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(CASE REPORT)

Papillary Carcinoma Arising in a Thyroglossal Duct Cyst in an Adult: A Rare Case Report and Review of Management

ACHRAF HAFDI *, MOHAMMED CHEHBOUNI, OMAR OULGHOUL, YOUSSEF LAKHDAR, YOUSSEF ROCHDI and ABDELAZIZ RAJI

Department of Otorhinolaryngology and Head-Neck Surgery, Arrazi Hospital, Mohammed VI University Hospital Center, Faculty of Medicine and Pharmacy of Marrakech, Cadi Ayyad University, Marrakech, Morocco.

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Abstract

Background: Thyroglossal duct cysts (TGDCs) are the most common congenital midline neck masses. Malignant transformation within a TGDC is rare, occurring in approximately 1% of cases, with papillary carcinoma being the most common histologic subtype. Because TGDCs generally present as benign lesions, carcinoma is often diagnosed incidentally after surgical excision, creating both diagnostic and therapeutic challenges.

Case Presentation: We report the case of a 33-year-old woman with no significant medical history who presented with a slowly enlarging, painless midline neck mass over six months. Physical examination revealed a mobile, non-tender infrahyoid mass that moved with tongue protrusion, without skin involvement or cervical lymphadenopathy. Imaging demonstrated a complex cystic lesion with internal calcifications suggestive of a thyroglossal duct cyst. The patient underwent a Sistrunk procedure. Histopathology revealed a 1.5 cm papillary carcinoma confined to the cyst wall, with clear surgical margins and no evidence of thyroid involvement. No additional surgery was performed. The postoperative course was uneventful, and follow-up showed no recurrence.

Conclusion: Papillary carcinoma arising in a TGDC is rare and difficult to diagnose preoperatively. In carefully selected low-risk patients, the Sistrunk procedure alone may be sufficient. Long-term follow-up is essential to ensure favorable outcomes.

Keywords: Thyroglossal Duct Cyst; Papillary Carcinoma; Midline Neck Mass; Sistrunk Procedure; Thyroid Cancer

1. Introduction

Thyroglossal duct cysts (TGDCs) are congenital anomalies resulting from incomplete involution of the thyroglossal duct during the embryologic descent of the thyroid gland. They represent the most common midline neck mass and are typically diagnosed in childhood or early adulthood, although presentation in later decades is not uncommon [5,6]. TGDCs are generally benign and asymptomatic, presenting as slowly enlarging, painless cervical swellings that characteristically move with swallowing or tongue protrusion.

Malignant transformation within a TGDC is rare, occurring in approximately 1% of cases, with papillary carcinoma accounting for the vast majority of reported malignancies [1–3]. Other histologic subtypes, such as squamous cell carcinoma and follicular carcinoma, are exceedingly uncommon [3]. Because of its rarity and nonspecific clinical features, TGDC carcinoma is often indistinguishable from benign TGDC on physical examination, and the diagnosis is most frequently established after surgical excision [1,5].

*Corresponding author: ACHRAF HAFDI

The pathogenesis of TGDC carcinoma remains controversial. Two principal theories have been proposed: the de novo theory, suggesting malignant transformation of ectopic thyroid tissue within the cyst wall, and the metastatic theory, proposing that the carcinoma represents metastasis from an occult primary thyroid malignancy [3,4]. Increasing evidence supports a de novo origin in most cases, particularly when the thyroid gland is normal on clinical and radiologic evaluation [2–4]. This distinction has important therapeutic implications, especially regarding the need for thyroidectomy.

Imaging modalities such as ultrasound and computed tomography (CT) play a key role in evaluating TGDCs. While most lesions demonstrate benign cystic features, the presence of solid components, mural nodules, or calcifications may raise suspicion for malignancy [6,9,10]. Nevertheless, imaging findings remain nonspecific, and preoperative diagnosis of TGDC carcinoma remains challenging.

The optimal management of TGDC carcinoma is still debated. While the Sistrunk procedure remains the standard treatment for TGDCs, the role of additional thyroidectomy and radioiodine therapy is controversial and largely depends on tumor characteristics and associated risk factors [2,6,9]. Given the rarity of this condition, most evidence comes from case reports and small case series, underscoring the need for continued reporting and analysis.

We present a rare case of papillary carcinoma arising in a thyroglossal duct cyst in an adult patient, successfully managed with the Sistrunk procedure alone, and discuss the diagnostic challenges and therapeutic considerations in the context of the current literature.

