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Thyroid Tissue Gone Rogue: Malignant Struma Ovarii

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ABSTRACT

Background: Malignant struma ovarii is an exceptionally rare ovarian tumor arising from thyroid tissue within a monodermal teratoma. Papillary thyroid carcinoma is the most common malignant transformation, and management remains controversial due to the absence of standardized guidelines.

Case Presentation: A 34-year-old woman with polycystic ovarian syndrome presented with intermittent lower abdominal pain and was found to have bilateral ovarian cysts. She underwent robotic bilateral ovarian cystectomy at an outside facility. Histopathology revealed an immature teratoma of the left ovary (stage IC, post-cystectomy) and a mature teratoma of the right ovary containing a microscopic 3 mm focus of papillary thyroid carcinoma (infiltrative follicular subtype). Imaging showed no evidence of peritoneal disease or lymphadenopathy. Neck ultrasonography did not reveal any thyroid lesion. Serum tumour markers and thyroid function tests were within normal limits.

Management and Outcome: Following slide review and multidisciplinary evaluation, the patient underwent laparoscopic bilateral salpingo-oophorectomy with omental biopsy, along with total thyroidectomy. Histopathology confirmed malignant Struma ovarii with papillary thyroid carcinoma-type malignant transformation and vascular invasion. In view of the microscopic tumour burden and absence of extra-ovarian spread, radioactive iodine therapy was deferred. The patient was initiated on thyroid hormone replacement therapy and planned for long-term surveillance with serial serum thyroglobulin monitoring.

Conclusion: This case highlights the diagnostic and therapeutic challenges of malignant Struma ovarii. Treatment should be individualized based on disease extent and histopathological risk factors. Long-term follow-up is essential due to the possibility of delayed recurrence.

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Introduction

Struma ovarii is a rare monodermal teratoma characterized by the presence of thyroid tissue constituting at least 50% of the ovarian tumour. Malignant transformation is uncommon and accounts for approximately 0.17–10% of cases. Malignant Struma ovarii represents nearly 1% of mature teratomas and is most frequently diagnosed in premenopausal women aged between 30 and 50 years. The varied and subtle presentation of malignant Struma ovarii creates a complex diagnostic dilemma, often leading to it being mistaken for ovarian cancer until after histological examination. Unlike its benign counterpart, malignant Struma ovarii necessitates strict monitoring and active treatment due to its potential for metastasis.

Case Summary

A 34-year-old woman with polycystic ovarian syndrome, gravida 3 para 2 living 2 abortion 1, presented with intermittent lower abdominal pain for two months. She had no known comorbidities or any significant family history of malignancy. She had a history of laparoscopic surgery 18 years earlier for a suspected dermoid cyst.

Since her initial ultrasonography revealed bilateral ovarian cysts, she was further evaluated with MRI and tumour markers and underwent robotic bilateral ovarian cystectomy at an outside facility. Histopathology revealed immature teratoma of the left ovary (stage IC post cystectomy) and mature teratoma of the right ovary with a 3 mm focus of malignant transformation into papillary thyroid carcinoma (infiltrative follicular subtype).

On examination, the patient had ECOG performance status 0. Thyroid examination was normal, with no palpable nodules. Abdominal examination revealed no palpable masses. Port scars were healthy. Per speculum examination showed a healthy cervix, and bimanual examination revealed a bulky uterus and no adnexal masses.

A slide review was performed. Contrast-enhanced CT abdomen and pelvis showed bilateral adnexal cystic lesions with enhancing septations measuring 5 × 3.5 cm on the left and 4.5 × 3.6 cm on the right, with no peritoneal deposits, omental deposits, or lymphadenopathy. Neck ultrasound showed no thyroid lesions.

Laboratory evaluation showed CEA 1.1 ng/mL, CA-125 22.7 U/mL, β-hCG <2 mIU/mL, AFP 1.2 ng/mL, CA 19-9 51.9 U/mL, TSH 1.53 mIU/mL, and thyroglobulin 32.75 ng/mL (normal range).

The patient underwent laparoscopic bilateral salpingo-oophorectomy with omental biopsy and total thyroidectomy under general anaesthesia. The postoperative period was uneventful.

