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Case Report

Riedel's Thyroiditis: A Rare Clinical Entity

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ABSTRACT

Introduction

Riedel's thyroiditis is a chronic inflammatory disease of the thyroid gland characterized by fibrotic infiltration which leads to its parenchyma being replaced by fibrous tissue rendering the gland nonfunctional. The exact etiology is unknown, but currently, the most accepted view is that of systemic autoimmune hypothesis. Due to its low incidence and it resembling other more common thyroid pathologies there is often a delay in diagnosis. Due to the paucity of literature on the disease as it is a rare entity this report presents an insight on the diagnostic dilemma posed by Riedel's Thyroiditis and the possible means for the management of the disease.

Case Report

45-year male presented with complaints of an anterior neck swelling for the last 20 years with a discharging opening in the neck following decannulation of a tracheostomy and also with difficulty breathing on lying down for the last 14 days. The patient was investigated and diagnosed as a benign thyroid swelling with a sinus tract and taken up for hemithyroidectomy. Ultimately was diagnosed as a case of Riedel's Thyroiditis. The patient has been in regular follow up and is doing well.

Discussion

Clinical knowledge of such a presentation of Riedel's thyroiditis would enhance our ability to make an accurate diagnosis. We report a case of a male patient with Riedel's thyroiditis keeping in mind its rare occurrence and highlighting the diagnostic challenges.

Keywords

Riedel's Rhyroiditis; Hemithyroidectomy; Thyroid Fistula; Euthyroid

Riedel's thyroiditis was first described in 1896 by Reidel. It is a rare disease with a low incidence of 1.06 cases per 100,000 outpatients and accounting for 0.06% of all thyroidectomies.[1] It is a chronic inflammatory disease characterized by invasive fibrosis that partially destroys the thyroid gland and extends into adjacent neck structures.[2,3] Thyroid gland is non tender with a stony hard consistency as a result of fibrosis. The literature is often limited to case reports and small case series.

Riedel's thyroiditis presents a clinical challenge since it can mimic the fibrous variant of Hashimoto thyroiditis or malignant neoplasm during preoperative clincoradiological examination. Also, diagnosis can only be confirmed with an open biopsy of the thyroid gland highlighting the fact that there are no non-invasive or minimally invasive tests or criteria to establish the diagnosis.

Case Report

A 45-year-old male presented to the outpatient department at our institute with an anterior neck swelling for 20 years which was insidious in onset and gradually progressive. It was associated with shortness of breath on lying down since the last 15 days. Patient had a past history of a road traffic accident 8 months ago following which he was tracheostomised in view of prolonged intubation due to diffuse axonal injury by the department of Neurosurgery at our hospital. Three weeks after decanulation he developed a persistent opening in front of neck which was associated with purulent discharge since 6 months.

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On clinical examination of the swelling, it was approximately 10 x 7 cm in size mainly on the right side of neck with retrosternal extension, which was firm and non-tender. It was mobile as it moved with deglutition but not with protrusion of tongue. A fistulous opening of size 1 x 1cm close to midline was present through which pus could be expressed, with normal surrounding skin (Fig.1).

A thyroid function test was done which showed the patient to be euthyroid: T3 of 0.67 ng/ml (0.97-1.69 ng/ml), T4 of 4.6 mcg/dl (5.53-11.0 mcg/dl) and TSH of 3.84 mIU/L (0.46-4.68 mIU/L). AntiTPO was <28IU/ml(0-34IU/ml) and thyroglobulin levels of 1.4ng/ml(1.4-29.2ng/ml)

