Head and Neck Lesions

PRIMARY PAPILLARY CARCINOMA ARISING IN A THYROGLOSSAL DUCT CYST SIMULATING A CYSTIC LYMPHANGIOMA:

A Case Report With Literature Review

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ABSTRACT :

Background: Lateral cervical cystic masses in adults pose a diagnostic challenge, the majority being benign. Papillary thyroid carcinoma (PTC) arising in a thyroglossal duct cyst (TGDC) is rare and can mimic benign lesions, leading to mismanagement.

Case Presentation: A 34-year-old man with no significant medical history presented with a right lateral cervical mass evolving over 7 years. Ultrasound-guided fine-needle aspiration suggested a cystic lymphangioma. Cervicothoracic MRI revealed a large bilobed cystic formation (150 × 90 × 66 mm) with a central enhancing solid component (23 × 13 × 10 mm). The lesion displaced adjacent structures without evidence of invasion. Surgical excision revealed unexpected histology of PTC. Subsequent work-up identified an atrophic thyroid gland and a juxta-hyoid

solid mass. Total thyroidectomy combined with Sistrunk procedure confirmed a primary PTC arising in a TGDC, without intrathyroidal tumor foci.

Conclusion: This case emphasizes the need for systematic suspicion of malignancy in any cervical cystic mass containing a solid component, regardless of initial cytology. Primary PTC arising in a TGDC, although rare, should be included in the differential diagnosis to guide appropriately oncologic surgical management.

Keywords: *Papillary thyroid carcinoma; Thyroglossal duct cyst; Cystic cervical mass; Cystic lymphangioma; Diagnostic pitfall.*

1 .INTRODUCTION :

Lateral cervical cystic masses in adults represent a frequent diagnostic challenge in ENT practice, the majority of these lesions being benign [1]. Papillary thyroid carcinoma (PTC) is the most common endocrine malignancy, but its occurrence in ectopic thyroid tissue, particularly within a thyroglossal duct cyst (TDC), is rare, accounting for approximately 1–2% of resected TDCs [2].

Furthermore, a lateral cystic presentation can be misleading, clinically and radiologically mimicking benign lesions such as cystic lymphangioma or branchial cleft cysts [3]. This morphological similarity exposes patients to a significant risk of misdiagnosis and inadequate initial surgical management.

We report a challenging diagnostic case of primary PTC arising in a TDC, presenting as a large, long-standing lateral cervical mass, initially diagnosed and treated as a cystic lymphangioma. This case highlights a major diagnostic pitfall and allows discussion of the limitations of preoperative investigations and the key elements for optimal therapeutic management.

2. CASE REPORT

A 34-year-old man with no significant medical history, thyroid risk factors, or history of cervical irradiation presented with a right lateral cervical mass that had been progressively enlarging over seven years. The mass was painless, without local inflammatory signs or cutaneous fistulization. The patient’s primary concern was cosmetic.

On clinical examination, there was a right cervical mass that was fluctuant, non-pulsatile, and mobile relative to both superficial and deep planes, with no palpable satellite lymphadenopathy.

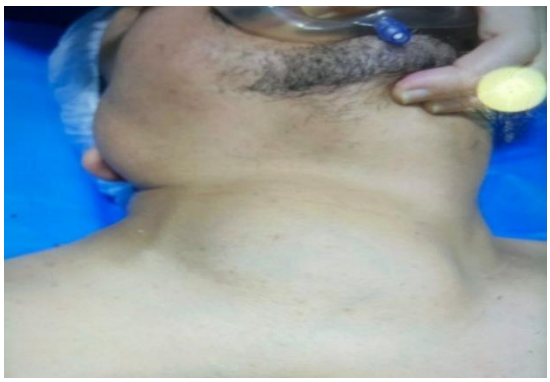

Figure 1: Anterior view of the right cervical mass, fluctuant, non-pulsatile and mobile, without palpable satellite lymphadenopathy

A fine-needle aspiration (FNA) was performed to evacuate the cystic fluid. Cytological analysis of the paucicellular fluid indicated a benign lesion consistent with a cystic lymphangioma, based on the presence of macrophages and the absence of atypical thyroid epithelial cells.

