# Lump at the back of my tongue. The lingual thyroid – a case report & narrative review of literature

Joe Jabbour<sup>1</sup>, Nelson Agostinho<sup>1</sup>, Harinder Bains<sup>1</sup>, Peter Earls<sup>2</sup>, Louise Killen<sup>2</sup>, Ruwan Perera<sup>1</sup>, Ranga Sirigiri<sup>1</sup>, and Hayder Ridha<sup>1</sup>

<sup>1</sup>Dubbo Base Hospital <sup>2</sup>St Vincent's Hospital Sydney

January 26, 2021

#### Abstract

A 68-year-old male presented with a difficult airway secondary to a firm mass at the base of the tongue on intubation for an emergency general surgical procedure. Further workup confirmed a lingual thyroid with previous upper airway obstruction requiring a temporary tracheostomy. The mass was excised using transoral robotic surgery.

## Introduction

A lingual thyroid (LT) is a rare anomaly of functioning ectopic thyroid tissue found at the base of the tongue and represents the most common location of functioning ectopic thyroid tissue.<sup>1</sup> The incidence of LT is approximately 1 in 3000 thyroid cases with an overall prevalence of 1 in 100,000. LT is four times more common in females than in males and often manifests in the second decade of life.<sup>1,2</sup> The diagnosis of LT is suspected clinically and confirmed with radionucleotide scanning.<sup>1</sup>

## Case Report

A 68-year-old male presented to a rural emergency department with a 48-hour history of right upper quadrant pain associated with nausea and vomiting on the background of poorly controlled insulin dependent diabetes, hypothyroidism, GORD and hypertension. Regular pharmacotherapy included insulin, pantoprazole, and thyroxine. Investigation revealed cholecystitis with choledocholithiasis. An ERCP was initially performed to relieve the biliary tree obstruction followed by an interval cholecystectomy. At induction for cholecystectomy the anaesthetic team encountered a difficult airway due to a firm mass at the base of the tongue. An airway was eventually established allowing completion of a laparoscopic cholecystectomy without issue. The patient was routinely extubated and had an uneventful post-operative recovery.

During the post-operative period the encountered base of tongue mass was investigated. The patient revealed a history of dysphagia and imminent airway obstruction requiring an emergency tracheostomy 15 years prior to this admission. Nasoendoscopy revealed a large base of tongue lesion with associated displacement of the epiglottis (Figure 1). An ultrasound of the neck revealed no tissue in the thyroid bed with calcified structure at the tongue base. A computer tomography (CT) of the neck was then performed that supported the finding of no tissue in the thyroid bed and a calcified mass at the base of the tongue with a volume of 40cc measuring 29mm (anteroposterior) and 26mm (transverse) (Figure 2). Finally, a technetium 99m sestamibi scan was performed that revealed no uptake in the tongue mass to suggest parathyroid adenoma. Biochemical investigations revealed that the patient was euthyroid. The patient was eventually accepted for a transoral robotic surgery (TORS) at a tertiary centre. This was performed without complication and the lingual thyroid mass sent for histopathology (Figure 3). Histopathology of the lesion revealed oral type

squamous mucosa with subepithelial stroma, thyroid tissue, minor salivary glands and skeletal muscle (Figure 4).

#### Discussion

The mainstay of treatment involves hormone suppression treatment (HST) with exogenous thyroid hormone to induce atrophy of the gland by causing negative feedback in the pituitary/thyroid axis. <sup>3,4</sup>Patients have regular follow up for clinical examination and thyroid function tests. The literature suggests treatment effectiveness in 61% of patients. <sup>2</sup> Surgical excision of the LT is indicated when patients receiving HST remain symptomatic. Another option for patients who fail HST or are not candidates for surgical excision is radioactive ablation with I-131. <sup>5</sup>

Surgical: Excision alone or excision with autotransplantation into muscle

Surgery for LT can be offered for patients with airway compromise, dyspnoea, dysphagia, speech impairment, globus pharyngeus or OSA.<sup>6</sup> Technetium 99 (99mTc) and neck ultrasound will determine whether surgical excision alone is required or excision with autotransplantation into muscle. The three main approaches to surgical excision of lingual thyroid are: transoral, transmandibular, and lateral pharyngotomy.<sup>7</sup> Open surgery has been associated with increased morbidity and prolonged hospitalization. Thus, CO2 laser, electrocautery assisted resection with rigid endoscope and operating microscope and suspension laryngoscopy have been attempted.<sup>8</sup> Although the aforementioned approaches are limited by safe visibility and difficulty in manipulation rendering resection more difficult.

Transoral robotic surgery (TORS) is a safe and feasible minimally invasive approach for excision of the lingual thyroid with larger three-dimensional point of view and easier manipulation due to freedom of motion of robotic instruments.<sup>9,10</sup> The predominant risk with transoral robotic lingual thyroid resection is lingual artery injury. This injury can be prevented by preoperative imaging methods and careful dissection with knowledge of anatomy.<sup>10</sup> The anatomic relationship may be effected by mass effect and thus the routine course of the lingual artery may vary.<sup>10</sup> Absolute local contraindication is limited mouth opening or trismus.

