Case Report

Thyroid abscess secondary to infected synovial sarcoma of the thyroid: A case report

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ABSTRACT

Primary sarcomas of the thyroid are very rare, accounting for <1.5% of all thyroid cancers. Due to its rarity and similarities to benign inflammatory conditions, diagnosing it accurately is challenging. We report a unique case of thyroid synovial sarcoma presenting like a thyroid abscess. A 50-year-old female presented with a painful, rapidly growing anterior neck swelling and fever over the past month. The neck mass measured 10 × 10 cm, was firm, mildly tender, and had erythematous skin. An enlarged right supraclavicular lymph node was also noted. Imaging showed a complex solid-cystic mass. Initial antibiotic treatment failed as the white blood cell count increased and fever persisted. Ultrasound-guided aspiration yielded hemopurulent fluid. Due to worsening sepsis, surgical drainage and open biopsy were performed. Histopathological analysis confirmed a diagnosis of synovial sarcoma, with immunohistochemistry showing positivity for transducin-like enhancer 1, cytokeratin, B-cell lymphoma, epithelial membrane antigen, and CD99. This case highlights the need to consider rare sarcomas when diagnosing thyroid abscesses, especially if standard treatments fail. Early and precise diagnosis through histopathological and molecular tests is vital for determining the best treatment approach. The literature suggests that early recognition and intensive multimodal management may improve outcomes for this rare but aggressive cancer.

Key words: Synovial sarcoma, Thyroid abscess, Thyroid gland

Non-epithelial tumors of the thyroid, such as sarcoma or lymphoma, are very rare. Synovial sarcoma is a type of soft-tissue sarcoma, making up 8–10% of all these tumors [1]. It is a high-grade malignant neoplasm of mesenchymal pluripotent cells, with a specific chromosomal translocation that leads to the SYT-SSX fusion gene. Synovial sarcoma can have either a monophasic or classic biphasic pattern, based on the cell structures. Synovial sarcoma in the head and neck region is relatively unusual, and primary synovial sarcoma of the thyroid gland is extremely rare, with <20 cases reported globally.

Due to the rarity of this disease, there is no global consensus on management. In this study, we report a case of a middle-aged woman who initially presented with features suggestive of a multi-nodular goiter with thyroiditis, and subsequently developed a thyroid abscess with an underlying thyroid synovial sarcoma. This posed a significant challenge in diagnosis and management, as no similar case has been reported online to date. We hope this case report and literature review can raise awareness of thyroid synovial sarcoma and improve the diagnosis and treatment of this rare disease.

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CASE PRESENTATION

A 50-year-old female presented with a rapidly enlarging, painful neck mass and fever for 1 month. She also has dysphagia, but denied shortness of breath. She has no loss of weight, and no similar diseases running in the family.

On examination, her blood pressure was 130/86 mmHg, pulse 95 beats/min, and temperature was 37.8°C. Neck examination showed a huge lobulated anterior neck mass measuring 10 × 10 cm (Fig. 1), which was firm and mildly tender with erythematous skin. The edge was vague, and the trachea was deviated to the left with a palpable right supraclavicular lymph node measuring 2 × 2 cm. Clinically, she was euthyroid.

Total white cell count noted to be markedly elevated, 34 × 10^9/L, and thyroid hormone was normal. The US neck showed a heterogeneous mass measuring 7.3 cm × 10.6 cm × 8.7 cm (Fig. 2) with no moving debris seen. The right supraclavicular lymph node measures 2.1 × 3.9 × 2.6 cm with loss of fatty hilum. Contrast-enhanced computed tomography (CECT) of the neck showed the right thyroid lobe was diffusely enlarged, measuring 9.8 × 8.0 × 10.4 cm (Fig. 2). There were multiple heterogenous solid-cystic