To assess the anatomical extent of this large lesion, a cervical and thoracic magnetic resonance imaging (MRI) study was performed. The MRI revealed:

A well-defined bilobed cystic mass was identified in the right lateral cervical and supraclavicular regions, measuring 150 mm in transverse length, 90 mm in height, and 66 mm in the anteroposterior plane.The lesion showed marked hyperintensity on T2-weighted images and hypointensity on T1-weighted images, consistent with a fluid content.A central solid tissue component was observed, appearing hypointense on T1 and heterogeneously hyperintense on T2

Following gadolinium administration, the solid component, measuring 23 × 13 × 10 mm, showed intense and heterogeneous enhancement. The mass displaced adjacent structures (trachea and vascular axes) without evidence of local invasion, intrathoracic extension, or identifiable vascular pedicle. The orthotopic thyroid gland was described as normal in size and signal intensity.

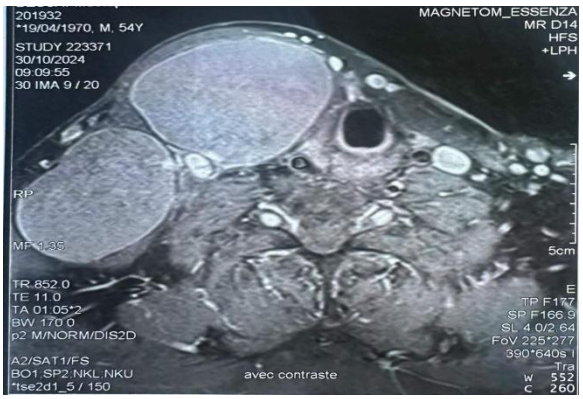

Figure 2: cervical MRI showing a giant bolobed cystic lymphangioma with an anhancing solid component after gadolinium injection

Based on the clinical findings, benign cytology, and the MRI appearance of a well-defined cystic lesion, a preoperative diagnosis of cystic lymphangioma was made. The patient subsequently underwent a complete surgical excision of the mass via a cervical approach.

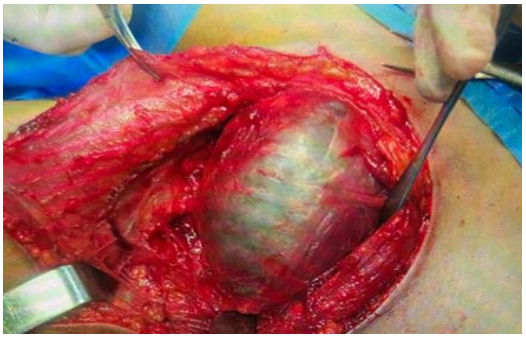

Figure 3 : Cystic mass of the thyroglossal duct identified during surgery

Histopathological examination of the surgical specimen revealed an unexpected diagnosis of classical papillary thyroid carcinoma. The tumor was localized within the wall of the cystic structure, without involvement of the cyst lumen.

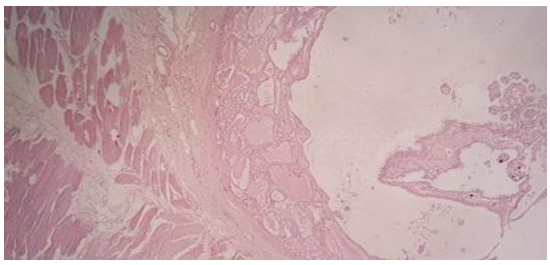

Figure 4: Papillary thyroid carcinoma arising in a thyroglossal duct cyst in close contact with adjacent striated muscle, without evidence of invasion

