RESEARCH PAPER

Clinical Presentation, Surgical Management, and Histopathological Evaluation of Thyroglossal Duct Cyst Carcinoma: A Case Report

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ABSTRACT

Background: Thyroglossal duct cyst (TGDC) is the most common congenital anomaly of the neck and accounts for the majority of midline cervical swellings. It arises from the persistence of the thyroglossal duct during embryological descent of the thyroid gland. Although commonly diagnosed in children, it may also present in adults. Malignant transformation within TGDC is rare, occurring in approximately 1% of cases, with papillary carcinoma being the predominant histological subtype. The Sistrunk procedure remains the standard surgical treatment.

Case Presentation: A 52-year-old male presented with a gradually enlarging, painless midline neck swelling of 10 days duration. Clinical examination revealed a well-defined cystic swelling measuring approximately 2 × 2 cm at the level of the hyoid bone, which moved with swallowing and protrusion of the tongue. Routine laboratory investigations were within normal limits. Ultrasonography demonstrated a well-defined cystic lesion measuring approximately 10 mm in the midline near the hyoid bone, with a normally located thyroid gland. The patient underwent surgical excision via the Sistrunk procedure. Histopathological examination revealed papillary carcinoma (conventional type) arising within the thyroglossal duct cyst, characterized by papillary architecture, fibrovascular cores, nuclear clearing, grooving, overlapping, and psammoma body formation, with a tumor size of approximately 2 cm. In view of the malignant diagnosis, the patient subsequently underwent total thyroidectomy, which demonstrated multinodular goitre without evidence of primary thyroid malignancy, confirming primary carcinoma arising within the thyroglossal duct cyst. The postoperative course was uneventful, and the patient recovered well.

Conclusion: This case highlights a rare occurrence of papillary carcinoma arising in a thyroglossal duct cyst despite classical benign clinical and radiological features. Routine histopathological examination is essential in all cases of TGDC. While the Sistrunk procedure remains the gold standard treatment, identification of malignancy necessitates further evaluation and individualized management. Long-term follow-up is crucial to monitor for recurrence and associated thyroid pathology.

Keywords: Thyroglossal duct cyst, Papillary carcinoma, Sistrunk procedure, Midline neck swelling, Thyroid carcinoma, Congenital neck mass

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Introduction

Thyroglossal duct cyst (TGDC) is the most common congenital anomaly of the neck and represents a major proportion of midline cervical swellings encountered in clinical and surgical practice. It develops due to the persistence of epithelial remnants of the embryonic

thyroglossal duct, which forms during the descent of the thyroid gland from the foramen cecum at the base of the tongue to its definitive pre-tracheal location in the anterior neck during early fetal development. Normally, this duct involutes by the tenth week of gestation; however, incomplete obliteration may lead to cystic dilatation along the pathway of thyroid descent. These cysts may occur anywhere along this tract, although the majority are located in close proximity to the hyoid bone in the midline of the neck [1].

Clinically, thyroglossal duct cysts usually present as a painless, gradually enlarging midline neck swelling. The swelling is typically mobile and characteristically moves upward during swallowing and protrusion of the tongue because of its attachment to the tract connected to the tongue base. Although the lesion is frequently diagnosed in children and adolescents, adult presentation is also well documented, particularly when the cyst becomes infected or progressively enlarges. Recognition of this classical clinical feature is important because it helps differentiate TGDC from other causes of midline neck swelling such as dermoid cysts, lymphadenopathy, thyroid nodules, or enlarged lymph nodes [2].

Epidemiological studies indicate that remnants of the thyroglossal duct are present in approximately 7% of the general population, making it a relatively common developmental anomaly. Among congenital neck masses, TGDC accounts for nearly 70–75% of midline cervical lesions and represents the most frequently encountered congenital cyst of the neck. These cysts may remain asymptomatic for several years and often become clinically evident following secondary infection, inflammation, or rapid enlargement. Recurrent infection is one of the most common complications associated with thyroglossal duct cysts and is frequently responsible for patients seeking medical attention [3].

Although TGDC is typically benign, rare cases of malignant transformation have been reported. The incidence of carcinoma arising in a thyroglossal duct cyst is estimated to be approximately 1%, with papillary thyroid carcinoma being the most commonly reported histological type. In many patients, the diagnosis of malignancy is made incidentally following histopathological examination of the excised cyst. Therefore, careful surgical excision and pathological evaluation are essential for confirming the diagnosis and excluding associated malignancy [3].

The diagnosis of TGDC is primarily clinical but is supported by imaging studies. Ultrasonography is commonly used as the first-line imaging modality because it helps determine the cystic nature of the lesion, evaluate its anatomical relationship with surrounding structures, and confirm the presence of a normally located thyroid gland. Proper evaluation is important before surgical intervention to avoid complications and to ensure accurate diagnosis [4].