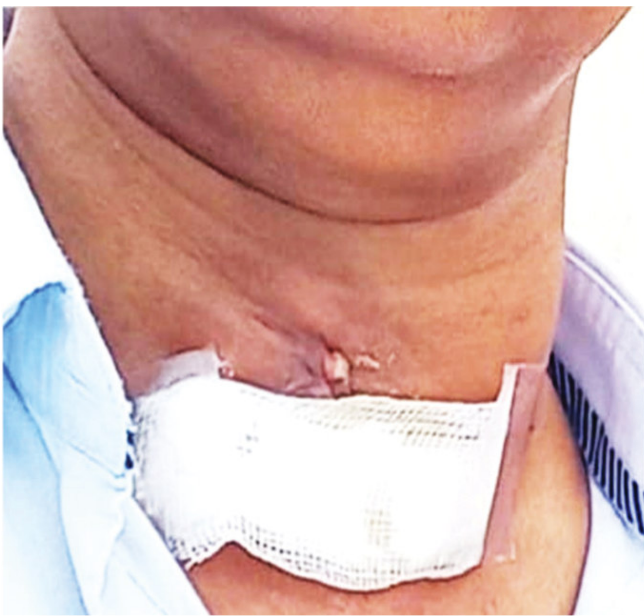

Fig. 1: Right sided neck swelling with a fistulous opening

Imaging modalities like ultrasonography (USG), X-ray soft tissue neck (STN) and Contrast enhanced computed tomography (CECT) Neck were performed. USG Neck showed 4 x 5.6 x 7 cm (85 cc vol) sized abscess in the right lobe of the thyroid with a separate TIRADS V lesion measuring 4.7 x 3.7 x 3.8 cm which was solid, hyperechoic, wider than taller, showing punctate and peripheral rim calcifications with extrathyroid extension (Fig.2). It also showed deviation of trachea towards the left. A fistula tract of 2.8 cm long and 4.6 mm thick with its external opening near midline and internal opening communicating with the right thyroid lesion, was also noted (Fig 3 & 4).

CECT Neck revealed multiple heterogeneously enhancing hypodense nodules seen in right lobe of thyroid showing multiple coarse calcification and air foci with extension into anterosuperior mediastinum, largest measuring 5.6 x 6.4 cm with compression and displacement of trachea towards the left side, suggestive of a Colloid nodule or Follicular adenoma.

X-ray STN anteroposterior and lateral view showed gross displacement of the trachea to the left with a soft tissue opacity on the right side with areas of coarse calcification, anterior to the airway at level C4-C6 with a normal prevertebral space and normal vertebrae (Fig 5 & 6).

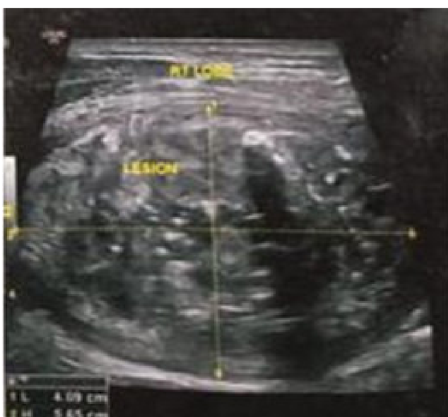

Fig. 2. TIRADS V lesion in right thyroid lobe with internal areas of calcification

