



CASE REPORT

Papillary carcinoma in lateral cervical ectopic thyroid

Denise Santos Rosa¹, Beatriz Soares de Azevedo Sardano¹,
Claudia Menezes da Rocha Crioulo¹, Vanessa de Vicenze Cruz¹,
Jossi Ledo Kanda¹

Abstract

Introduction: Ectopic thyroid is a rare event, with an incidence of 1 in 300,000 cases. The probability of a carcinoma appearing in this tissue is less than 1%. It may be extremely difficult to distinguish a primary neoplasm involving the ectopic thyroid from a metastatic cervical lymph node. **Case report:** We report a case of papillary carcinoma in lateral cervical ectopic thyroid region. It was initially diagnosed as a thyroid carcinoma with lateral cervical metastasis. **Comments:** The diagnosis of papillary carcinoma in lateral cervical ectopic thyroid can be challenging, and immunohistochemistry can be a useful tool.

Keywords: Carcinoma, Papillary; Head and Neck Neoplasms; Thyroid Dysgenesis.

¹Faculdade de Medicina do ABC (FMABC), São Bernardo do Campo, SP, Brasil

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Informed consent was given by the patient for case publication.

Introduction

Ectopic thyroid is any thyroid tissue located outside its usual topography¹. It is a rare event, with a hypothesis of 1: 300,000 cases, affecting mainly women (4:1)². It can suffer the same pathological processes of an orthotopic thyroid: inflammation, hyperplasia and carcinogenesis³. The probability of a carcinoma arising in this tissue is less than 1%².

The distinction between a primary malignancy involving the ectopic thyroid from a metastatic cervical lymph node can be challenging, constituting a differential diagnosis of great importance in the management, staging and prognosis of the disease^{3,4}.

Case report

A 22 years old male Caucasian patient presented with a complaint of a painless, progressively growing nodule in the left cervical region for 3 months. The locoregional examination presented a palpable, mobile and painless lymph node in the left of carotid-jugular region, of 3.0 × 2.0 cm of diameter, and other smaller lymph nodes in the left superior carotid-jugular regions and in the right lower spinal region, with the same characteristics. The thyroid gland was normal at palpation. The patient's thyroid function was normal (TSH = 2.46 mIU/L) and only anti-thyroperoxidase was slightly elevated (43.7). The neck ultrasound identified a 3.1x1.7cm solid-cystic lymph node at the left III level, in addition to bilateral smaller lymph nodes. No thyroid nodules were observed. Computed tomography of the neck with intravenous contrast



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revealed a nodule-free heterogeneous thyroid associated with posterior cervical and carotid-jugular lymph node. The largest cystic aspect was of left level III, of 24mm. The fine-needle aspiration biopsy (FNA) of the largest lymph nodes was positive for the malignant neoplasms cells, which correspond to metastasis of thyroid papillary carcinoma. The thyroglobulin in washout dosage from the FNA was of 266IU/mL. At first, the patient underwent lymphadenectomy in level V, on the right side. The intraoperative frozen section examination revealed reactive lymphoid hyperplasia. It was then followed by excision of the level III lesion, on the left side, and the frozen section examination revealed a benign lesion. It was opted for total thyroidectomy, which the frozen section found only a colloid nodule of 0.7cm in the left lobe. Thereby, as no carcinoma was found in the thyroid gland and there was no malignancy in the left cervical nodule, the procedure was closed until conclusion of paraffin. The histopathological examination confirmed a colloid goiter of 0.7cm in the left lobe and mild lymphocytic chronic thyroiditis (Figure 1). The cervical node of level III, on the left side, 3.0 × 2.0 × 0.5cm of size, was identified as a well-differentiated papillary carcinoma - no evidence of lymph node tissue or brachial cyst was found (Figure 2). Lymph node of level V, on the right side, 1.0 × 0.8 × 0.4cm of size, confirmed reactive lymphoid hyperplasia. The thyroid was totally included in the analysis (no other nodules were found) and the histological picture was compatible with primary neoplasm in lateral cervical ectopic thyroid. In the immunohistochemical analysis, the thyroglobulin marker (Tg), thyroid transcription factor 1 (TTF-1) and galectin-3 (Gal-3) were positive and cytokeratin-19 (CK19) negative (Figure 3A, B, CD). The marker of reactive and neoplastic mesothelial cells (HBME-1) was not available. In the follow-up of the treatment of this patient, adjuvant radiotherapy was not chosen, but only hormonal suppression. Two years after the surgery, the hormonal suppression is still in use with 175mcg of levothyroxine and there is no evidence of disease recurrence.

