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Basilingual Ectopic Thyroid: About A Historical Case

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Abstract **Case Report**

Ectopic thyroid is a rare embryological aberration, with incidence between 1/100 000 and 1/300 000. It is associated with the absence of thyroid tissue in normal site in approximately 70% of cases. It is more common in women (3 to 7 W versus 1 M). This observation has concerned a 15 years old patient presenting a dysphagia for 6 months. Hormonal evaluation showed a hypothyroidism (TSH was 8.43µUI/ml). Cervical ultrasound suggested hypoplasic thyroid gland. Tomography scan concluded to the absence of thyroid tissue in normal position and presence of a mass at the base of the tongue reaching the contact of the free upper part of the epiglottis. The 99mTc thyroid scintigraphy confirmed the diagnosis, showing the absence of thyroid tissue in normal site and uptake the presence of an ectopic focus at the level of the base of the tongue. In the majority of cases, lingual thyroid is diagnosed in the childhood and young adulthood or around menopause. It appears as a mass in the base of the tongue causing local signs of compression often associated with a hypothyroidism. A 99mTc or iodine 123 thyroid scan can make diagnosis. Treatment based on hormone replacement aims to avoid ectopic tissue growth of and to prevent surgery which can be hard.

Keywords: ectopic- thyroid - diagnosis- scintigraphy.

INTRODUCTION

Thyroid ectopia is a rare pathology [1, 2] secondary to the abnormal migration of thyroid cells during embryogenesis [2]. Thyroid tissue remains in lingual position, sublingual or even high cervical position and does not develop sufficiently. Lingual thyroid occurs with a frequency of 1/100 000 to 1/300 000 [2, 3]. It is four or five times more common in the woman than in man [4]. The age of diagnosis is between birth and over 80 years old with an average age of 40.5 years [3]. 70% to 75% of patients have a lingual thyroid, it does not exist eutopic thyroid in the pre-tracheal position [3, 5]. The occurrence of thyroid carcinoma on thyroid Ectopic is very rare. On the very rare cases reported in the literature, the vesicular variant was predominant [6].

CASE REPORT

A 15-year-old female patient with no particular pathological history has had a dysphagia for six months. Clinical examination objective a posterior linguistic mass. A hypoplastic thyroid gland has been reported to the cervical ultrasound at the level of the oral floor. Cervical tomography scan, performed with injection of contrast, revealed the presence of a mass at the base of the tongue reaching the contact of the free upper part of the epiglottis, measuring 21x22x26 mm in favor of an ectopic thyroid [Figure.1]. The dosage of thyroid hormones has confirmed hypothyroidism; the TSH level was 8.43 µU / ml. The patient was put under Levothyroxine. Tc99m thyroid scintigraphy confirmed the absence of thyroid tissue in a normal anatomical position and the presence of an ectopic focus at the level of the base of the tongue [Figure.2]. The patient was referred for exerese surgery.

