International Journal of Health Sciences and Research
www.ijhsr.org ISSN: 2249-9571

Case Report

A Rare Case of Double Ectopic Thyroid with Absent Eutopic Thyroid Gland

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ABSTRACT

Ectopic thyroid tissue is defined as any tissue not located anterior and lateral to the second, third and fourth tracheal rings. We present a case where an 8 year old female presented with history of midline neck swelling for the past 1 year with no features of hypothyroidism or hyperthyroidism. Patient had normal thyroid profile, USG and tc99m uptake study showed absent eutopic thyroid gland with subhyoid and lingual thyroid gland.

Key words: ectopic thyroid, lingual thyroid, hypothyroidism.

INTRODUCTION

Ectopic thyroid tissue is defined as any tissue not located anterior and lateral to the second, third and fourth tracheal rings. This can occur anywhere along the path of descent of the normal thyroid gland. Subhyoid thyroid and lingual thyroid, both are common anomalies of the thyroid gland. [1] Both maybe present without normal thyroid gland. However, the concurrent occurrence of all the three anomalies together is a rarity. In a massive Internet search using medline/pubmed services authors could find only twenty such cases of concurrent embryological anomaly reported in the medical literature [1-3] with this background we present one such rare case where the diagnosis and management was found to be interesting.

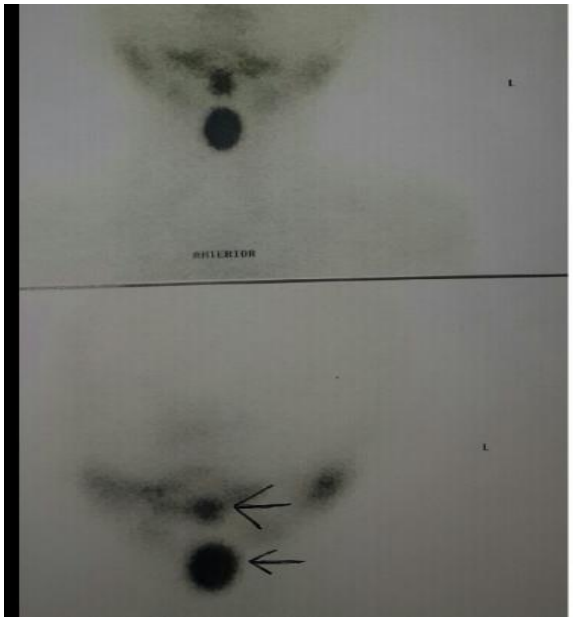
CASE REPORT

8 year old female reported in the endocrinology out door of our tertiary care teaching hospital, with the complaint of midline neck swelling for the past 1 year. The swelling had gradually increased over a period of time. The patient had no other associated complaints like pain, dyspnoea, dysphagia or change in voice. There were no apparent clinical features of hypothyroidism or hyperthyroidism. On examination, a smooth ovoid swelling, 3 cm in maximum diameter with well defined margins was noted in the upper midline of the neck. The swelling was firm in consistency, non-tender and moved with deglutition. A presumptive diagnosis of ectopic thyroid was made.

An *ultrasonography* was also done which reported: A well defined heterogeneous oval shaped soft tissue lesion in the neck 2.1 cm × 1 cm, with the absence of normal thyroid gland, thyroid scan and thyroid function tests to confirm the ectopic thyroid tissue.

The radionuclide scan [99m technetium scan] showed no tracer uptake in the normal thyroid region. A focus of intense tracer uptake was noted in the neck swelling and another focus of increased

tracer uptake was noted just at the base of tongue. The Thyroid function test showed TSH-4.3 [reference value: 0.34–5.6 u IU/ml], FT4-0.81 [reference value: 0.6–1.12 ng/dl] and FT3-3.63 [reference value: 2.5–3.9 ng/ml]. Thus, finally the diagnosis of concurrent lingual thyroid and sub hyoid thyroid with the absence of orthotopic thyroid gland was made.



Radio iodine thyroid uptake scan showing ectopic subhyoid and lingual thyroid

DISCUSSION

Embryologically, during the development of the "Branchial Apparatus" at 4–5 weeks, the thyroid primordium arises as an invagination of the endoderm in the floor of the pharynx and develops caudally, with the descent of the great vessels. It soon becomes bilobed and finally comes at the level of the base of neck to form the thyroid gland at the end of 7th week. This tract usually involutes, but at times the caudal end of this tract may persist in the form of pyramidal lobe. Failure of this tract to involute leaves epithelial remnants or an open area in the duct which develops into thyroglossal duct cysts on accumulation of secretions. [1,4] Lingual thyroid too is an embryological anomaly and originates from failure of thyroid gland to descend from foramen caecum to its normal pre-laryngeal site. [1,2,5]

The simultaneous appearance of double thyroid ectopia with no thyroid gland in the normal site in neck is a rare embryological aberration. Clinically these cases usually present withsymptoms of hypothyroidism. This suprahyoid swelling usually presents by 5 years of age. [2] A distinct lingual swelling at the base of tongue with no clinical symptoms was reported in the previous two cases on examination. [1,2] In this context it would also be important to note that only 0.01% cases of lingual thyroid are symptomatic, warranting treatment. [6] The lingual lesion in our case was picked on radionuclide scanning. These cases may present with hypothyroidism. It would be imperative to note that ectopic thyroid cases rarely present with hyperthyroidism and hypothyroidism has been reported in 33% of such cases. [7]

Ultrasonography plays a very important role in diagnosis of these cases as is evident from our case. USG to detect normal thyroid tissue in the neck. [8,9] In the absence of this tissue on USG, the patients are subjected to radionuclide scanning which confirms the diagnosis. Under the said protocol we could thus diagnose this case of double ectopic thyroid gland.

Physiologically it would be prudent to note that ectopic thyroid gland secretes a hormone that is chemically normal (normal serum levels of T_4, T_3 and TSH). However, in conditions of physiological stress (puberty, menstruation, pregnancy, infection, trauma and surgery) the circulating levels of thyroid hormone become insufficient to the metabolic needs of the body. This leads to increased TSH production and secondary hypertrophy of the gland causing goitre. [10] Hence a patient of double ectopic thyroid gland will certainly benefit from thyroxine treatment. Surgery and radioactive ablation may be needed in cases where there is marked hypertrophy of the ectopic gland not responding to suppressive therapy or in rare cases of malignant change in ectopic gland. In such cases, preoperative counselling should address the need of lifelong thyroid

supplementation. The concern for preservation of functional gland remnants in such cases of double ectopic presenting with hypothyroidism may not be absolute, but should be given due consideration.

In summary, the case in focus merits mention on account of rarity of the occurrence of ectopic thyroid concurrently in both lingual and sub hyoid with the absence of normal anatomical thyroid gland.

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How to cite this article: Kumar R, Sengupta N, Shankar K et al. A rare case of double ectopic thyroid with absent eutopic thyroid gland. Int J Health Sci Res. 2017; 7(6):355-357.

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