

Simultaneous occurrence of Graves' disease in eutopic and ectopic thyroid tissues: A case report and review of literature

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ABSTRACT

Ectopic thyroid tissue an uncommon condition results from abnormal migration of the primitive thyroid bud. This may be the only functional thyroid. Ectopic thyroid tissue may sometimes coexist with the eutopic thyroid gland. Hyperthyroidism in association with ectopic thyroid tissue is very uncommon. We report a rare case of simultaneous involvement of ectopic and eutopic thyroid tissue in a married women of 35 years who was referred to our department for a technetium 99m thyroid scan. Coexisting ectopic and eutopic thyroid tissue due to identical histology may have similar response to various stimulatory and inhibitory factors like hormones and immunoglobulin's. Iodine-131 is an easy to administer and effective treatment for patients with simultaneous Graves' disease in the ectopic and eutopic thyroid tissues.

Keywords: Ectopic thyroid, eutopic thyroid, Graves' disease, simultaneous

INTRODUCTION

Ectopic thyroid tissue (ETT) is a consequence of abnormal thyroid bud migration. The gland may present anywhere along the line of descent from the foramen caecum in the floor of mouth to its pre-tracheal position and at unusual sites like mediastinum.^[1] The prevalence of ETT is approximately 1/100,000-300,000 persons and is reported to occur in one in 4000-8000 patients with thyroid disease.^[2] ETT is commonly associated with hypothyroidism. Graves' disease involving the ETT is a rare presentation. On Pub Med search we came across 7 such reported cases.^[1-8] Simultaneous occurrence of Graves' Thyrotoxicosis involving ectopic and eutopic thyroid tissue has been reported in 4 such cases.^[1-5] Most of these patients are definitively treated by administration of oral Iodine-131.^[2-4,7] We report a case of Graves' disease simultaneously involving ectopic mediastinal thyroid tissue and the normal eutopic thyroid gland. The toxic thyroid tissue was ablated by an oral single fixed dose of 370 MBq (10 mCi) Iodine-131.

CASE REPORT

A 35-year-old married woman presented to our department of Nuclear Medicine with chief complaints of palpitations, tremors, increased sweating, increased frequency of stools, and weight loss. The patient was non-smoker, had two children a male and a female aged 14 and 12 years respectively. At the time of presentation the patient was non-lactating and not pregnant. In drug history, patient gave history of treatment with anti-thyroid medication (Carbimazole and Propranolol). On her own, the patient discontinued these drugs 4 months before the current symptoms. At presentation, she had a regular pulse of 104/min. Her weight was 47 kg. She had only mild visible proptosis. There was a transverse surgical scar in the neck. Thyroid was not visible or palpable. There was no other swelling in the neck. There was an abdominal surgical scar on right side (post cholecystectomy) and rest of the general and systemic examinations were unremarkable. Her serum free tri-iodothyronine (FT3) was 3.98 pg/ml (normal, 1.7-4.2 pg/ml), and FT4 was 2.30 ng/dl (normal, 0.70-1.80 ng/dl). Her thyroid stimulating hormone (TSH) was less than 0.01 μ IU/ml (normal, 0.30-5.5 μ IU/ml). Anti TPO antibodies were 690 U/ml (0-60 U/ml). The 24 h radioactive iodine-131 uptake of thyroid was elevated at 72%. Her chest X-ray was reported normal. The Endocrinologist referred her for radioiodine treatment of her hyperthyroidism. A thyroid scan done 15 min after intravenous injection of 185 MBq of Technitium-99 m sodium