2. Case presentation

A 33-year-old female with no significant past medical history presented with a gradually enlarging, painless midline neck mass that had been evolving for 6 months. The mass was located just inferior to the hyoid bone. She reported no symptoms of dysphagia, dysphonia, or pain. On physical examination, the mass was soft, mobile, and non-tender, measuring approximately 3 cm in diameter. It moved with tongue protrusion and swallowing. There was no overlying skin involvement, and no fistula was observed. The thyroid gland was normal on palpation, and no cervical lymphadenopathy was detected (Figure 1). The patient was afebrile and in good general condition.

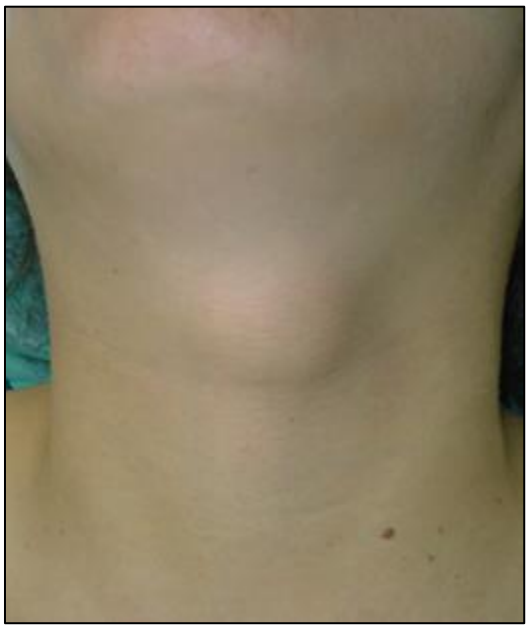

Figure 1 Clinical photograph showing a midline cervical mass in the infrahyoid region

The ultrasound revealed a well-defined, hypoechoic lesion in the midline, just below the hyoid bone, measuring approximately 3 cm. The mass exhibited mixed echogenicity with both cystic and solid components, consistent with a thyroglossal duct cyst. It had an irregular but well-defined capsule, and there was a small number of echogenic debris, suggesting prior infection or hemorrhage. The lesion was mobile with swallowing and tongue protrusion, indicating its connection to the thyroglossal duct. No enlarged cervical lymph nodes were seen, and the thyroid gland appeared normal with no signs of nodules or malignancy.

Contrast-enhanced computed tomography (CT) revealed a 3 cm midline complex cystic lesion with varied densities. The lesion showed small calcific specks and featured an enhancing nodular area within the cyst, located posterolateral. This nodular component abutted the left side of the hyoid bone, with slightly blurred margins suggesting possible local involvement. The lesion was consistent with a thyroglossal duct cyst, and there was no evidence of lymph node enlargement or distant metastasis. Additionally, a well-defined, isoechoic lesion was observed in the right thyroid lobe, which appeared benign (Figure 2).

