**SAR Journal of Surgery**

Abbreviated Key Title: *SAR J Surg*
Home page: https://sarpublication.com/journal/sarjs/home
DOI: 10.36346/sarjs.2023.v04i03.004

ISSN 2707-7748 (P)
ISSN 2709-6912 (O)

**Case Report**

# Thyroglossal Duct Carcinoma: A Case Report and Literature Review

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**Article History:** | Received: 09.07.2023 | Accepted: 11.08.2023 | Published: 15.08.2023 |

**Abstract:** Thyroglossal duct cancer is a relatively rare finding, presenting clinical features that resemble those of a benign cyst, a more common congenital abnormality that may manifest as a lump in the midline or neck area. Thyroglossal duct cysts are a commonly found aberration during the development of the thyroid gland. The prevalence of adenotonsillar hypertrophy is greater than that of branchial cleft abnormalities, and among the pediatric population, it is the second most common cause of neck masses, surpassed only by enlarged cervical lymph nodes. In specific instances, thyroglossal duct cysts have been seen to exhibit a low prevalence of cancer. The diagnosis of thyroglossal duct cancer is commonly made by histological examination of the tumor. In addition, needle aspiration cytology, ultrasonography, and computed tomography are employed as diagnostic tools to differentiate malignant patients from alternative potential diagnosis.

**Keywords:** Head and neck tumors, Thyroglossal duct, Papillary carcinoma, Thyroid cancer, Neck surgery.



## INTRODUCTION

A thyroglossal duct cyst is a congenital anomaly affecting the thyroid gland, commonly presenting as a palpable mass along the thyroglossal tract. The thyroglossal tract represents a vestigial structure derived from the embryonic development of the thyroid gland. During its descent, the thyroid gland establishes a connection with the base of the tongue via a duct, which subsequently undergoes involution and ceases to exist. Occasionally, it may endure and experience dilatation, a process that contributes to the development of thyroglossal duct cyst formation. The occurrence of ectopic thyroid tissue within a thyroglossal duct cyst exhibits a range of prevalence, spanning from 1.5% to 45% of reported cases. This phenomenon can be elucidated by considering the embryological development of the thyroid gland. In general, ductal cysts are typically of a benign nature; however, it is important to note that approximately 1% of cases may exhibit malignant characteristics. The majority of cases pertaining to TDC carcinoma are typically identified during the third and fourth decades of an individual's life, with occurrences in children under the age of 14 being exceedingly rare. Papillary carcinomas, which are frequently observed, exhibit a favorable prognosis due to the infrequent occurrence of cervical and distant metastases. The subsequent course of action for patients who are fortuitously diagnosed with carcinoma following surgery is contingent upon the extent of tumor metastasis. Limited tumors are effectively managed through regular monitoring, whereas the more aggressive tumors necessitate a comprehensive approach involving total thyroidectomy and radioiodine ablation for optimal management.

The predominant form of primary thyroglossal duct cyst carcinoma is papillary carcinoma, accounting for approximately 75-80% of cases. However, it is important to note that other types of tumors can also arise as primary tumors in this context. These include mixed papillary-follicular carcinomas (7%), squamous cell carcinoma (5%), follicular carcinoma (1.7%), Hürthle cell carcinoma, and anaplastic carcinoma (0.9%). Similar to thyroglossal duct cysts, thyroglossal duct cyst carcinomas frequently manifest as an indolent,

symptomless mass situated along the midline of the neck. However, there have been documented instances of these neoplasms originating in a lateral position. Dysphagia, dysphonia, weight loss, and rapid growth in size are indicative of potential malignancy. The prevailing surgical technique employed for the treatment of a thyroglossal duct cyst is known as Sistrunk's procedure. This procedure involves the meticulous removal of the thyroglossal duct cyst, the central segment of the hyoid bone's body, and a surrounding core of tissue encompassing the thyroglossal tract. The objective is to establish an opening into the oral cavity at the foramen cecum. Sistrunk's procedure, a well-established surgical intervention, is typically conducted as an independent procedure in patients with low risk. However, it is important to note that patients with high risk may require an additional total thyroidectomy and subsequent postoperative radioactive iodine ablation therapy.

