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Case Report

Recurrent Graves' Disease in Thyroglossal Duct Following Total Thyroidectomy

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ABSTRACT

Introduction: A 46-year-old female with a past medical history of Graves' disease refractory to medical management, thus requiring total thyroidectomy presented to clinic for recurrence of her hyperthyroidism and an increasing midline neck mass two years after her index operation.

Case Description: CT imaging of the neck mass revealed a locally extensive enhancing abnormality immediately anterior to the hyoid bone within the infrahyoid muscles measuring 1.4x1.9x4.0 cm. This was consistent with an exceedingly rare proliferation of a thyroglossal duct remnant secondary to recurrence of the patient's Graves' disease. The patient was treated with surgical resection of the neck mass shown to be ectopic Graves' activated thyroid tissue by pathology. The patient was restarted on thyroid hormone replacement therapy and has remained euthyroid to date following surgical resection.

Conclusion: The incidence of this event is estimated to be approximately one per a million persons and thus is a rare occurrence in endocrinology. This case highlights the potential shortcomings of surgical management of Graves' disease. Following surgical resection, the autoimmune status of the patient remains unchanged. Ectopic thyroid tissue can be found anywhere along the embryologic descent of the thyroid gland and as low as the mediastinum. Thus, patients treated with total thyroidectomy should still be monitored regularly for recurrence of Graves' disease secondary to ectopic thyroid tissue.

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Introduction

Graves' disease is an autoimmune disease characterized by activating antibodies to thyroid tissue. The incidence of Grave's disease is 20-50 cases per 100,000 persons in the general population [1]. While 95% of patients are successfully managed medically, refractory cases are typically treated with total thyroidectomy [2]. Following thyroidectomy, these activating antibodies have the biological potential to activate latent or ectopic thyroid tissue even after removal of a patient's thyroid gland. While typically located in the anterior neck, thyroid tissue can be found anywhere along the embryologic descent of the thyroid gland beginning at the foramen cecum, down the neck and as low as the mediastinum [3]. Additionally, a thyroglossal duct is the embryologic failure of complete obliteration of the structure during gestation. The rate of existence of thyroglossal ducts cysts in the general population is thought to be around 7% percent based on postmortem studies [4]. The following case

describes an instance of recurrent Graves' disease following total thyroidectomy, where ultimately the patient was found to have hyperplastic thyroid tissue along a thyroglossal duct.

Case Presentation

The patient (initials WU) is a 46-year-old woman with a past medical history of polycystic ovarian syndrome and Raynaud's disease who initially presented for evaluation of an enlarging midline neck mass. She had a prior history of Graves' disease diagnosed after an episode of thyrotoxicosis. After initial attempts at medical management with Methimazole and Propranolol, her symptoms remained inadequately controlled. When offered radioactive iodine ablation therapy, she declined due to having young children at home, and ultimately underwent total thyroidectomy in April 2018, at an outside facility.

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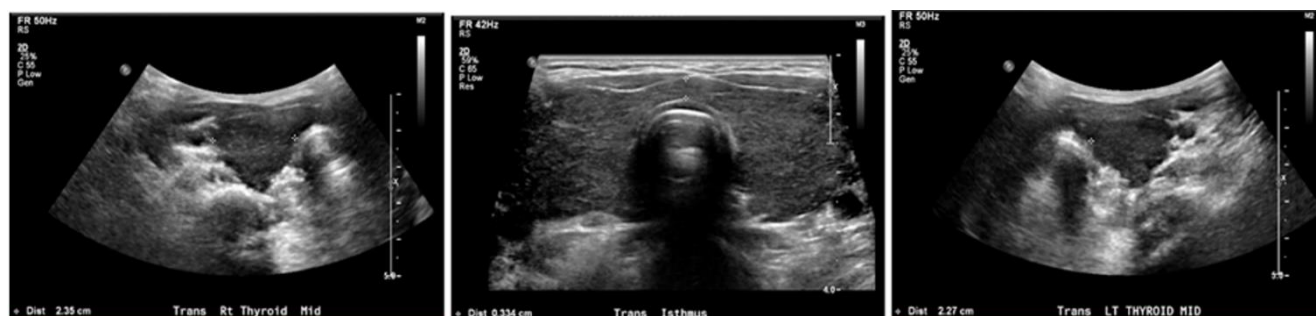


Figure 1: Ultrasound of thyroid gland transverse view showing an enlarged right, left lobe and isthmus with mild diffuse heterogeneity throughout the thyroid parenchyma.