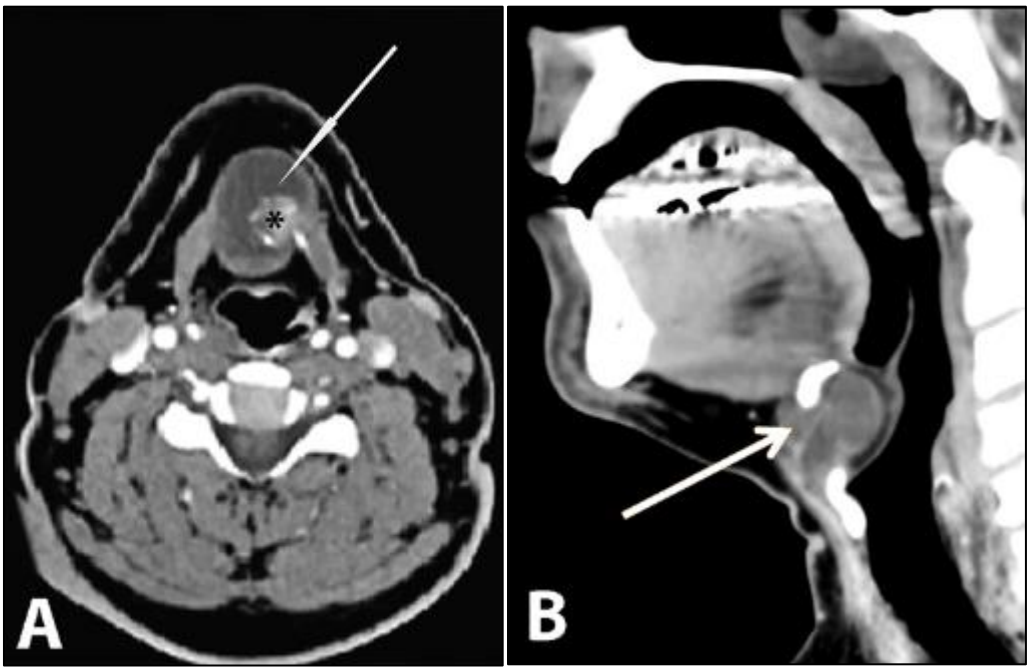

Figure 2 Contrast-enhanced computed tomography (CT) of the neck demonstrating a midline infrahyoid mass consistent with a thyroglossal duct cyst. (A) Axial section showing a well-defined complex cystic lesion with an internal solid component (white arrow) and punctate calcifications (asterisk). (B) Sagittal section illustrating the close relationship of the lesion to the hyoid bone and its midline location, with internal calcific foci (asterisks), raising suspicion of malignant transformation

The patient underwent the Sistrunk procedure (Figure 3), which involved excision of the thyroglossal duct cyst, the central portion of the hyoid bone, and a core of tissue extending to the foramen cecum to ensure complete removal of any residual duct tissue. The lesion, located midline below the hyoid bone, was excised, including the adjacent nodular compartment. The thyroid gland was examined, and no abnormalities were noted. The surgery proceeded without complications, and the patient recovered well postoperatively.

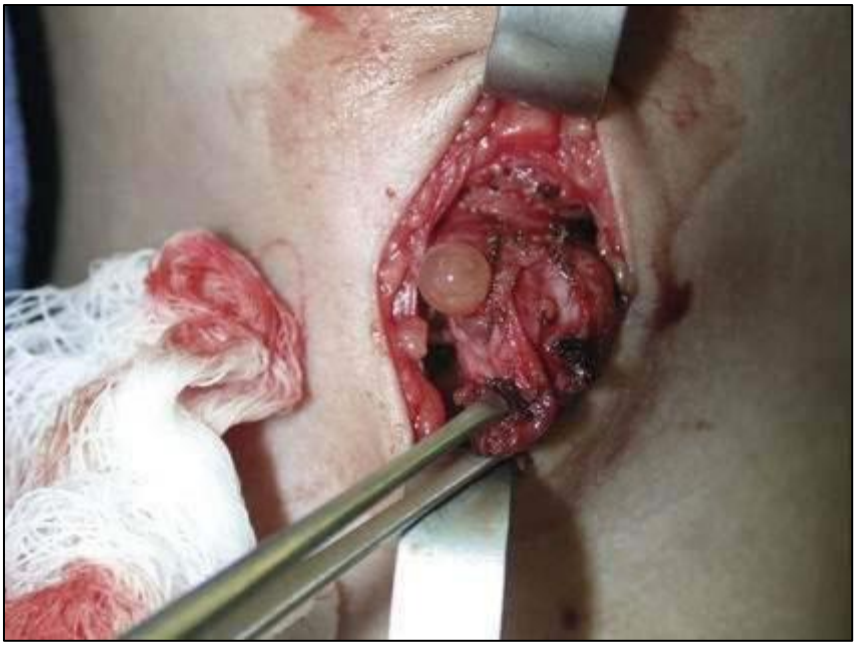

Figure 3 Intraoperative view demonstrating a midline thyroglossal duct cyst with well-defined margins, exposed during the Sistrunk procedure. The cyst is seen in close relation to the hyoid bone before complete excision

The definitive pathology report (Figure 4) revealed a papillary carcinoma within the thyroglossal duct cyst, measuring 1.5 cm in diameter. The tumor was confined to the cyst wall, with no evidence of extrathyroidal extension. Surgical margins were clear, with a safe distance of 1.2 cm from the tumor to the nearest tissue edge, confirming that the excision was complete and performed with healthy margins. The thyroid gland showed no signs of malignancy.