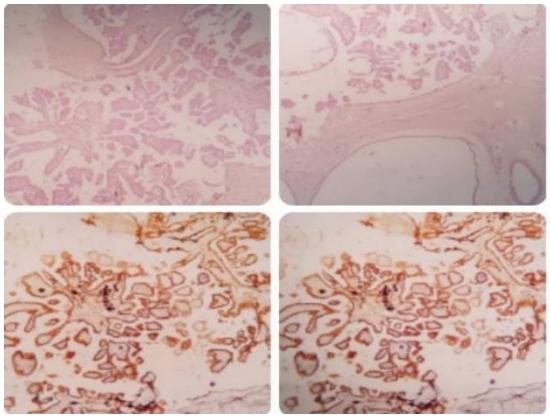

Figure 5 : Histopathological and immunohistochemical features of papillary thyroid carcinoma with cytoplasmic thyroglobulin expression

This unexpected finding prompted further staging investigations. A follow-up thyroid ultrasound revealed an atrophic thyroid gland without suspicious nodules. Additionally, a solid tissue formation measuring 15 mm was identified adjacent to the anterior aspect of the hyoid bone, suggestive of ectopic thyroid tissue along the thyroglossal tract.

The patient subsequently underwent a completion total thyroidectomy combined with resection of the hyoid mass using the Sistrunk technique. Definitive histopathological examination of the thyroid specimen revealed no primary tumor foci. Examination of the hyoid mass confirmed a thyroglossal duct cyst, with a focus of papillary carcinoma arising from its wall. The final diagnosis was primary papillary thyroid carcinoma developing in a thyroglossal duct cyst.

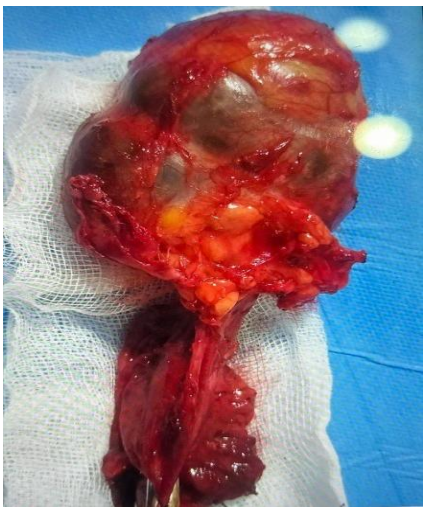

Figure 6 : Surgical specimen including the thyroid gland and the resected thyroglossal duct cystic mass

3. LITERATURE REVIEW

Primary Papillary Carcinoma in Thyroglossal Duct Cysts (TDCs)

Epidemiology

Thyroglossal duct cysts (TDCs) are developmental anomalies of the thyroid frequently encountered in ENT practice; however, malignant transformation of a TDC remains rare. The reported prevalence of malignancy varies across studies, with the most commonly cited value being approximately 1% of resected TDCs, although some series report slightly higher ranges depending on the cohort and selection criteria. The majority of tumors arising in TDCs are papillary carcinomas [1].

Tumor Origin: Primary vs Secondary

Two mechanisms have been discussed in the literature: (1) a primary carcinoma arising within ectopic thyroid tissue located in the wall of the thyroglossal duct cyst (TDC); and (2) extension or metastasis of an intrathyroidal papillary carcinoma to a thyroglossal duct remnant. Reported series include cases of both entities. The diagnosis of a primary carcinoma is supported by the absence of intrathyroidal tumor foci after total thyroidectomy and by the localization of the tumor within the cyst wall [2].

Clinical Presentation and Radiological Warning Signs

Clinically, malignant thyroglossal duct cysts most often present as an anterior or paramedian cervical mass, typically diagnosed in adulthood. Imaging studies (ultrasound, MRI, CT) play a crucial role in evaluation. The presence of an intracystic solid component or contrast enhancement should raise suspicion for a neoplastic lesion and represents a radiological “red

flag", warranting targeted investigations (fine-needle aspiration directed at the solid component, comprehensive thyroid assessment) rather than blind surgical excision.
In several series, tumor size is often modest, although cases of large cystic masses have also been reported [1].