Surgical removal remains the definitive treatment for thyroglossal duct cyst. The Sistrunk procedure, which involves excision of the cyst along with the central portion of the hyoid bone and the tract extending toward the foramen cecum, is considered the gold standard treatment. This technique significantly reduces recurrence rates compared with simple cyst excision and has become the standard surgical approach worldwide. Because TGDC represents the most common congenital midline neck lesion and may occasionally be associated with complications or rare malignancy, detailed clinical evaluation and documentation of cases remain important for improving understanding of its presentation and management.

Case presentation:

A 52-year-old male, presented to the surgical outpatient department with complaints of a swelling in the anterior midline of the neck for approximately 10 days. The swelling was initially small and gradually increased in size. It was painless and not associated with dysphagia, odynophagia, dyspnea, voice change, fever, weight loss, or history of trauma. There was no past history of thyroid disease, tuberculosis, diabetes mellitus, hypertension, or prior neck surgery.

On general physical examination, the patient was conscious, alert, and oriented. He was moderately built and nourished, and his vital parameters were within normal limits (pulse rate: 78/min, blood pressure: 120/80 mmHg, respiratory rate: 16/min, temperature: 98.4°F, oxygen saturation: 99% on room air). There was no pallor, icterus, cyanosis, clubbing, generalized lymphadenopathy, or pedal edema.

Local examination of the neck revealed a solitary, well-defined swelling in the midline of the anterior neck at the level of the hyoid bone, measuring approximately 2 × 2 cm. The overlying skin was normal, with no signs of inflammation, ulceration, or sinus formation. On palpation, the swelling was soft to cystic, non-tender, and freely mobile with well-defined margins. A characteristic clinical feature was observed, with the swelling moving upward on both swallowing and

protrusion of the tongue. No cervical lymphadenopathy was noted.

Systemic examination of the cardiovascular, respiratory, abdominal, and central nervous systems was unremarkable.

Routine laboratory investigations were within normal limits, including hemoglobin (14.5 g/dL), total leukocyte count (6,290 cells/mm³), platelet count (1.96 × 10^5/mm³), random blood sugar (96 mg/dL), and HbA1c (5.31%). Renal and liver function tests, serum electrolytes, and coagulation profile were also within normal ranges.

Ultrasonography of the neck demonstrated a well-defined cystic lesion measuring approximately 10 mm in diameter in the midline at the level of the hyoid bone, suggestive of a thyroglossal duct cyst. The thyroid gland was normally located, with normal size and echotexture, and no focal nodules were identified.

Based on the clinical and radiological findings, a provisional diagnosis of thyroglossal duct cyst was made. The patient underwent surgical excision using the Sistrunk procedure under general anesthesia. Intraoperatively, a cystic lesion with a fibrous tract extending toward the hyoid bone was identified. Complete excision of the cyst along with the tract and the central portion of the hyoid bone was performed, and the wound was closed in layers after achieving adequate hemostasis.

Histopathological examination of the excised specimen revealed a cyst wall with chronic inflammatory infiltrate, areas of hemorrhage, and cholesterol cleft formation. The lumen showed papillary structures with fibrovascular cores lined by cells exhibiting optically clear nuclei, nuclear overlapping, and nuclear grooving. Psammoma bodies and focal calcifications were also present. The tumor measured approximately 2 cm in greatest dimension. These features were diagnostic of **papillary carcinoma (conventional type) arising in a thyroglossal duct cyst**.

In view of the malignant diagnosis, the patient subsequently underwent total thyroidectomy for further evaluation and definitive management. Histopathological examination of the thyroidectomy specimen revealed features consistent with multinodular goitre, with no evidence of primary malignancy in the thyroid gland, confirming the diagnosis of primary thyroglossal duct cyst carcinoma. The postoperative period was uneventful. The patient was managed with antibiotics and analgesics, and oral intake was gradually resumed. He was discharged in stable condition. On follow-up, the surgical wound had healed well, with no clinical evidence of recurrence.

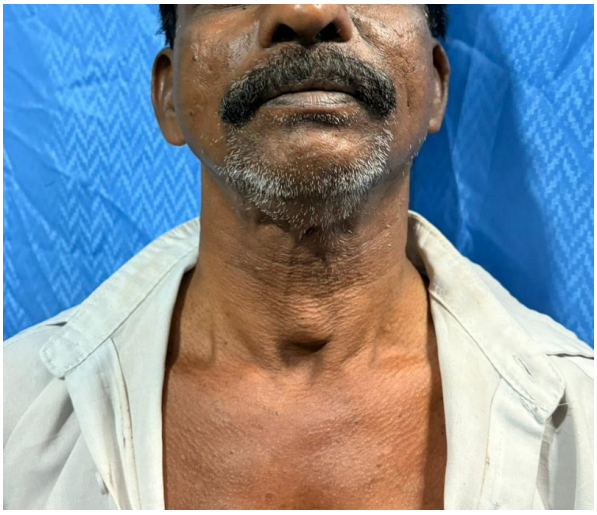

Figure 1:
Clinical photograph showing a midline neck swelling located at the level of the hyoid bone consistent with a thyroglossal duct cyst.

