

# Peritoneal and mediastinal highly differentiated follicular carcinoma of ovarian origin

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**ABSTRACT** A 70-year-old female patient presented to her primary care doctor with persistent elevated alkaline phosphatase of suspected metastatic etiology. Computed tomography demonstrated epicardial and peritoneal nodules. Biopsy of one of the peritoneal nodules revealed thyroid tissue and extraovarian struma ovarii was considered. The patient had a history of remote total abdominal hysterectomy and bilateral salpingo-oophorectomy 31 years prior for endometriosis with no available pathology from that surgery. The patient recalls being told that she had a left ovarian cyst. A thyroid ultrasound was performed that demonstrated multiple nodules without concerning features; however, due to high clinical suspicion, a total thyroidectomy was performed. Upon full histological evaluation a 0.5 cm papillary microcarcinoma was found. Given the rarity of metastatic papillary cancer to the peritoneum and the small size and grade of the tumor, a diagnosis of highly differentiated follicular carcinoma of ovarian origin was favored. The patient was subsequently treated with radioiodine therapy.

**Keywords:** Highly differentiated follicular carcinoma of ovarian origin, metastatic papillary cancer, struma ovarii

## INTRODUCTION

Struma ovarii is defined as an ovarian teratoma composed primarily of thyroid tissue.<sup>[1-6]</sup> The thyroid elements may be benign or malignant in histology, the latter most commonly appearing as papillary and less frequently follicular thyroid-type carcinoma.

Extra-ovarian spread of struma ovarii is rare, but when it does occur, it is usually intra-abdominal and the majority of cases are diagnosed post-operatively. The extra-ovarian tissue can have a histologically benign appearance or can demonstrate features seen in thyroid-type carcinoma. The former has been historically classified as “peritoneal strumosis,” although more recent literature<sup>[2-3]</sup> recognizes these cases as neoplastic, terming them highly differentiated follicular carcinoma of ovarian origin (HDFCO).

HDFCO is characterized by “extraovarian dissemination of thyroid elements with histological resemblance to non-neoplastic thyroid tissue.”<sup>[2,4]</sup> Our literature search reveals only two documented cases of HDFCO with spread to the peritoneum, making this a rare but important entity to consider in cases with appropriate pathological findings.<sup>[2,7-8]</sup>

## CASE REPORT

Here we report a gravida 3, para 2 Caucasian 70-year-old female patient who was evaluated at an outside institution for persistently elevated alkaline phosphatase levels with no clinical complaints. A bone scan was obtained at that time and demonstrated abnormal uptake in the cranium, long bones and pelvis, raising the possibility of metastatic disease and the patient was referred to our institution. The patient demonstrated an elevated thyroglobulin level of 101.5 ng/ml (normal 0-33.0 ng/ml). In addition, the tumor marker CA 15-3 was also elevated at 34.7 u/ml (normal 10-19.9 u/ml).

Contrast enhanced computed tomography scan of the chest, abdomen and pelvis demonstrated multiple peritoneal nodules, the largest of which measured 3.2 cm × 2.2 cm [Figure 1]. In addition, hyperenhancing epicardial nodules were also noted [Figure 2]. Biopsy of one of the peritoneal lesions revealed thyroid tissue without atypia and we investigated whether struma ovarii was possible. The patient had

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undergone an elective total abdominal hysterectomy and bilateral salpingo-oophorectomy for endometriosis at age 39 years. The pathology findings were not available. The patient reported being told of a left ovarian cyst without the need for follow-up.

Given the number of lesions associated with the omentum, the patient underwent a diagnostic laparoscopy and partial omentectomy. Pathology from the excised omental nodules demonstrated histologically benign appearing thyroid tissue with multiple thyroid follicles of various sizes [Figure 3]. Pelvic washings were negative for malignant cells.

As part of the diagnostic evaluation a bone marrow biopsy was performed and was negative for malignancy. In addition, a thyroid ultrasound was performed that demonstrated multiple small nodules with a dominant nodule in the right upper thyroid lobe. Three of the nodules were biopsied and the cytologic findings were consistent with benign nodules.

