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Thyroid fibromatosis: A case report

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Objectives. Fibromatosis is a rare tumor with a prevalence of 2–4 per million per year. Its occurrence in the thyroid is very rare and to the best of our knowledge only five cases have been reported in the literature.

Subject and Results. In this case report, we present a thirty-seven-year-old man without any significant past medical history, who presented with an anterior neck swelling and occasional cough for four months before admission. Sonographic-guided fine needle aspiration cytology studies reported a follicular neoplasm. Microscopic and immunohistochemical evaluation of the thyroidectomy specimen showed thyroid fibromatosis. The patient had no symptoms after surgery and no signs of recurrence after 2 years of follow-up.

Key words: fibromatosis, thyroid, neoplasm

Fibromatosis encompasses a broad group of benign fibroblastic lesions defined based on the presence of well-differentiated fibroblastic and myofibroblastic proliferation without malignant changes and minimal mitotic activity. Even in clinical scenarios with aggressive behavior and local recurrence, metastasis occurs rarely (Goldblum et al. 2013; Goldblum et al. 2017). Most cases of fibromatosis occur sporadically due to gene mutation in a gene called Beta-catenin (Mullen et al. 2013; van Broekhoven et al. 2015). They have a prevalence of 2–4/million per year (Reitamo et al. 1982; Shields et al. 2001; Fallen et al. 2006; Nieuwenhuis et al. 2011). Considering the susceptible tissues in the neck area, about 10% of fibromatoses occur in this area and their treatment is mainly based on complete surgical excision (Samsi et al. 1992; Rodriguez-Bigas et al. 1994; Seper et al. 2005). In this case report, we present a case of thyroid fibromatosis in a young man.

Subject and Results

A thirty-seven-year-old man without significant past medical history and with suspected hypothyroidism in the family, presented with swelling of the anterior neck and an occasional cough from four months before admission. The patient had stable vital signs and no local pressure or systemic symptoms. The right thyroid lobe was irregular and nodular. Ultrasound study showed normal parenchymal echogenicity and no solid mass in the left lobe. Ultrasound study of the right lobe showed a hypoechoic mass of 56×43 mm, and in the vicinity of it, another hypoechoic nodule, measuring 38×28 mm, with a compressive effect on adjacent vessels. No evidence of normal tissue was seen on the right lobe of the thyroid. No adenopathy was reported in bilateral neck sonography. Ultrasound-guided FNA of the right hypoechoic mass gave the diagnosis of a follicular neoplasm. The patient underwent total right thyroidectomy with near-total left thyroidectomy and the specimen was sent for pathological examinations. Macroscopically, the right thyroid lobe measured 9×7×4 cm and weighed 16 g. The cut surface showed a well-circumscribed, creamy, and solid mass, measuring 8×3.5×2 cm. Left thyroid lobe measured 4×2×1 cm and weighted 5 g, without any discrete mass.

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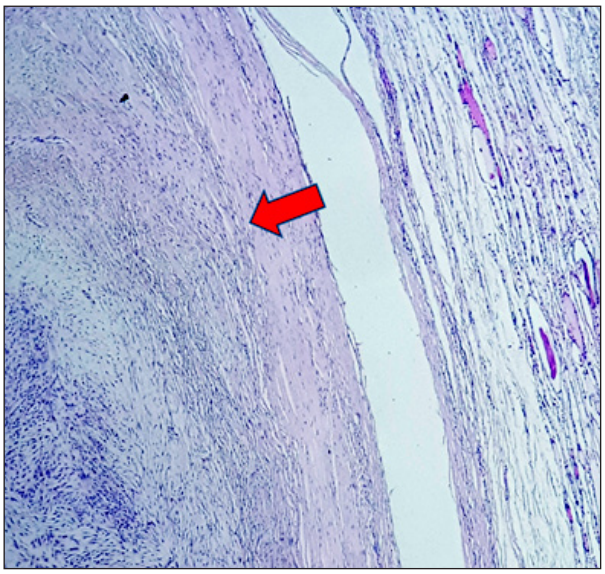

Figure 1. Microscopic examination of the right lobe revealed thyroid parenchyma with a well-circumscribed, patternless mass with non-atypical spindle cells with collagen bundles and staghorn vessels (arrow: non atypical spindle mass).

