Case Report

DOI: 10.21276/APALM.1392

Riedel's Thyroiditis in a 78 Year Old Male: A Rare Experience

Anju Pradhan[1]*, Krishna Maharjan[1], Punam Paudyal[1], Niharika Shah[1] and Shyam Thapa Chettri[2]

[1]Department of Pathology, B.P. Koirala Institute of Health Sciences (BPKIHS). Dharan, Nepal
[2]Department of Otorhinolaryngology and Maxillofacial Surgery, B. P. Koirala Institute of Health Sciences. Dharan, Nepal

ABSTRACT

We present a rare case of Riedel's thyroiditis in a 78-year-old male, native of hilly region of Nepal who presented with chief complaint of long standing swelling of the thyroid with discharging sinus. Right hemithyroidectomy with excision of sinus was done. Gross examination showed asymmetrically enlarged right lobe of thyroid with adherent fibroadipose and muscular tissue. Microscopy revealed diffuse hyalinised fibrosis of the thyroid parenchyma with presence of variable number of atrophic to few normal thyroid follicles in between. The stroma showed dense lymphoplasmacytic infiltration with foci of calcifications and hemorrhage. There was extension of fibrosis beyond the thyroid capsule, encasing the skeletal muscle bundles at many places. Diagnosis of Riedel's thyroiditis was made.

Riedel's thyroiditis is a rare entity but can occasionally be encountered. Various imaging modalities may not be helpful for the definite diagnosis. Diagnostic thyroidectomy should be performed for the accurate diagnosis and further management.

Keywords: *Thyroiditis, Hashimoto Thyroiditis, Fibrosis, Hürthle Cell, Thyroid.*

Introduction

Riedel's thyroiditis (RT) is a chronic thyroiditis characterized by an inflammatory proliferative fibrosing process that partially destroys the thyroid gland and extends into the surrounding tissues beyond the thyroid capsule.[1] It is an uncommon disease found in about 0.05% of all thyroidectomies.[2] Clinical symptomatology and imagery overlap makes it difficult to differentiate from malignancy. Riedel's thyroiditis usually cannot be diagnosed accurately by preoperative cytology.[3]

Case Report

A 78-year-old male presented in the Department of Otorhinolaryngology and Maxillofacial Surgery of B. P. Koirala Institute of Health Sciences, Dharan, Nepal, with complain of mass at right side of anterior neck and a discharging sinus since 20 years which was not associated with pain or fever. On physical examination, mass moved with deglutition and was non-tender. Thyroid Function Test showed subclinical hypothyroidism. T3 and T4 levels were within the normal range while thyroid stimulating hormone (TSH) was raised (10.7 µIU/mL).

Neck Ultrasound showed enlarged right lobe of thyroid with multiple nodules and coarse calcifications. A large, eccentric, heterogeneous, predominantly hypo echoic area with echogenic foci within a hypo echoic tract with internal echoes extending from the lesion to skin surface was observed, which was suggestive of an infective pathology.

Preoperative diagnostic fine-needle aspiration cytology (FNAC) performed yielded non- diagnostic material. Right hemithyroidectomy with excision of sinus was done.

On gross examination, the gland was found to be asymmetrically enlarged with adherent fibroadipose and muscular tissue on the right. The cut surface was grey white to yellowish, firm to hard, with gritty sensation on sectioning (Figure 1). The attached soft tissue showed presence of a sinus tract which was not reaching up to the thyroid. Normal thyroid parenchyma was not identified grossly.

Microscopic examination revealed diffuse hyalinised fibrosis of the thyroid parenchyma with presence of variable number of atrophic to few normal thyroid follicles in between (Figure 2 & 3). The stroma showed dense lymphoplasmacytic infiltration (Figure 4) with foci of calcifications and hemorrhage. The fibrosis extended beyond the thyroid capsule and were seen encasing the skeletal muscle bundles at many places (Figure 5 & 6). Sections from sinus tract revealed a tract lined by granulation tissue and mixed inflammatory cells. The tract was not extending into the thyroid parenchyma microscopically as well. There was absence of hürthle cells, lymphoid follicles, multinucleated giant cells or granulomas. Based on these findings a diagnosis of Riedel's thyroiditis was considered.