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pertechnetate revealed a eutopic thyroid gland with relatively smaller right lobe and a left lobe with marked tracer uptake. The salivary glands were suppressed. In addition to these findings an abnormal area of marked uptake was seen in the right upper mediastinum that appeared to be an ETT [Figure 1]. A non-contrast computed tomography (CT) scan revealed both lobes of a eutopic thyroid gland with an ectopic thyroid gland in the right superior aspect of anterior mediastinum [Figure 2]. A diagnosis of Graves' disease involving the eutopic as well as the ectopic mediastinal thyroid was confirmed. The patient was subsequently administered 370 MBq (10 mCi) of Iodine-131 solution orally 5 weeks post I-131 therapy patient was cured of her thyrotoxic symptoms and complained of lethargy and puffiness of face. Her TSH levels were reported as 117.63 μ IU/ml. A Tc-99m sodium pertechnetate thyroid scan confirmed thyroid ablation [Figure 3]. Her post Iodine-131 hypothyroid symptoms were controlled by replacement dose of oral thyroxine.

DISCUSSION

ETT is a rare congenital disorder arising due to a fault in the descent of embryonic thyroid primordium. In majority of the cases the ETT is found at the base of the tongue, commonly referred to as lingual thyroid.^[2,4] The other sites of ETT are mediastinum, upper neck, lower neck, submandibular region, parotid gland, trachea, heart, lung, duodenum, adrenal gland, uterus, lateral to carotid arteries.^[9] ETT may coexist with the eutopic thyroid as in our patient. Genetic defects implicated in ETT with eutopic thyroid gland include mutation in paired box transcription factor Paired box gene 8 (PA \times 8) and thyroid transcription factor 1 and thyroid transcription factor 2. In some sporadic and familial cases heterozygous mutations in PAX8 and thyroglobulin genes have been documented.^[9] In 70% of the cases ETT may be the only functioning tissue.^[2] Majority of patients with ETT are clinically and biochemically euthyroid.^[2,4] Graves' disease is rarely associated with ectopic ETT.^[2,6] On literature review including Pub med search we came across clinical 7 published cases of ETT associated with Grave's disease [Table 1]. Three of these patients had mediastinal ETT, 2 had ETT in neck, one had a dual ETT and in one patient the ETT was present in the submandibular region. The five of these patients were females. The age range was from 31 years to 79 years, with majority presenting in the 3rd and 4th decades of life. Simultaneous occurrence of Graves' disease in eutopic thyroid gland and ETT appears to be clearly documented in 4 such cases.^[1,3-5] Our patient is a rare such case. The possible cause of concurrent involvement of eutopic and ETTs are the high-levels of circulating immunoglobulin's stimulating the identical eutopic and ETTs.^[4] Tc-99m sodium pertechnetate, Iodine-123, Iodine-131 thyroid scintigraphy confirms the presence of eutopic and ETTs. Thyroid scintigraphy is also useful in differentiating thyroid from extra thyroidal neck swellings. CT scan, high-resolution ultrasonogram, Magnetic resonance imaging is other complimentary modalities in aiding the diagnosis. FT3, FT4 and TSH levels confirm the functional

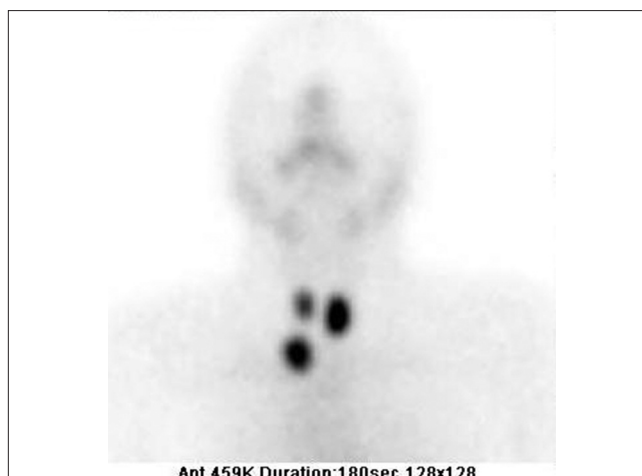


Figure 1: Technetium-99 m pertechnetate thyroid scan showing toxic normal thyroid in neck and toxic ectopic thyroid tissue in mediastinum

