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Papillary Carcinoma Arising in a Thyroglossal Duct Cyst: A Case Report and Review of the Literature

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Case Report

Keywords: Thyroglossal duct cyst, Papillary thyroid carcinoma, Sistrunk procedure, Neck mass, Case report

Posted Date: April 9th, 2026

DOI: https://doi.org/10.21203/rs.3.rs-9349382/v1

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Additional Declarations: The authors declare no competing interests.

Abstract

Background

Thyroglossal duct cysts (TGDCs) represent the most common congenital cervical anomalies resulting from incomplete involution of the thyroglossal duct during embryologic development. Malignant transformation within a TGDC is rare, occurring in approximately **1–1.5% of cases**, with **papillary carcinoma** being the most frequent histological type.

Case Presentation

We report the case of a **30-year-old woman**presenting with a **midline cervical swelling** evolving over four years. Clinical examination revealed a **firm, painless anterior cervical mass** that was **mobile with tongue protrusion**. Imaging studies suggested a **multiloculated thyroglossal duct cyst with a solid component**. The patient underwent a **Sistrunk procedure**. Histopathological examination revealed **papillary thyroid carcinoma measuring 1.5 cm** arising within the thyroglossal duct cyst. Following multidisciplinary discussion, **completion thyroidectomy was recommended but declined by the patient**.

Conclusion

Papillary carcinoma arising in TGDC is rare and often diagnosed incidentally after surgical excision. The optimal therapeutic strategy remains controversial, particularly regarding the need for additional thyroidectomy. **The Sistrunk procedure remains the cornerstone of treatment**, while further management should be individualized according to clinical, radiological, and histopathological findings.

Introduction

Thyroglossal duct cysts (TGDCs) are **congenital cervical malformations resulting from the persistence of the thyroglossal duct**, an embryologic structure that normally extends from the **foramen cecum at the base of the tongue to the thyroid isthmus or the pyramidal lobe of Lalouette** during thyroid gland descent [1, 2].

Malignant transformation within a TGDC is rare, occurring in approximately **1–1.5% of cases**, and is predominantly represented by **papillary carcinoma** [1, 3, 4].

We report the case of a **30-year-old woman** who underwent surgery for a TGDC, in whom **histopathological examination revealed papillary thyroid carcinoma arising within the cyst**, and we review the relevant literature.

Case Report

A **30-year-old woman** with no significant past medical history presented with a **midline cervical swelling** that had been evolving for **four years**.

Clinical examination revealed a **multilobulated anterior cervical mass**, painless and non-compressive, **mobile on tongue protrusion**, firm in consistency, measuring approximately **5 cm in its greatest diameter**. No signs of local inflammation or **palpable cervical lymphadenopathy** were observed.

Cervical ultrasonography revealed a large **septated thyroglossal duct cyst** with vascularized septa and an **isoechoic hypervascular solid component**, associated with a **nodular goiter**.

A **cervical computed tomography (CT) scan** demonstrated an **anterior cervical mass located below the hyoid bone**, predominantly cystic and multilocular, containing a **central tissue component enhanced after contrast administration**.

The patient underwent a **Sistrunk procedure**, consisting of **en bloc resection of the cyst, its tract, and the central portion of the hyoid bone**.

Histopathological examination of the surgical specimen revealed a **papillary thyroid carcinoma measuring 1.5 cm in greatest dimension arising within the thyroglossal duct cyst**.

Following **multidisciplinary discussion**, completion treatment with **total thyroidectomy** was recommended. However, the patient **declined further surgery after being informed of the potential benefits and associated risks**.

Discussion

During the **third week of embryonic development**, the thyroid gland originates as an epithelial proliferation at the floor of the primitive pharynx at the level of the **foramen cecum**. The thyroid primordium then forms a duct that descends anterior to the pharyngeal gut, known as the **thyroglossal duct**, which normally involutes and disappears.