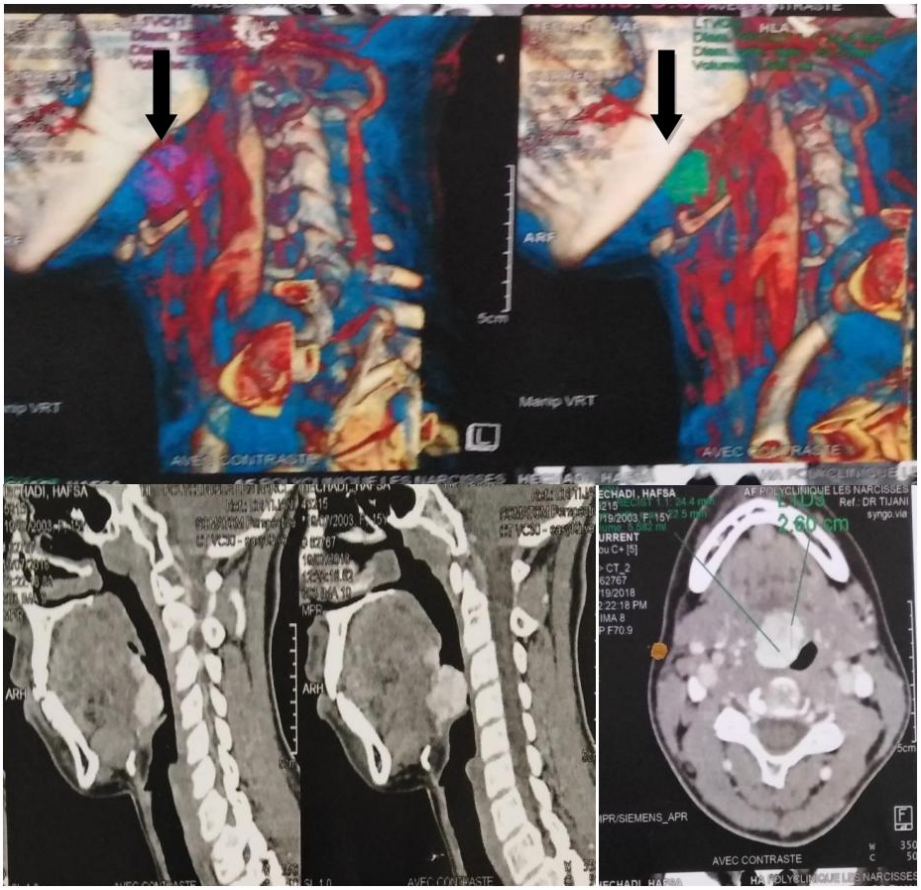

Fig-1: Tomography scan showing a mass at the base of the tongue reaching the contact of the free upper part of the epiglottis and bulging into the oropharynx lumen, measuring 21x22x26 mm in favor of an ectopic thyroid process

DISCUSSION

During the third and fourth weeks of embryonic development, emerge the primary thyroid tissue of the junction of two-thirds anterior and third posterior of the tongue. At the end of the fourth week, the thyroid lobes are united by the thyroglossal canal attached to the ventral floor of the pharynx. From the sixth week, this channel atrophies while maintaining its pharyngeal connection in the form of an orifice (the foramen caecum) located at back of the tongue. At the end of the seventh week, two lobes united by the isthmus are in position cervical while the cells of the canal get differentiate into thyroid tissue to form the pyramid of Lalouette [2].

A migration defect will cause a thyroid ectopia. High thyroid ectopia represents the most frequent localization [90% of cases]; lingual more often, prehyoid or subhyoid [2, 7]. Approximately 70% of patients do not have of thyroid tissue in the normal pre-tracheal position [2, 3, 5, 7]. The lingual thyroid is found in one case out of100, 000 to 300,000. In case of thyroid pathology associated including hypothyroidism, frequency is 1 in 3,000 to 1 in 4,000 [2, 3]. It is more common in women than in the man with a sex ratio that varies between 3/1 and 7/1[3,4]. In most cases, she is diagnosed in children, the young adult but often to the menopause [2, 5.8] [30% at the time of puberty, 55% between 18 and 40 years [2], 10% to menopause and 5%in the elderly [5]. So, puberty and pregnancy are typically factors triggering the symptomatology [2, 9].

Clinically, ectopia can be manifested by hypothyroidism, or by a compressive phenomenon such as coughing, dysphagia, dysphonia or dyspnea [2, 3, 5, 10, 11].Hyperthyroidism is rare [2, 3, 5,11], neoplasia, is very rare [2, 3,5] less than 30 cases have been reported in the literature [6].The existence of a palpable posterior lingual mass evokes the diagnosis. It can be infracentimetric or reach several centimeters as the case of this patient where the mass measured 21x 22 x 26 mm.

An untimely biopsy could be avoided; in Indeed, it exposes the patient to hemorrhagic risks, perforation, stenosis, tracheobronchial fistulas or infection [12]. Differential diagnoses of thyroid ectopia are the thyroglossal canal cyst, lymphangioma, hemangioma, fibroid, lipoma, epidermal cyst, tumors of the salivary glands and the lymphoma [2, 7, 9].99m Technetium Thyroid Scintigraphy [9] or iodine123 confirms the diagnosis [2,6,13]. It usually shows hyperactivity at a mouth height without uptake in the anterior cervical area.

