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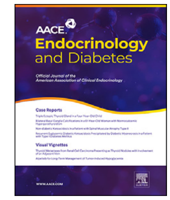

Case Report

Triple Ectopic Thyroid Gland in a 4-Year-Old Child

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ABSTRACT

Background/Objective: Triple ectopic thyroid is exceedingly rare. The objective of this report is to present a case of triple ectopic thyroid that presented as a right submandibular mass in a euthyroid child.

Case Report: A 4-year-old boy presented with a right-sided neck mass for 2 months. Neck ultrasound revealed a soft tissue mass adjacent to the submandibular salivary gland. Post intravenous contrast computed tomography scan, an additional 2 enhancing nodules were identified in the sublingual and hyoid areas. No thyroid tissue was identified in its normal pretracheal region. A fine-needle aspiration biopsy under ultrasound guidance of the right neck mass revealed numerous follicular epithelial cells in a background of homogenous colloid, characteristic of normal thyroid tissue. A Tc-99m pertechnetate thyroid scan demonstrated that the other 2 nodules were also ectopic thyroid tissue. Further workup revealed a thyroid stimulating hormone of 6.19 mIU/L (0.7-6.4) and a free thyroxine level of 1.0 ng/dL (0.8-2.0).

Discussion: Delayed presentation of ectopic thyroid tissue as a mass presents diagnostic challenges, particularly when it is not found around the course of the thyroglossal duct and is associated with 2 additional ectopic thyroid nodules. Very few cases of triple thyroid ectopy have been reported particularly in pediatric patients.

Conclusion: Although most cases of ectopic thyroid in children are identified shortly after birth, a late presentation can occur. In a euthyroid patient presenting with a mass and absence of a thyroid gland in the normal position, thyroid ectopy should be included in the differential diagnosis.

Introduction

The ectopic thyroid gland is a developmental abnormality that results from an aberration in the descent of the thyroid to its normal pretracheal position during the first few weeks of fetal life.[1,2] Ectopic thyroid glands are usually single and constitute more than two-thirds of cases of thyroid dysgenesis.[3] Children with ectopic thyroid glands are typically identified at birth due to the finding of an elevated thyroid stimulating hormone (TSH) on the newborn screen for congenital hypothyroidism (CH). It can also present later in life with a neck mass with or without symptoms of hypothyroidism. Here, we present a 4-year-old boy who was seen for a painless neck mass and was found to have 3 separate foci of ectopic thyroid tissue. To our knowledge, this is the youngest case of triple thyroid ectopy that has been reported. Additional cases of multiple ectopic thyroid foci in the literature involving pediatric patients are summarized.

Case Report

A 4-year-old boy was referred for evaluation of a right-sided neck mass. His parents reported that it had been present for approximately 2 months. He had no symptoms of airway obstruction or hypothyroidism and had been growing normally along the 19% tile for height. On examination, the lesion was nontender, firm,

Abbreviations: CH, congenital hypothyroidism; TSH, thyroid stimulating hormone.

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and nonmobile (Fig. 1). The remainder of his physical examination was unremarkable. The patient had no past medical history and was not on any medication. His newborn screen for CH was reportedly normal. His parents indicated that they were unaware of any family history of thyroid disorders. He was diagnosed with reactive lymphadenitis and treated with 2 weeks of oral antibiotics without apparent change in the mass.

Neck ultrasound was performed which revealed a 20 × 18 × 15 mm smoothly marginated hypoechoic highly vascularized right submandibular solid mass just below the salivary gland (Fig. 2). A post contrast computed tomography scan demonstrated the homogenous enhancing right submandibular mass along with an additional 2 midline nodules with similar characteristics in the sublingual area and at the hyoid (Fig. 3). No thyroid tissue was identified in the pretracheal area. A fine-needle aspiration biopsy under ultrasound guidance of the right neck mass revealed scattered groups of follicular epithelial cells in a background of abundant colloid characteristic of normal thyroid tissue. Technetium-99m pertechnetate scintigraphy confirmed that the other 2 midline lesions were also ectopic thyroid (Fig. 4).