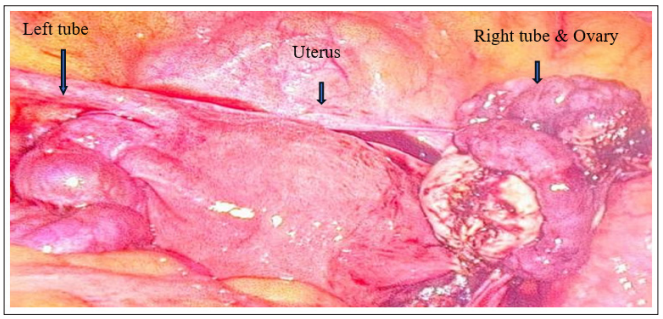

Figure 1: Intraoperative laparoscopic Picture Showing Right Ovary Which Harboured the Malignant Struma Ovarii

Histopathological evaluation revealed thyroid tissue arranged in an infiltrative microfollicular pattern with enlarged vesicular nuclei and scattered nuclear grooves. Tumour was encasing a nerve bundle and extending into the lumen of a blood vessel through its muscular wall, consistent with vascular invasion. These features supported the diagnosis of malignant struma ovarii with papillary thyroid carcinoma-type malignant transformation.

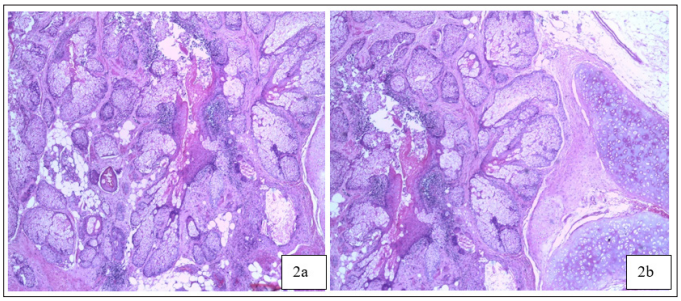

Figure 2 a, b: Low Power Sections Show Mature Teratomatous Elements Including Hyaline Cartilage and Abundant Thyroid Tissue Arranged in Lobules Separated by Fibrous Septae, Consistent with Struma Ovarii.

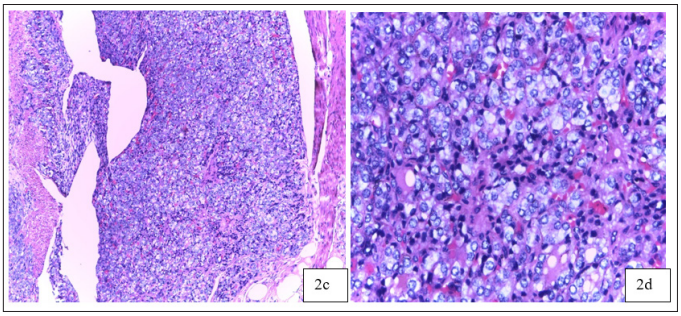

Figure 2c: Intermediate - A Focal Area Shows Infiltrative Proliferation of Thyroid-Type Epithelium with Solid and Microfollicular Architecture, Suspicious for Malignant Transformation.

Figure 2d: Tumour Cells Show Enlarged Optically Clear Nuclei with Overlapping, Nuclear Grooves, and Irregular Nuclear Contours, Consistent with Papillary Thyroid Carcinoma-Type Nuclear Features.

Given the microscopic tumour focus and absence of extra-ovarian spread, radioactive iodine therapy was not administered. The patient was initiated on thyroid hormone replacement therapy and planned for long-term surveillance with serial thyroglobulin monitoring.

Discussion

Struma ovarii is a rare ovarian neoplasm, with an estimated incidence of malignant transformation occurring in <1 case per 10 million women annually [1]. The literature shows that malignant Struma ovarii is typically reported as case studies or small, single- centre case series, indicating it is a significant medical rarity. Due to the same reason, there are no universally accepted management guidelines, and treatment is individualized based on tumour characteristics and extent of disease

Malignant Struma ovarii has been categorized into 4 types: (1) papillary carcinoma thyroid -like; (2) proliferative malignant resembling follicular adenoma; (3) highly differentiated follicular carcinoma with extragonadal spread and (4) histologically malignant with high-grade features. Genetic studies reveal that malignant struma ovarii shares oncogenic activations with primary thyroid carcinoma including the BRAF, KRAS, and RET genes. TERT promoter pathogenic variants have also been demonstrated and appear to be associated with more aggressive biological behaviour, similar to that observed in ectopic thyroid malignancies [2].

Malignant struma ovarii diagnosis is challenging due to its nonspecific symptoms and lack of distinct imaging findings [3]. 60-70 % cases are unintentionally discovered during surgery or imaging procedures carried out for unrelated purposes. 17- 35 % cases present with symptoms like abdominal pain, bloating, pelvic mass, urinary symptoms, abnormal uterine bleeding, or ascites. Thyrotoxicosis symptoms are uncommon and only appear in approximately 5–8% of cases [4].