Discussion

Riedel's thyroiditis (RT), also known as Riedel struma, fibrous thyroiditis or invasive thyroiditis is an extremely

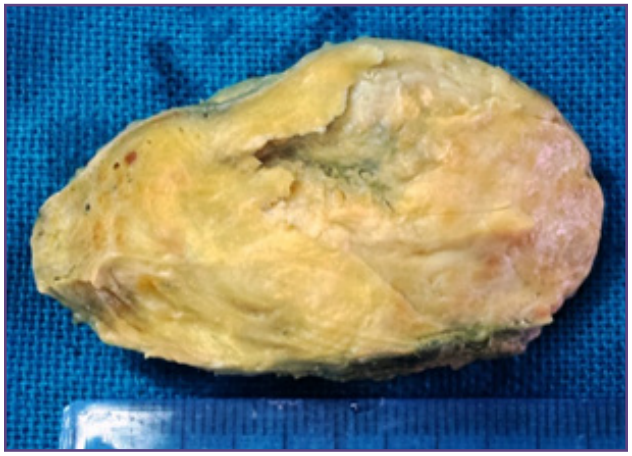

Fig. 1: Gross appearance of thyroid revealing a solid grey white to yellowish cut surface. Normal thyroid parenchyma is not seen.

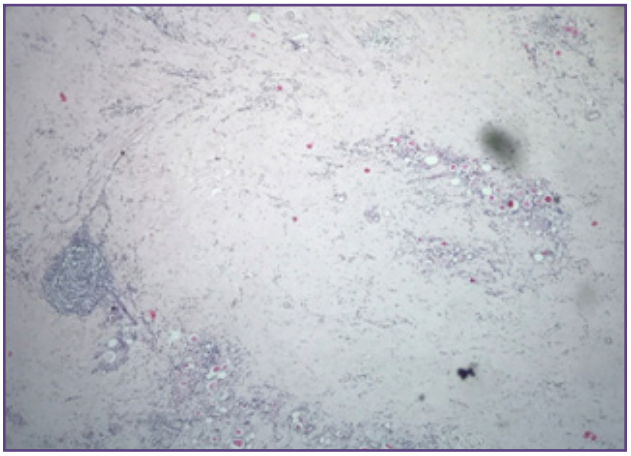

Fig. 2: Extensive fibrosis of parenchyma with residual atrophic and normal thyroid follicles and lymphocytic infiltrates. H & E stain.

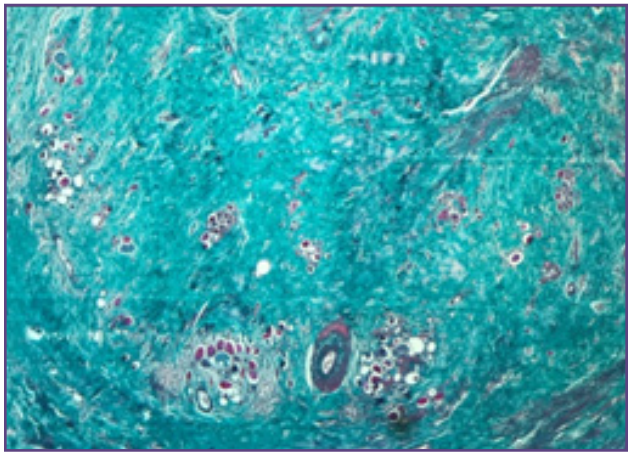

Fig. 3: Masson Trichome stain demonstrating extensive fibrosis of thryroid parenchyma with residual and atrophic thyroid follicles.

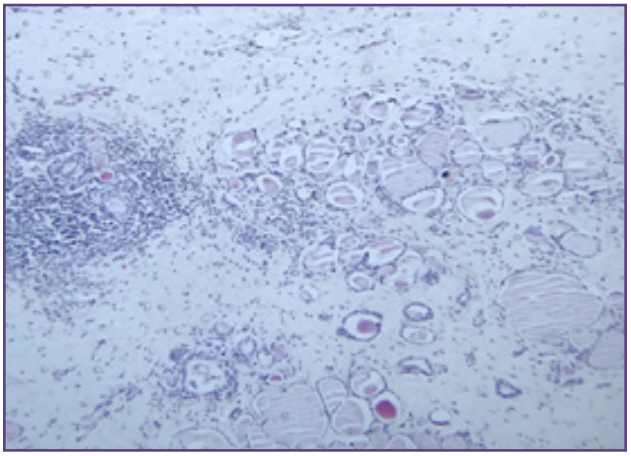

Fig. 4: High power view revealing residual atrophic and few normal thyroid follicles with mononuclear cell infiltrates.

