Case Report

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Dual Ectopic Thyroid Tissue: Diagnostic Approaches to a Rare Clinical Anomaly

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Abstract

Dual ectopic thyroid tissue represents an exceedingly rare developmental anomaly, with only a handful of cases described in the literature. The simultaneous occurrence of two ectopic thyroid foci in the presence of a normally sited gland is particularly uncommon. We present the case of a 60-year-old Chinese woman who presented with a painless swelling in the left mid-lateral neck of six months' duration. Clinical assessment and contrast-enhanced computed tomography suggested thyroid-like tissue in the mid-lateral neck area, while thyroid function tests remained normal. Fine needle aspiration cytology yielded benign follicular nodules, and radionuclide single photon emission computed tomography demonstrated two hyperfunctioning ectopic thyroid foci at the base of the tongue and in the left mid-lateral neck region, with preserved uptake in the orthotopic gland. The left neck mass was excised and confirmed histologically as thyroid tissue without evidence of malignancy. The patient made a full recovery with no further treatment required. This case illustrates the value of considering ectopic thyroid tissue in the differential diagnosis of neck masses, particularly when encountered in atypical locations.

Keywords: Ectopic thyroid, thyroid gland, lingual thyroid, neck mass, radionuclide imaging, case report

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Introduction

The thyroid gland, the earliest endocrine structure to arise during embryogenesis, originates between the third and fourth weeks of gestation (1). The gland arises chiefly from the endoderm, forming the isthmus and the lateral lobes, with additional contributions from paired lateral anlages that supply C-cells and parts of the lateral thyroid (1,2). During development, the thyroid descends from the foramen caecum at the tongue base to its definitive cervical position.

Ectopic thyroid tissue is defined as thyroid tissue located outside of its normal anatomical position and results from aberrations in this migratory process. It is most commonly found at the lingual site but may also occur in the subhyoid, mediastinal, or lateral neck regions. Ectopic thyroid tissue may therefore arise if descent is incomplete, migration is arrested at any point, or the tissue develops aberrantly at an alternative site (1-3).

The prevalence of solitary ectopic thyroid is estimated at 1 in 100,000 to 1 in 300,000 individuals, with a clear female predominance (3). Dual ectopic thyroid tissue is far rarer.

Meng et al. (4) reported a prevalence as low as 0.05% for dual ectopic thyroid tissue among patients undergoing thyroid scintigraphy, highlighting the rarity of this entity. Our case is especially noteworthy for the coexistence of dual ectopic thyroid foci alongside a normally positioned thyroid gland. Such a presentation exemplifies a rare embryological event, offering unique insights into the complexities of thyroid development and migration.

Case Presentation

A 60-year-old Chinese female with treated hypertension presented with persistent, painless left neck swelling for six months. She reported no symptoms suggestive of thyroid dysfunction or malignancy of the upper aerodigestive tract. On physical examination, a firm, mobile 6x4 cm left mid-lateral neck mass was noted (Figure 1); it was fluctuant on external palpation and not associated with cervical lymphadenopathy. Flexible nasolaryngopharyngoscopy revealed no visible mucosal lesions, masses, or airway compromise. The lingual ectopic thyroid tissues identified on radionuclide single photon emission computed tomography (SPECT) were limited in size, submucosally and located at the base of the tongue, explaining its absence on endoscopic examination.

Fine needle aspiration cytology (FNAC) demonstrated benign thyroid follicular nodules without nuclear atypia. Thyroid function tests were within normal reference limits. Contrast-enhanced computed tomography (CT) demonstrated a heterogeneous enhancing lesion in the left mid-lateral neck region with imaging characteristics resembling thyroid parenchyma (Figures 2a, 2b). The orthotopic thyroid gland was identified in its normal cervical

Figure 1. A large left mid-lateral neck swelling is seen (yellow arrow)

location, without significant nodal enlargement (Figure 2c). No direct communication was observed between the left lateral neck lesion and the left thyroid lobe. Radionuclide thyroid SPECT detected increased uptake in both the lingual and the left mid-lateral neck regions, with preserved uptake in the orthotopic thyroid gland (Figures 3, 4) confirming dual ectopic thyroid tissue.