Failure of this involution results in persistence of the duct and the formation of a **thyroglossal duct cyst (TGDC)**.

TGDCs may occur anywhere along the course of the duct, extending from the **base of the tongue to the pyramidal lobe of Lalouette**.

Malignant transformation within TGDCs is rare, accounting for **1–1.5% of cases**, and occurs mainly in **adults during the fourth decade of life** [1,3,6].

The first description of carcinoma arising in a TGDC was reported by **Brentano in 1911** [3].

The **papillary histological subtype** is the most common, accounting for approximately **83% of cases**. Other reported histological types include:

- Mixed **papillary–follicular carcinoma (8%)**
- **Squamous cell carcinoma (6%)**

- Rare cases of **Hürthle cell carcinoma**
- **Follicular carcinoma**
- **Anaplastic carcinoma**
- **Epidermoid carcinoma**

No cases of **medullary carcinoma arising from TGDC** have been reported in the literature [1,3,6–7].

Most authors consider that TGDC carcinomas arise **de novo within ectopic thyroid tissue present in the cyst**, suggesting that they represent **primary thyroglossal duct carcinomas**. However, other authors, including **Belnoue**, have suggested that the thyroglossal duct may serve as a **pathway for the spread of primary thyroid carcinoma**, which explains the differing therapeutic approaches proposed in the literature [5].

Clinically, TGDC carcinoma usually presents similarly to a **benign thyroglossal duct cyst**, and the diagnosis is frequently **incidental following histopathological examination of the surgical specimen** [1].

However, certain clinical features may raise suspicion of malignancy, including:

- A **hard or fixed cervical mass**
- **Irregular borders**
- **Rapid increase in size**
- Presence of **cervical lymphadenopathy** [3]

The management of TGDC carcinoma remains **controversial**, particularly regarding the need for **thyroidectomy following the Sistrunk procedure** [6].

Some authors advocating a **more aggressive approach**, such as **Belnoue [5]** and **Miccoli [8]**, recommend **systematic total thyroidectomy** due to the relatively high frequency of associated thyroid carcinoma. Indeed, **11–27% of patients undergoing thyroidectomy are found to have synchronous thyroid carcinoma**. In addition, thyroidectomy facilitates **postoperative surveillance using thyroglobulin levels and radioactive iodine therapy when indicated**.

In a **meta-analysis**, **Patel [9]** demonstrated that the **extent of the initial surgical treatment** was the only significant factor influencing survival outcomes.

Conversely, other authors supporting a **more conservative strategy**, such as **Baïzri [1]**, recommend limiting treatment to the **Sistrunk procedure alone**, citing the **slow progression and excellent prognosis** of papillary carcinoma arising in TGDCs, provided that **regular follow-up is ensured**.

Overall, the **prognosis of TGDC carcinoma is favorable**, with a lower incidence of distant metastases compared with primary thyroid carcinoma [5]. According to **Patel [9]**, the **5-year and 10-year survival rates are approximately 100% and 95.6%, respectively**.

Conclusion

Papillary carcinoma arising in a thyroglossal duct cyst is a **rare clinical entity** that is most often diagnosed **incidentally following surgical excision**. Because **clinical and radiological findings cannot reliably distinguish a benign TGDC from malignant transformation**, the **Sistrunk procedure remains the standard initial treatment**. However, the **optimal postoperative management remains controversial**, particularly regarding the indication for **completion thyroidectomy**. Therefore, **therapeutic decisions should be individualized** based on clinical, radiological, and histopathological findings, as well as patient preferences.

