

RUPA MAHARJAN
AMIYA K SHAH

Department of ENT & Head and Neck Surgery
National Academy of Medical Science
National Trauma Center
Kathmandu, Nepal.

Corresponding Author:

Dr Rupa Maharjan

Department of ENT & Head and Neck Surgery
NAMS, National Trauma Center, Nepal.
Email: drupamaharjan@gmail.com

ECTOPIC LINGUAL THYROID

ABSTRACT

Lingual thyroid is a rare embryological anomaly. It develops from failure of the thyroid gland to descend from the foramen caecum to its normal pre-laryngeal site. The ectopic gland located at the base of the tongue is often asymptomatic but may cause local symptoms such as dysphagia, dysphonia with, upper airway obstruction and hemorrhage. We presented a case of a 9 year-old girl, she complained of sensation of a foreign body in throat and progressive difficulty in swallowing and occasional difficulty in breathing. The diagnosis of lingual thyroid was made clinically and radionuclide scanning. It was confirmed with histopathological finding. In this case report, presentation, diagnosis and management of this conditions are highlighted.

Keywords: Ectopic, Lingual, Thyroid.

INTRODUCTION

Lingual thyroid is a rare embryological anomaly and originates from failure of the thyroid gland to descend from the foramen caecum to its normal pre-laryngeal site. The presence of thyroid tissue anywhere along its embryologic path other than the usual thyroid position in the neck is defined as heterotopic thyroid tissue.^{1,2} Ectopia occurs during descent of the thyroid from the foramen caecum to the normal position, as a result of maternal antibodies inhibiting thyroid antigens or persistent thyroglossal ductal epithelial tissue. Although ectopic thyroid tissue is rare, with a reported incidence of 1 in 300,000, it is most frequently reported between the foramen caecum and the normal position of the thyroid gland, often in the base of the tongue, as a lingual thyroid.^{1,3} The other common sites for an ectopic thyroid glands are thyroglossal duct, the coupling between the foramen cecum and thyroid gland, and laryngotracheal region. It occurs more frequently in females.⁴ The ectopic gland located at the base of the tongue is often asymptomatic but may cause local symptoms such as dysphagia, dysphonia, upper airway obstruction and hemorrhage often with hypothyroidism.⁵ we present a case of 9 year-old girl with ectopic lingual thyroid.

CASE REPORT

A 9-year-old girl presented with complaints of sensation of a foreign body in throat associated with difficulty in swallowing and occasionally difficulty

in breathing for the last seven months. There was no medical history related to hypothyroidism like constipation, lethargy, weight gain, no history of delayed milestones and mental retardation. She weighed 38 kg and her height was 105 cm. On oral cavity and Oropharynx examination revealed a solid, pink, spherical mass, covered with intact mucosa with prominent blood vessels, located at the base of the tongue, measuring 3 cm × 2 cm × 2 cm (Fig.1), which was firm, non-tender and

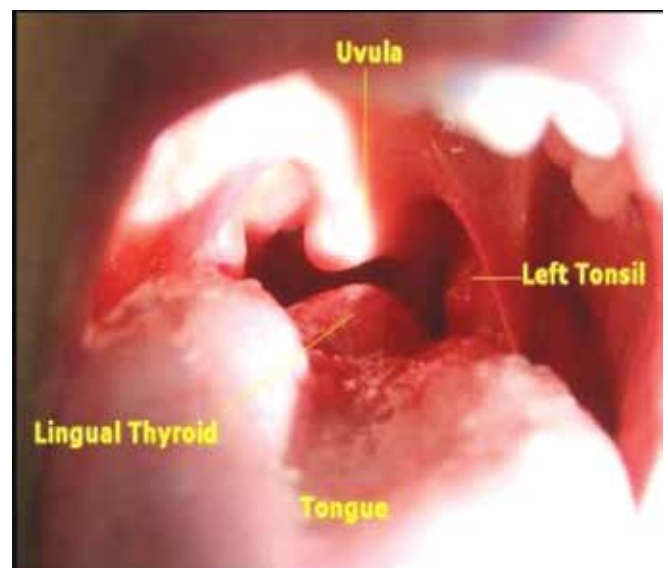


Figure 1. Swelling at the base of tongue

fixed to the underlying tissue. Clinical examination of the neck revealed no palpable thyroid gland in the normal pretracheal position and no cervical

lymphadenopathy. Ultrasonography scan of the neck showed absence of the thyroid in its normal anatomical position. Thyroid function tests showed T3 (2.25pg/ml) and T4 (0.29 ng/dl) while TSH was elevated (27.68m IU/ml). Other laboratory tests were within normal range. Thyroid scan with technetium Tc-99m sodium was performed, showing no tracer uptake in the region of thyroid bed and focus of tracer uptake were seen at the base of the tongue (Fig. II).

