Case Report

Autoimmune thyroiditis a fibrous variant of Hashimoto’s thyroiditis: A rare case

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ABSTRACT

Hashimoto thyroiditis (HT) is an autoimmune disorder characterized by inadequate thyroid hormone production. A fibrous variant is one of the rarest entities of Hashimoto’s thyroiditis disease. A 42-year-old female patient presented to our service with neck swelling associated with difficulty swallowing; she was discovered to have an enlarged thyroid gland with mass effect. She underwent an ultrasound and fine-needle aspiration (FNA), which was consistent with Hashimoto’s thyroiditis -Bethesda category II-. Due to compressive symptoms, we proceeded to total thyroidectomy. The final histopathology revealed numerous polymorphic lymphoid cells, plasma cells, follicular cells, and scattered Hürthle cells, characteristic of fibrous variants. The surgery was complicated with voice hoarseness and hypocalcemia, which was managed successfully with corticosteroids and calcium supplements. The mainline treatment of HT is medical, but surgical intervention can be considered in some cases. A multidisciplinary approach is needed for successful management. Continuous patient monitoring post-operatively is vital to detect and intervene with early surgical complications.

1. Introduction

Hashimoto’s thyroiditis (HT) is named after the Japanese physician Hakaru Hashimoto (1912) who was the first to describe this disease after examining the thyroid specimens of four middle-aged women who had a thyroidectomy due to compressive symptoms. Hashimoto’s thyroiditis was previously known as chronic lymphoid thyroiditis or autoimmune thyroid disorder (AITD), which predominantly affects females (Female to male ratio is 10:1). HT was rarely diagnosed until the late 1950s, but nowadays, it is the most common autoimmune and endocrine disorder associated with hypothyroidism. Patients with HT commonly present with a painless progressive diffuse thyroid swelling and with mass effect on the trachea and esophagus. A fibrous variant is accounted for 10% of the cases of Hashimoto’s thyroiditis [1–3]. Up to our knowledge, this is the 3rd case of fibrous variant reported worldwide. This case report has been reported in line with the SCARE Criteria [4].

2. Case presentation

A 42-years-old married, non-smoker female office worker, known to have hypothyroidism on Levo-Thyroxin, referred to our clinic with a complaint of gradual progressive neck swelling affecting her ability to swallow solid food. She denied any history of allergic reactions, autoimmune diseases, or Malignancy. Physical examination revealed a large

Abbreviations: HT, Hashimoto thyroiditis; FNA, Fine Needle Aspiration; AITD, Autoimmune Thyroid Disorder.

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anterior neck swelling which was stony hard in consistency, moving vertically with swallowing and stationary with tongue protrusion with multiple enlarged cervical lymph nodes. Her laboratory tests showed low TSH, elevated levels of free T4, anti thyroglobulin, and anti thyroperoxidase antibodies. Our differential diagnosis included Malignancy as the mass was stony hard in consistency, and lymphadenopathy was suspected due to the presence of multiple enlarged neck lymph nodes. A neck ultrasound was ordered, revealing an enlarged heterogenous thyroid where the right lobe measures about (3.9 x 2.7 x 6.5 cm) while the left lobe measures about (3.4 x 1.8 x 5.2 cm) with the mild shifting of the trachea to the left side. Furthermore, calcifications were identified and accessed with a 23 G needle. Three passes were taken from the solid calcified part and sent to the lab on dry slide smearing and alcohol container (Fig. 1). The biopsy took dispersed heterogeneous lymphoid cells (small lymphocytes, centrocytes, centroblasts, and plasma cells), rare tangible-body macrophages, along with clusters of cells with an extensive crushing artifact, occasional identifiable Hürthle cells in small groups, and rare multinucleated giant cells are seen. The background of the taken biopsy shows blood and no colloid. The final result of FNA was consistent with Hashimoto’s thyroiditis Bethesda category II (Fig. 2).

Besides the U/S, intravenous contrast injected computed tomography scan of the neck was done, showing a bilateral diffusely enlarged thyroid gland with a mild shifting of the trachea to the left side. Both thyroid lobes appeared heterogeneous enhanced multinodular lobulated masses extending to the isthmus with no calcifications. Multiple enlarged cervical lymph nodes were most of them sub centimetric with preserved fatty hilum (Fig. 3). After careful evaluation of CT and histopathology, we were able to rule out Malignancy and lymphadenopathy.

After all available treatment modalities were discussed with the patient, we elected to do a total thyroidectomy after a full pre-operative assessment, including ENT and endocrine evaluation. As the patient was morbidly obese, she was given anticoagulant prophylaxis.