Declarations

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

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Figures

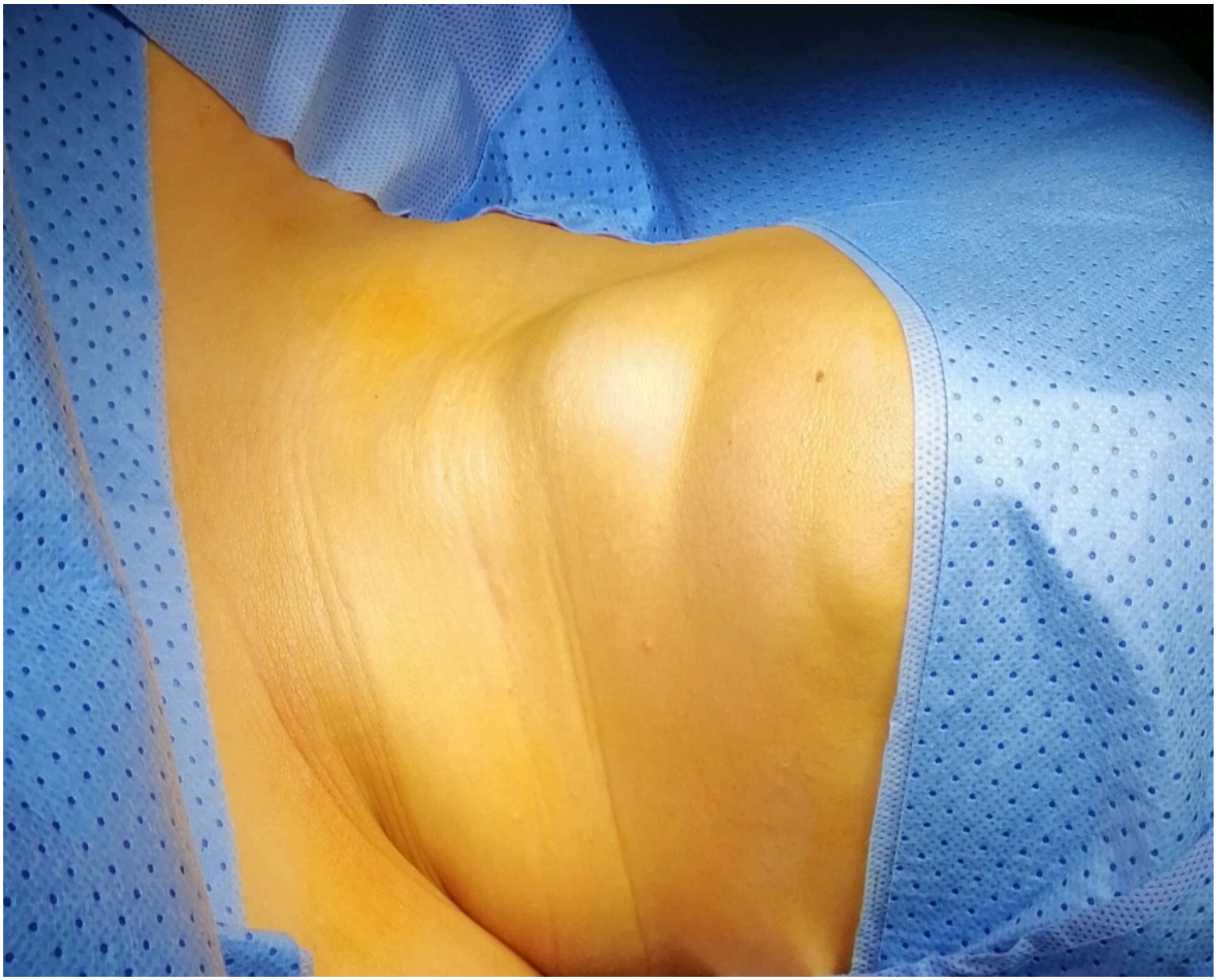


Figure 1

Midline cervical swelling suggestive of a thyroglossal duct cyst.

