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Case Report

Pathology Section

Papillary Carcinoma of Thyroglossal Cyst in a Background of Hashimoto's Thyroiditis: A Case Report

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ABSTRACT

Thyroglossal Duct Cyst (TGDC) is one of the most common congenital neck anomalies that occur due to incomplete involution of the thyroglossal duct. Carcinoma arising in TGDC is rare, accounting for about 1% of cases, with non neoplastic thyroid gland lesions even rarer. This report demonstrates a case of papillary carcinoma with Hashimoto Thyroiditis (HT) of TGDC in the background of HT in the thyroid gland. A 51-year-old woman presented with complaints of midline neck swelling for the past 20 years. Ultrasound revealed an infected thyroglossal cyst. Fine Needle Aspiration Cytology (FNAC) was inconclusive, and the cystic lesion was excised and sent for histopathology. Histopathology showed a thyroglossal cyst with papillary carcinoma along with a focus of HT. The patient underwent total thyroidectomy after a month, which showed features of HT and nodular hyperplasia. There was no evidence of papillary carcinoma in the thyroidectomy specimen. TGDC can harbour malignancies accounting for about 1% of cases. Since TGDC has ectopic thyroid tissue, they should be evaluated for neoplastic and non neoplastic lesions. Also, the evaluation of the thyroid gland is essential for identifying the presence of malignancy to confirm a primary or secondary metastatic carcinoma in a TGDC.

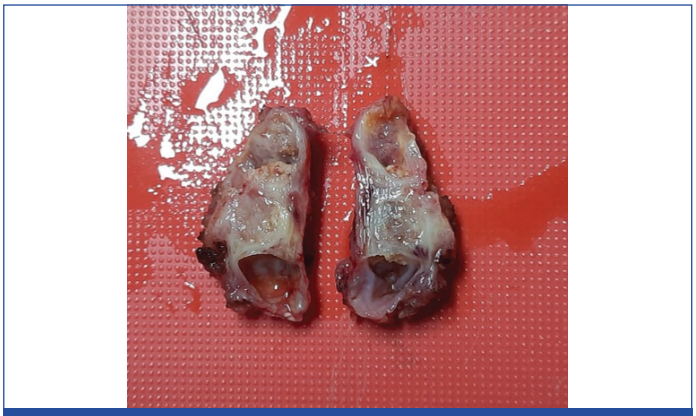
Keywords: Midline neck swelling, Sistrunk procedure, Thyroid, Thyroid carcinoma

CASE REPORT

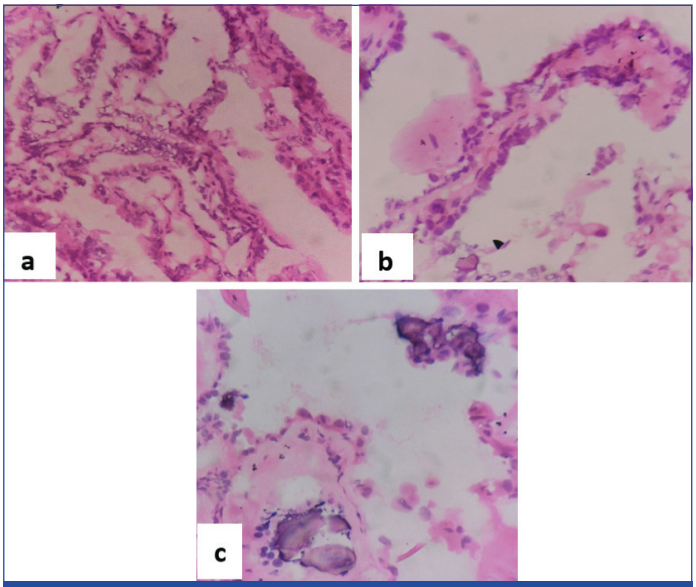
A 51-year-old woman presented with a 20-year history of front-of-neck swelling which was insidious in onset, gradually progressive, and associated with throbbing pain. She was not a known case of hypothyroidism, and there was no history of any drug intake. Physical examination showed a mass of size 4×4 cm along the midline of the neck, which moved on deglutition and also with protrusion of the tongue. Palpation revealed a mobile, non tender cystic mass. It was not fixed to the underlying structures, and there were no palpable neck nodes. Thyroid function tests showed TSH-1.49 IU/mL (0.27-4.2 IU/mL), FT3-2.6 pg/mL (2.0-4.4 pg/mL), and FT4-0.9 ng/dL (0.93-1.7 ng/dL) within normal limits. Ultrasound showed a well-defined anechoic lesion of 2.3×2.3×1.9 cm with a smooth margin, thick septation, solid component, and calcification. The surrounding tissue showed inflammatory changes suggestive of an infected thyroglossal cyst. The right lobe of the thyroid showed a Thyroid Imaging Reporting and Data System (TIRADS) score 1 lesion. The rest of the thyroid gland was normal. Subsequently, FNAC was inconclusive.