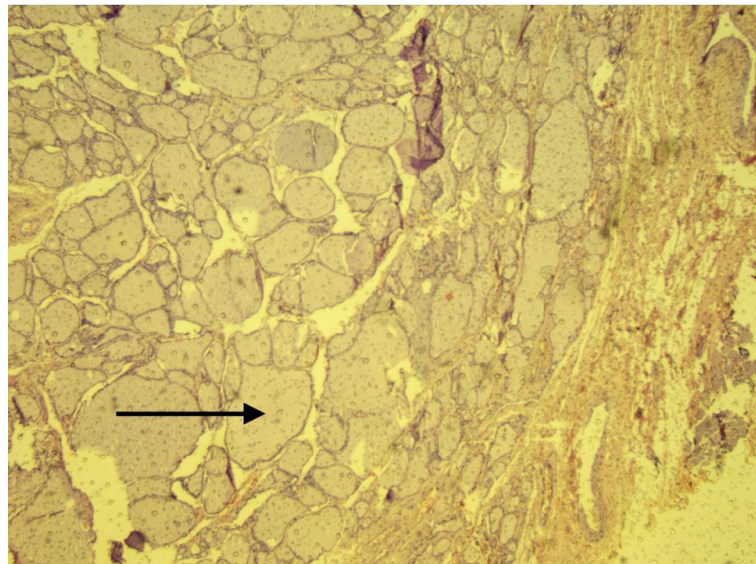


Figure 1. Optical microscopy of the nodule in the left lobe of thyroid. Colloid nodular goiter (arrow). Objective 4. H-E.

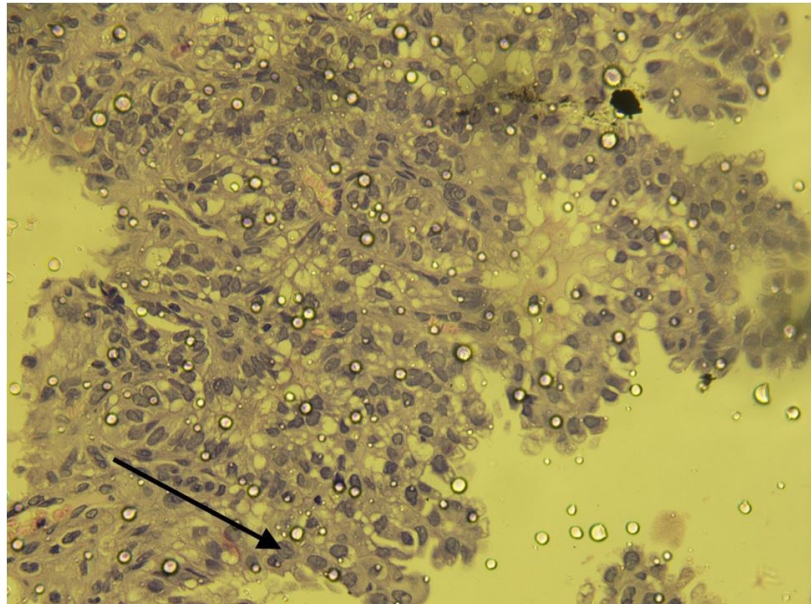


Figure 2. Optical microscopy cervical node of level III left. Papillary arrangements showing the slot nucleus and eosinophilic intranuclear inclusion (arrow) pathognomonic of papillary carcinoma. Objective 40. H-E.

