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CASE REPORT OPEN ACCESS

CASE REPORT OF RIEDEL'S THYROIDITIS

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ABSTRACT

Background: There is no standard surgical management of Riedl's thyroditis because it is rare. Indications of surgery in Riedel's thyroiditis are obstructive symptoms, inability to diagnose the patient using non-invasive tools such as fine needle aspiration (FNA), and suspicions of malignancy. The extensive fibrosis that involves the surrounding structures prevents the surgeon from performinga complete surgery free of complications. ***Case report:*** A47-year-old Saudi male presented to our outpatient department as a referral from a secondary care center with a case of goiter with FNA, which showed thyroid lymphoma. The patient was hypothyroid and has a history of subdural hematoma. The review of his FNA in our center showed Hashimoto's thyroiditis. The patient's case was followed closely and he was given levothyroxine. After severalthyroid urltrasound, there was a newly appearing lesion which extended to the left internal jugular vein, and disturbed its flow. The repeated Ultrasound Guidance FNA failed to diagnose the extended lesion and the presence of a suspicious lymph node necessitate to operate on the patient. A frozen section of the suspicious lymph node confirmed the benign diagnosis, soa hemithyroidectomy preserving the left internal jugular vein was done. The histopathology result of the hemithyroidectomy showed Riedel's thyroiditis. The patient then was regularly followed up for two years with no relapse. ***Conclusion:*** Riedel's thyroiditis is a challenging disease both in diagnosis and in management. In our case report, thehemithyroidectomy was the diagnostic method. We recommend that the management of such disease be done in highly specialized centers with an expert endocrine surgeon and an expert pathologist. Follow up should be continued regardless of thyroid symptoms because of extra thyroid manifestations that can present late in the course of the disease.

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INTRODUCTION

Riedel's thyroiditis is a rare dense fibrosing inflammation of the thyroid, with invasion of adjacent structures, with extrathyroidal fibrosis such as retroperitoneal and mediastinal fibrosis (Hay, 1985). Because it is a rare condition, consensus in the treatment does not exist and most literature, since it was recognized in 1896 by Bernhard Riedel, consists of just case reports. Presentation in most reported cases resembles malignancy because of hard goiter, obstructive symptoms, and invasion of the fibrosis to adjacent structures which is why some patients have hypoparathyroidism (Beahrs *et al.,* 1957; Zimmermann-Belsing, 1994). Cellular examination of the fine needle aspiration, hence tissue biopsy (thyroidectomy, lobectomy), is needed for diagnosis in some cases. Mismanagement can occur when Riedel's thyroiditisis not considered. The operative incidence was 0.06%; 37 cases were diagnosed with a series of 57,000 thyroidectomies collected at Mayo Clinic between 1920 and 1984.

The overall incidence is 1.06 per 100,000 and women are affected more than men[1]. We are presenting a rare case of Riedel's with a diagnosis dilemma, which responded positively to hemithyroidectomy without medication; there was also no relapse for three years.

Case presentation**:** A 47-year-old man was referred to our hospital for further management of thyroid lymphoma. This was discovered accidentally by Computed Tomography (CT) during evaluation of a sudden left-sided weakness and severe headache three months before. The CT showed a right-sided subdural hematoma and thyroid enlargement. Retrospectively, he had experienced progressive voice change and a slightly asymmetrical neck during the last three years. He was treated for his subdural hematoma, and then investigated for his thyroid problem; a fine needle aspiration was done and the diagnosis was thyroid lymphoma. The patient's history in our hospital revealed that the patient had not complained of this

neck swelling during the last three years and had not sought medical advice. He had no difficulty in swallowing or breathing, but he did complain of weight gain, tiredness, constipation, and loss of interest in his usual life activities including his work. His examination showed a partially-fixed hard goiter, and his trachea was central and resonant retrosternal with no palpable lymph nodes in the neck. Investigations showed a slight increase in white blood cells (12.25X10^9/L). Other hematological investigations including coagulation profile were within normal limits; his bone profile was normal. His thyroid function test (TFT) showed a thyroid stimulating hormone level of 82mU/L and free T4 8 m Upmol. His antithyroglobuline was 3595 U/ml. Slides had been submitted to our pathologist for re-examination which confirmed Hashimoto's thyroiditis (Figure 1). The result of the ultrasound showed a heterogeneous enlarged thyroid. The patient at this stage was discharged on levothyroxine, and advised to follow up with an ultrasound in one year.