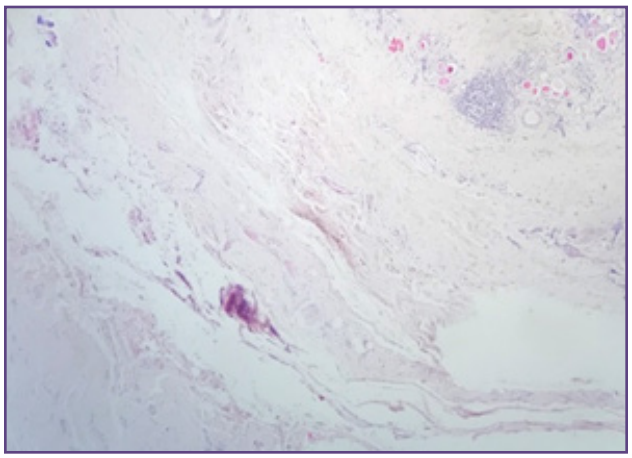

Fig. 5: Extensive fibrosis extending into the capsule and surrounding structures, with entrapment of skeletal muscle bundles at the left lower end. H & E stain.

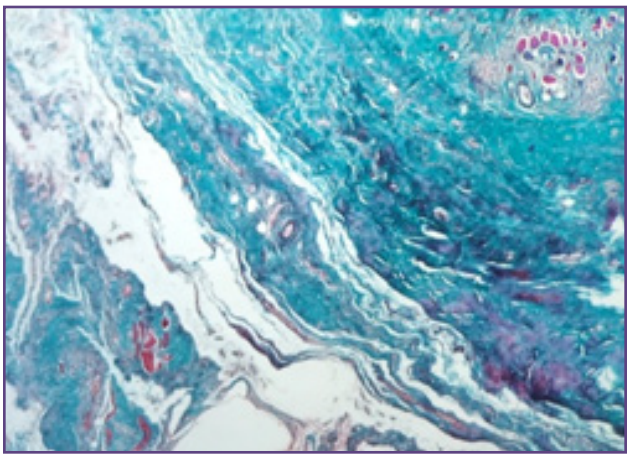

Fig. 6: Masson trichome stain highlighting the extracapsular fibrosis with entrapment of skeletal muscle bundles at the left lower end.

rare form of infiltrative and inflammatory disease of the thyroid and was first described by Bernard Riedel in 1896.[4,5] Reports of Riedel's thyroiditis in the literature are often limited to case reports and small case series. The true incidence is unknown, but in a 1985 review of 56,700 thyroidectomies performed at Mayo Clinic, only 37 cases of Riedel's thyroiditis were identified.[6]Due to extensive fibrosis of the thyroid tissue along with surrounding soft tissue, it generally presents as a firm mass in the thyroid, with compressive symptoms.[5,7] Riedel's thyroiditis presents as a painless mass as is not preceded by acute inflammatory process.[5]

Similar to cases reported by Pi GY et al[3] and Wojciechowska-Durczynska[8] et al our case presented with non-specific cervical discomfort, painless mass which was firm in consistency. In our case thyroid function test showed subclinical hypothyroidism in contrast to the euthyroidism in the case reported by Wojciechowska-Durczynska et al.[8] Hypothyroidism was seen in the case reported by Pi GY et al.[3]In 2 case reports by Zakeri H et al one case presented with hypothyroidism and the other with thyrotoxicosis.[7]

It is difficult for physicians to distinguish Riedel's thyroiditis from malignant neoplasms of the thyroid clinically because both clinical examination and imaging of Riedel's thyroiditis suggests malignancy. Ultrasound of Riedel's thyroiditis shows a hypo-echoic and hypo-vascular mass with extension into adjacent soft tissues as in our case. However, this 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.[9,10]Thus, it is hard to distinguish Riedel's thyroiditis from other forms of thyroiditis.

Preoperative diagnostic modalities such as imaging and FNAC are inconclusive, as in our case and in cases reported by Pi GY et al[3] and Wojciechowska-Durczynska et al[8].

Grossly, the gland is asymmetrically enlarged with adherent fibroadipose and muscle tissue and is stony hard in consistency. The tissue is difficult to cut with gritty sensation and cut section reveals a firm to hard, grey white fibrotic tissue with complete obliteration of the normal thyroid gland.[4,5]Our case showed similar finding. The sinus tract that was present in the soft tissue did not reach the thyroid gland.