The left mid-lateral neck mass was excised under general anesthesia (Figure 5) due to its significant size, progressive enlargement, cosmetic concern, and the need for definitive histopathological confirmation, as ectopic thyroid tissue may occasionally harbor malignancy despite benign FNAC findings. In contrast, the lingual ectopic thyroid was left in place, and no further treatment was planned, as it was asymptomatic, demonstrated physiological radionuclide uptake, and showed no features warranting biopsy or

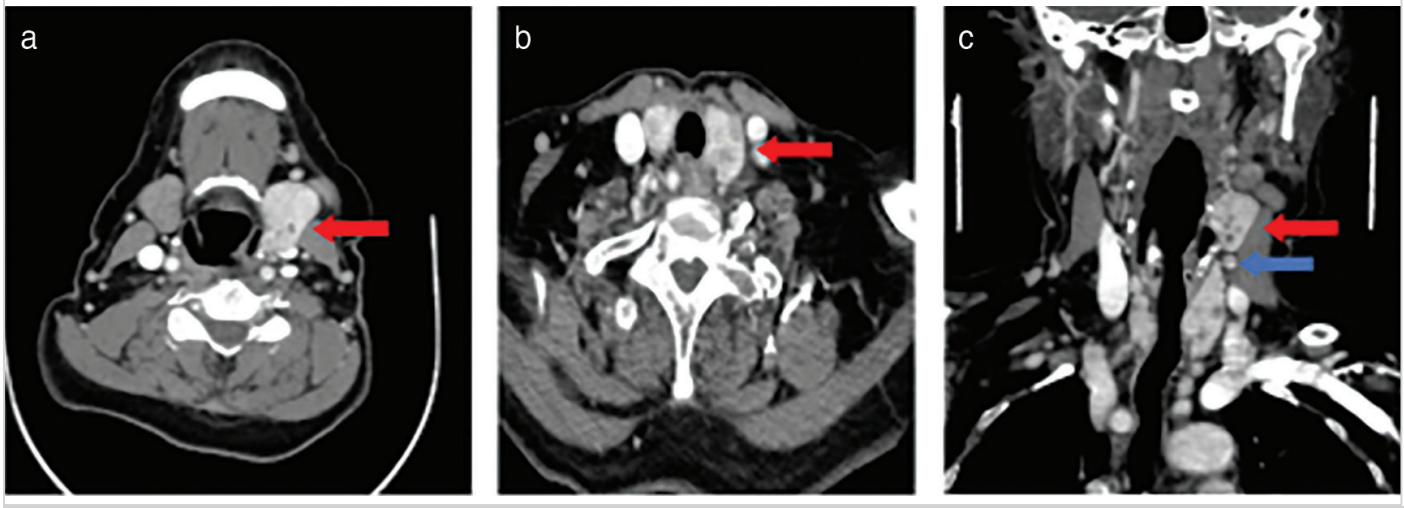

Figure 2. a. A heterogeneous enhancing lesion at the left lower lateral neck region in axial view CT (red arrow), **b.** which shows similar enhancement with the thyroid parenchyma (red arrow). **c.** Contrasted CT in coronal view, which shows no direct communication within the left mid-lateral neck region lesion (red arrow) with the upper pole of the left thyroid lobe (blue arrow)
CT: Computed tomography

