An intrapericardial ectopic thyroid mimicking metastasis in a patient with papillary thyroid cancer: Localization, differential diagnosis by ^{18}F-FDG PET/CT and ablation by ^{131}I

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Abstract
We report a very rare case of incidental intrapericardial thyroid in a papillary thyroid cancer patient. Post ablation scan revealed iodine-131 (^{131}I) uptake in the mid-chest. Fluorine-18-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT) was performed and showed a ^{18}F-FDG avid lesion between the right atrium and the ascending aorta, which was shown to be an ectopic thyroid and not metastasis. The lesion disappeared on a 6 month follow-up ^{123}I whole body scan while serum thyroglobulin was negative. Although intrapericardial ectopic thyroid is reported to show high iodine uptake, low ^{18}F-FDG avidity of the lesion could be helpful in the exclusion of metastases.

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Introduction

Ectopic thyroid refers to thyroid tissue located outside the thyroid bed, between the second and fourth tracheal cartilages. Its incidence is 1 per 300,000 to 400,000 of the population and is reportedly more common amongst women [1]. Ectopic thyroid usually presents in the lingual area (90%) and is rarer in the more caudal locations. Although rare, ectopic thyroid tissue can be found in the esophagus, lung, adrenal gland, pancreas, gallbladder, bladder, ovary (struma ovarii) and even in the heart (struma cordis) [2]. The cause of these distant ectopic thyroid tissue foci is reported to be related to aberrant migration or heterotopic differentiation of uncommitted endodermal cells [3]. Although there are 35 case reports discussing intracardiac ectopic thyroid (struma cordis) [1, 4, 5], there are only 3 cases of intrapericardial ectopic thyroid [6-8].

We report a case of intrapericardial ectopic thyroid that was incidentally detected after radioactive iodine therapy (RAI) in a thyroid cancer patient. Intrapericardial thyroid is very rare and this is the first case that was ablated by ^{131}I treatment.

Case Report

A 57 years old woman underwent ultrasonography which detected a focal low echogenic nodule in the right lower pole of the thyroid gland. After fine needle aspiration, the nodule was pathologically diagnosed as papillary thyroid cancer. The patient had no clinical symptoms and her physical examination and laboratory tests were normal. The levels of thyrotropin (TSH), thyroglobulin (Tg), free thyroxine (T4), and anti-thyroglobulin (anti-Tg) antibodies were 1.1μIU/mL (normal range 0.6-4.9), 34.5ng/mL (3.5-77.0), 1.18ng/dL (0.8-1.7), and <20.0IU/mL (0.0-40.0), respectively. A total thyroidectomy with central neck dissection was performed. The resulting specimen was papillary thyroid cancer, measuring 0.7x0.7x0.5cm. There was extension of the cancer into the neighboring soft tissues but the resection margins were clear, and no lymphovascular or lymph node involvement was detected. The patient was assessed to have stage III (according to American Joint Committee on Cancer) and intermediate risk (according to American Thyroid Association risk classification system) disease.

Therapy with 1110MBq of ^{131}I was performed after supplementary thyroid hormone withdrawal and a low iodine diet for 2 weeks. At the time of radioactive iodine (RAI) therapy, the levels of TSH, Tg, free T4, and anti-Tg antibodies were 46.4μIU/mL, 0.41ng/mL, 0.21ng/dL, and <20.0IU/mL, respectively. A whole body scan acquired three days after RAI treatment administration revealed focal iodine uptake in the neck from the thyroid remnant and an additional intense star-shaped iodine uptake in the mid-chest, suggesting metastasis (Figure 1). At this time, the patient complained of persistent epigastric discomfort after RAI administration. Fluorine-18-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT) was performed to further characterize the unexpected mid-mediastinal uptake. A 2cm sized soft tissue lesion with low ^{18}F-FDG avidity (SUVmax: 2.4) was detected proximal to ascending aorta (Figure 2). The lesion was thought to be an ectopic thyroid instead of metastasis. On follow-up, enhanced CT and diagnostic ^{123}I whole body scan 6 months later, the intrapericardial soft tissue lesion disappeared (Figure 3). The patient reached successful ablation (negative whole body ^{131}I scan and negative serum Tg after TSH stimulation). The patient had no cardiac symptoms.