Given that the carcinoma was limited to the cyst with no evidence of thyroid involvement or lymph node metastasis, no further surgical procedures were required. The tumor was fully excised with healthy margins, and additional thyroidectomy or neck dissection was not needed. The patient was discharged after an uneventful recovery, with regular follow-up to monitor for recurrence.

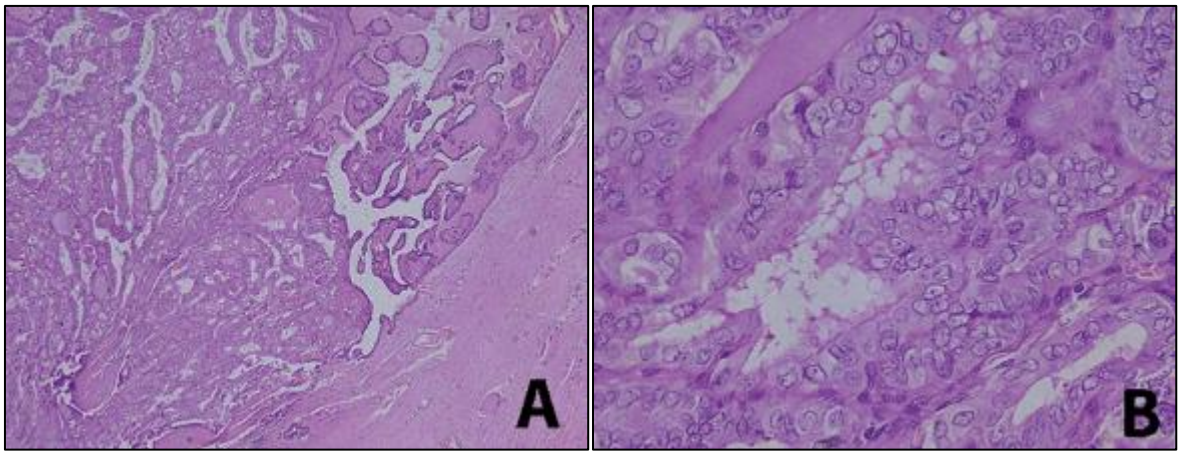

Figure 4 Histological findings from the excised specimen. (A) Low-power photomicrograph (×40) of the thyroglossal duct cyst wall with associated papillary carcinoma, predominantly exhibiting a follicular pattern (H&E staining). (B) High-power photomicrograph (×400) showing cellular details with nuclear clearing and characteristic nuclear grooves, typical of papillary carcinoma (H&E staining)

3. Discussion

Papillary carcinoma arising in a thyroglossal duct cyst (TGDC) is a rare pathological entity, representing approximately 1% of all TGDCs, with papillary carcinoma being the most frequently reported histological subtype [1–3]. Although TGDCs are common congenital anomalies of the neck, malignant transformation within these lesions remains exceptional. The rarity of this condition, combined with its benign clinical appearance, contributes to frequent delays in diagnosis and the predominance of postoperative incidental findings following surgical excision [1,5].

The histogenesis of TGDC carcinoma has been extensively debated. Two principal theories have been proposed: the de novo theory, which suggests malignant transformation of ectopic thyroid tissue entrapped within the cyst wall, and the metastatic theory, which considers the TGDC lesion as a secondary manifestation of an occult primary thyroid carcinoma [3,4]. Increasing molecular and clinicopathological evidence supports the de novo origin in most cases, particularly when the thyroid gland is normal on clinical examination and imaging, as in the present patient [2–4]. This distinction is of major clinical importance, as it directly influences the extent of surgical treatment and the indication for thyroidectomy.

From a clinical standpoint, TGDC carcinoma is almost indistinguishable from its benign counterpart. Patients typically present with a painless, slowly enlarging midline neck mass that moves with swallowing or tongue protrusion, reflecting its embryological tract [5–7]. Features traditionally associated with malignancy, such as rapid growth, fixation, skin involvement, or cervical lymphadenopathy, are inconsistently observed and lack sufficient predictive value [6,8]. As demonstrated in this case, the absence of alarming clinical signs does not exclude the presence of malignancy, particularly in adult patients.

