A Case of Ectopic Thyroid Cancer in the Abdominal Cavity

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Ectopic thyroid glands occur in the head and neck, typically at the base of the tongue but rarely in the abdominal cavity. Malignant tumors can also arise from ectopic thyroid glands. Here, we present an extremely rare case of intraperitoneal ectopic thyroid cancer. A 61-year-old woman was diagnosed with a mass in the left para-aortic region based on computed tomography (CT) at another hospital for a preoperative diagnosis of rectal cancer. Positron emission tomography-computed tomography revealed mild fluorodeoxyglucose accumulation in the mass, and rectal cancer metastasis was suspected. The mass was resected laparoscopically along with the primary tumor. Postoperative pathology revealed a follicular papillary carcinoma of the thyroid gland. The patient had a history of an ovarian cyst; therefore, we considered teratoma as a differential diagnosis, in addition to lymph node metastasis of thyroid cancer into the abdominal cavity. However, based on the histological findings, we considered primary intra-abdominal thyroid cancer to be the most likely diagnosis. The mechanism of intra-abdominal thyroid tissue development remains unknown because of the small number of cases, and future case reports are warranted.

Key words: ectopic thyroid carcinoma, abdominal, follicular variant, teratoma

INTRODUCTION

Ectopic thyroid is a congenital abnormality in which the thyroid tissue is present in a position other than the intrinsic position of the thyroid gland, and this is found in approximately 0.17 per 1000 individuals [1]. Generally, ectopic thyroid tissue is caused by either a defect in the descent of the thyroid gland primordium during the embryonic period or by straying into the proximal tissues after the thyroid gland has settled in its intrinsic position but before the capsule is formed [1]. Approximately 90% of these lesions occur at the base of the tongue and are often derived from the remnant tissue of the thyroglossal duct [2]. There have also been reports of it occurring in the head and neck (trachea, submandibular gland, lateral neck, and palatine tonsils) and mediastinum [2]. It rarely occurs intraperitoneally or in the pelvis, and a preoperative diagnosis is, therefore, difficult. An ectopic thyroid in the abdominal cavity is usually discovered incidentally during treatment for other diseases. Malignant tumors can also arise from ectopic thyroid glands. Here, we present an extremely rare case of intraperitoneal ectopic thyroid cancer.

CASE PRESENTATION

Case: 61-year-old female
Chief complaint: None in particular

Contrast-enhanced computed tomography (CT) performed at another hospital for preoperative examination of rectal cancer revealed an intra-abdominal mass, and the possibility of lymph node metastasis from rectal cancer, Castleman disease, and malignant lymphoma could not be ruled out. The patient underwent laparoscopic high anterior resection and D3 dissection for rectal cancer, and the intra-abdominal mass was removed concomitantly. The postoperative histopathological diagnosis suggested an ectopic thyroid gland, possibly malignant, and the patient was referred to our hospital. The patient was unaware of any abdominal symptoms. The patient had a medical history of laparotomy for an ovarian cyst approximately 30 years ago. A "hair-bearing tumor" was removed, the details of which were unknown.

Preoperative imaging findings

A thyroid ultrasound revealed no findings suggestive of thyroid cancer. Multiple small hypoechoic masses suspicious for adenomatous nodules (Fig. 1).

Computed tomography (CT): The thyroid gland was in a normal position, and there were no neoplastic lesions in the neck. Two masses approximately 2 cm in diameter were observed on the left side of the abdominal aorta and were contiguous above and below the aorta. Both lesions contained internal calcified areas (Fig. 2A, B–C). Fluorodeoxyglucose positron emission tomography (FDG-PET) revealed mild FDG accumulation in the intra-abdominal mass (Fig. 2D).

