

CASE REPORT

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Ectopic Thyroid Papillary Carcinoma with Cervical Lymph Node Metastasis as the Initial Presentation, Accompanied by Benign Thyroid Gland

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ABSTRACT

Background: Ectopic thyroid papillary carcinoma presenting as bilateral neck lymph nodes metastasis is very rare. Ectopic thyroid tissue may appear in any location along the trajectory of the thyroglossal duct from the foramen cecum to the mediastinum. It is subject to malignant transformation and is classically accompanied by a similar transformation of the native thyroid gland. Similar to that of the native thyroid gland, the most common malignancy found is Papillary thyroid carcinoma. Unusual cases in which ectopic thyroid carcinoma presents with normal native tissue support an alternative hypothesis that ectopic thyroid tissue may develop malignancies independently from the native thyroid gland. **Objective:** We present an extremely rare case of a 30-year-old woman previously diagnosed with Hashimoto's thyroiditis, presenting with a palpable mass in the lateral neck suspicious for malignancy. **Results:** After several examinations and surgical removal of the mass, histopathologic evaluation of the continuous sections of the thyroid, demonstrated metastatic disease from papillary carcinoma of the thyroid. Total thyroidectomy and biopsy revealed benign thyroid tissue without any foci of microcarcinoma. A hypothesis of ectopic thyroid tissue and its malignant transformation was made. **Conclusion:** By presenting this case, our goal is to highlight and make the physicians aware of the possibility of developing primary carcinoma of the ectopic thyroid tissue, without an active tumor of the thyroid gland.

Keywords: Ectopic thyroid tissue, malignant transformation, metastatic papillary thyroid cancer.

1. BACKGROUND

Ectopic thyroid tissue (ETT) is a rare condition due to development of thyroid outside the physiologic thyroid gland anatomy and inciting as an idiopathic impairment during embryologic migration process. Before the thyroid gland dives into the neck it will contribute in the floor of pharynx formation in 24th-28th day in utero. Thus, as it starts to descend into the neck, initially it maintains connection to tongue through the thyroglossal duct, structure which will disappear later in life (1). In contrast when thyroglossal duct remnant persist in foramen cecum without shifting down, will lead to a mass or a cyst formation. Most common sites of ectopic thyroid tissue are: the base of the tongue, presenting as a primordial deposit tissue in the foramen cecum, following by the neck, thyroglossal, laryngotracheal, and the structure closed. Sometimes the ectopic migration may be the only physiologic thyroid tissue with diminished function highlighting lingual ectopic tissue, with 70% incidence (2). Generally, it has been detected during high demand states for thyroid hormones, such as puberty or pregnancy, altering their levels. Apart of these atypical locations studies have shown other unusual pathways of follicular cells such as in clavícula (3), in parenchymatous pancreas (4, 5), a very rare form might be an embryologic remote intradermal thyroid location (6) and mediastinal (7-11).

Papillary thyroid carcinoma (PTC) is defined as the most common form of thyroid cancers approximately with 80% prevalence occurring either in normal gland position (12, 13) or along the pathway of descending of thyroid tissue accounting for 3% of cases (13). The risk factors raising the in-

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cidence of ectopic thyroid for malignancy transformation are still not well understood and it is thought to occur independently from the native thyroid gland. Furthermore, few cervical cases have been confirmed through the presence of ectopic tissue metastasis in the nearby lymph node despite the intact thyroid native gland radiologic support (CT\MRI) biopsy, immunohistochemistry (2, 14). The occurrence of metastasis from the aberrant tissue can sometimes involve both cervical regions or unilateral only (2). Regarding to the ambiguous nature, the treatment has been focused in total thyroidectomy, removal of ectopic mass and local lymph node followed by radioactive iodine ablation leading to a satisfactory prognosis.

2. OBJECTIVE

We present an extremely rare case of a 30-year-old woman previously diagnosed with Hashimoto's thyroiditis, presenting with a palpable mass in the lateral neck suspicious for malignancy.