Radiological evaluation plays a central role in the preoperative assessment of TGDCs, although its diagnostic specificity remains limited. Ultrasound is often the first-line modality and may reveal complex cystic lesions with internal solid components, mural nodules, or calcifications [6,9]. Contrast-enhanced computed tomography (CT) provides superior anatomical delineation and may demonstrate features suggestive of malignancy, such as irregular cyst walls, enhancing nodular components, or microcalcifications [9,10]. Among these findings, the presence of calcifications has been repeatedly associated with papillary carcinoma and should prompt heightened suspicion [9]. Nevertheless, imaging findings alone are insufficient for definitive diagnosis, and histopathological analysis remains the gold standard [3,6].

The role of fine-needle aspiration cytology (FNA) in the evaluation of TGDCs remains controversial and warrants careful consideration. While FNA may be useful in selected cases, particularly when solid components or suspicious lymph nodes are present, its overall sensitivity in TGDC carcinoma is low [6,9]. The cystic nature of these lesions frequently leads to non-diagnostic aspirates or false-negative results, as samples may contain only acellular fluid or inflammatory debris [5,6]. Several authors have reported that preoperative FNA rarely alters surgical management, as the Sistrunk procedure remains indicated for both benign and malignant TGDCs [2,3,9]. Consequently, routine preoperative FNA is not considered mandatory in clinically typical TGDCs and should be reserved for cases with atypical features or suspicion of synchronous thyroid malignancy [4,6].

The optimal management strategy for TGDC carcinoma continues to be debated, particularly regarding the need for additional thyroidectomy. The Sistrunk procedure remains the cornerstone of treatment and is widely accepted as sufficient for low-risk patients with tumours confined to the cyst, clear surgical margins, and no evidence of thyroid or lymph node involvement [1,2,6]. In such cases, outcomes are excellent, and the morbidity associated with more aggressive surgery can be avoided [3,9]. Conversely, total thyroidectomy, with or without radioiodine therapy, may be indicated in patients with high-risk features, including tumour size greater than 1 cm, invasion beyond the cyst wall, cervical lymph node metastasis, or concomitant thyroid carcinoma [4,10].

Prognosis in TGDC carcinoma is generally favourable, particularly in cases of localized disease managed appropriately. Reported survival rates exceed 90%, and disease-specific mortality is exceedingly low [2,3]. However, recurrence, although rare, has been documented, sometimes occurring several years after initial treatment [9]. This underscores the importance of long-term follow-up, including periodic clinical evaluation and imaging when indicated, even in patients treated conservatively.

This case adds to the growing body of evidence supporting a tailored, risk-adapted approach to TGDC carcinoma. It highlights the limitations of preoperative diagnostic tools, including FNA, and reinforces the central role of the Sistrunk procedure in achieving optimal oncological outcomes. Careful patient selection, thorough pathological assessment, and long-term surveillance remain essential components of effective management.

4. Conclusion

Papillary carcinoma arising in a thyroglossal duct cyst is a rare entity that remains difficult to distinguish from benign lesions on clinical grounds alone. This case highlights the importance of maintaining a high index of suspicion in adult patients presenting with midline neck masses, particularly when imaging reveals complex cystic features or calcifications. The Sistrunk procedure remains the cornerstone of treatment and can be curative in low-risk cases when the tumour is confined to the cyst and clear surgical margins are achieved. Routine thyroidectomy or neck dissection is not mandatory in the absence of thyroid involvement or lymph node metastasis. Long-term follow-up, however, is essential to detect potential recurrence. Early diagnosis, adequate surgical management, and careful pathological assessment are key to achieving excellent outcomes in this rare malignancy.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

The authors declare that they have no conflicts of interest to disclose.

Statement of ethical approval

If studies involve use of animal/human subject, authors must give appropriate statement of ethical approval. If not applicable then mention 'The present research work does not contain any studies performed on animals/humans subjects by any of the authors'

Statement of informed consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Research involving human participants

This case report was conducted in accordance with the principles outlined in the Declaration of Helsinki. In line within situational policy, ethical committee approval was not required as this report involves a retrospective description of a single clinical case with no identifiable personal information and no experimental intervention.

Consent for Publication

Consent for publication of the case details and figures were obtained from the patient's legal guardian.

Availability of Data and Materials

All data generated or analyzed during this study are included in this published article. Further information is available from the corresponding author upon reasonable request.

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Authors' Contributions

AH performed the surgery, led the clinical management, and prepared the manuscript. LY, OO, and MC contributed to case documentation, imaging interpretation, and literature review. YR and AR supervised the clinical management and critically revised the manuscript. All authors reviewed and approved the final version of the manuscript.

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