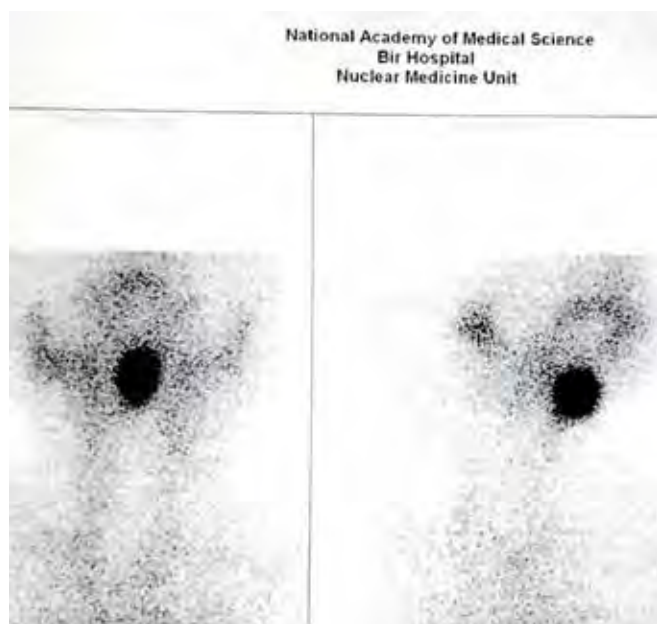


Figure II. Preoperative thyroid Scan

She was diagnosed as ectopic lingual thyroid with subclinical hypothyroidism. Endocrinologist started thyroxine sodium 75 mcg once daily in order to maintain the euthyroid state. Patient was re-evaluation after 3 months. After 3 months thyroid function tests was done, which was within normal range. There was mild increase in swelling size with progressive dysphagia. Even after use of 75 mcg OD for 3 months, patient's symptom was progressive. So she was planned for trans oral excision of lingual thyroid under general anesthesia. She underwent partial resection of mass and continued the thyroxin 75 mcg in postoperative period. Postoperative period was uneventful and discharged after three days of hospital stay. Histopathology of specimen showed tongue tissue lined by squamous epithelium with lymphoid follicles in sub epithelial stroma, minor salivary gland and skeletal muscle tissues. There were islands of dilated follicles lined by cuboidal to flattened follicular cells. Some of follicles were filled with colloid. Patient was followed on 7th Post-operative day and there was marked

improvement in her symptoms of dysphagia and dyspnea but still persistence of mild discomfort in throat. After three months following surgery patient presented with puffy face, lethargy, excessive sleep and weight gain but there was no complaints of dysphagia, dyspnea and discomfort. Thyroid function test showed TSH >100 IU/ml, so thyroxin dose was increased to 150 mcg. At six months following surgery showed normal thyroid function test and clinical symptoms were improved. Then patient was switched on to 50mcg thyroxin by endocrinologist team. She was followed up at one year, one and half year and two year after surgery without any complains and with normal thyroid function.

DISCUSSION

Lingual thyroid is developmental disorder due to failure of descend of thyroid gland from foramen caecum of tongue to prelaryngeal site in neck. A lingual thyroid, situated at the base of the tongue is the most common location of ectopic thyroid tissue, accounting 90% of reported cases. The mean patient age at presentation of lingual thyroid is 40.5 years, ranging from birth to 83 years.⁶ But in our case it was seen in nine years of age. The prevalence of thyroid ectopia is higher in females than in males.⁷ In our case it was seen in a girl. When a lingual thyroid is found, the usual thyroid bed location must be evaluated because orthotropic thyroid tissue will be absent in 70%–80% of the time and subsequent resection of the ectopic thyroid tissue will render the patient athyroid.⁸ In our case also lingual thyroid was only the thyroid tissue present in her body. Ectopic thyroid may become goitrogen and may also be associated with clinically evident thyroid dysfunction, which could be either hypofunction or hyperfunction. Ectopic thyroids are reported to have malignant transformation. The treatment of ectopic thyroid depends on factors such as mass size, local symptoms, age of the patient and functional status of thyroid gland. Rarely, benign or malignant neoplastic changes can occur. One must maintain a high degree of suspicion in any patient presenting with intrapulmonary nodules.⁹ It is rare, but there have been reports of ectopic tissue in the lung, submandibular region, the trachea, the mediastinum, the heart, the duodenum, the adrenal gland, the parotid gland and the gall bladder.³ In cases of lateral ectopic

