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Case Report

Thyroglossal duct cyst carcinoma in a young female: reassessing the need for total thyroidectomy after Sistrunk operation

Suhas S. S., Ashitha K.*, Spurthi Kabber

Department of ENT, Mysore Medical College and Research Institute, Mysore, Karnataka, India

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***Correspondence:**
Dr. Ashitha K.,
E-mail: ashithaksreekumar14@gmail.com

ABSTRACT

Thyroglossal duct cyst carcinoma (TGDCCa) is a rare malignancy, accounting for approximately 1% of thyroglossal duct cysts. Preoperative diagnosis is challenging because clinical and imaging findings often mimic benign lesions. Papillary thyroid carcinoma is the most common histological subtype, and optimal management, particularly the role of total thyroidectomy remains controversial. A 24-year-old female presented with a slowly progressive midline neck swelling of three years' duration. Ultrasonography and computed tomography (CT) revealed a cystic infra-hyoid lesion with a small solid component and microcalcifications. She underwent a Sistrunk procedure. Histopathological examination demonstrated a 4 mm focus of papillary carcinoma confined to the cyst wall with tumour-free margins, no hyoid bone invasion, and a normal thyroid gland. After counselling, she opted for surveillance rather than thyroidectomy. Postoperative thyroglobulin levels and serial imaging remained normal. This case highlights the indolent nature of low-risk TGDCCa and supports individualized management, with Sistrunk alone being adequate in select patients.

Keywords: Thyroglossal duct cyst carcinoma, Papillary thyroid carcinoma, Sistrunk procedure, Midline neck mass, Congenital neck anomaly

INTRODUCTION

Thyroglossal duct cyst (TGDC) is the most frequent congenital midline neck anomaly, accounting for nearly 70% of congenital neck masses, with an estimated prevalence of 7% in the general population.[1] It arises due to the persistence of the epithelial tract that connects the foramen cecum at the tongue base to the thyroid gland during embryologic descent.[2] Although TGDCs are typically benign, malignant transformation within these cysts is exceptionally rare, occurring in approximately 1% of cases.[3] Among the histological variants, papillary thyroid carcinoma (PTC) constitutes nearly 80–90% of thyroglossal duct cyst carcinomas (TGDCCa).[4] The condition is predominantly reported in adults and shows a slight female preponderance.[5]

The preoperative diagnosis of carcinoma in a TGDC remains challenging because its clinical features often overlap with benign cysts.[6] Imaging findings such as mural nodules, internal septations, microcalcifications, and solid components may raise suspicion of malignancy.[7] The standard surgical management for TGDC is the Sistrunk procedure, involving cyst excision with removal of the central hyoid bone and tract to the foramen cecum.[8] However, the optimal treatment for TGDCCa is controversial, especially regarding the need for total thyroidectomy and radioactive iodine ablation.[9] We present the case of a 24-year-old female with a midline neck swelling found to harbour a papillary carcinoma arising within a TGDC, managed successfully with Sistrunk procedure alone.

CASE REPORT

A 24-year-old female presented with a midline slowly progressive anterior neck swelling of three years' duration. The lesion had developed gradually and remained

asymptomatic except for mild discomfort on swallowing. She sought treatment for the swelling primarily for cosmetic reasons, forseeing her marriage. There were no symptoms of thyroid dysfunction, dyspnoea, dysphonia, dysphagia, constitutional complaints, significant weight loss, or loss of appetite. On examination, a smooth, well-circumscribed, 4×3 cm midline mass was palpable about 2 cm below the hyoid bone. It was soft, non-tender, and mobile, showing vertical movement with deglutition and tongue protrusion. The overlying skin was normal and mobile, and with no cervical lymphadenopathy. Thyroid palpation was unremarkable.

