Papillary Carcinoma on Intranodal Thyroid Ectopy

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Abstract

Thyroid ectopy is a rare pathology associated with a failure of the thyroid gland to migrate during embryonic development. It is characterized by the persistence of a thyroid remnant along the thyroglossal tract. We report a case of papillary carcinoma with lymph node metastasis developed on intranodal thyroid ectopy.
This is a 27-year-old patient who underwent excision of a right laterocervical adenopathy, the histopathological examination of which revealed a papillary carcinoma of thyroid origin (PTC). Total thyroidectomy with lymph node dissection revealed normal thyroid parenchyma and upper mediastinal lymph node metastasis.
The originality of this observation lies in the exceptional nature of this intranodal localization of thyroid tissue transformed into metastatic papillary thyroid carcinoma with normal thyroid parenchyma.

Keywords: thyroid ectopy, papillary carcinoma, lymph node metastasis.

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Introduction

Thyroid ectopy is an ectopic localization of the thyroid, due to the persistence of a thyroid remnant along the thyroglossal tract. The development of a carcinoma from this ectopic tissue is rare, and in the extreme majority of cases it is a papillary carcinoma. More exceptionally, ectopic thyroid tissue may be identified intranodally [1]. The development of papillary carcinoma from a lymph node origin is rare. Such clinicopathological presentation poses a diagnostic trap at the time of extemporaneous examination, leading to discussion of intranodal metastasis of a primary papillary thyroid carcinoma. We report a case of papillary carcinoma with lymph node metastasis developed on intranodal thyroid ectopy.

Case presentation

A 27-year-old patient with no history of childhood irradiation or family goiter, and no specific medical or surgical history, presented with a progressively enlarging right laterocervical adenopathy two years previously. He was referred to an otorhinolaryngology department for lymph node resection. Histopathological examination revealed the lymph node location of a papillary carcinoma of thyroid origin measuring 8 mm in the long axis without any positive signs of massive or significant lymph node involvement. A total thyroidectomy with extended lymph node dissection was performed after a cervical ultrasound showing a normal thyroid gland. Histopathological examination revealed normal thyroid parenchyma with one positive lymph node out of forty-nine (1N+/49). The positive lymph node was located in the upper mediastinal region and measured 2 mm long without any positive signs of massive or significant lymph node involvement. A differentiated thyroid carcinoma classified as pT(x)N(1b)M(x) with intermediate lymph node involvement was selected. Given this situation, we put forward two diagnostic hypotheses: either differentiated thyroid cancer on thyroid ectopy with cervical lymph node metastasis, or intra-lymph node thyroid ectopy transformed into papillary thyroid

carcinoma with cervical lymph node metastasis.
He underwent a 3.7GBq radioiodine therapy course for diagnostic and therapeutic purposes six weeks after total thyroidectomy. Biological assessment prior to iratherapy performed after hormone withdrawal revealed a well-stimulated ultrasensitive Thyroid Stimulating Hormone (TSH us) of 229 mIU/l; tumor marker assays of 1.97 ng/ml for thyroglobulin (Tg) and negative anti-thyroglobulin antibodies (anti-Tg Ab) respectively. Whole-body I-131 scintigraphy performed after the I-131 radioiodine therapy and

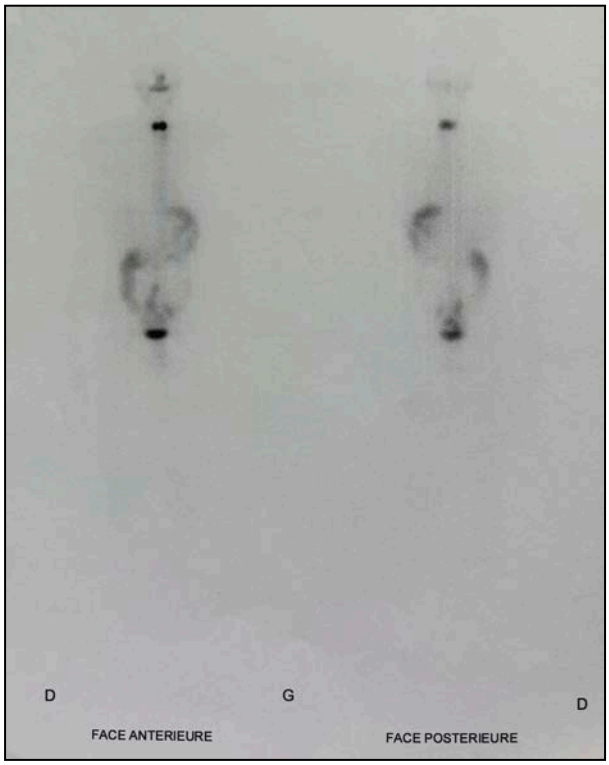

Figure 1: Whole-body I-131 scintigraphy showing a single focus of radioiodine uptake related to one thyroid residue with no other distant pathological foci.

hospitalization in a radioprotected room revealed a single focus of iodine fixation opposite the thyroid lodge (Figure 1), related to one thyroid residue (Figure 2), with no other distant pathological foci of fixation. The diagnosis of intranodal thyroid ectopy transformed into papillary thyroid carcinoma with upper mediastinal lymph node metastasis was finally accepted. Progression at 6 months post I-131 radioiodine therapy was favorable, enabling the patient to be declared "cured" on satisfactory morphofunctional and biological criteria. He is currently under clinicobiological surveillance for life.

