

A thyroid mass that moves with tongue protrusion: An ectopic thyroid gland

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Abstract

Thyroglossal duct cyst (TDC) is a developmental anomaly that usually appears in early childhood. The common presentation is midline swelling of the neck, which moves with both tongue protrusion and deglutition. Diagnosis is usually clinical and radiological. Fine needle aspiration cytology (FNAC) can be used as a tool for the exclusion of malignancy in adult patients. In some cases thyroid scan is done to rule out the presence or absence of the normal thyroid gland. A complete work-up is mandatory before cyst removal given that it contains only thyroid tissue. We report the case of a 32-year-old woman with only thyroid tissue in thyroglossal duct cyst.

Introduction

Thyroglossal duct cyst (TDC) is a congenital lesion. It is one of the most common causes of anterior neck swelling in children.¹ Its presentation is mostly a midline anterior neck lesion located below the hyoid bone, which is characterised by the painless mass and moves with tongue protrusion and deglutition. On the contrary, the thyroid gland does not move with tongue protrusion. During embryogenesis, the thyroid gland descends from its initial position (the tongue base) to its final pre-tracheal position creating a thyroglossal duct. The duct normally disappears completely before the 10th week of foetal life.² Failure in obliteration of the thyroglossal duct after the descent of the thyroid gland results in TDC.³ Diagnosis is always straightforward from clinical examination. The presence of all functioning thyroid tissue in an aberrant location along with the embryological line of thyroid gland descent is defined as ectopic thyroid. Only 1% to 2% of ectopic thyroid tissue is found in TDC.⁴

Case summary

A 32-year-old Malay woman presented with 5-year history of anterior neck mass. The swelling was increasing in size but there was no history of hyperthyroid or hypothyroid symptoms. She denied any history of dysphagia, change in voice or loss of weight. There was no known medical illness.

Examination of the patient revealed a 2 cm × 3 cm oval-shaped mass at the level of the hyoid bone, which was moving with both deglutition and tongue protrusion. There was no bruit, hyperthyroid and hypothyroid features. No lymph node was palpable and laryngoscopy was normal. This confirmed the clinical impression of a TDC.

Fine needle aspiration cytology (FNAC) of anterior neck swelling was compatible with colloid goitre. Ultrasonography revealed a structure which was hyperechoic than the anterior strap muscle. It measures 1.6 cm (AP) × 2.7 cm (W) × 3.7 cm (CC) (Figure 1). Further scanning on both sides of the neck did not show the presence of normal thyroid tissue. These features suggest TDC with ectopic thyroid tissue. The thyroid function test was normal. Thyroid scan confirmed the presence of thyroid tissue in the TDC as the only functioning thyroid. No uptake was demonstrated at the normal thyroid bed. She was then prescribed a suppression therapy (to halt the size increment) with 200 µg of L-thyroxine and was regularly followed up to monitor the progress and see if there are any changes in the malignancy. However, the swelling had been of the same size despite the hormonal suppression therapy with L-thyroxine. The patient has so far been followed up for 14 years without any suspicious sign.

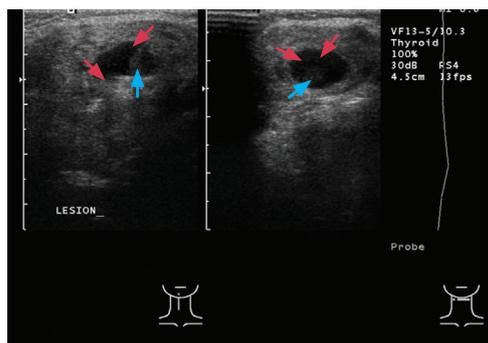


Figure 1. There is an oval structure of homogenous echotexture with medium echogenicity in the midline of the anterior neck. A rounded anechoic lesion (red arrow) with hypoechoic area (blue arrow) is noted. The hypoechoic area may be due to proteinaceous material.

Discussion

TDC is one of the most common causes of anterior midline neck swellings in childhood. It can be found anywhere from the base of the tongue to the suprasternal notch.¹ Although it is uncommon in adults, it may appear at any age.^{5,6} The presentations include a painless anterior neck mass, discharging sinus, abscess formation and on rare occasions compressive symptoms. It is usually complicated by the infection and abscess formation due to communication between the cyst and floor of the mouth, resulting into contamination with oral flora. This complication is commonly seen in adults.⁵