Cytology (FNA): Performance and Limitations

Fine-needle aspiration (FNA) of thyroglossal duct cysts (TDCs) has lower sensitivity and diagnostic yield compared with solid thyroid nodules, particularly in cystic lesions with fluid content. Cellular yield may be low, and tumor cells are often confined to the cyst wall, resulting in false-negative results. Several series and reviews have reported limited preoperative diagnostic accuracy, with sensitivities frequently around 50–60% in cystic TDCs, which explains why the diagnosis is often made only after histopathological examination.

Therefore, a benign cytological result obtained from cystic fluid does not exclude an underlying malignant epithelial lesion when imaging demonstrates a solid intracystic component [3].

Histological Type and Biological Behavior

Papillary carcinoma is by far the most frequently reported histological type in malignant thyroglossal duct cysts (TDCs), accounting for the vast majority of cases across published series. The biological behavior is generally indolent, with low local aggressiveness. Regional lymph node involvement and distant metastases are uncommon, although they have been reported, which justifies an appropriate staging workup, including cervical ultrasonography and additional imaging when clinically indicated [4].

Surgical Management — Controversies and Treatment Algorithms

The standard surgical treatment for a benign thyroglossal duct cyst (TGDC) is the Sistrunk procedure, which consists of excision of the cyst, the central portion of the hyoid bone, and the thyroglossal tract up to the foramen cecum.

In cases of carcinoma arising in a TGDC, the most commonly reported management strategies include:

Sistrunk procedure alone for low-risk cases (young patients, tumor size < 1–1.5 cm, confinement to the cyst wall, normal thyroid gland on ultrasound, and absence of suspicious cervical lymphadenopathy);

Sistrunk procedure combined with total thyroidectomy, with or without cervical lymph node dissection and adjuvant radioactive iodine therapy, for higher-risk cases (tumor size > 1–1.5 cm, extracystic extension, coexisting thyroid abnormalities on imaging, older age, or lymph node involvement).

Several published algorithms advocate tailoring the surgical approach according to these risk factors. Although no absolute consensus exists in the literature, there is a growing tendency toward individualized management, with total thyroidectomy recommended when there is suspicion of synchronous intrathyroidal disease or when concerning histopathological or radiological features are present. [5]

Prognosis and Follow-up

The prognosis of papillary carcinomas arising from a thyroglossal duct cyst is generally favorable, particularly when the tumor is confined to the cyst and appropriately managed. Follow-up includes regular clinical examination and cervical ultrasound, and in selected cases, serum thyroglobulin measurement when total thyroidectomy has been performed. The indication for radioiodine therapy depends on thyroid status, tumor size, and the conventional risk factors associated with differentiated thyroid carcinoma.

Our case highlights a triple diagnostic challenge: an atypical lateral location, a predominantly cystic morphological presentation, and an initially misleading cytological result, which led to delayed diagnosis and an oncologically incomplete initial surgical management.

Low cellular yield, dilution of diagnostic cells within cystic fluid, and the presence of macrophages may misleadingly suggest a lymphangioma or a branchial cleft cyst, thereby masking the malignant epithelial nature of the lesion.

In our case, however, MRI demonstrated a major warning sign: the presence of a solid tissue component with gadolinium enhancement. Retrospectively, this radiological red flag was insufficiently weighted in the context of a benign cytological result.

Any contrast enhancement within a cervical cystic mass, even when minimal, should systematically raise suspicion for neoplasia and preclude simple excision, in favor of primary oncologic resection.

Case Novelty and Papillary Carcinoma in a Thyroglossal Duct Cyst

Our case exhibits several unique features that underscore its educational value:
Exceptional size: The lesion measured 15 cm, a dimension rarely reported in the literature for a TGDC, whether benign or malignant.
Predominantly lateral location: While TGDCs are typically midline structures, the massive lateral cervical presentation in this case is atypical and contributed to the initial diagnostic error.
Prolonged course: The 7-year clinical history highlights the slow growth potential of these tumors, which may create a false sense of security.
The diagnosis of primary papillary carcinoma in a TGDC is supported by the absence of tumor in the native thyroid gland and the histopathological confirmation of tumor origin within the thyroglossal duct remnant. The reference surgical management for a TGDC complicated by carcinoma consists of the Sistrunk procedure combined with total thyroidectomy, allowing for optimal local oncologic control [1,2].