Laboratory tests such as haemoglobin, total WBC count, renal function tests, liver function test, thyroid function test, chest X-ray and electrocardiography (ECG) were within normal limits. Ultrasonography revealed a cystic lesion (38×18 mm) with a solid component (11×9 mm) containing septations and microcalcifications, with normal thyroid lobes. Contrast-enhanced CT demonstrated a well-defined infra-hyoid cystic mass with septal calcifications, confined to the subcutaneous plane and extending to the thyrohyoid membrane. The imaging findings were suggestive of a thyroglossal duct cyst. Fine-needle aspiration cytology did not reveal any malignant features.

The patient underwent a Sistrunk procedure with complete cyst excision and central hyoid bone removal. Gross appearance was that of a nodular grey-white tissue with attached piece of bone. The nodular tissue measures 4.0×3.3×2.8 cm. The external surface shows muscle tissue with no gross evidence of tumour. The hyoid bone identified, measures 2.0×1.6 cm. The bone appears free of any gross pathology. On cutting, the nodular tissue shows cystic spaces containing coagulated yellowish mucinous material. Focal solid-white and papillary areas are noted, measuring approximately 0.6 cm in maximum dimension (Figure 1).

Histopathology showed cystic tissue, lined by flattened to cuboidal to columnar epithelium. Areas shows denudations. Respiratory epithelium is also identified, focally. The wall shows fibrous tissue and skeletal muscle fibres. Multiple foci of a papillary neoplasm, composed of well-formed branching papillae with fibrovascular cores, covered by mildly pleomorphic cuboidal cells exhibiting nuclear crowding, chromatin-clearing, nuclear irregularity, intranuclear grooving and occasional pseudo-inclusions. The cytoplasm of these cells is pale eosinophilic (Figure 2).

The surgical margins appear free of tumour. No other tumour component identified. No large vessel invasion or perineural invasion identified. The final diagnosis was a well-differentiated (G1) papillary thyroid microcarcinoma arising within a thyroglossal duct cyst, with multifocal involvement (three to five foci), the largest measuring 4 mm. Tumour appears limited to the cyst component, without thyroid gland involvement.

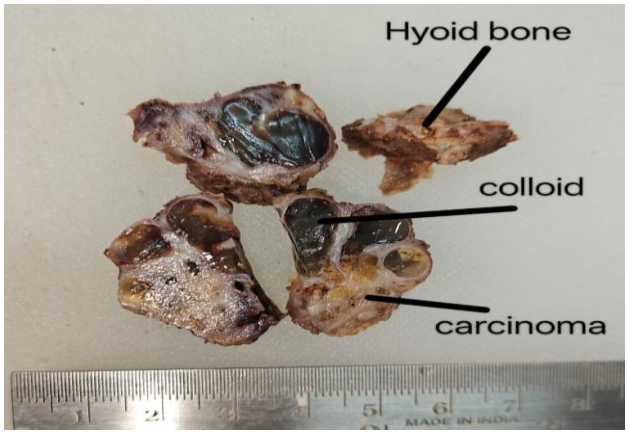

Figure 1: Post-operative specimen showing firm carcinomatous area in colloid filled cystic lesion.

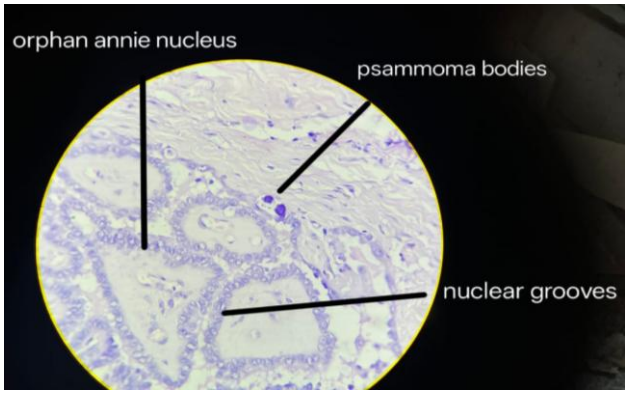

Figure 2: Histopathology showing classical features such as papillary fronds with fibrovascular cores lined by cuboidal to columnar epithelial cells showing optically clear ("orphan Annie eye") nuclei, nuclear crowding, grooves, and pseudo-inclusions.

