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Case Report

Lingual Thyroid: Case report and brief review of the literature ☆

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ABSTRACT

Lingual thyroid is by far the most common presentation of ectopic thyroid. Though mostly asymptomatic it is associated with congenital hypothyroidism and importantly, absence of orthotopic thyroid making it the only functional thyroid tissue a patient has in many cases. It appears indistinguishable to orthotopic thyroid tissue on imaging, with avid homogeneous enhancement on contrast computed tomography. Here we report clinical presentation and imaging findings of lingual thyroid in a 38-year-old man.

Background

Lingual thyroid is usually described as ectopic thyroid tissue in the midline of the base of the tongue between the circumvallate papillae and the epiglottis [1,2]. Its prevalence is approximately 1 in 100,000 which increases to 1 in 4000 in patients with thyroid disease [3]. Most ectopic thyroids are asymptomatic but dysphagia, dysphonia, cough, snoring, sensation of foreign body, sleep apnea, bleeding and dyspnea have all been reported [4,5]. Ectopic thyroid glands frequently present as congenital hypothyroidism with hypoplasia of the thyroid gland accompanying the ectopia in the majority of cases. This accounts for 40%-45% of permanent congenital hypothyroidism in iodine-sufficient countries [6]. Hypothyroidism has been found in 61.9% of ectopic thyroid patients and ranges from 14.5% to 70% in lingual thyroid patients [7–10]. A more serious concern is thyroid carcinoma which

Abbreviation: CT, computed tomography; MRI, magnetic resonance imaging; US, ultrasound; FNAC, fine needle aspiration cytology; TIA, transient ischemic attack; TSH, thyroid stimulating hormone; Tc-99m, Technetium 99m; MR, magnetic resonance; SPECT, single-photon emission computed tomography.

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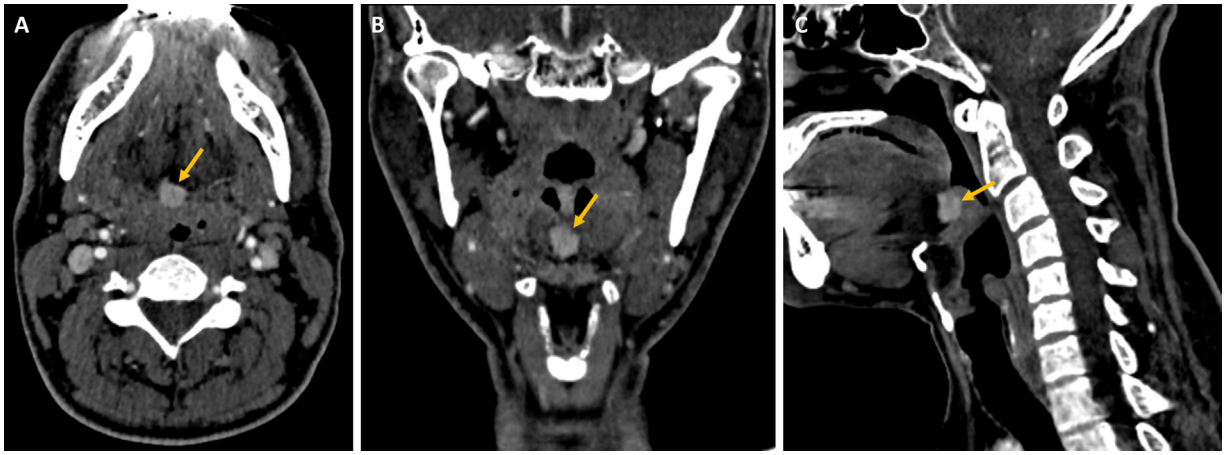

Fig. 1 – Axial (A), coronal (B) and sagittal (C) images from extracranial CT angiography, shows a incidentally detected 1.0 cm x 1.0 cm x 1.2 cm homogeneously enhancing mass at the base of the tongue (arrow). No thyroid tissue was visible on either side at the level of the thyroid cartilage (shown in Fig. 2). Findings were compatible with ectopic lingual thyroid.

is a rare complication of lingual thyroids with only 51 cases reported between 1910 and 2016; papillary carcinoma being the most common [11].

Treatment depends on the symptoms, size, or complications such as hemorrhage [12]. Thyroxine is used to suppress and reduce the growth of the tissue in cases with mild symptoms as well as treat concurrent hypothyroidism [12]. If suppression is not effective for relief surgical resection or iodine-131 ablation could be considered [13]. In asymptomatic and euthyroid patients continued follow-up is suggested to monitor for any developing complications [5]. Scintigraphy is vital at determining not only the location of the ectopic thyroid tissue but any other functional thyroid tissue. This is an important consideration for surgery as 70% of patient with lingual thyroids have an absence of orthotopic thyroid tissue, thyroid in the normal position in the body [14]. Computer tomography (CT), ultrasound (US), and magnetic resonance imaging (MRI) all have utility in determining the location and extension of ectopic tissue. While, a fine needle aspiration cytology plays a role in confirming the diagnosis and differentiating between benign and malignant tissue [5].