Discussion

The thyroid gland appears after the second week of gestation, as an epithelial proliferation of the endodermal lining in the median part of the floor of the primitive pharynx, which later becomes the "foramen caecum". It progressively follows a caudal and ventral direction, along the thyroglossal tract. By the seventh week, it reaches its final position below the thyroid cartilage, opposite the 5th and 6th tracheal rings. Any failure in this migration can lead to thyroid ectopy, the pathogenesis of which remains poorly understood [2]. Sublingual thyroid ectopy remains

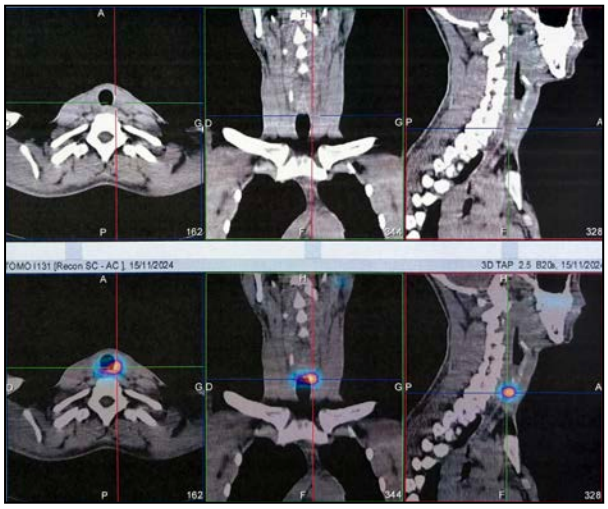

Figure 2: Cervico-mediastinal SPECT CT showing a single focus of radioiodine uptake related to one thyroid residue.

the most common form, occurring in 70-90% of cases[3,4]. Ectopic locations may be submandibular, tracheal, lateral cervical, carotid, esophageal, gastric, duodenal, pancreatic, mesenteric, intracardiac, aortic, pulmonary, pituitary, axillary, and in the iris of the eye. It is exceptionally intranodal.
The frequency of thyroid ectopia is estimated at between 1/100,000 and 1/300,000, and affects more women than men, with a sex ratio of 1 in 4 [5]. Progression of thyroid ectopy to malignant degeneration is rare and is classically observed in the median cervical region or in residues of the thyroglossal tract [6]. Only a few cases of thyroid ectopia transformed into papillary carcinoma have been reported in the literature [1,7]. The most common histological type is papillary carcinoma [1]. The situation of intra-nodal thyroid ectopy transformed into papillary thyroid carcinoma with lymph node metastasis, as in the case we report, raises the problem of primary and metastatic adenopathy. Refer-

ring to the migratory path during thyroid organogenesis, the primary origin of the laterocervical adenopathy is likely, and that of the upper mediastinal adenopathy (cervical area VII), a metastasis. The possibility of double thyroid ectopy within the lymph node remains unlikely, given that the two lymph nodes do not belong to the same lymph node chain; the jugulocarotid chain being high up and the upper mediastinal chain lower down.

After initial excision of a cervical adenopathy indicative of differentiated thyroid cancer, total thyroidectomy and lymph node dissection with histopathological examination remain the standard of care. Additional isotopic management as adjuvant treatment is essential, even in the presence of normal thyroid parenchyma, because in addition to its therapeutic effect, it also enables us to map any local or distant thyroid ectopia. In the case reported, a single 3.7GBq radioiodine therapy course resulted in “cure" on satisfactory morphofunctional and biological criteria. He was put on L-thyroxine at a suppressive dose with a target of 0. 1< TSH < 0.5 as he was classified as being at intermediate risk of tumor recurrence according to the 7th edition of the UICC/AJCC classification. He is monitored clinically, biologically and, if necessary, by ultrasound for the rest of his life. This case illustrates a very specific situation of an intranodal thyroid ectopy transformed into papillary carcinoma with lymph node metastasis. Cervical ultrasonography in favor of a normal thyroid prior to total thyroidectomy, histopathological examination in favor of an essentially normal thyroid, and a whole-body I-131 scan, which did not reveal any iodine fixation sites of distant pathological localization, ruled out the possibility of occult thyroid carcinoma and other types of thyroid ectopy.

Conclusion

The occurrence of a differentiated thyroid cancer on ectopic thyroid is exceptional. The most common histological type is papillary thyroid carcinoma. The case reported illustrates a rare case of intranodal thyroid ectopy transformed into papillary thyroid carcinoma with lymph node metastasis, which responded perfectly to adjuvant radioisotopic treatment. I-131 radioiodine therapy was used for both diagnostic and therapeutic purposes, and as a complement to histological examination, eliminated any remaining diagnostic ambiguity.

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Declarations

Consent for publication: The author clarifies that written informed consent was obtained and the anonymity of the patient was ensured. This study submitted to Swiss J. Rad. Nucl. Med. has been conducted in accordance with the Declaration of Helsinki and according to requirements of all applicable local and international standards. All authors contributed to the conception and design of the manuscript, participated in drafting and revising the content critically for important intellectual input, and approved the final version for publication. Each author agrees to be accountable for all aspects of the work, ensuring its accuracy and integrity.

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