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Case Report

# **Lingual Thyroid**

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#### **ABSTRACT**

Lingual thyroid is due to an aberration in the normal development of thyroid. Ectopic thyroid is defined as the presence of thyroid tissue in a location other than the normal anterior neck region between 2<sup>nd</sup> and 4<sup>th</sup> tracheal rings. It is due to a defect in thyroid diverticulum migrating from the base of the tongue, to its final pretracheal position. Here is a case of lingual thyroid presented as a swelling at the base of the tongue, of size 2x3cm since 2 months. Diagnosis of ectopic lingual thyroid was made. Tc99 pertechnetate radioisotopes can confirm ectopic lingual thyroid. There was no thyroid tissue in the normal location. Hence ectopic Lingual thyroid was the only functioning thyroid. Management was by Surgical excision of the gland and thyroxine replacement.

*Key words:* Lingual thyroid, Ectopic thyroid, Tc<sup>99</sup> pertechnetate scan.

## INTRODUCTION

In 1869 Hickman reported first case of ectopic thyroid at the base of tongue pressing down the epiglottis on the larynx and causing death by suffocation 16 hrs after birth. [1] Ectopy of thyroid is defined as an aberrant localization of thyroid tissue outside the thyroid compartment. Lingual thyroid is most common type of ectopic thyroid accounting for almost 90% of the cases. Incidence of lingual thyroid is one in 1,00,000 cases. [2] There are four groups of lingual thyroid: lingual, sublingual, intralaryngeal, thyroglossal. [3] Diagnosis is clinical examination based on radioisotope scanning. Radio nucleotide thyroid imaging employing technetium  $^{99m}$  pertechnetate,  $I^{131}$ ,  $I^{123}$  is useful in the evaluation for ectopic thyroid. differential diagnosis for lingual thyroid is lipoma, dermoid cyst, heamangioma of tongue, lymphangioma, thyroglossal duct

cyst and lymphadenopathy. Here we present a case of lingual thyroid; An eight year-old female patient presented with a chief complaint of noticing a swelling at the base of tongue since two months. Ectopic thyroid was suspected and confirmed by Tc99-m pertechnetate scan. Management was by surgical excision followed by thyroxine replacement.

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#### **CASE REPORT**

An eight-year-old female patient came to surgical OPD with a chief complaint of noticing a swelling at the base of tongue since two months. This was not associated with any symptoms, no similar complaints in her family. Patient was euthyroid. Oral cavity examination revealed 2x3 cm solitary swelling at the base of the tongue, extending more towards the right side. Swelling margins are regular, surface is smooth, soft and nontender fixed to the

tongue, non compressible, not bleeding on touch. (figure:1)



Figure: 1 Diagnosis of ectopic lingual thyroid was made.

On investigation with Tc99 pertechnetate radioisotope scan; ectopic lingual thyroid was diagnosed. (figure: 2) There was no thyroid tissue in normal location at anterior part of neck



Figure: 2 Patient was managed by surgical excision of the lesion and thyroxine replacement.

# **DISCUSSION**

Lingual thyroid is due to aberration in the normal development of thyroid. Normally the thyroid development starts on 24<sup>th</sup> day of gestation. The thyroid gland has two diverse cell types which originate from two different embryological structures: thyroid anlage ultimobranchial body which are the sites of origin of thyroid follicular cells and C cells respectively. It develops from endodermal diverticulum from the median plate of floor of the pharyngeal gut. This diverticulum descends in the midline and reaches its final position in front of the trachea in the seventh week of gestation. [4] Aberration in descent of thyroid development leads to ectopic thyroid. Genetic research has shown that the gene transcription factors TITF-1(Nkx2-1), Foxe1 (TITF-2) and PAX-8 are essential for thyroid differentiation and morphogenesis. [4,2] Mutation in these genes may be involved in ectopic thyroid. Maternal antibodies against thyroid are also implicated aberration in thyroid development. [1]

Lingual thyroid is the most common type of ectopic thyroid accounting for almost 90% of the cases. Incidence of lingual thyroid is one in 1,00,000 cases. [2] In 1869 Hickman reported first case of ectopic thyroid at the base of tongue pressing down the epiglottis on the larynx and causing death by suffocation 16 hrs after birth. [1] The other locations of head and neck region where ectopic thyroid tissue may be found includes trachea, submandibular, lateral cervical region axilla, palatine tonsils carotid bifurcation, iris of the eye, pituitary gland. [5-8] Other places where ectopic thyroid reported in literature include heart. gallbladder, uterus, vagina, adrenal gland, duodenum. [9-11]

Lingual thyroid, though usually asymptomatic, can present with cough, pain, dysphagia, dysphoea, dysphonia and hemorrhage, [12,13] Adults having lingual thyroid may present with sleep apnea and respiratory obstruction. [14] The clinical manifestations of lingual thyroid peek at a mean age of 40 years with two statistical peaks at 12and50years. [13]

Radio nucleotide thyroid imaging employing technetium <sup>99m</sup> pertechnetate, I<sup>131</sup>, I<sup>123</sup> is useful in the evaluation for ectopic thyroid. To 99-m pertechnetate vields better quality imaging and at the same time delivers a lower radiation burden to body compared to I<sup>131</sup>. I<sup>123</sup> hence is widely used for ectopic thyroid in the children. But the higher cost and shorter half life makes its availability more difficult. High resolution ultrasonography is also used for evaluation of the ectopic thyroid. Other investigations used for ectopic thyroid are CT and MRI. Usually, biopsy and FNAC are not recommended because of risk of bleeding and infection but if malignancy of thyroid is suspected we need to perform a biopsy and FNAC for better diagnosis. [6,7] Thyroid function tests are useful for serum levels of thyroid hormones.

Asymptomatic euthyroid patients with ectopic thyroid do not require any

treatment but do need to be kept under observation. Surgical intervention indicated for severe obstructive symptoms, cystic degeneration, ulceration, bleeding and malignancy conditions. [15] The ectopic thyroid maybe the only functioning thyroid in the patient, so determination of normally located thyroid gland is necessary for planning removal of ectopic thyroid to avoid hypothyroidism and lifelong hormone replacement. To retain the thyroid tissue in the patient, different procedures These described in literature. are: transposition of ectopic thyroid with vascular pedicle flap into lateral pharyngeal wall; muscular space at the floor of mouth, anterior rectus sheath and strap muscles. [16,17] Even after auto transplantation of of the patients require thyroid 70% exogenous thyroid hormone replacement. Surgical excision can be made either transorally or externally with pharyngotomy through a trans hyoid approach. [18,19] Other methods are transoral laser excision. [20,21] Trans oral radiofrequency ablation may also reduce the tissue volume. [22] Radioiodine is an alternative to surgical excision of ectopic thyroid indicated in patients who are unfit or unwilling for surgery. [23] Treatment of ectopic thyroid with hyperthyroidism can be treated with anti-thyroid drugs or surgical excision, or radioactive iodine, which can also be used in patients' refractory to antithyroid drugs. <sup>[24]</sup>

# **CONCLUSION**

A rare case of Lingual thyroid presenting as the only functioning gland in the absence of normal thyroid is presented.

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