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A Case of Malignancy in a Thyroglossal Duct Cyst—Recommendations for Management

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Thyroglossal duct cyst carcinomas are rare tumors with just more than 200 cases published to date. This is a case report of a thyroglossal duct cyst harboring an occult carcinoma for which a Sistrunk operation was performed. Histopathological examination revealed a papillary carcinoma arising from a thyroglossal duct cyst after which the patient underwent a total thyroidectomy. With current evidence-based guidelines lacking, we discussed some of the issues relevant to the surgical planning and postoperative management of such a patient.

Keywords: thyroid ■ cancer ■ tumor

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INTRODUCTION

Thyroglossal duct cysts (TGDCs) represent one of the most common differentials for a congenital midline neck cyst or mass. After diagnosis, this is usually treated by a Sistrunk operation. A well-differentiated carcinoma arising in a TGDC, whilst a rare occurrence, is usually clinically indistinguishable from its benign counterpart and an incidental finding after surgical resection. This is a case report of a papillary TGDC carcinoma for which a Sistrunk procedure, followed by a total thyroidectomy, was performed. This report reviews the current data as published in the world literature and highlights some of the controversies surrounding further management.

CASE REPORT

An 18-year-old obese female presented with a 4×2 cm nontender, firm midline mass in the infrahyoid region that had been associated with dysphagia for 2 months. It demonstrated vertical movement on swallowing and protrusion of the tongue, and the thyroid gland was palpably normal with no cervical lymphadenopathy. She was in good health except for obesity; past surgical and medical history was unremarkable. A neck ultrasound showed a 2.8×2.1-cm, well-defined complex infrahyoid mass (mainly cystic with multiple septations (Figure 1) separate from the thyroid. There was no focal cervical lymphadenopathy. computed tomography scan demonstrated similar features compatible with a diagnosis of a TGDC (Figure 2). Thyroid function tests were normal. A Sistrunk procedure was performed (frozen section was not available intraoperatively). Histopathology revealed a 4-cm diameter fluctuant mass, which, on sectioning, revealed a multiloculated cyst filled with gelatinous material. In addition, a papillary thyroid carcinoma was noted at one pole with tumor infiltration but incomplete penetration of the cyst wall (Figure 3). Two months later, the patient underwent a total thyroidectomy without nodal/compartmental dissection. The thyroid appeared clinically normal with no palpable nodes at operation. Pathological examination of the thyroid gland revealed a normal gland with no evidence of carcinoma.

Postoperative serum thyroglobulin was within normal range. She is currently being followed up as an outpatient on thyroid hormone suppression therapy awaiting a radioactive iodine scan. Thyroglobulin levels are being monitored as well.

DISCUSSION

TGDCs are uncommon and thought to arise as a result of the failure of the thyroglossal duct to involute. Ellis and van Nostrand suggested that this failure occurs in approximately 7% of the adult population.[1] However, TGDCs are the most common congenital midline swellings that present to the general surgeon. These benign thyroglossal duct remnants may comprise up to 75% of midline neck masses in children and are found in up to 7% of adults.[1]

The finding of an incidental malignancy within this cyst is rare and within a thyroglossal duct remnant after simple excision even more infrequent.[2] The true incidence of malignant transformation of TGDC is unknown[3,4] but estimated at approximately 1%. A neoplasm arising from a TGDC was first described in the literature by Brentano[5] in 1911; a search by Luna-Ortiz et al revealed only 215 cases in the world literature.[6] The largest study to date

found only 14 cases of thyroid cancers in 1075 patients with thyroglossal cysts[7] over the period 1971-1995. Papillary neoplasms were predominant.

In the past, there has been some speculation as to the origin of malignancy within a TGDC. However, it has been demonstrated histologically, that up to two-thirds of TGDCs contain thyroid tissue with its inherent propensity to malignant transformation just as possible.

In most cases documented in the literature as well as our own presented here, the diagnosis of malignancy was not suspected preoperatively either on clinical examination or intraoperatively.[8-10] Preoperative imaging may not be able to diagnose a malignancy. Fine-needle aspiration cytology has a positive yield of approximately 66%.

Some surgeons consider a Sistrunk procedure as adequate, while others add a subtotal or total thyroidectomy. With no large studies and no long-term follow-up of the treatment modalities available, recommendations have been suggested with respect to indications for total thyroidectomy. These include finding a concomitant primary thyroid cancer, which occurs in 11% to 33% of cases, invasion into cyst wall, and tumors larger than 1 cm. Complete removal of all thyroid tissue also allows long-term monitoring with thyroglobulin levels and radioiodine ablation therapy if needed. There is great debate in the management of primary papillary thyroid carcinomas that are less than 1 cm in size, with some suggesting hemithyroidectomy alone is enough, avoiding total thyroidectomy with radioactive iodine and thyroid suppression.

