



Isolated Papillary Thyroid Carcinoma in Thyroglossal Duct Cyst

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Abstract

Thyroglossal duct cysts are the most frequently encountered congenital neck masses having a malignancy rate of 1%. The most common malignant form is papillary thyroid carcinoma. Synchronous papillary thyroid carcinoma developing in the thyroglossal duct cyst is rarely seen. Synchronized papillary carcinoma can be described as a multifocal tumor or a metastatic lesion on a thyroglossal duct cyst. In this study, we presented a 29-year-old female patient with a midline neck mass who underwent thyroglossal duct cyst excision and had papillary thyroid carcinoma in the thyroglossal duct cyst. Total thyroidectomy was performed, and histopathological examination showed no malignancy of the thyroid gland.

Keywords: Thyroglossal duct cyst, papillary thyroid cancer, congenital neck masses

Introduction

Thyroglossal duct cysts are the most commonly seen congenital anomalies located in the midline of the neck. It can be found along the region from the foramen cecum to the sternum and occurs due to the partial or total lack of obliteration of the thyroglossal duct, which the thyroid gland forms while extending from the base of the tongue to its place in the neck in the fetus (1).

The development of carcinoma from a thyroglossal duct cyst is rare and is detected in approximately 1% of cysts. In total, 85% of malignant cases comprise papillary thyroid carcinoma (2). While there are papillary carcinoma foci in the thyroid gland in some cases, others have no extra foci. In literature, the pathophysiology of synchronous papillary carcinoma in a thyroglossal duct cyst and the thyroid gland is still controversial.

In this study, it was aimed to discuss papillary thyroid carcinomas of isolated and synchronous thyroglossal duct cysts by reviewing literature.

Case Report

Two years ago, a 29-year-old female patient had applied to an external health center with the complaint of a swelling that was located in the midline portion of the neck and mobile with swallowing for approximately 4–5 years. A fine-needle aspiration biopsy specimen had been taken from the lesion, and she was reported to have cytological findings consistent with those of a thyroglossal cyst. The patient had refused to be operated then. The patient then applied to our clinic with the same complaint. She had no other accompanying complaint such as rash and discharge around the mass. She did not have difficulty in breathing, dysphagia, sore throat, and hoarseness. She had a history of smoking for 15 packs-year and social alcohol use. She had no history of radiotherapy on the neck region. In the physical examination, an approximately 1 cm in diameter and medium-hard lesion, which was mobile on swallowing and tongue movements, was palpated in the midline of the neck, at approximately 3-finger width above the jugulum. No palpable lesion was observed in the neck and thyroid gland.

In the ultrasonographic examination, there was a mass lesion that was thought to be solid, not demonstrating an apparent vascularization in the color Doppler ultrasonography, including punctate calcific foci in hard-margined hypoechoic nodules in the midline of the neck. Magnetic resonance imaging of the neck revealed a mass lesion that was subcutaneously extending between the strap muscles anterosuperior to the thyroid cartilage, superior to the thyroid gland, and that was demonstrating a hypointense diffuse-contrast involvement at T1A and hyperintense diffuse-contrast involvement at T2A. Ultrasonography of the neck and magnetic resonance imaging did not reveal any pathologic lesion in the thyroid gland, and thyroid function tests were normal.

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Written and verbal informed consent was obtained from the patient for performing the Sistrunk operation. The cystic lesion in the subcutaneous tissue was reached, and it was removed with hyoid bone corpus by following the cyst duct. The macroscopic size of the lesion was approximately 3×2×1 cm (Figure 1).

In the microscopic examination, a cystic tumor tissue that was neoplastic in appearance covered the epithelium; it was observed to have a fibrous wall. Normal thyroid tissue was found in the cyst wall (Figure 2). Papillary structures included clear nucleated neoplastic cells with nuclear clefts and intranuclear inclusion bodies. It was observed that the tumor was encapsulated and that it developed in the thyroid tissue, demonstrating a neoplastic feature (Figure 3).

There was no change in the thyroid function tests of the patient who did not have any problem during the postoperative period. In the control ultrasonography of the neck performed during this period, a 10 mm in diameter, well-demarcated, suspected hypoechoic nodular mass was observed in the left lobe, caudal to the thyroid gland. No extra-regional lymphadenomegaly and pathologic echo of the mass were found in the neck. To facilitate the patient's follow-up and to evaluate the existent nodule, total thyroidectomy was performed in the postoperative 2nd month. There was no malignancy in the thyroid gland and in the nodule in the caudal region of the left lobe. The patient was evaluated in cooperation with the clinic of nuclear medicine and applied postoperative radioactive iodine therapy.

