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Case report

Severe trachea compression caused by Riedel's thyroiditis: A case report and review of the literature

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HIGHLIGHTS

- Riedel's thyroiditis (RT) is a rare form of chronic thyroiditis, associated with fibroinflammatory process involving the thyroid and surrounding cervical tissues, leading to compressive symptoms.
- It is important to differentiate this condition from other thyroid disorders, especially malignant lesions.
- Thyroidectomy is indicated for patients with compressive symptoms, suspicious malignancy and failure of conservative management.
- Clinicians should be aware of RT.

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ABSTRACT

Background: Riedel's thyroiditis (RT) is a rare form of chronic thyroiditis, associated with fibroinflammatory process involving the thyroid and surrounding cervical tissues, leading to compressive symptoms.
Case presentation: We present a case of RT in a 73-year-old female with dyspnoea caused by severe trachea compression. She had reported dyspnoea during physical stress, and had noticed a large mass on the front of the neck. Despite the combination of various imaging modalities, the thyroid mass was not differentiated from thyroid malignancy and other thyroid disorder. Total thyroidectomy and tracheotomy were performed. During surgery, the thyroid had severe adhesion to surrounding tissue and the pathology revealed RT.
Conclusions: Clinicians should be aware of RT. It is important to differentiate this condition from other thyroid disorders, especially malignant lesions. Thyroidectomy is indicated for patients with compressive symptoms, suspicious malignancy and failure of conservative management.

1. Introduction

Riedel's thyroiditis (RT) is a rare thyroid disease, which is found in 0.06% of all thyroidectomies, and reports are often limited to case reports and small case series. It was first described by Bernhard Riedel in 1896. Till now, there have been around 200 cases reported [1]. The main characteristic of RT is invasive fibrosis of the thyroid. Generally, it presents with a stony-hard and fixed neck mass. Some patients would have the compressive symptoms such as dysphagia and dyspnea. However, there were few reports about severe trachea compression caused by RT in China. Here we present a case of Riedel's thyroiditis that caused severe trachea compression and review the literature regarding the diagnosis and treatment of RT.

2. Case report

A 73-year-old female patient was admitted to our hospital. The patient was referred to our institution with a 3-month history of Hashimoto's thyroiditis. She had reported fatigue and dyspnoea during physical stress, and had noticed a large mass on the front of the neck for two months. The patient presented with general weakness, dysphagia and dyspnea on admission. On physical

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examination, there was a firm, painless, diffuse thyroid swelling and wheezing sounds were noted on auscultation.

A thyroid function test was abnormal, which showed hypothyroidism: T3 of 0.897nmol/L, FT3 of 3.00pmol/L, T4 of 89.37nmol/L, FT4 of 12.69pmol/L, TSH of 6.77uIU/mL,TG-Ab of 1359.0 IU/mL, and TPO-Ab of 600 IU/mL.Ultrasonography (US), computed tomography (CT), and bronchofiberscopy were performed. These imaging modalities in the patient with suspected thyroid carcinoma showed a diffusely enlarged mass covering both thyroid lobes encircling the trachea and esophagus. The minimal diameter of the trachea is 8 mm. This mass caused tracheal stenosis, but there was no evidence of tracheal invasion [Fig. 1A and B,C]. Despite the combination of these imaging modalities, the thyroid mass was not differentiated from thyroid malignancy and other thyroid disorder such as severe thyroiditis. Preoperative pathologic diagnostic procedure including fine-needle aspiration cytology (FNAC) was not performed because we have thought that FNAC might not obtain adequate tissue from the fibrotic thyroid and definite diagnosis usually needs surgical biopsy [1]. At that time, surgery was imperative to alleviate the compressive symptoms. In fact.

Total thyroidectomy was performed and an intraoperative frozen section was also performed due to the suspicion of thyroid malignancy. First, the biopsy revealed that there were no malignant epithelial cells, and the possibility of RT was suggested. Next, bilateral total thyroidectomy (including isthmus) was performed clearly without complications due to the purpose of relieving the compressive symptoms even though the benign result of the frozen section. Then, tracheotomy (indwelling tracheostomy tube) was performed in order to avoid asphyxia caused by tracheal collapse. In fact, surgery for RT is technically most challenging. Severe inflammatory fibrous process with invasion to surrounding tissue makes the thyroid adhere to surrounding tissue densely with obscure plane.