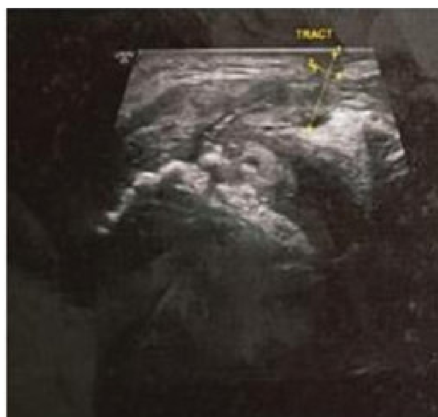

Fig. 3. External opening of the fistulous tract

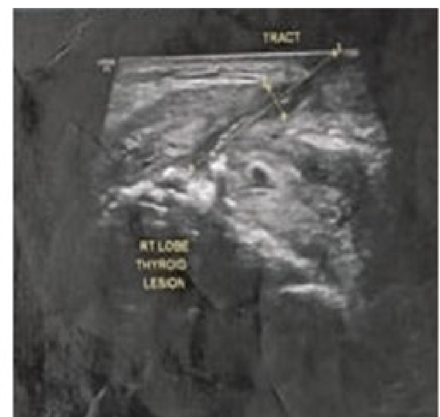

Fig. 4. Internal opening of the fistulous tract into the right lobe of thyroid

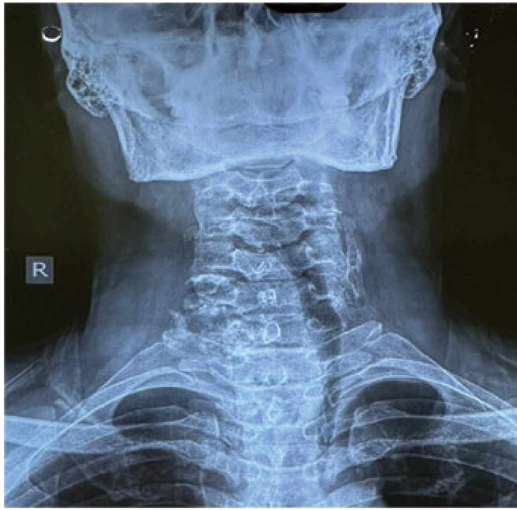

Fig. 5. X-ray STN anteroposterior view showing tracheal deviation

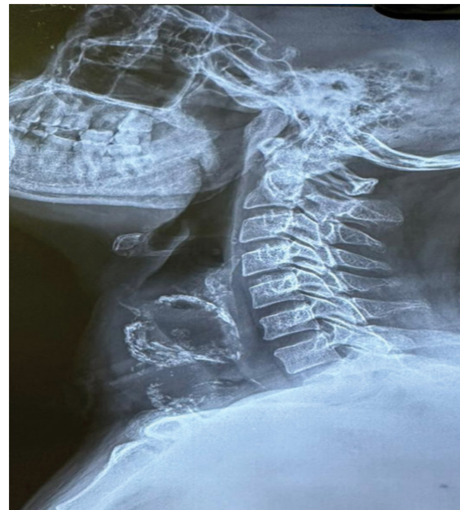

Fig. 6. X-ray STN lateral view showing coarse calcifications

USG guided FNAC showed small group of slightly scattered benign follicular cells in a background of blood and colloid, suggestive of benign follicular nodular disease (Bethesda Category II) in the right lobe and pus aspirated from a swelling near the fistula, consisting of dense acute and chronic inflammation in a background of blood, suggestive of a non-specific abscess.

The patient was subsequently planned for right hemithyroidectomy under general anaesthesia after due considerations and pre anaesthetic checkup. Due to extensive tracheal deviation the patient was intubated via fibreoptic approach.

Horizontal skin crease incision was given with an elliptical encirclement of the fistulous opening. Intraoperatively the right lobe of thyroid was found to be stony hard with area of surrounding fibrosis. The sternothyroid muscle was found adherent to the gland and was partially removed. The right lobe of thyroid was

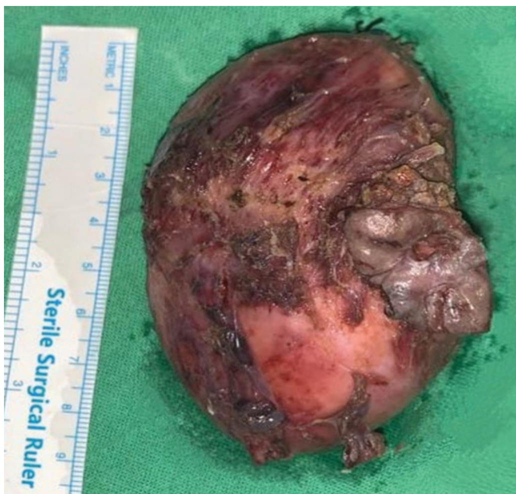

Fig. 7. 9 x 6 cm Rt Lobe of thyroid with isthmus and adherent sternothyroid muscle

