed that papillary carcinoma of the thyroglossal duct remnant represented metastatic lesion of the thyroid gland, these cases reported as thyroglossal duct carcinoma fulfill the aforementioned diagnostic characteristics and support primary carcinoma originating in the thyroglossal duct.

Preoperatively, the lesion was diagnosed to be a benign thyroglossal duct remnants in almost all of cases. Accordingly, the initial operation was excision of the cyst in

all patients except several patients who had thyroidectomy and nodal dissections. Concerning surgical treatment, it is widely accepted that when there is no evidence of extension of carcinoma beyond the cyst wall, excision of the cyst is of choice and further surgical intervention is not indicated, since postoperative prognosis in such cases is favorable without recurrences.

References

1) Hans, S.S., Lee, P.T. and Proctor, B. : Carcinoma arising in thyroglossal duct remnants. Am. Surg. 42 : 773-777, 1976.
2) Roses, D.F., Snively, S.L., Phelps, R.G., Cohen, N. and Blum, M. : Carcinoma of the thyroglossal duct, Am. J. Surg. 145 : 266-269, 1983.
3) Yoda, M. and Ishikawa, T. : A case of thyroglossal mass with malignant feature. Hormone Clin. 12 : 188, 1964 (in Japanese).
4) Kaneko, H. : A case of malignant goiter of a girl. Kitakanto Igaku 15 : 478-481, 1965 (in Japanese).
5) Nagamine, N., Miyagi, Y. and Sho, Y. : A case of carcinoma arising in the thyroglossal duct remnants : a case report and review of the literature. J. Clin. Cancer 22 : 793-798, 1979 (in Japanese).
6) Inuyama, S., Takasaki, T., Ozu, R., Kawashiro, N., and Inuyama, N. : papillary adenocarcinoma arising from the remnants of the thyroglossal duct. a case report. J. Otolaryngol. 49 : 635-637, 1977 (in Japanese with English abstract).
7) Yoshida, A., Kubota, T., Kaku, M., Matsui, K., Shimono, T. and Sakugawa. H. : Carcinoma in thyroglossal duct remnants. J. Otolaryngol. 54 : 935-936, 1982 (in Japanese with English abstract).
8) White, L.L. and Talbert, W.M. : Sguamous cell carcinoma arising in thyroglossal duct remnant epithelium. Otolaryngol. Head Neck Surg. 90 : 25-31, 1982.
9) Nussbaum, M., Buchwald, R.P., Ribner, A., Mori, K. and Litwins, J. : Anaplastic carcinoma arising from median ectopic thyroid (thyroglossal duct remnant). Cancer 48 : 2724-2718, 1981.