3. CASE PRESENTATION

A 30-year-old woman came to our clinic for FNAC examination of the thyroid following-up her history of Hashimoto's thyroiditis. She has been diagnosed with Hashimoto's thyroiditis 3 years prior, and has been compliant with her scheduled appointments in the clinic. Cytologic evaluation of the thyroid with ultrasound-guided fine needle aspiration from multiple sites revealed normal thyrocytes and presence of macrophages that fell into the Bethesda II category according to WHO (Figure 2). Thyrocytes were arranged in crowded three dimensional groups and in microfollicular pattern accompanied by a background of sparse macrophages, lymphocytes and stromal elements.

Four months later she presented to the clinic with discomfort and swelling on the right side of her neck that has been growing for several weeks.

Physical examination resulted in a non-tender palpable mass. She was clinically euthyroid. Ultrasound of the thyroid (Figure 1) showed diffuse heterogeneous parenchyma with focal glandular enlargement presenting as hypoechoic nodules varying in size in a background of hyper vascularized thyroid gland. Fine echogenic fibrous septa create a pseudo-lobulated appearance. CT-scan of the neck showed enlarged lymph node with poorly defined nodal margins, classic of extracapsular nodal spread, suggesting metastatic disease. Consequently, the patient underwent surgery of the neck with complete removal of the mass.



Figure 1 (a, b, c). Ultrasound of the thyroid showed diffuse heterogeneous parenchyma with focal glandular enlargement presenting as hypoechoic nodules varying in size in a background of hyper vascularized thyroid gland. Fine echogenic fibrous septa create a pseudo-lobulated appearance

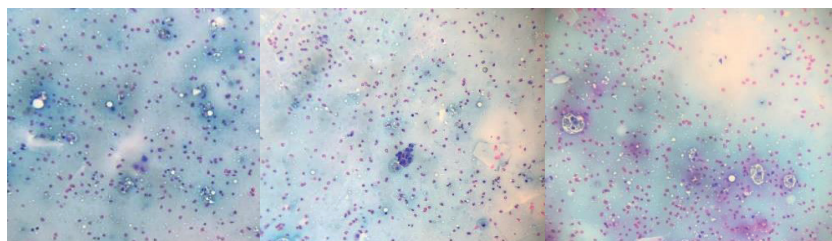


Figure 2 (a, b, c) (Pap stain). FNAC of the thyroid gland in 3 different sites showed aggregates of thyrocytes with monomorphic aspect, some of them showing mild nuclear anisokaryosis in a background of sparse lymphocytes, macrophages erythrocytes and stromal elements

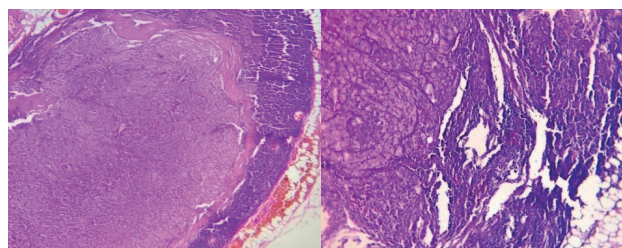


Figure 3 (a, b). H&E- stained sections of the lymph node showed presence of metastatic disease from papillary carcinoma of the thyroid. Neoplastic cells were arranged in finger-like projections with a central fibrovascular core giving the appearance of true papillae. High magnification of the tumor cells showed nuclear enlargement and overlapping features and the presence of nuclear grooves

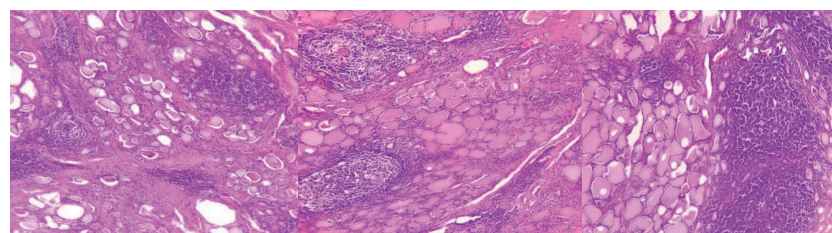


Figure 4. H&E-stained sections from the thyroid gland displayed micronodular pattern with diffuse lymphocytic infiltration of the gland with numerous lymphoid follicles with germinal centers accompanied by fibrosis

Microscopic examination (Figure 3) of the mass showed presence of neoplastic cells within the lymph node parenchyma, with extracapsular invasion. The neoplastic cells were arranged in finger-like patterns with central fibrovascular core creating true papilla (Figure 4 and 5). A diagnosis of metastatic involvement of the lymph node originating papillary carcinoma of the thyroid was made.

