Ochsner Journal 16:154–157, 2016

Thyroglossal Duct Remnant with Follicular Hyperplasia Presenting After Total Thyroidectomy

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Background: The thyroglossal duct fails to involute in up to 7% of adults, creating a thyroglossal duct remnant (TGDR) attached to the hyoid bone. Thyroid malignancies have been reported in approximately 1% of TGDRs. In previous reports of TGDR carcinoma, patients had radiographic evidence of a TGDR at initial clinical presentation. Alternatively, hypertrophy of a TGDR is well described in patients with hypothyroidism because of the growth of functional ectopic thyroid tissue. We present the case of a patient who had no radiographic evidence of a TGDR prior to thyroidectomy but presented 14 months after surgery with a recurrent cervical mass.

Case Report: A 58-year-old female underwent total thyroidectomy for micropapillary thyroid cancer. Fourteen months later, she presented with an enlarging cervical mass. She underwent a Sistrunk procedure, and surgical pathology revealed a TGDR with compensatory glandular hypertrophy.

Conclusion: To our knowledge, this is the first report of a TGDR follicular adenoma initially appearing as a result of compensatory thyroid glandular hypertrophy following total thyroidectomy for a micropapillary thyroid carcinoma. Our case presented a novel clinical dilemma regarding the best management for a patient with a new TGDR along with a recent history of micropapillary thyroid cancer.

Keywords: *Adenoma, thyroid cancer–follicular, thyroid cancer–papillary, thyroglossal cyst, thyroid neoplasms*

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INTRODUCTION

During the third week of gestation, the thyroid gland begins to form at the foramen cecum of the dorsal tongue. The ventral diverticulum of the foramen cecum descends along the midline of the ventral neck and through the hyoid bone to form the primitive bilobed thyroid gland. The thyroglossal duct marks the path of this descent and normally disappears by the tenth week of gestation. This tract fails to involute in up to 7% of adults,[1] creating a thyroglossal duct remnant (TGDR) that can be fixed to the hyoid bone.

Although rare, thyroid malignancies have been reported in 1% of TGDRs.[2] Of these cases, approximately 81.7% are papillary and 6.9% are mixed papillary-follicular carcinomas.[3] In most cases, thyroid cancer is successfully diagnosed with fine needle aspiration, and the presence of a TGDR is confirmed by either ultrasound or computed tomography (CT) because the radiographic features of these lesions are well documented.[4]

CASE REPORT

A 58-year-old female was initially referred for surgical management of a suspicious thyroid nodule detected during physical examination. The patient denied compressive symptoms, cold or heat intolerance, weight or appetite changes, and changes in vision. Her family history was significant for papillary thyroid cancer in her mother and 2 sisters. Ultrasound of the neck revealed a 3.0-cm right thyroid lobe nodule with calcifications as well as a 1.7-cm left lobe nodule. Serum laboratory test results showed a thyroid-stimulating hormone (TSH) level of 0.458 mIU/L (reference range 0.4-4.0 mIU/L) and a thyroxine level of 15.0 mcg/dL (reference range 4.8-13.0 mcg/dL). Ultrasound-guided fine needle biopsy of the mass revealed a follicular lesion.

After a discussion of the relevant therapies and because of her extensive family history of thyroid cancer, the patient consented to total thyroidectomy. Surgery was successful and without complications. Surgical pathology revealed a

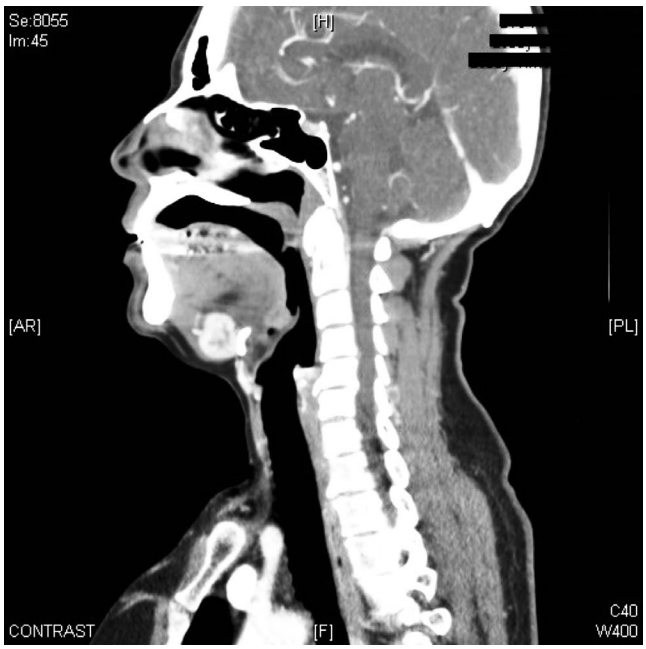

Figure 1. Sagittal computed tomography image demonstrates the thyroglossal duct remnant fixed anteriorly to the hyoid bone at the midline.

follicular variant of micropapillary thyroid cancer of the right nodule. The patient was discharged after an overnight stay and started on 125 mcg levothyroxine.