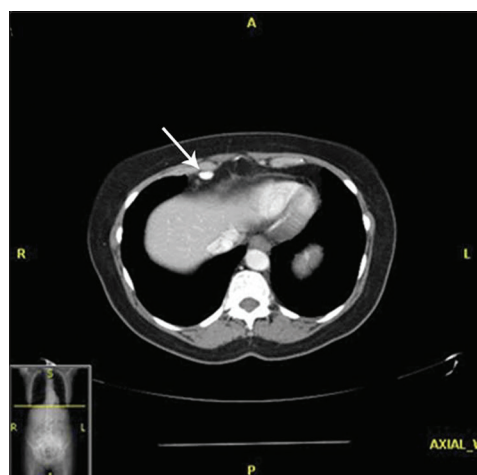
Given the patient's extensive extra-ovarian struma ovarii, a total thyroidectomy was recommended prior to radioiodine therapy. Pathological evaluation of the thyroid revealed a 0.5 cm focus of well-differentiated papillary carcinoma (a microcarcinoma) without lymphovascular invasion, stage PT1 [Figure 4].

Total body I-123 uptake scan was performed for treatment planning purposes and demonstrated innumerable areas of uptake within the chest, abdomen and pelvis [Figure 5]. A discussion was held regarding the appropriate I-131 treatment dose. Given concerns about bowel radiation injury, the patient was treated with 30.5 mCi of radioiodine 131.

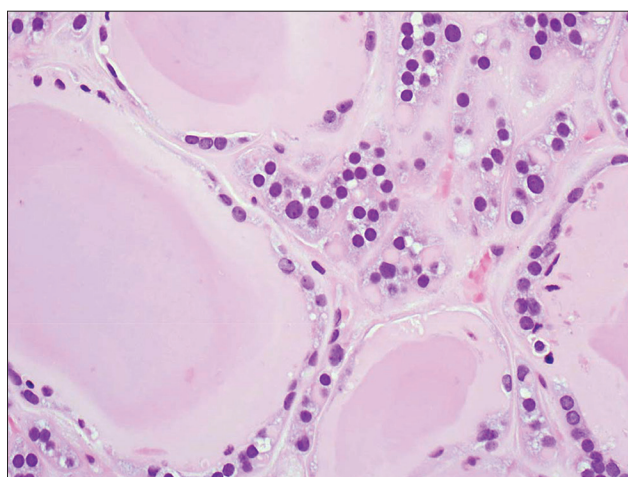
A follow-up I-123 uptake scan 4 months later showed resolution of the chest uptake and significant decline in the number of lesions throughout the abdomen and pelvis that were consistent with a short-term positive response [Figure 6]. Thyroglobulin tumor marker at the follow-up scan was elevated at 91 ng/ml. Plans were made for continued follow-up.



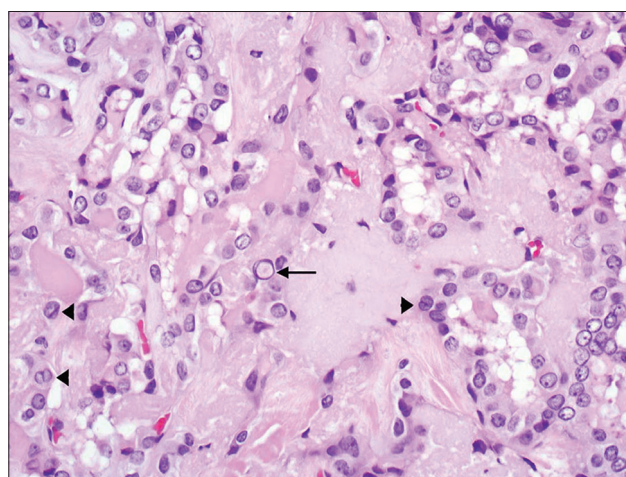
**Figure 1:** Computed tomography image of the pelvis demonstrating multiple peritoneal lesions, the largest of which is demonstrated above