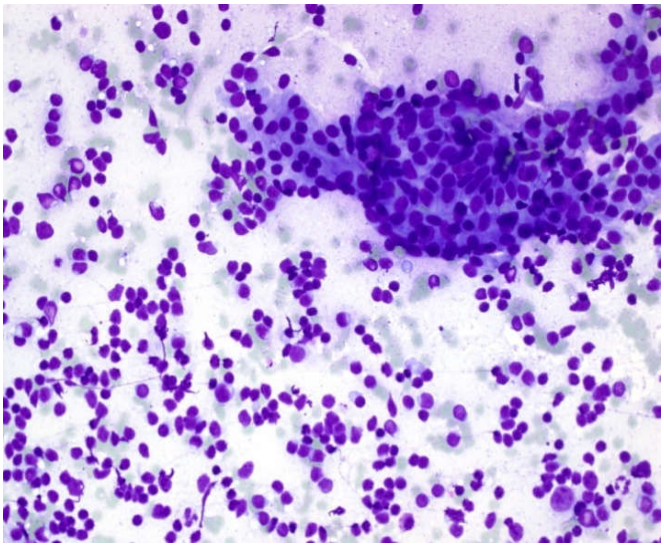

Figure 1.

Medical therapies such as corticosteroids (frequently used to limit the progression of fibrosis)* were not considered because the patient was asymptomatic locally.

One year later, the ultrasound showed multiple nodules in both lobes, and a speculated hypoechoicnodule in the left lobe invading the pre-thyroidal fat. FNA under Ultrasound Guidance was repeated 3 times; the result in every case was the same. There were no blood cells or thyroid follicles. At this time, the patient was asymptomatic (no obstructive symptoms). There were no significant clinical changes in his neck mass and his thyroid function test was normalized on 125 microgram levothyroxin. We decided to follow up with him up in a shorter interval (6 months). At this time, the ultrasound was repeated and showed an incremental increase in nodule size, which had invaded the left internal jugular vein and caused a disturbance of normal flow. There was a newly suspicious lymph node (Figure 2). At this stage, we repeated the FNA under Ultrasound Guidance and the result showed only blood cells in the nodule and reactive lymph node. A CT was done and showed no contrast enhancement of the thyroid gland, which made it difficult to evaluate. However, it was evident that there were no mass-occupying lesions. Unfortunately, the scan failed to fully evaluate the left internal jugular vein (Figure 3). Based on our investigation, the decision was made to operate on the patient.