The clinical presentation of TDC is very classical as demonstrated in this case report where the mass is located on the anterior midline of the neck at the level of the hyoid bone, which moves with tongue protrusion and deglutition.⁵ However, lateral neck swelling was reported in a 50-year-old woman with a pre-operative diagnosis of solitary thyroid nodule, which revealed an intrathyroid thyroglossal cyst when a right hemithyroidectomy was performed.⁷ Similarly, a lateral neck mass was reported, which did not move with tongue protrusion and dysphagia with pre-operative diagnosis of thyroid goitre that turned out to be a TDC on operation.⁸ Thus, TDC should be included in the differential diagnosis of a lateral neck mass in an adult patient in addition to branchial cleft cyst, lymphoepithelial cyst, thyroid gland lesions, cystic degeneration of metastatic cancer in a delphian lymph node and lymphadenopathy.⁹

Ectopic thyroid has been described in numerous sites between the base of the tongue and its final pre-tracheal position, as well as in the mediastinum and distant

sub-diaphragmatic areas. The majority of thyroid ectopias are located in the midline along the tract of the thyroglossal duct due to arrest of migration along the line of descent.¹⁰ However, the presence of ectopic thyroid tissues in distant locations could be due to aberrant migration or heterotopic differentiation of uncommitted endodermal cells.¹¹ In most of the cases of ectopic thyroid, orthotopic thyroid gland usually coexists; hence, the patients are euthyroid. This is because the thyroid hormones produced by ectopic thyroid are usually subnormal.¹² Nevertheless, in our case report the patient presents only functional thyroid tissue, yet she remains euthyroid. The other main concern about ectopic thyroid is malignant transformation. Even though it is uncommon, but it has been reported in TDC¹³ as well as in lateral aberrant thyroid tissue, mediastinal and struma ovarii. To date, papillary carcinoma that was reported in ectopic thyroid outnumbered the other type of thyroid carcinomas.¹⁴

In addition to clinical assessment, the diagnosis of TDC and ectopic thyroid needs a critical radiological and histopathological evaluation. Usually, in most cases ultrasonography (US) has been frequently utilised in the diagnosis of TDC.¹⁵ Adequate information about the cyst can be provided by the US alone though scintigraphy is considered valuable in cases of hypothyroidism and where normal thyroid gland is not visualised on US.¹⁶ Other radiological imaging modalities that may help in designating the extension and location of ectopic tissue for pre-surgical evaluation include computed tomography (CT) and magnetic resonance imaging (MRI).^{17,18} Occasionally, in some cases of intrathoracic goitre chest radiography may also be a useful evaluating tool.¹⁴ In selected cases of patients, FNAC is considered for the exclusion of malignancy, especially in adults.⁵ About 5% of thyroid tissues is revealed histologically in TDC evaluation (but with thyroid gland in the normal location).¹⁹ Other findings in FNAC include cholesterol crystals, phagocytes and columnar ciliated epithelium. Moreover, FNAC also provides considerable assistance in confirming the diagnosis of ectopic thyroid in TDC.

In this case, scintigraphy using Tc-99m, I-131 or I-123 still remains the most important diagnostic tool in detecting ectopic thyroid tissue and showing the absence or presence of thyroid in its normal location. Thyroid scan is very sensitive and specific in differentiating an ectopic thyroid from other causes of midline neck masses^{17,18} and thus very useful

in detecting additional sites of thyroid tissue. As a result of normal or abnormal iodine uptake in the head and neck, the possibility of false positive diagnostic iodine scans must be taken into consideration. Pathological causes of increased uptake of Iodine may include sinusitis, dacryocystitis, prosthetic eye, dental disease and meningiomas; while uptake due to physiological causes include nasal mucosa, salivary glands, intestine, liver and urinary bladder.²⁰ Thyroid scan indications include cases such as lingual TDC where the gland cannot be located radiologically in its normal anatomic location. This observation was reported by Radowski et al.²¹ and Batsakis.²² They stated that since the gland is the leading element in the descent of the tract, a TDC implies a thyroid gland in a more distal location. This is contrary to our case report in which the ectopic thyroid tissue is within the TDC located just below the hyoid bone. In further evaluation with thyroid scan, it was revealed that only the thyroid tissue was present within the TDC. Other indications of thyroid scan

include patients with elevated level of TSH, abnormal thyroid function tests or symptoms of hypothyroidism.¹⁶

Sistrunk's operation,²³ which was described in 1920 and modified in 1928, is the best surgical option for TDC. However, an appropriate follow-up with the avoidance of surgery is the only choice for TDC with the functioning thyroid tissue. Our current patient has been followed up regularly. She has been on suppressive thyroxine therapy, 6-monthly thyroid function tests and the annual thyroid scan.

Conclusion

There should be a high index of suspicion of ectopic thyroid in every TDC. If US cannot detect any normal orthotopic thyroid tissue in the neck or is inconclusive than thyroid scan is essential for the investigation. FNAC may help to confirm ectopic thyroid but more importantly in excluding malignancy.

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