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Papillary Thyroid Carcinoma in Thyroglossal Duct Cyst: A Case Report and Review of Management Strategies

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Abstract	Case Report

Thyroglossal duct cysts (TGDCs) are common congenital neck anomalies, with approximately 1% undergoing malignant transformation, most frequently into papillary thyroid carcinoma (PTC). We report a case of a 50-year-old woman with a four-year history of a painless, progressively enlarging midline cervical mass. Imaging revealed a mixed solid-cystic lesion with calcifications and suspicious lymphadenopathy. A Sistrunk procedure and total thyroidectomy confirmed PTC within the TGDC and thyroid gland. Postoperative radioactive iodine therapy was administered, and the patient remains recurrence-free. TGDCa management remains controversial, with the Sistrunk procedure as the cornerstone of treatment. Total thyroidectomy and radioiodine therapy are recommended in high-risk cases or synchronous thyroid involvement. Early diagnosis and tailored management are essential for optimal outcomes. This case highlights the importance of considering TGDCa in the differential diagnosis of cervical masses.

Keywords: Thyroglossal Duct Cyst, Papillary Thyroid Carcinoma, Sistrunk Procedure, Total Thyroidectomy, Radioiodine Therapy.

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INTRODUCTION

Thyroglossal duct cysts (TGDC) are among the most frequent congenital anomalies of the neck, representing the majority of midline cervical masses, particularly in children, with a prevalence of up to 75% [1]. Although typically benign, approximately 1% of TGDC cases transform into thyroglossal duct cyst carcinoma (TGDCa), most commonly papillary thyroid carcinoma [2]. This transformation, while rare, has been documented in nearly 300 cases since its first description by Brentano in 1911[1-3].

TGDCa often mimics benign lesions, with diagnosis typically confirmed postoperatively, requiring suspicion for irregular or firm masses with lymphadenopathy [4].

While the standard treatment involves the Sistrunk procedure, the role the role of total thyroidectomy, radioiodine therapy and suppressive thyroxine therapy, remains controversial due to limited consensus and a lack of large-scale studies [5]. This report presents a rare case of papillary carcinoma in a TGDC, focusing on its presentation, management, and related therapeutic debates.

CASE REPORT

We report the case of a 50-year-old woman with no significant medical history, presenting with a painless anterior mid-cervical mass that had progressively enlarged over four years. She reported no associated symptoms of compression.

On physical examination, a firm, well-defined, midline cervical mass was observed, measuring 4 cm in its longest axis. The mass was mobile with deglutition and tongue protrusion, and no palpable thyroid gland or cervical lymphadenopathy was detected. Thyroid function tests were normal.

Cervical ultrasound revealed a mixed solidcystic mass in the midline, measuring $20 \times 32 \times 36$ mm. The mass displayed isoechoic solid components with calcifications and signs of degeneration. Additionally, bilateral level IA cervical lymph nodes measuring 6×7 mm were identified, along with thyroid nodules classified as EU-TIRADS III. A cervical computed tomography CT scan confirmed a midline cystic lesion measuring $20 \times 35 \times 38$ mm, with an enhancing centrally solid component and focal calcifications, as well as suspicious cervical lymphadenopathy (Firgure 1).

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Figure 1: CT scan of the neck: Midline cystic lesion with an enhancing solid component and focal calcifications

Surgical management began with Sistrunk's procedure, including removal of the cyst and hyoidectomy. Pathological examination revealed a moderately differentiated papillary carcinoma within the TGDC. One week later, a total thyroidectomy was performed, and histopathology confirmed a papillary thyroid carcinoma.

The patient had an uneventful postoperative course and continued treatment with radioactive iodine. She remains under regular follow-up without recurrence or new developments in her condition.

DISCUSSION

Thyroglossal duct cysts (TGDCs) are congenital anomalies resulting from the incomplete obliteration of the thyroglossal duct during embryogenesis, typically presenting as midline cervical masses, accounting for nearly 7% of such masses in adults and representing the most common congenital neck anomalies [1-6].

Although TGDCs are generally benign, approximately 1% undergo malignant transformation, with papillary thyroid carcinoma (PTC) being the most common histological type (80%). Other reported malignancies include mixed papillary/follicular carcinoma (8%), squamous cell carcinoma (6%), and rare types such as Hurthle cell, follicular, and anaplastic carcinomas (6%) [1-7]. In our case, the patient presented with PTC.

TGDC carcinoma (TGDCa) predominantly occurs in the fourth decade of life, with a higher prevalence in women [8]. The etiology of TGDCa remains debated, with theories suggesting either de novo development from ectopic thyroid tissue within the cyst wall or metastasis from an occult thyroid tumor [9].

Clinically, thyroglossal duct cyst carcinoma (TGDCa) is indistinguishable from benign TGDCs, typically manifesting as an asymptomatic midline cervical mass. However, rapid enlargement, firm or fixed consistency, and irregular margins are indicative of malignant transformation [10]. In our case, the mass exhibitedslow progression over four years.

Imaging modalities such as ultrasonography, CT, and MRI can reveal suspicious features like mural nodules, microcalcifications, or solid components, but definitive diagnosis often requires histopathological examination post-surgery [11]. Fine-needle aspiration cytology (FNAC) has limited diagnostic accuracy (53%) but can guide surgical planning if malignancy is suspected [12].

The Sistrunk procedure, which involves excision of the cyst, the hyoid bone, and a core of surrounding tissue, is the gold standard for treatment, significantly reducing recurrence rates to 3-10% [8]. Controversy exists regarding the need for total thyroidectomy (TT), with some advocating for TT in high-risk patients (e.g., those with thyroid gland abnormalities, lymph node metastasis, or tumors >4 cm) to enable radioactive iodine (RAI) therapy and thyroglobulin monitoring [13-14]. However, TT is not routinely recommended for low-risk patients, as it increases morbidity without proven survival benefits [8-15]. Lymph node dissection is reserved for cases with confirmed metastasis, while postoperative RAI and thyroid hormone suppression are recommended for highrisk cases [1-14]. Overall, TGDCa has an excellent prognosis, with a 95-100% survival rate when adequately treated, emphasizing the importance of individualized management and multidisciplinary collaboration [8-15].

CONCLUSION

Thyroglossal duct cyst carcinoma (TGDCa) is a rare malignancy, with papillary thyroid carcinoma (PTC) being the most frequent histological type. While it mimics benign TGDCs clinically, features like rapid growth or irregular margins suggest malignancy. The Sistrunk procedure is the cornerstone of treatment, with excellent prognosis. In high-risk cases or synchronous thyroid involvement, total thyroidectomy and radioiodine therapy may be necessary. Early diagnosis and tailored management are essential for optimal outcomes.

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