

# World Journal of *Clinical Cases*

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## Multiple ectopic goiter in the retroperitoneum, abdominal wall, liver, and diaphragm: A case report and review of literature

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### Abstract

#### BACKGROUND

Ectopic thyroid is a rare developmental disorder, typically found in lingual areas, and most distantly in the abdomen. Thyroid ectopia in multiple regions is extremely rare. To date, there are no reports of ectopic goiter in four regions of the abdominal cavity in a single patient.

#### CASE SUMMARY

We present a case of thyroid ectopia in four areas of the abdomen, comprising normal orthotopic thyroid tissue. A 36-year-old woman, who underwent ovarian teratoma resection 10 years previously due to symptomatic ovarian teratoma, was referred to our hospital for treatment of an incidental hepatic mass. Magnetic resonance imaging and computed tomography (CT) of the abdomen showed a heterogeneously enhanced lobulated lesion in the sixth and seventh hepatic segment adjacent to the diaphragm. The mass was surgically excised, and histologic examination determined an ectopic nodular goiter. At the one-year follow-up, the abdominal CT scan, whole-body radionuclide I131 examination, and abdominal wall biopsy showed similar lesions in the left renal fascia and anterior abdominal wall.

#### CONCLUSION

Multiple para-ectopic thyroid is often misdiagnosed, owing to its extremely rare incidence and non-specific clinical manifestations and imaging features. A combination of multiple examinations is necessary for diagnostic accuracy.

**Key Words:** Ectopic thyroid; Magnetic resonance imaging; Computed tomography; <sup>99m</sup>Tc sodium pertechnetate scintigraphy; Case report

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**Core Tip:** Ectopic thyroid glands in the abdomen are extremely rare, and the occurrence of ectopic thyroid tissues in multiple regions simultaneously is even rarer. However, it is best to diagnose ectopic thyroid, its function and location, before surgery. Additionally, abdominal metastasis due to thyroid cancer and ectopic thyroid cancer must be differentiated from benign lesions.

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## INTRODUCTION

Ectopic thyroid is a rare developmental disease with an incidence of only one in 300000<sup>[1]</sup>. The rate of incidence is higher in females (75%-80%)<sup>[2]</sup>. Thyroid ectopic tissue is most often located in the neck and head, accounting for 90% of all ectopic thyroids<sup>[2]</sup>. Ectopic thyroid glands in the abdomen are extremely rare, and the occurrence of ectopic thyroid tissues in multiple regions simultaneously is even rarer<sup>[3,4]</sup>. There has been only one report of ectopic thyroid tissues located in multiple areas of the abdomen<sup>[5]</sup>. To our knowledge, based on the current literature review, the report presented herein is the first of multifocal ectopic goiter located in four regions of the abdomen, specifically the liver, diaphragm, left retroperitoneum, and frontal abdominal wall, and coexisting with the normal positioning of multinodular goiter.

## CASE PRESENTATION

### Chief complaints

A 36-year-old woman, who had undergone ovarian teratoma resection 10 years previously due to symptomatic ovarian teratoma, was referred to our hospital for treatment of an incidental hepatic mass.

### History of present illness

She presented with occasional dull pain below the xiphoid.

### History of past illness

She had undergone ovarian teratoma resection 10 years previously.

### Personal and family history

No special personal and family history.

### Physical examination

Physical examination showed no abnormalities.

