

## Case Report

# Lingual thyroid with hypothyroidism in a child

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## ABSTRACT

Ectopic thyroid is an uncommon embryological abnormality characterized by the presence of thyroid tissue in a site other than its usual pretracheal location. Of all ectopic thyroids 90% are found to be lingual. Lingual thyroid is estimated to occur in 0.2 per cent of normal children, being more common in females. It is a rare congenital anomaly appearing with prevalence of 1:100000. This embryological anomaly originates from failure of thyroid gland to descend from foramen caecum to its normal pre laryngeal site. Interestingly, only 0.01% of these patients present with any overt symptoms. If symptomatic: dyspnoea dysphagia, dysphonia and stomatolalia are some common clinical features associated with it. In females these symptoms manifest during stress as in pregnancy, puberty and menstruation. Some rare clinical presentations reported in literature are hyperthyroidism, malignancy and hemoptysis. Hereby authors are reporting a 7-year-old female child who presented to our department with dysphagia who on evaluation diagnosed to have lingual thyroid with hypothyroidism.

**Keywords:** Dysphagia, Ectopic thyroid, Lingual thyroid

## INTRODUCTION

The thyroid gland is one of the largest endocrine glands in the body and lies approximately at the same level as the cricoid cartilage.<sup>1,2</sup> Lingual thyroid is defined as the presence of thyroid tissue in the midline anywhere between the circumvallate papillae of tongue and the epiglottis. The condition arises from the embryonic failure of normal thyroid tissue to descend from the foramen cecum area of the tongue base through the lower neck and presents as a lobular midline mass in the mucosal surface of the tongue base.<sup>3</sup> Lingual thyroid is a rare anomaly with an incidence of 1 in 3000 of the thyroid cases seen and with overall prevalence of 1 in 100,000. Of all ectopic thyroids 90% are found to be lingual thyroids.<sup>4</sup> Very rarely, two ectopic foci may be present simultaneously.<sup>5,6</sup> Approximately two thirds of the patients with lingual thyroid lack thyroid tissue in

neck. Lingual thyroid is four times more common in females than in males. Diagnosis of lingual thyroid includes local examination of the tongue base associated with the absence of normally located gland, and imaging examinations. Imaging studies include neck ultrasound, neck CT-scan, neck-IRM and scintigraphy using Tc 99m, I-131, I-123.<sup>7</sup> The main differential diagnosis are haemangioma of tongue, thyroglossal duct cyst, dermoid cyst, lipoma, lymphadenopathy, lymphangioma, cystic hygroma, besides neoplasms.

## CASE REPORT

A seven year old female child came to authors department with progressive dysphagia These symptoms were present for three months and gradually increasing every day. The past medical history of the patient was insignificant. On examination, the patient presented a

solid, spherical mass with 3 cm of diameter, covered with intact mucosa, located at the base of the tongue (Figure 1 and 2). Examination of the neck revealed no palpable thyroid gland in the normal pretracheal position and no cervical lymphadenopathies.



**Figure 1: Solid mass seen in the base of tongue.**



**Figure 2: Solid spherical mass with intact mucosa seen in the base of tongue.**

Ultrasound neck revealed absence of thyroid in its normal anatomic location. TSH (Thyroid Stimulating Hormone) levels were elevated 15.2 uIU/ml (Reference range-0.49 to 4.67 uIU/ml), FT<sub>4</sub> (Free Thyroxine<sub>4</sub>) was borderline low at 0.9 ng/dl (Reference range-0.89 to 1.78 ng/dl). Other biochemical parameters were non-contributory. A radionuclide scan was carried out using 99m Tc pertechnetate intravenously. Tracer accumulation was seen in the lingual region in midline confirming the presence of lingual thyroid. There was no activity present in the thyroid bed.

She was diagnosed as a case of Lingual thyroid with juvenile hypothyroidism and placed on L-Thyroxine 50 mcg OD. A follow-up protocol was established, Patient was advised re-evaluation after 3 months to titrate the L-thyroxine dosage. There was significant improvement in dysphagia but the child still had foreign body sensation in the throat. TSH (Thyroid Stimulating Hormone) levels were at 5.9 uIU/ml (Reference range-0.49 to 4.67 uIU/ml), FT<sub>4</sub> (Free Thyroxine<sub>4</sub>) was at 1.1 ng/dl (Reference range-0.89 to 1.78 ng/dl). Hence the dose was increased to 75 mcg OD following which child became completely asymptomatic and she is on regular follow up.

## DISCUSSION

The thyroid tissue reaches the normal location in the pretracheal region by migrating caudally from the foramen cecum in the tongue base at the seventh week of fetal life. Ectopic lingual thyroid is caused by noncompletion of this migration.<sup>8,9</sup> Lingual thyroid tissue is the most frequent ectopic location of the thyroid gland, although its clinical incidence is low with 1 in 100 000 cases occurring.<sup>10</sup> There are four groups of lingual thyroid: lingual, sublingual, thyroglossal, and intralaryngeal. Endocrine changes such as puberty, pregnancy, and menstruation can lead to an increase in gland size and symptoms. This explains why lingual thyroid is 7-fold higher among women.<sup>11</sup>

Lingual thyroid does not usually lead to any symptoms unless an increase in gland size occurs. In symptomatic cases, patients present with complaints of dysphagia, dysphonia, foreign body sensation in the throat, cough, pain, bleeding, and dyspnoea.<sup>9,12,13</sup> Rarely, the lingual thyroid may cause hyperthyroidism or be the site of thyroid cancer.<sup>14,15</sup> The fact that the ectopic thyroid tissue can be the only functional thyroid tissue must be kept in mind when determining the therapeutic approach. Asymptomatic cases can be monitored with suppressive hormonal therapy aiming for reduction of ectopic tissue volume. Decrease in symptoms can occur with suppressive treatment in some cases. Conservative treatment had proved its efficiency in many studies.<sup>11,12,16-18</sup> Indeed, administration of a suppressive dose of thyroid hormones aims to decrease the TSH level, therefore it can reduce the ectopic glandular volume and consequently reduce all the compressive symptoms.

As long as you do not get an adequate response to clinical treatment, the method of surgical thyroidectomy is so preferred. Other indications for surgery are on the basis of urgency: dysphonia or dysphagia with decreased oral intake, suspicion of malignancy, cystic degeneration, uncontrolled hyperthyroidism and repetitive or severe bleeding. However, even more experienced hands, total excision of ectopic thyroid tissue is extremely difficult regardless of the approach.<sup>19</sup>

There is no consensus in the literature about the best therapeutic strategy, given the rarity of the disease and to

the limited number of cases described. Treatment depends on the size, the presence or absence of symptoms and complications like ulceration, hemorrhage, malignancy or obstruction of the upper airway. The fact that ectopic thyroid may or may not be the only functional thyroid tissue must also be valued to determine the therapeutic approach. The main objective will be to relieve the obstruction of upper airways without putting the patient's life at risk or leave you with scars mutilating.<sup>20-22</sup>

## CONCLUSION

Children who presents with dysphagia or foreign body sensation ectopic lingual thyroid must be taken into consideration and the approach of hormone suppression can be tried first line like in our patient who had dramatic improvement without surgery. Therapeutic approach should be considered according to symptomatology.

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