A Rare Case of Hemangioma in the Thyroid: Clinical Presentation and Management Insights

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Thyroid hemangiomas are rare. However, preoperative investigations should be considered because of the potential risk of significant intraoperative hemorrhage. Here, we report a case of thyroid hemangioma in a 58-year-old woman. Physical examination revealed a large, hard, non-tender mass measuring approximately 60 mm in the anterior neck. Laboratory tests indicated hypothyroidism (triiodothyronine: 2.32 pg/mL, thyroxine: 0.62 ng/dL, thyroid-stimulating hormone: 22.4 μIU/mL) and markedly elevated anti-thyroglobulin (1,357 IU/mL) and anti-TPO (405 IU/mL) antibodies. Ultrasonography revealed a well-defined, heterogeneous internal mass measuring 60 mm within the right lobe of the thyroid gland, with a hyperechoic image bordering the mass. Contrast-enhanced CT revealed a 44 mm mass in the right lobe with calcification at the rim and gradual contrast enhancement towards the interior. Fine-needle aspiration cytology (FNAC) yielded mostly blood cells, with no evident epithelial components. A right thyroid lobectomy was performed, and post-operative histopathology confirmed a diagnosis of hemangioma. This case highlights the importance of considering thyroid hemangiomas in the differential diagnosis of thyroid masses due to the risk of intraoperative hemorrhage. It also emphasizes the role of various imaging modalities such as ultrasonography and CT in the preoperative evaluation of such cases.

Key words: thyroid hemangioma, thyroid glands, hemangioma

INTRODUCTION

Hemangiomas are benign vascular neoplasms that occur in various organs throughout the body, with a predilection for the body surface, including the skin and soft tissues; however, hemangiomas in the thyroid gland are very rare. The preoperative evaluation of thyroid hemangiomas is important because of the potential risk of significant intraoperative hemorrhage. We report a case of thyroid hemangioma in a 58-year-old woman and discuss the findings of ultrasonography, computed tomography (CT), other imaging modalities, and fine-needle aspiration cytology (FNAC).

CASE REPORT

A 58-year-old Filipino female with a history of Sjögren's syndrome, antiphospholipid antibody syndrome, and chronic renal failure first noticed a painless neck mass around 2003. In 2022, she was referred to the Department of Nephrology because of hypokalemia associated with Sjögren's syndrome and renal tubular acidosis. During a thorough examination, a right-lobe thyroid mass was noted, and she presented to our department in October 2022.

Physical examination revealed a large, hard, non-tender mass measuring approximately 60 mm in the anterior neck, without cervical lymphadenopathy or vocal cord paralysis. Laboratory tests indicated hypothyroidism (triiodothyronine: 2.32 pg/mL, thyroxine: 0.62 ng/dL, thyroid-stimulating hormone: 22.4 μIU/mL). The thyroglobulin concentration was 2.1 ng/mL (reference ≤ 30 ng/mL), while anti-thyroglobulin and anti-TPO antibodies were markedly elevated at 1,357 IU/mL and 405 IU/mL, respectively.

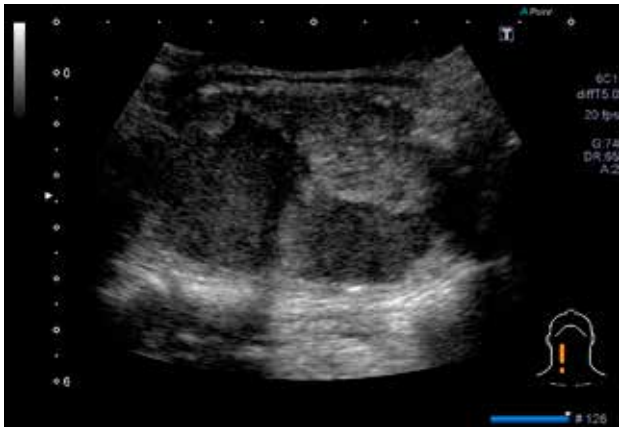

Fig. 1 Ultrasonographic findings
A well-demarcated heterogeneous mass measuring 67 × 43 × 52 mm is observed. Hyperechoic areas encircling the mass are also observed.

