

Dual Ectopic Thyroid: A Case Report in a 12 years old girl

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ABSTRACT

Ectopic thyroid gland is uncommon in our routine clinical practice and among them dual ectopic thyroid appears to be rarer. We report an unusual case of two different locations of ectopic thyroid tissues in the lingual and left submandibular regions of a 12 years old girl who presented with painless, nodular left upper neck swelling. Provisionally, the patient was diagnosed by high resolution ultrasound (HRUS) of neck and confirmed by radionuclide thyroid scan. Thyroid scintigraphy with ^{99m}Tc -pertechnetate is highly sensitive but minimally invasive procedure to detect and image the ectopic thyroid tissue with fairly good specificity and negligible radiation exposure.

Key words: Dual ectopic thyroid, ^{99m}Tc -pertechnetate, Thyroid scintigraphy

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INTRODUCTION

The first endocrine gland to develop during embryogenesis is the thyroid with the timing around 24th day of gestation (1). Ectopic thyroid tissue is an uncommon congenital anomaly in which the presence of thyroid tissue other than its normal cervical position between the 2nd to 4th tracheal cartilages (2,3). In approximately 7th week of gestation, the thyroid gland reaches its final location in pretracheal region. Rarely the normal descent of thyroid gland may fail resulting in the development of ectopic thyroid gland with lingual/sublingual thyroid as the most common ectopic location (4,5). Normal thyroid gland is absent in 70% cases of ectopic thyroid and more commonly found in women. Hypothyroidism is reported in 33% of these patients. An estimated prevalence of ectopic thyroid tissue is 1 per 100000-300000 population and rising to 1 per 4000-8000 in patient with thyroid disease (6,7).

CASE REPORT

A 12 years old girl presented with a painless, visible and palpable swelling in left upper neck (left submandibular region) which was gradually increasing in size for 1 year.

There was no family history, features of airway obstruction, history of radiation exposure, pain, fever and change in voice. No definite feature of hypothyroidism or hyperthyroidism was noted. Physical examination revealed an oval shaped, non-tender, mobile, firm swelling in the left upper neck measuring about 3.5X2.0 cm that moved with deglutition (Fig.1). Thyroid bed was unremarkable.



Fig. 1(a). Visible, painless swelling in left upper neck predominantly located in left submandibular region (arrow), Fig. 1(b). Visible swelling close to the base of the tongue (arrow).

HRUS of neck showed no thyroid tissue in the thyroid bed. On the other hand, the visible and palpable swelling in the left upper neck corresponded to a solid, predominantly hypoechoic, non-homogenous, soft tissue area located in the left submandibular region measuring about 3.0X1.8cm. Left submandibular gland could be visualized separately with normal ultrasound appearance. Another solid, hypoechoic, soft tissue area was noted at the base of the tongue measuring about 2.1X1.7 cm. The patient was unaware of the swelling at the base of the tongue. HRUS reported as suspected dual ectopic thyroid tissue (Fig. 2).

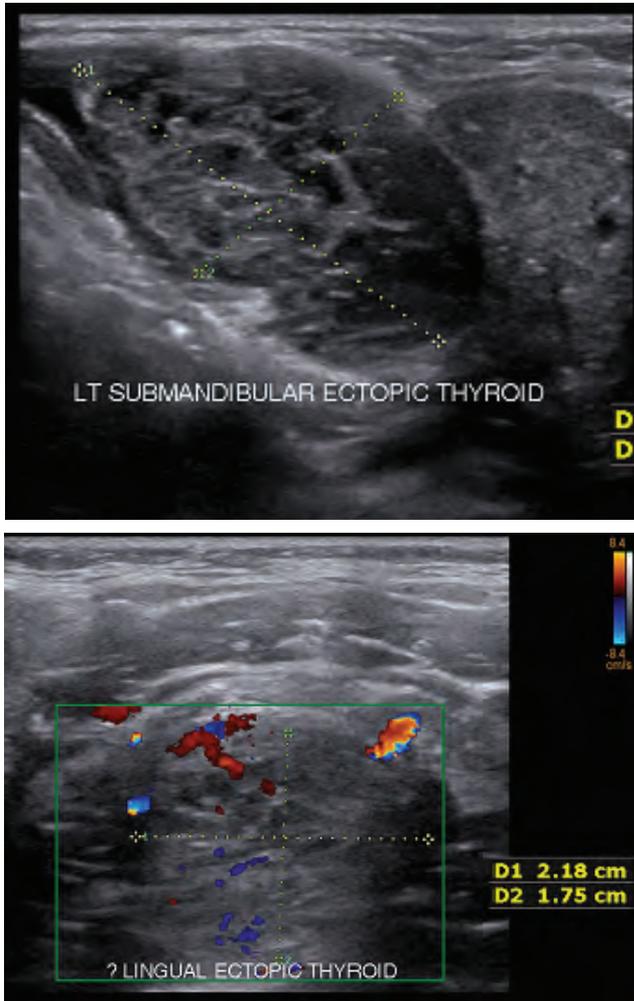


Fig 2(a). Ultrasound image showing thyroid tissue like area in left submandibular region, **Fig 2 (b).** Ultrasound image of lingual region showing thyroid tissue like oval shaped area.

