CASE REPORT

Cutaneous thyroid carcinoma sixteen years after benign total thyroidectomy: a unique case

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Abstract
Background: Cutaneous metastasis of papillary thyroid carcinoma is rare and is a hallmark of a locally aggressive tumor.
Case report: We present a unique case of cutaneous thyroid carcinoma sixteen years after total thyroidectomy for multi-nodular goiter. The tumor originated from the upper anterior thoracic wall's skin and was found to invade the rostral half of the sternum's external periosteum and the caudal part of the right sternocleidomastoid muscle. Wide local excision of the neoplasm was performed with macroscopically free margins and right selective neck dissection (lymph node levels IV and V). The skin deficit was reconstructed with a right pectoralis major island flap. The histopathologic findings displayed a papillary thyroid carcinoma with Hürthle cell predominance and microscopically positive margins at the excised portion of the sternocleidomastoid muscle. The patient was placed on close follow-up, and nine months postop, there are no clinical signs of recurrence.
Conclusion: To our knowledge, this is the first case of cutaneous thyroid carcinoma following benign thyroidectomy. A possible mechanism for this incidence is the malignant transformation of benign thyroid cells inoculated into the skin. Dissecting along the fascial planes during thyroidectomy and preserving the thyroid capsule establishes a clean surgical field and minimizes the chance of inoculation of thyroid cells into the adjacent structures. Close follow-up and a high level of suspicion for skin lesions in patients with thyroid disease are warranted in all cases. HIPPOKRATIA 2020, 24(2): 88-90.

Keywords: Thyroid, malignancy, metastasis, skin, thyroidectomy

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Introduction

Thyroid cancer usually displays a benign clinical course but may also give rise to local, regional, or distant metastases[1]. The most frequent metastatic sites are lymph nodes, lungs, and bones via lymphatic or hematogenous routes. Metastasis to the skin is extremely rare, accounting for 0.7-2 % of all cutaneous malignant neoplasms, and carries a poor prognosis[1,2]. Malignant transformation of small thyroid remnants after benign thyroidectomy has also been reported[3]. Finally, iatrogenic inoculation of thyroid cells has also been proposed as a mechanism of thyroid cancer's dissemination to the skin or other adjacent structures[2,4]. We present a unique case of cutaneous thyroid carcinoma sixteen years after total thyroidectomy for multi-nodular goiter.

Case report

A 79-year-old Caucasian male was referred to our institution with a sizeable, non-ulcerated, non-tender, fast-growing lesion of the upper anterior thoracic wall (Figure 1). He had a medical history of type 2 diabetes mellitus under oral treatment, trigeminal neuralgia, hypertension, chronic obstructive pulmonary disease treated with inhaled bronchodilators and corticosteroids, and gastroesophageal reflux disease under proton pump inhibitors. No family history of thyroid malignancies was reported. The patient had a history of total thyroidectomy for multi-nodular goiter seventeen years before his presentation in our institution. The pathology report described a multi-nodular goiter with microfollicular adenomatous and colloid thyroid nodules, without thyroid malignancy findings. Postoperatively the patient was placed under hormone replacement therapy with levothyroxine (0.1 mg/day).

One year before his presentation, the patient self-referred to a general surgeon complaining of a skin lesion <1 cm below the right medial clavicular area. A punch biopsy of the lesion was performed under local anesthesia, and the pathology showed a papillary thyroid carcinoma (PTC). He was then referred to an Endocrinologist and was treated with a 150 mCi dose of radioactive iodine (RAI) at the local tertiary referral center. We have no access to thyroglobulin levels before and post-treatment. Due to no response to RAI treatment and disease progression, he was referred

to our institution for surgical excision of the lesion.

Computerized tomography revealed a midline soft tissue mass, 5.2 cm in maximum diameter, extending from the sternum's manubrium to the jugular notch (Figure 2). Additionally, an ultrasound scan identified a suspicious right level IV lymph node and a small tissue residue in the ipsilateral thyroid bed. Fine needle aspiration cytology (FNAC) of the thyroid residue had benign findings (Bethesda Category II), while FNAC of the lymph node revealed infiltration by PTC.