### Laboratory examinations

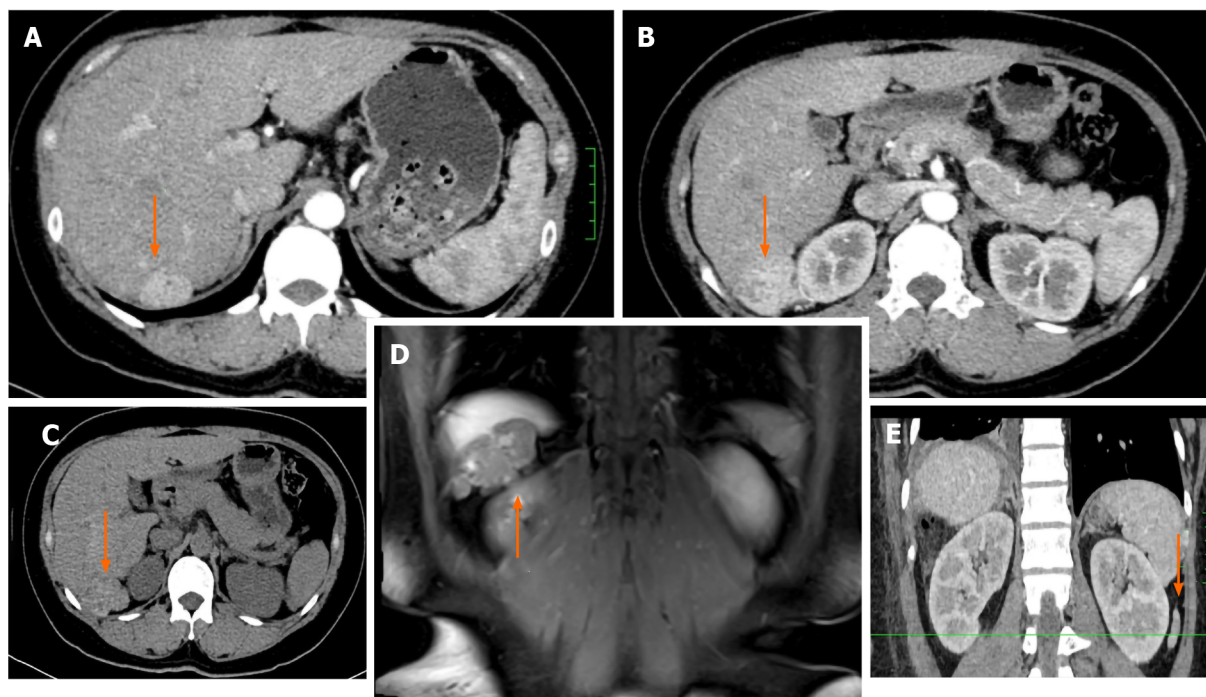
The liver function test and all tumor markers were negative.

### Imaging examinations

Abdominal computed tomography (CT) showed a 1.2 cm × 2.9 cm × 2.8 cm (transverse × anteroposterior × craniocaudal) lobulated lesion in the diaphragm (Figure 1A), and a 4.2 cm × 3.9 cm × 3.9 cm solid lesion in the sixth hepatic segment (Figure 1B), with a 60-70 Hounsfield unit (HU) value (Figure 1C).

On post-contrast CT, the lesion revealed a heterogeneous enhancement pattern (Figure 1A and B). The lesions showed equal signal intensity on T2-weighted imaging and slightly low signals on T1-weighted imaging (Figure 1D). The magnetic resonance





**Figure 1 Computed tomography and magnetic resonance imaging of the abdomen.** Computed tomography (CT) axial sections and sagittal section show three ectopic hyperdense lesions located in the liver, diaphragm, and retroperitoneum, respectively. Magnetic resonance imaging (MRI) coronal section shows thyroid ectopia in the liver and diaphragm. A-C: CT axial sections (orange arrow); D: MRI coronal section (orange arrow); E: CT sagittal section (orange arrow).

imaging (MRI) enhancement pattern was similar to the CT pattern, and diffusion-weighted imaging showed a slightly higher signal ( $b$  value = 800). The lesions showed no restricted diffusion, and no contrast agent uptake on the hepatobiliary specific phase, displaying low signal.

## FINAL DIAGNOSIS

Based on these examinations, we clinically diagnosed a benign adenoma with hemorrhage.

