

Case Report

A Diagnostic Trifecta Case Report: Struma Ovarii with Pseudo-Meigs Syndrome and Subsequent Ovarian Remnant Syndrome

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Abstract: Struma ovarii is a rare ovarian teratoma composed predominantly of thyroid tissue, representing less than 1% of all ovarian tumors. The clinical presentation of struma ovarii can vary widely, from an asymptomatic palpable abdominal mass, hyperthyroidism (elevated free T4 and/or T3; low TSH), and, in rare cases, pseudo-Meigs syndrome. Diagnosis is often based on postoperative histopathological confirmation of thyroid tissue. We present a case of a 44-year-old Hispanic female with an incidental finding of an adnexal mass, pleural effusion, and ascitic fluid, but with normal tumor markers, cancer antigen 125 (CA-125), and carcinoembryonic antigen (CEA). Postoperative pathological examination confirmed struma ovarii, with pseudo-Meigs syndrome. Unexpectedly, the patient developed ovarian remnant syndrome (ORS). This case contributes to the limited literature describing struma ovarii associated with pseudo-Meigs syndrome and normal tumor markers, highlighting the diverse clinical spectrum of this tumor. It also underscores the need for a high index of suspicion for struma ovarii in atypical cases, regardless of tumor marker levels. The development of ORS further emphasizes the importance of long-term follow-up after definitive surgical management.

Keywords: struma ovarii, ovarian tumor, pseudo-Meigs syndrome, ovarian remnant syndrome

1. Introduction

Struma ovarii is a rare and often underrecognized variant of ovarian dermoid tumors in which thyroid tissue is the major constituent [1, 3, 6]. Struma ovarii was first described in 1889 by Boettcher, who noted thyroid tissue within an ovarian dermoid [2, 3]. Later, in 1899, Gottschalk published a report of an ovarian tumor composed entirely of thyroid-like tissue [3]. Struma ovarii accounts for less than 1% of all ovarian tumors and 2% to 4% of all ovarian teratomas; 5% to 10% are bilateral, and 5 to 10% are malignant [4]. Although struma ovarii contains functioning thyroid tissue, only about 5% of cases produce sufficient thyroid hormones to cause clinical hyperthyroidism [1].

The clinical presentation of struma ovarii is highly variable, ranging from an asymptomatic pelvic or abdominal mass to pelvic pain or abnormal vaginal bleeding. In rare cases, it may be associated with pseudo-Meigs syndrome, characterized by the triad of ascites, pleural effusion, and an ovarian tumor. Laboratory findings can be misleading, as serum tumor markers such as cancer antigen 125 (CA-125) and carcinoembryonic antigen (CEA) may be normal or elevated. Elevated CA-125 levels are frequently reported and may incorrectly suggest ovarian carcinoma. A pelvic ultrasound is necessary based on these findings, and struma ovarii would be indicated by a complex ovarian mass with cystic and solid areas [6]. Computed tomography (CT) or magnetic resonance imaging (MRI) should also be performed to assess the tumor's extent and to screen for possible metastases.

Management of struma ovarii is primarily surgical and may involve unilateral or bilateral oophorectomy using laparoscopic or open approaches, depending on patient age, fertility considerations, tumor characteristics, and concern for malignancy. An added complexity arises when struma ovarii is followed by ovarian remnant syndrome (ORS), a rare condition. Here, residual ovarian tissue remains after oophorectomy and later becomes symptomatic. ORS is most often

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reported in patients with endometriosis, pelvic inflammatory disease, multiple past surgeries, or pelvic adhesions, although its occurrence after struma ovarii is not well characterized in the literature. Patients usually present with chronic pelvic pain, which may be linked to a pelvic mass or found incidentally as an asymptomatic mass [5].

This case report details the diagnostic evaluation, surgical management, and postoperative complications in a 44-year-old Puerto Rican-Hispanic female with struma ovarii and pseudo-Meigs syndrome, normal tumor markers, and subsequent ORS. It highlights the diagnostic and management challenges and underscores the need for long-term postoperative surveillance.

2. Case Presentation

A 44-year-old Hispanic woman, gravida 3 para 3, with a medical history significant for class I obesity (BMI 34kg/m²), hypertension, and iron deficiency anemia presented to her obstetrician-gynecologist following referral from her primary care provider for evaluation of a pelvic mass found incidentally. The patient reported mild, nonspecific abdominal discomfort but denied genitourinary symptoms. Her last menstrual period occurred two weeks prior to presentation, and she reported regular menstrual cycles. Gynecologic history was notable for menarche at age 12; one cesarean delivery at age 18, followed by two vaginal births. Her surgical history included laparoscopic sterilization and incision and drainage of three prior vulvar abscesses.

On presentation, vital signs were notable for elevated blood pressure measuring 165/92 mmHg; the remainder of her vital signs were within normal limits. Abdominal examination revealed moderate tenderness and distension, with surgical scars consistent with her prior operative history. On bimanual pelvic examination, a palpable right adnexal mass was identified. The uterus was anteverted, normal in size and contour, and freely mobile, with no cervical motion tenderness appreciated.