Histopathological examination (hematoxylin and eosin staining) showed thyroid follicular structure disappeared. It was replaced by extensive fibroblast and collagenous fibrosis. There were more nodules fibroblasts, lymphocytes or lymph follicles and squamous metaplasia nests [Fig. 1, D]. Immunohistochemical stainings with various markers were performed for differential diagnosis with the following results:Vimentin (+),LCA (+),CD68 (+),CK5/6 (squamous metaplasia nest+), Ki-67 (+),Syn (−),CT (−),TG (−)[Fig. 1, E]. Taken together, Riedel's thyroiditis was the final pathological diagnosis.

The patient was discharged from the hospital 8 days after surgery without decannulation. The tracheostomy tube was removed after 27 days, and the respiration function was completely restored. She has received routine check-ups and thyroid function tests along with thyroid hormone replacement (L-thyroxine 0.1 mg/day).

3. Discussion

RT, also known as fibrous thyroiditis, invasive thyroiditis, and Riedel's struma is an extremely rare condition of unknown aetiology [1]. It is characterized by breathing difficulties and dysphagia resulting from pressure from a rapidly enlarging thyroid and more common in females, with an operative incidence of 0.06% reported [2]. Local compressive symptoms including dyspnea by tracheal compression and dysphagia are frequent in RT, but severe trachea compression caused by RT are not common, as in the present case [3]. Although there are many hypotheses regarding this disorder, the prevailing view is that it is part of a generalized fibroinflammatory process that also involves other organs [4,5]. And the most probable cause of RT is an autoimmune process [6]. A study performed by Ken Takeshima et al.has shown that immunoglobulin G4-related systemic disease (IgG4-RSD) is likely involved [7]. The main characteristics of RT is invasive fibrosis that