Discussion

The thyroid gland, which is the first endocrine gland to develop during embryogenesis, completes its migration from the tongue base to its known final anatomic location in the 7th week. Thyroglossal ducts created during this migration lose the feature of ducts and then disappear in time. However, some parts of the ducts can be persistent. As a result of the continuing secretions of the cells constituting the duct, a thyroglossal duct cyst can develop between the hyoid bone and thyroid gland (3).

The wall of a thyroglossal duct cyst is covered by ciliated respiratory epithelium and/or squamous epithelium, and its some part larger than 62% includes normal thyroid tissue.

The development of carcinoma inside a thyroglossal duct cyst is rare, but papillary carcinoma, mixed papillary follicular carcinoma, squamous cell carcinoma, adenocarcinoma, and anaplastic carcinoma account for 85%, 7%, 5%, 2%, and 1% of malignant cases, respectively (4).

The development of thyroid cancer in thyroglossal duct cysts is still controversial. Some authors suggest that it results from the islets of normal thyroid tissue in thyroglossal duct residues. This theory gets stronger with the determination of ectopic thyroid tissue islets in the histopathological examination of 62% of a thyroglossal duct cyst.

Papillary carcinoma of a thyroglossal duct cyst can synchronously coexist with papillary carcinoma of the thyroid gland, or they can be coincidentally detected as an isolated focus.

The diagnostic criteria of primary thyroglossal duct cysts were defined by Widström. According to these criteria, the presence of a cyst including normal thyroid follicles on their wall and being

lined with epithelium and the absence of any primary tumor in the thyroid gland must be demonstrated (5).

In a study conducted on 6 patients with carcinoma of a thyroglossal duct cyst, carcinoma in the thyroid gland was detected in 4 patients (66%), and the rate of patients with carcinoma synchronized with the thyroid gland was found to be high (6).

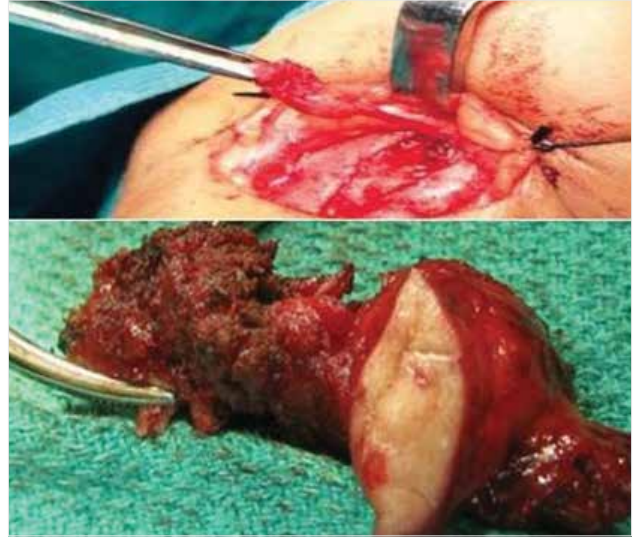


Figure 1. Intraoperative and postoperative macroscopic images

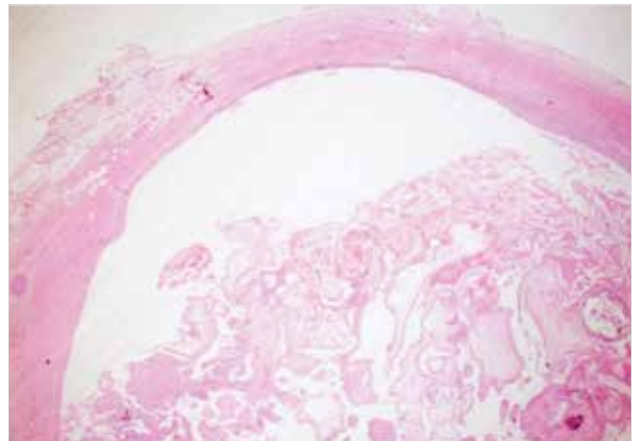


Figure 2. Partial general view. Cystic tumor tissue under the capsule of thick connective tissue (HE 40×)

