Dual Ectopic Thyroid: A Rare Entity, Case Report With Review Of Literature

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Abstract: Ectopic thyroid is a rare entity, multiple ectopia is more rare. Worldwide, only 32 cases have been reported up to date. Here we report another case of this rare entity of dual ectopic thyroid in a young female easily detected on MDCT and nuclear scan with emphasis on complementary role of CT to nuclear scan with relevant review of literature.

Keywords: MDCT- Multidetector CT, Ectopia, Thyroid, ETT-Ectopic thyroid tissue.

INTRODUCTION

Ectopic thyroid refers to the presence of thyroid tissue in locations other than the normal anterior neck region between the second and fourth tracheal cartilages. To date, about 500 cases have been reported in the English literature, mostly occurred in females. Dual ectopia of thyroid is further more rare [1], only 32 cases have been reported till date. Although it is difficult to detect asymptomatic thyroid ectopy, postmortem studies have suggested that 7-10% of adults may harbor asymptomatic thyroid tissue along the path of the thyroglossal duct [2]. Ectopic thyroid tissue coexisting with a eutopic thyroid may be equal to that without a normally located gland [3].

Lingual thyroid is the most common type accounting for 90% of cases, while sublingual types are less frequently encountered. The sublingual types may be suprahyoid, infrathyroid or at the level of the hyoid bone [3]. Existence of ectopic thyroid glands at two different locations is very rare. Two cases of triple ectopia have been reported, one of these by Ibrahim et al, [4] where three separate ectopic thyroid masses were present in the lateral neck region with a co-existing eutopic goiter.

CASE REPORT

A 11 years girl presented in ENT department with midline anterior neck mass in infrathyroid region. Clinical examination revealed a smooth mass moving with deglutition in the neck in midline with normal thyroid gland not being palpable. No mass and lymph node found in rest of neck.

Her Thyroid function test were within normal limits, T3-137ng (Normal 80-230ng/dl), T4-9.2mcg (Normal 7-14mcg/dl), TSH-3.51 (Normal .5-5.0 u IU/ml).

On neck ultrasound examination, there was absence of normal thyroid gland and the palpable midline neck mass was appearing a well defined, echogenic hypervascular solid mass. FNAC of the infrathyroid anterior neck mass revealed sheets of follicular epithelial cells with macrophages in a colloid mixed hemorrhagic background:- features of Colloid goitre with cystic changes.

She was further investigated with non contrast CT scan and nuclear scan to see the nature of the mass and multiplicity, combining the functional and anatomical information both. MDCT revealed, a well defined, rounded, hyperdense mass (110-140HU) at the level of thyroid and cricoid cartilage in midline in infrathyroid location (Figure1a). And another smaller density mass seen in the midline at the base of tongue at the suprahyroid location (figure1b), which was not found earlier. The high CT attenuation of mass was enabling it to be easily detected and sharply marginated from the surrounding tissue even on unenhanced CT. The eutopic thyroid has similar CT attenuation (more that of muscle) due to iodine content and was not found.

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A thyroid scan with Tc 99 revealed nodular uptake in sublingual region. No radiotracer uptake was seen in normal thyroid bed. The faint uptake at lingual region was overlooked and was identified retrogradely after simultaneously analyzing both scans (figure 2a, 2b and figure 3a, 3b).
On the basis of aforementioned finding, Dual ectopic thyroid tissue in the sublingual and infrahyoid region in midline was established with absent eutopic thyroid.

**DISCUSSION**

Dual ectopic thyroid is extremely rare disease, majority occurs in neck along the path of descent. Up to date, only 32 cases have been reported worldwide. 27 cases have been reported in a review [5] published in 2008 and thereafter 4 cases have been found in Jain and Pathak [6] study in 2010 and 1 more case has been reported by Joshi and Lele [7] in the 2013.

Although the cause is not fully known, genetic factors have been associated with thyroid gland morphogenesis and differentiation. So far, no mutation in known genes has been associated with human thyroid ectopy. Different pathological changes that affect normal eutopic thyroid can occur in the ectopic tissue. The majority are asymptomatic; however, symptoms may arise following enlargement of the gland during periods of stress.

Hormone production from ETT is usually insufficient, leading either to a subclinical or clinical hypothyroid state. The incidence of clinical hypothyroidism with ETT varies from 24 to 60%. Usually the patients complain of a palpable mass, growth retardation and lump sensation in the throat. Patients may have dyspnea, stridor or cough due to intratracheal thyroid gland. Adolescents and young adults may present with symptoms of hypothyroidism [2]. Lifelong thyroxine therapy is required according to individual thyroid status after establishing the diagnosis. It helps in achieving the euthyroid status as well as decreasing the size of ectopic thyroid swelling by lowering the elevated TSH level [8]. Surgery is usually not done, since the ectopic thyroid may be the only functioning tissue in the body. It is indicated if the patient has pressure or obstructive symptoms, or there is suspicion of malignancy in ETT. At times ectopic thyroid of these patients are excised when they present with mass in neck region, rendering them frankly hypothyroid as against subclinical hypothyroid state that they were suffering from earlier [9]. Hence, possibility of ectopic thyroid should be kept in mind and evaluated accordingly.

Other locations in the head and neck regions where ectopic thyroid tissue may be found include the trachea, submandibular, lateral cervical regions, axilla, palatine tonsils, carotid bifurcation, iris of the eye and pituitary gland. Radionuclide studies are highly sensitive and specific in demonstrating the functional tissues in patients with ectopic thyroid [10]. Tc-99m pertechnetate scan is a well known method for evaluating the size, distribution and activity of thyroid and distinguishing it from other tissues in the head and neck and superior mediastinum. Single photon emission tomography-computed tomography (SPECT-CT) is an example of fusion imaging where functional information of nuclear medicine and anatomical information of radiology can be evaluated together as done by Joshi and Lele [7].

On CT thyroid tissue has an attenuation coefficient higher than that of muscle, which is thought to be due to high iodine content in thyroid tissue. Due to this ectopic thyroid tissue gives a characteristic appearance on unenhanced CT, appearing as hyperdense nodule, readily visible and sharply demarcated from surrounding tissue as seen in our case. Nodular hyperdense mass in midline along the thyroglossal duct on the unenhanced CT which is moving on deglutination on clinical examination suggest diagnosis of ectopic thyroid. Radionuclide thyroid scan shows tracer uptake in functioning tissue and is highly specific and sensitive non invasive tool for ectopic thyroid. In our case, initially only one (infrahyoid)ectopic thyroid tracer uptake was identified on radionuclide scan, and the other one (sublingual) was overlooked due to less tracer uptake. While on CT scan, both the ectopic thyroid nodule was very well seen.

Joshi and Lele [7] also suggested the combination of Radionuclide scan and SPECT/CT as primary noninvasive tool for diagnosis of ectopic thyroid . Jain and Sujata [6] found the similar characteristic CT appearance of ectopic thyroid in their series.

**CONCLUSION**

Unenhanced CT scan is a non invasive sensitive and specific modality for the detection of ectopic thyroid and should be combined with radionuclide scan. And if feasible radionuclide scan should be combined with SPECT. The incidence of multiple ectopia is likely more as reported earlier, more cases of multiple thyroid ectopia is now being detected due to increased use of these noninvasive investigation.

**REFERENCES**

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