Microscopic examination of the right lobe revealed thyroid parenchyma with a well-circumscribed, patternless mass with non-atypical spindle cells with collagen bundles and staghorn vessels (Figure 1). There was no evidence of papillary thyroid carcinoma. According to the microscopic findings, a mesenchymal tumor was seen in the right lobe. There was no significant pathologic finding in the left lobe.

Immunohistochemical (IHC) studies were performed to confirm the diagnosis and to rule out other differential diagnoses, such as solitary fibrous tumor, nodular fasciitis, and papillary carcinoma with fibromatosis/fasciitis-like stroma (PTC_FLC) (Figure 2A–D). The IHC studies for beta-catenin, actin, desmin, CD34, TTF-1, Tg, S100, and CK was ordered. Beta-catenin and actin staining were positive and desmin, CD34, TTF-1, Tg, S100, and CK were negative. These findings were consistent with the diagnosis of thyroid fibromatosis. After surgery, the patient had no recurrence of tumor for two years.

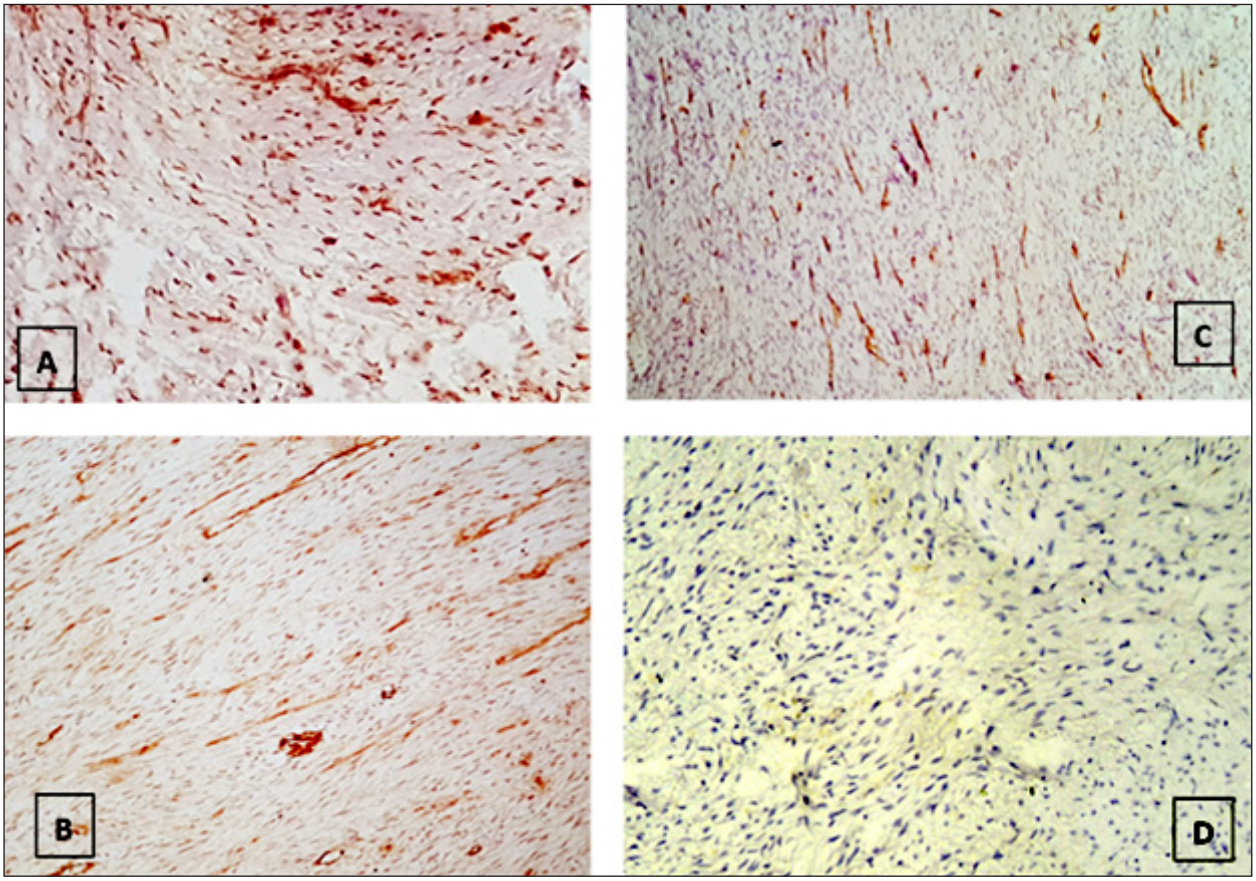

Figure 2. Immunohistochemical staining of **(A)** beta-catenin (positive), **(B)** actin (positive), **(C)** CD34 (negative), and **(D)** thyroglobulin (negative).

Discussion

Due to the presence of muscular tissue and fascia, fibromatosis can happen in the neck (Conley et al. 1966). Whenever a spindle cell lesion, suspicious of thyroid fibromatosis is present, low-grade fibrosarcoma and fibrous histiocytoma should be considered as the differential diagnoses. Large areas of hyalinization, inflammatory infiltrate, and scarce mitosis implies the benign nature of the lesion (Samsi et al. 1992). Differential diagnoses of progressive and severe thyroid enlargement consist of uncommon conditions, such as anaplastic or