Abdominopelvic CT (Figure 1) revealed a large, 20.5 cm cystic mass with a calcified soft-tissue component extending into the right adnexa, findings that were concerning for ovarian carcinoma. Additional findings included trace right-sided pleural effusion, mild ascites, and soft-tissue thickening along the omentum, raising suspicion for peritoneal carcinomatosis. The combination of an ovarian mass, ascites, and pleural effusion suggested Meigs syndrome. Laboratory evaluation demonstrated normal TSH (1.341 μ IU/mL), T4 (1.37 ng/dL), and human chorionic gonadotropin (hCG) (<2.6 μ IU/mL) levels. Additional tumor markers were also normal: CA-125, 32 ng/mL; CEA, 0.14 ng/mL; AFP, 0.2 ng/mL. Given the concern for malignancy, the patient underwent bilateral salpingo-oophorectomy, appendectomy, omentectomy, and peritoneal washings. Gross specimens (Figure 2) were collected and submitted for histopathological evaluation. The cystic ovarian mass measured 19.5 \times 15.5 \times 5.5 cm and weighed 1,206 g (4.2 lb). Histopathologic examination demonstrated predominant thyroid tissue differentiation, confirming the diagnosis of struma ovarii associated with pseudo-Meigs syndrome.

Four months postoperatively, the patient reported painless vaginal bleeding that had persisted for approximately three months. Physical examination revealed active vaginal bleeding, while bimanual pelvic examination remained unremarkable. Endovaginal ultrasound (Figure 3) demonstrated an anteverted uterus measuring 9.6 \times 6.1 \times 7.1 cm in longitudinal, anteroposterior, and transverse dimensions respectively. A simple cyst measuring 2.2 \times 1.8 \times 2.5 cm was identified in the cul-de-sac, with no evidence of residual ovarian tissue or additional adnexal masses. Laboratory evaluation revealed a normal follicle-stimulating hormone (FSH) level (5.44 mIU/mL) and an elevated estradiol level (166.60 pg/mL), findings suggestive of ongoing ovarian hormonal activity despite prior bilateral salpingo-oophorectomy.

The patient's abnormal uterine bleeding was initially managed with diagnostic hysteroscopy and fractional curettage. Histopathologic examination demonstrated chronic endocervicitis and endometrial hyperplasia without atypia. In the setting of persistent estrogen production and prior oophorectomy, ORS was suspected. Medical management with megestrol acetate was initiated to address hormonal imbalance and endometrial changes; however, the patient continued to experience persistent symptoms along with adverse medication effects. Consequently, a supracervical hysterectomy with peritoneal washing was performed.

Gross surgical specimens (Figure 4) were obtained and submitted for pathologic evaluation. Histopathologic analysis revealed benign residual ovarian parenchyma with evidence of hemorrhagic corpora lutea, confirming the diagnosis of ORS. The patient's postoperative course was uneventful, with complete clinical recovery and no reported complications.

2.1 Images

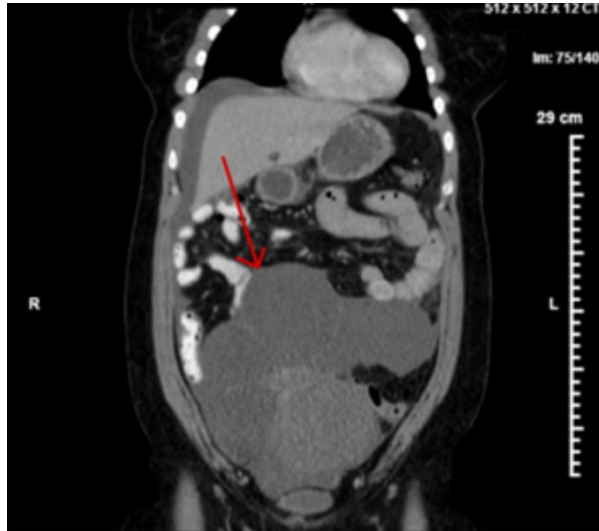


Figure 1. Abdominopelvic CT scan showing a large, 20.5 cm multi-lobulated cystic mass with calcified soft tissue component extending to the right adnexa (red arrow).