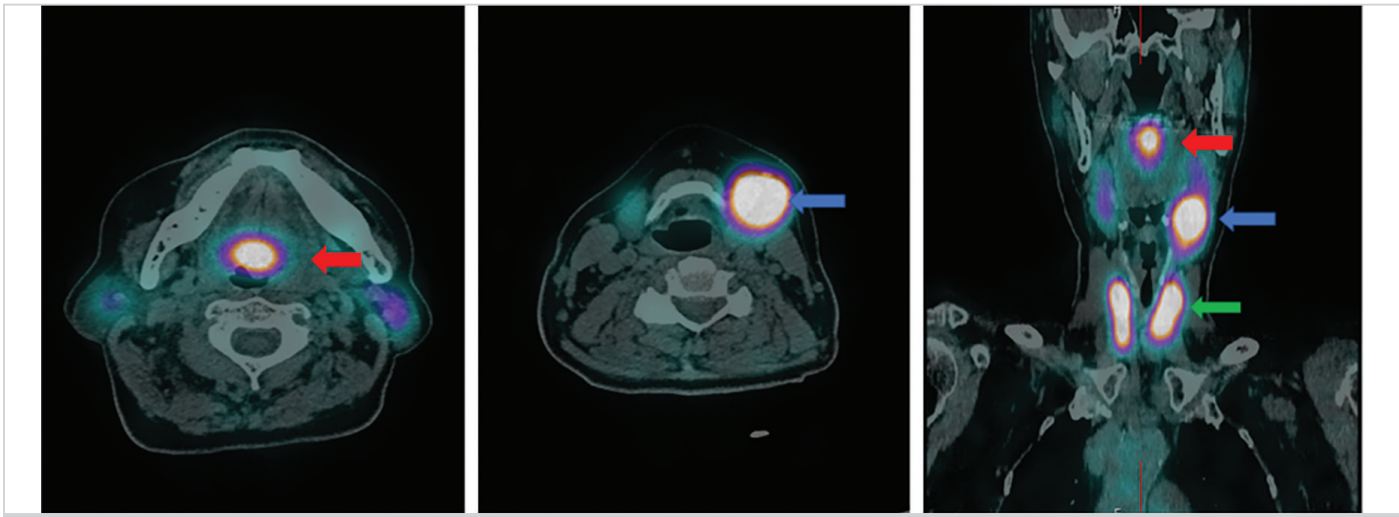

Figure 3. Radionuclide imaging thyroid single photon emission computed tomography fused with CT image, which shows uptake at the lingual region in axial view (red arrow) and left mid-lateral neck region lesion in axial view (blue arrow) and normally located thyroid nodules (green arrow)
CT: Computed tomography

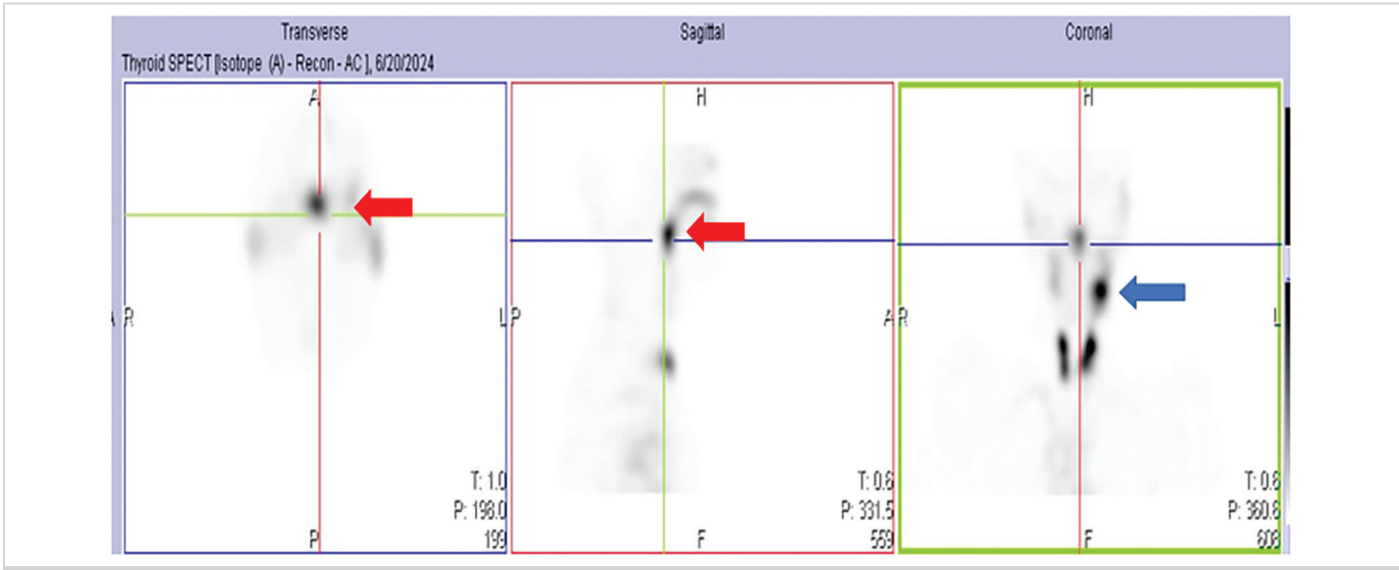

Figure 4. Radionuclide imaging with thyroid single photon emission computed tomography image, which shows uptake at the lingual region in sagittal view (red arrow) and left mid-lateral neck region lesion in coronal view (blue arrow)