The patient underwent a Sistrunk procedure initially for the removal of the infected TGDC, and a frozen section was done. Lobectomy was not done. A cystic lesion measuring 3.2×2.4×1.4 cm was received. The outer surface showed multiple cysts of varying sizes. On the cut surface, two cystic spaces were observed, each measuring 0.7×0.6×0.5 cm, with a cyst wall thickness ranging from 0.2 to 0.3 cm. The upper cystic space showed multiple tiny grey-white projections, with a grey-white solid area noted between the two cystic spaces [Table/Fig-1]. No enlarged lymph nodes were found, so neck dissection was not done.

Routinely processed histopathology sections showed multiple cysts lined by tall columnar cells and confirmed papillary carcinoma with psammoma bodies [Table/Fig-2]. In addition, there was a focus showing features of HT with Hurthle cell change, follicle destruction, and dense infiltration by lymphocytes and plasma cells [Table/Fig-3,4]. The diagnosis was TGDC with papillary carcinoma in the background of HT with Hurthle cell change.

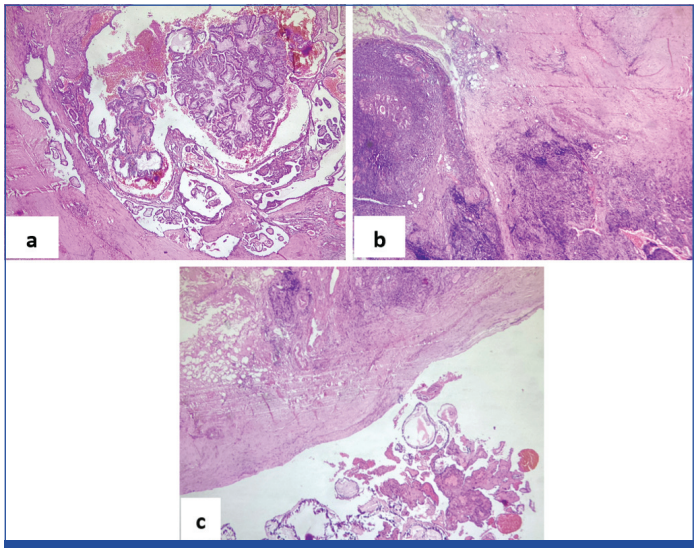


[Table/Fig-1]: Gross image of TGDC with solid and cystic areas with multiple tiny projections.

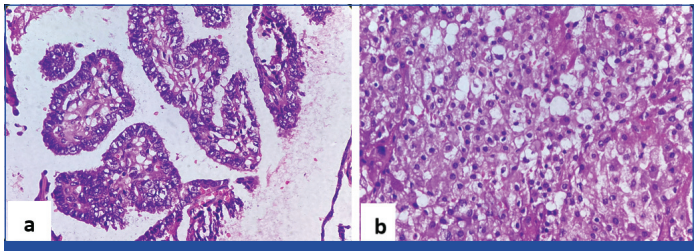


[Table/Fig-2]: Frozen section images of TGDC: a) Papillary architecture (H&E stain, x25); b) True papillae (H&E stain, X400); c) Calcifications- psammoma bodies (H&E stain, X400).

Two months postoperatively, the patient underwent a repeat ultrasound, which revealed a TIRADS 3 lesion in the right lobe of the thyroid with internal vascularity. The rest of the thyroid gland appeared normal. The patient was readmitted for surgery, and a total thyroidectomy specimen was obtained. Grossly, the intact capsule displayed multiple tiny colloid-filled nodules. The histopathology of the thyroidectomy specimen demonstrated numerous colloid-filled follicles with foci of lymphoid follicles with germinal centres. Some follicles showed Hurthle cell change. In addition, the right lobe of the thyroid exhibited nodular hyperplasia, resulting in the diagnosis of HT and nodular hyperplasia of the right thyroid lobe. No malignancy was detected.



[Table/Fig-3]: a) TGDC with papillary thyroid carcinoma (H&E stain, x25); b) TGDC with Hashimoto Thyroiditis (HT) (H&E stain, x25); c) TGDC with papillary carcinoma and foci of Hashimoto Thyroiditis (HT) (H&E stain, x25).



[Table/Fig-4]: a) Nuclear features characteristics of papillary carcinoma-nuclear enlargement, overlapping, optically clear chromatin, irregular nuclear contours (H&E stain, X400); b) Hurthle cell change (H&E stain, X400).

Postoperatively, patient came only for the first review. She was stable without any clinical complaints. Thyroid function test showed elevated TSH of>100 IU/mL and decreased FT3- 0.3 pg/mL and FT4- 0.08 ng/dL. She did not come for subsequent reviews and was lost to follow-up.