The patient was informed of the pathological findings in detail, and two options for further treatment were discussed with her. The first option was total thyroidectomy, postoperative radioactive iodine ablation, and lifelong thyroxine tablets. The second option was close observation and follow-up with regular postoperative cervical ultrasound and thyroid hormones surveillance every 6 months, without additional surgical treatment. The patient had no high-risk factors and a low risk of recurrence as the postoperative pathology confirmed that the capsule was intact, the papillary thyroid carcinoma showed no external invasion, cervical ultrasound and enhanced CT showed no obvious space-occupying lesions in the thyroid, and no obvious enlarged lymph nodes on both sides of the neck. After full communication, the patient chose the second option as she was unmarried and didn't want to undergo thyroidectomy.

Postoperatively, serum thyroglobulin was normal. The patient was discharged uneventfully and placed on three-monthly follow-up with ultrasound and thyroid function assessment.

DISCUSSION

Carcinoma arising within a TGDC is a rare but clinically significant occurrence, representing less than 1% of all TGDCs.[3] The mean age of presentation is around the fourth decade, with a noted female predominance.[5] Clinically, it is indistinguishable from benign TGDCs, usually presenting as a midline, painless, mobile neck mass that elevates with tongue protrusion and swallowing.[10] A history of recent rapid enlargement, firm consistency, or fixation to surrounding tissues may indicate malignant change.[11] In our case, the patient's cyst had been stable for three years before a sudden increase in size over one month, prompting surgical evaluation.

The pathogenesis of TGDCCa remains debated. Two main theories exist: de novo carcinoma arising from ectopic thyroid tissue within the cyst wall, and metastasis from an occult primary thyroid carcinoma[12]. Evidence supporting a de novo origin includes the frequent absence of synchronous thyroid lesions, distinct histologic features, and separate molecular profiles.[13] In the present case, normal thyroid function, absence of thyroid nodules on ultrasonography, and negative histopathology confirmed a de novo origin.

Ultrasonography (US) remains the first-line diagnostic modality. Suspicious features include irregular cyst wall, mural nodules, solid areas, internal vascularity, and microcalcifications.[7,14] A recent study by Choi et al demonstrated that microcalcifications were present in 76% of malignant TGDCs compared with 8% of benign cysts, providing significant diagnostic value.[7] In our patient, US and CT revealed cystic and solid components with septations and microcalcifications, consistent with malignancy. Contrast-enhanced CT further delineates lesion extent, relation to hyoid bone, and potential lymph-node involvement.[15]

Histopathology remains the gold standard for diagnosis. Papillary carcinoma is the predominant variant, followed by mixed papillary-follicular and rarely squamous cell carcinoma.[4,5] Typical microscopic features include papillary fronds lined by cuboidal cells with nuclear clearing, grooves, pseudo-inclusions, and psammoma bodies.[6] In our specimen, a 4 mm focus of papillary carcinoma was confined within the cyst wall, with no capsular invasion or positive margins, corresponding to a low-risk microcarcinoma.

The Sistrunk procedure forms the basis of surgical management for all TGDCs, benign or malignant.[8] It significantly reduces recurrence compared with simple cyst excision. For TGDCCa, Sistrunk alone may suffice in selected low-risk cases characterized by tumour confined to the cyst, size <1.5 cm, absence of lymph-node or thyroid involvement, and age <45 years.[9] In our patient, all these criteria were fulfilled; hence, Sistrunk excision alone was considered adequate.