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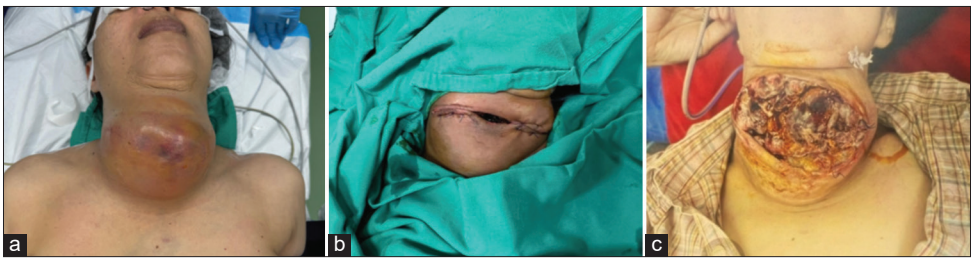

Figure 1: Photo of the tumor anteriorly pre-operative (a), post-operative (b), and 1 month post-operative (c)

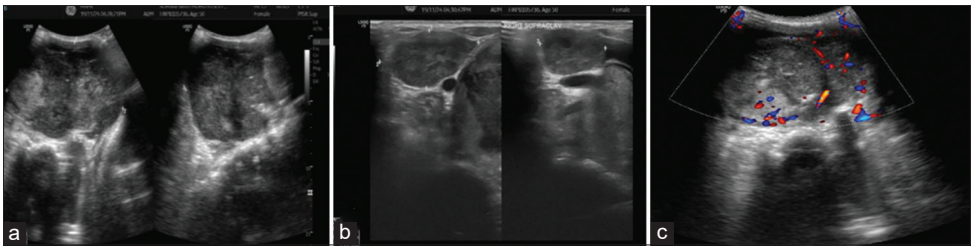

Figure 2: Pre-operative ultrasonography of the patient (a and b), color Doppler imaging of the tumor (c)

nodules seen in the right lobe, the largest cyst measures 5.8 × 4.9 cm (AP × W) with a rim of calcification seen. The mass is compressed onto the bilateral neck vessels. However, there was no clear plane between the trachea, esophagus, prevertebral space, and anterior muscle. The right lobe extends until the angle of the mandible superiorly and the sternal edge inferiorly.

Initially, a thyroid abscess with thyroiditis was suspected due to the patient's sepsis presentation. Intravenous amoxicillin/clavulanate potassium was started, but the patient failed to respond. An ultrasound-guided aspiration drained 50 mL of pus; however, the culture results were negative. Histological analysis revealed a highly cellular tumor with spindle cells, nuclear pleomorphism, and frequent mitosis. Immunohistochemistry (IHC) was positive for transducin-like enhancer 1, cytokeratin, and vimentin, and negative for thyroid markers, consistent with primary synovial sarcoma of the thyroid (Fig. 3). However, the characteristic translocation could not be confirmed due to the unavailability of the test.

Despite aggressive treatment, the patient's condition worsened, with increasing white cell count. After the discussion, a staged surgical approach was planned to address the sepsis first. Intraoperatively, the tumor was found to be cystic-solid with hemorrhagic and necrotic components, resulting in significant bleeding (Fig. 1b). The patient required a blood transfusion and intensive care management. Unfortunately, further imaging showed rapid tumor growth, with possible invasion of laryngeal structures and increased lymph node size (Fig. 4). In view of the aggressive nature of the tumor and the patient's deteriorating condition, the decision was made to provide palliative care. The patient was discharged home and referred to palliative services, succumbing to the disease within 4 weeks (Fig. 2c).

