Sublingual Thyroid: A Case Report With Literature Review
S Salain, G Rodrigues, S Kumar

Citation

Abstract
Ectopic thyroid tissue can occur at various sites in the human body. Though it does not cause symptoms usually, it can do so in a few patients. Also, this may be the only thyroid tissue present. We report a case of sublingual thyroid with a review of literature.

CASE REPORT
A 22-year-old male presented with complaint of a swelling in the right upper neck of four years duration. It was progressively increasing in size. There was no history of pain, sudden increase or decrease in size, breathing difficulty or change in voice. There was no other positive contributory history.

On examination, there was a single, non-tender, cystic swelling situated in the right submandibular region extending on to the midline of the neck, measuring 5x2 cms, free from skin, moving up and becoming more prominent on deglutition. There was doubtful movement on tongue protrusion. There was no associated cervical lymphadenopathy. Other systems revealed no abnormality. Hence, a clinical diagnosis of thyroglossal cyst and ectopic thyroid was made.

The patient was subjected to fine needle aspiration cytology (FNAC) which revealed benign thyroid cells. A thyroid profile, hemogram and an x-ray of the neck done were normal. A thyroid scan was done which revealed this to be the only thyroid tissue present and an absence of normal thyroid gland in its normal position. We empirically started him on Eltroxin tablets (0.2 microgram once daily), to shrink the size of the gland as the patient was concerned with the size of the neck mass. In a month’s time, the size did reduce and thyroxine was decreased to 0.1 mg and finally stopped. On regular follow-up the ectopic gland has drastically reduced in size and the patient is comfortable.

DISCUSSION
The thyroid gland is the first of the body's endocrine glands to develop, on approximately the 24th day of gestation. The gland originates as a proliferation of endodermal epithelial cells on the median surface of the developing pharyngeal floor. The site of this initial development lies between 2 key structures, the tuberculum impar and the copula, and is known as the foramen cecum. The thyroid initially develops caudal to the tuberculum impar, which is also known as the median tongue bud. This embryonic swelling arises from the first pharyngeal arch and lies in the midline on the floor of the developing pharynx, eventually helping form the tongue as the 2 lateral lingual swellings overgrow it.

An aberrant or ectopic thyroid gland may occur anywhere along the path of initial descent of the thyroid, although it is most common at the base of the tongue, just posterior to the foramen cecum. In this location, an aberrant or ectopic thyroid gland is known as a lingual thyroid and represents a failure of the thyroid to descend. This failure to descend contrasts with the incomplete descent of the thyroid, in which case the resulting final resting point of the gland may be high in the neck or just below the hyoid bone.

Accessory thyroid tissue can also occur, arising from remnants of the thyroglossal duct. While the accessory thyroid tissue may be functional, it is generally insufficient for normal function if the main thyroid gland is entirely removed. This accessory tissue may appear anywhere along the path of the thyroglossal duct tract.

Ectopic thyroid tissue can arise from either the median anlage or lateral anlage (far less common). The tissue may or may not be functioning. Prevalence is higher in females than in males, with a female-to-male ratio of 7:1. The ectopic tissue is typically found incidentally, though these masses can be symptomatic and can cause dysphagia, dysphonia,
stridor, dyspnea, hemorrhage, or hoarseness. An association has been noted between lingual thyroid tissue and cretinism.

The lingual thyroid is four times more common in females than in males. It presents as an asymptomatic nodular mass of the posterior lingual midline, usually less than a centimeter in size but sometimes reaching more than 4 cm. Larger lesions can interfere with swallowing and breathing, but most patients are unaware of the mass at the time of diagnosis, which is usually in the teenage or young adult years. Up to 70% of patients with lingual thyroid have hypothyroidism and 10% suffer from cretinism.

The lingual thyroid consists of a nonencapsulated collection of embryonic or mature thyroid follicles which may extend between muscle bundles, raising suspicions of malignant invasion. The follicular cells, however, are normal or atrophic in appearance. All diseases capable of affecting the normal thyroid gland can, of course, affect the glandular tissue entrapped in the tongue. Thyroid adenoma, goiter, hyperplasia, inflammation, and carcinoma occur in lingual thyroids and must, therefore, be evaluated in the same fashion as would any biopsied thyroid gland. Parathyroid tissue may be seen but has not been neoplastic in reported cases.

CT scans often reveal a homogenous hyperdense mass within the lingual musculature. Management of the mass is surgical; however, prior to the decision to operate, thyroid scanning has to be performed to assess if the mass is functioning and to determine if this tissue is the only viable thyroid material.

Either surgical excision or radioiodine therapy is an effective treatment for lingual thyroid, but no treatment should be attempted until an $^{131}$ iodine radioisotope scan has determined that there is adequate thyroid tissue in the neck. In those patients lacking thyroid tissue in the neck, the lingual thyroid can be excised and autotransplanted to the muscles of the neck. Most cases require no treatment and biopsy should be considered with caution because of the potential for hemorrhage, infection or release of large amounts of hormone into the vascular system (thyroid storm).

Occasional patients with parathyroid tissue associated with their lingual thyroid have developed tetany after their inadvertent removal.

Rare examples of thyroid carcinoma arising in the mass have been reported, almost always in males, but an enlarged lingual thyroid is more likely to reflect a normal compensatory response to thyroid hypofunction. Endocrine evaluation for hypothyroidism should, therefore, be done in such cases. In this light, it is important to know that three of every four patients with infantile hypothyroidism have ectopic thyroid tissue.

**CORRESPONDENCE TO**
Dr. Gabriel Rodrigues, MS, DNB. Associate Professor of Surgery 157, KMC Quarters Madhav Nagar Manipal – 576 104. Karnataka, India. Tel: 00919448501301 Fax: 00918202570061 Email: rodricksgaby@yahoo.co.in

**References**
Author Information

Sandhya Salain, MBBS, MS
Department Of General Surgery, Kasturba Medical College

Gabriel Rodrigues, MS, DNB
Department Of General Surgery, Kasturba Medical College

Sampath Kumar, MS
Department Of General Surgery, Kasturba Medical College