

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

Rui Dong, PhD, Kai Li, PhD, Gongbao Liu, BM, Shan Zheng, MD.

## ABSTRACT

يعد ظهور الأنسجة الغدية في غير مكانها الطبيعي أمراً غير شائع، كما أن ظهورها المضاعف يعد أمراً نادراً للغاية. نستعرض في هذا المقال حالة مريضة تبلغ من العمر ١١ عاماً، وقد كانت مصابة بظهور الأنسجة الغدية المزدوجة في غير مكانها الطبيعي التي حاكت في شكلها الأنسجة الغدية الأحادية في غير مكانها الطبيعي في الفص الأمامي من الغدة الدرقية. وقد تبين ظهور الأنسجة الغدية في غير مكانها الطبيعي في العرض الجانبي، مع بؤر في المناطق اللامية اللسانية الأمامية، ولكنها لم تظهر في المنطقة ما قبل القصبة الهوائية. وكان من المقرر أن يجري المريض عملية جراحية بعد تشخيصه بالورم الشفاني في العنق، ولكن كشف فحص الغدة الدرقية، والتصوير المقطعي للرقبة عن وجود الأنسجة الغدية في غير مكانها الطبيعي قبل العملية الجراحية. وهكذا نستنتج بأن يجب عمل مسح للغدة الدرقية إما بالتصوير المقطعي أو بالموجات فوق الصوتية بشكل روتيني في الحالات التي تظهر فيها كتل في الرقبة لتجنب التشخيص الخاطئ.

Ectopic thyroid is not a common disease, and a double ectopic thyroid is extremely rare. We report a recent case of dual ectopic thyroid in an 11-year-old girl mimicking mono-ectopic lingual thyroid on the anterior view of the thyroid scan. The dual ectopic thyroid was shown on the lateral view, with foci in the lingual and anterior hyoid regions, but no thyroid tissue in the pre-tracheal region. She had originally been scheduled for surgery with a diagnosis of neck schwannoma, however, pre-operative thyroid scan and CT scan of the neck revealed the presence of dual ectopic thyroid tissue. Thyroid scan, and either CT, or ultrasonography should be performed routinely in cases presenting with a neck mass to avoid unnecessary misdiagnosis.

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*From the Department of Pediatric Surgery, Children's Hospital of Fudan University, and Key Laboratory of Neonatal Disease, Ministry of Health, Shanghai, China.*

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*Address correspondence and reprint request to: Dr. Shan Zheng, Department of Pediatric Surgery, Children's Hospital of Fudan University, and Key Laboratory of Neonatal Disease, Ministry of Health, 399 Wan Yuan Road, Shanghai 201102, China. Tel. +86 (021) 64931007. Fax. +86 (021) 64931901. E-mail: szheng@sbmu.edu.cn*

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.

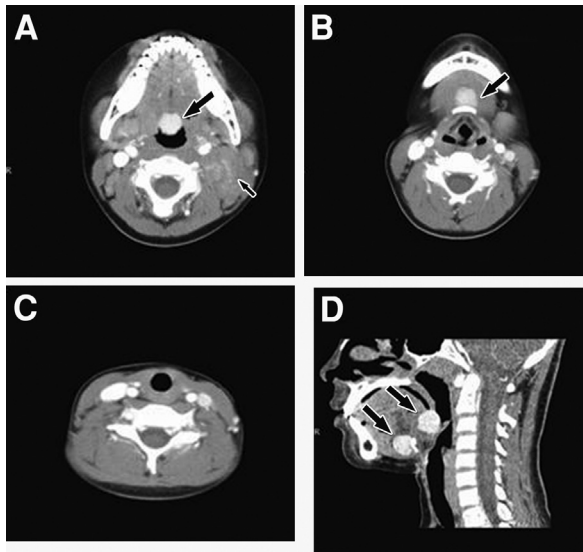
**Case Report.** An 11-year-old girl presented with a history of left-sided neck swelling for 2 months. The swelling had gradually increased in size over the past 2 months, and was not associated with dysphagia or any change in her voice. Fine needle aspiration (FNA) cytology of the palpable neck swelling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

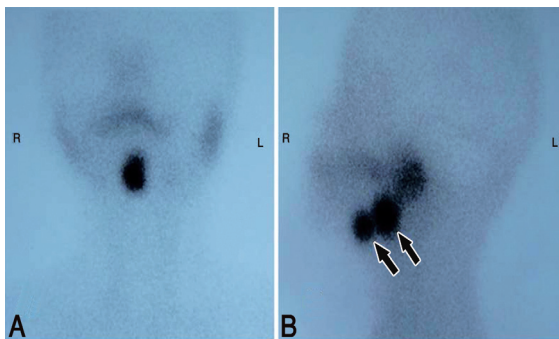
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**Figure 1** - Laryngoscopy image showing a 2x2 cm mass protruding from the base of the tongue (arrow).



**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.

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.<sup>1</sup> Dual ectopic thyroid is extremely rare, and only 13 cases have been reported in the English literature.<sup>2,3</sup>

In the pediatric population, it has been reported that up to 24% of children with primary non-goitrous hypothyroidism have thyroid ectopia.<sup>4</sup> 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 pretracheal area in a 15-year-old girl was reported.<sup>5</sup>

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.<sup>6</sup> 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 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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