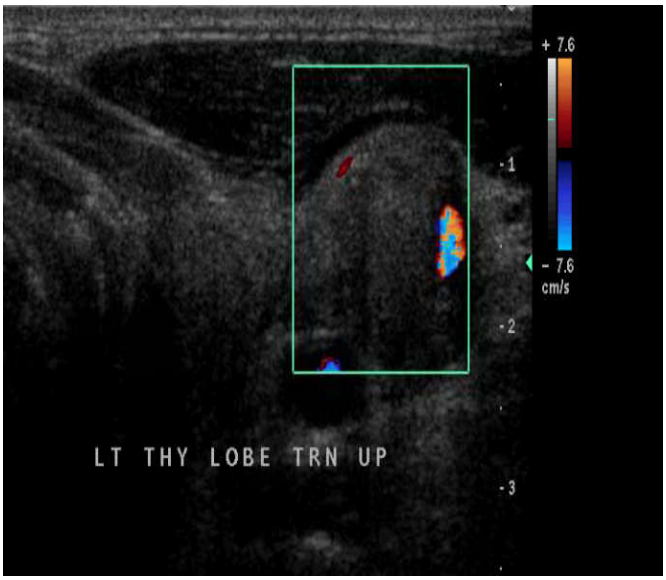

Figure 3. CT scan showed no contrast in left IJV

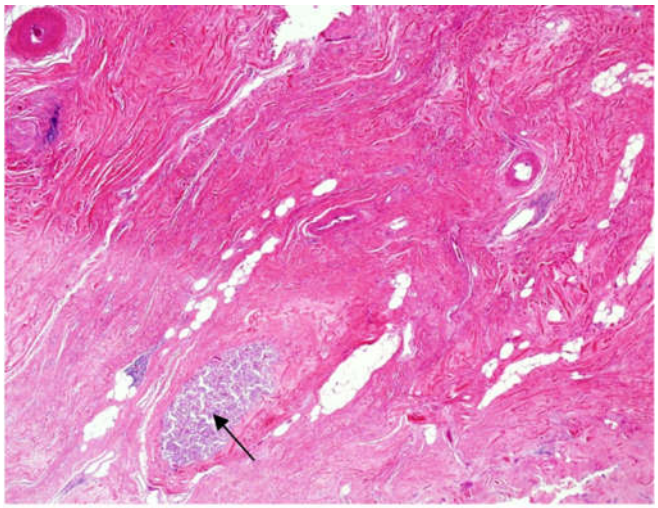

Figure 4. Central part with encased parathyroid (arrow)

Intraoperative findings included amalgamated avascular whitish tissue with no anatomical landmarks. There was a central lymph node which was sent to be frozen and the result came back as a reactive lymph node. An intraoperative decision was made to perform a left hemithyroidectomy as a diagnostic resection. Since the diagnosis was not clear, the left lobe was excised with difficulty in two pieces which have been labeled as left thyroid lobe (lateral) and midline mass (medial). No parathyroid tissue was identified in the specimen, but the recurrent laryngeal nerve was identified and preserved throughout the procedure. No dissection was done around the vein and the part of the nodule which encircled the vein was left without interference. The histopathology result showed the left thyroid lobe had a dense fibrosis and showed atrophy with residual foci of Hashimoto's thyroiditis without evidence of malignancy and an unremarkable parathyroid gland. It also showed midline mass fibro adipose tissue with chronic inflammation and dense fibrosis enclosing a parathyroid gland and, again, no evidence of malignancy (Figure 4). The histopathologist also commented that the residual foci of thyroiditis showed reactive germinal centers and prominent Hurthele cell metaplasia. Elsewhere there was sparsely cellular, keloid-like fibrosis and atrophic thyroid tissue with prominent squamous metaplasia and cystic change. These features together are compatible with Hashimoto's thyroiditis fibrous variant. On the other hand, extension of fibrosis to

extrathyroidal tissues as evidence of entrapment of parathyroidal gland (Figure 4) is unusual and raises the possibility of coexistence with Riedel's thyroiditis which is a very unusual occurence. Fibrous Hashimoto's and Riedel's thyroiditis was recently linked to the IgG-related systemic disease. Four years of patient follow up showed no recurrence, no obstructive symptoms, and no mass. He was euthyroid clinically and biochemically and has a nicely-healed scar as we see in Figure 5. Late in the follow up, he presented at the clinic with abdominal pain and an abdominal CT was requested; it showed a retroperitoneal fibrosis encasing the distal aorta and the iliac bifurcation showed tethering of the right ureter medially. There was perirenal stranding of the right kidney with mild dilatation of the right ureter. The patient was referred to the urology department.