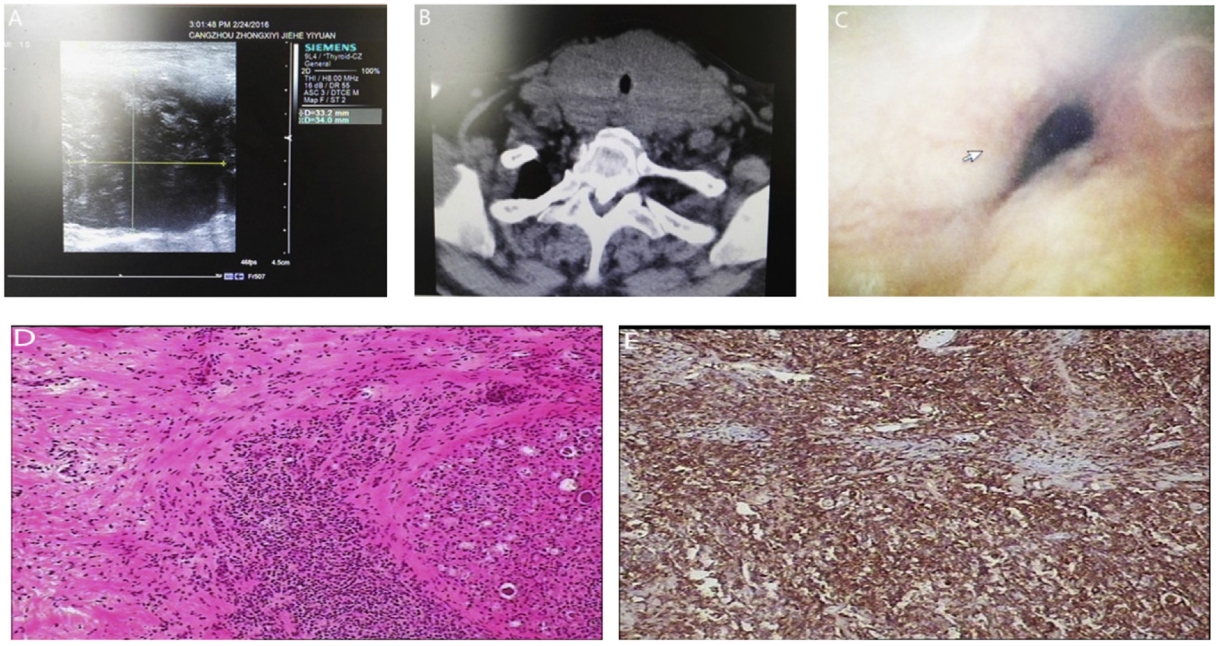

Fig. 1. A. Longitudinal ultrasonography scan of the bilateral thyroid gland showed diffuse homogeneous hypoechoic change and multiple bilateral cervical lymph nodes. **B**. Computed tomography scan of the neck in the patient with suspected thyroid carcinoma showed severe trachea compression caused by a diffusely enlarged mass encircling trachea. **C**. Tracheoscopy revealed a tracheal stenosis by external pressure. The minimal diameter of the trachea is 8 mm. **D**. Histopathological examination showed thyroid follicular structure disappeared. It was replaced by extensive fibroblast and collagenous fibrosis. There were more nodules fibroblasts, lymphocytes or lymph follicles and squamous metaplasia nests (magnification, ×10). **E**. Immunohistochemical stainings with various markers were performed for differential diagnosis with the following results: Vimentin (+),LCA (+),CD68 (+),CK5/6 (squamous metaplasia nest+),Ki-67 (+),Syn (−),CT (−),TG (−) (stain, hematoxylin and eosin; magnification, ×10).

partially destroys the thyroid gland and extends into adjacent neck tissues [1,8]. The present case had a history of Hashimoto's thyroiditis, pointing to hypothyroidism. But, there was no IgG4 test. Fatourechi et al.reported that 14 patients were hypothyroid at presentation, and 9/10 had positive TPO or TG antibodies, similar to the present case [9].

It is hard to distinguish RT from other thyroid disorders, due to the appearance is nonspecific and can be seen in other disease processes that present with diffuse fibrotic involvement, such as Hashimoto thyroiditis, lymphoma, and thyroid carcinoma. Although the most important diagnostic tool for thyroid disease is FNAC under US guidance, RT usually cannot be diagnosed accurately by preoperative cytology [10]. In addition, various imaging modalities, including US and CT, can be performed for the diagnosis of RT, but may not be helpful for the definite diagnosis of RT and differentiation from thyroid malignancy. In fact, the disease is easily misdiagnosed due to low incidence and limited experiences for most clinicians. Therefore, we hold opinion that surgical biopsy is still the key tool for definite diagnosis of RT in that the presentation of RT may mimic thyroid malignancy.

The standard therapy of RT is not established yet. Conservative treatment for RT, including glucocorticoids and tamoxifen, can be performed [11]. Surgical excision is generally preferred to relieve compressive symptoms and confirm the diagnosis by excluding malignancy in RT, as in the current report [12]. Although the thyroid resection is very difficult due to the unclear anatomical relationship caused by RT and often results in postoperative complications, thyroidectomy should be performed for an accurate diagnosis as well as relieving trachea compression caused by RT. Some authors suggested wedge resection or isthmusectomy, instead of radical thyroidectomy, to relieve compressive symptoms and to avoid complications of hypoparathyroidism or recurrent laryngeal nerve injury [4,13]. We support that surgical intervention is indicated for patients with compressive symptoms, failure of conservative management, or when differentiation from malignancy can not be achieved [4,13].

4. Conclusions

In conclusion, clinicians should be aware of RT. It is important to differentiate this condition from other thyroid disorders, especially malignant lesions. Thyroidectomy is indicated for patients with compressive symptoms, suspicious malignancy and failure of conservative management.

Ethical approval

Approval of the study was obtained from the Institutional Review Board.

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Author contribution

Ren Chongxi has made substantial contributions to conception and design; Wang Hongqiao and LiuYan have been involved in drafting the manuscript and revising it critically for important intellectual content.

Conflicts of interest

The authors declare that they have no competing interests.

Guarantor

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