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Case Report

A Rare Case of 18F-FDG Uptake in an Ectopic Thyroid Carcinoma of the Anterior Middle Neck Lacking Thyroglossal Duct Remnants

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Keywords
Ectopic thyroid carcinoma · ^{18}F-FDG PET/CT · Papillary thyroid carcinoma

Abstract
An 80-year old female presented with a well-defined tumor of the anterior middle neck, and a diagnosis of thyroglossal duct cyst was made. When the tumor size increased, malignancy was suspected. Ultrasonography revealed a smooth, heterogeneously hypoechoic area at her anterior neck. Contrast-enhanced computed tomography showed a well-defined contrast-enhanced tumor inferior to the hyoid bone. Fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT) showed a fluorine-18 fluorodeoxyglucose-avid tumor with a maximum standardized uptake value of 12.8. Surgical tumor resection was performed, and the histopathological finding was ectopic papillary carcinoma lacking thyroglossal duct remnants, which is very rare. To our knowledge, few cases of ectopic thyroid carcinoma with ^{18}F-FDG PET/CT findings have been reported. Ectopic thyroid carcinoma lacking thyroglossal duct remnants should be considered a differential diagnosis in cases of ^{18}F-FDG uptake in an anterior middle neck tumor.

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Introduction

Ectopic thyroid tissue is a developmental abnormality involving aberrant embryogenesis of the thyroid gland at an early stage. It has been found in a variety of sites ranging from the base of the tongue to the neck, trachea, mediastinum, heart, adrenal glands and pancreas; the mediastinum is the most frequent location after the neck [1, 2, 3]. The prevalence of ectopic thyroid tissue is about 1 per 100,000–300,000 people, more commonly in females [4]. The probability of this tissue developing into ectopic thyroid carcinoma (ETC) is less than 1% [5]. To our knowledge, case reports of ETC with fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT) findings are few. We present a rare case of ETC lacking thyroglossal duct remnants detected by ^{18}F-FDG PET/CT.

Case Presentation

An 80-year-old woman presented with a 17-mm tumor at the anterior middle neck, and received a diagnosis of thyroglossal duct cyst at a local hospital. However, the tumor size increased to approximately 20 mm, and malignancy was suspected. She was admitted to our institution. Ultrasonography showed a smooth heterogeneous tumor at the anterior middle neck, smooth isoechoic thyroid nodules without calcification, and no cervical lymph nodes swelling (Fig. 1). Fine needle aspiration cytology (FNAC) was done to both the thyroid gland and the anterior middle neck tumor. Papillary thyroid cancer of the anterior middle neck tumor was diagnosed. There was no pathological finding of malignant cells in the thyroid gland. Thyroid nodules were diagnosed as adenomatous goiter. Laboratory data regarding liver and renal function, electrolytes, tumor markers, thyroid function, and complete blood count were within normal ranges. Contrast-enhanced computed tomography (CT) of the neck was performed to evaluate the anterior neck tumor characteristics, tumor spread and lymph metastasis. Contrast-enhanced CT showed a clear-boundary tumor inferior to the hyoid bone and no cervical lymph metastasis (Fig. 2a, b). This tumor did not include a cystic component, and showed no invasion into the hyoid bone and adjacent soft tissue. On CT and ultrasonography, this tumor had no continuity with the thyroid gland. ^{18}F-FDG-PET/CT was performed to examine distant metastasis and other malignant tumors. An integrated full-ring PET/CT scanner (Biograph Duo; Siemens Japan, Tokyo, Japan) was used for data acquisition. After a 4 h fast, the patient's blood glucose level was 108 mg/dL. She was administered a 336 MBq dose of ^{18}F-FDG and rested quietly for approx. 60 min. ^{18}F-FDG PET/CT showed high accumulation in the anterior middle neck tumor, with a maximum standardized uptake value (SUVmax) of 12.8, but the rest of the tissue showed normal ^{18}F-FDG uptake (Fig. 2c, d). As a thyroglossal duct carcinoma or lymph node metastasis from carcinoma of another primary unknown source was suspected, and the size of the anterior middle neck tumor was increasing, surgical tumor resection was performed. Histopathological findings showed an ETC lacking thyroglossal duct remnants (Fig. 3). As there was no finding of cervical lymph node metastasis or cancerous lesions of the thyroid gland, and she was the advanced age of eighty, neither cervical lymph node resection nor thyroidectomy was performed. After the operation, she was followed up at a local hospital for approximately 54 months using CT and ultrasonography. Iodine-131 (^{131}I) whole-body scintigraphy was not done because subsequent adjuvant radioiodine therapy for metastasis was not required. She has had no local recurrence or metastases, and the characteristics and size of the thyroid nodules have remained unchanged.

