

Recurrent Papillary Thyroid Carcinoma with Lymph Node Metastases Involving Skin and Blood Vessels

Abstract

We present a clinical case of a 73-year-old female with a history of papillary thyroid carcinoma (PTC) since 1982 when thyroidectomy and adjuvant radioiodine (radioactive iodine [RAI]) treatment were performed. The patient was lost to follow-up until 2007 when a recurrence/persistence of the same PTC was present. The second operation and RAI treatment were performed. Around 2015, she noticed a lump in the same region, and in 2016, SPEC-CT revealed 131-I uptake in the left cervical lymph metastases with massive skin invasion. In May 2017, two skin ulcerations on the left neck and jugulum due to skin infiltration and tumor necrosis were present. Fine-needle biopsy proved infiltration by papillary carcinoma without dedifferentiation. Ultrasound examination revealed a massive tumor invasion of the internal jugular vein with tumor thrombosis. This case emphasizes the importance of methodical follow-up as recommended for early detection and adequate treatment of recurrent and/or persistent disease till the end of the patient's life.

Keywords: Local metastases, papillary carcinoma, recurrence

Introduction

Papillary carcinoma/papillary thyroid carcinoma (PTC) is the most common malignancy of the thyroid gland (85% of the differentiated cancers). The 10-year survival rate is above 90% and depends on different risk factors. Adequate surgery is the first-line treatment.^[1] Postoperative radioiodine remnant ablation eliminates postsurgical thyroid remnant, thus facilitating an early detection of recurrence or persistence, together with serum thyroglobulin measurement and/or radioiodine whole-body scan. The detection of disease recurrence is a major goal of long-term follow-up after obtaining tumor control. The follow-up algorithm was based mainly on cervical ultrasound at 6–12 months to evaluate thyroid bed and central and lateral cervical nodal compartments and to exclude morphological recurrence. Serum thyroglobulin together with negative thyroglobulin antibody (Tgab) is the part of follow-up algorithm tested every 6–12 months to exclude biochemical recurrence.^[1]

Life span in low and intermediate group of radically treated patients is similar to that of the general population. In patients with recurrent/persistent disease, overall survival drops to 50%–60%.^[2]

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The present case report demonstrates how a patient with initially curable pT2pN1bM0G1 papillary thyroid carcinoma could develop a large tumor burden, rarely seen in our routine practice, when lost from proper follow-up. Moreover, vessel tumor thrombosis seen in our patient is rare, and <30 cases were reported in the literature.

Case Report

We present the case of a 73-year-old female patient with a history of pT2N1bMo G1 papillary thyroid carcinoma since 1982, when total thyroidectomy was performed, followed by radioiodine ablation (RIA) with 3 GBq (80 mCi). After that the patient disappeared from follow-up. In 2007, she appeared with a left lymph node involvement and had a salvage neck dissection and second radioiodine therapy (RIT) with 3.7 GBq (100 mCi). Again, the patient refused medical follow-up. In 2015, the mass on the left neck appeared again. This time the patient started alternative medicine locally to avoid new surgery. The tumor was growing, and in 2016, 131-I single-photon emission computed tomography (SPECT-CT) was performed that revealed left laterocervical, supraclavicular, jugular, and upper mediastinum lymph metastases [Figure 1]. No uptake in the lungs and bones

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was detected. Serum thyroglobulin on thyroid-stimulating hormone (TSH) stimulation (19.31 mU/L) was >500 ng/ml. This time the patient was inoperable, with blood vessel and skin involvement; therefore, only palliative RIT with 4,8GBq (130 mCi) was offered in November 2016. Several weeks after RIT, an ulceration on the left side of the neck appeared [Figure 2] and few months later a second one in the jugulum [Figure 3]. Her serum thyroglobulin on suppression was 41 ng/ml (TSH – 0.011 mU/L) with negative Tgab.

On admission, in the clinic of endocrinology, a large mass on the left side of the neck was palpated and two bleeding ulcerations on the skin were found [Figures 2 and 3]. Laboratory tests revealed suppressed TSH levels (<0.005 mU/l), elevated FT4 (40.1 pmol/l), microcytic hypochromic anemia (Hg – 82 G/l; red blood cell– 3.1 T/l; and Hct – 0.27), normal white blood cell and platelet count (WBC – 5.4 G/l and PLT – 264 G/l), and increased erythrocyte sedimentation rate (ESR – 37 mm/h). On ultrasound examination, the following formations were seen: (1) in the left side of thyroid bed sized 33 mm × 86 mm × 32 mm [Figure 4], (2) laterally of the carotid artery and jugular vein sized 30/23 mm, and (3) in the jugular fossa sized 43 mm × 47 mm × 52 mm. Dorsally of the formation in the left lobe four lymph nodes were detected, sized respectively 5/6.8, 6/6, 7/9, and 7/11 mm. Dorsally of the lymph nodes, on transverse section of the internal jugular vein a propagating from the ventral wall hyperechogenic formation was seen, probably organized thrombus. The vessel wall was visibly invaded from the nearby pathological lymph nodes. The skin above the masses was not clearly visible at some places.