The rarity of a diagnosis of carcinoma arising from a TGDC precludes consensus on the further course of management with the potential benefits of more aggressive treatment vs the lifelong consequences of total thyroidectomy and the inherent risks associated with surgery, radioactive iodine ablation/treatment, and lifelong hormone suppression therapy. In our patient, the final pathology of the thyroid was negative for malignancy, which adds fuel to the debate.

There are some who advocate that consideration be given to fine-needle aspiration cytology of all midline cystic masses in the neck. Whilst the risk of carcinoma may be small, the benefit of a planned, single operative procedure is obvious. The low diagnostic yield of fine-needle aspiration cytology is advantageous, especially with large cystic lesions with small mural tumor. We, however, believe the yield to be too low to warrant nonselective application and also that it should be reserved for evaluation of suspicious nodules in the thyroid. However, histological evaluation is absolutely necessary.

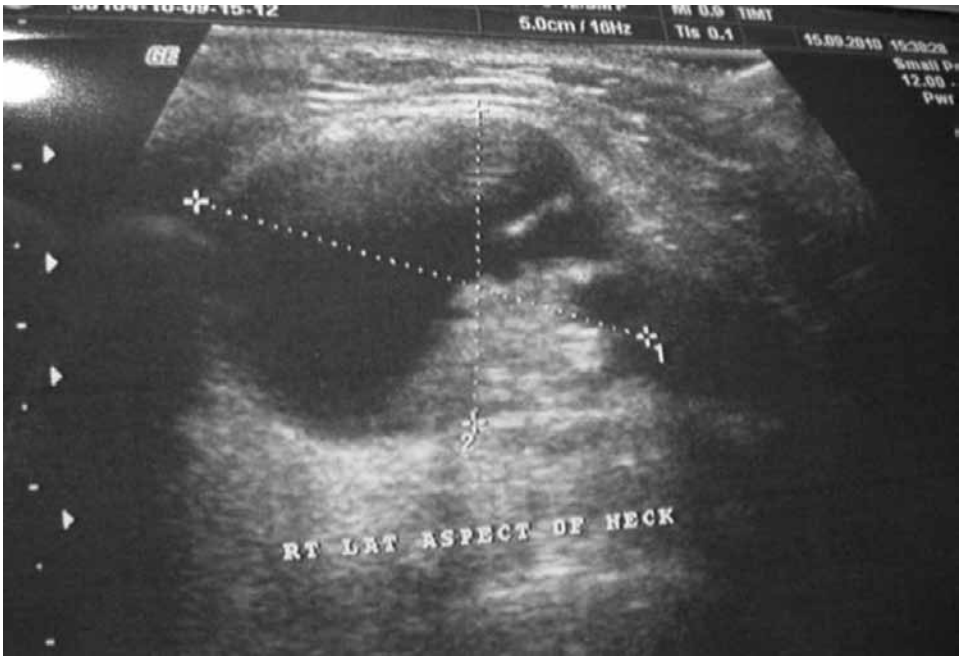
Guidelines based on level 1 evidence are currently unavailable due to the absence of large studies and long-term follow-up. If it is possible to stratify the risk for patients, higher-risk patients can be offered more aggressive treatment options.

It is well known that papillary cancer has a prolonged clinical course so that long-term follow-up is recommended (>20 years).

CONCLUSION

The Sistrunk operation may be adequate treatment for most patients with an incidental finding of a malignancy arising from a TGDC, but to date there exist no clear consensus guidelines on the further management. There

Figure 1. Cystic Mass



may, however, be a role for identifying higher-risk groups who may benefit from more aggressive treatment as one would treat a well-differentiated thyroid carcinoma. In the absence of large prospective studies, the evidence obtained from case reports and small case series suggest that the safer option is extirpation of the gland.

