

## Case Report

# Metastatic Papillary Carcinoma in Thyroglossal Duct Remnant from Thyroid Gland: A Case Report

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### ABSTRACT

Metastatic papillary carcinoma of the thyroglossal duct remnant from thyroid gland is a clinical rarity. It is almost always diagnosed in adult patients. Pre-operative diagnosis is

seldom possible. We report such a case because of its extreme rarity and suggestion of pre-operative diagnosis on fine needle aspiration cytology, CT and thyroid nuclear scans.

KEY WORDS: papillary carcinoma, thyroglossal duct remnant

### INTRODUCTION

Carcinoma arising in a thyroglossal duct (TGD) is extremely rare, with only approximately 200 reported cases and an estimated incidence of less than 1%. While the carcinoma may evolve from ductal epithelium, approximately 90% of cases originate from thyroid remnants<sup>[1]</sup>. The majority is papillary in nature, less than 5% being squamous cell in type<sup>[2]</sup>. Most cases have occurred in adults. There seems to be no pre-disposing factors. Typically, neither clinical history nor physical examination leads to pre-operative diagnosis<sup>[3]</sup>. In this paper, a case of papillary carcinoma of thyroid in a thyroglossal duct remnant, which was diagnosed pre-operatively on fine needle aspiration cytology, is described.

### CASE REPORT

A 29-year-old female patient presented to our surgical clinic for evaluation of a mass in the neck. She had noticed this gradually increasing mass incidentally, two years earlier. She denied any other symptoms. Physical examination revealed a firm round mass 2 cm in diameter anteriorly in the midline, which was vertically mobile on tongue protrusion and deglutition, and without any inflammatory signs. The thyroid gland felt clinically normal and there was no palpable cervical lymphadenopathy. The thyroid gland ultrasonographic evaluation identified a mass 3 cm x 1.5 cm in size, separate from the thyroid gland, adjacent to the isthmus; and thyroid gland also had a solid mass 0.5 cm in diameter in the left lobe. FNAC from the mass suggested papillary carcinoma, most probably in a thyroglossal cyst. Radioactive thyroid scan showed the clinically palpable mass to be extra thyroidal in origin, along

with a cold nodule in left lobe of thyroid.

CT scan also showed a mass in the midline anteriorly in the neck, measuring 3 x 1.6 cm separate from thyroid gland and small nodule in left upper pole of thyroid gland 0.5 cm in diameter. There was no cervical lymphadenopathy (Fig. 1).

Other tests including blood chemistry thyroid function tests and X-ray chest and neck were all normal. Intra-operatively, a mass of about 3 cm in diameter in midline anteriorly, connected to the isthmus of thyroid gland below with a fibrous band and above to the base of tongue by the thyroglossal tract via the hyoid bone, was found. Total excision of thyroglossal duct remnant mass with the hyoid bone and total thyroidectomy were done because the frozen section showed papillary carcinoma in left lobe of the thyroid gland. Her post-operative period was uneventful. Histopathological examination of the resected mass in the midline measuring 3 x 2.5 x 1.5 cm showed the papillary architecture composed of mildly pleomorphic cells with optically clear irregular shape nuclei. Foci of calcification and psammoma bodies were also seen (Fig. 2). The left upper lobe thyroid mass measuring 0.5 x 0.1 cm, showed almost similar architectural feature of papillary carcinoma. The patient has received radioactive iodine and was started on thyroxine replacement therapy. On her recent follow-up at 3 months, she was totally asymptomatic.

### DISCUSSION

Thyroglossal duct remnants are the most common midline neck masses in childhood, representing more than 75% of such masses<sup>[4]</sup>. About 7% of the adult population have these remnants<sup>[5]</sup>. Embryologically, the thyroid descends

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Fig 1: CT scan showing a mass in the anterior neck separate from the thyroid gland.

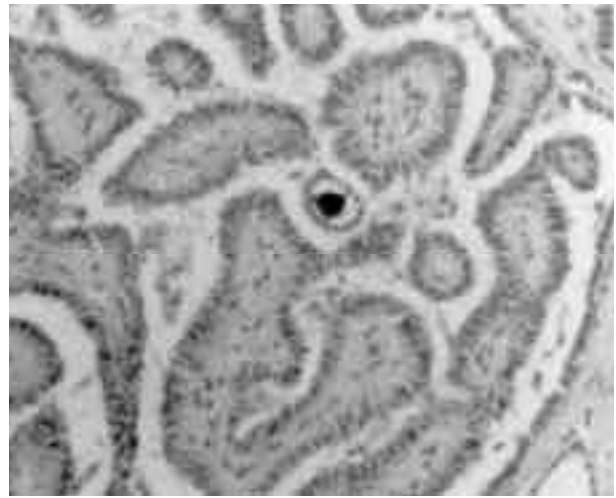


Fig 2: Histopathology of the thyroglossal mass showing papillary carcinoma of thyroid, and psammoma body

from foramen caecum reaching the final position in the seventh to eighth week of intrauterine development. The thyroglossal remnant duct can become, obliterated, a cyst, a duct or ectopic tissue<sup>[6]</sup>.

Brentano in 1911 and Uchermann in 1915 are credited as being among the first to describe a neoplasm in a TGD remnant<sup>[1,7]</sup>. The median age of presentation is around 40 years. Most of the patients are asymptomatic.

There are two theories to explain the thyrogenic origin of thyroglossal duct adenocarcinoma. The first theory, the cyst *de novo* theory stems from the fact of histological identification of ectopic thyroid nests in as many as 62% of the surgical specimen. The absence of medullary carcinoma in thyroglossal duct also supports this theory because it arises from para-follicular cells which do not develop from the thyroid primordium and would not be present in ectopic tissue<sup>[8,9]</sup>. The second is the metastatic theory wherein the tumor is derived from metastases of an occult primary carcinoma of the thyroid gland. This is supported by cases of synchronous papillary carcinoma but can also be due to multifocal tumor<sup>[10]</sup>. Although Crile<sup>[11]</sup> believed that the thyroglossal duct could act as a natural conduit for spread of thyroid carcinoma, the metastatic theory seems less likely<sup>[12]</sup>. In the light of the theories of origin of the carcinoma, arising in the thyroglossal duct remnant, this case could well fit into the metastatic one.

In most of the cases reviewed, the malignancy was not suspected before operation. The carcinoma was diagnosed only at pathological examination. They were indistinguishable clinically<sup>[1]</sup>. However, in our case a pre-operative suspicion of carcinoma of the TGD remnant was suggested on FNAC, and thyroid nuclear scan.

Treatment by the Sistrunk procedure is suggested for non-metastatic disease. Thyroidectomy

is recommended only if there is objective suspicion of a mass or nodule in the gland (e.g., 'cold' nodule on scan, palpable mass, or history of radiation), followed by radioactive iodine treatment. Radical or modified radical neck dissection is indicated only in the presence of positive lymph nodes<sup>[1,10]</sup>. Given the excellent cure rate for papillary thyroid carcinoma, regular physical examination and periodical imaging is advised.

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