Uncommon Presentation of Ectopic Thyroid in Adulthood: About 2 Cases

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Abstract: *Introduction*: Ectopic Thyroid (ET) is a rare condition resulting from the abnormal thyroid development or migration. We report through this article two cases of ectopic thyroid .

Observation 1: A 44-year-old woman presented with chronic tearing of the right eye, underwent an orbital CT scan showing a lesion located at the base of the tongue. Physical exam showed that the thyroid gland was not palpable and normal oropharyngeal space. The tongue MRI showed a heterogeneous mass of the base of the tongue, measuring 43x37 mm. Thyroid Scintigraphy showed an elective uptake of radiotracer at sublingual area and no uptake in the normal thyroid location. Hormonal exploration was normal. Medical supervision was proposed to our patient.

Observation 2: A 40-year-old woman presented with a history of dysphonia. On clinical examination, thyroid gland was not palpable in cervical area. Nasofibroscopy revealed a mass of the tongue base compressing the vocal cords. During nasofibroscopy, she presents an acute difficulty of breathing, a tracheotomy was done urgently. Surgical excision of the mass was realised. Histopathological findings revealed the presence of a normal thyroid gland within the fibromuscular tissue, with focal vesicular thyroid carcinoma.

Discussion: Ectopic thyroid may be asymptomatic or revealed by hypothyroidism or signs of compression. The risk of malignancy remains rare. In asymptomatic forms, biological and morphological monitoring is suggested. Surgery is necessary in case of compression or suspicion of malignancy.

Keywords: Thyroid, ectopia, adulthood, asymptomatic, vesicular carcinoma.

INTRODUCTION

Ectopic Thyroid (ET) is a very rare entity [1,2] caused by the abnormal migration of thyroid cells during embryogenesis [2]. The thyroid gland may stop at various locations along the thyroglossal duct during its descent from the lingual base [1]. The lingual region is the most common form [2]. Frequency is estimated for approximately from 1/100 000 to1/300 000 [1,2]. Diagnosis may be achieved at any age with a female predominance. Its clinical presentation is variable, according to the early or late revelation and according to thyroid function. Furthermore, the occurrence of carcinoma remains extremely rare [3]. Imaging allows diagnosis and guides therapeutic approach.

In this study, we report 2 cases of lingual thyroid revealed in the adulthood and we review the literature on this topic.

CASES PRESENTATIONS

Case 1

A 44-year-old woman was followed for watery eye caused by lacrimal duct obstruction. She underwent an orbital CT-scan that showed an incidental lesion of the tongue base. Clinical examination was normal, thyroid gland was not palpable. CT-scan of the neck showed the presence of a heterogeneous mass measuring 43x37 mm at the base of the tongue with complete thyroid atrophy (Figure **1**). Ultrasonography showed the absence of the thyroid gland at its usual site. Thyroid scintigraphy with Tc-99m pertechnetate, showed an intensive and elective uptake of radiotracer at sublingual area and no uptake in the normal thyroid location (Figure **2**). Thyroid hormone levels and biochemical values of the patient were normal: TSH of 0.37 mUI/L (0.25 to 5) and free T4 of 18 pmol / l (10.6-19.4).

In the absence of complications, yearly monitoring of thyroid function and thyroid scintigraphy or MRI of the neck was recommended.

Case 2

A 40-year-old woman consulted for a history of chronic dysphonia since 6 months. She did not have a history of neck irradiation, thyroid disease, or family history of thyroid cancer. Physical examination was normal and thyroid gland was not palpable. Nasofibroscopy revealed a mass of glossotonsillar sulcus. During nasofibroscopy, she presented an acute dyspnea and underwent an urgently tracheotomy. CT-scan of the neck showed a large epiglottiss mass

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Figure 1: CT-scan of the neck (axial (left) and sagittal (right)) showing an heterogeneous mass of the tongue base measuring 43x37 mm).

Figure 2: Thyroid scintigraphy with Tc-99m pertechnetate showing an intensive and elective uptake of radiotracer at sublingual area and no uptake in the normal thyroid location.

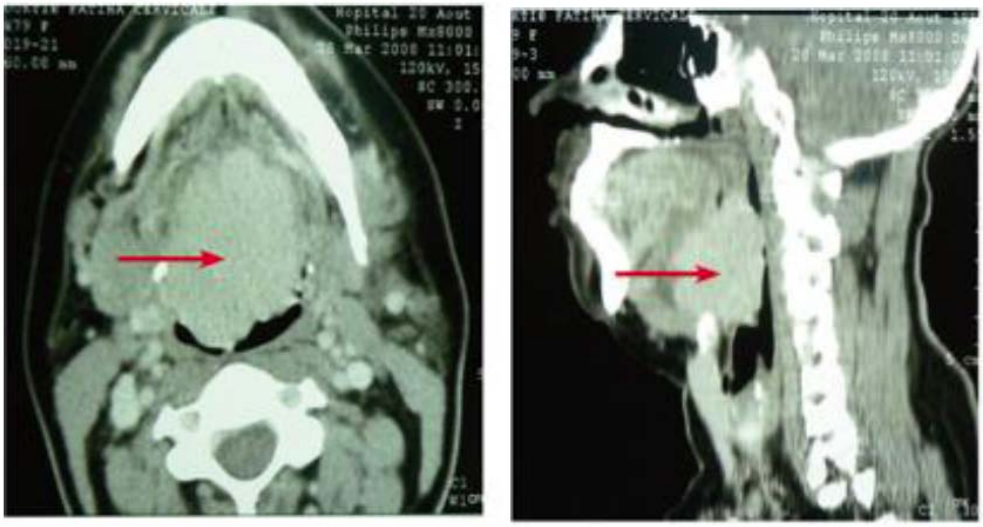

Figure 3: CT-scan of the neck showed a large epiglottic mass extending to the base of the tongue and protruding to the oropharyngeal area.