At that time, her pre-operative ultrasound (Figure 1) demonstrated an enlarged thyroid gland, with the right lobe at $7.2 \times 1.9 \times 2.4$ cm, the left lobe at $6.2 \times 2.0 \times 2.7$ cm, and the isthmus measured 3mm. There was notably mild diffuse heterogeneity throughout the thyroid parenchyma with no discrete nodules identified. At her initial operation, she was described to have a bilateral multinodular goiter with surrounding adhesions from thyroiditis. Specimen pathology demonstrated diffuse follicular hyperplasia with papillary infoldings and hypertrophy of follicular cells with colloid showing peripheral scalloping consistent with Graves' disease. Post-operatively, she was started on 100 mcg of levothyroxine daily.

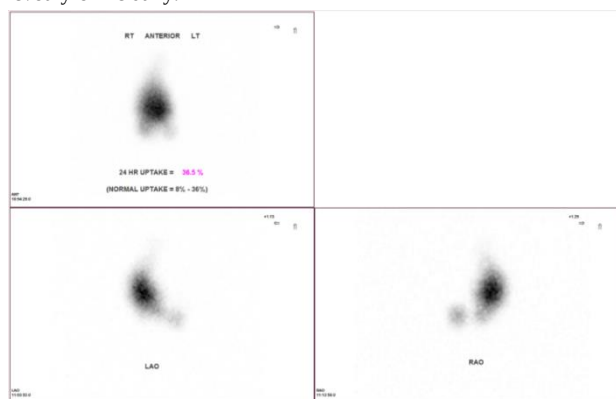


Figure 2: Radioactive iodine uptake scan 2 years post thyroidectomy showing Graves' disease of residual/regenerated thyroid tissue predominantly in the isthmus and to a lesser extent within the right and left lobe of the thyroid beds.

For the first months following surgery, she followed with endocrinology, had resolution of her symptoms, and was euthyroid while on thyroid hormone replacement therapy. Over the course of 2 years, the patient was slowly weaned off levothyroxine due to deviations in thyroid function tests. Two years post-thyroidectomy, the patient's thyroid function tests increased to a TSH of 0.04, a Free T4 of 1.2 ng/dL and a total T3 of 141 ng/dL and TSI 541. In May 2020, a radioactive iodine uptake scan was performed, which demonstrated Graves' disease of residual or regenerated thyroid tissue at the region of the isthmus and also in the left and right thyroid beds to a lesser degree (Figure 2). The patient was restarted on medical management with methimazole. CT scan demonstrated a mass anterior to the thyroid cartilage extending superiorly to the level of the hyoid bone (Figure 3).

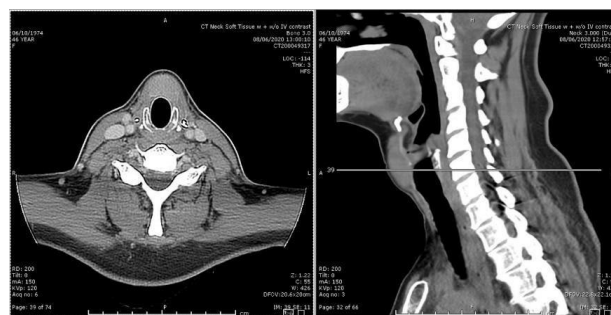


Figure 3: CT scan of the neck 2 years post-operatively from total thyroidectomy, transverse and sagittal views showing an enlarged midline mass inferior to the hyoid bone.