The operation was carried out in a district general hospital by an available surgery consultant with experience of more than 30 years. Under general anesthesia, the patient was in a supine position with neck hyperextension using sandbags; Total thyroidectomy was performed using electrocautery and 3-0 vicryle tie. The Thyroid was stony hard with severe adhesions to adjacent structures, and the superior and recurrent laryngeal nerves were identified and preserved.

Gross histopathology showed both lobes with solid cut surface and a yellow-white nodular appearance. Their capsule had been stained green in color, and both contained parathyroid glands. There was no lymph node seen in the resected specimen.

Microscopically, the sections from both lobes were similar. They show architectural destruction with the disruption of the normal lobular pattern. Thyroid parenchyma was characterized by extensive fibrosis with atrophy of thyroid follicles; prominent lymphoplasmacytic inflammatory infiltrates distributed within and around the lobule with the formation of germinal centers mixed with oncocytes (Fig. 4). Prominent squamous metaplasia of the follicular cells was seen. The fibrosis was limited to the Thyroid and did not extend beyond the thyroid capsule. There was no evidence of obliterate phlebitis or vasculitis. During extubation, the patient developed shortness of breath which was treated with 3L oxygen and urgent intravenous Hydrocortisone. She was also noted to be in respiratory distress and having flaring nostrils. ENT evaluation revealed edematous arytenoids and normal freely mobile vocal cords. After stabilization, she was transferred to the surgical ward. On day one post total thyroidectomy, the patient was still complaining of shortness of breath and voice hoarseness. On examination, her chest had equal bilateral breathing sounds with inspiratory stridor more heard on the left side with scattered end-expiratory wheezing. This was managed using Symbicort 1 puff twice daily, in addition to Salbutamol nebulization of 2.5 mg every 8 hours when needed. Calcium and parathyroid levels were within the normal range.

The patient was discharged in stable condition. Upon her discharge, she was on a tapering scale of corticosteroid, Calcium gluconate 600 mg twice daily orally, and Vitamin D 50000 IU twice weekly. She is monitored through OPD routinely up to this day. The patient had developed post-total thyroidectomy hypoparathyroidism. She had been treated with Calcium carbonate 1200 mg three times a day, Alphacalcidol 0.5 mcg once daily, and Levothyroxine 250 mcg once daily, which was tolerated well by the patient. Her voice hoarseness improved markedly, and she is following up regularly with endocrine in addition to general surgery.

3. Discussion

Katz and Vickery et al., 1974 reported fifty-six patients with fibrous variants of HT subtype that represented 12.5% of all pathologically diagnosed Hashimoto thyroiditis. They described the typical features of this lesion characterized by a marked fibrous replacement of one-third to the majority of the parenchyma and changes of Hashimoto’s disease in the remaining tissues, the fibrous process is within the thyroid capsule, and there is a dense hyaline form of connective tissues in contrast to active proliferative fibrosis seen in Riedel’s thyroiditis. The epidemiology of this disease is not known; it commonly affects middle-aged females. A fibrous variant is one of the rarest entities of HT, accounting for only 10% of all of its cases. The diagnosis of such an entity is challenging and requires a multidisciplinary approach. It is usually simulated as malignant neoplasm of the Thyroid due to the presence of islands of metaplastic epithelium in the fibrotic tissues simulating foci of carcinoma cells. Clinical manifestations of the patients with HT vary from progressive, painless swelling to shortness of breath and dysphagia due to its mass effect on nearby structures. On examination of the neck, the thyroid gland is usually stony hard, as reported in the literature [5-7].

Diagnosis is usually initiated by laboratory investigations such as a Thyroid function test, Anti thyroglobulin, and anti thyroperoxidase antibodies, followed by Thyroid ultrasound-guided fine-needle aspiration. CT scan of the neck and chest is a very useful diagnostic imaging modality, especially in the presence of compressive symptoms and retrosternal gland extension [8,9].

