A case of Riedel’s thyroiditis associated with benign nodule: Mimic of anaplastic transformation

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Abstract Riedel’s thyroiditis is a rare variant of thyroiditis and characterized by the replacement of thyroid parenchyma by extensive fibrosis. Typically, the thyroid is diffusely involved and a painless, hard anterior neck mass shows clinical features similar to those of anaplastic thyroid carcinoma: a rapidly enlarging, hard, fixed thyroid mass and symptoms such as dysphagia, dysphonia, and dyspnea. We experienced a case of Riedel’s thyroiditis that had presented rapidly growing, hard, fixed, thyroid mass mimicking anaplastic thyroid cancer in a 41-year-old female patient with longstanding benign thyroid nodule for 6 years. The clinical features were indistinguishable from that of anaplastic transformation and open biopsy could exclude anaplastic thyroid cancer. After surgery final diagnosis of Riedel’s thyroiditis could be made by typical microscopic findings and immunohistochemical studies. We reported the case with review of the related literatures.

Introduction

Riedel’s thyroiditis is a rare variant of thyroiditis that was initially described by Riedel in 1896.1 It is characterized by the replacement of thyroid parenchyma by extensive fibrosis, which also invades into adjacent tissues. Typically, the thyroid is diffusely involved, although unilobular disease has been described. It typically presents as a painless, hard anterior neck mass that progresses over weeks to years to produce symptoms of compression. The clinical features of the disease are very similar to those of anaplastic thyroid carcinoma: a rapidly enlarging, hard, fixed thyroid mass and symptoms such as dysphagia, dysphonia, and dyspnea. Therefore, the differential diagnosis of Riedel’s thyroiditis should include lymphoma, poorly or undifferentiated thyroid cancer as well as chronic or granulomatous thyroiditis.

We present a case of Riedel’s thyroiditis mimicking anaplastic transformation that had occurred on the same lobe of thyroid of preexisting benign nodule.

Case

A 41-year-old female presented with rapidly growing nodule with the size of 2.5 cm in diameter on her left...
lobe of thyroid gland. Six years ago the nodule had been found at the same location of thyroid gland with the size of 1 cm in diameter and diagnosed as a benign nodule by fine needle aspiration cytology at local clinic. The nodule has been followed up and its size has not been changed until recently. Six months ago the nodule started to grow and she felt anterior neck discomfort and dysphagia due to the growing nodule. However, there were no other definite symptoms of inflammatory reaction such as local pain, edema, fever, malaise, or myalgia. In her past and family history she had undergone total hysterectomy due to left ovarian tumor 15 years ago and otherwise unremarkable.

Physical examination revealed hard, fixed nodule on the mid portion of her left lobe of thyroid gland, which was about 2.5 cm in diameter, and the right lobe was normal. There were no abnormal findings in relation to the nodule such as tenderness, voice change, and dyspnea. Review of other system also showed no abnormal findings. Biologic evaluation showed no abnormal finding except for slightly decreased level of alkaline phosphatase (72 IU/L; normal range: 96–254); white blood cell (WBC) count and differential count were within normal range (WBC 8300/mm³, segment neutrophil 57.7%, lymphocyte 36.9%, and eosinophil 0.8%). The results of thyroid function test were also within normal range: T3 (RIA) 2.0 ng/mL, Free T4 1.43 ng/dL, TSH 0.35 mIU/L, anti-thyroglobulin antibody 5.68 IU/mL, anti-microsomal antibody 3.19 IU/mL, TSH receptor antibody 11.0 U/L.

Ultrasound examination of neck showed a 2 × 1.1 × 0.9 cm sized, inhomogeneous, hypoechoic nodule in the mid portion of the left lobe of thyroid which was not clearly delineated with surrounding tissue (Fig. 1). In addition, several normal sized lymph nodes were detected on the lateral neck along the internal jugular chain. Fine needle aspiration cytology of the nodule suggested “negative” result for malignancy. However, the clinical features were so similar to anaplastic transformation from longstanding benign nodule that open thyroidectomy was decided for differential diagnosis of poorly differentiated or undifferentiated carcinoma, lymphoma or other diseases. Left lobectomy and isthmusectomy of thyroid were performed and followed by histopathological examination.

At surgery, it was difficult to expose the left lobe of the thyroid due to severe adhesion to the strap muscles and trachea. The right lobe of the thyroid was well preserved and showed normal feature of thyroid gland. The left lobe was so hard and infiltrative to surrounding tissues that en bloc resection of the left lobe was performed with strap muscles. Fortunately, left recurrent laryngeal nerve was preserved safely. Intraoperatively frozen sectional biopsy of the resected specimen showed no malignancy, but definite diagnosis was deferred. So, surgery was finished. Postoperatively, the patient was recovered well without any problems.

