

DOI: 10.5455/2320-1770.ijrcog20150437

Case Report

## Torrential bleed from lingual thyroid: an unusual cause of IUD

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**Received:** 22 February 2015

**Accepted:** 01 March 2015

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### ABSTRACT

Lingual thyroid is rare anomaly which results from failure of descent of thyroid gland to normal position in neck. The common mode of presentation is foreign body sensation, dyspnoea, dysphagia or rarely haemorrhage. Acute severe haemorrhage causes hypotensive shock which may lead to intra-uterine foetal distress or foetal death. We hereby present, a case of 22 year old primigravida who presented with massive oral haemorrhage secondary to hypertrophied lingual thyroid. Patient was successfully managed conservatively with oral thyroxine.

**Keywords:** Lingual thyroid, Superior thyroid artery, Thyroxine, IUD

### INTRODUCTION

Ectopic lingual thyroid is a rare anomaly with an incidence of 1:100000 to 1:300000 with more female preponderance of 4:1 to 7:1.<sup>1</sup>

Most common presentation of lingual thyroid is as a foreign body sensation in throat. It may also present with dyspnoea or dysphagia. It rarely presents as profuse oral bleed.

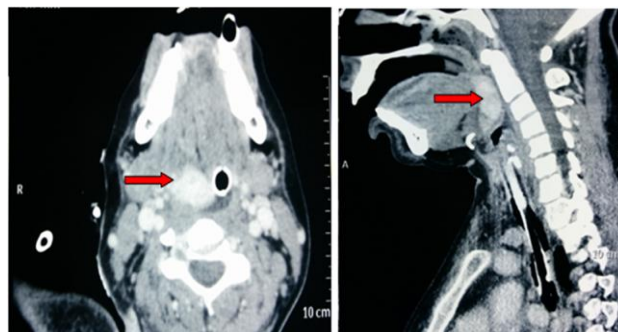
Here, we present a case of young primigravida who presented with oral bleed secondary to hypertrophied lingual thyroid.

### CASE REPORT

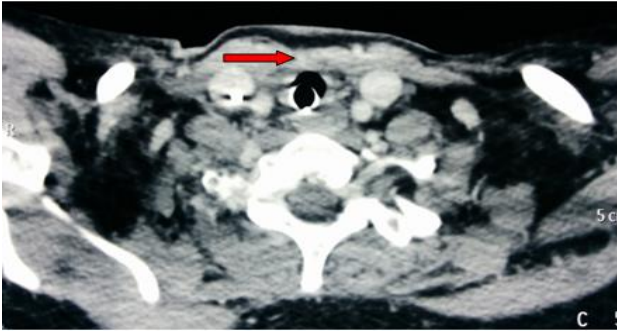
22 year old pregnant female with 8 months of gestation presented with profuse oral bleed to our emergency department. Immediate intubation to secure the airway was done. Complete oral packing with betadine soaked roller pack was done as no pin point bleeder was seen. Foetal heart sounds could not be appreciated at time of presentation. Careful examination revealed a firm globular swelling in the base of tongue. There was no

previous history of similar episodes or bleeding from any other orifice. There was no history of dysphagia or odynophagia. Coagulation profile was done which showed that the platelet counts were 60000 with deranged prothrombin time and activated partial thromboplastin time.

CT scan from base skull to mediastinum was done which revealed a globular swelling in the region of base of tongue which enhanced on contrast enhancement.



