Recurrent thyrotoxicosis following near-total thyroidectomy

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ABSTRACT

Total thyroidectomy for hyperthyroidism is usually curative. We report the unusual recurrence of thyrotoxicosis following a near-total thyroidectomy. The patient, a 27-year-old woman, elected to have a total thyroidectomy and began levothyroxine after the procedure. Approximately 2 years later, recurrent thyrotoxicosis was evident off levothyroxine. Vascularized thyroid tissue was noted on ultrasound, and a radioactive iodine scan indicated increased uptake in the right thyroid region. She began antithyroid medication and was subsequently treated with radioactive iodine once a euthyroid state was achieved. We discuss the implications of this rare scenario—recurrence of thyrotoxicosis after near-total thyroidectomy.

KEYWORDS Graves' disease; recurrence; thyrotoxicosis; total thyroidectomy

Graves' disease, an immune-mediated disorder associated with elevated thyroid-stimulating immunoglobulins (TSIs), is a common cause of hyperthyroidism. Hyperthyroidism is usually treated with medication, radioactive iodine, or surgery. Recent evidence indicates that radioactive iodine treatment may increase the risk of solid tumors¹ and exacerbate Graves' ophthalmopathy. Thyroidectomy may also be considered an accepted therapy during the second trimester of pregnancy.² Additionally, there has been a preference for total thyroidectomy in the treatment of hyperthyroidism. Total thyroidectomy produces better outcomes than subtotal thyroidectomy because of the reduced rate of recurrent thyrotoxicosis.³ As such, total thyroidectomy has been assumed to be curative in hyperthyroidism. In this article, we present the rare case of recurrent thyrotoxicosis following near-total thyroidectomy.

CASE PRESENTATION

A 27-year-old woman presented with a 27-lb unintentional weight loss, palpitations, insomnia, and excessive perspiration. Her past medical history included two pregnancies, asthma, and hidradenitis suppurativa. Vital signs were normal with the exception of elevated systolic blood pressure (157 mm Hg) and an elevated heart rate (107 beats/min). Physical exam findings were unremarkable with the

exception of a diffuse goiter and hyperreflexia. There was no proptosis. The patient had a free T₄ of 5.13 ng/dL and a thyroid-stimulating hormone level of <0.01 mIU/L. The diagnosis of Graves' disease was suspected from the clinical presentation and the TSI of 582% (normal <130%). She deferred a diagnostic thyroid scan and radioiodine treatment and was commenced on atenolol (50 mg daily) and methimazole (20 mg twice daily). After 7 months of poor medication compliance, she returned with symptom exacerbation. At her request, she was referred to an otolaryngologist experienced in thyroid surgery.

Operatively, all thyroid tissue was removed except for a small remnant surrounding the presumed site of the right recurrent laryngeal nerve. The left recurrent laryngeal nerve was not visualized. Within 2 weeks, the patient began thyroid hormone replacement therapy with normalization of thyroid function.

For 2 years, the patient complied with her levothyroxine replacement. Her TSI concentration fell from 438% preoperation to 321% postoperation but began climbing after several months to 408%. Concomitantly, she noted symptoms consistent with thyrotoxicosis. She was weaned off her thyroxine. A free T₄ of 2.13 ng/dL and a thyroid-stimulating hormone level of <0.01 mIU/L were noted along with a TSI of 490%. A thyroid ultrasound revealed two vascular lesions in the right thyroid bed measuring 2.9 × 1.5 × 1.4 cm and

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1.9 × 1.4 × 1.5 cm. A radioactive iodine scan showed increased uptake in the area. The patient elected to have radioactive iodine treatment for recurrent hyperthyroidism. Following 15 mCi radioactive iodine, she developed hypothyroidism and was commenced on thyroxine replacement. A euthyroid state was subsequently achieved.