## CASE REPORT

In this case report, we report a 27-year-old female patient who sought medical attention at our Endocrine surgery department. The patient reported a painless, midline swelling located in the upper neck that had been present for the last 24 months. The swelling had a slow and insidious beginning, progressively increasing in size over time. There was an absence of any documented instances of dysphagia, dyspnea, or hoarseness in the patient's medical history. There was no historical evidence indicating the presence of hypothyroidism or hyperthyroidism. The patient did not provide any information on previous exposure to radiation. There was no other noteworthy historical or personal background. Upon careful inspection, a solitary swelling was seen in the suprahyoid area, precisely in the midline. The swelling was around 4 · 3 cm in size, with an oval form. Furthermore, it had clear boundaries and a soft substance. The superficial integument had a typical visual aspect and was readily susceptible to manual compression. The object exhibited movement throughout the processes of swallowing and tongue extension. Upon palpation, the observed enlargement exhibited characteristics of being non-tender and having a soft substance.

The external integument had a typical and unremarkable visual presentation, with a pliable texture that allowed for effortless manipulation by pinching. The thyroid gland had typical clinical characteristics in terms of its form and size. The remainder of the systemic evaluation yielded no abnormal findings. A preliminary diagnosis of thyroglossal cysts was established. The results of the blood tests, including routine examinations and assessments of thyroid function (namely T3, T4, and TSH levels), were within normal ranges. The neck of the United States government (USG) was exposed. The lower pole of the left lobe exhibits a solid lesion with partially defined edges, hypoechoic homogeneous echogenicity, and no color Doppler flow measuring 3.3x2.9mm. The left thyroid lobe measures 25.9x14.5x11.3mm, while the right lobule measures 34.9x17.2x15mm. The isthmus measures 2.5mm. Additionally, there is the presence of adenopathy hypoechoic echogenics with central microcalcifications, no flow to Doppler color measuring 4mm, and bilateral lymph nodes with fatty thread retention of less than 10mm. Sistrunk procedure is carried out, the presence of thyroglose cysts of approximately 5x4 cm, firmly attached to the body of hioids and prethyroid muscles, hioides are partially resecated, the final pathology is documented papillary thyroid carcinoma incidental.