The role of routine total thyroidectomy in the management of thyroglossal duct cyst carcinoma continues to be debated, particularly in patients without thyroid or nodal involvement. It allows complete disease clearance, facilitates radioactive iodine therapy, and enables serum thyroglobulin surveillance.[10,11] However, recent meta-analyses have demonstrated no survival benefit in patients without thyroid or nodal involvement, and with low-risk histology.[13,14] Furthermore, performing thyroidectomy in such cases may expose patients to unnecessary surgical risks. Therefore, individualized treatment based on preoperative imaging, intraoperative findings, and histopathological risk factors is recommended.[9]

The role of lymph-node dissection is similarly debated. Prophylactic neck dissection is not recommended unless clinical or radiologic evidence of nodal metastasis exists.[8] In large multicentre reviews, cervical lymph-node metastases were observed in only 7–10% of TGDCCa patients which corresponding to about 6–12 patients out of 120 patients, in typical pooled cohorts.[13] The prognosis of TGDCCa is excellent regardless of whether only the Sistrunk procedure is performed, Sistrunk + total thyroidectomy, or Sistrunk + thyroidectomy + neck dissection is carried out as it behaves similarly to low-risk papillary thyroid carcinoma. TGDCCa needs individualized treatment, not a one-size-fits-all approach: Sistrunk alone is adequate for - small tumour, no high-risk features, no clinical/radiologic thyroid involvement, no lymph-node enlargement. Total thyroidectomy is recommended when - thyroid gland shows suspicious lesions, tumour >1 cm, patient prefers radioactive iodine therapy, high-risk histology, need for postoperative thyroglobulin surveillance. Neck dissection is performed only if lymph nodes are clinically or radiologically involved (not routine).[13] Reported 10-year survival rates exceed 95%, and recurrence rates are below 5% when adequate surgery is performed.[15] Factors associated with poor outcomes include extrathyroidal extension, large tumour size (>4 cm), lymph-node metastasis, and coexisting thyroid carcinoma.[12] Given the indolent nature of papillary carcinoma, even limited disease can be managed conservatively with vigilant follow-up.[13] Our patient had no palpable lymphadenopathy and negative imaging findings, hence neck dissection was not indicated.

Adjuvant radioactive iodine (RAI) therapy is generally reserved for high-risk patients or those undergoing total thyroidectomy.[9] Because our patient underwent only Sistrunk excision, and her postoperative imaging and serum thyroglobulin were normal, RAI was not indicated. Similarly, the benefit of thyroxine suppression therapy remains unclear in isolated TGDCCa; it may be considered in cases with thyroid involvement or recurrence risk.[14]

Follow-up protocols typically include physical examination and neck ultrasonography every 6–12 months, with periodic thyroid function and serum thyroglobulin monitoring.[8] In our case, three-monthly surveillance was initiated, and the patient remained

disease-free at the last follow-up, 6 months after surgery. Early detection of recurrence, though uncommon, allows timely intervention.

This case emphasizes the importance of considering malignancy in any TGDC that exhibits atypical imaging features like solid components, mural nodules, or calcifications within the cystic neck mass in CT and magnetic resonance imaging (MRI) or rapid growth. Awareness among clinicians can facilitate prompt diagnosis and appropriate management. Publication of such cases contributes to the growing literature that may help refine management guidelines for this rare condition.[14,15]

CONCLUSION

Primary papillary carcinoma arising in a thyroglossal duct cyst is a rare neoplasm that often mimics benign midline neck lesions. Diagnosis is typically confirmed only after surgical excision and histopathologic evaluation. This case demonstrates that thorough pathological examination of all TGDCs is essential, even when clinical and radiologic findings appear benign.

Our patient's disease was localized to the cyst with a normal thyroid gland, exemplifying the indolent nature and favourable prognosis of this tumour when treated with a Sistrunk procedure alone. Total thyroidectomy and radioiodine therapy should be reserved for high-risk patients with thyroid invasion or nodal metastasis. Long-term follow-up with periodic ultrasound and thyroglobulin estimation is recommended to detect recurrence or delayed thyroid involvement. The routine recommendation of total thyroidectomy, particularly in young unmarried women who wish to avoid additional surgery and lifelong thyroxine supplementation, warrants careful consideration. Is sistrunk alone sufficient, is still a question of debate.

Awareness of this uncommon malignancy and adherence to complete surgical excision are key to achieving excellent outcomes.

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