Laboratory evaluation revealed a TSH of 6.19 mIU/L (0.7-6.4) and free T4 of 1.0 ng/dL (0.8-2.0). Following these results, the patient was managed with observation alone. A repeat TSH 3 months later was again normal for age at 4.33 mIU/L. Follow-up was scheduled with both endocrinology and otolaryngology.

Discussion

We describe a rare case of a euthyroid child with triplet ectopic thyroid that presented as a right submandibular mass that was diagnosed on fine needle aspiration and managed without surgical or medical intervention. In children, ectopic thyroid tissue is usually identified soon after birth following a diagnosis of CH. Although determining the etiology of CH does not change the initial management, it can provide valuable prognostic information that is appreciated by parents. For example, children with eutopic glands may be eligible for a trial off levothyroxine after age 3 as approximately one-third of them are ultimately found to have transient CH.[4] Regardless of the etiology, parents should also be informed of the need for frequent monitoring of the TSH and clinic follow-up as well as what signs and symptoms should prompt them to contact their child's health care provider. Thyroid imaging modalities that are employed in infants with CH include technetium-99m pertechnetate scintigraphy and thyroid ultrasonography.[5] High accuracy in identifying eutopic thyroids using ultrasound in infants with congenital hypothyroidism has been reported.[6]

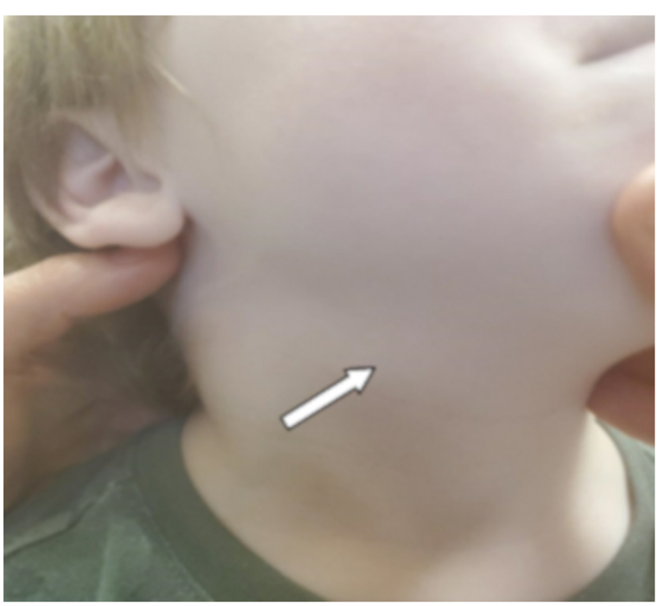

Fig. 1. Ectopic thyroid gland in submandibular area (arrow).

> **Highlights**
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> - Occurrence of multifocal ectopic thyroids is extremely uncommon
> - Thyroid ectopy can be misdiagnosed as cervical lymphadenitis, leading to unnecessary treatment
> - Imaging is needed to differentiate ectopic thyroid from other neck masses
>
> **Clinical Relevance**
>
> This case highlights the rarity of multifocal ectopic thyroid, emphasizing the need for careful evaluation of pediatric neck masses to avoid misdiagnosis. Early recognition and appropriate imaging can prevent unnecessary interventions, ensuring better management of thyroid function and long-term outcomes in affected patients.

In older children and adults, thyroid ectopy is most often found incidentally in the course of imaging for an unrelated concern. Although most commonly located along the path of embryologic descent of the thyroid, with lingual tissue comprising around 90% of cases,[7,8] ectopic thyroid tissue has been described in such diverse locations as the abdomen, adrenal glands, skull base, and breast tissue.[9-11]

Ectopic thyroid glands are typically asymptomatic but can present with symptoms depending on their location, ranging from neck masses and primary hypothyroidism to obstructive symptoms such as dysphagia and respiratory issues.[2] Our patient presented with a neck mass that was mistakenly interpreted as cervical lymphadenitis, a condition that is particularly common in pediatric patients. Consequently, he was treated with antibiotics before the correct diagnosis was made.