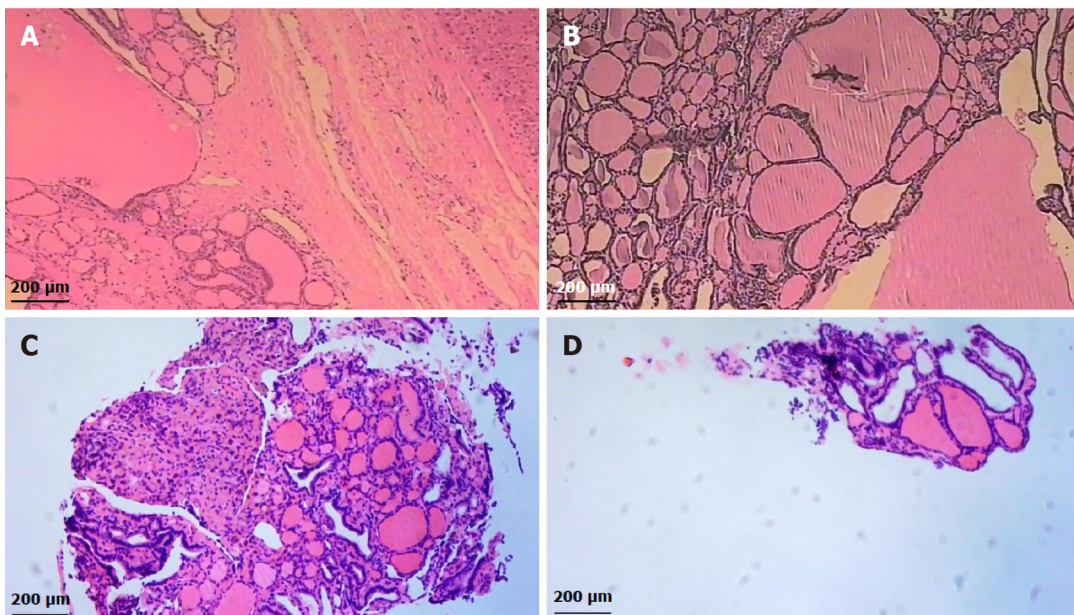
## TREATMENT

Surgery was performed, which revealed a 6.5 cm × 5.5 cm × 4.5 cm mass in the right posterior lobe of the liver, and a 2.5 cm × 2.0 cm × 1.5 cm mass in the right diaphragm adjacent to the sixth hepatic segment, both with an incomplete capsule. The gross specimens were grayish white and gelatinous.

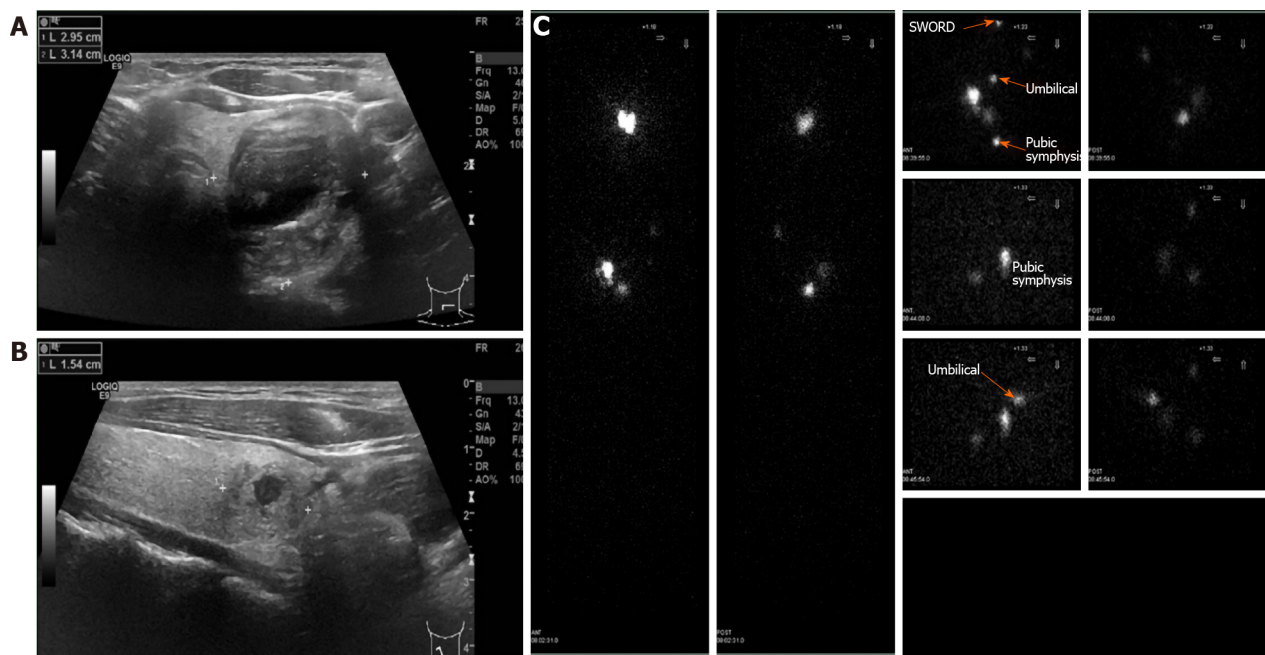
## OUTCOME AND FOLLOW-UP

Histological examination revealed ectopic goiters in the right diaphragm and in the right lobe of the liver (Figure 2A and B). Immunohistochemistry staining revealed the strong presence of thyroid transcription factor-1 (TTF-1), thyroglobulin, thyroperoxidase (TPO), CK19, and CK7; while galectin-3 and CD56 were negative. The postoperative thyroid function test was normal. Thyroid and cervical lymph nodular ultrasonography indicated nodular goiter (Figure 3A), confirmed by simultaneous ultrasound-guided puncture, with no malignant cells, and without thyroid cancer metastasis.