Histopathological findings

Fibrous tissue was the major component of the mass, accompanied by a small amount of adipose tissue outside the capsule. The thyroid tissue was observed at the margins; however, no lymph node structures were observed (Fig. 3A, B). Most ectopic thyroid

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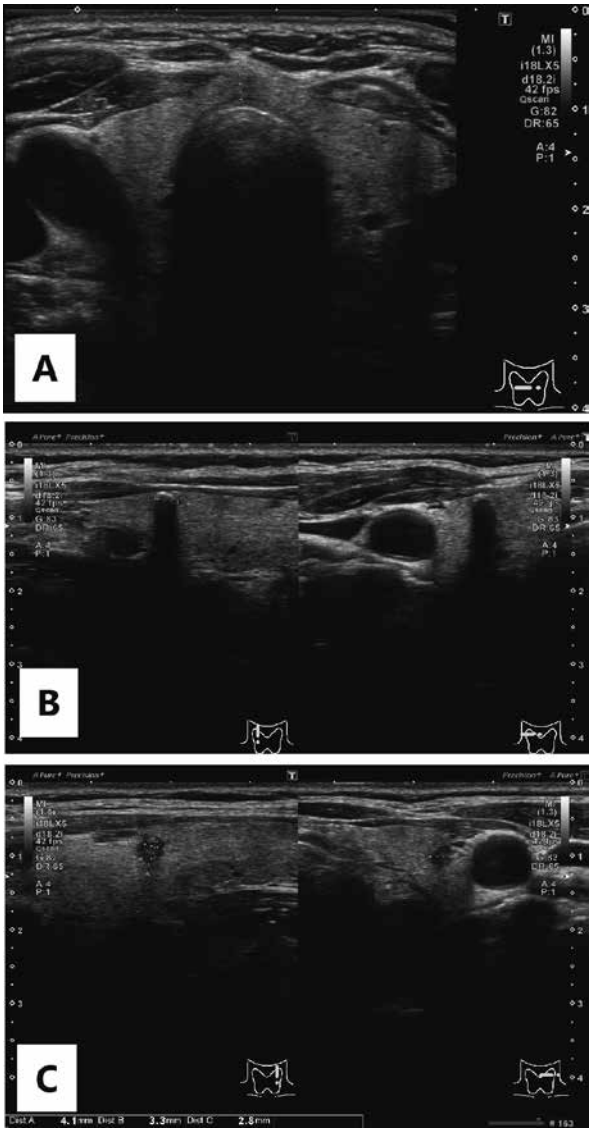

Fig. 1 No mass suspected of being thyroid cancer was found on the thyroid ultrasound. (A) Left lobe: Multiple hypoechoic masses suspicious for adenomatous nodules, max 7 mm in diameter. (B) Right lobe: Hypoechoic masses suspicious for adenomatous nodules, 3 mm in diameter. Calcified lesion with acoustic shadow, 3 mm in diameter. (C)

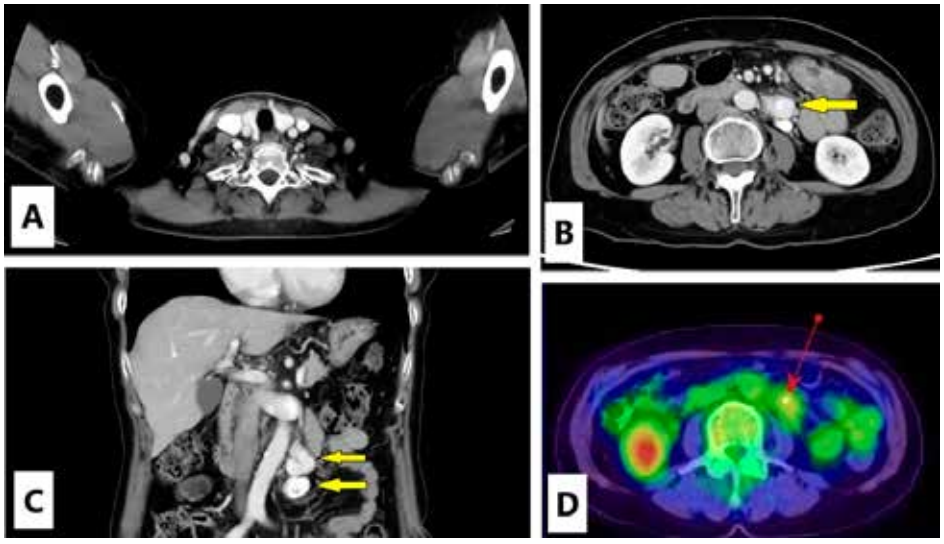

Fig. 2 The thyroid gland was located in the normal position, and there were no neoplastic lesions in the neck. (A) On contrast-enhanced abdominal CT, the arrow indicates an approximately 2-cm mass on the left side of the abdominal aorta. (B) Arrows show masses contiguous above and below the aorta. Both lesions exhibited calcified internal areas. (C) On fluorodeoxyglucose-positron emission tomography (FDG-PET), the arrow indicates mild FDG accumulation in the intra-abdominal mass. (D)