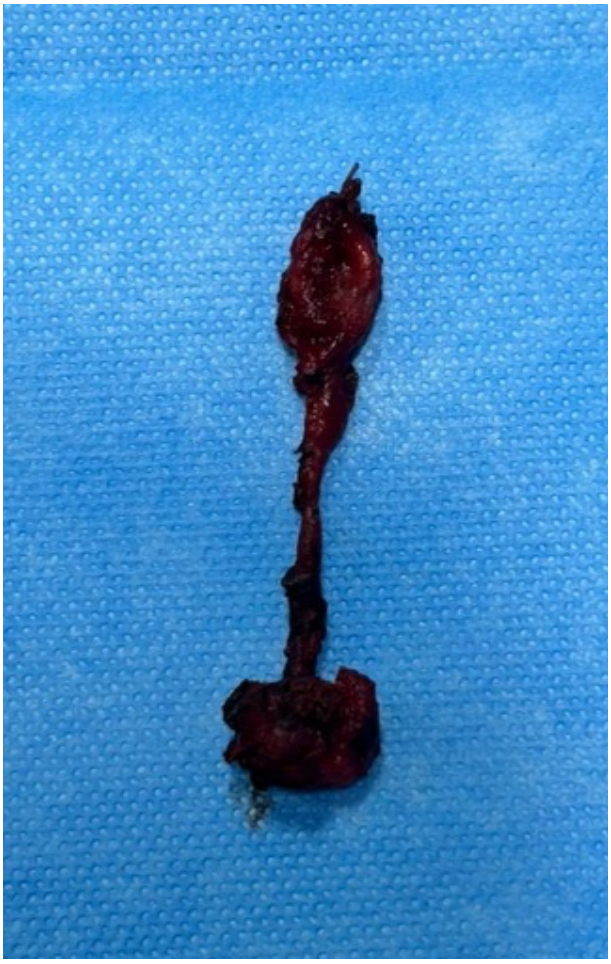

Figure 2:
Intraoperative specimen demonstrating the excised thyroglossal duct cyst along with the tract and central

portion of the hyoid bone following Sistrunk procedure.

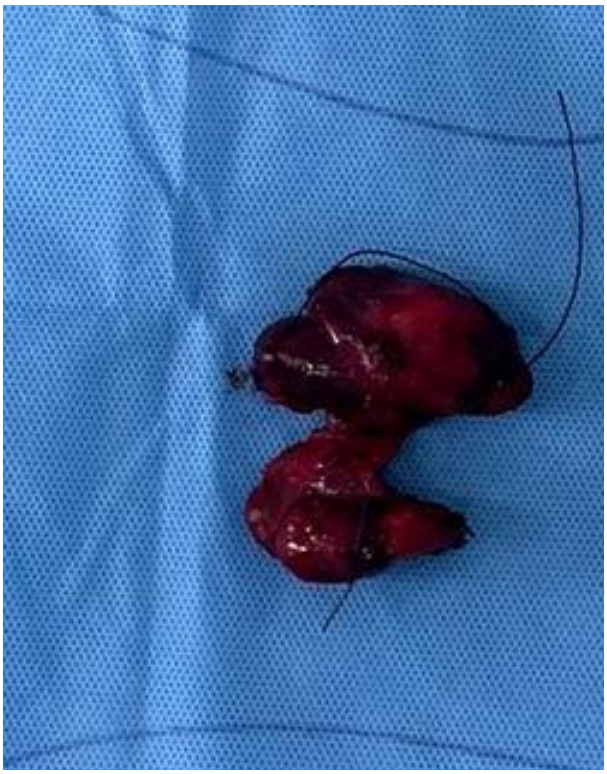

Figure 3:
Excised surgical specimen showing the cystic mass attached to the thyroglossal duct tract after complete removal during surgery.

Discussion

Thyroglossal duct cyst (TGDC) represents the most common congenital anomaly of the neck and accounts for the majority of midline cervical swellings encountered in clinical practice. Although it is predominantly diagnosed in pediatric populations, adult presentations are well recognized, often associated with secondary infection or progressive enlargement. In the present case, a 52-year-old male presented with a gradually enlarging midline neck swelling exhibiting classical clinical features, including movement with swallowing and protrusion of the tongue. Ultrasonography demonstrated a cystic lesion at the level of the hyoid bone with a normally located thyroid gland, and the patient underwent surgical excision via the Sistrunk procedure.