DISCUSSION

The TGDC are a common cause of congenital midline neck swelling. The thyroglossal duct typically obliterates by 7 to 10 weeks of embryonic life, but approximately 7% of adults exhibit incomplete involution [1]. The incidence of malignancy in TGDC is very rare, accounting for about 0.7 to 1.5%, of which 75-85% are of the papillary subtype [2]. Papillary carcinoma in a TGDC can originate de novo from remnant thyroid tissue or as a metastasis from the thyroid gland [3]. HT is a common cause of hypothyroidism. An extensive meta-analysis by Hu X et al., showed a global prevalence of HT to be 7.5% among adults. HT is four times more common in women, with a higher prevalence observed among adults in the lower middle-income group [4]. Carcinomas arising from TGDC are rare, and there are very few large series of TGDC carcinoma in the literature [5,6].

This report describes a 51-year-old female with a 20-year history of anterior neck swelling associated with pain and a short duration of fever. The clinical presentation of carcinoma in a TGDC is similar to that of a benign TGDC. Most malignancies in TGDC are diagnosed postoperatively by histopathology [5,6]. Before the surgery, Fine-needle Aspiration (FNA) of the midline neck swelling yielded scanty material and predominantly showed a reactive lymphoid population. FNA of TGDC has a 53% true positive and 47% false negative rate due to factors like cystic nature of the lesion, a small malignant component, and specimen quality leading to low cellular yield [5,6].

In the present case, examination of the thyroid gland revealed only HT with nodular hyperplasia, with no evidence of malignancy. In a study by Rossi ED et al., 45% of TGDC carcinomas had no thyroid malignancy. The remaining 55% of TGDC carcinomas showed synchronous thyroid cancer, with three cases of Papillary Thyroid Carcinoma (PTC) and two cases of follicular variant of PTC [6]. There is a possibility that the majority of TGDCs arise de novo from remnant pre-existing thyroid tissue rather than as metastatic disease from the thyroid gland [5,6]. Regional lymph node metastasis was seen in 13-67% of cases [5]. In the present case, patient did not show any lymph node involvement.

Cytogenetic analysis by Rossi ED et al., found the V600E BRAF mutation in all four cases of primary TGDC papillary carcinoma, with the wild type present in three cases. V600E BRAF mutations were also present in five cases of TGDC papillary carcinomas and the same mutation in four synchronous thyroid carcinomas. V600E BRAF mutations are associated with extra thyroidal extension, advanced stage at presentation, lymph node metastasis, large tumour size, and multifocality [6]. There are very few case reports describing the presence of co-existing HT with primary papillary carcinoma of TGDC [7,8]. In the present case, in addition, there was also HT in the thyroid gland. The comparative review of literature highlighting a few cases with almost similar histology is presented in [Table/Fig-5] [9-11]. Themeli Y et al., described a case of a 31-year-old female with PTC in both the thyroid gland and the thyroid ectopic tissue within the TGDC. There was the presence of HT in both TGDC as well in the thyroid gland. The patient underwent thyroidectomy and radioactive ablation [9]. Prasad ML et al., described a case of papillary carcinoma of the thyroglossal duct remnant with HT in both the non neoplastic ectopic thyroid tissue and the thyroid gland. Age and clinical details were not available [10]. The above two case reports favour the theory of metastatic carcinoma to the thyroglossal duct. A report by Cizmic´ M et al., described a similar case of papillary carcinoma in the TGDC with HT in the thyroid gland. However, there was no evidence of coexisting HT in the TGDC [11]. Various literature data suggest about a 20%-37.5% correlation between PCT and HT in the thyroid gland, but similar data for TGDC is missing. The association of HT is 1.99 times higher among those with PTC than in patients with other pathological types of thyroid cancer. This is due to RET/PTC translocation which is a RET rearrangement found in the large majority of tissues with HT and without detectable PTC, which may show a progression to cancer from chronic thyroiditis.

Author, ref no., year of publication	Age (years) /Gender	Duration of chief complaint	PTC in TGDC	HT in TGDC	HT in thyroid	PTC in thyroid
Themeli Y et al., [9]-2022	31/F	2 months	Yes	Yes	Yes	Yes
Cizmic´ M et al., [11]-2007	53/F	N/A	Yes	No	Yes	N/A
Prasad ML et al., [10]-1990	N/A	N/A	Yes	Yes	Yes	Yes
Present case, 2024	51/F	20 years	Yes	Yes	Yes	No

[Table/Fig-5]: Comparison of previous case reports with the present case.
PTC: Papillary thyroid carcinoma; TGDC: Thyroglossal duct cyst; HT: Hashimoto thyroiditis; N/A: Not available

P63 expression is detected in 81% of HT and PCT, which is not found in normal thyroid or Graves. This unique expression of P63 suggests a potential role in the pathogenesis of PCT and HT [7,8].

CONCLUSION(S)

There are very few case reports in the literature regarding the co-existence of HT and papillary carcinoma in TGDC. Sudden or prolonged swelling of a TGDC in adults requires immediate evaluation for the possibility of malignancy. Investigations like FNAC and imaging studies are important in the follow-up. The Sistrunk procedure should be done, and excised tissue should be carefully evaluated for malignancy. Thyroid gland evaluation is also necessary, as there is no clear evidence whether malignancy in the TGDC arises de novo or from metastasis of an occult primary. Proper follow-up is also needed for the treatment of relapses or metastasis.

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