At the one-year follow-up, abdominal CT showed a 2.4 cm × 0.9 cm ovoid slightly hyperintense lesion (84 HU) adjacent to the left renal fascia (Figure 1E). To confirm further thyroid ectopia in the left retroperitoneum, we recommended that the patient undergo  $^{99m}\text{Tc}$  sodium pertechnetate scintigraphy. To our surprise, this showed a



**Figure 2 Hematoxylin and eosin stain results.** The histopathological findings confirmed ectopic goiter. A: Liver; B: Diaphragm; C: Left renal fascia; D: Anterior abdominal wall. Bar, 200 µm.



**Figure 3 Ultrasound and  $^{99m}\text{Tc}$  sodium pertechnetate scintigraphy of the thyroid.** A-B: Ultrasound of the thyroid showed nodular goiters in both normal lateral lobes; C:  $^{99m}\text{Tc}$  sodium pertechnetate scintigraphy showed three abnormal foci in the left renal and lower abdomen.

patchy abnormal concentration equivalent to the area of the left kidney, as well as two patchy abnormal concentrations in the right lower abdominal wall (Figure 3B). Considering the possibility of ectopic thyroid in the right lower abdominal wall of the left kidney, an ultrasound-guided puncture provided proof (Figure 2C and D).

## DISCUSSION

The mechanism of abdominal ectopic thyroid formation remains controversial, but several theories have been proposed. The most common theory to explain the presence of ectopic thyroid tissue is excessive descent of thyroid precursor cells outside the usual path of thyroid anlage<sup>[6]</sup>, or abnormal migration of precursor cells<sup>[7]</sup>. However, it

is difficult to prove this due to the rarity of abdominal ectopic thyroid, and the distance of the path of embryonic development.

Secondly, there is a theory that the precursor cells of the foregut endoderm differentiate into thyroid ectopia<sup>[8,9]</sup> because most abdominal organs, as well as the thyroid, originate in the endoderm. However, it is difficult to explicate why the adrenal gland is located in the retroperitoneum, since it originates from mesoderm, while the cortex and medulla derive from the epiblast<sup>[10]</sup>.

A third theory is that germ cell neoplasms can differentiate into mature teratoma-type thyroid tissue<sup>[11]</sup>. Tamaki *et al*<sup>[12]</sup> confirmed that, after the first division of meiosis, a single germ cell could generate into ectopic thyroid tissue and other tissue components (*e.g.*, skin appendages and bone). However, peritoneal strumosis that originated from ovarian teratoma may develop into malignant papillary thyroid cancer<sup>[5,13,14]</sup>. In addition, there can be complications due to thyroid tissue implantation during surgery<sup>[15,16]</sup>. This theory may clarify why goiters in the present case were located in the anterior abdominal wall.

Ectopic thyroid gland tissue may be classified as vagus thyroid or parathyroid; the former has no normal orthotopic thyroid gland tissue, while the latter coexists with normal orthotopic thyroid gland tissue. We reviewed the current literature in the electronic databases Medline and Embase, and found 15 cases of multiple-site ectopic thyroid (Table 1)<sup>[3,4,17-24]</sup>. Most cases with three sites were located in the neck, while those in four or more regions were apt to locate in post-operative regions or previous ovarian teratoma.

According to the review literature, ectopic thyroid is mostly asymptomatic<sup>[2]</sup>. However, non-specific symptoms may also occur depending on the anatomical location, such as pain, difficulty breathing, diarrhea, bleeding, and other complications. In the present case, the patient experienced an occasional dull pain below the xiphoid, because the ectopic thyroid was located at the sixth and seventh hepatic segment adjacent to the diaphragm. Liang *et al*<sup>[25]</sup> reported a 60-year-old woman with recurrent pain in her right upper abdomen due to a gallbladder mass, which proved after surgical resection to be ectopic thyroid tissue. In another case, a 60-year-old woman underwent routine appendectomy due to sudden right lower abdominal pain; the histological examination revealed abnormal ectopic thyroid tissue in the fat around the appendix<sup>[11]</sup>. In the present case, thyroid tissue was found in the orthotopic position of the ectopic thyroid, and the thyroid function test was normal.

In our patient, the various imaging examinations were inconclusive and could not provide a confident diagnosis; only postoperative pathologic examination confirmed ectopic thyroid. Due to the rarity of ectopic thyroid tissue in the abdominal cavity and the limited experience with diagnostic imaging in these cases, the location and function of ectopic thyroid are usually determined postoperatively or after biopsy<sup>[8]</sup>.