A notable aspect of this case is the presence of malignant transformation despite classical benign clinical and radiological findings. Histopathological examination revealed papillary carcinoma arising within the thyroglossal duct cyst, characterized by papillary architecture, fibrovascular cores, nuclear clearing, grooving, overlapping, and psammoma body formation. These findings are consistent with papillary thyroid carcinoma arising from ectopic thyroid tissue within the cyst wall, supporting the theory that TGDC carcinomas originate from embryologically retained thyroid remnants.

Saavedra-Leveau et al. described a similar case of incidental papillary carcinoma arising in a TGDC in a younger patient, emphasizing that malignancy occurs in approximately 1% of cases and is frequently diagnosed only after histopathological evaluation [5]. In contrast, the present case occurred in an older individual, highlighting that malignant transformation can occur across a broader age spectrum and should not be excluded based on age alone.

Hu et al. reported that papillary carcinoma constitutes approximately 80–90% of TGDC-associated malignancies, while other histological subtypes such as squamous cell carcinoma are rare [6]. The tumor size in previously reported cases ranged from 1.5 to 2 cm, which is consistent with the present case, where the tumor measured approximately 2 cm, further supporting typical tumor characteristics described in the literature.

Gómez-Álvarez et al. reported that most TGDC carcinomas are located near the hyoid bone and commonly measure between 1 and 2 cm [7]. The anatomical location and tumor size observed in the present case closely align with these findings. Additionally, characteristic histopathological features such as nuclear grooves, overlapping, and psammoma bodies described in their series were also evident in our case, reinforcing the diagnostic consistency [8].

Taha et al. emphasized that TGDC accounts for nearly 70–75% of congenital midline neck masses and may present as a painless swelling in adults, as observed in the present case [9]. This highlights the importance of maintaining a high index of suspicion for TGDC even in adult patients presenting with midline neck swellings.

From a management perspective, the Sistrunk procedure remains the gold standard for TGDC, significantly reducing recurrence rates by excising the cyst, tract, and central portion of the hyoid bone. However, the optimal management of TGDC carcinoma remains a subject of ongoing debate. While Sistrunk procedure alone may be adequate in low-risk patients with no evidence of thyroid involvement, additional total thyroidectomy is often recommended in selected cases to exclude synchronous thyroid malignancy, facilitate radioactive iodine therapy, and enable long-term surveillance using thyroglobulin levels.

In the present case, although initial imaging demonstrated a normal thyroid gland, the unexpected diagnosis of papillary carcinoma on histopathology warranted further evaluation. The patient subsequently underwent total thyroidectomy, which revealed multinodular goitre without evidence of primary thyroid malignancy, thereby confirming the diagnosis of primary carcinoma arising in the thyroglossal duct cyst rather than metastatic disease. This step was crucial for accurate staging, risk stratification, and planning of further management.

Wynings et al. reported recurrence rates of less than 5-10% following the Sistrunk procedure, with low postoperative morbidity [11]. The present case demonstrated a similarly favorable postoperative course, with no complications or recurrence during follow-up.

Recent minimally invasive techniques such as the transoral endoscopic vestibular approach (TEVAS) have been explored to improve cosmetic outcomes; however, conventional open Sistrunk procedure continues to be the most reliable and widely practiced technique [12].

Zoupas et al. emphasized that although papillary carcinoma arising in TGDC is rare, long-term follow-up is essential due to the potential risk of recurrence and the possibility of synchronous or metachronous thyroid malignancy [13]. This underscores the importance of individualized management strategies and vigilant postoperative surveillance.

Conclusion

Thyroglossal duct cyst should be considered an important differential diagnosis in patients presenting with midline neck swelling, including in the adult population. Although the majority of cases are benign, the possibility of malignant transformation, though rare, must always be considered, particularly as it may not be clinically or radiologically apparent.

The present case highlights a rare instance of papillary carcinoma arising in a thyroglossal duct cyst in a 52-year-old male with classical benign clinical features. This emphasizes the indispensable role of routine histopathological examination of all excised TGDC specimens to ensure accurate diagnosis.

The Sistrunk procedure remains the gold standard surgical treatment and is associated with excellent outcomes in low-risk patients. However, the identification of malignancy warrants a comprehensive postoperative evaluation. In the present case, subsequent total thyroidectomy confirmed the absence of primary thyroid malignancy, supporting the diagnosis of primary TGDC carcinoma and guiding further management.

Overall, early diagnosis, appropriate surgical intervention, and meticulous histopathological assessment are crucial for optimizing patient outcomes. Long-term follow-up is essential to monitor for recurrence and to detect any potential thyroid involvement at an early stage.

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