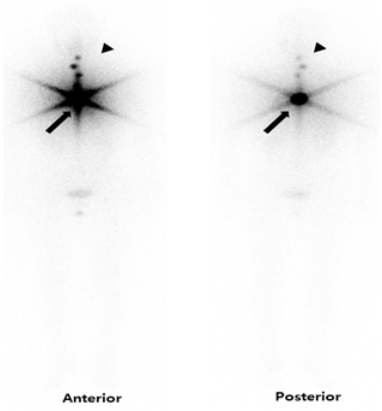

Figure 1. Iodine-131 post-ablation whole body scan shows intense ^{131}I uptake with a star shaped artifact in the mediastinum (black arrow) and three foci of iodine-131 uptake in the neck, suggesting remnant thyroid tissue in the thyroid bed (black arrowhead).

Discussion

Ectopic thyroid in the heart was first reported by Dosch in 1941 and is a rare condition with only 35 reported cases so far [1, 4, 5]. Most are found on the right side of the interventricular septum and spread to the right ventricular outflow tract [1]. Intrapericardial ectopic thyroid is even rarer, with only 3 reports worldwide. All were found to be close to the ascending aorta [6-8].

The thyroid gland develops from the ventral midline of the pharynx between the first pharyngeal pouches and descends caudally through the second branchial arch. Thyroid ectopy may be because of the persistent contact of the thyroid primordium with the anterolateral part of the primitive heart infundibulum (bulbus cordis). It can be caused by the occurrence of thyroid rudiments dragged into the chest during the descent of the heart and great vessels in early stages of organogenesis. Thus, intrapericardial thyroid tissue on the ascending aorta is the result of over-descending [8, 9]. Ectopic thyroid in the heart is more common in women and its clinical manifestations peak is at 40 to 50 years of age [1]. Because the clinical symptoms are usually nonspecific, ectopic thyroid may be detected incidental during evaluation of related conditions, such as arrhythmias, right ventricular obstruction, or pulmonary embolism [10]. The differential diagnosis includes thrombi, benign tumors (such as teratoma, myxoma, lipoma, papillary fibroelastoma, or rhabdomyoma), and primary/secondary malignant tumors.

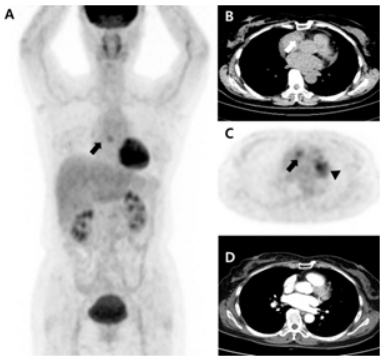

Figure 2. A) Maximum intensity projection image of ^{18}F-FDG PET/CT shows a focal ^{18}F-FDG uptake in mid-chest (black arrow). B, C) Axial ^{18}F-FDG PET/CT shows a nodular soft tissue lesion with mildly increased ^{18}F-FDG uptake adjacent to the right proximal aorta (white and black arrows). Focal ^{18}F-FDG uptake at left side mediastinum is physiologic uptake of left cardiac ventricle (black arrowhead). D) At the 6 months follow-up enhanced chest CT, the nodular lesion is no longer visible.

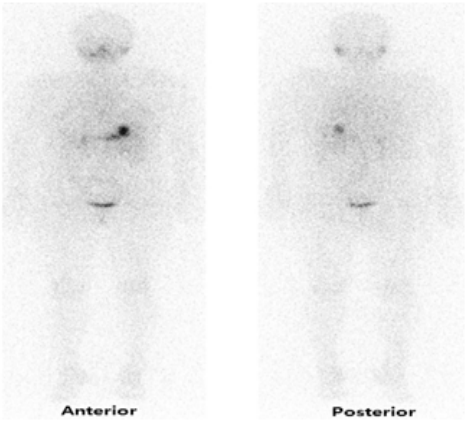

Figure 3. The intense ^{131}I uptake in the mediastinum and remnant thyroid tissue has resolved as showed by the 6 month follow-up ^{123}I diagnostic whole body scan.