CT scan demonstrated advanced local disease [Figure 5]. There were no signs of infiltrative or nodular changes in the lungs, pleural, and pericardial effusions; there was no evidence of bone changes, suspicious for secondary dissemination. The fine-needle biopsy revealed infiltration with differentiated papillary carcinoma (Bethesda IV).

The patient was considered inoperable by a surgeon and not a good candidate for RIT or external beam radiation therapy (EBRT) by a radiation oncologist. Skin ulcerations were assessed as skin invasion with tumor necrosis after the last RIT. Bleeding and the internal jugular vein involvement were contraindication for tyrosine kinase inhibitor due to the high risk of life-threatening bleeding. The patient was put on palliative care and symptomatic therapy.

Discussion

PTC is a common endocrine tumor with a very good prognosis and a high survival rate. A large study with 900 patients with papillary carcinoma showed that 20- and 40-year tumor recurrence rates were only 6% and 8%, respectively, despite multifocality (23%) and lymph node involvement (4%–31%).^[2] According to the literature, significant risk factors for recurrence or persistent disease

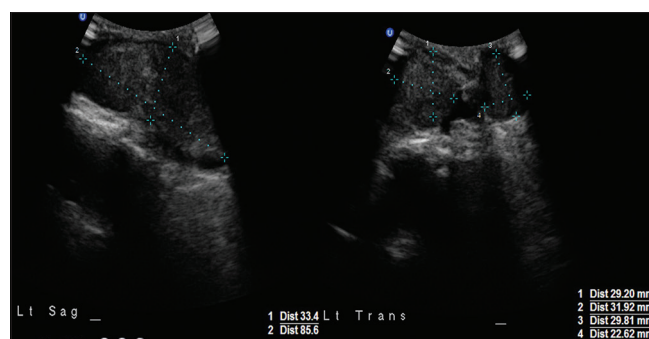


Figure 1: Ultrasound examination revealing a formation in the left side of thyroid bed sized 33 mm × 86 mm × 32 mm

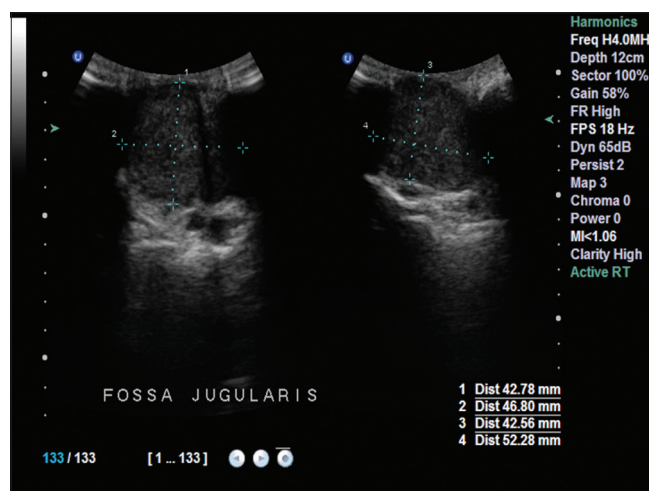


Figure 2: Ultrasound examination revealing a formation in the jugular fossa sized 43 mm × 47 mm × 52 mm

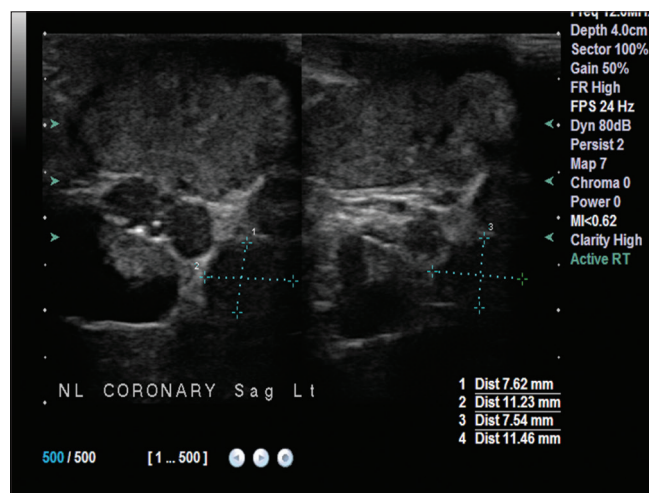


Figure 3: Ultrasound examination revealing four lymph nodes were detected, sized respectively 5/6.8, 6/6, 7/9, and 7/11 mm dorsally of the formation in the left lobe

include the numbers of lymph node metastases (>5), LN metastases with extracapsular extension, tumor size (>4cm), and lymph node metastases in the central compartment.^[3,4] The relapse reported in our patient in 2007 was detected 25 years after the initial diagnosis and treatment, so we could consider it as a true recurrence rather than persistence.

