BMH Med. J. 2021;8(1):41-44. Case Report

Successful Medical Management for Bleeding Lingual Thyroid

K N Vijayan, Rakesh Chandru K, Akash S, Mohamad Safan A

Department of General Surgery, Kerala Institute of Medical Sciences, Thiruvananthapuram, Kerala, India

Address for Correspondence: Dr. Rakesh Chandru K, 36 Puliyadi Street, Vadasery, Nagercoil, Tamilnadu 629001, India. E- mail: rakeshchandru.k@gmail.com

Abstract:

Lingual thyroid is the most common ectopic location of the thyroid gland due to aberrant embryogenesis during thyroid migration. Most of the patients with lingual thyroid are asymptomatic and remain unnoticed. Few patients present with complications like dyspnoea, dysphagia, bleeding and rarely malignancy. We report a case of ectopic lingual thyroid presenting with bleeding and anemia which was successfully treated by medical management.

Keywords: Lingual thyroid, Ectopic thyroid, TSH suppression, Thyroxine

Introduction

Lingual thyroid was first reported by Hickman in 1869 in a newborn infant who presented with large mass at the base of the tongue causing suffocation. Lingual thyroid is a relatively rare disease with an overall prevalence of 1 in 100,000. It is more common in females than in males. Lingual thyroid is associated with an absence of normal thyroid gland in 70% of cases.[1] The majority of the cases are diagnosed clinically when there is an enlargement of lingual thyroid due to increased physiological demand, particularly in puberty, pregnancy, and menopause. Few cases present with obstructive symptoms like dysphagia, dyspnoea, and bleeding. The objective of this case report is to emphasize the role of TSH suppressive therapy in lingual thyroid presenting with bleeding.

Case Report

A middle aged male presented with complaints of sudden onset of bleeding from the throat, throat discomfort and difficulty in swallowing. He was very pale and had features of hypothyroidism. On examination, there was blood stained posterior pharyngeal wall with no obvious swelling. There was no cervical thyroid swelling or nodes in the neck. Video laryngoscopy revealed a midline mass in the posterior third of the tongue. (**Figure 1A**) Computerized tomography (CT) showed a well defined hypodense lesion of size 2.8*2.1*2.7 cm at the base of tongue suspicious of lingual thyroid (**Figure 1B,C**). A radionucleotide scan

was performed using 99m technetium pertechnetate which confirmed the lingual thyroid gland and hypoplastic thyroid tissue in the neck (**Figure 1D**). His hemoglobin was 5.8 gm/dl and the thyroid function test showed low free T4 of 0.2 ng/dl (reference value 0.93–1.7 ng/dl), low free T3 of 0.63 ng/ml (reference value 0.8–2 ng/ml) and TSH more than 100. He was diagnosed as a case of lingual thyroid with severe hypothyroidism, bleeding, and anemia. Packed cell transfusion has been given for the correction of anemia. He was initially started on a suppressive dose of levothyroxine 125 mcg and later increased to 150 mcg. There was a significant improvement in symptoms and a rise in hemoglobulin level to 11.4 mg/dl over one month. On video laryngoscopy examination, there was shrinkage of lingual thyroid and the thyroid functions were also normalized with TSH 3.92 μ IU/ml. Therefore a surgical excision of the mass was not required and the patient was kept under regular follow up.

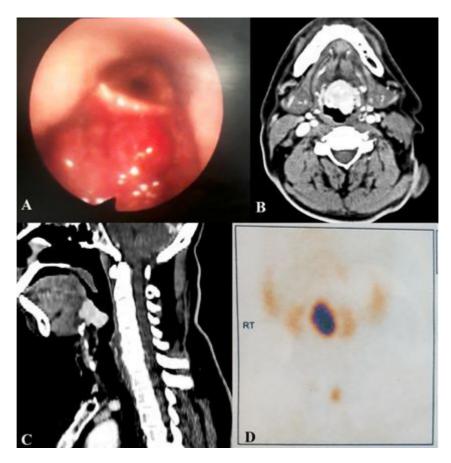


Figure 1: (See text for details).