Here, we present a case of a 38-year-old man with a diagnosis of lingual thyroid.

Case presentation

A 38-year-old male with a past medical history of congenital hypothyroidism (on levothyroxine) presented to our hospital for blurred vison direct from an outpatient office. He was evaluated by neurology for transient ischemic attack episode.

CT angiograms of intracranial and extracranial structures with and without contrast were ordered for further workup. The findings on the intracranial CT angiogram were unremarkable. The extracranial CT did not reveal any significant atherosclerotic disease, stenosis or occlusion of the large vessels. However, incidentally, a 1.0 cm x 1.0 cm x 1.2 cm homogeneously enhancing mass at the base of the tongue was noted, on arterial phase of the examination (Fig. 1). No thyroid tissue was visible on either side at the level of the thyroid cartilage (Fig. 2). Findings were compatible with ectopic lingual thyroid.

Serum thyroid stimulating hormone (TSH) level on admission was 0.312 mIU/L (Normal range: 0.350- 5.000 mIU/L), with normal free T4 level of 1.11 ng/dL (Normal range: 0.70-1.25 ng/dL). Due to the lower TSH levels his levothyroxine was decreased from 300 μg daily to 250 μg.

He was discharge and outpatient follow-up scheduled with otolaryngology and endocrinology.

Discussion

The thyroid gland tissue is derived from endodermal cells from the foregut which normally start migrating from the foramen cecum, at the posterior of the tongue, by day 24 of development, forming the thyroglossal duct as it travels caudally. It descended anteriorly to the developing hyoid bone, then travels inferiorly and posteriorly around the hyoid bone. It continues to its orthotopic position anterior to the thyrohyoid membrane and thyroid cartilage where it stops migrating at day 45-50 of development [15,16]. While ectopic thyroids can be found all along the path of descent at the hyoid bone, midline infrahyoid neck and more rarely the lateral neck, the lingual location represents 90% of the cases [16,17]. Rarer cases have shown intratracheal, intrathoracic and intracardial thyroid [5].

Most ectopic thyroid tissue is asymptomatic with its discovery on imaging is frequently incidental [18]. In fact in a study looking at 200 autopsies found 10% of individuals had lingual thyroid tissue ranging from microscopic to 1 cm indicating how common the condition could be [19]. Intuitively, ectopic thyroid gland tissue will be indistinguishable to orthotopic thyroid gland tissue on imaging, though differently shaped [16].

Scintigraphy may not have the resolution of CT or MRI, however it allows for the identification of ectopic thyroid foci

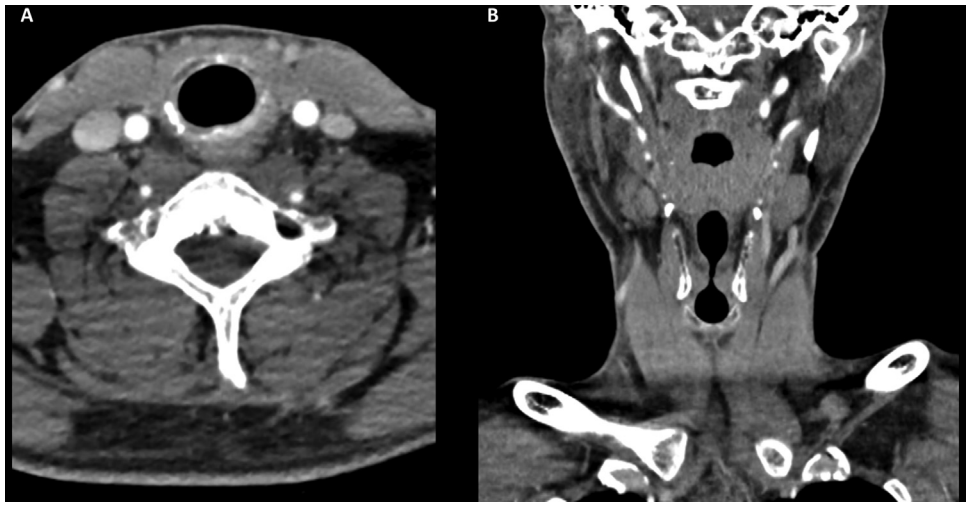

Fig. 2 – Axial (A), and coronal (B) images from extracranial CT angiography, shows no evidence normal thyroid gland in its normal position.

undetected by other studies making it the most important diagnostic imaging [5]. Technetium 99m (Tc-99m), iodine-131 and iodine-123 can all be used in scintigraphy to image thyroid tissue, though with different benefits [20]. Tc-99m produces better images than iodine-131 and delivers less radiation exposure, however, it accumulates in the background of ectopic thyroid tissue making identifying small masses difficult [21]. There are also false-positives reported due to physiologic uptake in regions such as the nasopharynx and salivary glands or pathologic uptake such as from inflammation, benign tumors or malignant tumors [22]. The false-positive rate was reportedly decreased with the addition of single-photon emission computed tomography imaging in patients with metastatic thyroid cancer and additional scintigraphy-negative lesions were reported [23]. With the lower radiation exposure Tc-99m and iodine-123 are better for use in children; however Tc-99m is less expensive and more readily available [20].