Limitations of Preoperative Work-up and Key Takeaways

This case highlights the limitations of a sequential, non-integrative diagnostic approach. The discordance between imaging, revealing a suspicious solid component, and cytology, which remained benign, should have prompted increased caution and possibly a repeat targeted fine-needle aspiration of the solid portion.

Based on these findings, a decision-making algorithm is proposed to guide the management of cystic cervical masses in adults. (Figure 7)

In adults, any cystic cervical mass should raise suspicion of malignancy, regardless of location or duration. Initial evaluation relies on high-resolution imaging, starting with cervical ultrasound of the thyroid and lymph node areas, and, if necessary, complemented by MRI or contrast-enhanced CT to identify high-risk radiological signs, such as an intra-cystic solid component, an enhancing parietal nodule, calcifications, irregular cyst walls, or extracystic extension.

Fine-needle aspiration cytology (FNAC) further complements the evaluation, targeting the solid component when present. However, FNAC of cystic lesions may have low cellularity and a high false-negative rate; therefore, benign cytology does not exclude malignancy. In cases of discordance between imaging and cytology or when high-risk radiological signs are present, the lesion should be considered potentially malignant.

Systematic preoperative thyroid evaluation searches for synchronous nodules, ectopic thyroid tissue, or suspicious cervical lymph nodes. Surgical strategy is then adapted according to the risk

profile: low-risk lesions may undergo a Sistrunk procedure alone, whereas high-risk lesions, defined by at least one severity criterion (size >1–1.5 cm, enhancing solid component, atypical location, extracystic invasion, thyroid or lymph node abnormalities, or imaging–cytology discordance), require a Sistrunk procedure combined with total thyroidectomy, possibly with selective lymph node dissection.

Finally, definitive histopathology confirms the diagnosis and guides adjuvant treatment, including radioactive iodine therapy according to tumor size, extension, and lymph node involvement..