DISCUSSION

This reoccurrence of thyrotoxicosis in Graves' disease following total thyroidectomy is exceedingly rare. Based on studies from the early 1970s, recurrence of thyrotoxicosis following subtotal thyroidectomies is well documented.⁴ The recurrence of thyrotoxicosis following subtotal thyroidectomy may be the result of latent, persistent disease.⁵ Additionally, recurrent Graves' disease may occur from thyroid tissue in ectopic and aberrant sites. These sites may include the anterior neck,⁶ thyroglossal duct remnants,⁷ mediastinum,⁸ and lateral neck.⁹ Thyroid scintigraphy reveals the active thyroid tissue sites.

In our patient, the recurrent thyroid tissue was identified in the right thyroid bed by ultrasound and radioactive iodine scan. The match between the location of the tissue in the right thyroid bed and the tissue spared on the right side during a total thyroidectomy raises interesting questions. We believe that the right decision was to avoid injury to the recurrent laryngeal nerve by sparing a small amount of tissue in its vicinity. It remains possible that regrowth of this tissue contributed to an overactive thyroid state subsequently. Some thyroid tissue may persist after total thyroidectomies, and perhaps the term near-total thyroidectomy is a more apt description.¹⁰ The surgeon noted that a 0.5 × 0.5 × 0.5 cm piece of tissue was left in place during the surgery, and this is not an uncommon practice. Interestingly, it does not appear to take much thyroid tissue for clinical hyperthyroidism, at least in our patient. It remains to be seen whether an increasing TSI would provide an early warning of such rare recurrent thyrotoxicosis. This case also illustrates the need for ongoing long-term monitoring of patients following intervention for hyperthyroidism.

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1. Kitahara CM, Berrington de Gonzalez A, Bouville A, et al. Association of radioactive iodine treatment with cancer mortality in patients with hyperthyroidism. *JAMA Intern Med.* 2019;179:1034. doi:10.1001/jamainternmed.2019.0981.
2. Genovese BM, Noureldine SI, Gleeson EM, et al. What is the best definitive treatment for Graves' disease? A systematic review of the existing literature. *Ann Surg Oncol.* 2013;20:660–667. doi:10.1245/s10434-012-2606-x.
3. Feroci F, Rettori M, Borrelli A, et al. A systematic review and meta-analysis of total thyroidectomy versus bilateral subtotal thyroidectomy for Graves' disease. *Surgery.* 2014;155:529–540. doi:10.1016/j.surg.2013.10.017.
4. Hedley AJ, Ross IP, Beck JS, et al. Recurrent thyrotoxicosis after subtotal thyroidectomy. *BMJ.* 1971;4:258–261. doi:10.1136/bmj.4.5782.258.
5. Sugino K, Mimura T, Ozaki O, et al. Management of recurrent hyperthyroidism in patients with Graves' disease treated by subtotal thyroidectomy. *J Endocrinol Invest.* 1995;18:415–419. doi:10.1007/BF03349738.
6. Jakibchuk K, Ali S, Samantray J. Recurrence of Graves' disease in ectopic thyroid tissue. *BMJ Case Rep.* 2018;2018. doi:10.1136/bcr-2017-221566.
7. Basili G, Andreini R, Romano N, et al. Recurrence of Graves' disease in thyroglossal duct remnants: relapse after total thyroidectomy. *Thyroid.* 2009;19:1427–1430. doi:10.1089/thy.2009.0143.
8. Cunha FM, Rodrigues E, Oliveira J, et al. Graves' disease in a mediastinal mass presenting after total thyroidectomy for nontoxic multinodular goiter: a case report. *J Med Case Rep.* 2016;10:70. doi:10.1186/s13256-016-0878-7.
9. Winters R, Christian RC, Sofferman R. Thyrotoxicosis due to ectopic lateral thyroid tissue presenting 5 years after total thyroidectomy. *Endocr Pract.* 2011;17:70–73. doi:10.4158/EP10071.CR.
10. D'Andrea V, Cantisani V, Catania A, et al. Thyroid tissue remnants after "total thyroidectomy." *G Chir.* 2009;30:339–344.