Discussion

ETC has been reported in the thyroglossal duct, lateral thyroid tissue, lingual thyroid, mediastinum and ovarian stroma [1]. The common histopathological types are papillary carcinoma (80%), a follicular variant of papillary carcinoma (8%), and squamous cell carcinoma (6%), with the remaining 6% represented by Hurthle cell adenoma and anaplastic carcinoma [1]. Most ETC cases originate from thyroglossal duct remnants and the medial lingual thyroid [6]. This case was an ETC lacking thyroglossal duct remnants, considered to be very rare. Only a few cases of ETC without thyroglossal duct remnants have been reported in the literature [7, 8]. The differential diagnosis of ETC is cervical lymph node metastasis derived from thyroid cancer, and the possibility of metastasis from an occult papillary thyroid cancer could not be ruled out initially. However, there was no malignant finding of the thyroid gland on FNAC and no ^{18}F-FDG-avid lesions of the thyroid gland on ^{18}F-FDG PET/CT. The diagnosis of the thyroid nodule was adenomatous goiter, and the size and characteristics of the thyroid nodules did not change during the follow-up period. The histopathological finding of anterior neck tumor showed papillary thyroid cancer with normal thyroid tissue. Based on these clinicopathological findings, metastatic papillary thyroid carcinoma from the thyroid gland could be excluded. The choice of therapy for ETC is based on the patient's age and the location, size and extent of the tumor. Treatment of ETC is based predominantly on surgical excision, because ETC is usually diagnosed only after surgical excision of the lesion [9]. A neck dissection would only be indicated if metastatic nodules were noted [10]. In the case of a positive margin, or the presence of more advanced disease, adjuvant radioiodine treatment would be considered [9, 10]. Patients with ETC don't always undergo thyroidectomy; it is performed when subsequent radioiodine treatment for metastasis is required, and ETC with concurrent thyroid cancer is suspected [9]. However, Patel et al. found that the addition of thyroidectomy to tumor resection did not have a significant impact on outcome [11]. Opinions differ whether thyroidectomy should be performed or not. As this case was judged as low risk according to the papillary thyroid cancer classification [12], no thyroidectomy or radioiodine treatment was performed. The overall prognosis of ETC is favorable, with 5- and 10-year Kaplan-Meier overall survival rates of 100 and 95.6%, respectively [11].

Regarding ETC-related nuclear medicine imaging, Michigichi et al. suggested the usefulness of thallium-201 chloride (^{201}TlCl) scintigraphy for the detection of ETC in patients with lymph node metastasis [13]. Piciu et al. showed an image from ^{131}I whole-body scintigraphy for the diagnosis of ETC with lymph node metastasis in the mediastinum after thyroidectomy due to thyroid papillary microcarcinoma developing from lymphatic thyroiditis [14]. Vázquez et al. reported that ^{18}F-FDG PET/CT is useful for detecting metastatic lesions and deciding the management of ETC patients [15]. Reports of the usefulness of nuclear medicine imaging including ^{18}F-FDG PET/CT for diagnosing ETC are very few. If ^{18}F-FDG uptake in the anterior middle neck tumor is observed incidentally, ETC lacking thyroglossal duct remnants should be considered a differential diagnosis.

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Statement of Ethics

The authors have no ethical conflicts to disclose.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Concept and design, HK; Acquisition of data, HK, HI, TC; Drafting of the manuscript, HK, TC; Critical revision of the manuscript for important intellectual content, HK, KI, TC; All authors approved final version of manuscript.