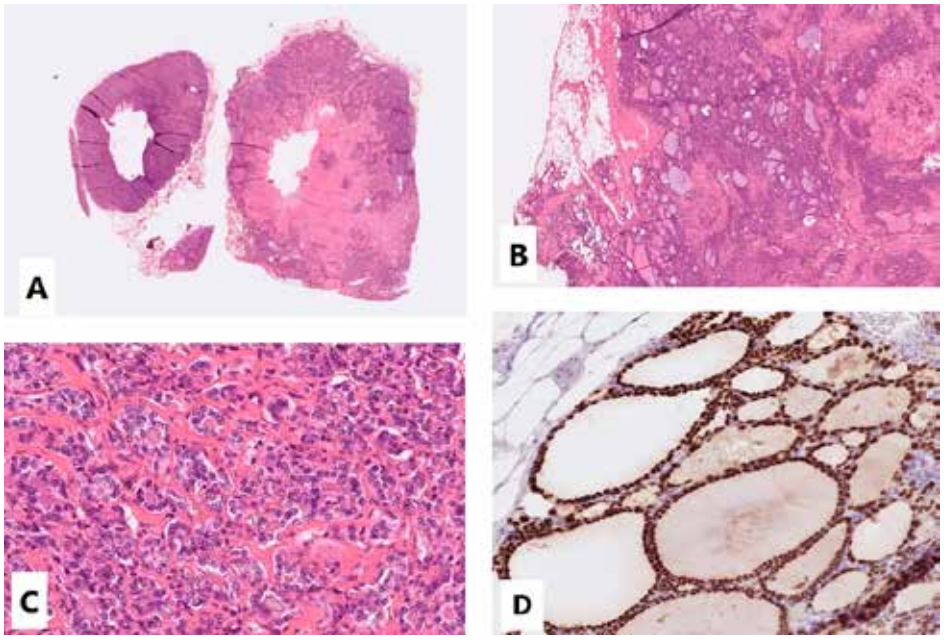

Fig. 3 Histological sections of the specimen.
Hematoxylin- and eosin- (HE) stained specimen under low-power magnification (0.5 ×) (A), (3.2 ×) (B)
The mass mainly comprised thyroid and fibrotic tissues. This was accompanied by a small amount of adipose tissue outside the capsule. However, no lymph node tissue was observed.
HE-stained specimen under high magnification (20 ×) (C)
A portion of the thyroid tissue contained dense atypical cells that retained a follicular structure. The atypical cells exhibited nuclear grooves, frosted nuclei, and nuclear inclusion bodies.
Thyroid transcription factor(TTF)-1 immunohistochemistry (15 ×) (D)
Thyroid tissue and atypical cells were diffusely TTF-1 positive.

tissues exhibit dense atypical cells and are considered malignant. The tumor grew in a follicular manner, and papillary structures were not evident. The atypical cells exhibited nuclear findings characteristic of papillary carcinoma, such as nuclear grooves and ground-glass nuclei, as well as a small number of intranuclear inclusions (Fig. 3C). Immunohistochemistry revealed that the thyroid tissue and atypical cells were diffusely positive for thyroid transcription factor-1 (Fig. 3D). These findings suggest a primary follicular variant of papillary thyroid carcinoma, diagnosed as intraperitoneal ectopic thyroid cancer. Owing to the strong calcification of the tumor, additional thin-section specimens could not be obtained, and a histological diagnosis of the calcified area was not possible.

Clinical course

After referral to our hospital, additional computed tomography (CT) scans did not reveal any obvious lesions. The thyroid gland was in a normal position and no other cervical lesions were detected on ultrasonography. Thyroid function was normal (free triiodothyronine: 2.17 pg/mL, free thyroxine: 1.37 ng/mL, thyroid-stimulating hormone: 0.762 U/mL, thyroglobulin: 11.9 ng/mL). The patient is currently not undergoing any treatment, and no obvious recurrence or metastasis was observed one year after the surgery.

