

# Lymphnode metastasis of thyroid cancer misinterpreted as lateral aberrant thyroid 40 years before identification of primary tumor. Case report and review of the literature

G. RIVA<sup>1</sup>, M. VILLANOVA<sup>1</sup>, G. FRANCIA<sup>2</sup>, G. VALOTTO<sup>1</sup>, L. MEZZETTO<sup>1</sup>, M. TOAIARI<sup>2</sup>, A. ECCHER<sup>1</sup>, L. NOVELLI<sup>3</sup>

<sup>1</sup> Department of Pathology and Diagnostic, University and Hospital Trust of Verona, Italy; <sup>2</sup> Department of Endocrinology, Pederzoli Hospital, Peschiera del Garda (VR), Italy; <sup>3</sup> Department of Pathology and Diagnostic, Careggi University Hospital, Firenze, Italy

## Key words

Aberrant thyroid • Occult thyroid carcinoma • Ectopic thyroid

## Summary

The differential diagnosis between lateral ectopic thyroid tissue with orthotopic normal gland and metastatic thyroid carcinoma is challenging. Lateral cervical site is a very rare location for ectopic tissue since only a few cases have been reported. The peculiarity of this clinical case is the finding of a thyroid carcinoma forty years after surgical resection of the ectopic thyroid lesion. This asynchronous association, never reported in literature, raises the question of the differential diagnosis between a true ectopic aber-

rant thyroid and an early lymph node metastasis from an occult thyroid carcinoma, evident in the primitive site many years later. Several elements, which will be matter of discussion, seem to favour the latter hypothesis.

This case, although isolated, suggests that any lateral cervical mass, comprising thyroid tissue, should be regarded as a metastasis of thyroid carcinoma until proven otherwise. Carefull investigation of thyroid gland is mandatory.

## Case report

Ectopic thyroid was described for the first time by Heller in 1749. The prevalence is about 1 case per 100.000-300.00 people. This rare condition is characterized by the presence of thyroid tissue outside the usual location of the normal thyroid, mostly in the pretracheal position, without anatomical and vascular connection with the gland. Lateral cervical site is a very rare location for ectopic tissue to occur, since only few sporadic cases have been reported in literature. It is so rare and for this its presence as a nosological entity is questionable. On the other hand, according to literature, most cases of regional neck lesions associated to a clinically normal thyroid gland at first evaluation, should be considered to be metastases of occult thyroid carcinoma. Our case provides the opportunity to discuss this topic.

We report the case of an 74-year-old man, who presented, in 2014, with a left lateral-cervical mass that he noticed two months earlier. He was affected by non Hodgkin B

lymphoma, diagnosed in 2010 and since then in follow-up. His past medical history revealed that in November 1974 he underwent to an excision of a 3-4 cm left lateral-cervical lesion. The surgical examination of the mass did not reveal any anatomical connection with normal thyroid and the lesion appeared firmly adherent to internal jugular vein, connected by small and short vessels. The carotid artery and the vagus nerve were not involved. Pathological examination revealed the presence of thyroid tissue composed of normal hyperplastic cells in a follicular pattern, scanty colloid and massive infiltration of lymphocytes, often arranged in lymphoid follicles. On the basis of these findings the final diagnosis was “ectopic aberrant lateral-cervical thyroid with basedowian histological appearance”. At that time physical examination of the thyroid gland was normal and patient did not show any symptom of thyroid dysfunction. Neither endocrine assessment nor imaging study were carried out. In the following years no clinical and/or instrumental follow-up was performed. Forty years later, in 2014, a

## Correspondence

Manuela Villanova, Department of Pathology and Diagnostic, University and Hospital Trust of Verona, A. Stefani square 1, 37126 Verona - E-mail: manvilla86@libero.it