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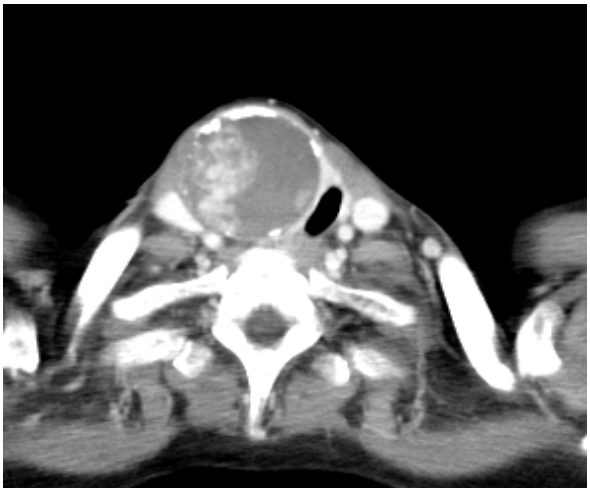

Fig. 2 Axial contrast-enhanced computed tomography
Computed tomography revealed a mass in the right lobe of the thyroid gland with rim calcification. Gradual contrast enhancement is observed from the periphery to the interior of the mass.

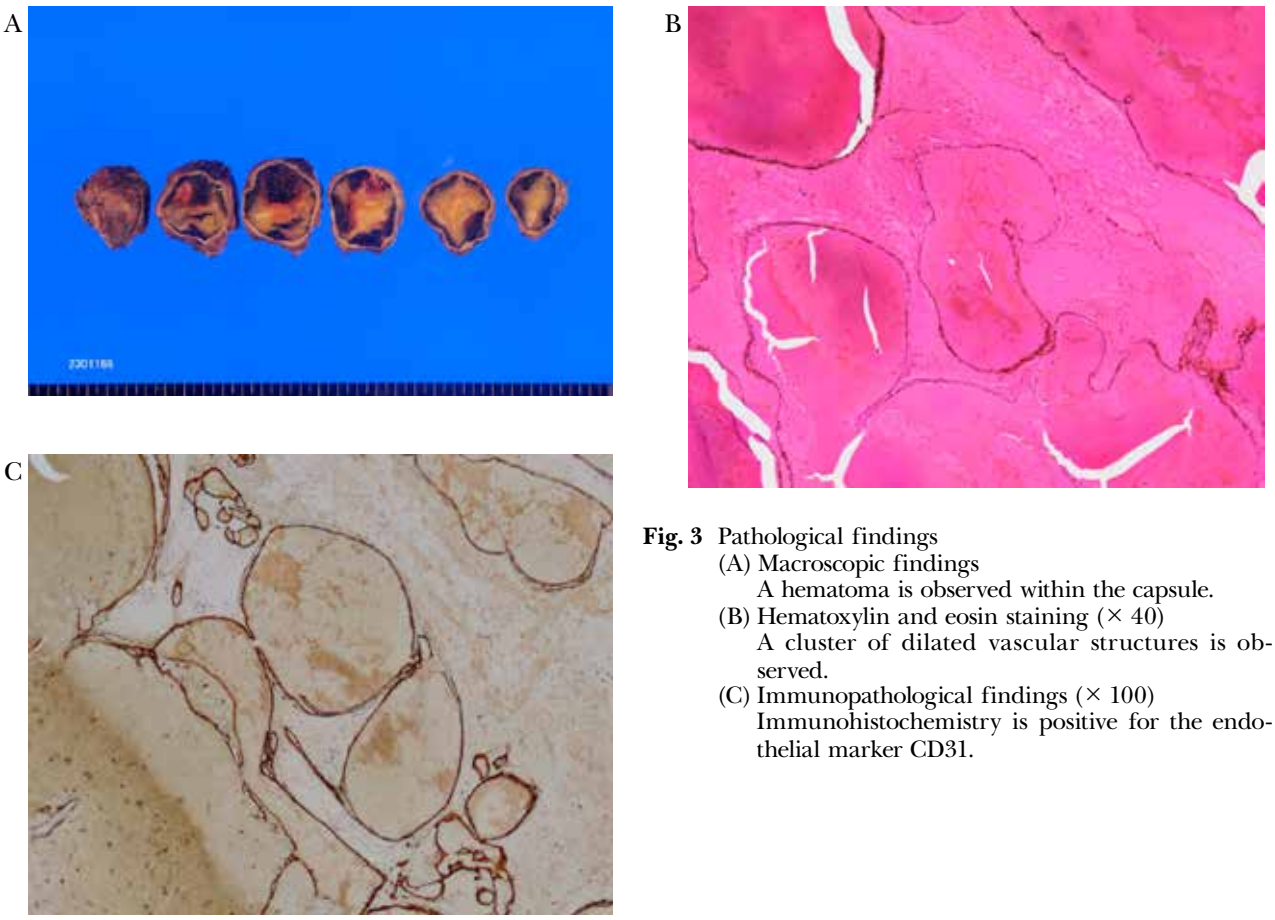

Fig. 3 Pathological findings
(A) Macroscopic findings
A hematoma is observed within the capsule.
(B) Hematoxylin and eosin staining (× 40)
A cluster of dilated vascular structures is observed.
(C) Immunopathological findings (× 100)
Immunohistochemistry is positive for the endothelial marker CD31.

Ultrasonography revealed a well-defined, heterogeneous internal mass measuring 60 mm in the right lobe of the thyroid gland. A hyperechoic image bordering the mass was observed. Contrast-enhanced CT revealed a 44 mm mass in the right lobe of the thyroid gland. The mass had calcification at the rim and gradual contrast from the rim to the interior. FNAC yielded mostly blood cells, with no evident epithelial components.

Based on echographic findings, a benign tumor was suspected; however, because of its large size, surgery was indicated. A right thyroid lobectomy was performed through a collar incision. The tumor showed no obvious adhesion to the surrounding tissue. The right recurrent laryngeal nerve was preserved, and intraoperative blood loss was 60 mL.

On histopathological examination, the lesion appeared hematoma-like and was covered by a fibrous capsule. Microscopically, it had a hematoma-like appearance with clusters of dilated vascular-like struc-

tures. Immunostaining confirmed positivity for CD31 and CD34, consistent with a cavernous hemangioma, and negativity for AE1/3 and D2−40, with no malignant features.