**Figure 1: Axial and sagittal CT scans of intubated patient showing lingual thyroid.**



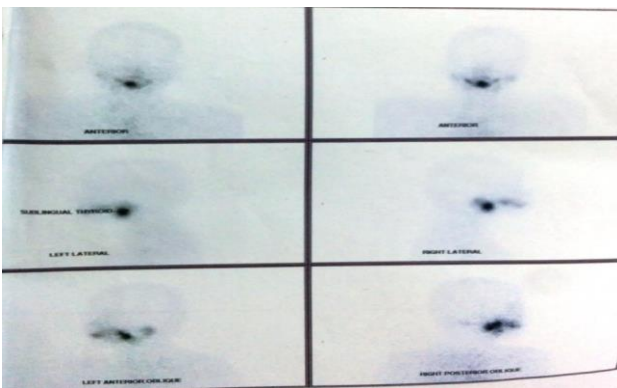
**Figure 2: Axial CT showing absence of thyroid tissue in normal cervical position.**

There was absence of thyroid gland in the neck. CT angiography revealed the feeder for this mass was bilateral lingual artery. There was absence of superior thyroid artery and the first branch arising from the external carotid was lingual artery. Inferior thyroid artery were also absent bilaterally with aplasia of costocervical trunk on both sides.



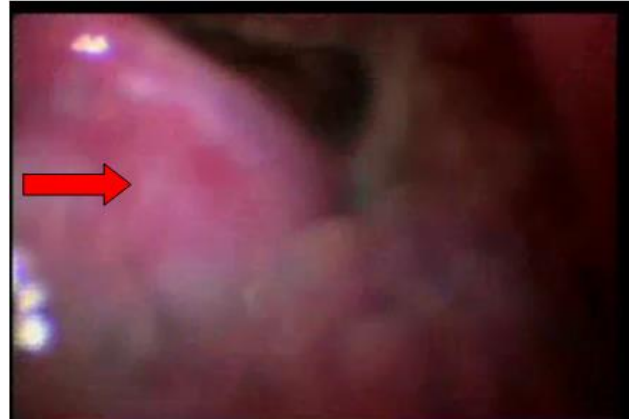
**Figure 3: CT angiography showing absence of superior thyroid artery and inferior thyroid artery. The first branch from the external carotid artery is lingual artery.**

Patient was electively tracheostomised on third day of intubation. Elective induction of labour to expel the dead foetus with products of conception was done. The haematological profile was corrected. Patient was then posted for radioiodine scan which showed intense take up by the lesion present in the tongue suggestive of ectopic lingual thyroid and there was no uptake in the neck.



**Figure 4: Radio-iodine scan showing lingual thyroid.**

Patient was explained about the nature of the anomaly and was advised to take 125 micrograms of thyroxine orally to revert back the hypertrophied thyroid gland and follow up regularly especially when planning for next pregnancy.



**Figure 5: lingual thyroid on 70 degree Hopkins endoscopy.**

## DISCUSSION

Ectopic thyroid is a rare developmental anomaly. The ectopic thyroid can be found anywhere between the foramen caecum to pre-tracheal region.<sup>2</sup> The most common ectopic site is base of tongue (90%).<sup>3</sup> Other possible sites of ectopia are sublingual, thyroglossal, intralaryngotracheal, mediastinal, esophageal, heart and cervical lymph nodes.<sup>3</sup> The incidence of lingual thyroid is 1:100000 to 1:300000.<sup>5</sup> It is four times more common in females than in males, and mean age of clinical manifestations is 40 years. The first case of lingual thyroid was described by Hickman (1869).<sup>6</sup>

Embryologically, thyroid gland develops from the endodermal tissue in the floor of pharynx between tuberculum impar and hypobranchial eminence during the 3<sup>rd</sup> to 7<sup>th</sup> week of development. The hypo-brachial region later develops into foramen caecum. This developing tract (thyro-glossal tract) descends through the body of hyoid to the pretracheal region till the end of the 7<sup>th</sup> week of gestation. Any aberration during this course of descent results in the ectopic thyroid.