A literature review has identified only 16 cases of multiple ectopic thyroid glands of which only 7 cases have been reported in pediatric patients. Most of them (86%) presented with neck swelling, 3 had hypothyroidism and 4 had normal thyroid function tests, similar to our case (Table).[10] All cases of dual or triple ectopic thyroid glands presented as midline neck swelling. In the majority, the most inferior ectopic gland was at the level of the floor of the mouth or just inferior to the hyoid. Our patient is unique as the most inferior ectopic thyroid gland was to the right and inferior to the submandibular salivary gland and presented with a right lateral mass. To our knowledge, this is the youngest reported case of triplet ectopic thyroid tissue.

Our patient's TSH was in the normal range for a young child, and therefore no therapy was initiated. Ongoing surveillance of thyroid function will be essential since he may need to be treated with levothyroxine in the future.

Conclusion

Ectopic thyroid tissue can rarely escape early detection and present during childhood with a neck mass. Multiple thyroid ectopy is extremely rare. In cases where no thyroid tissue is seen in the normal location, as in our patient, choosing needle biopsy over excision biopsy is advised when dealing with a homogeneous enhancing mass that might be an ectopic thyroid. This case

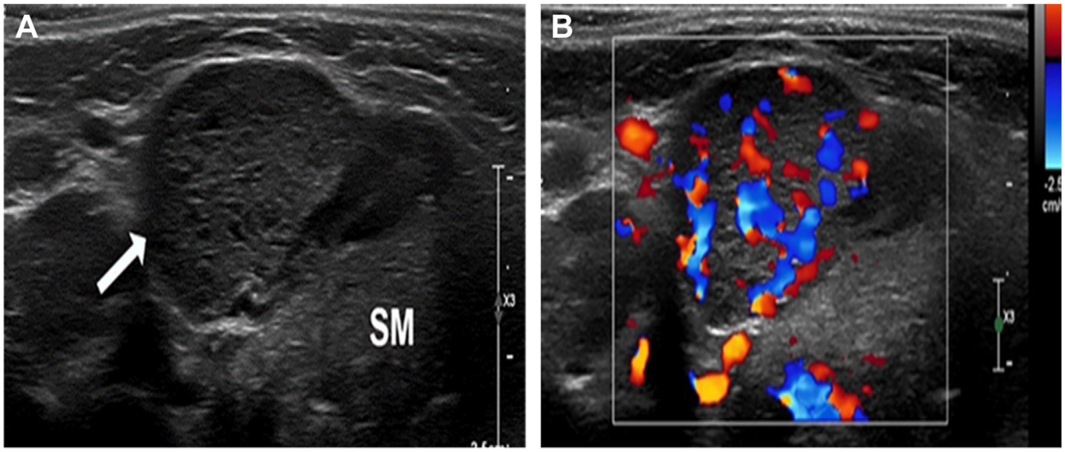

Fig. 2. Transverse ultrasound of the right submandibular area. *A*, Solid hypoechoic mildly heterogeneous mass (white arrow) lateral to the salivary submandibular gland (SM). *B*, Color Doppler shows marked increased vascularity of the mass.

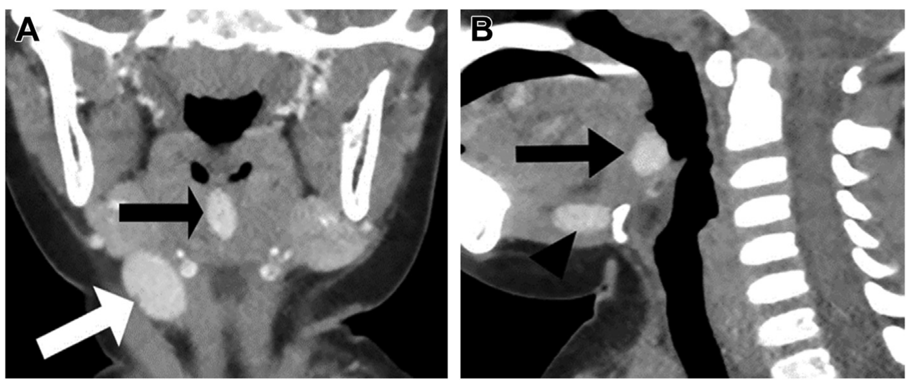

Fig. 3. Post intravenous contrast computed tomography scan of the neck. *A*, Coronal reformat demonstrates marked homogeneous enhancement of right ectopic submandibular thyroid (white arrow) inferior to the salivary gland and midline lingual ectopic thyroid (black arrow). *B*, Sagittal reformat demonstrates midline marked homogeneous enhancement of the lingual (arrow) and at the level of the hyoid at the floor of the mouth (arrowhead).