Radionuclide thyroid images using ^{99m}Tc pertechnetate, ^{131}I, or ^{123}I are widely used for the diagnosis and localization of eutopic thyroid gland [11, 12]. When imaging for ectopic thyroid, the use of ^{131}I is discouraged because of its high radiation dose. Although pertechnetate is more readily available, ^{123}I demonstrates better images when uptake is low or there is need for visualization of retrosternal tissue. Selection between the two radiopharmaceuticals should be ac-

cording to clinical circumstances. In one case report, intrathoracic ectopic thyroid showed high ^{18}F-FDG avidity (SUVmax 8.0) and metastasis was included in the differential diagnosis [13]. Unlike that case, the lesion in our case did not show pathological ^{18}F-FDG uptake according to the low SUVmax 2.4. To our knowledge, there has not been a report describing the usefulness of the ^{18}F-FDG PET/CT for evaluating intrapericardial thyroid. If a focal ^{131}I uptake is detected outside the thyroid bed in a post-ablation whole body scan, metastasis from thyroid cancer must be excluded. The focal lesion showing intense iodine uptake in our case showed relatively low ^{18}F-FDG avidity and was eventually diagnosed as ectopic thyroid. Because malignant lesions such as metastasis usually show high ^{18}F-FDG uptake, low ^{18}F-FDG avidity of incidentally detected intrapericardial lesions may be useful in excluding metastases and preventing unnecessary medical and/or surgical treatment [4].

Asymptomatic ectopic thyroid usually deserves no treatment, but surgical removal is needed if there are symptoms (bleeding, ulceration) or malignancy [4]. In intrapericardial ectopic thyroid surgical excision is generally performed for differential diagnosis [8], of malignant potential of the ectopic thyroid tissue [14] or for cardiac problems such as adjacent vessel/chamber compression [6, 8]. Thyroid function must be monitored for potential postoperative hypothyroidism. In our case, the intrapericardial ectopic thyroid was successfully removed by conservative RAI treatment.

Whole body ^{131}I scan, besides remnant thyroid tissue or metastases may also detect ectopic thyroid tissue, ectopic gastric mucosa, inflammation, infection, and non-thyroidal neoplasm [15]. The fact that in our case there was no clinical evidence nor laboratory tests of inflammation and the ^{131}I uptake was very intense (star artifact) suggested that the lesion was of thyroid tissue origin (remnant tissue, metastasis, or ectopic thyroid). Although right ventricular metastasis obstructing the outflow tract has been reported in highly differentiated follicular thyroid carcinoma [16], the fact that our patient had papillary cancer in a low initial cancer stage (T3N0), with normal postoperative serum Tg, and negative follow-up ^{123}I whole body scan suggested that the intrapericardial soft tissue lesion with low ^{18}F-FDG uptake was more likely an ectopic thyroid than metastasis.

Treatment using RAI, to the best of our knowledge, has not been reported in either intracardiac or intrapericardial ectopic thyroid. Although in our case RAI was performed for the ablation of remnant tissue in the thyroid bed, it was successful for the removal of both tissues. Low dose RAI therapy may be sufficient for a small intrapericardial or other mediastinal ectopic thyroid if eutopic thyroid is absent or has been removed.

In conclusion, we report a case of intrapericardial ectopic thyroid incidentally detected in a patient with papillary thyroid cancer. The patient had a mediastinal lesion with high ^{131}I uptake on the whole body ^{131}I scan and ^{18}F-FDG PET/CT was helpful in excluding metastases because of its relatively low ^{18}F-FDG avidity. The lesion was totally ablated by ^{131}I as shown by the ^{123}I scan.

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