



Case report

Management of highly differentiated thyroid follicular carcinoma of ovarian origin with a minimally invasive approach

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1. Introduction

Struma ovarii was first described in the early part of the 20th century. Accounting for only 3% of all ovarian teratomas, it is monodermal containing primarily thyroid tissue. Rarely, the thyroid tissue undergoes malignant transformation and can metastasize to the liver and to intra- and retro-peritoneal locations. The two most common types of thyroid carcinoma arising from struma ovarii include papillary carcinoma and follicular carcinoma. The majority of these do not spread beyond the ovary (Zhang and Axiotis, 2010). Describing the frequency that ovarian ectopic thyroid tissue becomes malignant has been difficult given conflicting pathologic terminology and its varying malignant potential. This was first addressed in an article by Roth and Karseladze that explored the varied descriptors of extra-ovarian dissemination (Roth and Karseladze, 2008). Traditionally, spread of histologically benign thyroid tissue from struma ovarii to the peritoneum has been termed “peritoneal strumosis.” Recognizing that spread of benign appearing thyroid tissue beyond a struma ovarii is evidence of a low-grade malignant neoplasm, the current favored pathologic term has transitioned from “peritoneal strumosis” to ‘highly differentiated thyroid follicular carcinoma of ovarian origin’ (HDFCO) (Roth and Karseladze, 2008). The published literature describing HDFCO is confined to case reports and describe treatment with both surgical resection and radioactive iodine ablation (RIA) following total thyroidectomy (Willemse et al., 1987; Brogssitter et al., 2004). Previously, the pelvic peritoneal stripping required for removal of peritoneal implants and complete cytoreductive surgery was completed using an open abdominal approach to enhance visualization and access to the lesions (Sugarbaker, 2013). We present the first reported case of HDFCO treated

conservatively with a minimally invasive robotic-assisted approach and thyroid preservation.

2. Case

In 2005, a 32 year old gravida-2 para-2 female presented to her gynecologic oncologist for surgical removal of a 14 cm complex adnexal mass found during her pregnancy. She reported a history of intermittent, severe, right-sided abdominal pain since November 2003. She had a normal CA-125 with a transvaginal ultrasound describing a 14 cm sized right ovarian mass with concerning features to include thick septations and solid components. She had no other significant medical history with a surgical history of a prior cesarean section. After her successful vaginal birth, she underwent an exploratory laparotomy postpartum with a right salpingo-oophorectomy and dilation and curettage. The operative report described a previously ruptured, 14 cm complex multi-cystic right ovarian mass. The frozen section was consistent with mature teratoma without evidence of atypical proliferation or malignancy. Final pathology revealed a struma ovarii. She developed worsening pelvic pain over the next decade. In 2014, she underwent a laparoscopic left salpingo-oophorectomy with final pathology consistent with a physiologic cyst. In 2016, she underwent another diagnostic laparoscopy for persistent pelvic pain that revealed petechiae on her uterus, anterior and posterior peritoneum (Fig. 1a, b). Biopsy of the petechiae revealed HDFCO. She was then referred to gynecologic oncology for treatment.

The patient strongly desired avoiding an open procedure and the decision was made to achieve complete resection with a robotic-assisted minimally invasive approach including a modified radical hysterectomy

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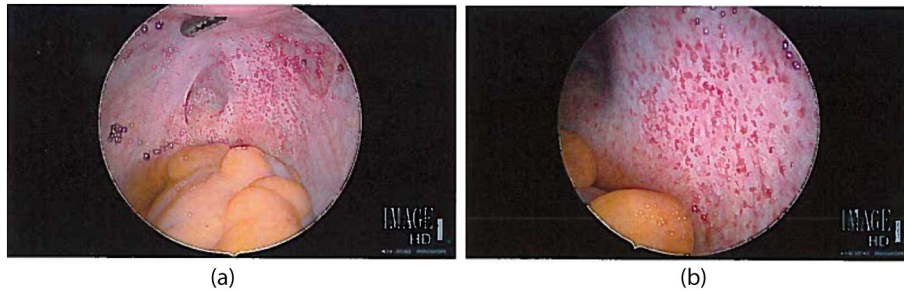


Fig. 1. (a and b) Laparoscopic photos from the diagnostic laparoscopy in 2016 first demonstrating evidence of the petechial HDFCO.