Six days after surgery, the patient was discharged without vocal cord paralysis or other complications. Postoperative pathology confirmed the diagnosis of a hemangioma. Two years have passed since then and the patient has been under outpatient observation without any recurrence.

DISCUSSION

Hemangiomas are benign tumors that develop throughout the body. Most of them occur congenitally in the skin and soft tissues, and 60% of them are said to occur in the head and neck region [1]. They are extremely rare in the thyroid gland, with only 39 case reports published from 1975 to 2024. This is the 40th reported case. In particular, they are reported to occur during FNAC, which is indispensable in the treatment of thyroid tumors, and they are thought to be caused by disruption of the vessel wall because of abnormal angioblastic mesenchyma [1]. In the present case, the patient visited another hospital approximately 20 years ago for a thyroid mass, and it is possible that puncture aspiration cytology was performed there; thus, the possibility of a secondary diagnosis cannot be ruled out.

The conditions conventionally lumped together under the term "hemangioma" include tumors and vascular malformations that are conventionally treated as tumors. Vascular anomalies are classified according to the International Society for the Study of Vascular Anomalies (ISSVA) guidelines published in 1996 and revised in 2018. Under this classification, "vascular anomalies" are broadly classified into vascular tumors with endothelial cell proliferation and vascular malformations that do not show cell proliferation [3]. The majority of cases conventionally reported as thyroid hemangiomas are cavernous or capillary hemangiomas and are classified as venous malformations according to the current ISSVA classification.