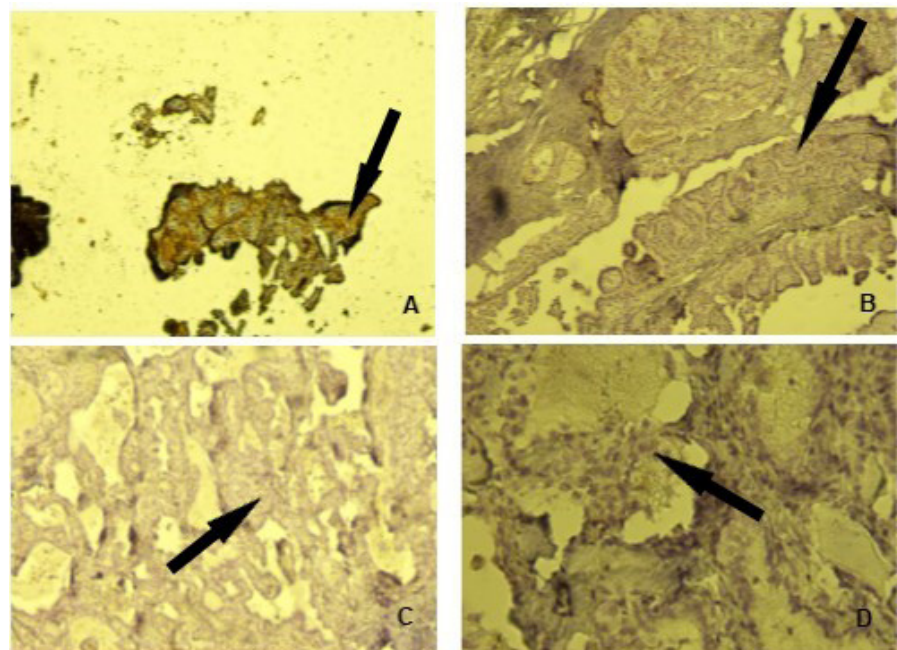


Figure 3. A. Immunohistochemistry. Neoplasm with papillary arrangement whose Tg antibody was positive (arrow). Objective 10. **B.** Immunohistochemistry. Papillary arrangements showing malignant neoplastic cells positives for the TTF-1 marker (arrow). Objective 10. **C.** Immunohistochemistry. Papillary arrangements showing malignant neoplastic cells positives for the Gal-3 marker (arrow). Objective 10. **D.** Immunohistochemistry. Negative marker CK19 (arrow). Objective 40.

Discussion

In the embryonic stage, the thyroid gland is derived from two endodermal sprouts, one larger medial and two smaller laterals. The medial sprout produces most of the thyroid parenchyma, while the lateral are derived from the fourth pharyngeal pouch and contribute from 1 to 30% of the gland's weight. Commonly, failure of the medial sprout to the descent along the thyroglossal duct results in ectopic thyroid at different locations along the midline. However, by contrast, in rare cases, the failure of lateral sprout fusion results in lateral ectopic thyroid¹.

The thyroid tissue can suffer pathological processes such as inflammation, hyperplasia and carcinogenesis. More than 99% of thyroid carcinomas occur within the ectopic gland³. The probability of a carcinoma in the ectopic tissue is lesser than 1%² and among these; the most common subtype is the papillary⁵.

The literature describes several locations of ectopic thyroid carcinoma: lingual, within the cyst of the thyroglossal duct, intratracheal, within a branchial cleft cyst, besides others rare locations in the midline. However, there are few reports of carcinoma in the lateral cervical thyroid, and consequently precise epidemiological data concerning the incidence and prevalence of this phenomenon are rares^{3,4}.

The diagnosis confirmation is possible due to the absence of a primary orthotopic thyroid carcinoma from a careful histopathological examination. Thus, lymph node metastasis of occult primary carcinoma tends to be over diagnosed, and may represent a carcinoma in lateral cervical thyroid³. So how can these two phenomena be differentiated?

The immunohistochemical study can be a tool in the aid of this distinction. The thyroglobulin (Tg) is a marker that when positive in the cytoplasm of neoplastic cells indicates the thyroid origin. The TTF-1 identifies differentiated thyroid neoplasms. Gal-3 and CK19 have been useful markers in the diagnosis of papillary thyroid carcinoma (PTC), as well as the HMB-14. Cabib et al.⁴ examined the expression of Gal-3, CK19 and HBME-1 in two different groups ("A" formed by cases of lateral cervical masses with PTC, but without tumor in the thyroid, and "B" by cases that presents PTC, as much as in the thyroid as in the lateral cervical masses) and concluded that in the first group, possibly, the PTC developed from an ectopic tissue and in the second it was possibly metastatic carcinoma.

The diagnosis of papillary carcinoma in lateral cervical ectopic thyroid can be challenging. This possibility should be considered in cases of lateral cervical masses and immunohistochemistry can be a useful tool. Due to the rarity of published cases, there are still no standardized procedures. It is believed that the treatment should be based predominantly on surgical removal of the malignant lesion. The management of this disease, including total thyroidectomy, neck dissection and radiotherapy, should be based in individualized risk stratification or at the discretion of the surgeon.

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***Correspondence**

Denise Santos Rosa
Faculdade de Medicina do ABC (FMABC)
Rua Xingu, 823, Apto 41, Bloco Gama,
Vila Valparaíso
CEP 09060-050, Santo André (SP), Brasil
Tel.: +55 (11) 5049-0262,
(11) 5049-1471
E-mail: denise.rosa1010@gmail.com

Authors information

DSR, VVC - Assistants medicals at the
Discipline of Head and Neck Surgery
of the Faculdade de Medicina do ABC;
BSAS, CMRC - Medical students at
the Faculdade de Medicina do ABC;
JLK Medical and professor at the
Discipline of Head and Neck Surgery
of the Faculdade de Medicina do ABC.

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