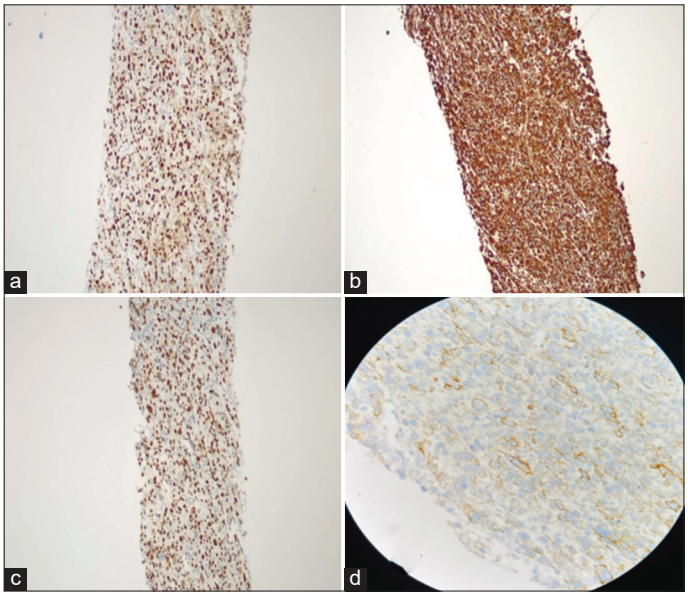

Figure 3: Histopathology examination of the patient. (a) Vimentin positive, (b) transducin-like enhancer 1: Diffuse positive, (c) P53: >80% strong positive, and (d) CD 99 positive

DISCUSSION

Soft-tissue sarcomas are solid tumors of mesenchymal origin, comprising over 50 different histological subtypes. It has been shown to arise at almost any anatomic location, including viscera, and approximately 10% affects the head and neck region [1,2]. However, the thyroid is an exceptionally rare site.

The early-stage clinical features can resemble a benign thyroid nodule. This may lead to a misdiagnosis of multi-nodular goiter preoperatively. However, its rapid growth and clinical presentation may later mimic advanced or anaplastic thyroid carcinoma (ATC). It may present with neck compression symptoms. Infected synovial sarcoma of the thyroid with abscess formation is rare, and this case may be the only one reported. It can present with hyperthyroidism if inflammation occurs, or hypothyroidism if the cancer cells damage most thyroid follicles.

The diagnosis of synovial sarcoma of the thyroid gland can be challenging using only ultrasound and fine needle aspiration

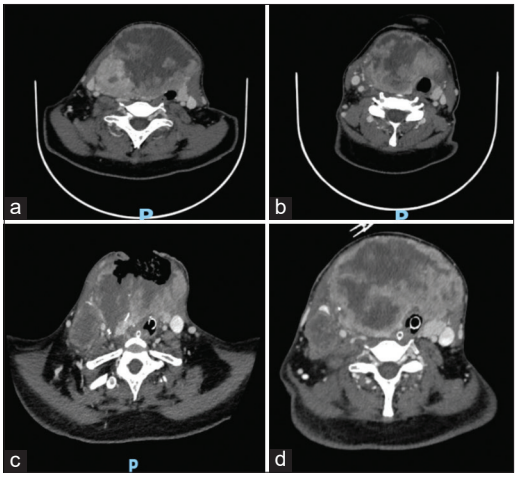

Figure 4: Contrast-enhanced computed tomography neck of the patient preoperatively (a and b) and postoperatively (c and d)

cytology (FNAC). If sarcoma is suspected, a core needle biopsy under ultrasound guidance is recommended to examine the solid areas [3]. However, as synovial sarcoma of the thyroid is a rare condition without distinctive imaging features, it is not commonly considered a diagnosis. The characteristic chromosomal translocation (X;18), detected by polymerase chain reaction (PCR) or fluorescent *in situ* hybridization, is the definitive test for synovial sarcoma, present in over 90% of cases [4]. However, this investigation may not be available in all hospitals.

Some authors have proposed using intraoperative frozen sections to help surgeons make clinical decisions and avoid reoperation. However, this approach has a high false-negative rate, as histology diagnosis requires more than just a simple microscopic examination and may need special tissue staining. Therefore, it is not recommended. The immunohistochemical profile of these tumors is crucial in supporting the diagnosis, as synovial sarcomas typically express markers such as vimentin, epithelial membrane antigen, B-cell lymphoma 2, and CD99 [5]. This IHC profile was particularly important in this case, as the other PCR or FISH tests were not available. According to research by Maduekwe *et al*., synovial sarcoma is thought to have a tendency to metastasize to lymph nodes, necessitating the need for possible lymph node FNAC or biopsy [6].