Under general anesthesia, wide local excision of the tumor was performed in macroscopically healthy-appearing tissue. During deep plane dissection, the neoplasm was found to invade the periosteum of the sternum's manubrium and the sternal and clavicular heads of the right sternocleidomastoid muscle (SCM). Elevation and removal of the affected periosteum area and excision of the right SCM's infiltrated part achieved macroscopically free margins (Figure 3). A right selective neck dissection (levels IV and V) was also performed through the same incision. The anterior neck and thoracic area's skin defect was reconstructed with a right pectoralis major island myocutaneous pedicled flap (Figure 4). Drains were placed, the wound was sutured in layers, and the patient had an uneventful postoperative course. He was discharged on the third postoperative day in good condition.

Microscopically, the tumor was identified as a metastatic PTC with Hürthle cell predominance. The neoplasm infiltrated the full thickness of the cutis and subcutis, sparing the epidermis. Also, microscopically positive margins were noted at the SCM excision border of the biopsy specimen. Immunohistochemically, the tumor cells were positive for thyroglobulin and TTF-1. Two of the nine excised lymph nodes were infiltrated by the neoplasm with extranodal extension. The Oncology Board offered re-operation with a wider excision of the tumor, but the patient declined. On his nine-month postoperative follow-up, there were no clinical signs of recurrence (Figure 5). Two months before his last follow-up, a whole-body ^{131}I scintigraphy did not reveal any radionuclide uptake in the thyroid bed or other parts of the body. He has opted for close follow-up by the treating Endocrinologist at the tertiary regional referral center and complementary RAI treatment, depending on thyroglobulin levels.

Discussion

Thyroidectomy is the standard-of-care for many thyroid diseases. Indications include benign or malignant neoplasms, autoimmune thyroiditis, medically refractory hyperthyroidism, symptomatic goiters causing dysphagia or dyspnea, and thyromegaly with significant cosmetic compromise.

Differentiated thyroid cancer (DTC) is the most common endocrine gland malignancy and accounts for the vast majority of thyroid cancers. PTC accounts for nearly 85 % of the cases, and follicular thyroid carcinoma (FTC) for about 12 %, including their Hürthle cell variants. Overall, DTC carries an excellent prognosis and very low disease-specific mortality[5].

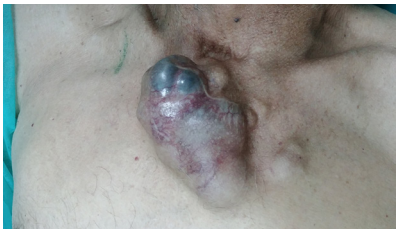

Figure 1: Preoperative clinical image showing a sizeable lesion of the anterior thoracic wall in a 79-year-old male patient.

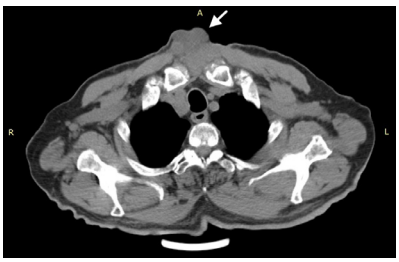

Figure 2: Axial computed tomography image (without contrast medium due to thyropathy) showing a mass at the area of the anterior thoracic wall and jugular notch (white arrow).

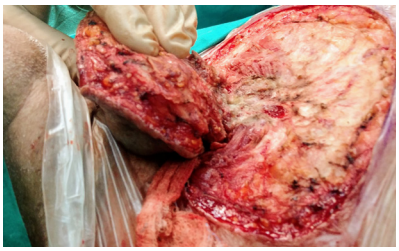

Figure 3: Intraoperative image demonstrating a wide local excision of the tumor with macroscopically free margins.