DISCUSSION

Ectopic thyroid tissue arising in the head and neck can cause pharyngeal discomfort due to local compression and hypothyroidism; however, most cases are asymptomatic and can be diagnosed using fine-needle aspiration cytology, thyroid scintigraphy, or biopsy specimens. However, symptoms of ectopic thyroid gland at other sites, especially in the abdominal cavity, have been reported in fewer cases and are unknown. Thirty-four cases of intra-abdominal ectopic thyroid tissue were reported from 1960 to 2017 according to Ma *et al.*, 13 of which were adrenal, eight gallbladder, four hepatic hilum, two mesenteric, two pancreatic, one liver, one spleen, one retroperitoneal, one duodenal, and one jejunal [3]; however, none of them presented with characteristic symptoms. Preoperative diagnosis is difficult because the disease is often discovered incidentally on CT or other imaging studies associated with other diseases. Therefore, it is usually diagnosed histologically after surgical resection. Two hypotheses have been proposed for the appearance of ectopic thyroid tissue in the abdominal cavity: the malformation theory, whereby a single embryonic cell transforms into thyroid tissue, as in a teratoma [4], and the theory of ectopic differentiation of the endoderm from the primitive foregut into thyroid tissue [3]. The possibility of excessive descent of the thyroid gland protuberance beyond the diaphragm is considered unlikely but has not been ruled out. Of the ectopic abdominal thyroid cases, 82.4% were females and most had normal thyroid function [4]. In the present case, the preoperative thyroid function was not measured; however, it was normal postoperatively and was not accompanied by abnormal thyroid function. A literature review by Fu *et al.* covering 110 years until 2022 reported 132 cases of ectopic thyroid cancer, of which 113 were in the head and neck, 11 thoracic, two abdominal (liver and rectum), and six pelvic. Ectopic thyroid cancer of the abdominal cavity is extremely rare [5]. Regarding the histological types of ectopic thyroid cancer, 10 cases of

papillary carcinoma, two cases of follicular papillary carcinoma, and one case of medullary carcinoma have been reported for ectopic thyroid cancer at the base of the tongue, which is a relatively common site [6]. In the histopathological diagnosis of the present case, papillary carcinoma tissue with follicles was found, and a follicular variant of papillary thyroid carcinoma was diagnosed. Therefore, differential diagnosis of metastatic thyroid cancer is necessary. Although no suggestive primary lesions were observed in the thyroid gland, occult thyroid cancer could not be ruled out. However, as no lymph node tissue was identified in the specimen, a diagnosis of primary ectopic thyroid cancer rather than metastatic thyroid cancer was made. The patient had undergone laparotomy for an ovarian cyst approximately 30 years prior, albeit with unknown details, but the "hair-bearing tumor" suggests that it may have been a mature cystic teratoma. Mature cystic teratomas can contain thyroid tissue, hair, cartilage, and teeth. Although rare, a case of recurrence due to intraperitoneal dissemination after surgery for mature cystic teratoma has been reported [7], but there is little reason to suspect that this case was a dissemination of teratoma, since teratomas are defined as consisting of three germ layers in principle, and no case of intraperitoneal dissemination of only the thyroid tissue was identified. The patient was found incidentally on a CT scan with no local symptoms. Although the imaging findings revealed calcification, it was difficult to differentiate it from a lymph node lesion, thus making preoperative diagnosis difficult. As the possibility of rectal cancer metastasis could not be ruled out, the decision to simultaneously remove the abdominal mass was considered appropriate. Although there have been no reported cases of ectopic thyroid cancer recurrence in the literature to date, when an abdominal mass is discovered during the follow-up of a patient and there is a possibility of malignancy, histological diagnosis is required to determine whether it is a recurrent metastasis of colon or thyroid cancer.

CONCLUSION

Here, we present an extremely rare case of ectopic thyroid cancer in the abdominal cavity. Although the patient had a history of suspected teratoma, a histological diagnosis of primary ectopic thyroid cancer was made. The patient is currently in good health, with no recurrence or metastasis one year after the surgery.

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