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Unusual Presentation and Novel Treatment of Granulomatous Thyroiditis: A Rare and Interesting Case

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Authors' contributions

This work was carried out in collaboration among all authors. Authors RBP and SM designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors DVM and RBP managed the analyses of the study. Author RBP managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Granulomatous thyroiditis (GT) is one of the various types of autoimmune thyroiditis. Although, it usually occurs in women of reproductive age in peri-gestational period. We present a case of GT in a sixty year old gentleman presenting with pyrexia of unknown origin lasting for 12 months. On comprehensive evaluation, no other sources of chronic inflammatory disease causing fever were found. Surgical total thyroidectomy was performed as no other treatment was relieving his

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fever. Surprisingly his fever resolved completely at followup. In this context, we report this unusual clinical presentation of PUO due to GT and the role of total thyroidectomy as a treatment option.

Keywords: Thyroiditis; thyroidectomy; fever; hypothyroidism; autoimmunity.

1. INTRODUCTION

Granulomatous thyroiditis (GT) is one of the various types of autoimmune thyroiditis [1]. It usually occurs in women of reproductive age in peri-gestational period. Clinical picture is predominantly painful goiter with radiating pain to ears with low grade fever lasting for few days, often associated with mild hyper or hypothyroidism [1,2]. The treatment is symptomatic and supportive with no curative option. There are scanty anecdotal reports on the role of thyroidectomy in curing GT. In this context, we report an unusual clinical presentation and impact of total thyroidectomy on GT.

2. CASE DETAILS

A 60 year old gentleman had history of continuous low grade fever, since 12 months with quotidian spikes up to 100–101^{0}F. These spikes were often associated with chills. He could get symptomatic relief with paracetomol. He was worked up with a battery of investigations for chronic liver disease, renal disease, viral causes, tuberculosis and any autoimmune connective tissue disorders such as SLE, scleroderma etc., by various physicians. All the investigations were within normal limits with no definite etiology for this fever. He was even empirically started on anti-tuberculous medications (ATT) since 3 months with no significant alleviation from fever. One of the physician noticed anterior neck swelling (goiter) in him and referred the case to our endocrine surgery department.

We examined and evaluated the case in detail. On detailed history taking, he complained of pain in throat radiating to the ear in the past. There was a diffuse, firm, non tender grade 2 goiter and no regional cervical lymphadenopathy. There was no parotid or submandibular sialadenomegaly. No evidence any neurocutaneous markers of any autoimmune syndromes. Thyroid function test showed subclinical hyperthyroidism with serum thyroid stimulating hormone (TSH) of 0.01 uIU/L (0.3-4.4) and normal T4, T3 levels. Anti-thyroid peroxidase antibody (anti-TPO) titer was 45 IU/L (0–34). Erythrocyte stimulating hormone (ESR) was high at 36 mm/hr. Fine needle aspiration of goiter showed picture of thyroiditis with few macrophages and colloid. Radioactive iodine uptake and scan typically reveals a low RAIU and poor visualization of the thyroid. Thyroid ultrasound typically shows a heterogeneously hypoechoic pattern with mildly increased vascularity. After careful intradepartmental discussion and patient (family) counseling, we withdrew ATT and planned total thyroidectomy with the provisional diagnosis of granulomatous thyroiditis (GT) as cause for this pyrexia of unknown origin (PUO).

Total thyroidectomy was performed with uneventful morbidity. Intraoperatively, the gland was vascular with adhesions to the surrounding strap muscles. No postoperative complications or hypocalcemia occurred. Surprisingly, from 5th postoperative day his fever has resolved completely with no further spikes. ESR was 15 mm/hr at 2 weeks after surgery. Histopathology was reported as Granulomatous thyroiditis with non-caseating granulomas, macrophages, destroyed follicles and lymphocytic infiltrates. Body temperature has normalized and he never experienced any episodes of fever till last followup at 12 months. He had no other sequelae or stigmata of any systemic or chronic inflammatory or neoplastic disease during followup period.

3. DISCUSSION

Granulomatous thyroiditis (GT) is an inflammatory condition of the thyroid usually with characteristic presentation and clinical course. GT was variably called as De Quervain's thyroiditis or subacute thyroiditis (SAT) or creeping thyroiditis in the literature [1,2]. GT presents with protean clinical manifestations. Clinical picture was predominantly painful goiter with radiating pain to ears with low grade fever lasting for few days, sometimes associated with mild hyper or hypothyroidism [1]. Pain may radiate to the jaw or the ears. Malaise, fatigue, myalgia and arthralgia are common. A mild to moderate fever is expected, and at times a high fever of 104°F (40.0°C) may occur. Usually, the

onset of symptoms extends over 1 to 2 weeks and continues with fluctuating intensity for 3 to 6 weeks. Approximately 50% of the patients present during the first weeks of the illness with symptoms of thyrotoxicosis. This painful condition lasts for a week to a few months, usually demonstrates a very high erythrocyte sedimentation rate (ESR) and has a tendency to recur. Another subtype of GT is painless (silent, autoimmune) subacute thyroiditis that occurs spontaneously or following pregnancy. When it is referred to as postpartum thyroiditis (PPT). One of the recent papers showed that GT is predominantly painless [3]. The cause of SAT is idiopathic but has been linked with viral infections, including dengue fever and of late with covid fever [4,5]. It was also linked with drugs such as immunomodulators [6]. The unusual features of our case is a prolonged fever lasting for 12 months occurring and not related to gestation. Moreover this GT occurred in an elderly male person and manifested in a rare presentation of pyrexia of unknown origin (PUO).