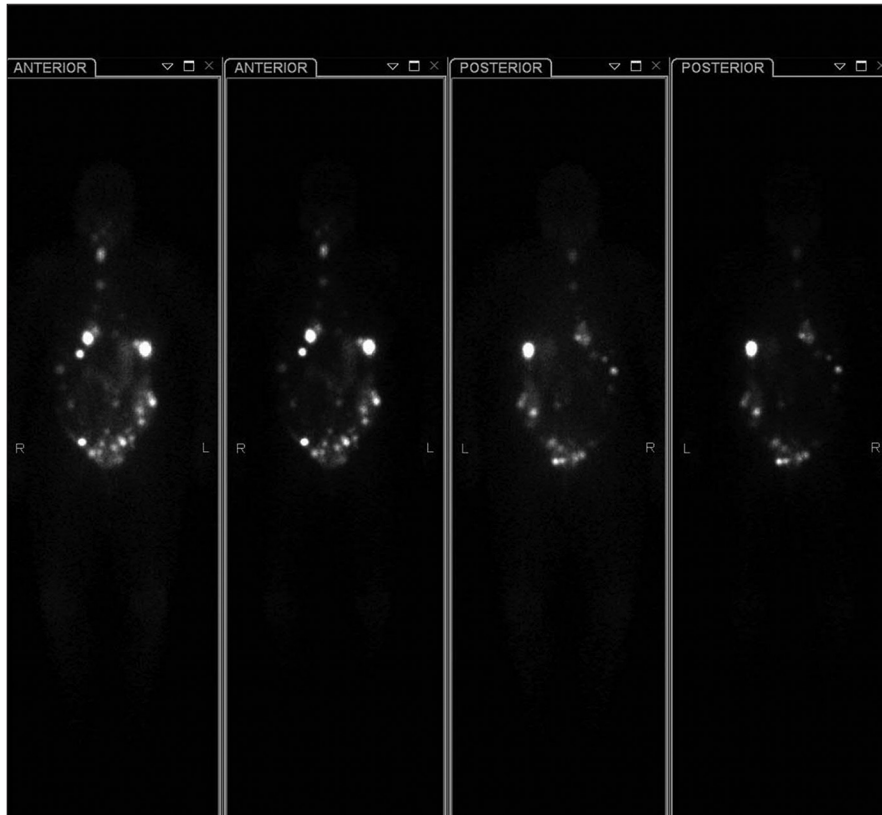
**Figure 2:** Hyperenhancing epicardial nodules



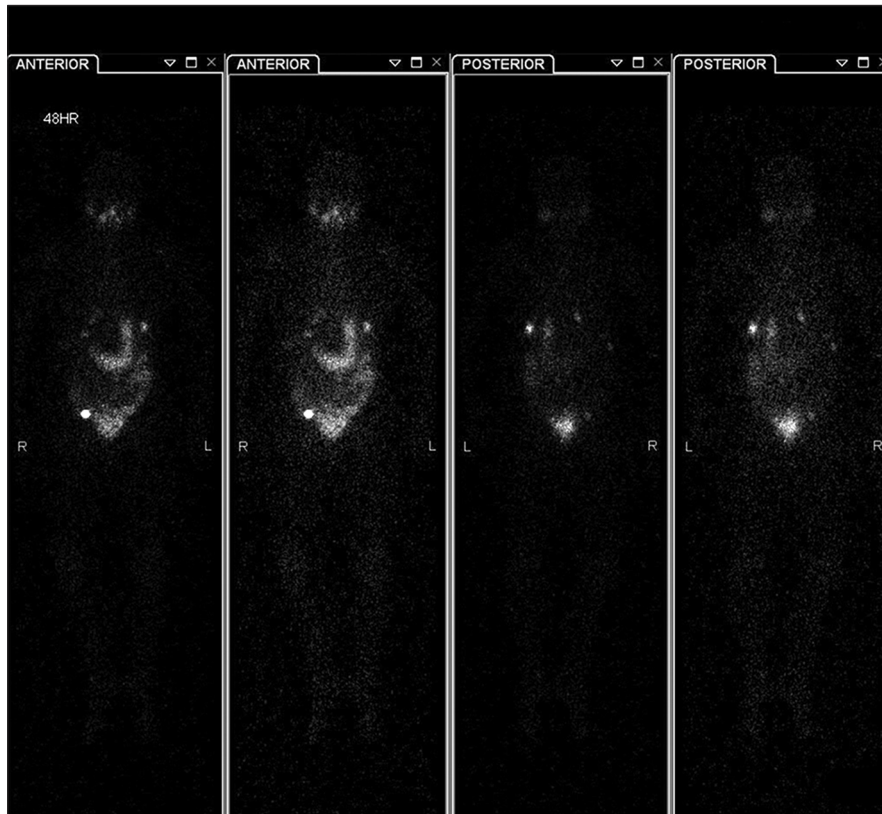
**Figure 3:** Papillary thyroid carcinoma, with characteristic nuclear grooves (arrowheads) and a nuclear inclusion (arrow) (H and E, ×400)



