



Peritoneal strumosis: Presentation and management with multiple radioactive iodine treatments

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ARTICLE INFO

Keywords:

Struma ovarii
Peritoneal strumosis
Radioiodine
Radioactive I-131
Thyroid
Thyroidectomy

ABSTRACT

Background: Struma ovarii is a rare benign ovarian teratoma comprising of thyroid tissue. Infrequently, benign struma ovarii can present with extra ovarian dissemination, thus posing a treatment challenge. Here we describe a case of peritoneal strumosis treated with thyroidectomy and two courses of radioiodine ablation.

Case presentation: 70 year old female with history of bilateral salpingo-oophorectomy 30 years prior presented with elevated thyroglobulin levels. Computed Tomography of the chest, abdomen and pelvis showed multiple peritoneal lesions. Subsequent biopsy revealed benign thyroid tissue. Thyroidectomy revealed papillary thyroid carcinoma; however, it did not match the peritoneal lesions. She was subsequently treated with radioiodine in two doses, each 30 mCi, 20 months apart. Follow up imaging and thyroglobulin levels showed a significant response to radioactive iodine treatment.

Conclusion: Peritoneal Strumosis is a rare presentation of extra ovarian struma ovarii with benign thyroid histology. We propose a treatment paradigm similar to a thyroid malignancy, including total thyroidectomy and radioactive ablation with I-131. Moreover, radioactive ablation can be performed in stages with smaller doses to avoid radiation-induced side effects with an excellent response.

1. Introduction

Struma ovarii is a rare tumor defined as an ovarian teratoma composed of at least 50% thyroid tissue. It accounts for 3% of all ovarian teratomas and is usually benign [1]. The thyroid tissue of the struma ovarii demonstrate equivocal histology and physiology to primary thyroid tissues. Roughly 5% of struma ovarii cases are malignant, and metastatic disease is even more rare [2]. The majority of metastatic struma ovarii is composed of papillary or follicular carcinoma. Rarely, cases of benign struma ovarii with extra ovarian dissemination have been reported and occasionally mistaken as malignant, metastatic disease [3]. There is no standard treatment strategy described due to the infrequency of presentation. Here we report a case of benign struma ovarii with peritoneal dissemination 30 years after bilateral oophorectomy treated with thyroidectomy and two courses of radioiodine ablation.

2. Case report

A 70 year old female with no reported symptoms initially presented with persistent elevated alkaline phosphatase to her primary care provider. Nuclear medicine bone scan was performed and demonstrated

diffuse abnormal uptake in the cranium, ribs, long bones and pelvis. Computed tomography of the chest, abdomen and pelvis was significant for multiple peritoneal lesions, the largest of which measured 3.2×2.2 cm (Fig. 1). No definite bone lesions were seen. CT guided biopsy of a peritoneal lesion at the level of the umbilicus revealed thyroid tissue without atypia concerning for the diagnosis of struma ovarii. She was referred to our institution for further evaluation.

Her past medical history included an elective total abdominal hysterectomy and bilateral salpingo-oophorectomy for endometriosis 31 years ago. No surgical pathology was available to review. Upon initial workup, significant labs included an elevated thyroglobulin level of 101.5 ng/ml (normal 0–0.33 ng/ml). MRI of the lumbar spine suggested a diffuse infiltrative process. Furthermore, bone marrow biopsy was negative for any evidence of metastatic disease or malignancy but revealed systemic mastocytosis explaining her abnormal alkaline phosphatase and bone scan.

Due to the multiple peritoneal lesions and risk of sampling error with biopsy, she subsequently underwent an examination under anesthesia with a diagnostic laparoscopy and partial omentectomy. Pathologic evaluation of the excised lesion was consistent with benign thyroid tissue without morphologic atypia. Immunostaining was positive for thyroglobulin confirming thyroid histology (Fig. 2). Pelvic

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Fig. 1. Initial CT Pelvis with contrast showing multiple enlarged peritoneal lesions. The largest lesion measured 3.2×2.2 cm.

washings were negative for malignant cells.

In subsequent work up of the thyroid, ultrasound exhibited multiple thyroid nodules. Biopsy of two thyroid nodules demonstrated benign thyroid tissue. To eliminate the possibility of an underlying thyroid malignancy, she underwent a total thyroidectomy. Pathologic evaluation demonstrated benign thyroid tissue with nodular hyperplasia in the right lobe and isthmus as well as a 0.5 cm focus of well differentiated, grade 2, papillary thyroid carcinoma in the left lobe with negative surgical margins (Fig. 3). There was no evidence of lymphovascular invasion, however focal extrathyroidal extension was noted Total I-123 uptake scan demonstrated uptake in the cervical thyroid bed, mediastinum and multiple areas within the abdomen and pelvis (Fig. 4A). She received 30 mCi (mCi) of radioiodine 131 to ablate the abdominal thyroid lesions as well as any post-surgical residual cervical thyroid tissue. A low dose was chosen to minimize the risk of small bowel complications. Post-treatment scan revealed uptake in all previously noted areas. She experienced no adverse reactions.

