Case Reports

Dual ectopic thyroid with lingual and anterior hyoid ectopic thyroid tissue in a Chinese girl

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ABSTRACT

Ectopic thyroid means the presence of thyroid gland tissue in abnormal position but if it is related to embryogenesis defect, it is called lingual thyroid. We report a recent case of a patient initially diagnosed with a neck schwannoma, who was subsequently found to have both lingual and anterior hyoid ectopic thyroid tissue, with no thyroid tissue visible in the usual pretracheal location on CT, or thyroid scan. To avoid unnecessary misdiagnosis, thyroid scan and a neck CT, or ultrasound (US) examinations should be performed routinely in cases presenting with a neck mass.

Case Report. An 11-year-old girl presented with a history of left-sided neck swelling for 2 months. The swelling had gradually increased in size over the past 2 months, and was not associated with dysphagia or any change in her voice. Fine needle aspiration (FNA) cytology of the palpable neck swelling was suggestive of schwannoma.

Interestingly, US examination revealed a well-encapsulated lesion anterior to the hyoid with heterogeneous echotexture and hypoechoic areas, and no thyroid gland in the usual position. Laryngoscopy showed an irregular erythematous nodule at the base of the tongue, which moved on tongue protrusion and deglutition (Figure 1). A CT scan of the neck revealed 2 homogeneous hyperdense enhancing lesions, one in the

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tongue (1.8 cm diameter), and one anterior to the hyoid (1.7 cm diameter). No glandular tissue was identified in the normal pretracheal position (Figure 2). A sodium pertechnetate Tc-99m thyroid scan showed uptake in the region of the tongue base, and in the region of the anterior neck swelling (Figure 3). There was no uptake at the usual site of the gland. Thyroid function tests showed: total tri-iodothyronine level - 139.80 ng/dL (normal range: 70-220 ng/dL); total thyroxine level - 3.95 μg/dL (normal range: 4.5-15.4 μg/dL); free tri-iodothyronine level - 2.99 pg/mL (normal range: 1.78-5.60 pg/mL); free thyroxine level - 0.80 ng/dL (normal range: 0.5-2.3 ng/dL), thyroid-stimulating hormone level - 9.21 μIU/mL (normal range: 0.25-7.31 μIU/mL). Based on these findings, a diagnosis of dual ectopic thyroid was established. The protocol was reviewed and approved by the human ethics committee of the Medical Center of Fudan University, and written informed consent was obtained from the parents of the patient.

Discussion. The prevalence of clinically apparent lingual thyroid is approximately one in 100,000, with females being affected 4-8 times more often than males. Dual ectopic thyroid is extremely rare, and only 13 cases have been reported in the English literature. In the pediatric population, it has been reported that up to 24% of children with primary non-goitrous hypothyroidism have thyroid ectopia. Simultaneous thyroid ectopia at the base of the tongue, and in the anterior hyoid region is extremely rare. A similar study of dual ectopic thyroid present in the lingual and infrahyoid areas with no thyroid tissue in the pre-tracheal area in a 15-year-old girl was reported.

Most ectopic thyroid glands are asymptomatic and benign. However, malignant transformation has been reported in 1-3% of cases, and approximately 80% of such tumors are papillary carcinomas. If there is a risk of hemorrhage or malignant transformation, biopsy should be undertaken. Investigation of the anatomical position all ectopic thyroid tissue is therefore important. Neck US and CT scan, thyroid scans and thyroid uptake tests are all valuable diagnostic modalities, and should be performed routinely to avoid unnecessary misdiagnosis. In this case, in order to make a definite diagnosis for the left-sided neck swelling, FNA was carried out. The case was diagnosed as a neck schwannoma. However, we did not palpate the thyroid gland in the usual position, during a routine physical examination. And then, US, CT and thyroid scan, and thyroid uptake tests were used to look for the thyroid gland. Eventually, we identified that the girl have lingual and anterior hyoid ectopic thyroid.

Figure 1 - Laryngoscopy image showing a 2×2 cm mass protruding from the base of the tongue (arrow).

Figure 2 - Contrast-enhanced axial CT showing: A) a well-delineated, 1.8 cm diameter, strongly enhancing homogenous round mass is seen at the base of the tongue (big arrow), as well as a left neck mass (Schwannoma, small arrow); B) lower section of contrast-enhanced axial CT scan of the neck showing a 1.7 cm diameter, heterogeneous, moderately enhancing round mass in the anterior hyoid space (arrow); C) no thyroid gland is seen in the pretracheal region; D) sagittal view clearly showing the location of both masses.

Figure 3 - Sodium pertechnetate thyroid Tc-99m scan showing: A) anterior view. Focal uptake is shown between the submandibular glands. No uptake is seen in the normal pretracheal region in the lower neck; B) Lateral view. Two foci of radiotracer uptake in the tongue base (arrows) showing dual ectopic thyroid glands.
thyroid tissue with no thyroid tissue visible in the usual pretracheal location.

The management of patients with ectopic thyroid tissue depends on the severity of symptoms and on complicating factors, such as puberty, pregnancy, ulceration, and hemorrhage. Thyroid hormone therapy will typically suppress the production of thyroid-stimulating hormone, and is useful in cases of glandular enlargement. Radioactive iodine ablation is reserved for older patients who remain symptomatic, and for whom surgery is not thought appropriate, however, it is contraindicated in children as the systemic doses required have potentially damaging effects on the gonads and other organs. In this case, thyroid function tests were normal, and no immediate treatment was given. The patient will undergo regular follow-up.

In summary, ectopic thyroid tissue is an extremely rare entity with diagnostic and treatment challenges. In a patient with a neck mass, a pre-operative thyroid scan, and a neck CT, or US scan should be undertaken to avoid unnecessary misdiagnosis.

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References


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