**Figure 4:** Abdominal thyroid tissue characterized by histologically bland thyroid tissue including follicles and colloid (H and E, ×400)



**Figure 5:** Total body radioactive iodine scan prior to therapy shows uptake in the abdomen as well as small foci in the mediastinum. At 24 h I-123 uptake in the cervical thyroid bed was measured at 1.4% (normal = 8%-29%)



**Figure 6:** Follow-up total body radioactive iodine scan demonstrates resolution mediastinal uptake and significantly decreased uptake in multiple abdominal and pelvic nodules

## DISCUSSION

Struma ovarii occurs when thyroid tissue is the predominant element in an ovarian teratoma. In a small percentage of these cases, the thyroid tissue has a malignant histology and is capable of spreading to the peritoneum, bones, cranium and multiple organs.<sup>[3,9]</sup> Struma ovarii has been stated to metastasize in 5-23% of cases.<sup>[11]</sup> The majority of metastases are diagnosed upon excision of the primary ovarian mass. There are less than thirty reports of struma ovarii metastasizing to distant sites<sup>[10]</sup> and even fewer that demonstrate the significantly delayed presentation that was seen in this case. We could find only one reported case in which peritoneal dissemination was discovered 26 years after initial operation; However, several cases of delayed dissemination to other organ systems exist.<sup>[2,3]</sup> For example McDougall *et al.* reported the discovery of vertebral and liver metastases in a patient 41 years after oophorectomy for ovarian tumor.<sup>[7]</sup>

Roth and Karseladze<sup>[2]</sup> have recently proposed a new entity termed “HDFCO”. This term has been applied to patients with metastases of ovarian struma that have a bland, nonmalignant histologic appearance. This diagnosis characteristically cannot be made until distant metastasis has occurred. The case presented here is consistent with the diagnosis of HDFCO.

The current case is complicated by the lack of any significant ovarian pathology from the patient’s hysterectomy and bilateral oophorectomy 31 years ago. After pathology review of the omentectomy specimens confirmed thyroid tissue without atypia, the location of the primary tumor remained unclear-thyroid versus ovarian.

The histologic features of the papillary thyroid microcarcinoma in this case were distinctly different from the histology of the omental nodules. The possibility of distant metastases from the papillary thyroid microcarcinoma was felt to be highly unlikely due to its small size and lack of cervical lymph node metastases.<sup>[5,6,11]</sup>

Although we were unable to review the original ovarian pathology from 31 years ago, which was reported as benign, our investigation supports this case as a likely additional example of HDFCO and illustrates the insidious nature of struma ovarii and the importance of maintaining a high degree of suspicion, even in patients who have had remote oophorectomy.

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