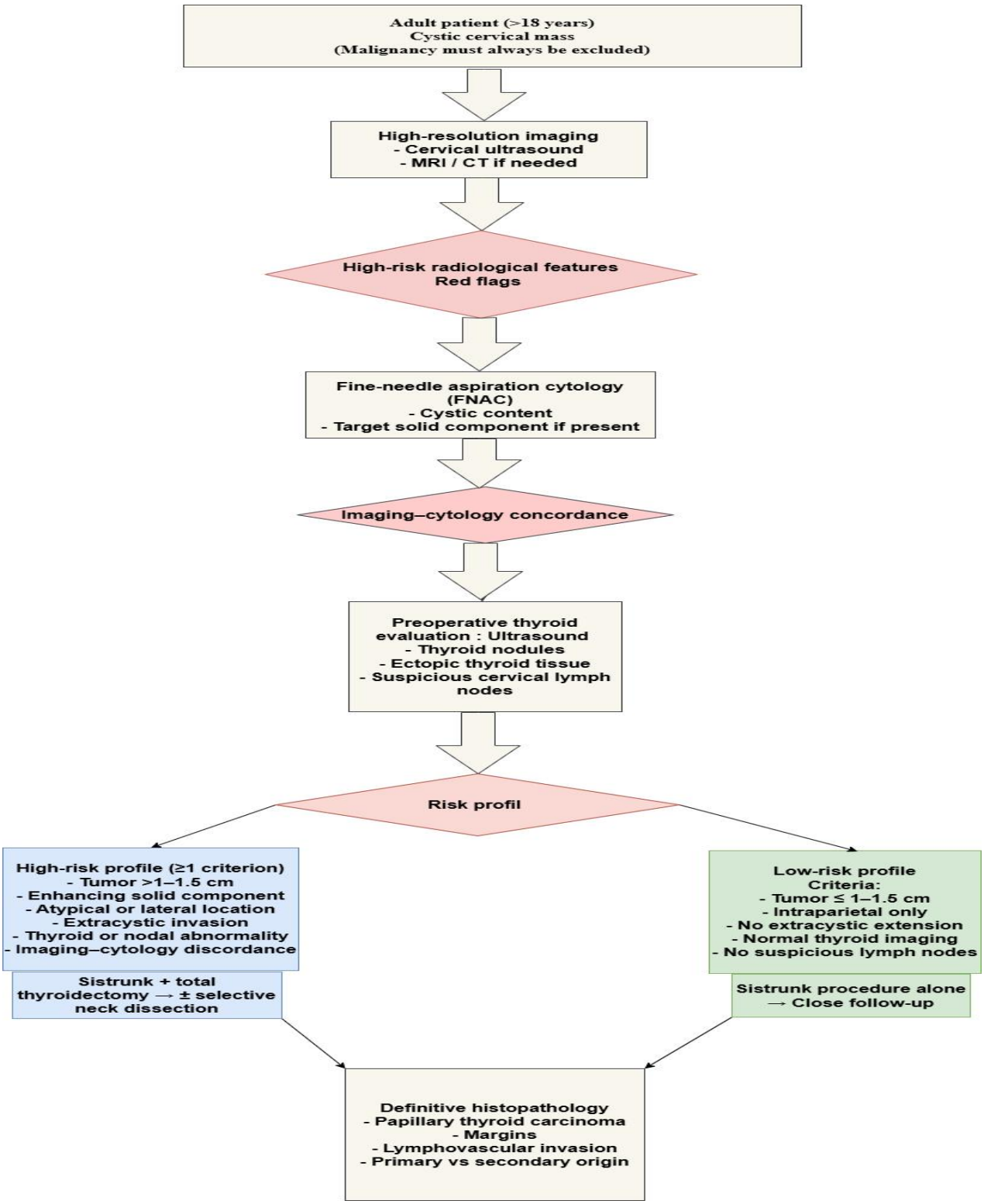
Adult patient (>18 years)
Cystic cervical mass
(Malignancy must always be excluded)
High-resolution imaging
- Cervical ultrasound
- MRI / CT if needed
High-risk radiological features
Red flags
Fine-needle aspiration cytology
(FNAC)
- Cystic content
- Target solid component if present
Imaging–cytology concordance
Preoperative thyroid
evaluation : Ultrasound
- Thyroid nodules
- Ectopic thyroid tissue
- Suspicious cervical lymph
nodes
Risk profil
High-risk profile (≥1 criterion)
- Tumor >1–1.5 cm
- Enhancing solid component
- Atypical or lateral location
- Extracystic invasion
- Thyroid or nodal abnormality
- Imaging–cytology discordance
Sistrunk + total
thyroidectomy → ± selective
neck dissection
Low-risk profile
Criteria:
- Tumor ≤ 1–1.5 cm
- Intraparietal only
- No extracystic extension
- Normal thyroid imaging
- No suspicious lymph nodes
Sistrunk procedure alone
→ Close follow-up
Definitive histopathology
- Papillary thyroid carcinoma
- Margins
- Lymphovascular invasion
- Primary vs secondary origin

Figure 7: Management of Adult Cystic Cervical Masse

Ethics and Consent

The patient provided written informed consent for the publication of this case report and any accompanying images.

The study was conducted in accordance with the ethical standards of the institutional Ethics Committee of [Mustapha University Hospital, Algiers, Algeria] and the 1964 Helsinki Declaration and its later amendments.

CONCLUSION

This report serves as an important reminder for clinicians, radiologists, and cytopathologists. Cervical cystic masses, particularly those with a long-standing course and a solid component on imaging, should be approached with a high level of suspicion. The presence of contrast enhancement within the cyst constitutes a red flag that should take precedence over reassuring cytology and prompt consideration of a neoplastic lesion. Although rare, the possibility of a primary papillary carcinoma arising in a thyroglossal duct cyst should always be included in the differential diagnosis. Appropriate initial surgical planning, including the Sistrunk procedure combined with total thyroidectomy, is essential to avoid repeat surgeries and to ensure the best possible patient outcome.

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