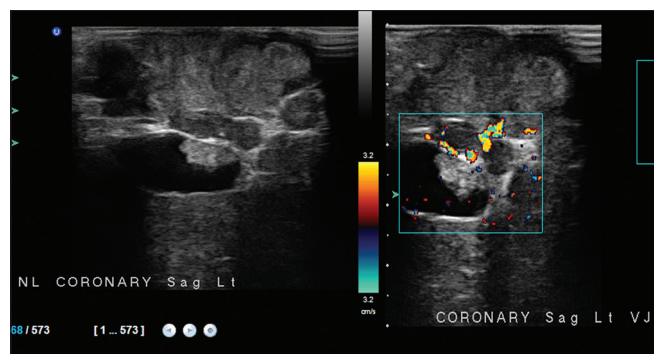


Figure 4: Ultrasound examination revealing transverse section of the internal jugular vein where a propagating from the ventral wall hyperechogenic formation was seen

We consider the case as not been controlled because of inadequate patient behavior, permanently avoiding follow-up.

Skin metastases from differentiated PTC as an appearance of distant disease are quite rare. In few described cases in literature, skin lesions were necrotic soft-tissue metastases or skin invasion by lymph node metastases.^[5] In our patient, the skin ulcerations were not evident until the third RIT. Vessel tumor thrombosis is rare, and <30 cases were reported in the literature.^[6] Our patient has evidence of the internal jugular vein involvement with a probably tumor thrombus inside, but without obstruction symptoms.

Radioiodine is an effective treatment option both as an adjuvant therapy in high-risk patients and also in patients with persistent and/or recurrent disease \pm distant metastases. After some time, a decreased or total loss of RIT effectiveness could happen, mainly because of dedifferentiation of long-lasting persistent disease.^[7,8] The persistent tumor in our case was still retaining radioiodine, but because of the risk of future necrotic effect and fatal hemorrhage RIT was not performed. For the same reason she was assessed as contraindicates for tyrosine kinase inhibitors as well.^[9] The patient was left to symptomatic and palliative nononcological care.

Conclusions

This case report reveals that a patient with initial curable pT2pN1bMo G1 papillary thyroid cancer, being lost from proper follow-up and neglecting seriousness of the disease, could develop a huge local tumor burden, rarely seen in our routine practice. Such advanced local tumor burden with skin necrosis and blood vessel involvement is a contraindication for salvage surgery, EBRT, and target therapy and condemns the patient to poor quality of life till the end.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.



Figure 5: Computed tomography scan of the neck and upper chest

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Conflicts of interest

There are no conflicts of interest.

References

- Haugen BR, Alexander EK, Bible KC, Doherty GM, Mandel SJ, Nikiforov YE, *et al.* 2015 American thyroid association management guidelines for adult patients with thyroid nodules and differentiated thyroid cancer: The American thyroid association guidelines task force on thyroid nodules and differentiated thyroid cancer. *Thyroid* 2016;26:1-33.
- Hay ID, Hutchinson ME, Gonzalez-Losada T, McIver B, Reinalda ME, Grant CS, *et al.* Papillary thyroid microcarcinoma: A study of 900 cases observed in a 60-year period. *Surgery* 2008;144:980-7.
- Links TP, van Tol KM, Jager PL, Plukker JT, Piers DA, Boezen HM, *et al.* Life expectancy in differentiated thyroid cancer: A novel approach to survival analysis. *Endocr Relat Cancer* 2005;12:273-80.
- Tuttle RM, Tala H, Shah J, Leboeuf R, Ghossein R, Gonen M, *et al.* Estimating risk of recurrence in differentiated thyroid cancer after total thyroidectomy and radioactive iodine remnant ablation: Using response to therapy variables to modify the initial risk estimates predicted by the new American Thyroid Association staging system. *Thyroid* 2010;20:1341-9.
- Krishnamurthy A, Ramshankar V. Ulcerative cutaneous nodule as a presenting feature of recurrent thyroid carcinoma. *Indian J Dermatol* 2014;59:616-8.
- Hyer SL, Dandekar P, Newbold K, Haq M, Wechalakar K, Thway K, *et al.* Thyroid cancer causing obstruction of the great veins in the neck. *World J Surg Oncol* 2008;6:36.
- Durante C, Haddy N, Baudin E, Lebouilleux S, Hartl D, Travagli JP, *et al.* Long-term outcome of 444 patients with distant metastases from papillary and follicular thyroid carcinoma: Benefits and limits of radioiodine therapy. *J Clin Endocrinol Metab* 2006;91:2892-9.
- Tuttle RM, Sabra M. S2-Defining RAI Refractory Thyroid Cancer: When is RAI Therapy Unlikely to Achieve a Therapeutic Response? *Thyroid Disease Manager*. Available from: <http://www.thyroidmanager.org/chapter/s2-defining-rai-refractory-thyroid-cancer-when-is-rai-therapy-unlikely-to-achieve-a-therapeutic-response/>. [Last accessed on 2017 Jun 04].
- Lorusso L, Pieruzzi L, Biagini A, Sabini E, Valerio L, Giani C, *et al.* Lenvatinib and other tyrosine kinase inhibitors for the treatment of radioiodine refractory, advanced, and progressive thyroid cancer. *Oncotargets Ther* 2016;9:6467-77.