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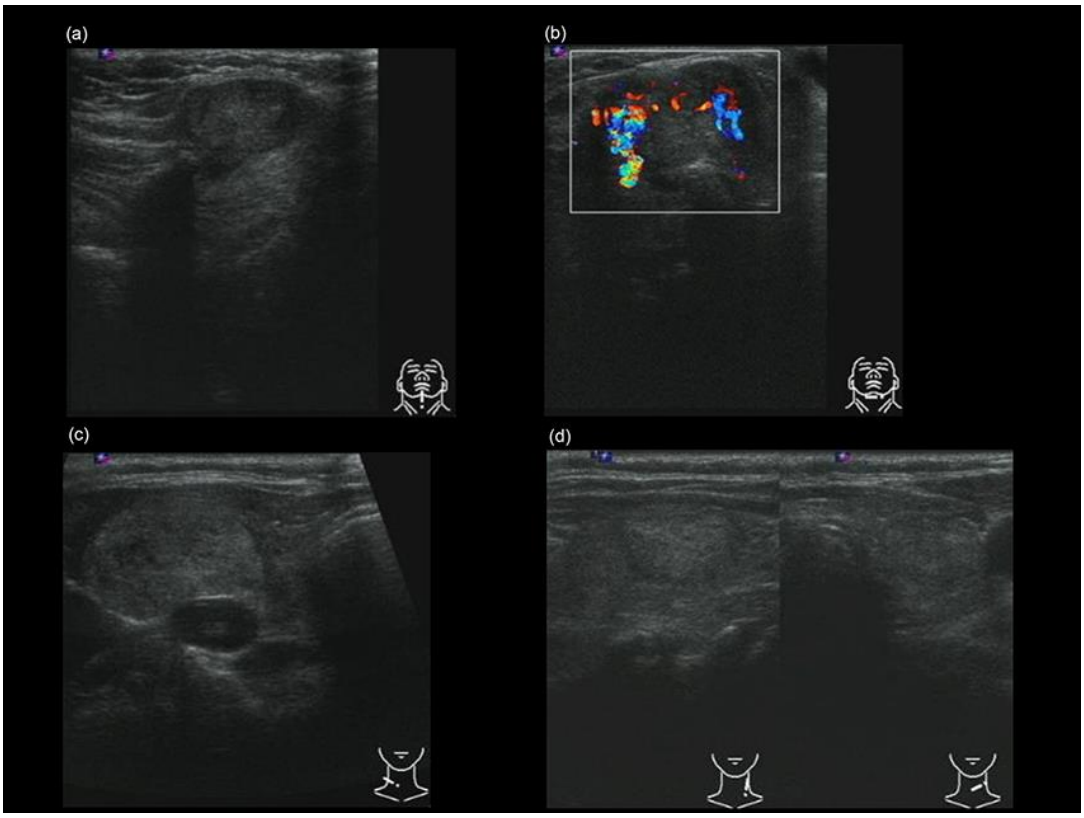

Fig. 1. Ultrasonography showed a 19 × 16 mm heterogeneous solid nodule (**a**), and Doppler ultrasonography revealed a hypervascular nodule (**b**) at the anterior neck. Ultrasonography showed 23 × 14 mm (right lobe) (**c**) and 15 × 8 mm (left lobe) (**d**) smooth isoechoic thyroid nodules including cystic degeneration.

