

A rare case of triple thyroid ectopia

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ABSTRACT

Various anomalies of thyro-glossal duct have been described, in which the duct may form a cyst or may present as a solid nodule to form an ectopic gland. The ectopic gland can develop along the tract of the duct to give rise to ectopic lingual, sublingual (pre-hyoid) or sub-hyoid (pyramidal) gland, with or without normal pre-tracheal thyroid gland. There are a few reports of double ectopia of thyroid but triple ectopia of thyroid is extremely rare. We have come across a case of triple thyroid ectopia, i.e., thyroid tissue at three locations along the tract of descent of thyro-glossal duct on CT, which has been rarely reported in the world literature, and hence this report.

Key words: Computed tomography of thyroid anomalies, thyroglossal duct, thyroid ectopia

INTRODUCTION

There are many anomalies of development of thyroid gland, which include ectopia along the path of descent of thyro-glossal duct or at other places in the neck.

We have come across triple thyroid ectopia, i.e., nodules of thyroid tissue at three places, namely base of tongue, sublingual or pre-hyoid region and pyramidal lobe. The literature regarding ectopia of thyroid is discussed in the light of CT and isotope study.

The thyroid gland is the first endocrine gland to develop by 24th day of gestation.

It develops from an endodermal thickening of cells that originate from 3rd to 4th branchial pouches. In craniofacial development, the cells move to the base of the tongue to a point known as the foramen caecum. This is where the main anlage of the thyroid develops. From the base of the tongue, the anlage descends from the foramen caecum as the thyroid diverticulum, called as thyroglossal duct, passing

anteriorly to the hyoid bone and thyroid cartilage. It settles as a bilobed organ just inferior to the thyroid cartilage, one lobe on each side of the trachea anterolaterally. The two lobes are joined by an isthmus which unites the lobes over the trachea, anterior to the second and third tracheal rings.

The prevalence of ectopic thyroid tissue ranges between 7 and 10%. Lingual thyroid is the most common ectopic thyroid accounting for 90% of all cases with prevalence between 1:100000 and 1:300000 and a clinical incidence between 1:4000 and 1:10000. Other sites of ectopic thyroid are suprahyoid and infrahyoid, lateral aberrant thyroid, substernal goiters, struma ovarii, and struma cordis. Ectopic thyroid has also been found in larynx, trachea, esophagus, pericardium, diaphragm, and branchial cysts. Rare cases of ectopic thyroid are described in parathyroid, cervical lymph nodes, submandibular gland, duodenal mesentery, adrenals, and carotid bifurcation.^[1]

CASE REPORT

A 42-year-old woman was examined by CT as a client of health check-up to study neck and brain vessels. Her history revealed that she was investigated and counseled by a physician for a nodule over the midline in the neck a few years ago. She was subjected to thyroid function tests, which were normal and excluded hypo or hyper thyroidism. She was correctly advised not to do any further investigation, worry about it or get rid of it surgically.

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CT revealed ectopic thyroid nodules over the surface of posterior tongue, anterosuperior and close to body of hyoid bone and in sub-hyoid region as an enlarged pyramidal lobe. The normal thyroid lobes and isthmus were absent and as a result common carotid and internal jugular veins were closely applied to trachea without the interposing thyroid lobes [Figure 1]. All these were un-connected and separated by 5-8 mms in craniocaudal direction in the midline. These were hyperdense and showed enhancement (post contrast CT value measuring more than 200). A nuclear scan was performed using 3 millicuries of Technetium 99^m pertechnetate administered intravenously. The static images were acquired at 20 minutes. The frontal projection showed two hot spots, a smaller and upper one between submandibular glands due to lingual ectopia and the lower larger one due to combined activity from pre-hyoid (sublingual) and pyramidal ectopic gland [Figure 2].

As she was now asymptomatic and nuclear scan had ruled out any abnormal thyroid function, repeat thyroid function tests were also not recommended.

DISCUSSION

The thyroid gland is the first endocrine gland to develop in a foetus by 4th week. The initial thyroid precursor, called as thyroid primordium, begins as a midline diverticulum or epithelium-lined tubular structure, called as thyroglossal duct. It develops over the base of the tongue in a V shaped sulcus between tuberculum impar and copula.