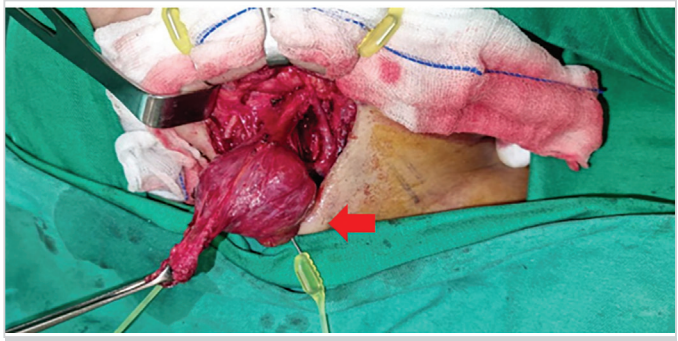

Figure 5. Intraoperative clinical picture showing a mass in the mid-lateral neck region lesion (red arrow)

excision. Histopathologic examination confirmed benign thyroid tissue without malignant features. Postoperative follow-up confirmed an uneventful recovery, with stable thyroid function and no need for additional treatment. Written informed consent was obtained from the patient.

Discussion

The coexistence of multiple ectopic thyroid tissues in the presence of a normally located thyroid gland is an extraordinary phenomenon, posing significant diagnostic and management challenges due to its rarity and limited documentation (5). Meng et al. (4) identified six cases of dual

ectopic thyroid tissue in a five-year review of 11,905 thyroid scintigraphy scans, with the most frequent ectopic sites being lingual, subhyoid, and sublingual. Recognition of this rare entity is essential to prevent misdiagnosis and unnecessary surgical interventions.

Although ectopic thyroid tissue is a congenital developmental anomaly, it often remains clinically silent for many years. Most cases are discovered incidentally or become symptomatic only when the tissue enlarges or causes a local mass effect related to its size or anatomical location (6,7). Enlargement later in life has been attributed to hormonal fluctuations, such as those occurring during puberty, pregnancy, or menopause; compensatory hypertrophy in response to increased thyroid-stimulating hormone (TSH) stimulation; and degenerative or cystic changes within the ectopic tissue itself (6,7). Pathological processes similar to those affecting the orthotopic thyroid gland, including multinodular goiter, cystic degeneration, or neoplastic transformation, may also occur in ectopic thyroid tissue and contribute to delayed clinical presentation (6,8). These mechanisms explain the late onset of symptoms in the presented case, despite the congenital nature of the condition.

Wildi-Runge et al. (5) reported that dual ectopy could be seen in up to 9% of the congenital hypothyroidism cases, suggesting a specific association within this subgroup. The mechanisms underlying dual ectopic thyroid tissue remain incompletely understood. One postulated mechanism involves insufficient signaling gradients during embryonic thyroid migration, leading to the arrest of precursor cells at multiple points. This is further supported by the occurrence of dual ectopy in congenital hypothyroidism, indicating early divergence of thyroid cell populations (5). Alternatively, a polyclonal origin has been proposed, whereby distinct thyroid cell populations migrate independently (2,5). Genetic factors, including mutations in the *Sonic hedgehog* gene, have also been implicated in the pathogenesis of ectopic thyroid development (2).

The diagnosis of ectopic thyroid tissue is often challenging, as it can mimic salivary gland neoplasm, lymphadenopathy, or metastatic deposits given overlapping clinical and radiological features (3). A multimodal diagnostic approach is therefore essential. Ultrasound provides a non-invasive initial assessment for evaluating thyroid morphology; however, its diagnostic accuracy can be limited, as ectopic tissue can exhibit sonographic features similar to other neck masses (9). Fine-needle aspiration cytology is a valuable adjunct but is occasionally inconclusive and cannot reliably exclude malignancy in all cases (4).