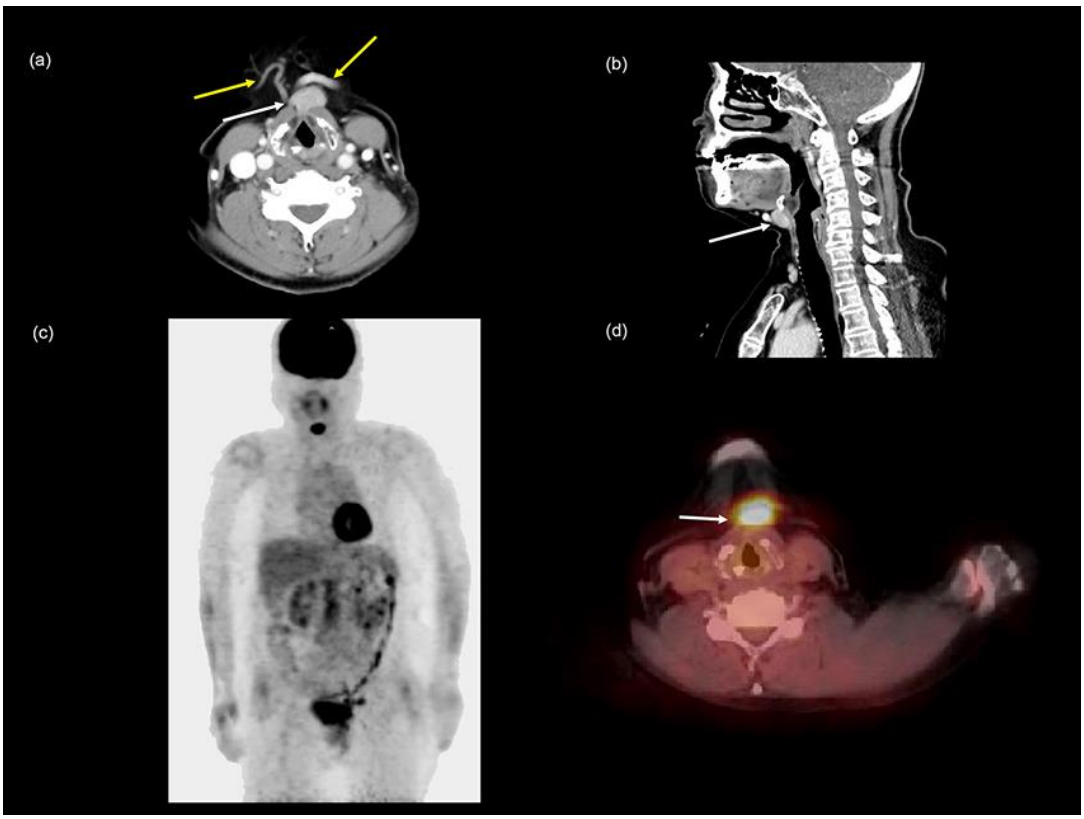

Fig. 2. Contrast-enhanced computed tomography of the neck (**a**; axial image, **b**; sagittal image) showed a 20 × 10 × 17 mm homogeneous contrast-enhanced tumor inferior to the hyoid bone (white arrow). The vascular structure in front of the tumor is the anterior jugular vein (yellow arrows). ^{18}F-FDG PET/CT showed high accumulation in the anterior middle neck tumor, with a SUVmax of 12.8 (**c**; Maximum Intensity Projection image, **d**; axial PET/CT fusion image [white arrow]).

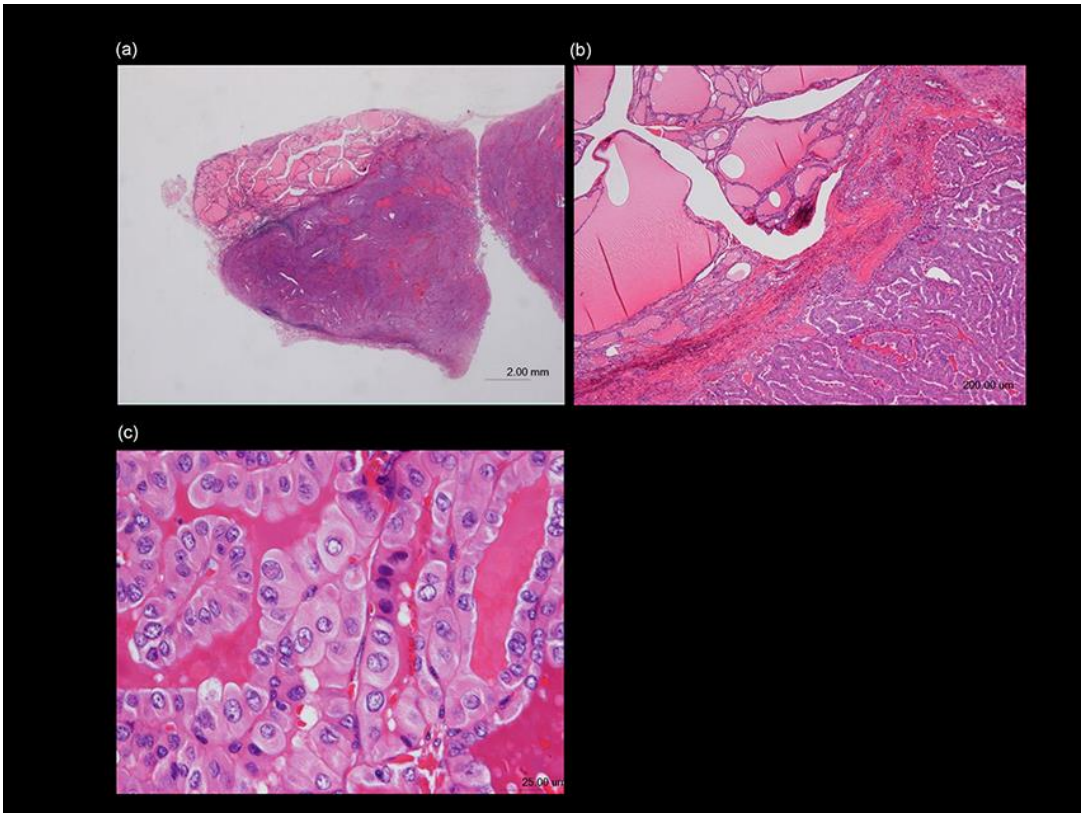

Fig. 3. Hematoxylin-eosin staining of the tumor showed normal thyroid tissue and papillary cancer (**a** ×40), and the growth and invasion of cancer with irregular follicular and papillary architectures (**b** ×100). Neoplastic cells showed enlargement of the nuclear groove and eosinophilic cytoplasm (**c** ×400). No thyroglossal duct tissue was found.