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Case Report

# **Intrapericardial Ectopic Goiter: A Very Unusual Presentation**

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Mediastinal ectopic goiter is a thyroid tumor that lies entirely below a plane extending from the superior surface of the first thoracic vertebra to the suprasternal notch, and commonly lies in the vicinity of the thymus. Intrapericardial ectopic goiter is extremely rare. We present an extremely rare case of a 63-year-old woman with an intrapericardial ectopic goiter and review the pertinent literature.

Keywords: intrapericardial tumor, ectopic goiter, thyroid tissue, coronary artery

## Introduction

The thyroid gland develops from the floor of the pharynx between the first pharyngeal pouches. Ectopic thyroid tissue is thought to occur if the gland develops incorrectly. While ectopic thyroid tissue has been found in many different midline locations, the most frequent location is the lingual area (90%).<sup>1)</sup> Although intrathoracic ectopic goiters, such as in the mediastinum, lungs, and heart, have been reported sporadically, there are few cases of intrapericardial ectopic goiter in the English literature.

We report here an extremely rare case of intrapericardial ectopic goiter, and also discuss the relevant literature.

### **Case Report**

A 63-year-old woman presented with a chest shadow on a routine chest roentgenogram in August 2017. She was

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under medical treatment for hypertension, diabetes mellitus, cholelithiasis, and spinal canal stenosis. Her family history was unremarkable. She had never smoked. The physical findings were all within normal limits. Blood tests found that the thyroid stimulating hormone (TSH) level was 0.01 µIU/mL, while free triiodothyronine (FT3) was 9.93 pmol/mL, and free thyroxine (FT4) was 5.14 pmol/mL (normal range: TSH, 0.34-3.8 µIU/mL; FT3, 2.0-3.8 pmol/mL; FT4, 0.8-1.5 pmol/mL). Chest computed tomography (CT) revealed a nodular intrapericardial lesion in the middle mediastinum, outside the shadow on routine chest roentgenogram (Fig. 1). The lesion showed a clear boundary and heterogeneous enhancement on CT after the intravenous administration of contrast material. Coronary CT angiography showed that the lesion was not connected to a coronary artery. On positron-emission tomography (PET), which was obtained 60 minutes after the injection of <sup>18</sup>F-fluorodeoxyglucose (<sup>18</sup>F-FDG), the lesion showed slight <sup>18</sup>F-FDG accumulation with a maximum standardized uptake value (SUVmax) of 1.30. Thus, we performed surgery with a transsternal approach to confirm the diagnosis of the lesion. There was no abnormality on the outside of the pericardium. On opening the pericardium from the foot side of the lesion seen through the pericardium, the nodular lesion was  $25 \times 20 \times 15$  mm in size, and was located on the anterior surface of the proximal ascending aorta. The lesion projected into the pericardial space in a pedunculated manner from the serous pericardium covering the ascending aorta (Fig. 2A). After ligation and detachment of the root of the pedicle, we removed it en bloc.



Fig. 1 Chest CT scan shows a well-defined nodular lesion (arrow) located in the intrapericardial space. The lesion shows heterogeneous enhancement on CT after the intravenous administration of contrast material.

The resected specimen was a firm nodule wrapped in a fibrous capsule. The cut surface was yellowish brown in color, and showed scattered coarse calcifications and some small cysts (**Fig. 2B**). Histologic examination of the lesion showed encapsulated thyroid tissue, and that of the stalk connecting with the tumor showed abnormal vessels and collagen fibers (**Fig. 3**). The postoperative clinical course was uneventful. The FT3, FT4, and TSH levels after surgery returned to the normal range. At 1-year follow-up after the operation, she showed continuously good status without recurrence or de novo appearance of thyroid tissue.