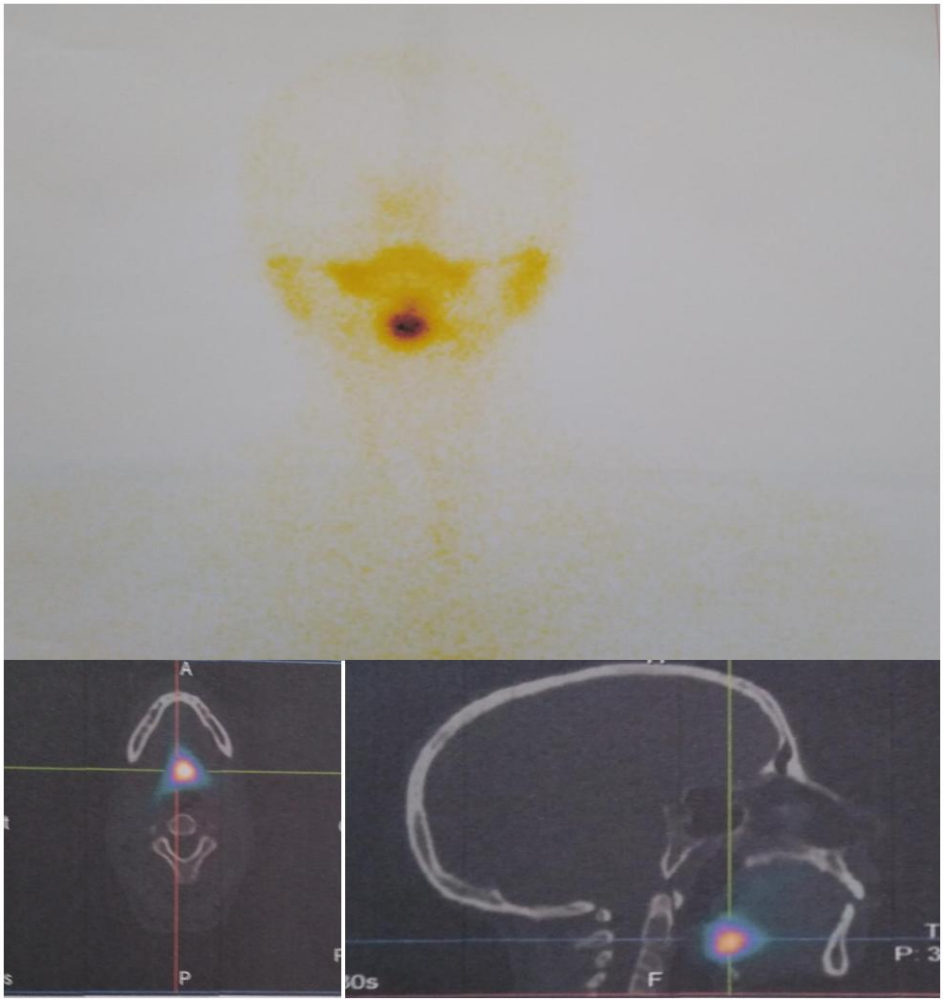

Fig-2: Tc99m thyroid scintigraphy showing the presence of ectopic thyroid tissue at the base of the tongue

On ultrasound, the thyroid is very difficult to detect when in lingual position as was the case for this patient. In eutopic localization, prehyoidal, it appears as a median, solid tissue nucleus, more or less homogeneous [2].

The tomodensitometry allow to appreciate the density of the mass, its limits, its seat and its relations with the vascular axes, however, it should be avoided at cause of iodized injection while thyroid scintigraphy has not yet been performed [2].MRI allows to explore the mass, to identify an eventual tumor vascular contingent, to detect perineural extensions and locate it from relative vascular axes [2].

In front of the necessity of a surgical gesture or in case of suspicion of neoplastic transformation, dimensions and relations of mass to the surrounding tissues should be specified by MRI or tomodensitometry [2]. The occurrence of thyroid carcinoma on Ectopic thyroid is very rare [1%] [9]. The first case was reported in 1997 by Grunn and Rutgers. On a thirty cases described later in the literature, the histological type was not mentioned in 16 cases. The papillary variant has been reported in one case, the cellulo-squamous carcinoma in 2 cases and vesicular carcinoma in 10 cases [9].

Treatment is based on hormonal substitution for life allowing the thyroid tissue to be rest and avoid surgical treatment [2] who is rarely necessary except in case of suspicion of neoplastic transformation, obstruction airway, severe and repeated haemorrhage or significant dysphagia [1 2, 5]. In order to prevent the development and/or recurrence of lingual goiter, administration of L-thyroxine is also indicated in case of incomplete excision [2].

Autotransplantation of thyroid tissue in the cervical region or abdominal muscles has been described in rare cases [5].The success rate is estimated at 30% [2]. Ablative Iratherapy is an alternative to surgical intervention in case of refusal or inoperability. It is recommended to prescribe a temporary corticosteroid therapy to avoid symptoms related to post-radiation thyroiditis [2].

CONCLUSION

The rarity of this embryological anomaly and its locoregional and functional consequences need to

discuss the diagnosis in front of hypothyroidism without a palpable thyroid.

Diagnosed early, the basal-lingual thyroid has an evolution most often favorable, especially under hormone replacement therapy. Regular annual clinical and biological surveillance is essential. The severity of this pathology concerns tumor forms with compressive and hemorrhagic signs requiring surgery, as well as the degenerative evolution of the tumor. In most cases, patients remain asymptomatic and thyroid ectopia passes unnoticed.

Currently, thanks to advances in imaging and neonatal screening for hypothyroidism, the prognosis of thyroid ectopia, when properly managed, is considered good.

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