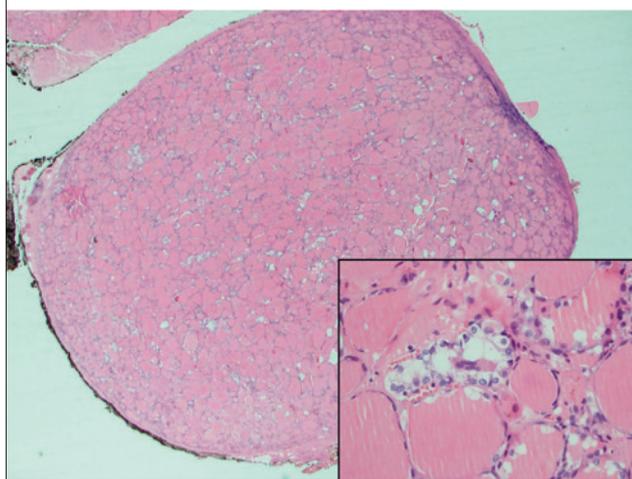
new mass, in left lateral-cervical region became evident. Neck ultrasound imaging showed several lymph nodes on the left side, the greater of which had a main axis of 2 cm with rough calcifications. Furthermore a 1 cm nodule, in the left thyroid lobe, showed a highly suspicion ultrasound appearance. Cytology by fine needle aspiration demonstrated features consistent with papillary thyroid carcinoma. The patient underwent to a total thyroidectomy and left lateral neck compartmental lymph node dissection. Histology confirmed the presence of a multifocal follicular variant of papillary carcinoma (FVPC) localized in the left thyroid lobe (Fig. 1), with capsular infiltration and involvement of the perithyroidal soft tissues. Three out of 19 lymph nodes were positive for metastasis. No lymphocytic infiltration of thyroid tissue was observed. We retrieved the previous case from our archive. It's reevaluation was consistent with a nodal metastasis from papillary thyroid carcinoma. The patient underwent to a remnant ablation therapy with 100 mCi <sup>131</sup>I after rhTSH, with excellent biochemical and structural response after one year of follow-up.

## Discussion

Lateral ectopic thyroid is due to an aberrant development of the gland, that happens when the cells of lateral part do not join those of the median one<sup>1</sup>. Its occurrence is usually in the submandibular region and, less frequently, in the para-jugular or para-carotid region<sup>2,3</sup>. This event is far rarer than lingual thyroid, which represents 90% of all cases of ectopic thyroid. Differential diagnosis with lymph node metastasis of thyroid cancer can be very challenging. The cases of ectopic thyroid detected in the lateral cervical region were, in the past, usually interpreted as malignant metastatic lesions<sup>4,6</sup>. In the series of Nussbaum et al. 27 out of 197 patients with thyroid carcinoma (14%) presented with regional

lateral mass and a clinically normal thyroid gland on early evaluation<sup>7</sup>. More recently, however, some authors argued that normal ectopic tissue should suggest ectopic thyroid rather than papillary thyroid carcinoma (PTC) metastasis<sup>8</sup>. In one case PTC was found to be associated with hyperplastic thyroid tissue in the lateral neck, considered as ectopic thyroid<sup>9</sup>. Amodi et al. reported the association of lateral multi nodular thyroid tissue and normally located goiter<sup>10</sup>. Finally Basaria S. et al. (2001) described a patient affected by PTC and normal lingual thyroid misdiagnosed as metastasis<sup>11</sup>. The crucial problem is whether typical nuclear changes of PTC are always clearly evident in aberrant localizations. Diagnosis of FVPC relies on the typical nuclear alterations, the assessment of which is, in many cases, subjective and leads to a high inter observer variability, mainly when nuclear features are "border line" or faded<sup>12,13</sup>. According to some authors it is not always possible, in cytology, separate FVPC from the other follicular lesions because of overlapping features or sporadic presence of nuclear abnormalities<sup>14</sup>. The absence of the nuclear features characteristic of FVPC does not rule out the diagnosis since these can be very mild and scattered. Critical analysis of our case should take in account the important progress in medical knowledge and diagnostic methods made in the last decades. Firstly, in the seventies, imaging techniques as neck ultrasonography and computerized tomography were not yet widely available in clinical practice. At that time scintigraphy examination was the only method to assess functional status and morphology of the thyroid gland, as well as to detect ectopic tissue. In our case scintigraphy was not performed so that evaluation of thyroid relied exclusively on neck palpation, which did not show abnormalities. No symptoms related to thyroid dysfunction were recorded, despite the presence of massive lymphocytic infiltration, which was regarded as "basedowian picture". This latter finding was more likely compatible with the presence of lymphoid tissue, harboring thyroid tissue of adenomatous appearance. Forty years later FVPC was diagnosed, with nodal metastatic involvement in the same site of the previous lesion. This clinical evolution suggests that the lateral mass excised in 1974 could be a metastasis from an occult FVPC, rather than an ectopic lateral thyroid. Besides we must take into account that, in the mid-1970s, FVPC was not yet broadly recognized as separated primary tumor<sup>15</sup>. Moreover in 1974 not the whole tissue excised was included and evaluated. In conclusion we recommend that diagnosis of lateral ectopic thyroid should be made with great caution in cases of lateral cervical mass. Every lateral-cervical lesion containing thyroid tissue should be considered suspected until proven otherwise. In these cases, thyroid should mandatory be investigated from a clinical, radiological and pathological point of view and total thyroidectomy should be considered in the diagnostic route, in order to rule out an occult papillary thyroid carcinoma.

**Fig. 1.** Hematoxylin and eosin stained (H&E) at 2x and 20x magnification showing typical nuclear features of papillary thyroid carcinoma.



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