A comprehensive diagnostic approach often requires multiple imaging modalities, including ultrasonography, CT, magnetic resonance imaging (MRI), and thyroid scintigraphy. Of these, thyroid scintigraphy is particularly valuable, serving as the cornerstone for identifying ectopic thyroid tissue and distinguishing it from metastatic disease or other neck lesions (10). In the presented case, CT failed to identify the lingual ectopic thyroid, underlining the value of employing a multimodal imaging approach. Routine thyroid function testing including TSH, T3, and T4 levels, is vital not only in assessing thyroid function but also in determining the necessity of lifelong thyroid hormone replacement therapy (4). The diagnostic performance values and imaging characteristics summarized in Table 1 are derived from previously published studies evaluating imaging modalities for ectopic thyroid tissue.

Radionuclide imaging combined with SPECT/CT currently offers the highest reported specificity and sensitivity for detecting ectopic thyroid tissue (10). Although ultrasound is widely accessible and non-invasive, its sensitivity decreases in cases where ectopic tissue is small or exhibits low echogenicity. CT and MRI provide excellent anatomical

Table 1. Summary of diagnostic modalities for ectopic thyroid

Modality	Sensitivity	Specificity	Diagnostic features
Ultrasound (9)	55.0%	100.0%	**Advantages:** Readily available, non-invasive, low cost. **Disadvantages:** Limited resolution for scattered echotexture, echogenicity, or small ectopic tissue. Combination with radionuclide imaging is recommended for comprehensive evaluation.
CT (10)	84.6%	100.0%	**Advantages:** Better at differentiating hard tissues; ectopic thyroid tissue typically has a higher density than the surrounding soft tissues. **Disadvantages:** Limited diagnostic accuracy, potential interference with other procedures, radiation exposure.
MRI (10)	71.4%	100.0%	**Advantages:** Superior for soft tissue assessment. **Disadvantages:** May fail to detect small thyroid tissue due to signal intensity similar to adjacent muscles.
Scintigraphy (10)	92.3%	100.0%	**Advantages:** Highly diagnostic; provides detailed information on size, location, nature, and radioactive iodine uptake of ectopic thyroid tissue. **Disadvantages:** Both benign and malignant ectopic thyroid tissues can show tracer uptake, limiting differentiation.

detail, assisting in the differentiation of ectopic thyroid tissue from other neck masses, though CT involves exposure to ionizing radiation and MRI may struggle to distinguish small foci from the adjacent muscle (3). As both benign and malignant ectopic thyroid tissues may demonstrate tracer uptake, histopathological confirmation is essential in any suspicious case (8).

Management of dual ectopic thyroid tissue must be tailored according to clinical symptoms, risk of malignancy, and thyroid function. Asymptomatic patients with benign cytology and preserved thyroid function may be managed conservatively with regular follow-up, whereas surgical intervention is reserved for those with compressive symptoms, suspicion of malignancy, or functional compromise (9,10).

Conclusion

Dual ectopic thyroid tissue in the lingual and lateral neck regions in the presence of a normally located gland is exceptionally rare. Comprehensive imaging, including radionuclide scintigraphy, and histopathological confirmation are essential for accurate diagnosis and appropriate management. Clinicians should maintain a high index of suspicion for ectopic thyroid in an atypical neck mass to avoid misdiagnosis and guide appropriate management.

Ethics

Informed Consent: Written informed consent was obtained from the patient.

Footnotes

Authorship Contributions

Surgical and Medical Practices: N.E.A.F., K.A.M., M.R.M.Y., Concept: A.H.M.Y., Design: A.H.M.Y., Data Collection and/or Processing: A.H.M.Y., C.J.K., Analysis or Interpretation: N.E.A.F., K.A.M., M.R.M.Y., Literature Search: A.H.M.Y., N.E.A.F., K.A.M., Writing: A.H.M.Y., C.J.K., N.E.A.F., K.A.M., M.R.M.Y.

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Main Points

- Dual ectopic thyroid tissue is a very rare condition, especially in the presence of a normally located thyroid gland.
- Ectopic thyroid tissue should be considered in the differential diagnosis of atypical lateral neck masses.
- Radionuclide imaging, particularly single photon emission computed tomography, is essential for accurate identification of ectopic thyroid tissue.
- A multimodal imaging approach improves diagnostic accuracy and helps avoid unnecessary surgical intervention.
- Management should be individualised based on clinical symptoms, thyroid function, and risk of malignancy.

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