Her thyroid function tests were suggestive of euthyroid state with the value of T3-1.72 ng/ml (normal= 0.9-2.20), T4-86.80 ng/ml (normal = 45-135) and thyroid-stimulating hormone (TSH)-3.96 μ IU/ml (normal = 0.3-5.0). Based on clinical and biochemical examination, a provisional diagnosis of ectopic thyroid was made and the patient was referred for thyroid scan at INMAS, Dhaka. The thyroid ^{99m}Tc -pertechnetate scan demonstrated no radiotracer activity in the thyroid bed but two distinct focal areas of intense radiotracer uptake in the sublingual and submandibular regions (arrows; Fig. 3). A final diagnosis of dual ectopic thyroid was made based on thyroid scan findings. FNAC correlation from the left submandibular swelling further confirmed the diagnosis of ectopic thyroid.

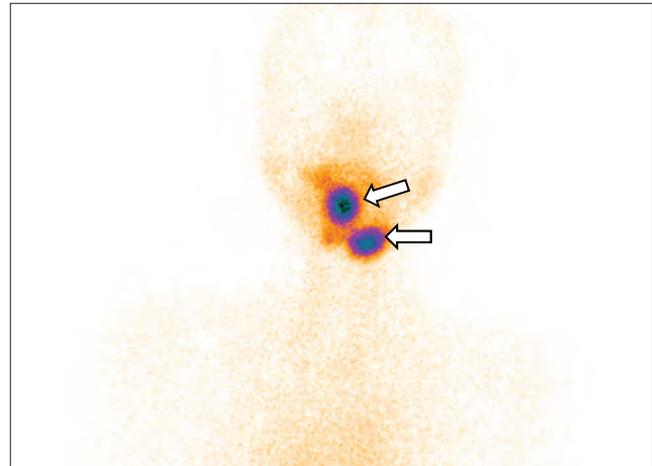


Figure 3: ^{99m}Tc pertechnetate thyroid scan (anterior view) shows two focal areas of abnormal radiotracer uptake in sublingual (arrow) and submandibular (arrow) regions with no radiotracer uptake in the region of the normal thyroid bed

DISCUSSION

The ectopic thyroid tissue is an uncommon clinical phenomenon caused by the failure of descent of thyroid gland to its normal site during early fetal life. Normally thyroid gland is located antero-laterally from the 2nd to 4th tracheal cartilage. An ectopic thyroid gland may occur due to any discrepancy in the descent of the thyroid tissue which is commonly found in midline, but the lateral location is also possible though very rare (3,7). Lingual thyroid is the most common type, about 90% of the reported cases (2). Other ectopic sites include sublingual, prelaryngeal, or mediastinal. Ectopic thyroid tissue may be present in intrathoracic and sub diaphragmatic areas (5). In this reported case of dual ectopic thyroid, sublingual and submandibular locations were spotted. Rare sites of ectopic thyroid tissue include parathyroid, cervical chains, submandibular region, duodenal mesentery, adrenals and carotid bifurcation (8). Usually ectopic thyroid tissue produces inadequate hormone, resulting either subclinical or clinical hypothyroidism causing in increase in TSH secretion from the pituitary gland. This 12-year old girl was in euthyroid status showing no features of hypo or hyperthyroidism. Follicular cell hyperplasia of the ectopic thyroid tissue occurs due to increased TSH level resulting in visible swelling of anterior neck anywhere

along the path of descent (3). During adolescence or pregnancy, most ectopic thyroid tissue is found due to increased physiological demand of thyroid hormones. Reported case matches this observation as she was in her adolescence, aging 12 years. Thus, patients may present without any symptoms or just a swelling somewhere in the neck area with or without altered thyroid function. Occasionally, patients present with palpable swelling or lumpy sensation in the neck. Clinical features in adolescent and young adults are slow heart rate, cold intolerance, constipation, mental fatigue and chronic tiredness. Sleep apnoea and respiratory obstruction in adult patients having lingual thyroid has also been reported (2,3, 7). Ectopic thyroid with malignant transformation is a rare clinical entity with an estimated incidence of 1%. Commonest histopathological subtype is follicular carcinoma. Biopsy should be taken for exact pathological diagnosis (9).

High resolution ultrasound can detect the size and location of ectopic thyroid tissue and can differentiate between solid and cystic masses (10). Radionuclide thyroid scan is the gold standard for the diagnosis of ectopic thyroid tissue because it can detect all ectopic foci of thyroid tissue in the body. Furthermore, thyroid scan can differentiate ectopic thyroid tissue from other causes of anterior neck swelling such as thyroglossal cyst, lipoma, epidermoid cyst, enlarged lymph nodes, vascular malformation and malignancies. Inappropriate surgery may be avoided by detecting ectopic thyroid tissue (5). Fine needle aspiration cytology (FNAC) can detect ectopic thyroid tissue and can differentiate between benign and malignant lesions. Besides these, computed tomography (CT) and magnetic resonance imaging (MRI) can identify the location, size and extension of ectopic thyroid tissue (2).

The management of ectopic thyroid should be according to the size and location of the mass, local symptoms, endocrine function of the gland, patient's age and related complications. Asymptomatic and euthyroid patients do not require any treatment, but they should be followed up. Lifelong thyroid hormone replacement is usually required according to individual thyroid function after proper diagnosis.

Surgery usually is not done, as the ectopic thyroid tissue may be the only functioning thyroid tissue in the body. Surgery is needed in pressure effect, severe respiratory distress and in suspicion of carcinoma in ectopic thyroid tissue (2,3). This reported patient was euthyroid, so no supplementation of thyroid hormones were needed.

CONCLUSION

We reported a rare case of simultaneous presence of two ectopic foci of thyroid tissues. Thorough clinical examinations and multiple imaging modalities were opted to establish the diagnosis of ectopic thyroid tissue, while a ^{99m}Tc thyroid scan remains the most important diagnostic technique.

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