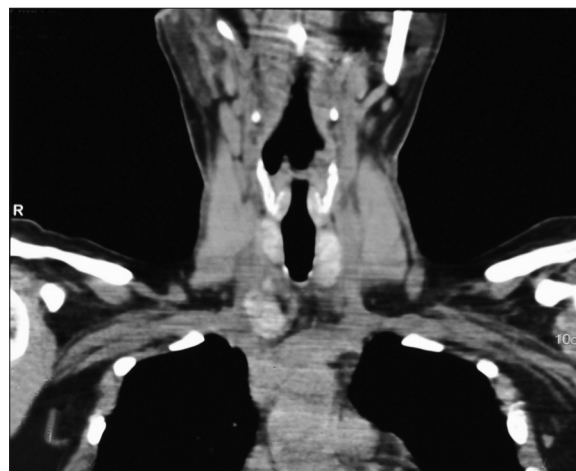


Figure 2: Computerized Axial Tomogram of neck and mediastinum showing ectopic thyroid in neck and ectopic thyroid tissue in mediastinum

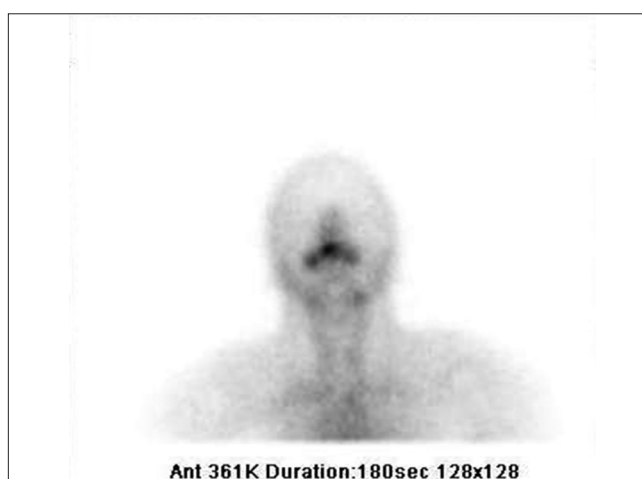


Figure 3: Tc-99m pertechnetate thyroid scan showing complete ablation of eutopic and ectopic thyroid tissue with Iodine-131

status of eutopic and ETTs. In most of the published data Iodine-131 is the preferred treatment.^[3-5,7] In the present, patient thyroid function tests along with the isotope scan

Table 1: Graves' disease involving ectopic thyroid tissue (summary of patients reported in literature)

Author	Age (years)	Sex	Site of ectopic thyroid	Treatment
Mollar, <i>et al.</i> ^[1]	79	F	Mediastinum (Normal thyroid present and involved)	ATD+BB
Prakash, <i>et al.</i> ^[2]	52	F	Mediastinum (Normal thyroid present and not involved)	Iodine-131+T4
Basaria, <i>et al.</i> ^[3]	48	F	Mediastinum (Normal thyroid and ectopic tissue involved)	Iodine-131+T4
Shinto, <i>et al.</i> ^[4]	31	M	Lower neck (Normal thyroid with pyramidal lobe involved)	Iodine-131+T4
Yamauchi, <i>et al.</i> ^[5]	28	F	Upper Neck (Normal thyroid present and involved)	Surgery/ATD
Kisakol, <i>et al.</i> ^[6]	45	M	Dual ectopia (Lingual and Sub mandibular)	Surgery (Sub mandibular)+T4
Kumar, <i>et al.</i> ^[7]	42	F	Sub mandibular (normal thyroid not present)	Iodine-131+T4

ATD: Anti thyroid drugs, BB: Beta blockers, T4: Thyroxine replacement

confirmed effectiveness of radioiodine-131 in treatment of Graves' disease involving the ETT and eutopic thyroid tissue.

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