However, it is best to diagnose ectopic thyroid, its function and location, before surgery. Abdominal metastasis due to thyroid cancer and ectopic thyroid cancer must be differentiated from benign lesions. Distant metastases of papillary carcinoma of the thyroid have been found in the pancreas<sup>[26]</sup>, liver<sup>[27]</sup>, kidney<sup>[28]</sup>, adrenal gland<sup>[29]</sup>, ovaries<sup>[30]</sup>, and other sites. Ectopic thyroid may also be cancerous. The canceration rate of lingual thyroid is about 1%<sup>[31]</sup>.

For differential diagnosis, there are a variety of examination methods: thyroid function test, and non-invasive imaging such as ultrasound, CT, MRI, whole-body thyroid radionuclide scan, and single-photon emission computed tomography (SPECT)/CT. Invasive examinations include puncture biopsy guided by ultrasound or CT, but these are not preferred.

Imaging methods can detect the extent and location of ectopic thyroid, and ultrasound is the cheapest and most easily available<sup>[2]</sup>. Ultrasound can distinguish benign lesions from malignant thyroid nodules<sup>[32]</sup>. However, ultrasound cannot distinguish ectopic thyroid gland from orthotopic thyroid gland, which is easy to misdiagnose.

When abdominal CT imaging depicts abdominal masses with higher density than that of surrounding soft tissue, with a clear boundary, then ectopic thyroid tissue should be considered<sup>[33]</sup>. On CT imaging, ectopic thyroid tissue, like normal thyroid tissue, contains a certain amount of iodine, and its attenuation relative to the adjacent skeletal muscle increases ( $70 \text{ HU} \pm 10 \text{ HU}$ ). After contrast agent is injected, most tissues show heterogeneous enhancement<sup>[8,22,34]</sup>. Moreover, ectopic goiter is generally found with calcification<sup>[35]</sup>. In the present case, non-contrast CT imaging displayed hepatic lesion hyperintensity ( $60\text{-}70 \text{ HU}$ ) that was higher than that of the surrounding normal liver tissue; contrast CT imaging showed heterogeneous enhancement.

On MRI, the ectopic thyroid has equal intensity or slight hyperintensity in T1-weighted and T2-weighted images (T1WI and T2WI), and the enhancement pattern is



Table 1 Case reports of multiple ectopic thyroid

Ref.	Age, yr	Gender	Location <sup>1</sup>	Endocrine	Ectopic location	Diagnosis
Agrawal <i>et al</i> <sup>[3]</sup> , 2019	11	Female	Not normal	Normal	Infrahyoid region, the base of the tongue and the other in the prehyoid location	TFT, US, Tc-99m TcO <sub>4</sub> scan, SPECT/CT, FNA
Barai <i>et al</i> , 2004	6	Male	Not normal	Hyperthyroidism	Region of the foramen cecum, suprahyoid and in the region of the cricoid cartilage	TFT, 131I WBS, pathology
Belgur <i>et al</i> <sup>[22]</sup> , 2016	25	Male	Not normal	NA	Tongue (posteriorly), the sublingual, the prehyoid and the right submandibular regions	US, CT, MRI, FNA
Dobrescu <i>et al</i> <sup>[14]</sup> , 2014	48	Female	Normal	Hyperthyroidism	Pulmonary areas, right pelvis, axial skeleton, pelvic bones, skull	US, CT, 131I WBS, TFT, pathology
Hua <i>et al</i> <sup>[24]</sup> , 2019	56	Female	NA	SH	Tongue base, neck region over the submental and hyoid bone level	131I WBS, FNA, CT
Jain <i>et al</i> <sup>[23]</sup> , 2010	20	Female	Normal	SH	Base of the tongue, the suprahyoid and the subhyoid	TFT, 131I WBS, CT
Kim <i>et al</i> <sup>[16]</sup> , 2015	35	Female	Normal	Hyperthyroidism	Right chest wall and axilla, the left chest wall	131I WBS, SPECT/CT
Konde <i>et al</i> <sup>[17]</sup> , 2012	16	Female	Not normal	Normal	Lingual foramen cecum level, sublingual (between the geniohyoid and mylohyoid muscles), below the hyoid bone level	CT, US, TFT, Tc-99m TcO <sub>4</sub> scan
Kuramoto <i>et al</i> <sup>[19]</sup> , 2013	10	Female	NA	NA	Near the base of the tongue, prehyoid and pretracheal region	NA
Nilegaonkar <i>et al</i> <sup>[21]</sup> , 2011	16	Female	NA	Hypothyroidism	Base of tongue, the level of the hyoid and suprahyoid location	US, CT, Tc-99m TcO <sub>4</sub> scan, FNAC, TFT
Passah <i>et al</i> <sup>[4]</sup> , 2018	5	Female	Not normal	SH	Base of the tongue, prehyoid region, anterior to the thyroid cartilage towards the right side	TFT, Tc-99m TcO <sub>4</sub> scan, SPECT/CT
Rahalkar <i>et al</i> <sup>[20]</sup> , 2014	42	Female	Not normal	Normal	Surface of posterior tongue, anterosuperior, close to the body of the hyoid bone and in the subhyoid region	CT, Tc-99m TcO <sub>4</sub> scan
Spencer-Segal <i>et al</i> , 2015	37	Female	Normal	Normal	Right sternocleidomastoid, periclavicular, pectoralis and axillary regions	TFT, I-123 SPECT-CT, CT, pathology
Thomas <i>et al</i> <sup>[5]</sup> , 2013	14	Female	Normal	Normal	Omentum, left adnexa, peritoneum, spleen, abdominal wall, bladder and rectum	TFT, CT, pathology
Weng <i>et al</i> , 2000	86	Female	Normal	Hyperthyroidism	Lung, visceral pleura, pelvic cavity and the skeleton	TFT, chest X-ray, US, pathology