Figure 2. Thyroglossal Duct Cysts

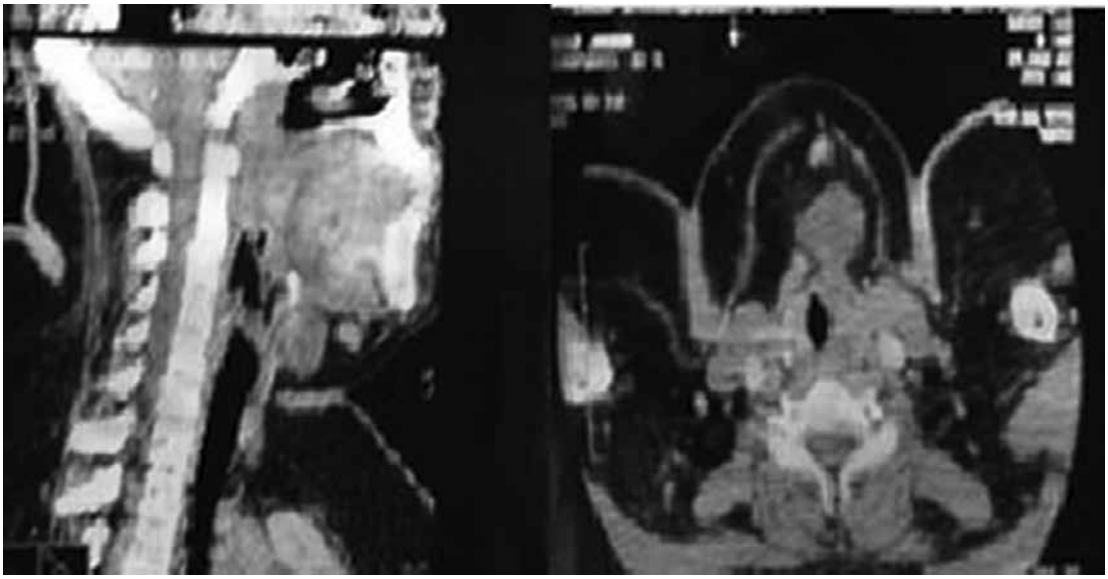
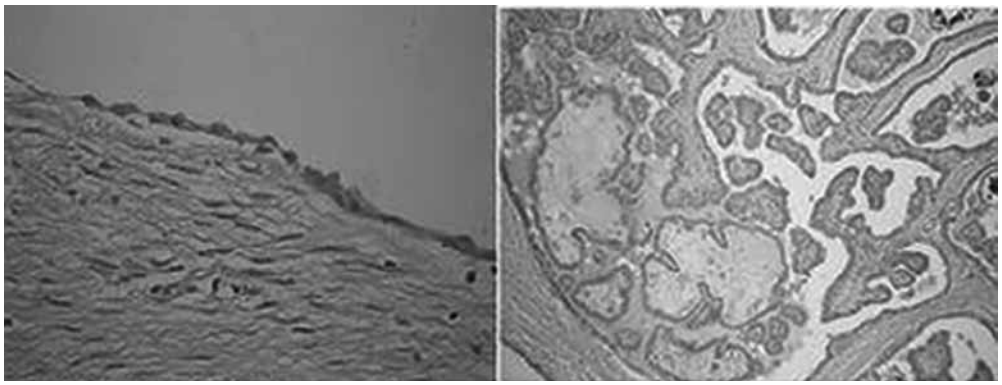


Figure 3. Histology of Thyroglossal Duct Carcinoma

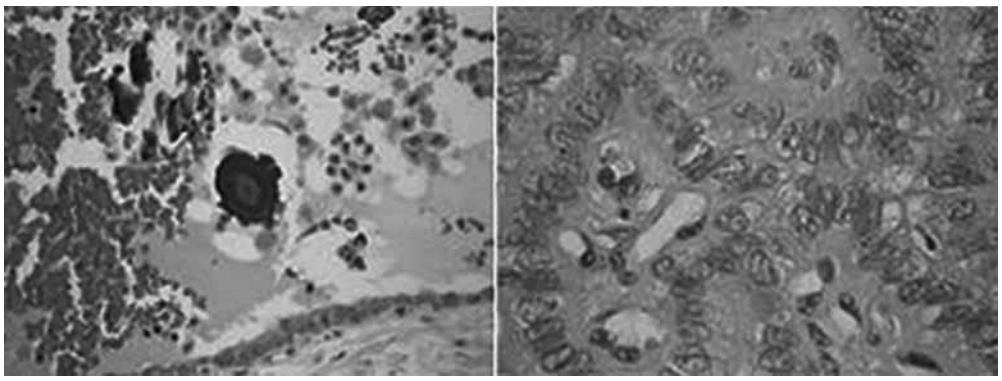
A Demonstrates cuboidal epithelial lining of the duct cyst

B Papillary architecture



B Psammoma body

D Showing nuclear grooving (×100)



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