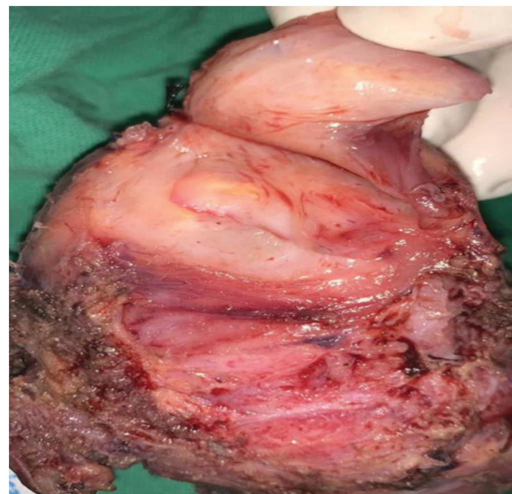

Fig. 8. Thyroid was stony hard in consistency and cut surface showed calcification

delivered in toto along with the isthmus and the cutaneous opening of the sinus tract (Fig. 7 & 8) and sent for histopathological examination.

Histopathological examination on gross showed the whole parenchyma replaced with solid cystic tumor with necrotic material and calcifications. Small portion of normal thyroid tissue seen at lower pole. Cut surface gritty due to calcification. Microscopy revealed thyroid tissue being almost replaced by extensive fibrosis and sclerosis (fig.9) with dense infiltration by lymphocytes (fig.10), plasma cells and histiocytes hemosiderin laden macrophages were present, areas of acute inflammatory cell infiltrate with abscess formation and necrosis were noted along with few areas of calcification and foreign body giant cells (fig.11). The final pathological diagnosis of Riedel's thyroiditis was made on the basis of histopathological findings.

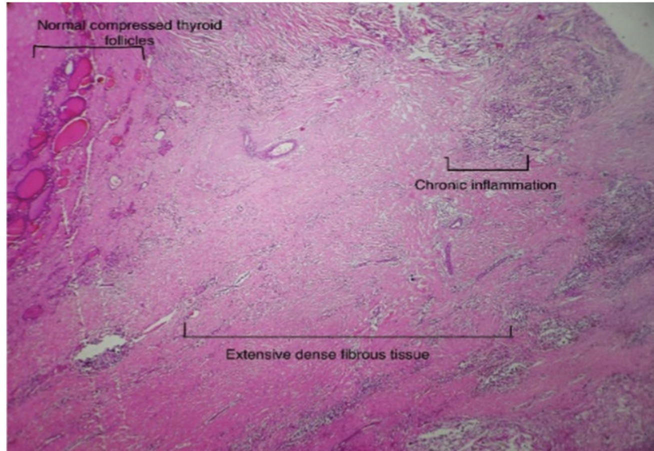

Fig. 9. 4x magnification with H and E staining showing extensive fibrous tissue and thyroid follicles pushed

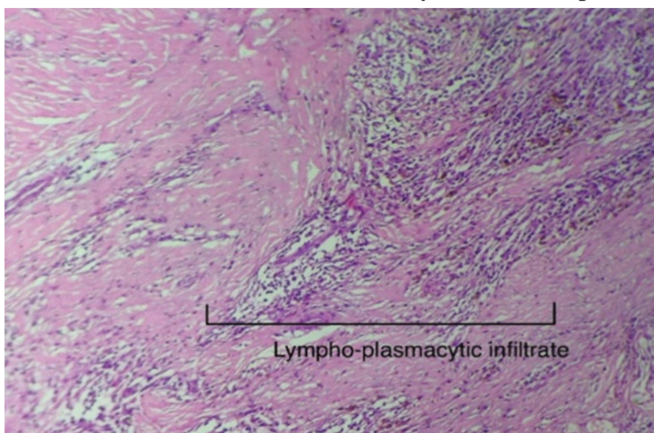

Fig. 10. 10x magnification with H and E staining showing Lympho-plasmacytic infiltrate

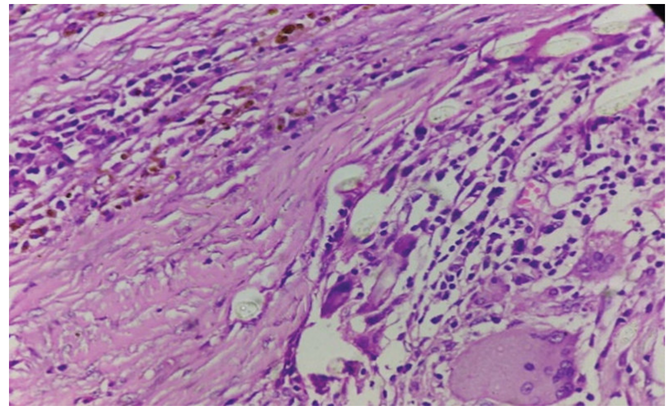

Fig. 11. 40x magnification with H and E staining showing Hemosidern laden macrophages and foreign body giant cells

Patient is in regular follow-up with no signs of recurrence and is doing well.