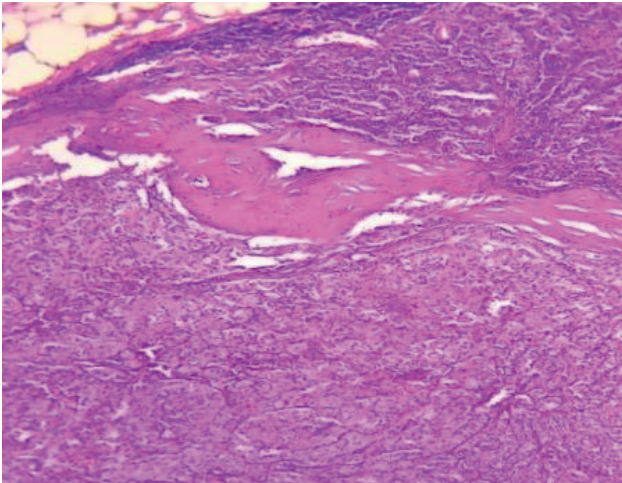


Figure 5. H&E- stained section of the lymphonode on neck dissection showed metastatic disease from papillary carcinoma of the thyroid

Occult papillary carcinoma was firstly suspected in this patient. Hence, the patient underwent total thyroidectomy with partial neck dissection. Histopathologic evaluation of the continuous sections of the thyroid, demonstrated benign thyroid tissue in a background of Hashimoto's thyroiditis. The tissues around the thyroid resulted in carcinomatous infiltration, and the two lymph nodes removed were positive for metastatic papillary carcinoma of the thyroid, but there was no presence of malignant cells in the thyroid gland which was entirely sectioned and reviewed, for microcarcinoma foci.

A diagnosis of ectopic thyroid papillary carcinoma with metastatic disease in bilateral cervical lymph nodes was made.

4. DISCUSSION

Thyroid gland has a complex embryology. Thyroid diverticulum develops from endodermal lining of foregut, descends into the neck maintaining a connection to the tongue through the thyroglossal duct (16). During this process of passage to the final pre-tracheal position, developmental embryogenic defects might occur, resulting to ectopic thyroid tissue formation (17). The most frequent location of ectopic thyroid tissue is found in the cervical midline, accounting for 90% of total cases. The rest of 10% of the cases are found in the anterior tongue, mediastinum, esophagus, struma ovarii and rarely in the abdominal cavity presenting mostly as incidental findings.

Primary malignant transformation of the ectopic thyroid tissue is very uncommon (18). Due to difficulties in distinguishing between primary carcinoma and metastasis, the incidence and prevalence data of such occurrence are lacking. Tissue sample evaluations, microscopic and pathological analysis, along with detailed imaging reports, are crucial to accurately identify primary ectopic papillary carcinoma.

Our case showed a rare presentation of the papillary thyroid carcinoma by the fact that the malignancy is detected in the ectopic thyroid tissue, without evidence of

a malignant pathology of the thyroid gland. Due to this rare occurrence, there is no standard treatment therapy or guidelines. The optimal treatment is in conjunction with early and precise diagnosis.

Fine needle aspiration remains the primary diagnostic test of choice to establish the diagnosis. Total thyroidectomy with bilateral or ipsilateral neck dissection followed by radioactive iodine treatment, excision of the ectopic tissue, are the most favorable treatment options for cases like this.

5. CONCLUSION

Malignancy arising from an ectopic thyroid tissue is a rare presentation. Therefore, implementing early precise diagnosis, is a crucial part in differentiating this occurrence from primary thyroid carcinoma. Treatment methods should be individualized, based on the case presentation and disease progression. Long term follow-up must be implemented to monitor recurrence and the thyroid hormone level management. By presenting this case, our goal is to highlight and make the physicians aware of the possibility of developing primary carcinoma of the ectopic thyroid tissue, without an active tumor of the thyroid gland.

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