Histological examination of permanent specimen of the left lobe of thyroid gland showed diffuse extensive fibrosis with marked infiltration of chronic inflammatory cells to strap muscle, and the remaining tissue of left lobe was atrophic (Fig. 2A). In the middle of left lobe preexisting nodular lesion was also found as nodular hyperplasia (Fig. 2B), and blood vessels were damaged by the extensive fibrosis (Fig. 2C). Normal thyroid tissue was preserved only in small portion of left lobe of the thyroid gland. Immunohistochemical study with various markers was performed for differential diagnosis and the results were as follows: positive for thyroglobulin and TTF-1 in atrophic follicular cells (Fig. 2D), positive for UCHL-1 and CD45RO in dominantly infiltrating reactive T-cells (Fig. 2E), however, focally positive for CD20 in sparsely distributed B cells (Fig. 2F), negative for CD56, negative for chromogranin and calcitonin. Taken together, we could confirm the diagnosis of Riedel’s thyroiditis in this case.

Discussion

The etiology of Riedel’s thyroiditis is controversial and has not been resolved. Some investigators suggest a primary autoimmune etiology on the basis of association of this disease with other autoimmune diseases such as pernicious anemia and Greaves’ disease. In addition, thyroid antibodies have been reported in up to 67% of patients. This observation, in addition to the presence of both B- and T-cells in the inflammatory infiltrates, suggests a possible autoimmune mechanism, although no direct relationship has been known. Riedel’s thyroiditis is also associated with other focal sclerosing syndromes, including mediastinal, retroperitoneal, periorbital, and retro-orbital fibrosis and sclerosing cholangitis, suggesting that it may be a primary fibrotic disorder. In this case thyroid antibodies were not detected, however, extensive T-cells’ infiltration was confirmed into the lesion by immunohistochemical study. This finding suggests that autoimmune mechanism would be a main etiology of Riedel’s thyroiditis in this case.

The extensive fibrosis of Riedel’s thyroiditis is progressive and may eventually cause compression of adjacent structures, particularly the trachea and esophagus. Physical examination reveals an extremely hard goiter, often described as “woody” in texture. Occasionally the involvement is unilateral, however, typically the thyroid is diffusely involved. In this case, indolent nodule showed rapid progression mimicking the clinical features of

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**Figure 1** Ultrasound examination of the left lobe of thyroid gland. A 2 × 1.1 × 0.9 cm sized, inhomogeneous, hypoechoic nodule was found in the mid portion of the left lobe of thyroid gland which was not definitely delineated with surrounding tissue. In addition, several normal sized lymph nodes were detected on the lateral neck along the internal jugular chain.
anaplastic transformation and the involvement showed only left lobe of the thyroid gland. Most patients are euthyroid unless almost complete replacement of the gland occurs, resulting in hypothyroidism. However, hyperthyroidism has been reported.

For the diagnosis of Riedel’s thyroiditis, fine needle aspiration is usually inadequate due to the extremely hardness of the gland and open thyroid biopsy of the goiter is required to confirm the diagnosis, which also helps exclude carcinoma. The main light microscopic findings that enable this entity to be distinguished from Hashimoto’s thyroiditis are (1) extension of the fibrotic process through the strap muscles and other surrounding tissues, (2) phlebitis with luminal distension by fibrous or lymphoid tissue, and (3) relatively normal remnant thyroid tissue. In this case, all of these findings were found and confirmed the diagnosis definitely.

Surgery is the mainstay of the treatment of Riedel’s thyroiditis. The chief goal of operation is to decompress the trachea by wedge excision of the thyroid isthmus and to

Figure 2  Microscopic findings of the resected left lobe of thyroid gland (H & E stain: A–C; IHC stain: D–F; ×100). (A) Extensive fibrosis invaded strap muscle (left lower part) with abundant collagen fibers and diffusely infiltrating inflammatory cells at almost all left lobe of thyroid gland. (B) In the middle of left lobe of thyroid gland, a lesion of nodular hyperplasia that had presumably been for a long period of time was found with surrounding tissue of extensive fibrosis. (C) Vessel was found to be damaged by extensive fibrosis. (D) Some atrophic follicles were found by expression of thyroglobulin in the middle of extensive fibrosis. (E) Reactive T-cells were diffusely infiltrated into the fibrotic tissue and confirmed by CD45RO expression. (F) B-cells were sparsely found in the fibrotic tissue and confirmed by CD20 expression.
make a tissue-diagnosis. More extensive resections are not advised and often impossible because of infiltrative nature of the fibrotic process that obscures usual landmarks and structures. Despite its invasive nature, recurrent obstruction after resection is rare. Medical therapy, especially in the early stages of the disorder, has occasionally been successful, including corticosteroids and tamoxifen. Although estrogen receptors have not been identified in the thyroid tissue, the mechanism underlying the response to tamoxifen has been postulated to be related to transforming growth factor (TGF)-β1. TGF-β1 is a potent growth inhibitor of immature fibroblasts and epithelial cells and has been shown to be upregulated by tamoxifen. Thyroid hormone therapy is indicated only if hypothyroidism is present, since suppression therapy is ineffective.

We present a case of Riedel’s thyroiditis in a 41-year-old female patient mimicking anaplastic transformation that had occurred on the same lobe of thyroid of preexisting benign nodule.

References