Fig. 2. Gross surgical specimen. Posterior uterus with arrows denoting miliary areas of strumosis.

with complete pelvic peritoneal resection. The procedure was performed using four eight millimeter laparoscopic ports and a single twelve millimeter assistant port. Intraoperatively, the patient had a normal appearing liver edge, stomach, diaphragm, and appendix. There was no evidence of bowel, omental, or upper abdominal involvement. Petechiae were noted along both the posterior and anterior peritoneum overlying the uterus and onto the pelvic sidewall. The retroperitoneum was entered, the ureters identified, and the pararectal spaces were developed. The overlying posterior peritoneum was stripped away from the recto-vaginal space and the pelvic side walls. The infundibulopelvic ligaments were then skeletonized and transected with cautery bilaterally. The bladder peritoneum was then peeled away from its attachments to the uterus and pelvic sidewalls using a combination of gentle traction and electrocautery to dissect the anterior peritoneum from the underlying bladder. The remaining portion of the procedure was completed in standard fashion for a robotic-assisted laparoscopic modified radical hysterectomy to better resect all grossly visible disease (Fig. 2).

The patient remained inpatient until hospital day two and was discharged home in stable condition. She was seen for follow up one week after her procedure with symptoms of urinary retention and overflow incontinence. After one week of indwelling catheter drainage, her urinary retention resolved. Since removal of the peritoneal implants, the patient reports resolution of her pelvic pain.

Final pathology revealed HDFCO with low-grade malignant potential with mature thyroid tissue involving the anterior and posterior peritoneal reflections of the uterus without evidence of malignancy (Fig. 3a, b). Her pre-, peri-, and postoperative thyroid function tests confirmed normal thyroid function. A neck ultrasound revealed a normal size thyroid with normal echotexture without a distinct nodule.

Three months after her surgery, she underwent an ^{123}I whole body scan with SPECT imaging that revealed a sub-centimeter focus of radiotracer uptake along the greater curvature of the stomach with no evidence of pelvic involvement (Fig. 4). On upper abdominal survey during her operation, no disease on the stomach or omentum had been visualized. Thyroglobulin drawn at that time was 62.4 ng/mL (reference range 1.5–38.5 ng/mL) with an undetectable thyroglobulin antibody. Given her stability, reassuring labs, excellent postoperative recovery, minimal disease on post-operative imaging, as well as the low malignant potential of the disease on pathologic evaluation, the decision was made by endocrinology to forgo complete thyroidectomy and RIA and re-evaluate with a whole body scan at one year prior to considering further surgical intervention. The surveillance strategy was implemented utilizing intermittent ^{123}I whole body scans with SPECT-CT and serum thyroglobulin as a marker for worsening or recurrent disease. On six month follow-up, her pain had not recurred and her thyroglobulin was 65.5 ng/mL with an undetectable thyroglobulin antibody. Repeat ^{123}I whole body scan with SPECT imaging at 12 months from surgery no longer revealed the previously visualized radiotracer uptake along the greater curvature of the stomach. There were no further areas of concern.

3. Discussion

We wish to share our conservative method of management of HDFCO presenting greater than ten years after initial struma ovarii resection. The benefits to minimally invasive surgery as compared to open procedures have been well supported with decreased length of hospital stay, decreased blood loss during a procedure, decreased postoperative pain, and faster patient recovery (Mohiuddin and Swanson, 2013). The robotic approach allowed us to better visualize and remove peritoneal thyroid tissue implants with minimal surgical morbidity. Despite her postoperative urinary retention, she had minimal blood loss, was discharged home on postoperative day two, and only required NSAIDs for pain control after discharge from the hospital.

Another unique feature of this case is its delayed presentation. Previous case reports describe HDFCO as the spread of thyroid tissue to the peritoneum found at the time of initial ovarian teratoma resection (Ranade et al., 2015; Karseladze and Kulnitch, 1994). Given the patient's decade-long course between her documented struma ovarii removal and the discovery and resection of her HDFCO, her disease's indolence allowed a conservative approach to treatment, minimizing patient surgical and medical morbidity.

The recommended treatment for HDFCO in the limited literature available has been surgical resection of implants via open abdominal surgeries followed by total thyroidectomy and radioactive iodine to ablate remaining thyroid tissue and prevent later recurrence or metastasis. In thyroid cancer patients, radioactive iodine isotopes are used in post-surgical surveillance imaging as well as an adjuvant for treatment via RIA. The presence of an intact thyroid gland may limit the effectiveness of ablating metastatic thyroid tissue; as such a total