poorly-differentiated thyroid carcinoma, primary thyroid lymphoma, or Riedel's thyroiditis, and thyroid fibromatosis is one of the rare diagnoses (Simoes-Pereira et al. 2016). Another important differential diagnosis in spindle cell lesions of thyroid is papillary carcinoma with fibromatosis/fasciitis-like stroma (PTC_FLC) (Simoes-Pereira et al. 2016), which is a biphasic neoplasm and occasionally, prominent stromal reaction may hide the epithelial neoplastic nature and especially in small biopsy specimens be misdiagnosed as nodular fasciitis, fibromatosis, or other proliferative stromal conditions. Another pitfall is nuclear beta-catenin immunohistochemical staining in the mesenchymal component of PTC_FLC, which is characteristic of fibromatosis and some authorities call it papillary carcinoma with desmoid-type fibromatosis (Goldblum et al. 2017).

To the best of our knowledge, five thyroid fibromatosis cases have been reported so far (Samsi et al. 1992; Sinha et al. 1998; Simoes-Pereira et al. 2016). All patients were female, whereas in our study the gender of the patient was male. In three studies, the age of the patients was between 60–70 years, and in the studies of Sinha et al. (1998) and Schwarzlmuller and Hofstadter (1978), the patient age was 26 and 34 years, respectively. In our study, the patient was 37 years old.

Like other organs, fibromatosis of the head and neck is usually associated with an invasion of the surrounding structures. In the three previous case studies of thyroid fibromatosis, this mass had an invasion of the surrounding structures (muscles, vessels). However, in the studies of Simoes-Pereira et al. (2016), Schwarzlmuller and Hofstadter (1978) and this case, there was only a compressive effect of the mass on adjacent vessels.