The patient's TSH suppression proved challenging during the early postoperative period. Her TSH level peaked at 2.320 mIU/L during the 3-month period after surgery, exceeding her target TSH range of 0.1-0.5 mIU/L.

Approximately 14 months after surgery, the patient returned to the clinic with concerns about a painless midline submental mass with xerostomia but no compressive symptoms. Ultrasound of the neck revealed a midline submental mass measuring 2.1 × 2.1 × 1.9 cm with irregular margins and mixed echogenicity. No suspicious lymph nodes were detected in the central or lateral compartments of the neck. She underwent ultrasound-guided fine needle aspiration. Cytopathologic examination demonstrated a follicular lesion of undetermined significance. Serum laboratory tests revealed a TSH level of 0.227 mIU/L. Additional surgery was scheduled to remove the new lesion.

A preoperative CT scan showed that the midline submental mass was fixed to the hyoid bone (Figure 1). The patient underwent a Sistrunk procedure with a level IA selective neck dissection and en bloc removal of a 2.1 × 2.2 × 1.8 cm mass attached to the anterior portion of the hyoid bone (Figure 2). Pathological assessment revealed hyperplasia of thyroid tissue in a TGDR with evidence of compensatory glandular hypertrophy (Figure 3).

DISCUSSION

Papillary thyroid cancer represents 75% of all thyroid malignancies but carries the most favorable prognosis. The histology of these lesions may be purely papillary or mixed papillary-follicular carcinoma.[3] For patients diagnosed with papillary thyroid cancer with nests of cells in both thyroid lobes, the current standard of care is total thyroidectomy. Micropapillary thyroid cancer with no nodal or distant metastases is defined as stage I thyroid cancer. Patients with stage I thyroid cancer who receive total thyroidectomy have 5-year and 10-year survival rates of 99% and 98%, respectively.[5]

Recurrence of a cervical mass after total thyroidectomy is well documented in the literature. Although some masses may be observed clinically, most are detected with ultrasound examination. These lesions may represent recurrence of thyroid cancer, hypertrophy of residual thyroid

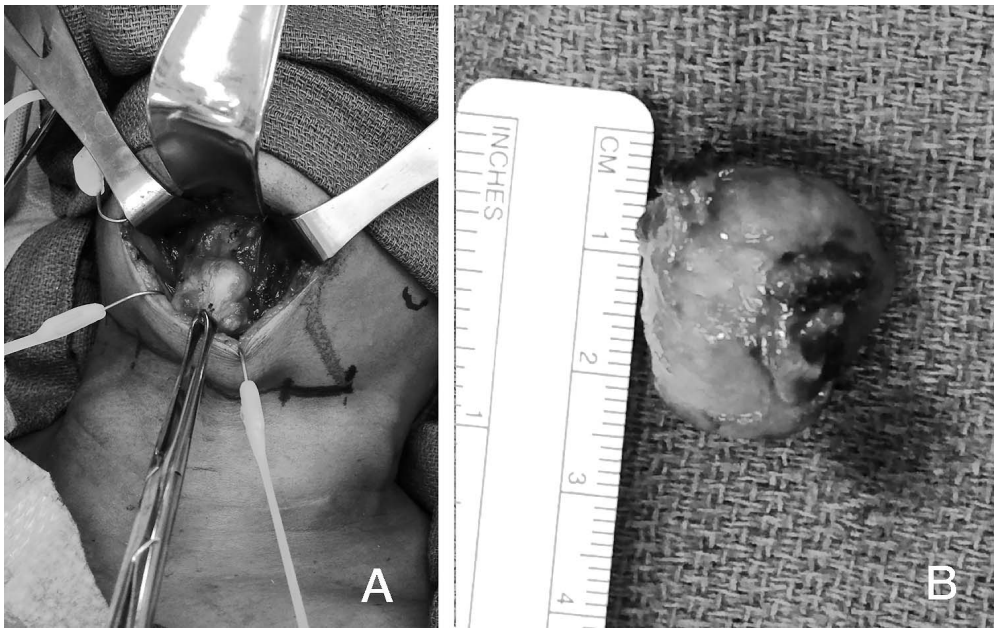

Figure 2. A: A horizontal incision was made and carried down to the level of the thyroglossal duct remnant. B: A 2.1 × 2.2 × 1.8 cm lesion was removed en bloc along with the midportion of the hyoid bone.