One of the difficult and important microscopic differential of Riedel's thyroiditis is with fibrosing variant of Hashimoto's thyroiditis. Lack of extension of fibrosis into the adjacent soft tissue, absence of extensive hürthle cell metaplasia and granulomatous inflammation are key features of Riedel's thyroiditis which helps to differentiate from fibrosing variant of Hashimoto's thyroiditis.[4,5,9,11] Our case typically lacked hürthle cell metaplasia and granulomatous inflammation. There was extensive fibrosis of the with replacement of the normal thyroid, however few normal thyroid follicles were seen in between the fibrous tissue along with dense infiltration by lymphocytes and plasma cells. The plasma cells present in Riedel's thyroiditis are polyclonal with numerous IgA and IgG4 producing cells.[5]Studies have been attempted to link Riedel's thyroiditis with IgG4-related systemic disease and have thought to be the underlying condition.[11] However, further studies are required to study the disease at various stages. Occlusive phlebitis is an important diagnostic feature but was not seen in our case.

Conclusion

Riedel's thyroiditis is an extremely rare entity but can occasionally be encountered. Preoperative diagnostic procedures including US, CT, MRI, and FNAC are not helpful for the definite diagnosis of Riedel's thyroiditis and differentiation from thyroid malignancy. Diagnostic thyroidectomy should be performed for the accurate diagnosis of Riedel's thyroiditis with histopathological evaluation for further appropriate management of the patient.

Acknowledgements

We would like to acknowledge the faculties of department of Pathology and colleagues of department of Otorhinolaryngology and Maxillofacial Surgery of B. P. Koirala Institute of Health Sciences, Dharan, Nepal, our junior residents and the technical staffs for their support. We would also like to express our sincere gratitude to the head of department, Prof Dr Smriti Karki for her constant encouragement in research work.

Reference

1. Nagashima T et Al. Subclinical Riedel's thyroiditis with hypothyroidism coexisting with Mikulicz's disease. Rheumatol Int 2012; 32:1851-1852
2. Lindau RH, Wolters K, Gross N, Wax MK. Riedel disease: Report of extensive disease requiring a total laryngectomy. Head and Neck Surgery 2012;147: 799
3. Pi GY, Lee YS, Hong SW, Chang H, Park CS. A case of Riedel's thyroiditis. J Korean Surg Soc 2012; 82:317-320
4. Pirola I, Morassi ML, Braga M, De Martino E, Gandossi E, Cappelli C. A Case of Concurrent Riedel's, Hashimoto's and Acute Suppurative Thyroiditis. Case Reports in Medicine, 2009, Article ID 535974, doi:10.1155/2009/535974

5. Rosai J, Tallini G. Breast. In: Rosia and Ackerman, editors. Surgical Pathology. 10th ed. Mosby; 2011: 488-564.
6. Fatourechi M, Hay I, Mclver Sebo T, Faourchi V. Invasive fibrous thyroiditis (Riedel thyroiditis): the Mayo Clinic experience, 1976-2008. Thyroid 2011;21(7):765-772.
7. Zakeri H, Kashi K. Variable clinical presentations of Riedel's thyroiditis: report of two cases. Case reports in medicine, Volume 2011, Article ID 709264, doi: 10.1155/2011/709264
8. Wojciechowska-Durczyńska K, Durczyński A, Sporny S, StrzelczykJ, Lewiński. Riedel's thyroiditis- a case report with genes expression studies. Thyroid Research 2012, 5:2
9. Ozbayrak M, Olgun DC, Mihmanli I. Riedel thyroiditis associated with massive neck fibrosis. J Ultrasound Med 2009; 28: 267-271
10. Darouichi M, Constatntin PE. Riedel's thyroiditis. Radiology case reports II 2016; 175-177
11. Dahlgren M, Khosroshahi A, Nielsen GP, Deshpande V, Stone JH. Arthritis care and Research 2010; 62(9): 1312-1318

***Corresponding author:**
Dr. Anju Pradhan, Annapurna Chowk, Punya Marga, Dharan-16/73, Sunsari district, Koshi Zone, Nepal
Phone: 0977-9852047077
Email: dranjupradhan@gmail.com

Financial or other Competing Interests: None.

Date of Submission : 06.03.2017
Date of Acceptance : 19.06.2017
Date of Publication : 20.10.2017