The transmandibular approach for excision of lingual thyroid provides wide exposure of tongue and reduces the need for a tracheostomy. This approach involves lip split mandibulotomy, dissection of mylohyoid muscles to reach the base of the tongue and exposure of lingual mass. The tumour is dissected out and excised. The osteotomized mandible is plated with primary closure of superficial layers. Regardless of the surgical approach, lifelong exogenous thyroid hormone replacement is required.

## Anaesthetic considerations

Securing the airway is a crucial component of LT surgical management. In children inhalation induction of general anesthesia is used to ensure spontaneous ventilation and avoid complications of asleep intubation and inability to secure the airway. A flexible video laryngoscopy, a difficult airway trolley, and a tracheostomy tray must be step up at the bedside. Paralytic agents of skeletal muscle should be avoided to prevent cessation of spontaneous ventilation and increased risk of a surgical airway. The trachea in children can usually be anaesthetised with a deep inhalation agent. Capnography monitoring of exhaled gas analysis is useful in determining the depth of anaesthesia. Fibreoptic awake nasotracheal intubation is the preferred method of securing the airway to allow increased visibility and maneuverability for the surgeon during excision of the lingual thyroid. The type of endotracheal tube used (cuffed or uncuffed) and size depend on the patient's age. However, to prevent subglottic stenosis, there needs to be an air leak at 20cm H2O. Securing the airway in an adult involves high-flow nasal prong oxygenation, fiberoptic nasoendoscopic application of topical anaesthesia with light sedation and subsequent nasotracheal intubation whilst awake.

#### Conclusion

Ectopic thyroid is a rare entity more common in females. Symptomatic patients require careful workup including diagnostic biochemistry and imaging to confirm normal thyroid producing tissue. Initial management needs to include anaesthetic considerations and securing an imminent airway obstruction. Treatment

modality of choice is dependent on patient factors, institution factors and surgeon factors.

Word count: 986 words

### References

- 1. Kumar, S.S., D.M.S. Kumar, and R. Thirunavukuarasu, *Lingual thyroid-conservative management or surgery? A case report*. The Indian journal of surgery, 2013. **75** (Suppl 1): p. 118-119.
- 2. Kamat, M.R., et al., Lingual thyroid: a review of 12 cases. Br J Surg, 1979. 66 (8): p. 537-9.
- 3. Alharbi, A., Lingual Thyroid: A Systematic Review of Hormonal Suppression Treatment. Journal of Otolaryngology-ENT Research, 2015.
- 4. Ueda, D., Y. Yoto, and T. Sato, *Ultrasonic assessment of the lingual thyroid gland in children*. Pediatric Radiology, 1998.**28** (2): p. 126-128.
- 5. Ehsan, S.R., Brandon, D., Halkar, R., Radioiodine treatment of a symptomatic lingual thyroid. The Journal of Nuclear Medicine, 2010.51: p. 1060.
- 6. Pellini, R., et al., *Ectopic lingual goiter treated by transoral robotic surgery*. Acta otorhinolaryngologica Italica: organo ufficiale della Societa italiana di otorinolaringologia e chirurgia cervico-facciale, 2013. **33** (5): p. 343-346.
- 7. Dziegielewski, P., et al., Lingual thyroid in adults: Management algorithm based on swallowing outcomes. Journal of otolaryngology head & neck surgery = Le Journal d'oto-rhino-laryngologie et de chirurgie cervico-faciale, 2011. 40: p. 19-26.
- 8. Ersoy Callioglu, E., et al., *Lingual Thyroid Excision with Transoral Robotic Surgery*. Case Reports in Otolaryngology, 2015.**2015**: p. 548582.
- 9. Prisman, E., A. Patsias, and E.M. Genden, *Transoral robotic excision of ectopic lingual thyroid: Case series and literature review.* Head & Neck, 2015. **37** (8): p. E88-E91.
- 10. Lauretano, A.M., et al., Anatomic Location of the Tongue Base Neurovascular Bundle. The Laryngo-scope, 1997. 107 (8): p. 1057-1059.

# Figure legends

- Figure 1. Nasoendoscopy of lingual thyroid: Round tongue base mass obstructing the laryngeal inlet.
- Figure 2. CT neck soft tissues with IV contrast: a) Axial view of rim-calcified mass at midline of tongue base measuring 30x32mm in anteroposterior diameter; b) Coronal view of rim-calcified mass at tongue base measuring 40mm in length.
- Figure 3 a) Macroscopic lingual thyroid specimen; b) The surgical margin is inked blue. Cut surface shows a multi-loculated calcified cyst 30x25x40mm.
- Figure 4. Microscopic histopathology of lingual thyroid specimen: a) Low magnification demonstrates oral type squamous mucosa (Red arrow), with subepithelial stroma, minor salivary glands (Black arrow), normal thyroid follicles (Yellow arrow) and a dominant thyroid nodule (Blue arrow); b) High magnification demonstrates thyroid tissue with features of nodule hyperplasia with a dominant (adenomatoid) partly calcified nodule within the subepithelial stroma and skeletal muscle.