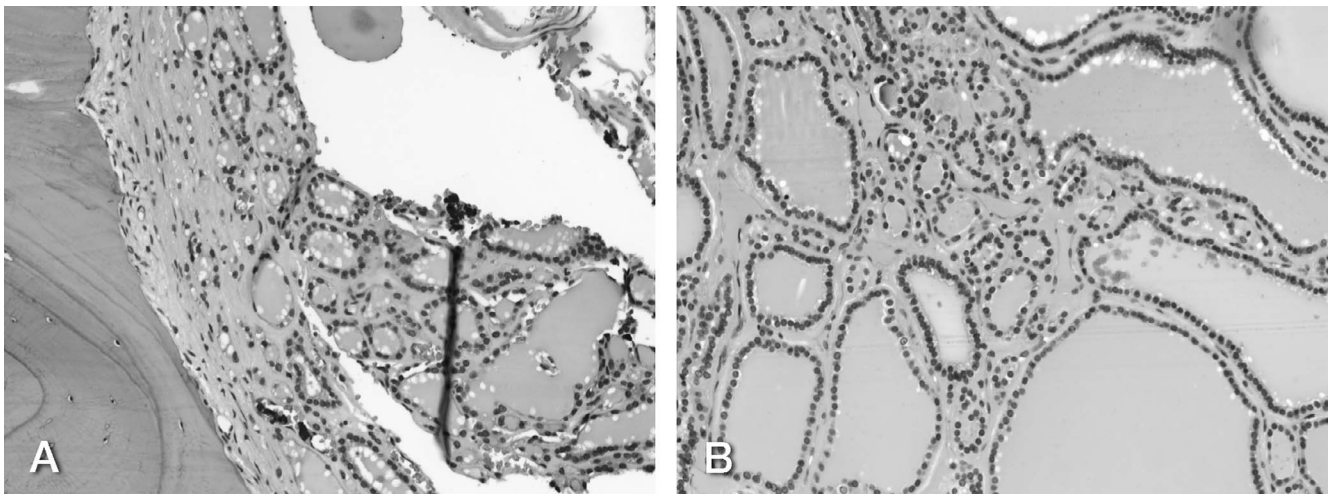

Figure 3. Histologic examination of the midline submental mass shows (A) thyroidal tissue present within the hyoid marrow space and (B) encapsulated mixed micro- and macrofollicular hyperplastic thyroid tissue consistent with compensatory hyperplasia of the thyroglossal duct remnant status post thyroidectomy (hematoxylin and eosin stain, 200×).

tissue, or novel thyroid pathology. Onkendi et al reported that during a span of 9 years, 410 patients at their institution required reoperative procedures for persistent papillary thyroid cancer following total thyroidectomy.[6] Hypertrophy of residual thyroid tissue following total thyroidectomy has also been reported with hypertrophic foci in both the thyroidectomy bed and a thyroglossal duct cyst.[7]

TGDRs are the most common congenital neck abnormality. While most TGDR malignancies are papillary thyroid carcinomas, squamous and anaplastic neoplasms have also been reported.[3]

Cervical ultrasound with fine needle aspiration is the gold standard for diagnosis of TGDR carcinoma. Patients with a positive diagnosis of TGDR carcinoma are stratified as high or low risk, with low-risk patients satisfying the following criteria: age 15-45 years, no history of neck radiation, tumor size $<$4 cm, and no distant or nodal metastases.[5] Low-risk patients should undergo a Sistrunk procedure alone if no thyroid or nodal abnormalities are detected during preoperative workup.[8] In previous reports of TGDR carcinoma, patients had radiographic evidence of a TGDR at initial clinical presentation. Alternatively, hypertrophy of a TGDR has been described in patients with hypothyroidism because of the growth of functional ectopic thyroid tissue.[2,3]

Prior to her total thyroidectomy, our patient had no radiographic evidence of a TGDR. Although her TSH peaked at 2.320 mIU/L during the 3 months immediately following total thyroidectomy, she maintained her goal TSH range of 0.1-0.5 mIU/L for a year afterward. Fourteen months after undergoing a total thyroidectomy, she presented with a recurrent cervical mass. An ultrasound was consistent with a TGDR. Considering the patient's previous diagnosis of micropapillary thyroid cancer, the emergence of an enlarging TGDR after total thyroidectomy carried a significant risk.

This case not only highlights the importance of optimizing the medical management with TSH suppression in micropapillary thyroid carcinoma but also illustrates the difficulty that can arise if surgery results in inadequate resection of all native thyroid tissue, regardless of whether the thyroid remnants are ectopic or eutopic. When dealing with micropapillary thyroid carcinoma, it is paramount to ensure a complete ablation. We believe that the scenario described above strongly points to compensatory thyroid gland hypertrophy.

To our knowledge, this is the first report of a TGDR follicular adenoma first appearing as a result of compensatory glandular hyperplasia following total thyroidectomy for a thyroid gland malignancy. Our team opted to perform a Sistrunk procedure, and physicians should be aware of this potential clinical scenario.

CONCLUSION

Patients with a TGDR could present with an enlarging submental mass in the setting of postoperative hypothyroidism after total thyroidectomy. Careful monitoring and control of postoperative TSH are essential after total thyroidectomy to avoid the need for performing a Sistrunk procedure in such patients.

ACKNOWLEDGMENTS

The authors have no financial or proprietary interest in the subject matter of this article.

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