Preoperative suspicion of malignancy is extremely challenging, as radiological findings often overlap with those of benign and malignant adnexal tumours and lack specific distinguishing features. Additionally, serum tumour markers are frequently non-contributory [5,6].

As a result, histological analysis of the surgical material is typically required to confirm the diagnosis of malignant struma ovarii [7]. Papillary thyroid carcinoma is the most commonly reported malignant subtype accounting for 50–70%, whereas follicular carcinoma is less prevalent which is around 20–30%. Rare variants include Hurtle cell carcinoma, poorly differentiated carcinoma, and anaplastic carcinoma. Papillary formations with fibrovascular cores, nuclear expansion, overlapping, nuclear grooves, and intranuclear cytoplasmic inclusions are typical characteristics of papillary thyroid cancer. As seen in this case, a follicular variation of papillary carcinoma may have microfollicular architecture while maintaining diagnostic nuclear characteristics.

Thyroid cancer and differentiation are supported by immunohistochemistry TTF-1 and thyroglobulin are helpful in confirming thyroid tissue [8]. Papillary thyroid cancer may be confirmed with the help of CK19, HBME-1, galectin-3, and CD56. Thyroid imaging and clinical correlation are necessary because immunohistochemistry cannot distinguish between original ovarian thyroid carcinoma and metastatic thyroid carcinoma.

Thyroidectomy may be considered in patients with synchronous thyroid carcinoma or suspicious thyroid nodules, aggressive ovarian histology (poorly differentiated carcinoma, capsular/vascular invasion, or anaplastic components), extra-ovarian spread, large tumors (>4–5 cm), or elevated postoperative Tg/TgAb levels. In order to rule out occult thyroid primary, permit

thyroglobulin surveillance, and make whole-body iodine screening possible in the future, a total thyroidectomy was carried out in this case. An estimated prevalence of Cervical thyroid cancer prevalence in MSO patients is about 2.6-8.8% of cases. Most co-existing lesions are considered as independent primaries rather than metastatic disease.

Among the case reports and series, five studies (Garg et al 2009, Goffredo et al 2015, Addley et al 2021, Ryu et al 2023, Ayketin et al 2025) reporting thyroid surgery along with laparotomy or laparoscopy (n = 121), thyroidectomy was performed in 22 patients (18.2%) and RAI therapy was administered to 13 patients (10.7%) [9].

The role of radioactive iodine therapy is debated and generally reserved for patients with metastatic disease, residual tumour, recurrence, or high-risk pathological features [10]. Post-thyroidectomy radioiodine ablation treats micrometastases (uptake seen in pelvic remnants or distant sites like liver), reducing recurrence risk from 35% without adjuvant therapy to 6% and yields more than 94 % 5 year disease free survival in metastatic cases after multiple doses. Due to the low disease load and lack of extra-ovarian dissemination in the present case, radioactive iodine was deemed unnecessary [11].

Long-term surveillance is crucial since relapses might happen after a protracted period of no disease. Clinical assessment, radiographic monitoring, and serial serum thyroglobulin measurements are typical forms of follow-up. Tumour dedifferentiation, often known as the "switch phenomenon," is a major worry during follow-up It occurs when dedifferentiation of differentiated thyroid cancer occurs [12,13]. This leads to radioactive iodine resistance by losing their capacity to concentrate iodine due to decreased sodium-iodide symporter activity (NIS). The "flip-flop" pattern on 18F-FDG PET/CT in metastatic malignant struma ovarii is typically observed during follow-up, particularly after radioiodine therapy, and reflects an inverse relationship between iodine avidity and glucose metabolism, wherein lesions that lose radioiodine uptake may demonstrate increased FDG avidity [14].

The use of external beam radiation is restricted, primarily in cases of palliative care or incurable illness. In progressive metastatic radioactive iodine-refractory disease, tyrosine kinase inhibitors like Sorafenib and Lenvatinib14 may be taken into consideration.

Fertility options for young people undergoing bilateral salpingo-oophorectomy include adoption, surrogacy, and IVF using donor oocytes. To lessen the morbidity linked to early menopause, hormone replacement treatment should be taken into consideration.

Conclusion

Malignant struma ovarii is an exceptionally rare ovarian tumour, often diagnosed only after histopathological examination. Surgical management remains the mainstay of treatment. Total thyroidectomy is useful for confirming ovarian origin and enabling thyroglobulin-based surveillance. Radioactive iodine therapy may be reserved for selected cases with high-risk or disseminated disease. Given the potential for late recurrence and dedifferentiation, long-term follow-up is mandatory.

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