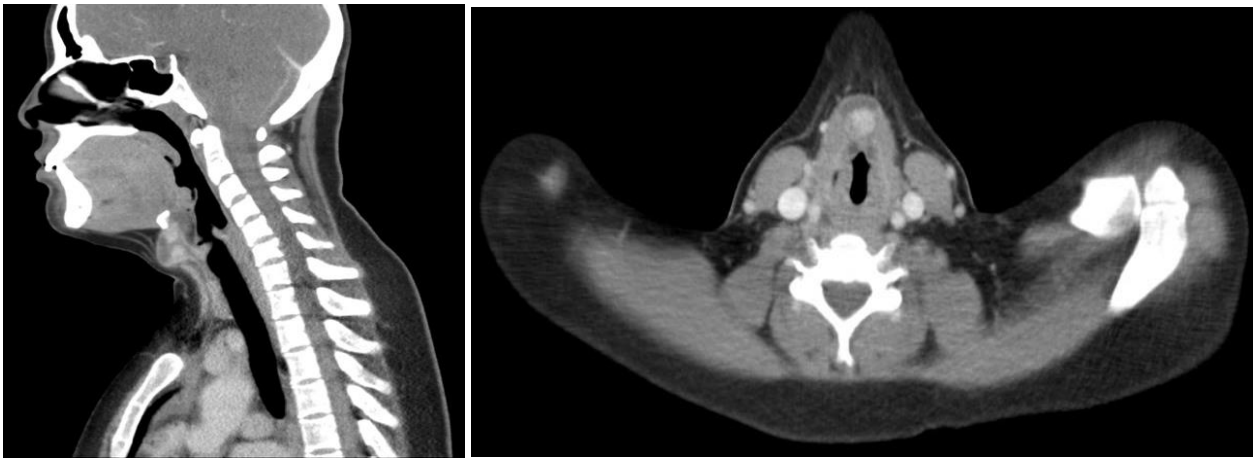

**Figure 1: Tomography / Anterior neck mass**

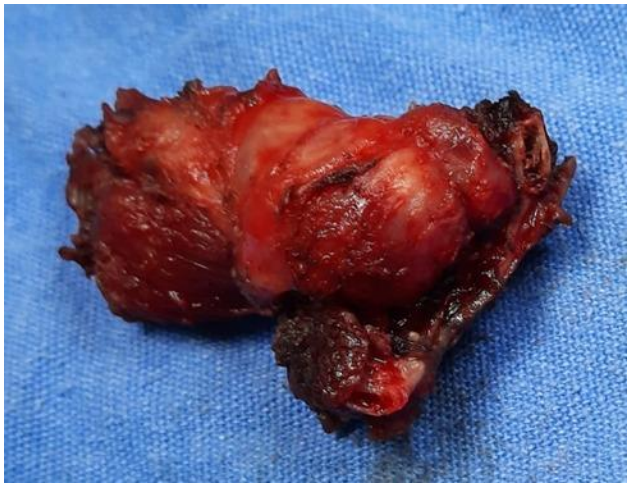

**Figure 2: A surgical piece**

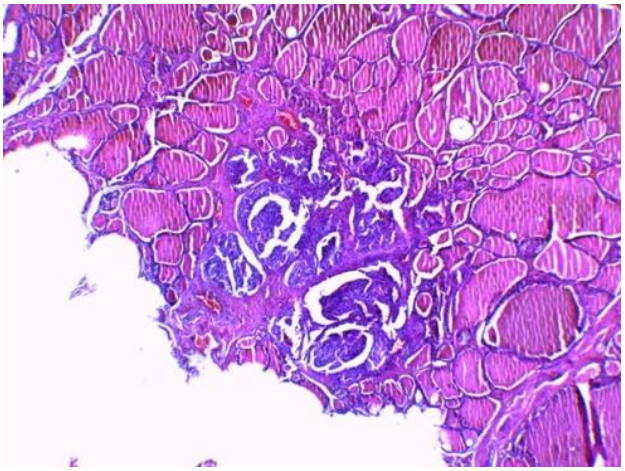

**Figure 3: Histological cut of thyroglose cyst with microcalcifications corresponding to bodies of psammoma, characteristic of papillary thyroid cancer**

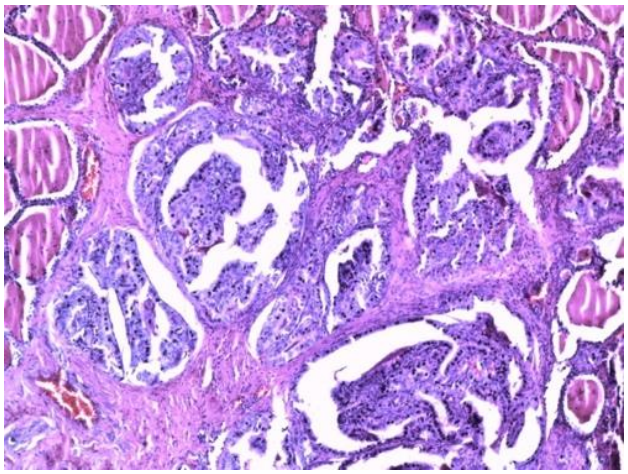

**Figure 4: Thyroid tissue is normal, with the follicles and the colloid, surrounding the island of thyroid papillary cancer with significant basophilia**

## DISCUSSION

The thyroid gland, an essential endocrine gland, undergoes its initial development during the third week of gestation. From an embryological perspective, it is important to note that the thyroid gland originates from the floor of the pharyngeal gut, specifically the foramen caecum. This gland takes shape as a bilobed diverticulum and maintains its connection to the gut through a slender canal known as the thyroglossal duct. Typically, the thyroglossal duct undergoes obliteration and subsequent disappearance during the tenth week of gestation. A thyroglossal remnant may persist in various