The primary benefit of US has been reported as determining the precise location and anatomy of ectopic tissue which is useful in guiding fine needle aspiration for confirmation of diagnosis [5,24]. It is useful in assessing for ectopic thyroid in cases of congenital hypothyroidism when orthoptic thyroid is absent [25]. On US normal thyroid tissue will appear homogenous with medium to high echogenicity greater than adjacent strap muscles [26,27]. This imaging can be used to differentiate between ectopic thyroid tissue and thyroglossal duct cysts. With thyroglossal duct cysts usually appearing as either well-circumscribed hypoechoic or anechoic cysts or a as pseudosolid cyst, in or deep to the thyrohyoid muscle [16,25]. The combination of US with scintigraphy is important as in cases of hypoplasia the thyroid tissue can appear normal on US but on scintigraphy the uptake with be decreased [25]. Conversely scintigraphy will fail to reveal thyroid tissue in patients with iodine-transport defects, maternal thyrotropin receptor-blocking antibodies, TSH receptor defects but a normal thyroid will be seen on US [28]. Furthermore, US assessment can be limited due to poor spatial and contrast resolution [24]. In considering improved assessment, colored doppler was found to have a higher sensitivity than gray scale US for detecting ectopic thyroid, with increased color flow signal seen in the ectopic areas that decreased with thyroid hormone replacement [29].

Similarly to US, CT scan and MRI are useful in determining the location, particularly when distant from the normal path of descension of the thyroid, and extension of ectopic tissue, which is helpful in pre-surgical planning. These imaging modalities are also beneficial in assessment when US is not able to identify orthotopic thyroid or when the radioisotope uptake in ectopic thyroid tissue is masked by uptake in the normal thyroid [20]. Ectopic thyroid tissue on CT appears indistinguishable from orthotopic thyroid tissue except for the positioning and bilobed shape [16]. The ectopic thyroid tissue is homogeneous, well-circumscribed and avidly enhancing on contrast CT, due to vascularity [30]. On CT without contrast the tissue had mildly increased attenuation compared to adjacent muscle due to iodine content, at approximately 70 HU ± 10 [30,31]. A downside of CT scanning is that the use of iodinated contrast means iodine-based scintigraphy cannot be performed within 6 weeks due to contrast uptake in the thyroid [18]. Ectopic thyroid tissue, on T1-weighted MR images appears as a rounded mass iso- to mildly hyperintense relative to muscle. This tissue appears mildly hyperintense and variably enhanced with gadolinium contrast on T2-Weighted MR images [31].

Other diagnoses to consider include a lingual thyroglossal duct cyst which, in contrast to a lingual thyroid, would have a well-circumscribed thin wall with possible rim enhancement on CT scan. The MRI would show low signal intensity on T1 weighted-sequences and high signal intensity on T2 weighted-sequences [16]. It should be noted that ectopic tissue is found in <5% of thyroglossal duct cyst walls [5]. If the thyroglossal duct cyst were to appear nodular malignancy should be assessed [16]. Other cysts on the differential include: branchial cleft cyst, which would normally be found

between the sternocleidomastoid muscle and submandibular gland, dermoid cysts, which have a high fat content in contrast to thyroglossal duct cysts, and epidermoid cyst, which have diffusion restriction [16]. A lingual abscess would reveal a central hypoattenuation with thick enhancing rim on CT along with clinical symptoms [16]. Lymphatic malformations would have hypoattenuating multilocular masses on CT and possibly present with swelling [16]. In contrast to lingual thyroid, squamous cell carcinomas of the tongue would be seen with heterogenic, only moderately enhancing tumors with poorly-defined borders on contrast CT. The T1-signal intensity will depend on the fat content of the tumor with higher fat leading to lower intensity [32].

The patient presented with the typical lingual thyroid incidentally found on imaging with congenital hypothyroidism, though less typical, the patient was male as the incidence lingual thyroid favors women in a ratio 4:1 [33]. Biopsy or scintigraphy could be used to confirm lingual thyroid but with the classic imaging findings of a homogenously enhancing mass in the most common location for ectopic thyroid lingual thyroid is the most likely diagnosis. Further support for this diagnosis is the lack of an orthotopic thyroid gland on imaging which would be expected in alternative diagnoses and, again, hypothyroidism which is common in patients with a lingual thyroid. The most common differential diagnosis, thyroglossal duct cyst, would have a thin wall and rim enhancement not seen in this case. A malignant process would more likely show heterogenicity of enhancement and less well-defined borders. While scintigraphy would be more specific for a lingual thyroid diagnosis, incidental finding on CT is likely more common and so this case's findings have utility in community practice. The lack of a lingual thyroid seen on laryngoscopy further shows imaging's place in the diagnosis of this condition. The case demonstrates the importance of recognizing ectopic thyroid on imaging since for most patients with lingual thyroid it represents the only thyroid tissue and if removed without consideration could lead to the loss of all functional thyroid tissue.

Patient consent

Consent was obtained for the publication of current case. No patient identifiers disclosed.

Written informed consent for publication of this case was obtained from the patient and is available upon request.

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