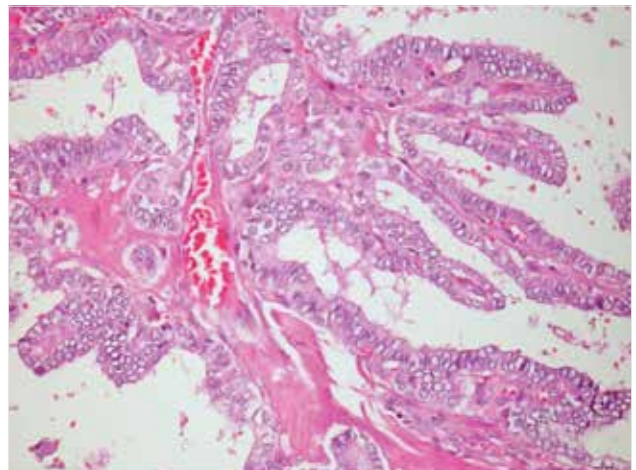


Figure 3. Papillary thyroid carcinoma (HE 200×)

In another study, the prevalence of occult thyroid carcinoma was found to be between 11% and 27% in patients with thyroglossal duct carcinoma and in whom thyroidectomy was applied (7).

Studies in literature do not provide sufficient information on the frequency of the development of carcinoma in the thyroid gland and thyroglossal duct cyst and their relationship with each other because they generally include a single or a few patients with papillary thyroid carcinoma.

Papillary carcinomas that develop in thyroglossal duct cysts and synchronously in the thyroid gland can be explained by the fact that papillary carcinomas can synchronously develop as multifocal. Alternatively, some authors evaluate them as a metastasis from the primary focus in the thyroid gland into a thyroglossal duct cyst (8).

Based on studies showing that carcinomas can also be detected in the thyroid gland in few patients with a thyroglossal duct carcinoma, it can be concluded that the primary focus may be a thyroglossal duct cyst rather than the thyroid gland and that papillary carcinoma that synchronously develops in the thyroid gland can be evaluated as a multifocal primary tumor.

Studies demonstrating the high rate of synchronous carcinoma support the view that carcinoma develop as a metastasis from the thyroid gland to a thyroglossal duct cyst. The authors suggesting that papillary carcinoma is the spread of an occult thyroid carcinoma believe that the thyroglossal duct is the natural way for the metastasis of thyroid carcinoma. However, the role of the thyroglossal duct as the natural way, which is the basis for the metastatic spread theory, can also be true for the spread of carcinoma through the cyst into the thyroid gland.

For these reasons, the application of thyroidectomy for carcinoma of a thyroglossal duct cyst is still controversial. There are some studies reporting that the prognosis is good in patients monitored without thyroidectomy. However, because of a few of these case series and patients, treatment strategies are not standardized. Because some authors evaluate these tumors as the metastasis of occult thyroid cancers, they accept total thyroidectomy as a part of an effective treatment (9). Moreover, surgery is recommended in adults due to synchronous carcinoma in the thyroid gland and a thyroglossal duct cyst. Total thyroidectomy, which is performed with the Sistrunk operation, is an effective treatment method for carcinoma of a thyroglossal duct cyst (10). In our case, because papillary thyroid cancer was detected on the base of the thyroglossal duct cyst after the Sistrunk operation, the patient was closely followed-up. Because control ultrasonography of the neck revealed a 10-mm nodule in the left lower lobe of the thyroid gland in the postoperative 2nd month, total thyroidectomy was performed to find any occult carcinoma focus in the thyroid gland and to facilitate the follow-up of the patient. It is suggested that elective neck dissection has no role in the treatment of the N0 neck. In our case, neck dissection was not needed because no nodule was suspected in the examination and imaging.

Postoperative radioactive iodine therapy and thyroid suppression therapy are recommended for these patients (7, 10). Therefore, when the diagnosis of papillary thyroid cancer is previously made in a thyroglossal duct cyst with fine-needle aspiration biopsy, si-

multaneous total thyroidectomy is recommended. When it is made after the excision of a thyroglossal duct cyst, total thyroidectomy is recommended to be performed later. Thus, thyroglobulin and body scanning can be used for determining the development of metastasis in long-term follow-ups. Our patient was given radioactive iodine therapy after total thyroidectomy, and she is still being followed-up in the clinic of nuclear medicine.

Conclusion

Cancer can sometimes develop from a thyroglossal duct cyst. The most common of these cancers is papillary microcarcinoma, which can occur due to a de novo thyroglossal duct cyst or develop as a metastasis of thyroid gland carcinoma or multicentrically with the thyroid gland. For treatment and follow-up procedures to be performed, total thyroidectomy must be performed in addition to the excision of thyroglossal duct cyst and neck dissection must be applied in the presence of lymphadenopathy. If there is metastasis, radioactive iodine therapy must also be planned.

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