In this case, the mass was located in the right lobe, and the patient complained of neck swelling and occasional cough. In the studies of Sinha et al. (1998) and Samsi et al. (1992), thyroid swelling was the main sign. There is no recommendation in relevant articles on how to follow up the patients with thyroid fibromatosis. In this case, the patient was followed up for two years without symptoms and no signs of recurrence. The longest follow-up period reported to date (11 years) was a case reported by Simoes-Pereira et al. (2016) and no signs of recurrence was reported. In a study by Samsi et al. (1992), a patient with thyroid fibromatosis was evaluated for one year postoperatively; which showed no sign of recurrence. However, there are reports of local recurrence even 4 years after thyroidectomy of thyroid fibromatosis. Therefore, the annual assessment of these patients is recommended. Because of many differential diagnoses of spindle cell proliferation in the thyroid, we should consider fibromatosis as one of the differential diagnoses and confirm it with a complimentary assessment.

References

Conley J, Healey WV, Stout AP. Fibromatosis of the head and neck. Am J Surg 112, 609–614, 1966.

Fallen T, Wilson M, Morlan B, Lindor NM. Desmoid tumors – a characterization of patients seen at Mayo Clinic 1976-1999. Fam Cancer 5, 191–194, 2006.

Goldblum JR, Weiss SW, Folpe AL. Enzinger and Weiss's Soft Tissue Tumors E-Book: Expert Consult: Online and Print, Elsevier Health Sciences, 2013.

Goldblum JR, Lamps LW, McKenney JK, Myers JL. Rosai and Ackerman's Surgical Pathology E-Book, Elsevier Health Sciences, 2017.

Mullen JT, DeLaney TF, Rosenberg AE, Le L, Iafrate AJ, Kobayashi W, Szymonifka J, Yeap BY, Chen YL, Harmon DC, Choy E, Yoon SS, Raskin KA, Hornicek FJ, Nielsen GP. β-Catenin mutation status and outcomes in sporadic desmoid tumors. Oncologist 18, 1043–1049, 2013.

Nieuwenhuis MH, Casparie M, Mathus-Vliegen LM, Dekkers OM, Hogendoorn PC, Vasen HF. A nation-wide study comparing sporadic and familial adenomatous polyposis-related desmoid-type fibromatoses. Int J Cancer 129, 256–261, 2011.

Reitamo JJ, Hayry P, Nykyri E, Saxen E. The desmoid tumor. I.: incidence, sex-, age-and anatomical distribution in the Finnish population. Am J Clin Pathol 77, 665–673, 1982.

Rodriguez-Bigas MA, Mahoney MC, Karakousis CP, Petrelli NJ. Desmoid tumors in patients with familial adenomatous polyposis. Cancer 74, 1270–1274, 1994.
Samsi A, Shah H, Vaidya A, Pai P, Deshmane U, Sane S. Fibromatosis of thyroid gland (a case report). J Postgrad Med 38, 36–37, 1992.
Seper L, Burger H, Vormoor J, Joos U, Kleinheinz J. Agressive fibromatosis involving the mandible - case report and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 99, 30–38, 2005.
Schwarzlmuller B, Hofstadter F. [Fibromatosis of the thyroid gland region. An electron-microscopic and enzyme-histochemical study (author's transl)]. Virchows Arch A Pathol Anat Histol 377, 145–155, 1978.
Shields C. Winter Dc, Kirwan WO, Redmond HP. Desmoid tumours. Eur J Surg Oncol 27, 701–706, 2001.
Simoes-Pereira J, Cabrera RA, Leite V. A case of thyroid fibromatosis, a rare lesion of this gland. Endocrinol Diabetes Metab Case Rep 2016, pii: 16–0019, 2016.
Sinha A, Rao A, Sinha A, Arora R. Fibromatosis of thyroid gland-a case report. Indian J Otolaryngol Head Neck Surg 50, 385–386, 1998.
van Broekhoven DL, Verhoef C, Grunhagen DJ, van Gorp JM, Den Bakker MA, Hinrichs JW, de Voijs CM, van Dalen T. Prognostic value of CTNNB1 gene mutation in primary sporadic aggressive fibromatosis. Ann Surg Oncol 22, 1464–1470, 2015.