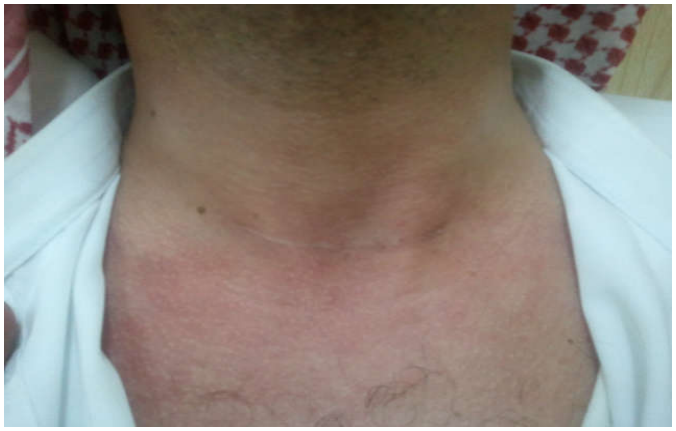

Figure 5. The patient two years post-operation

DISCUSSION

Riedel's thyroiditis is defined as a fibrotic process associated with a mononuclear cell inflammation that extends beyond the thyroid into perithyroidal soft tissues. The incidence is 1.06/100000 of outpatients. Prevalence is 0.05% or less of surgical thyroid disease, female to male ratio is 1:4, and the affected age group is 30-50 years. There is no known etiology for Riedel's thyroiditis, but the presence of inflammatory cells and antithyroidal antibodies support the autoimmune concept. The presence of fibrosclerotic lesions in the medastinum and retroperitoneum support the concept of systemic disease rather than a local thyroidal disease and is due to fibroblast proliferation induced by cytokines produced by B or T lymphocytes (Zimmermann-Belsing, 1994; Mitchinson, 1986; Dahlgren, 2010). The theory of systemic disease is supported by association of this type of thyroiditis with other IgG4-related systemic diseases. IgG4 plasma cells and high level serum IgG4 have been identified after thyroidectomy (Zimmermann-Belsing, 1994; Mitchinson, 1986; Dahlgren, 2010). Familial occurrence of multiple fibrosclerotic lesions which include Riedel's thyroiditis have been reported and have suggested a genetically-determined process (Comings *et al.,* 1967). Clinically, most reported cases presented with a slow-growing painless mass associated with obstructive hypothyroid and hypoparathyroid symptoms due to diffuse involvement of the thyroid, but only in one-third of patients which means the disease can spare some thyroidal follicles. Hypoparathyroidism due to invasion of perithyroidal tissues including the parathyroid, (the size varies) and bilateral involvement has been reported as well. Usually it is stony hard and partially moves with swallowing because it is adherent to the surrounding tissues (Schwaegerle *et al.,* 1988; Chopra *et al.,* 1978; Yasmeen *et al.,* 2002). In our case, the patient had hypothyroidism but his parathyroid function was normal. One parathyroid was invaded by fibrosis as shown in the histopathology and the thyroid seemed to have diffuse fibrosis. Laboratory findings of Riedel's thyroiditis are nonspecific. The thyroid function test will be affected in 30 % of patients. In our patient, the TFT was affected and he was hypothyroid. Auto antibodies are elevated in 60% of patients and was elevated in our patient. ESR could be elevated. Ultrasound demonstrates hypoechoic a vascular gland, either diffuse or segmental involvement of the gland, and CT and magnetic resonance imaging can show the extent of fibrosis. FNAc, in most cases, fails to provide diagnosis at late stages of disease due to severe fibrosis. Thus, unclear diagnosis is one of indications for surgery (Beahrs *et al.,* 1957; Schwaegerle *et al.,* 1988; De Lange *et al.,* 1989; Cho *et al.,* 2007). In the current case, the first diagnosis was not clear and later in the process, due to severe fibrosis, FNAc could not show any cells, so the diagnosis was done only after surgery. Few IgG4 antibodies were detected in the histopathology of the current case's specimen and that could attributed to the late stage or severity of the disease. Clinical and radiological evidence of retroperitoneal fibrosis supported our diagnosis of at least an IgG4-related disease if not pure Riedel's thyroiditis.

Conclusion

Such disease has significant challenges in diagnosis and management; in our case, hemithyroidectomy was the diagnostic method. We recommend that the management of such disease should be done in highly specialized centers with an expert endocrine surgeon and an expert pathologist. Follow up should be continued regardless of thyroid symptoms because extrathyroid manifestations can present late in the course of the disease.

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