manifestations, such as a cyst, a tract, or a duct, or even as an ectopic thyroid located within a cyst or duct. Thyroglossal duct cysts represent the prevailing anomalies encountered within the cervical region. Over 75% of thyroglossal duct cyst carcinomas (TGDCCa) are fortuitously detected during the conclusive histopathological examination following the extraction of an otherwise unremarkable and seemingly harmless thyroglossal duct cyst (TGDC). Specifically, the presence of a midline, enlarging, firm, and solid mass with irregular edges in the central area of the neck in an adult individual (occasionally observed in children, but less frequently), which does not move smoothly along the thyroid cartilage or hyoid bone, and does not exhibit synchronous movement with tongue protrusion, occasionally accompanied by discomfort and difficulty swallowing, can aid in distinguishing a thyroid gland-derived carcinoma (TGDCCa) from a typical thyroid gland-derived cyst (TGDC) or other non-cancerous conditions. The presence of concurrent lymph node swelling may contribute to heightened clinical suspicion.

Irrespective of the clinical manifestation, it is imperative to conduct a thorough examination using neck ultrasonography to assess the structure of the nodule. This evaluation includes identifying any concerning characteristics such as a prominent solid portion, microcalcifications, uneven wall thickening, and excessive blood flow. Additionally, the examination aims to determine the condition and attributes of the thyroid gland, ruling out any abnormal locations or the presence of metastases in the lymph nodes. Based on the aforementioned embryological factors, it is imperative to note that the sis-trunk procedure represents the minimal surgical intervention necessary for the proper management of a TGDCCa. A thorough assessment of a thyroglossal duct cyst for primary carcinoma necessitates distinguishing between carcinomas originating from a well-defined thyroglossal duct cyst or tract and carcinomas arising from the thyroid glands. Papillary carcinoma within the thyroglossal duct cyst (TDC) is typically managed through the utilization of the Sistrunk procedure, which has demonstrated a commendable control rate of 95% according to available reports. The Sistrunk procedure entails the comprehensive excision of the cyst, a segment of the hyoid bone, and any remaining thyroglossal duct remnants extending to the base of the tongue. The performance of a thyroidectomy procedure is not recommended unless there are palpable abnormalities detected in the gland or if there is a predicted lack of sufficient follow-up. If thyroidectomy is performed as part of a regular treatment regimen, it appears that it does not have a substantial effect on the overall treatment outcome.

This particular carcinoma typically demonstrates a highly favorable prognosis. Regarding adjuvant therapy, there is currently a lack of data supporting its efficacy in patients diagnosed with papillary carcinoma with thyroid disease complications (TDC). However, it is recommended to administer suppression hormone therapy in patients diagnosed with low-risk diseases. However, in cases where there is a suspicion of abnormalities in the thyroid gland, such as the presence of a large tumor measuring more than 1.5 cm, nodal disease, being over the age of 45, or a history of neck irradiation, it is recommended to undergo a total thyroidectomy procedure. Following this, I131 ablation is performed, and it is advised to suppress thyroid-stimulating hormone levels. In this particular instance, the thyroid gland was subjected to intraoperative examination, followed by a comprehensive postoperative imagological evaluation. The findings from these assessments revealed no discernible abnormalities in either the thyroid gland or the adjacent neck lymph nodes. Therefore, no additional surgical interventions were suggested, and the patient has not exhibited any signs of recurrence throughout the duration of the follow-up period.

## CONCLUSION

Thyroglossal duct carcinoma (TDCa) is a relatively rare condition, accounting for roughly 2.9% of all lesions associated with the thyroglossal duct. The process of diagnosing a condition by clinical examination can be challenging; however, the utilization of small needle aspiration biopsy and computed tomography (CT) can provide valuable assistance in reaching a diagnosis. The management of these cases remains a subject of controversy due to the scarcity of recorded instances. Consequently, a multidisciplinary approach and personalized management for each case are crucial in addressing these uncommon occurrences. The prognosis for individuals diagnosed with papillary carcinoma in relation to thyroglossal duct cysts is highly favorable.

**Conflicts of interests**

The authors have shown no conflicts of interest, ensuring the integrity and objectivity of their study results.

**Acknowledgements**

Our most sincere gratitude goes out to the well-respected team for the extraordinary efforts they made toward the successful completion of the remarkable case study that had special significance.

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