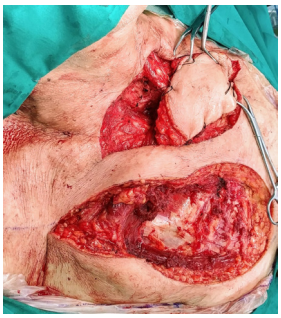

Figure 4: Intraoperative image showing the skin defect reconstruction with a right pectoralis major island myocutaneous pedicled flap.

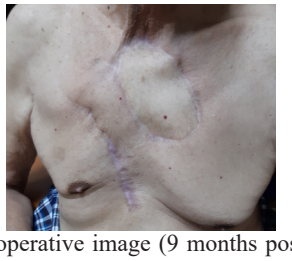

Figure 5: Postoperative image (9 months postop) showing excellent surgical wound healing and no clinical signs of local recurrence.

The follicular variant of papillary thyroid carcinoma (FVPTC) may be easily misclassified as follicular adenoma in up to 70 % of the cases, leading to progressive disseminated DTC. Moreover, FVPTC and FTC show a high RAS mutation rate compared to PTC where BRAF mutations usually predominate. On this basis, long-term measurements of serum thyroglobulin are suggested[6].

It has been proposed that Hürthle cell carcinoma (HCC), accounting for up to 4 % of all thyroid cancers, is genetically distinct from PTC and FTC. In comparison, it carries a higher risk of invasion and metastatic potential along with a poorer prognosis. Due to the low iodine uptake rate, it is less sensitive to RAI ablation. While no consensus exists on the optimal treatment method for HCC, surgery remains the most effective therapeutic modality for this disease[6,7]. FNAC specificity for HCC detection is low. Nevertheless, it should be noted that while Hürthle cell change in FNAC is considered an atypical feature by many clinicians, it does not increase the malignancy rate beyond the one estimated by the Bethesda Classification categories[8].

RAI ablation is the current therapeutic modality of choice for microscopic residual disease following total thyroidectomy for DTC[5]. Treatments outcomes are evaluated by assessing the structural and biochemical responses[5]. Microscopically positive excision margins are not considered a risk for recurrence[5]. On the other hand, gross residual disease is not amenable to RAI ablation and is associated with poorer treatment outcomes. Additionally, there is a significant correlation between microscopically positive margins and incomplete response to ^{131}I. Also, involved posterior margins are associated with lower response rates[9].

Cutaneous metastasis of PTC is rare and is a hallmark of a locally aggressive tumor. Usually, it is the first clinical sign in occult thyroid cancer cases, and the most common site reported is the scalp[1,2]. FTC is associated with a higher overall incidence of metastasis, although skin is infrequently involved[1,10]. Postoperative development of such lesions may be indicative of treatment failure or tumor recurrence[1]. Also, residual thyroid tissue following total benign thyroidectomy is not uncommon, and malignancies arising from such remnants are not impossible[3]. It has been proposed that cutaneous metastasis of thyroid cancer may happen intra-operatively due to thyroid nodule or thyroid capsule rupture and contamination of the skin[2]. Another possible mechanism is the dissemination of malignant thyroid cells to adjacent structures during needle aspiration biopsy[4]. Dissecting along the fascial planes and preserving the thyroid capsule establishes a clean surgical field and minimizes the chance of inoculation of thyroid cells into adjacent structures. Also, close follow-up and a high level of suspicion for skin lesions in patients with thyroid disease are warranted in all cases.

In the reported case, no malignant features were identified in the initial thyroidectomy specimen. The operative report did not comment on tissue re-implantation (e.g., accidental removal of a parathyroid gland). Moreover, FNAC of the residue in the thyroid bed was compatible with normal thyroid tissue remnants. Whole-body ^{131}I scintigraphy did not reveal any radionuclide uptake. Finally, the extremely long time (sixteen years) between the initial operation and the skin lesion's appearance suggests that malignant transformation happened postoperatively. On this basis, a possible mechanism would be intra-operative inoculation of benign thyroid cells to the skin, which then developed malignant features. To our knowledge, this is the first report of such a case.

Conflicts of interest

Authors declare no conflict of interest.

Acknowledgments

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