Five month follow up I-123 scan revealed significant improvement in the uptake in multiple areas of the abdomen compared to the previous study, but not complete resolution. (Fig. 4B). At one year follow up, positron emission tomography/computed tomography of the chest, abdomen and pelvis revealed significant decrease in the size of multiple abdominal lesions and no metabolic hyperactivity. At 20 months after initial radioiodine treatment, she underwent a second dose of 30 mCi of radioactive iodine. Post-treatment scan revealed uptake in the expected residual abdominal disease with no evidence of residual disease in the mediastinum or cervical thyroid bed. At last follow up, 41 months after treatment, she was completely asymptomatic and thyroglobulin levels decreased to 1.3 ng/ml. Follow up CT scan showed near resolution of prior abdominal soft tissues nodules. She declined follow up I-123 scan.

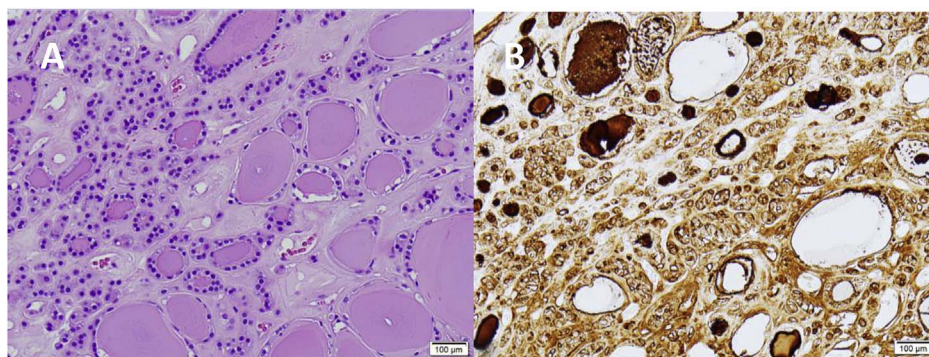


Fig. 2. Surgical Pathology. A) Hematoxylin and Eosin (H &E) stain of abdominal lesions showing normal thyroid tissue composed of thyroid follicles of various sizes. B) Immunohistochemical stain for Thyroglobulin stain of lesion is positive confirming diagnosis.

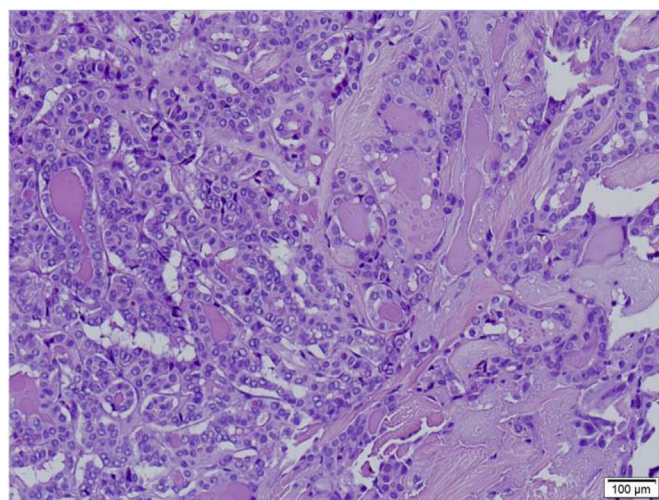


Fig. 3. Surgical Pathology (H &E) of thyroid reveals papillary thyroid carcinoma.

3. Discussion

Struma Ovarii with peritoneal dissemination, or peritoneal strumosis, is defined as struma ovarii with extra-ovarian spread with a histologically benign appearance [9]. The mean age of metastatic struma ovarii presentation is in the fourth decade [3]. Typical symptoms include pelvic pain, abdominal or pelvic mass, and abnormal uterine bleeding [4]. Initial treatment is resection of the primary tumor in order to aid in diagnosis [5]. Given the rarity of histologically benign but metastatic struma ovarii, there is no standard treatment regimen. For localized tumors, surgery is the optimal intervention [6]. However, this is not practical in the setting of metastatic disease. A more systemic approach is required.

Our patient presented asymptotically three decades after initial bilateral oophorectomy, therefore, the primary tumor was not identified. Thyroid surgical pathology revealed the presence of papillary carcinoma. However, pathology of the benign peritoneal metastasis did not match the malignant cervical thyroid pathology. Thus, this indicates two different processes, rather than cervical thyroid metastases to the abdomen, solidifying the diagnosis of peritoneal strumosis.

Authors of recent reports of malignant struma ovarii proposed an approach established for thyroid carcinoma, including total thyroidectomy and I-131 ablation with subsequent monitoring with thyroglobulin [7]. Radioactive Iodine (I-131) is an oral therapy used commonly for thyroid conditions such as Graves Disease, metastatic thyroid carcinoma, and postoperative ablation of thyroid remnants. It is a beta emitting radionuclide with a half-life of 8.1 days. A recent review of 40 cases of metastatic struma ovarii, comprising of benign and malignant pathology, advocated a similar treatment regimen [8]. However, no

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