Discussion

Ectopic thyroid tissue represents the thyroid tissue being in locations other than the normal anterior neck region. This occurs due to aberrant embryogenesis in the migration of the thyroid gland from the floor of primitive foregut to its final pretracheal position.[1,2] Ectopic glands can be found in the base of the tongue, sublingual, submandibular, prelaryngeal, thyroglossal, laryngotracheal, laterally in the neck, and in remote locations like axilla, esophagus, mediastinum, heart, aorta, pancreas, gall bladder, genitourinary and in the adrenal gland. Lingual thyroid is the most frequent ectopic location, accounting for about 90% of reported cases.[1,2]

Most patients are asymptomatic but in few patients, it can enlarge in size and cause severe obstructive symptoms like dyspnoea, dysphagia, dysphonia and rarely hemorrhage.[3] Lifethreatening massive hemorrhage can occur because of the prominent large blood vessels on the surface of lingual thyroid.[4,5] The pathological conditions that might affect normal

thyroid glands like adenoma, hyperplasia, and carcinoma can also occur in lingual thyroid tissue. But the risk of malignancy in lingual thyroid is extremely rare, accounting for less than 1% and usually results in papillary carcinoma.[1]

The majority of the patients, around 70% are hypothyroid which might be due to a small ectopic gland that fails to produce sufficient thyroxine and absent normal thyroid gland in the neck.[1[Hence a complete head and neck examination is essential. It is mandatory to perform an ultrasonogram of the neck to check the presence of the normal thyroid gland. When the thyroid gland is not detected in the neck, thyroid scintigraphy with 99m technetium helps confirm the diagnosis. The lingual thyroid typically shows increased radio nucleotide activity at the tongue base with no activity in the neck in most of the cases. A biopsy is not recommended because of the risk of bleeding and infection.[2]

The treatment options for lingual thyroid include non-surgical approaches like thyroxine suppression therapy or radioiodine ablation and surgery in few circumstances.[6] Medical treatment with a suppressive dose of thyroxine should be the initial treatment modality. It reduces hypertrophy, hyperplasia and the vascularity of the lingual thyroid tissue resulting in shrinkage of the mass. Surgery is indicated in very few circumstances like dysphagia preventing oral intake, severe respiratory compromise, repeated bleeding despite medical therapy or in suspected malignancy.[7] Radioactive iodine ablation therapy can be an alternative to surgery for unfit patients. The limitation of I¹³¹ therapy is that higher doses of radioiodine may be required especially in euthyroid or hypothyroid patients.[7]

There are only a few cases in the literature about lingual thyroid presenting with massive bleeding.[3-5] Our patient was successfully managed with levothyroxine suppressive therapy despite severe bleeding and anemia. There was a significant reduction in the size of the lingual thyroid with no further episodes of bleeding. Similar to our case, there were anecdotal reports where lingual thyroid was successfully managed with thyroxine suppression therapy. [7-10] So we emphasize the initial medical management with levothyroxine for all patients with lingual thyroid and surgery should be reserved only for refractory cases with obstructive symptoms or in suspected malignancy.

Conclusion

Asymptomatic lingual thyroid does not warrant any form of treatment unless the patient is hypothyroid who requires thyroxine supplementation. For patients who present with obstructive symptoms, thyroxine suppression therapy should be the first-line treatment. Surgery is indicated only in very few cases who were refractory to medical therapy or in case of suspected malignancy.

References

- 1. Ibrahim NA, Fadeyibi IO. Ectopic thyroid. Etiology, pathology and management. Hormones (Athens). 2011; 10(4):261–9.
- 2. Noussios G, Anagnostis P, Goulis DG, et al. Ectopic thyroid tissue: anatomical, clinical, and surgical implications of a rare entity. Eur J Endocrinol. 2011; 165(3):375–82.
- 3. Koc G, Taskaldiran I, Aslan Felek S, et al. Ectopic lingual thyroid presenting with massive hemetemesis. Acta Endocrinol (Buchar). 2019; 15(2):244-246.
- 4. Chiu TT, Su CY, Hwang CF, et al. Massive bleeding from an ectopic lingual thyroid

follicular adenoma during pregnancy. Am J Otolaryngol. 2002; 23:185-188.

- 5. Baek MK, Kim EY, Lee SG, et al. A Rare Case of Bleeding Ectopic Lingual Thyroid Presenting as Hematemesis. Yonsei Med J. 2015; 56(4):1163-4.
- 6. Magalhães C, Azevedo P, Nakamura R, Duarte D. Lingual thyroid: can we 'wait and see'? BMJ Case Rep. 2015; p ii: bcr2015210455.
- 7. Kumar V, Nagandhar Y, Prakash B, et al. Lingual thyroid gland: clinical evaluation and management. Indian J Pediatr. 2004; 71:e62–e64.
- 8. Kumar SS, Kumar DM, Thirunavukuarasu R. Lingual thyroid-conservative management or surgery? A case report. Indian J Surg. 2013; 75 (Suppl): 118-9.
- 9. Sigua-Rodriguez, E. A., Rangel Goulart, D., Asprino, L., de Moraes Manzano, A. C. Conservative management for lingual thyroid ectopic. Case reports in otolaryngology. 2015; 265207.
- 10. Dutta D, Kumar M, Thukral A, et al. Medical management of thyroid ectopia:report of three cases. J Clin Res Pediatr Endocrinol. 2013; 5(3):212-5.