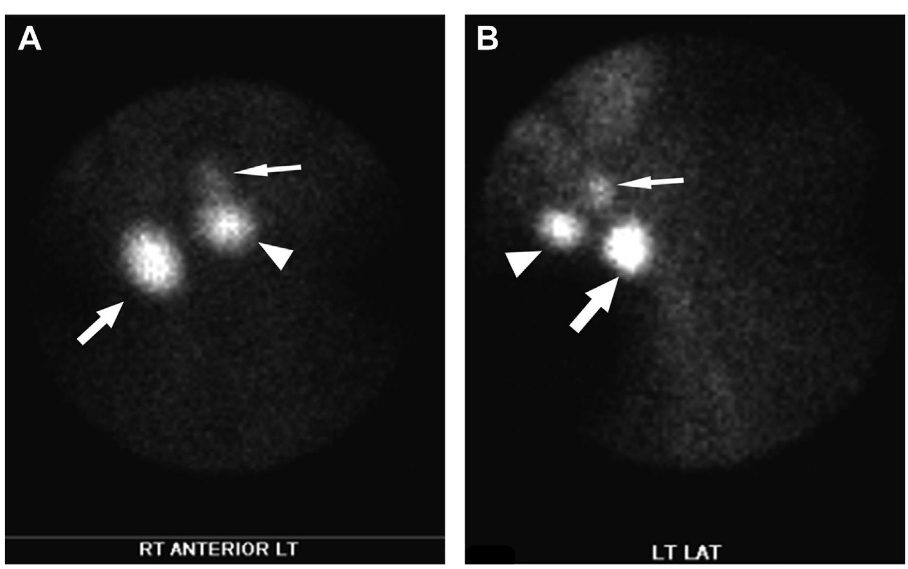

Fig. 4. Demonstration of the triple ectopic thyroid on nuclear medicine Technetium-99m scintigraphy. Anterior (*A*) and left lateral (*B*) pinhole images show 3 distinct locations of uptake in the right submandibular (thick arrow), midline lingual (arrowhead), and floor of the mouth (thin arrow) ectopic thyroid tissue.

Table
Summary of Pediatric Patients With Multiple Ectopic Thyroid Glands

Reference	Age Y/ Gender	Location	Imaging	TFT	Clinical Presentation
Barai et al[12]	6/M	Lingual, suprahyoid and in the region of the cricoid cartilage	Thyroid scan	Severely hypothyroid	Neck swelling and global developmental delay
Nilegaonkar et al[13]	16/F	Base of the tongue, At the level of hyoid and the suprahyoid region	Thyroid scan CT, US	Normal	Thyroglossal cyst mimic
Konde et al[14]	16/F	Lingual, sublingual and prehyoid	Neck US, CT, thyroid scan	Normal	Midline neck mass
Kuramoto et al[1]	10/F	Base of tongue, pre hyoid region and pretracheal region	CT, thyroid scan	Normal at presentation, became hypothyroid 5 months later	Midline neck mass
Thomas et al[15]	14/F	Omentum, left adnexa, peritoneum, spleen, abdominal wall and rectum	CT	Normal	Adnexal mass
Passah et al[16]	5/F	Base of tongue, prehyoid region and anterior to the thyroid cartilage	Neck US, thyroid scan, SPECT/CT	Subclinical hypothyroidism	Midline neck mass
Argwal et al[17]	11/F	Base of tongue, prehyoid and infrahyoid location	Neck US, thyroid scan, SPECT/CT	Normal	Midline neck swelling

Abbreviations: CT = computed tomography; F = female; M = male; SPECT = single-photon emission computed tomography; TFT = thyroid function test; US = ultrasonography.
Modified from Qin et al.[10]

underscores the importance of imaging studies in the evaluation of childhood neck masses and of considering the possibility that thyroid ectopy may be present.

Disclosure

The authors have no conflicts of interest to disclose.

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Statement of Patient Consent

Written consent by patient has been obtained.

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