Hence the fibrotic changes are the main characteristic feature and

Fig. 1. Ultrasound of right lobe of thyroid gland during FNA: Under complete aseptic technique and US guidance, Right complex thyroid nodule with microcalcifications was identified and accessed with 23 G needle. Three passes were taken from the solid calcified part and sent for histopathology.
the keystone in most Thyroid diseases, The routine cytology has a minimal role in differentiating between the different forms of HT, and the definitive diagnosis of the fibrous variant is based on the histopathological features, as diffuse lymphatic infiltration, well-developed germinal center, parenchymal atrophy, Hürthle cell with eosinophilic changes in thyroid follicles and characteristically extensive fibrosis of the thyroid parenchyma within the thyroid capsule and dense hyaline form of connective tissue to the surrounding structure. Most of those features were manifested in our case, and we could confirm the diagnosis based on those [10–12]. Multidisciplinary approach, including endocrine physicians, surgeons, and histopathologists, is the standard practice. The differential of Hashimoto’s thyroiditis includes malignant neoplasms of the Thyroid, medullary carcinoma, diffuse sclerosing variant of papillary cell carcinoma, a variant of anaplastic carcinoma, large malignant lymphoma, sarcoma, and sclerosing Hodgkin lymphoma, with features of fibrosis in the gland parenchyma. In our case, Malignancy was one of the differential diagnoses, but it was rolled out after the final histopathology. In addition to Malignancy, other benign conditions, such as adenoma (paraganglioma), solitary fibrous tumor, myxedema, and Riedel disease, should be excluded [13–17]. Fibrous variant and Riedel disease of the thyroid gland had many indistinguishable clinical and cytological features, but we can differentiate them using histopathological examination, where the earlier is characterized by extensive fibrosis, which affects the majority of thyroid parenchyma without any extension beyond the thyroid capsule, and the dense hyaline connective tissue to the surrounding structures unlike the latter. In 1957 Beahrs et al. described histological criteria to establish a differential diagnosis between Hashimoto’s thyroiditis and Riedel’s thyroiditis, which was validated by Schwaegerle and Meijer et al. [18–20] The mainstay of treatment of fibrous variant is medical using levothyroxine; surgery can be offered in cases of compressive symptoms and for cosmetic reasons. In addition, and due to the extensive fibrotic nature of the disease, surgeons may face difficulties during the procedure, which may increase the risk of postoperative complications. In our case report, the patient developed postoperative hypocalcemia. G Iannaci et al. (2013) and Tutuncu et al. (2000) recommended using steroids to decrease fibrosis and, by doing so, reduce postoperative complications [21]. There are limited published case reports discussing the fibrous

### Table: Bethesda classification of thyroid nodule fine needle aspiration.

| Diagnostic category | Description                                      | Risk of malignancy (%) |
|---------------------|--------------------------------------------------|------------------------|
| I                   | Non-diagnostic/unsatisfactory                   | 1–4                    |
| II                  | Benign                                           | 0–3                    |
| III                 | Atypia or follicular lesion of undetermined significance | 5–15            |
| IV                  | Follicular neoplasm or suspicious for follicular neoplasm | 15–30            |
| V                   | Suspicious for malignancy                       | 60–75                  |
| VI                  | Malignant                                        | 97–99                  |

Fig. 2. Bethesda classification of thyroid nodule fine needle aspiration.

Fig. 3. CT scan with IV contrast: A) Coronal view BI & II) Axial view C) Sagittal view of the right thyroid lobe Diffuse enlarged thyroid gland with mild shifting of the trachea to the left. Both lobes appear heterogeneously enhanced multinodular lobulated masses extending to the isthmus, no calcifications, Slightly the left lobe reach the thoracic inlet. Multiple Enlarged cervical lymph nodes, Left sub mandibular most of them subcentimetric with preserved fatty hilum.

Fig. 4. View of fibrous variant of Hashimoto thyroiditis: Lymphoid aggregations surrounded by dense hyaline fibrosis.
variant feature. All reviewed case reports stressed the importance of a multidisciplinary approach as the key to successful management of such cases; close observation post-operatively is vital in the detection and early intervention of any post-op complication.

4. Conclusion

In conclusion, this case report shows that the fibrous variant is one of the rarest entities of Hashimoto’s thyroiditis. Patients usually present with compressive symptoms and lymphadenopathy mimicking thyroid cancer. The gold standard diagnostic modality is histopathology. A multidisciplinary approach and careful pre-and postoperative evaluation are vital for successful management and excellent outcome.

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IRB approval.

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Author contribution

1. Almalali Maryam Isa Ahmed, Data analysis, interpretation and writing the paper.
2. Takrouni Arwa, Data collection revising the article.
3. Alsolami Sana, Data collection.
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8. Alyahyaei Abdulrahman, Data collection.
9. Ablasheed Fathi Ays A, Revising the article and supervision.

Declaration of competing interest

There is no conflict of interest.

Registration of research studies

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying imaging. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Appendix A. Supplementary data

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