Diagnostic Dilemma due to Atypical Presentation of Sublingual Thyroid Ectopia: A Case Report

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ABSTRACT

Thyroid tissue may be found in an abnormal location along the thyroglossal duct tract or other sites due to abnormal embryologic development and migration. Thyroid ectopia is an uncommon but well-documented clinical entity. In the event of any neck swelling, it should be kept as a differential diagnosis. Thyroid scintigraphy plays a very important role in diagnosing ectopic thyroid, with the complementary primary role of high-resolution neck ultrasonography (HRUS).

Reported case of an 18-year-old male referred to INMAS for evaluation of a slowly enlarging submental swelling with no history of dysphagia or dyspnea was biochemically hypothyroid. Patient had a history of submental swelling that was misdiagnosed as an abscess, creating a diagnostic dilemma, as he was found to have glandular tuberculosis after surgical drainage of the lesion.

Keywords: Thyroid ectopia, thyroid scintigraphy, submental abscess.

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BACKGROUND

Thyroid ectopia accounts for nearly 60% of thyroid dysgenesis. Ectopic thyroid results from aberrant embryogenesis during its passage from the floor of the primitive foregut to the final pre tracheal location (1). Its prevalence is about 1 per 10000–30000, rising to 1 per 4000-8000 with thyroid disease (2). Ectopic thyroid is common in females, especially in Asia. In 70–90% of cases, it is the only thyroid tissue present and can be found anywhere along its pathway of descent, and the most common location is sublingual region (1, 3).

The clinical presentation of thyroid ectopia is variable, might go unrecognized for decades, and can be detected incidentally (1). Ectopic thyroid usually becomes symptomatic during pregnancy and puberty because of

increased demand for thyroid hormone and susceptibility to hypertrophy of thyroid tissue (4).

We report an interesting case of a teenager with a sublingual ectopic thyroid that created diagnostic dilemma due to atypical clinical presentation.

CASE REPORT

An 18-year-old male was referred for the evaluation of a slowly enlarging submental swelling at the Institute of Nuclear Medicine & Allied Sciences (INMAS), Mitford. The swelling has been noticed for about one and a half year, and the patient reported an episode of acute illness one year back for which he was hospitalized. Currently, it was painless with no history of dysphagia or dyspnea. Aside from a history of mild intellectual disability, there was no other significant complaint. On initial inspection, a non-tender, smooth, oval swelling was noticed in the submental region that was not moving with deglutition. Overlying skin appeared normal.

HRUS of neck revealed a well-defined, complex and heterogeneous lesion (measuring about 4.6 X 3.8 cm) having internal cystic areas located in the midline and in close proximity to the base of the tongue and showing peripheral vascularity in Doppler mode. Myelohyoid muscle and overlying skin were separately visualized about 5 mm superficial to the lesion. Thyroid gland was not present in thyroid bed or other possible ectopic sites in the neck. Neck lymph nodes were unremarkable. Laboratory investigations reported unusually high serum TSH level (230 $\mu IU/mL)$ with very low FT3 and FT4 levels.

During his previous hospitalization one year back, HRUS of the swelling was done which revealed a heterogeneous,

hypoechoic lesion with thick, irregular walls and central necrosis, and reported as an abscess. Cytological examination of this swelling also reported abscess and then surgical drainage was done. Finally, the patient was diagnosed as a case of glandular tuberculosis, on the basis of histopathology report which demonstrated presence of palisading epithelioid cells and sheets of foamy histiocytes, enclosing a neutrophillic exudate rich in fibrin with nuclear debris and suspected a tuberculous abscess. Anti-TB regimen was started immediately but he was non-compliant and discontinued medications after seven months.

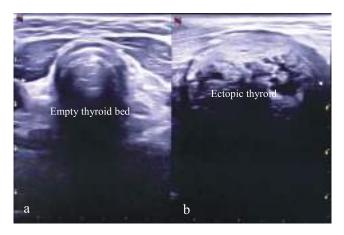


Figure 1: Transverse Gray scale HRUS images of neck a) showing no identifiable thyroid tissue in thyroid bed and b) heterogeneous nodular area corresponding to the palpable mass measuring about 4.6 X 3.8 cm.

The patient's past history of acute illness and imaging reports created a diagnostic dilemma as to whether the swelling was a tubercular abscess or an ectopic thyroid.

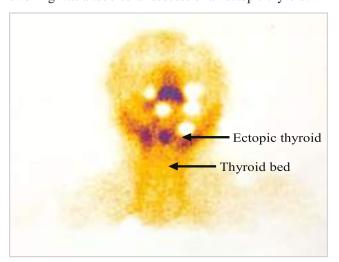


Figure 2: 99mTc scintigraphy image showing radiotracer uptake in sublingual location with no uptake in thyroid bed.

An ectopic thyroid was the leading consideration, as no thyroid tissue was present in the thyroid bed and the serum TSH level was very high. Then thyroid scintigraphy was performed in our institute with ^{99m}Tc pertechnetate that revealed focal tracer uptake in the sublingual region without any uptake in the normal thyroid location. The findings were consistent with the presence of ectopic thyroid tissue.

DISCUSSION

Thyroid tissue present in a location other than normal is known as "ectopic thyroid" or "thyroid ectopia." Thyroid ectopia may occur in any location along the path of the initial descent of the gland (5). When thyroid tissue is absent in its normal location, it is considered true ectopic; when the orthotopic thyroid gland is present in its normal location, it is considered accessory thyroid (6, 7). The absence of thyroid tissue in the thyroid bed makes the diagnosis easy. Although thyroid ectopia is a rare condition, this differential diagnosis should be kept in mind during the evaluation of any neck masses (4). In this reported case, the thyroid bed was not evaluated sonographically in the first scan, which misled the clinical diagnosis.

Multiple diagnostic modalities have been introduced for locating ectopic thyroid, including ultrasound, Computed Tomography (CT), Magnetic Resonance Imaging (MRI), and nuclear scintigraphy (7). Though ultrasound is a non-invasive bedside modality for evaluating a neck mass, ^{99m}Tc pertechnetate scintigraphy remains an important tool for the detection of ectopic thyroid tissues for its ability to scan the entire neck area in a single view. It is also cost effective and highly sensitive causing minimum radiation exposure, therefore considered as the gold standard for confirming ectopic thyroid (8, 9).

Thyroid involvement with tuberculosis is considered extremely rare due to inherent resistive mechanisms of the gland. Previously reported cases show a slight female predominance with the mean age of onset to be around the third to fourth decades of life (10, 11). The infection can be primary or secondary but the clinical presentation usually has no distinct characteristics. The disease course may mimic subacute thyroiditis, thyroid abscess, fever of unknown origin, or even thyroid malignancy (12). Current treatment regimen includes antituberculous drugs, followed by surgical intervention in case of large abscess

(10). Our reported case has several unusual aspect including the age and gender pattern, thyroid dysfunction as well as ectopic location of the gland itself.

CONCLUSION

This case report shows the importance of documentation about the presence and location of thyroid gland in a case of neck swelling and emphasizes once again the pivotal role of thyroid scintigraphy for ectopic thyroid detection. It also reinforces the fact that tuberculosis should always be kept in mind as a differential diagnosis in an endemic country like Bangladesh.

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