### **Discussion and Conclusion**

Ectopic thyroid tissue is thought to occur as a result of developmental defects in the early stages of thyroid gland embryogenesis, during its passage from the floor of the primitive foregut to its final pre-tracheal position.<sup>1)</sup> Ectopic thyroid tissue is reportedly more common among women, and is usually located in the lingual area. The prevalence of ectopic thyroid tissue is about 1 per 100,000–300,000 people, and mediastinal ectopic goiter accounts for only 1%–5% of all ectopic thyroid tissues.<sup>1)</sup>

To date, six cases of intrapericardial ectopic goiter, including the present case, have been reported since it was



Fig. 2 (A) After a transsternal approach, on opening the pericardium, the nodular lesion was located on the anterior surface of the proximal ascending aorta. The lesion projected into the pericardial space in a pedunculated manner from the serous pericardium covering the ascending aorta.
(B) The cut surface of the resected specimen was yellowish brown in color, and showed scattered coarse calcifications and some small cysts.

first described by Taylor in 1986.<sup>2–5)</sup> In these six cases, the median age (range: 42–69 years) was 49 years. All lesions were found to be close to the ascending aorta, and were often found by chance. Among the six cases, a patient with a lesion 5.5 cm in diameter had symptoms of chest pain and palpitations.<sup>3)</sup> Another patient with a lesion adjacent to a coronary artery had symptoms of dyspnea and angina.<sup>5)</sup> The other patients with intrapericardial ectopic goiters were asymptomatic, and their tumors were smaller. Thyroid function is often normal, but was slightly enhanced in our case without hyperthyroidism symptoms. In two cases, the lesion was connected to a coronary artery.<sup>4,5)</sup> In one of these two cases, the intrapericardial ectopic goiter was resected after starting partial



Fig. 3 Photomicrograph shows thyroid tissue in the lesion (hematoxylin & eosin, ×400 original magnification).

cardiopulmonary bypass.<sup>5)</sup> In previously reported cases of intrapericardial ectopic goiter, surgical resection was performed for both a firm diagnosis and treatment, as in our present case. Surgery was performed using a transsternal approach, and cardiopulmonary bypass might be required. Therefore, preoperative coronary CT or angiography should be performed for surgery of an intrapericardial tumor.

Mediastinal ectopic goiter is often revealed incidentally on chest radiograph or at autopsy. In CT, ectopic goiter is a well-defined lesion accompanied by calcifications with higher density than the surrounding soft tissues, except if the iodine content is low. After the intravenous administration of contrast material, ectopic goiter usually shows enhancement on CT.<sup>1,6)</sup> In MRI, ectopic goiter appears as a mass/nodule with higher signal intensity than that of the surrounding tissue in both T1- and T2-weighed images.<sup>1)</sup> Although ectopic goiter usually shows little <sup>18</sup>F-FDG accumulation on PET, it has also been reported that mediastinal ectopic goiter showed diffuse strong <sup>18</sup>F-FDG accumulation. <sup>18</sup>F-FDG accumulation in thyroid tissue can be seen in various benign and malignant conditions, including thyroid carcinoma, metastatic tumor, thyroiditis, multinodular goiter, and nodular hyperplasia.<sup>7)</sup>

Intrapericardial tumors are extremely rare among mediastinal tumors. According to the Atlas of Tumor Pathology, the most frequent intrapericardial tumor at the Armed Forces Institute of Pathology (1976–1993) was mesothelial cyst (58%), followed by malignant mesothelioma (13%) and sarcoma (13%). Excluding cystic lesions, malignant tumors (28%) were more common than benign tumors (13%) in adults.<sup>8)</sup>

In conclusion, while ectopic goiter can arise in the intrapericardial space, this location is quite rare. Intrapericardial tumors can adversely affect cardiac function, even if they are benign, and therefore they need to be resected. Although it is difficult to diagnose an intrapericardial tumor as an ectopic goiter without surgery, thyroid hormone levels might help with the diagnosis. Since we examined the relationship between the tumor and a coronary artery by coronary CT angiography, we were able to more safely treat intrapericardial ectopic goiter. We must pay close attention to the relation between an intrapericardial tumor and a coronary artery at the time of surgery.

## **Disclosure Statement**

The authors declare no conflicts of interest associated with this manuscript.

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