Figure 1: Post contrast axial images showing three hyperdense, (a) enhancing nodules over the surface of posterior tongue measuring 9×12 mms in size. (b) second ectopic nodule at a lower level just anterosuperior to the body of hyoid bone measuring 11×9 mms in size (c) the third nodule below the hyoid bone and anterior to thyroid cartilage as an enlarged pyramidal lobe measuring 19×24 mms in size. (d) an axial image showing absent lateral lobes of thyroid with the result that carotid and jugular vessels are closely applied to trachea

The thyro-glossal duct migrates or descends inferiorly anterior to hyoid bone and then to normal pre-tracheal location of thyroid in the neck by 7th week and disappears by 10th week. Most of the thyroid gland is derived from this median anlage, while two lateral anlages derived from the 4th pharyngeal pouch contribute up to 30% of the thyroid tissue.^[2] The most caudal end of thyroglossal duct gives rise to a pyramidal lobe, which may occur in about 30% of individuals. When present it is a very small tubular lobe above the isthmus.

Very rarely further downward or excessive descent may occur outside the tract of thyroglossal duct or embryonic rests may occur in lateral neck, walls of trachea/larynx/esophagus, mediastinum, ovary, pericardium, aortic valve, parotid gland, abdomen, branchial cyst, submandibular gland or adrenal gland.

Remnants of thyroid tissue may develop into ectopic thyroid anywhere along the path of thyro-glossal duct like lingual (over the surface of tongue), sublingual (pre-hyoid) or sub-hyoid (pyramidal lobe) regions.

Genetic research has shown that the gene transcription factors TTF-1 (Nkx 2-1), Foxe1 (TTF-2) and PAX-8 are essential for thyroid morphogenesis and differentiation. Mutation in these genes may be involved in abnormal migration of the thyroid.^[3]

Ectopia of thyroid at lingual, sublingual (pre-hyoid) or sub-hyoid location can be with normal or absent pre-tracheal gland. In 70% of cases of lingual thyroid the cervical thyroid is absent. In our case too normal thyroid lobes were absent.

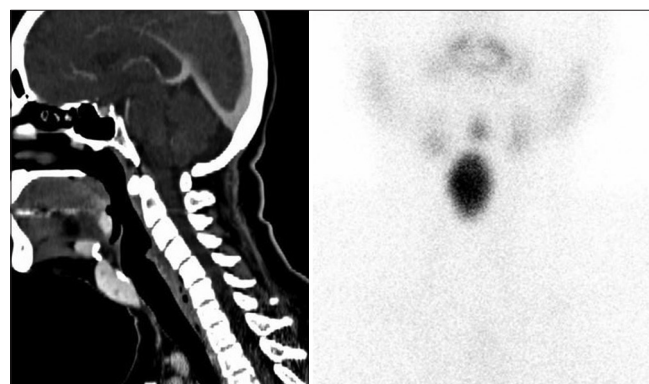


Figure 2: Left half of the image is a sagittal MPR image revealing triple ectopia in a single image. All ectopic nodules are separated by a few mms. The lingual nodule is just behind a small depression or sulcus and is partly buried in the tongue. Right half of the image is a frontal isotope scan shows two hot spots or areas of uptake. The upper one is between submandibular glands and in the midline typically corresponding to lingual ectopia. The lower and larger uptake is due to combined pre-hyoid gland and enlarged pyramidal lobe

Occasionally there may be dual thyroid ectopia confirmed by CT as well as a nuclear scan, which has been reported in a few articles.^[4,5] The dual ectopia was lingual and pre-hyoid in one case, while pre- and sub-hyoid in another. In both cases cervical thyroid was absent. Al-Akeely^[5] claimed that there were only eight cases of dual ectopia reported till 2002.

Review of literature reveals that only two cases of triple ectopia have been reported.

Barai,^[6] demonstrated triple ectopia in a hypothyroid child in frontal and lateral nuclear scans. However, no exact anatomical correlation was possible, as CT was not performed. Jain and Pathak,^[7] have also reported a case of triple ectopia of thyroid.

Our case is the third and exceptional case of triple thyroid ectopia with well-defined thyroid nodules seen on CT and further investigated by nuclear scan to be present over tongue and in pre-hyoid and sub-hyoid (pyramidal lobe) locations.

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