Thyroid hemangiomas often present with painless neck swelling as the only chief complaint. Other symptoms may include hoarseness, dysphagia, and respiratory distress due to tumor growth [1]. There are various reports on the speed of enlargement, with some cases reporting rapid enlargement within a few hours [1], while others have reported slow enlargement over a period of 20 years [4].

The imaging findings of thyroid hemangiomas are as follows. Ultrasonography typically shows a slightly heterogeneous, well-defined mass centered in the hypoechoic region, with abundant blood flow on Doppler images and high internal echoes when a hemorrhage is present. De-bin Yang *et al.* [5] performed contrast-enhanced ultrasonography and reported that it showed a slow-in and slow-contrast effect over normal thyroid tissue, which is useful for diagnosis. If there is no internal hemorrhage on non-contrast CT, the mass is uniformly hypoechoic. Contrast-enhanced CT reveals a slow-contrast effect; however, the contrast effect is weaker than that in the thyroid parenchyma. Some cases are accompanied by internal petechial-to-linear calcification and venoliths have been suspected in some cases. Magnetic resonance imaging scans have been reported to be useful for the diagnosis of internally heterogeneous masses with low-to-equal signal intensity on T1-weighted images and high signal intensity on T2-weighted images, with a contrast effect and flow voids in the surrounding and interior areas. Single-photon emission computed tomography and Tc-99m erythrocyte blood-pool imaging are useful for diagnosis, but they are difficult to perform in all patients owing to their degree of invasiveness and cost [6]. In the present case, ultrasonography showed a mixed mass, and contrast-enhanced CT revealed calcification within the mass, which was relatively homogeneous with a slow-contrast effect, an imaging finding that was suspicious for a hemangioma.

During puncture aspiration cytology for thyroid hemangiomas, only blood cells are often collected, making the diagnosis difficult [5]. In this case, too, puncture aspiration cytology showed only blood cell components.

Pathological examination is essential for a definitive diagnosis [7]. Microscopic evaluation of a hemangioma usually revealed a clear borderline proliferation of dilated, thin-walled vessels of various sizes lined by a single layer of flat endothelium. Nuclear atypia and mitosis are not observed. These vascular lumens are separated by fibrous septa-containing small vessels. Thrombosis, calcification, and interstitial hyalinization were observed. Markers indicative of vascular endothelial cells, such as CD31, CD34, ERG, FLI1, and Factor VIII, are positive [2]. The pathology results in this case were positive for the vascular endothelial cell markers CD31 and CD34, but negative for the epithelial marker AE1/3 and the lymphatic endothelial marker D2−40. The patient was diagnosed with a cavernous hemangioma, which was classified as a venous malformation rather than a tumor, similar to many previously reported thyroid hemangiomas.

Surgery is often the treatment of choice. In cases where a thyroid hemangioma is suspected, surgical and diagnostic treatments are recommended. The prognosis is good with no recurrence in most postoperative cases [8]. Hemangiomas, however, are blood-rich masses and they carry the risk of massive intraoperative bleeding [9]. Although preoperative diagnosis is difficult in many cases, we consider it important to suspect the possibility of a hemangioma before surgery. In retrospect, the imaging findings in this case were typical, and because only blood cell components were obtained by puncture aspiration cytology, the possibility of a thyroid hemangioma should have been considered preoperatively.

The possibility of hemangioma may be considered in the future when only hematologic components are found on puncture aspiration cytology, and imaging findings, such as ultrasonography and CT, may be reviewed to suspect thyroid hemangioma before surgery and prepare for the risk of intraoperative hemorrhage.

Thyroid hemangiomas are rare. When imaging suggests a tumor with abundant blood flow and FNAC shows only blood cell components, thyroid hemangioma should be included in the differential diagnosis. Preoperative planning should consider the potential risks of significant intraoperative bleeding.

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