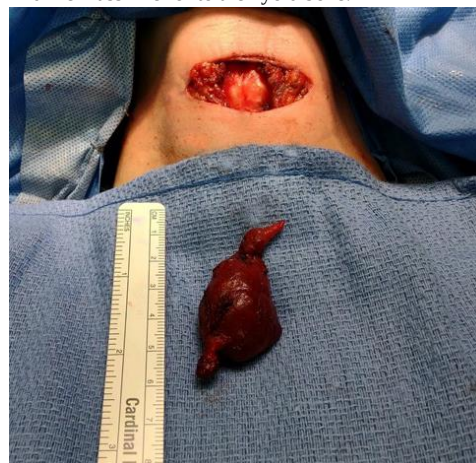


Figure 4: Intraoperative gross inspection of resected thyroid tissue. There is central expansion of the tissue as well as proximal and distal tapering.

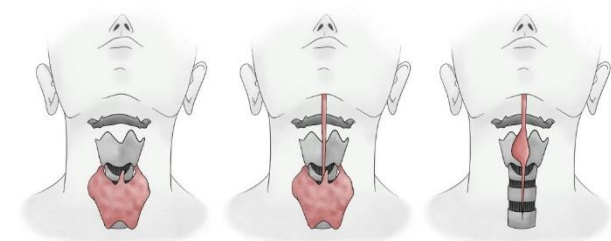


Figure 5: Illustration of normal thyroid anatomy (left), normal anatomy plus thyroglossal duct remnant (middle) and hyperplasia of remnant thyroglossal duct tissue after resection of primary thyroid gland (right).

With the concern of recurrent Graves' disease in remaining thyroid tissue, WU was offered revision total thyroidectomy with possible Sistrunk procedure [5]. A 4 cm incision was made directly over the mass, which was several centimeters above her previous thyroidectomy scar. After splitting the sternohyoid muscles in the midline, the mass was identified and appeared consistent with thyroid tissue. This appeared to track to the level of the hyoid bone but not beyond it, and dissection was then carried from caudal to cephalad, with thyroid tissue identified roughly 3-4 mm below the inferior edge of the hyoid. Deep to the hyoid bone, only a fibrous tract remained, and the hyoid bone was elevated to confirm no remaining thyroid tissue was present thus not requiring a Sistrunk procedure [5]. The tract was suture ligated and marked with a clip. Gross inspection of the resected tissue is displayed in (Figure 4) and illustrated in (Figure 5). Pathology showed benign thyroid tissue with diffuse follicular hyperplasia consistent with Graves' disease. The patient was seen 1-week post-operatively in clinic with a well healing surgical scar and complete resolution of her previous symptoms. She was restarted on 50 mcg daily levothyroxine after her surgery and is to-date clinically asymptomatic.

Discussion

The significance of this case is twofold: it demonstrates the infrequency that events such as this occur, while also highlighting that although total thyroidectomy can potentially be an effective treatment for refractory Graves' disease, surgical management also has its own potential shortcomings. The incidence of recurrent thyrotoxicosis after total thyroidectomy can be estimated to be about 1 per 1,000,000 persons by taking into account the incidence of disease, the percentage who undergo surgery and the estimated incidence of thyroglossal ducts. To date, events such as these have been described only a handful of times in case reports and remain an exceedingly rare event [6-8]. Thyroidectomy as a treatment for Graves' disease does not change the rheumatologic status of the patient. Instead, the approach removes the likely target of the patient's antibodies resulting in a euthyroid state. This mechanism is made apparent when the patient has ectopic or latent thyroid tissue resulting in a failure of total thyroidectomy to cure the disease. The origin of the ectopic thyroid tissue in this patient was likely not regenerated tissue that was retained following the patient's original

thyroidectomy, but rather latent thyroid tissue presents in a thyroglossal duct remnant based on the location of the majority of the tissue in relation to the hyoid bone and surrounding anatomy.

The case presented above demonstrates the importance of informed consent for the rare risk of recurrence and regular surveillance required of Graves' disease patients following the procedure. Physicians should follow their patients with thyroid functions tests as well as review of system screens for deviation from the euthyroid state, which may suggest recurrence of the patients' disease even following surgical resection.

Conflicts of Interest

None.

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