thyroidal tissue on the neck, metastases should be excluded. Thyroiditis has also been reported in ectopic thyroid tissue. Identification of ectopic thyroid gland should be considered for choosing the best treatment in the ectopic thyroid cases.⁶ Since an inaccurate preoperative diagnosis can result in hypothyroidism.^{3,7} An ectopic thyroid was reported incidentally in the pancreas of a 50-year-old woman, who underwent a bilateral truncal vagotomy and pyloroplasty for a duodenal ulcer. She had history of total thyroidectomy.¹⁰ It is often asymptomatic, and the most common symptoms are due to growth of the lingual thyroid tissue. This growth can occur as a result of metabolic stresses, including puberty, pregnancy, trauma and menopause. Sometimes it can cause symptoms with mass effect such as dysphagia, dysphonia, stridor, dyspnea, hemorrhage, and hoarseness.⁸ Ectopic thyroid may progress to malignant papillary thyroid carcinoma.⁹ Less than 1% of ectopic thyroids are reported to have malignant transformation and include all histologic variants with the exception of medullary carcinoma. Papillary carcinoma is the most common histologic type, accounting for approximately 85% of the cases.¹¹

CONCLUSION

Lingual ectopic thyroid is a rare developmental anomaly. Treatment could be conservative with hormone therapy if the symptoms are mild, while surgery is recommended in cases with moderate to severe symptoms. Evaluation of thyroid function is recommended before and after surgery due to the risk of preoperative and post-operative hypothyroidism.

When complications such as dysphagia or dyspnea occur due to lingual mass, surgery is necessary. The trans-oral approach can be useful in selected cases. Transplantation of tissue is not recommended but substitutive hormone treatment is often needed. Follow-up is recommended to monitor possible recurrence or complications.

REFERENCES

1. Lianos G, Bali C, Tatsis V, Anastasiadi Z, Lianou E, Papathanasiou V et al. Ectopic thyroid carcinoma. *G Chir.* 2013;34(4):114-116.
2. Abujrad H, Olberg B, Ooi TC. Heterotropic Pulmonary Thyroid in the Presence of a Normally Located Multinodular Goitre. *J Clin Case Rep.* 2012;2(7):134.
3. Bertha A, Hephzibah J, Shanthly N. Ectopic thyroid - a case series. *Journal of Clinical and Diagnostic Research.* 2011;5(2):291-293.
4. Stokić E, Kljajić V, Idjuški S, Benc D, Popović D, Protić M et al. Dysfunctional ectopic thyroid gland: a case report. *Srp Arh Celok Lek.* 2014 Nov-Dec;142(11-12):724-7
5. Toso A, Colombani F, Averono G, Aluffi P, Pia F. Lingual thyroid causing dysphagia and dyspnoea. Case reports and review of the literature. *Acta Otorhinolaryngol Ital.* 2009 Aug;29(4):213-7
6. Noussios G, Anagnostis P, Goulis DG, Lappas D, Natsis K. Ectopic thyroid tissue: anatomical, clinical, and surgical implications of a rare entity. *Eur J Endocrinol* 2011;165(3):375-382.
7. Amanda K. Heterotropic Thyroid. *Journal of Diagnostic Medical Sonography.* 2004;20 (2):120-123.
8. Di Benedetto V. Ectopic thyroid gland in the submandibular region simulating a thyroglossal duct cyst: a case report. *J Pediatr Surg* 1997;32(12): 1745-1746.
9. Özbek T, Çetin NK, Kanlioglu NK, Sen S, Erdogdu IH, Meteoglu I. Ectopic Thyroid Tissue in the Lung. *Journal of case reports.* 2016;6(3):321-324.
10. Erhun Eyübolu, Metin Kapan, Turgut Ipek, Yılmaz Ersan. Ectopic thyroid in the abdomen: Report of a case. *Surgery Today.* 1999;29(5):472-474. |
11. Matsumoto K, Watanabe Y, Asano G. Thyroid papillary carcinoma arising in ectopic thyroid tissue within a branchial cleft cyst. *Pathol Int.* 1999;49(5):444-6