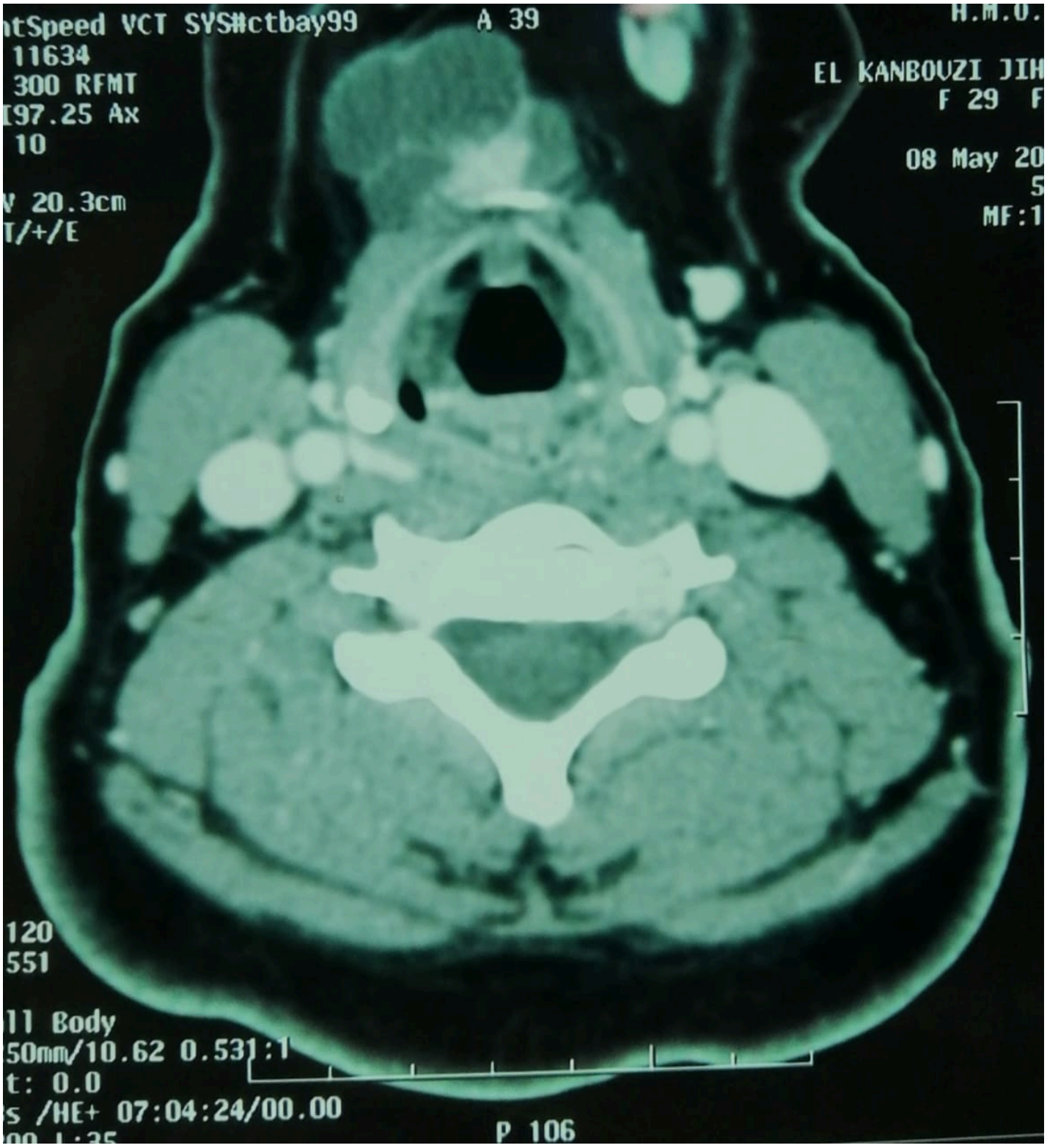


Figure 2

Cervical CT scan (axial view) showing a multilocular cystic mass suggestive of TGDC.



Figure 3

Intraoperative view of the thyroglossal duct cyst.