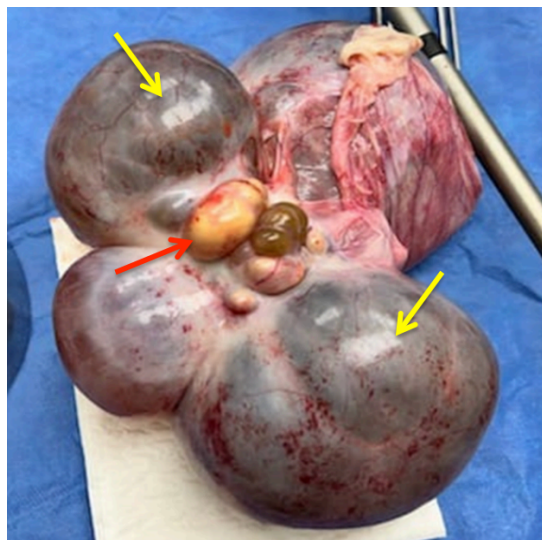


Figure 2. Macroscopic features of the struma ovarii resected surgically. The specimen demonstrates a markedly enlarged, multilobulated ovarian mass with multiple tense, cystic components (yellow arrows). The surface appears soft and gelatinous, with a golden yellow to beefy, reddish-brown color, resembling thyroid tissue (red arrow).

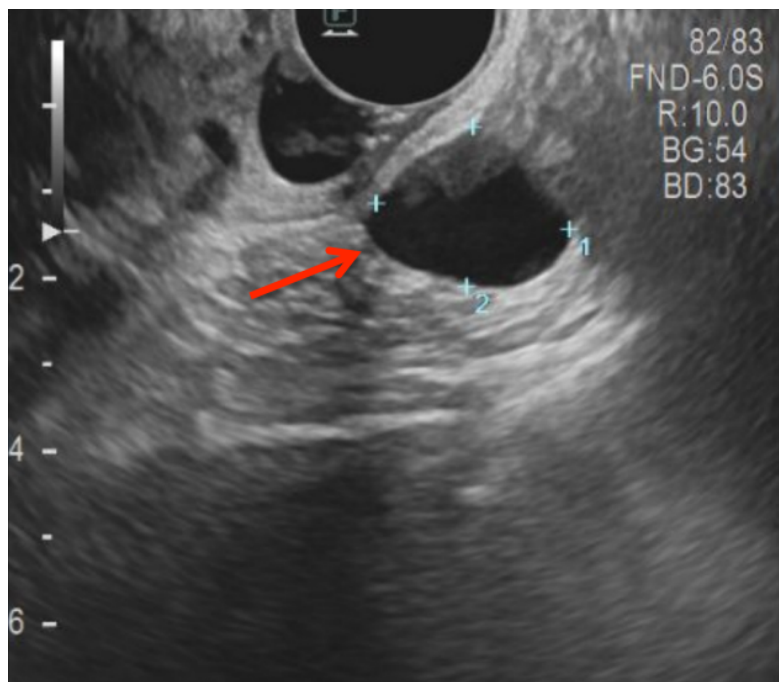


Figure 3: Endovaginal ultrasound showed a simple cyst (red arrow) measuring $2.2 \times 1.8 \times 2.5$ cm was identified in the cul-de-sac region.



Figure 4: Pathologic evaluation of the uterus after hysterectomy revealed a benign, residual ovarian parenchyma with evidence of hemorrhagic corpora lutea, confirming the diagnosis of ORS.

3. Discussion

Struma ovarii is a rare ovarian teratoma characterized by predominant thyroid tissue, accounting for less than 1% of all ovarian tumors [1,3,6]. Despite the high prevalence of thyroid disease, only

about 5% of cases result in clinical thyrotoxicosis [2,4]. Most cases are unilateral, and bilateral involvement is uncommon [4]. This rarity, combined with its heterogeneous presentation, often contributes to diagnostic uncertainty and delayed recognition.

Pseudo-Meigs syndrome—ascites and pleural effusion associated with an ovarian tumor other than fibroma—is a rare but recognized manifestation of struma ovarii [2,5]. As highlighted in this case, the coexistence of ascites, pleural effusion, and a large ovarian mass in the setting of normal tumor markers can closely mimic advanced ovarian malignancy, complicating preoperative diagnosis [3,6]. While CT and MRI may demonstrate a complex multicystic mass lacking the typical fat content of dermoid cysts, definitive diagnosis ultimately relies on histopathological confirmation [6,7].

A particularly unique aspect of this case is the rapid postoperative development of ORS, a rare complication caused by residual ovarian tissue following oophorectomy that typically presents with pelvic pain, mass effect, or persistent hormonal activity [8,9]. Although it is most often associated with endometriosis, extensive adhesions, or multiple prior pelvic surgeries, its occurrence following struma ovarii is rarely described in the literature [5]. In this case, early onset of ORS may have been influenced by technical challenges related to distorted anatomy and adhesions caused by the large ovarian mass [9].

This case underscores the importance of maintaining a high index of suspicion for struma ovarii in patients presenting with complex ovarian masses and atypical features, even in the presence of normal tumor markers. It also highlights the need for meticulous surgical technique and vigilant long-term follow up to promptly identify and manage rare postoperative complications such as ORS. Further research is needed to better elucidate the pathophysiologic relationships among struma ovarii, pseudo-Meigs syndrome, and ORS, and to improve preoperative diagnostic strategies for these uncommon but clinically significant entities [4,7].

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