In a study conducted in Changsha, China, they found that most cases of thyroid synovial sarcoma have a disease course of less than a year, and most patients experience recurrence within 2 years [2]. Out of eight cases with survival data, six patients had fatal events. The longest survival time was 126 months, and four patients died within 3.5 years [7]. Primary thyroid synovial sarcoma generally shows rapid disease progression and a relatively poor prognosis. In eight cases, FNAC was performed, but only one was suspected to be a malignant spindle cell tumor. Most patients were diagnosed with medullary thyroid carcinoma (MTC) or undifferentiated thyroid carcinoma (UTC) by FNAC [8]. These data indicate that the cytopathological diagnosis of this disease is challenging.

Imaging of the neck with CECT or magnetic resonance imaging, from the base of the scalp to the thorax, is essential for staging. Computed tomography (CT) may reveal a large, mixed cystic-solid mass in the neck. In addition, CECT of the abdomen and pelvis should be performed to assess for distant metastases, as the lungs are the most common site [9]. Equivocal lesions can be further evaluated with targeted ultrasound, with or without sampling, or fluorodeoxyglucose positron emission tomography/CT.

Primary synovial sarcoma of the thyroid can be mistaken for MTC, undifferentiated/ATC, or spindle epithelioid tumors with thymus-like differentiation [2]. MTC patients typically have elevated calcitonin levels. MTC tissue often consists of lympho/plasmacytoid cells, atypical cells, or spindle cells. MTC tumor cells generally express calcitonin, thyroglobulin, and neuroendocrine markers [10]. The premenstrual symptoms screening tool may be misdiagnosed as undifferentiated/ATC. UTC/ATC, anaplastic thyroid cancer commonly presents as a rapidly growing thyroid mass, is more common in elderly patients, and is composed of spindle cells with abundant cytoplasm, coarse chromatin, frequent mitoses, giant cells, and neutrophils. UTC/ATC is Pax-8 positive and has p53 mutations [11].

Due to the rarity of primary synovial sarcoma of the thyroid gland, there are no established treatment guidelines. Management concepts for sarcomas are adapted to this condition. Complete surgical resection remains the primary treatment for localized disease [12]. Surgeons must carefully avoid capsular rupture and tumor spillage during surgery. The role of radiation therapy is unclear, but some studies have reported improvements in local control and disease-free survival [13]. Synovial sarcoma is generally considered chemosensitive, with responses observed particularly to ifosfamide-based regimens [12,14]. However, the use of chemotherapy for localized disease remains controversial due to conflicting retrospective data [6,10].

In this case, management was challenging due to concomitantabscess formation. The attempted drainage surgery was technically demanding, as the infection had converted the locally advanced tumor into a large inflammatory mass with no clear planes. The rarity of this diagnosis can lead to difficulties in establishing the correct diagnosis and determining the appropriate treatment approach.

The 5-year survival rate is consistent with previous studies. Post-operative adjuvant chemotherapy should be considered to reduce recurrence and improve survival, where feasible [9]. Metastatic spread may occur years after the initial diagnosis [2,12]. Centralized, individualized treatment in a facility with expertise in both sarcomas and head and neck tumors is important to optimize the chance of cure and minimize treatment-related complications.

CONCLUSION

Surgeons should be aware of rapidly growing, cystic-solid thyroid masses that cause neck compression symptoms. Initial surgery with adequate margins is key for locally controlled primary thyroid sarcoma. However, prognosis is often poor due to high local recurrence rates, despite aggressive adjuvant therapy. Palliative or debulking surgery may be an option for some patients.

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