<sup>1</sup>Location of the thyroid. 131I WBS: Whole-body 131 I scintigraphy; CT: Computed tomography; FNA: Fine needle aspiration; FNAC: Fine needle aspiration cytology; NA: Not available; SH: Subclinical hypothyroidism; SPECT: Single-photon emission computed tomography; Tc-99m TcO<sub>4</sub> scan: Radionuclide 3 mCi technetium scan; TFT: Thyroid function test; US: Ultrasonography.

the same as CT imaging<sup>[22,36]</sup>. To evaluate the function and anatomy of the suspected ectopic thyroid tissue, especially thyroid tissue that is distant from the thyroid tongue tube, radionuclide and SPECT/CT imaging can show the uptake of radionuclide (99 TcmO<sub>4</sub> or 131I) by the ectopic thyroid and positivity<sup>[3,37,38]</sup>. Therefore, for suspected multiple abdominal ectopic thyroid, multiple imaging methods are required.

Radiologically, ectopic thyroid in our case, associated with multiple sites and lesions in the abdomen, was difficult to differentiate from hematological diseases such as lymphomas or leukemia. Additionally, multiple enlarged lymph nodes in the abdomen were typical manifestations of lymphomas or leukemia, which were limited to multiple round or oval soft tissue in certain areas at the preliminary stage. With regard to disease progression, it manifested in the retroperitoneal space, mesentery, superficial lymph node enlargement, and even involved the liver and spleen<sup>[39]</sup>, as well as pelvic organs, such as the uterus<sup>[40]</sup>. Lymphomas or leukemia are usually seen as homogeneous low-density lesions on CT imaging, with mild to moderate enhancement. However, ectopic thyroid showed high density on CT imaging. In addition, the density of lymph nodes with necrosis may be heterogeneous. In lymphomas, MRI features show low signal intensity of relative fat and slightly high signal intensity of muscle on T1WI. On T2WI, the signal intensity of relative fat is

equal or slightly lower, and that of muscle is high signal intensity. DWI showed characteristic homogeneous high signal intensity<sup>[41]</sup>. However, ectopic thyroid had equal intensity or slight hyperintensity in T1WI and T2WI.

## CONCLUSION

In conclusion, multiple para-ectopic thyroid is often misdiagnosed, owing to its extremely rare incidence and non-specific clinical manifestations and imaging features. A combination of multiple examinations is necessary for diagnostic accuracy. When thyroid tissue is found in an abnormal location, ectopic thyroid should be considered, especially if there is a previous history of ovarian teratoma. First, an ultrasound examination should determine whether there is normal thyroid tissue. This is followed in turn with a thyroid function test, CT or MRI (especially <sup>99m</sup>Tc sodium pertechnetate scintigraphy), and SPECT/CT. Finally, a biopsy and even pathological examination after surgical resection can provide confirmation. For the patient with a previous thyroidectomy, a follow-up that includes <sup>99m</sup>Tc sodium pertechnetate scintigraphy, or SPECT/CT examination, is essential to check for implantation.

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