The primordium of the gland receives its blood supply from the arteries that later give rise to the lingual and submental vessels. During its descent, the gland establishes new connections with the nearest vessels as older connections are attenuated and disappear. Persistence of these vessels at any stage in the migration of the gland explains the anatomic variations of its arterial supply.<sup>7</sup>

Normal development and migration of thyroid gland needs an intact Tbx1-Fgf8 pathway. This pathway has been identified as the key regulator of development of human thyroid gland. Tbx1 regulates the expression of

Fgf8 in the mesoderm, it is postulated that Fgf8 mediates critical Tbx1-dependent interactions between mesodermal cells and endodermal thyrocyte progenitors. Tbx1 is not expressed by thyroid primordium, but is strongly expressed by the surrounding mesoderm. It is also expressed by pharyngeal endoderm lateral to thyroid primordium. Thyroid organogenesis associated with the expression of a set of transcription factor encoding genes. They include Nkx2-1, Foxe1, Pax8 and Hhex1 genes. Expression of these genes in thyroid primordium is also dependent on Tbx1 gene expression.<sup>8</sup>

Burkhard et al. in their research on thyroid gland development in animal models concluded that in hedgehog-deficient mice, thyroid tissue always develops along the ectopically and asymmetrically positioned carotid arteries, suggesting that, in mice (as in Zebra fish), co-developing major arteries define the position of the thyroid. Hence, the involvement of vessels in thyroid relocalisation sheds new light on the interpretation of congenital thyroid defects in humans. This group concluded that the ventral aorta acts as an instructive structure for positioning thyroid tissue along the AP axis.<sup>9</sup>

In our case, we found anomaly in the development of cervical vasculature. There was absence of superior thyroid artery and inferior thyroid artery on both the sides along with aplasia of costo-cervical trunk on both sides. This finding strengthens the view of Burkhard et al. that the development of vessels especially the branches of ventral aorta influences the descent of the thyroid gland during embryogenesis.

Mostly lingual thyroid is asymptomatic and present generally due to puberty, pregnancy or stress. The most common symptoms are bleeding from the mass, dysphagia, foreign body sensation in throat and rarely dyspnoea or dysphonia. In our case, patient presented with massive recurrent attacks of oral bleed.

On clinical examination, lingual thyroid presents as a globular swelling on the base of tongue or may be diagnosed on palpation of the tongue base. It can be visualised on indirect laryngoscopy or 70 degree Hopkins rigid endoscopy.

The investigation of choice is radio-iodine thyroid scan to prove the ectopic lesion as thyroid but generally preceded by a CT scan of base of skull to mediastinum. In cases of lingual thyroid, CT scan will show absence of thyroid in the neck and presence of a soft tissue in the base of tongue which enhances on contrast. Generally the patients with ectopic thyroid are subclinical hypothyroid and need thyroid supplementation.

The line of management for lingual thyroid can be either conservative or surgical. In conservative line of management, the thyroxine is given at higher doses in order to suppress TSH to low level so that the stimuli for hypertrophy and increased vascularity are cut-off. This results in shrinkage of the gland with reduced vascularity

preventing any further episodes of bleed. We started the patient on oral thyroxine 125 micrograms per day and asked the patient to follow up as there were no complains of dysphagia or dyspnoea.

The surgical line of management is resorted to in cases of severe dysphagia and dyspnoea as well as in cases where the patients do not respond to medical line of management. The various different approaches for the surgical management are transmandibular translingual approach, lateral pharyngotomy and suprahyoid myotomy approaches. Thyroid transposition is also an option but is only indicated in euthyroid patients with ectopic thyroid.

Selective bilateral lingual artery embolisation is also favored pre-operatively to reduce the blood loss during the surgery.

Role of radioiodine ablation for management of lingual thyroid is still being explored but no definite guidelines for the role of radioiodine are defined.

## CONCLUSION

Lingual thyroid should be kept in differentials as a cause for inadvertent oral bleed during the gestation period. Radio-iodine scan is diagnostic test for the position of ectopic thyroid. Thyroxine supplementation is first line of management. Surgical line of management is resorted to only if symptoms persist.

*Funding: No funding sources*

*Conflict of interest: None declared*

*Ethical approval: The study was approved by the institutional ethics committee*

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**Cite this article as:** Bihani A, Dokhe Y, Hardikar P, Dabholkar J. Torrential bleed from lingual thyroid: an unusual cause of IUD. *Int J Reprod Contracept Obstet Gynecol* 2015;4:470-3.