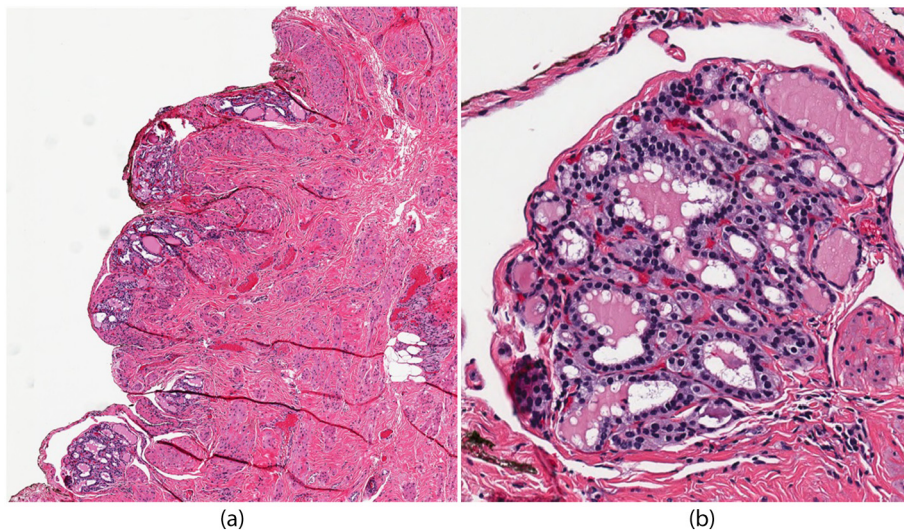


Fig. 3. Microscopic appearance of peritoneal strumosis. a: Variably sized nests of mature thyroid tissue, superficially embedded in the posterior uterine reflection. H&E magnification $\times 40$. b: Benign appearing thyroid follicles lined with cuboidal epithelium, comprised of cells with round nuclei and moderate cytoplasm. Abundant colloid is present within the follicles. H&E magnification $\times 200$.

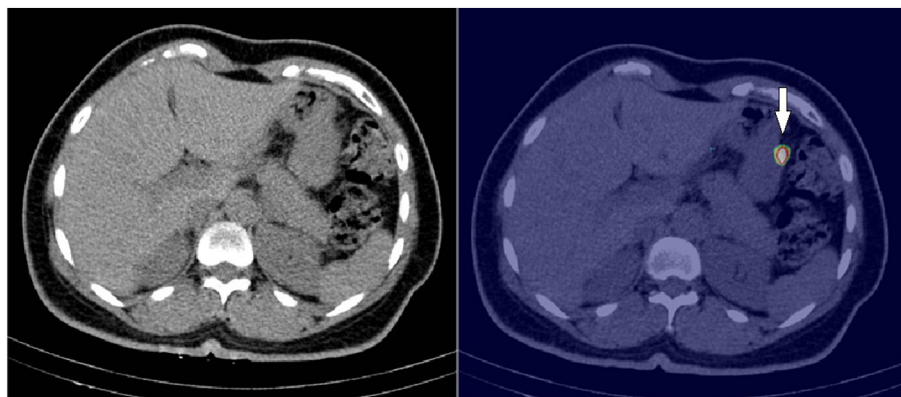


Fig. 4. ^{123}I whole body scan with SPECT imaging: possible mesenteric soft tissue density with associated focal radiotracer uptake adjacent to the greater curvature of the stomach. No RAI uptake elsewhere in the abdomen.

thyroidectomy is generally warranted prior to RIA (Haugen et al., 2016). Thyroglobulin is a glycoprotein molecule synthesized exclusively by thyroid follicular cells and is traditionally measured to detect thyroid cancer recurrence after thyroidectomy. In a recent case series out of Sloan-Kettering Cancer Center, Garg et al. recommended following serum thyroglobulin serially with cross-sectional imaging of the abdomen yearly for several years. Rising thyroglobulin or structural evidence of metastatic disease can be used as indications for more aggressive treatment with total thyroidectomy followed by RIA and further surgical resection if warranted (Garg et al., 2009). A minimally elevated thyroglobulin with a small focus of radioactive iodine avid soft tissue seen on whole body scan may have reflected a small focus of residual HDFCO tissue or microscopic disease, however the stable thyroglobulin level 6 months after surgery was suggestive of an indolent process. Repeat SPECT imaging at 12 months after surgery showed resolution of the gastric area of uptake. Coordinated efforts between gynecologic oncology and endocrinology have allowed conservative management that may serve as a guide to management of similar patients in the future. A less aggressive approach was utilized in this patient due to the indolent nature of the disease process, benign appearance on pathologic review of the specimen, and strong patient desire to avoid additional surgery. The peritoneal involvement seen a decade later from its initial presentation was likely a sequela of the teratoma's prior rupture in 2005. In this case we illustrate a minimally invasive surgical approach combined with conservative medical management that may serve as a guide to management of future cases of HDFCO.

Disclosures

I have no financial interests to disclose or conflicts of interest.

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