Discussion

Riedel's thyroiditis is extremely rare. It is a rare inflammatory process of the thyroid gland and surrounding structures.[2] It is usually associated with extensive fibrosis, replacing the normal thyroid tissue leading to hypo functionality of the gland. It has a low incidence of 1.06 cases per 100,000 outpatients accounting for 0.06 % of all thyroidectomies.[3]

There are multiple hypothesis on how it develops and its pathogenesis, ranging from the intrathyroidal hypothesis,[4-6] pharmacological hypothesis[6] and the inherited susceptibility and development hypothesis.[6] The most accepted etiopathogenesis is systemic autoimmune theory where higher expression of the gene PIK3CA stimulates fibroblasts growth factors which leads to fibrosis.[7-8] Unfortunately, the genetic testing for the same could not be done in the this case due to resource limitations.

Riedel's thyroiditis shows peak age incidence in 5th decade and gender predisposition with females affected more common than males and F to M ratio of 4:1[3] whereas

in our case male in his 4th decade was affected. It has been found to occur more commonly in smokers but in our case the patient developed Riedel's thyroiditis[3] despite being a non-smoker with no history of any substance abuse which doesn't coincide with the above finding.

Our patient presented with a thyroid swelling which had stony hard consistency similar to how thyroid swelling was originally described in Riedel's thyroiditis as 'eisenharte'.[9] 25-80% of the patients with Riedel's thyroiditis show hypothyroidism as thyroid gland undergoes fibrosis[9,10] but in our case the patient was euthyroid. Reidels patients usually have elevated Anti-TPO and thyroglobulin levels,[15,16,18] while our case was peculiar with normal Anti-TPO and thyroglobulin levels.

Ultrasonography for Riedel's Thyroiditis tends to show a hypoechoic and hypovascular lesion which are generally non-specific[2] since they mimic other thyroid conditions however in our case the findings were that of a hyperechoic solid lesion with areas of calcification and extrathyroidal extension.

Kumar et al. showed that one of the most important tools for diagnosis of thyroid pathologies, i.e. FNAC, is unreliable and cannot be depended upon to clinch the diagnosis of Riedel's thyroidits.[11] In our case too this fact was highlighted as the patient was diagnosed as a benign follicular disease (Bethesda category II) on FNAC

CECT of the neck was warranted in our case as patient had compressive symptoms and also to see the extent of the retrosternal extension. The general findings seen on CT are of hypo dense infiltrative mass[12] but in our case the CT findings showed heterogeneously enhancing hypodense nodules in right lobe of thyroid with multiple calcifications. MRI findings of Riedel's thyroiditis is not well known hence its role as a diagnostic tool is not useful.[13]

Mycophenolate mofetil, rituximab and low dose radiation have been used successfully[14] used for treatment of Riedel's thyroiditis as mentioned in literature.

Surgical intervention as a therapeutic modality is limed to debulking. Extensive resection is not preferred due to its infiltration in adjacent structures[15-17] and increased risk of post operative complications, upto 39%.[15] Our patient had compressive symptoms and tracheal deviation and thus the patient was subjected to surgery.

Conclusion

Riedel's thyroiditis is a rare inflammatory disease of thyroid gland which is difficult to differentiate between other more commonly encountered thyroid disorders. This case report highlights the importance of including Riedel's thyroiditis as differential diagnosis while evaluating patients presenting with hard thyroid swelling and compressive symptoms and such clinico-radiological-cytological contradiction. It is important that a timely diagnosis of Riedel's thyroiditis be made which helps in faster recovery and decreased associated complications.

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