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Case Report

Papillary Thyroid Carcinoma within Thyroglossal Duct Cyst: Case Report and Review of Literature

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Submitted: 25 October 2016

Accepted: 11 Novemebr 2016

Published: 13 Novemebr 2016

Keywords

- Thyroid cancer
- Thyroglossal duct cyst

Abstract

Approximately 1.5% of all thyroglossal duct cysts contain a thyroid malignancy (most commonly papillary thyroid carcinoma), which are usually diagnosed incidentally on histopathology. However, in the rare case of thyroid cancer within a thyroglossal duct cyst that is diagnosed preoperatively, the extent of surgical resection remains variable without consensus across institutions. We therefore present a case of classical papillary thyroid cancer within a thyroglossal duct cyst that was diagnosed preoperatively and discuss the current literature and controversy regarding the management of this condition.

ABBREVIATIONS

TDC: Thyroglossal Duct Cyst; PTC: Papillary Thyroid Cancer; FNA: Fine-Needle Aspiration; US: Ultrasound

INTRODUCTION

Thyroglossal duct cysts (TDCs) are benign congenital anomalies of the anterior neck thought to arise from epithelial remnants of the thyroglossal duct during caudal descent of the thyroid gland in early gestation. Up to approximately 60% of TDCs contain normally functioning thyroid cells, however series have shown that approximately 1-1.5% of all TDCs contain thyroid cancer [1,2]. As with *de novo* thyroid cancer, the most common histologic subtype of malignancy within TDCs is papillary thyroid cancer (PTC). The management of thyroglossal duct cyst carcinoma remains variable with some advocating for total thyroidectomy in addition to Sistrunk procedure and others recommending Sistrunk procedure alone. We therefore present here a case of PTC within a TDC diagnosed preoperatively which was treated by routine Sistrunk procedure. We also review the current literature regarding etiology, workup, and, recommended extent of surgical resection for this entity.

CASE PRESENTATION

A 42-year-old female without pertinent medical history presented to our facility with a three-month history of a 1-cm nodular mass just superior to the hyoid bone and slightly left of midline. The mass was noted to move upward upon protrusion of the tongue. There was no clinically evident cervical lymphadenopathy. The patient denied prior history of neck irradiation, family history of thyroid malignancy, or compressive symptoms. The patient was euthyroid. Prior to referral to our center, the patient had an extensive radiologic workup that included a neck ultrasound and computed tomography scan with intravenous contrast of the neck. Both studies showed a sub centimeter vascularized lesion within a left strap muscle near the hyoid bone consistent with diagnosis of thyroglosal duct cyst. Ultrasound displayed a normal thyroid gland without nodules and was negative for surrounding lymphadenopathy. Fine needle aspiration of the lesion was obtained revealed a population of atypical follicular cells with enlarged nuclei, pinpoint nucleoli, intra nuclear grooves, and intra nuclear inclusions consistent with papillary thyroid carcinoma. The patient subsequently underwent an uncomplicated Sistrunk procedure with excision of the cyst and a portion of the hyoid bone. Thyroidectomy was not performed. Surgical pathology revealed a margin-negative 5-mm classical papillary thyroid micro carcinoma within a thyroglossal duct cyst without evidence of vascular invasion. The patient continues to do well today and six month repeat neck ultrasound was unremarkable.

DISCUSSION

The cause of TDC carcinoma is unclear though the predominating theories are either metastatic disease from an occult primary or spontaneous development from ectopic thyroid tissue found within the TDC. In their series of nine cases of PTC within TDC, Rossi et al found that 53% had *BRAF* mutation [3].

However, this series also showed both synchronous malignancies in two cases with wild type PTC in the TDC and wild type follicular variant PTC in the normal thyroid tissue. The authors thus conclude that thyroglossal duct cyst PTC could perhaps be due to primary malignancy in the ectopic thyroid tissue, though they appropriately note the small sample size of their series. However, some series present cases of simultaneous PTC within both a TDC and normal thyroid tissue which argues for metastases from an occult primary [4,5]. Thus, the true etiology of PTC within TDC, be it metastatic or coincidental, remains elusive and is largely responsible for the controversy surrounding initial extent of resection.

The workup of any TDC should begin with thyroid function tests and neck ultrasound (US) with or without fine needle aspiration (FNA). The thyroid should also be fully investigated to evaluate for synchronous lesions. The sonographic appearance of TDC typically is described as a well-encapsulated cystic structure with a central solid component [6]. Fine needle aspiration (FNA) is only 53-66% sensitive when diagnosing PTC within TDC as opposed to at least 80% when diagnosing PTC within native thyroid tissue [7]. This is hypothesized to be due to the lesion's small size and dilution of cells by cyst contents which lead to hypo cellular smears. Notably, the diagnostic sensitivity increased to nearly 100% in one series when performing FNA with ultrasound guidance [8]. Some authors, including our own group, advocate obtaining FNA of TDC only in cases with high suspicion of malignancy (large or increasing size, irregular shape, immobility, history of radiation exposure) [9]. Computed tomography (CT) may be useful to evaluate the extent of the TDC and is recommended to further characterize the lesion if suspicious features are detected on US. CT can also help evaluate for further abnormalities such as lymphadenopathy and extent (if any) of mediastinal extension [10].

Surgical resection is typically accepted as the primary treatment for PTC, however the treatment for PTC within TDC is variable and there is little standardization in technique across institutions. Some would advocate that patient risk factors should help guide the choice of procedure. For example, Kennedy and colleagues proposed that low risk patients (15-45 years old without history of radiation exposure, tumor < 4cm in largest diameter, and no evidence of metastases) may be treated with Sistrunk procedure alone. The surgeon can expect 95% cure rate and 95-100% long term survival in these patients [11]. Avoiding total thyroidectomy in low-risk patients prevents risk associated with surgery (e.g. recurrent laryngeal nerve damage, hypoparathyroidism) and life-long hormone replacement therapy.

In contrast, concomitant thyroidectomy should be considered in those patients at higher risk for synchronous malignancies or when there is evidence of significant lymphadenopathy although formal recommendations and data for these patients remains to be elucidated [12,13]. A recent retrospective study by Bakkar et al., revealed that 12/19 (63.2%) of patients with thyroglossal duct cyst carcinoma who underwent Sistrunk procedure with total thyroidectomy had simultaneously malignancy of the thyroid gland on final pathology. Furthermore, it was noted that 3/7 (43%) of this cohort who had clinically significant thyroid cancer had completely unremarkable preoperative thyroid sonographic findings. This suggests that including total thyroidectomy in all cases of thyroglossal duct cyst carcinoma may not be overly radical. Additionally, in high risk patients performing thyroidectomy allows the possibility of radioactive iodine ablation to both eliminate residual disease and readily allows for monitoring of recurrence.

In conclusion, the exact cause of PTC within TDC is unknown and could be metastatic from an occult primary or originate *de novo* within the TDC itself. If suspected beforehand, the surgeon should avoid manual palpation-guided FNA and instead obtain ultrasound-guided FNA. The extent of resection required remains controversial. Some groups continue to advocate for additional total thyroidectomy only in high risk patients, however recent evidence suggests there may be a role for thyroidectomy in all patients found to have thyroglossal duct cyst carcinoma.

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Cite this article

Underwood HJ, Williams DM, Kundel A (2016) Papillary Thyroid Carcinoma within Thyroglossal Duct Cyst: Case Report and Review of Literature. JSM Head Neck Cancer Cases Rev 1(2): 1006.