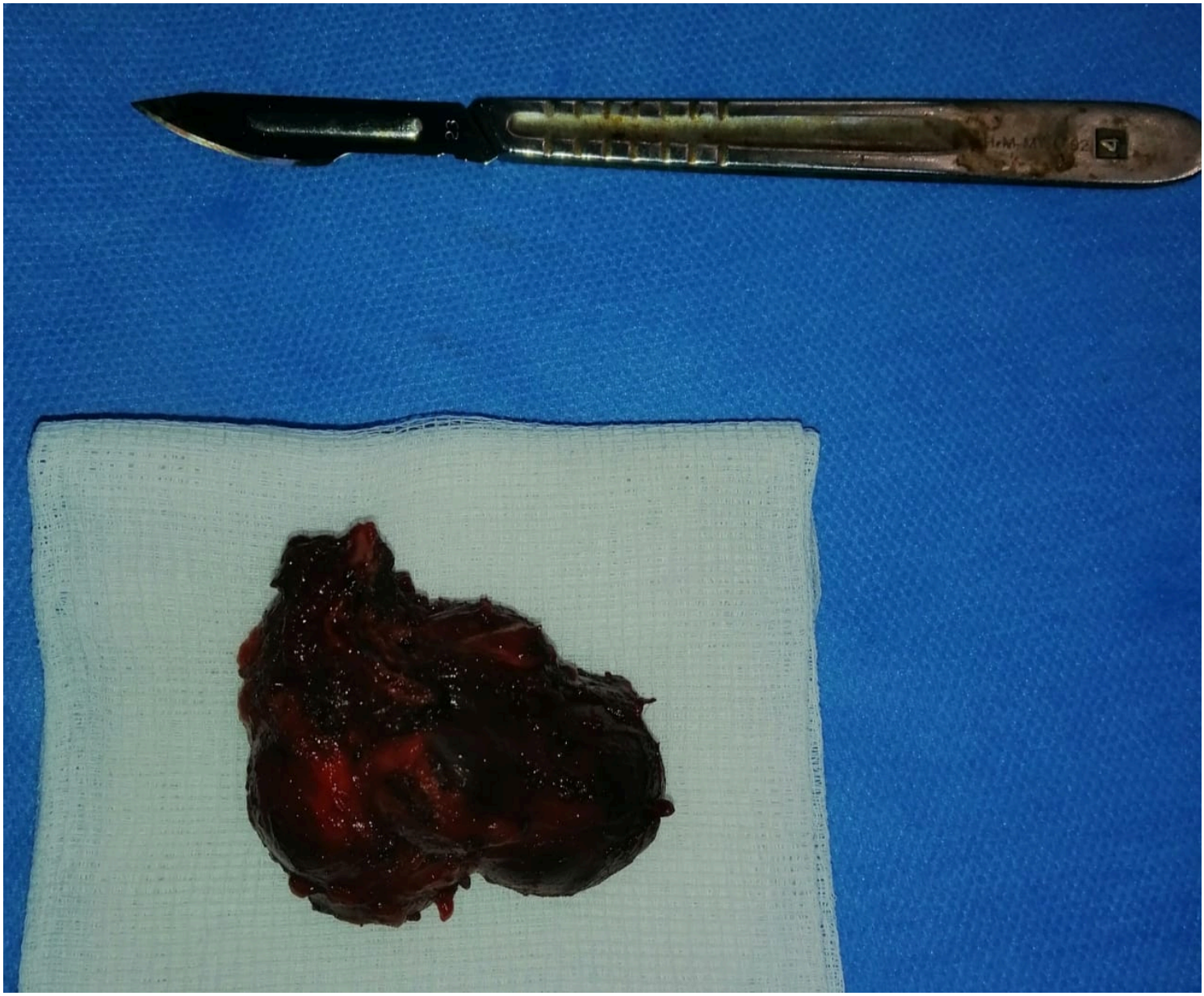


Figure 4

Surgical specimen following excision using the Sistrunk procedure.