The pathophysiology of GT is debatable and unclear. It is hypothesized that painful GT tends to follow upper respiratory tract (URIT) infections or sore throats, suggesting a viral infection. The development during the illness of cell-mediated immunity against various thyroid cell particulate fractions or crude antigens appears to be related to the release of these materials during tissue destruction [7,8]. An autoimmune reaction is possible as patients with GT often manifest HLA-Bw35 and those with postpartum thyroiditis are frequently anti TPO positive. A reported association between subacute thyroiditis and acute febrile neutrophilic dermatosis (Sweet's syndrome) may imply a common role for cytokines in both these conditions. We opine that the cause of PUO in our case is chronic release of pyrogenic cytokines by the GT process.

The usual treatment of GT is analgesic therapy for relief of pain achieved with non-steroidal anti-inflammatory agents. If this fails steroid administration is employed with daily doses of 20-40 mg prednisone. After one to 2 weeks of this treatment, the dosage is tapered over a period of 6 weeks. Beta blocking agents are usually administered for relief of thyrotoxic symptoms in the initial stage of SAT. Antithyroid drugs have no role in the management of established SAT as the excess thyroid hormone levels released from inflamed tissue.

Surgical intervention is never the primary treatment for subacute thyroiditis and was performed only if there is associated papillary cancer/ indeterminate thyroid nodule based on cytological examination [9,10]. We found no literature on the surgical thyroidectomy as a sole treatment of choice for GT. Our case highlights an unique role of surgical thyroidectomy as treatment of choice in GT as it was associated with PUO. Total thyroidectomy was not performed for goiter related complaints or associated malignancy but for GT perse.

Exact pathophysiology of GT is enigmatic but pyrogenic cytokines released by autoimmune thyrocyte destruction may cause PUO. Total thyroidectomy appears to be a viable curative option for GT causing PUO probably by reversing this pathophysiology and removing the source (thyroid gland) of this autoimmunity which provokes the release of these cytokines.

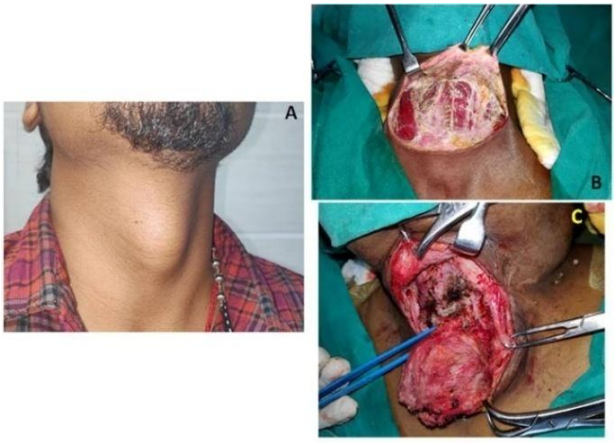

Fig. 1. A) Grade 1 diffuse goiter in anterior neck; B, C) Intraoperative images of total thyroidectomy

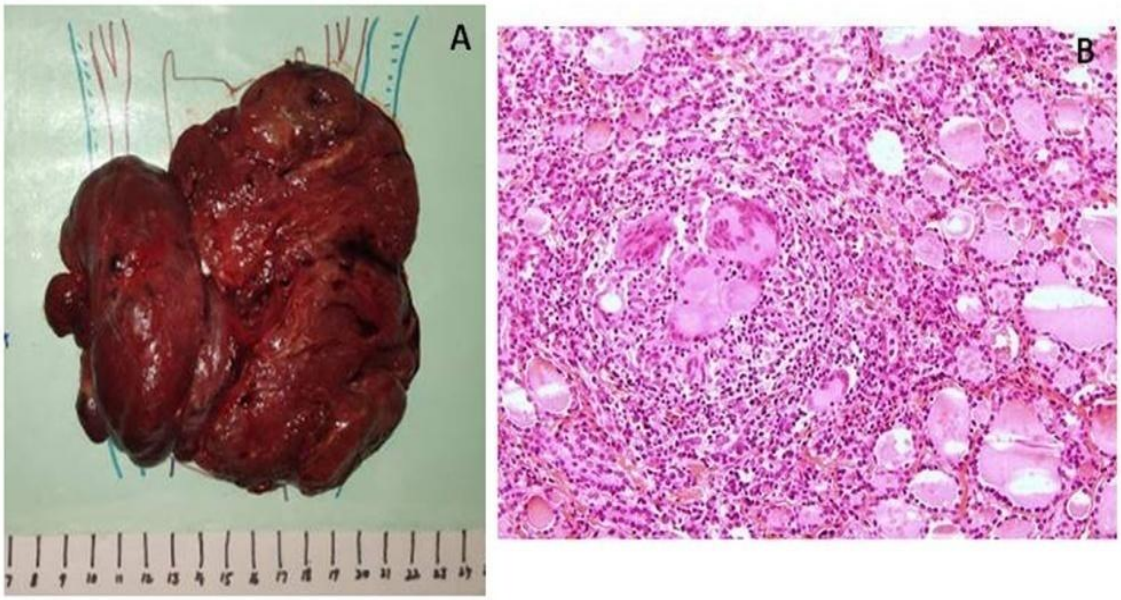

Fig. 2. A) Gross ex vivo specimen showing total thyroidectomy; B) Histopathological microphotograph (H&E; 40X) showing macrophages, destroyed follicles and lymphocytic infiltrates

4. CONCLUSIONS

Granulomatous thyroiditis can have protean clinical manifestations. Here GT unusually presented with pyrexia of unknown origin. Total thyroidectomy appears to be a viable curative option for GT causing PUO.

CONSENT AND ETHICAL APPROVAL

As per university standard guideline participant consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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