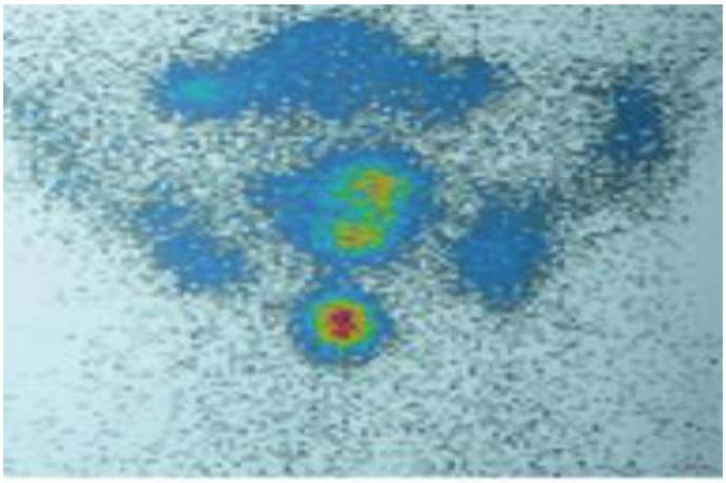

Figure 4: Thyroid scintigraphy with Tc-99m pertechnetate showed a sub hyoid double uptake of radiotracer and no uptake in the normal thyroid location.

extending to the base of the tongue and protruding to the oropharyngeal area (Figures **3**). Thyroid scintigraphy with Tc-99m pertechnetate (Figure **4**) showed a subhyoid double uptake of radiotracer and no uptake in the normal thyroid location. Biopsy of the mass during direct laryngoscopy showed a thyroid tissue.

The patient underwent a surgical excision of the mass. Histopathological findings revealed the presence of thyroid tissue with a focal vesicular thyroid carcinoma. Subsequent management included postoperative radioiodine therapy (radioiodine ablative 150 mci was given). The whole-body scan did not show functional thyroid tissue remnants. She was substituted by L-thyroxine at a dose of 150 µg / day. Thyroglobulin rate was 0.13 ng / ml.

DISCUSSION

Ectopic thyroid incidence is about 1 per 100 000 cases. It is responsible for 64% of congenital hypothyroidism [2,4]. It results from an aberration occurring during fetal migration, leaving a part or the totality of the thyroid tissue at the base of the tongue or other position along the thyroglossal duct [2].

The cause of ectopic thyroid tissue remains unclear in most cases. Genetic studies in mice showed that transcription factors appear to play an important role in embryogenesis and thyroid migration. TTF-1 (Thyroid-Transcription Factor-1) and PAX8 have a role in follicular cell proliferation and TTF-2 has a role in the migration and development of the TSH receptor after birth. Environmental factors are not excluded as well [3,5].

Ectopic thyroid occurs more frequently in female patients [3]. Mean age of diagnosis is 40 years. The age of diagnosis depends on the size and function of the ectopic thyroid tissue.

Symptoms of ectopic thyroid are related to the growth of the thyroid tissue, causing dysphonia, dysphagia, bleeding, or dyspnea [6]. The occurrence of neck pain or bleeding may be a sign of malignant transformation. It can also be revealed by hypothyroidism. Hyperthyroidism remains rare [7]. Asymptomatic forms are quite frequent.

Differential diagnoses of ectopic thyroid include thyroglossal duct cysts, lymphangioma, hemangioma, fibroma, lipoma, epidermal cysts, salivary gland tumors and lymphoma [8].

Malignant transformation of the lingual thyroid is very rare, occurring in about 1 to 3% of all cases with ectopic thyroid tissue [2]. Only thirty cases registered in the literature [9,10,11]. Vesicular thyroid carcinoma is more commonly found [12].

Ultrasonography, scintigraphy computed tomography (CT), magnetic resonance imaging (MRI) are the modalities that can be used in the diagnosis of ectopic thyroid [13]. The presence of scintigraphy uptake of radiotracer I n the base of the tongue, confirms the ectopic tissue thyroid. CT and / or MRI are necessary when surgery is indicated, or in case of suspicion of malignant transformation in order to: explore the mass, detect perineural extensions and localize vascular axes [2]. Biopsy should be avoided. Indeed, it can expose to a hemorrhagic risk, perforation, stenosis, fistulas or infections [14].

Treatment depends on several factors, including the size and the ectopic thyroid position, the presence of symptoms, thyroid function, histopathological findings and surgical risk [6].

Replacement therapy with L-thyroxine is indicated in cases of hypothyroidism, suspected malignant transformation, airway obstruction, severe and repeated bleeding, or severe dysphagia [15]. Surgical treatment is indicated only in complicated forms or in case of non-regression of the signs of compression under treatment. Radioiodine therapy may be an alternative to surgery [2]. Radioiodine ablative therapy should follow surgical resection in case of malignancy, followed by hormone replacement therapy [12]. Otherwise, monitoring of thyroid function is recommended in asymptomatic forms.

CONCLUSION

Ectopic thyroid is a rare condition. It may be asymptomatic or revealed by thyroid dysfunction or signs of compression. The risk of malignancy remains rare. In asymptomatic forms, biological and morphological monitoring is recommended. Surgery is necessary in case of compression or suspicion of malignancy.

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