

## Well-differentiated papillary mesothelioma found incidentally with concurrent struma ovarii: A case report

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### ABSTRACT

This is the first report of well-differentiated papillary mesothelioma (WDPM) found concurrently with struma ovarii. These rare tumors have no known association, and are considered benign, though malignant transformation of WDPM has been described. After treatment of WDPM, follow-up surveillance has been suggested in the literature, though a method has not been described and an evidence base is lacking. Pre-operative imaging by computed tomography and ultrasound did not identify the WDPM, calling into question the role of imaging-based follow-up. Furthermore, malignant transformation has been reported only outside of the typical 5-year follow-up window. The authors of this case report suggest rationalization of follow-up of WDPM, acknowledging the risks and unknown harms of scheduled imaging follow-up.

### 1. Introduction

Well-differentiated papillary mesothelioma (WDPM) is a rare mesothelial neoplasm, with approximately 180 cases reported [1]. Most commonly found incidentally during pelvic surgery in women of reproductive age, WDPM is usually peritoneal or pleural [1], but has been identified in the tunica vaginalis [2], epididymis [3], and pericardium [4].

Peritoneal WDPM may be uni- (41%) or multi-focal (59%), and, unlike malignant mesothelioma, is unrelated to asbestos exposure [1]. It is thought to have low malignant potential, though occasionally includes invasive foci, and there are reports of malignant transformation [5]. Treatment with excision biopsy alone may suffice, though for multifocal disease, radical surgery, chemotherapy, and radiotherapy have been described. Recurrence is rare, occurs at the original site, is surgically excisable, and is associated with disseminated disease at the time of original diagnosis [1,5,6]. Unifocal WDPM does not appear to recur [5].

Struma ovarii (goitre of the ovary) is a rare mature teratoma composed predominantly of thyroid tissue; it accounts for 0.5–1% of ovarian tumors and 2.7% of mature teratomas [7,8]. Up to one-third of cases have concomitant ascites and 5% are associated with hyperthyroidism. Most are benign, and where thyroid-type carcinoma exists, it rarely spreads beyond the ovary.

This case report describes the finding of two rare neoplasms during

laparoscopic salpingo-oophorectomy for a left ovarian mass, and considers the challenges of following up rare neoplasms with an unknown ratio of benefit to harm.

### 2. Case presentation

A 28-year-old Māori/New Zealand European woman was referred to gynecology with 18 months of cyclical pain in the left iliac fossa, described as most severe in the week prior to menstruation. There was a two-year history of persistent microscopic hematuria, for which laboratory investigations were unremarkable. No history of chemical or asbestos exposure, nor neoplasm in first- or second-degree relatives could be recalled. The patient used a combined oral contraceptive for menstrual regulation, and regularly smoked. Physical examination was non-contributory.

Initial radiological evaluation by computed tomography (CT) of the urinary tract demonstrated a 70 mm multicystic left adnexal mass with a small volume of free pelvic fluid. There were no urinary tract calculi nor other abnormalities. Pelvic ultrasound reported three focal lesions within the ovary: one was consistent with mature teratoma; another was solid and granular; and another was heterogenous and cystic. There was also a 9 mm endometrial polyp with feeder vessel. The patient's CA-125 of 14 kIU/L was in the normal range, as was her thyroid stimulating hormone serum concentration of 1.4 mIU/L.

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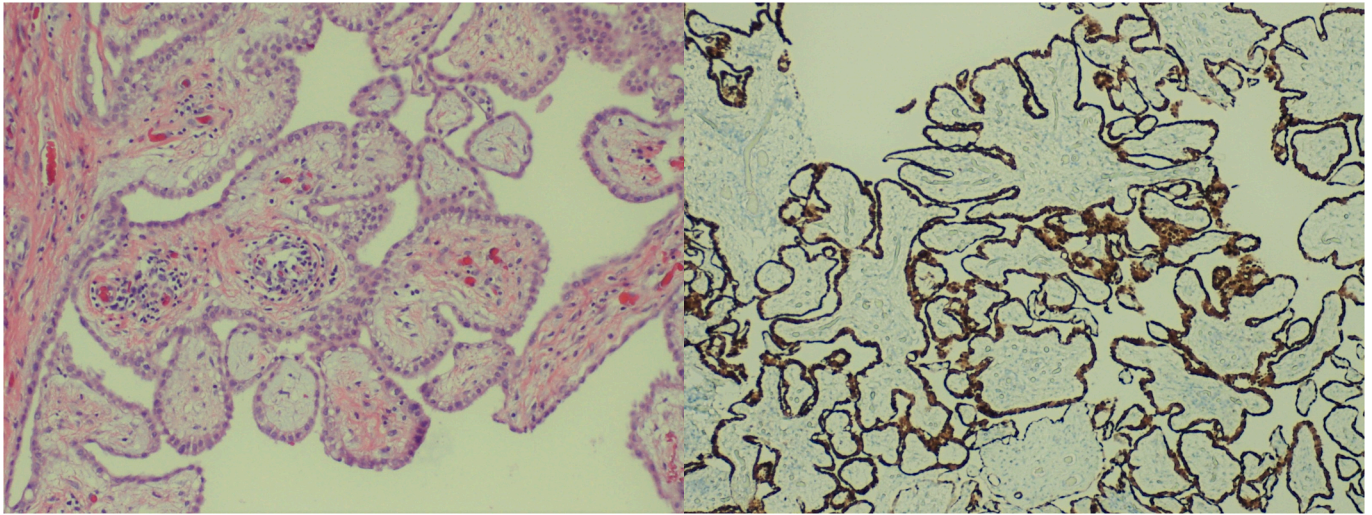
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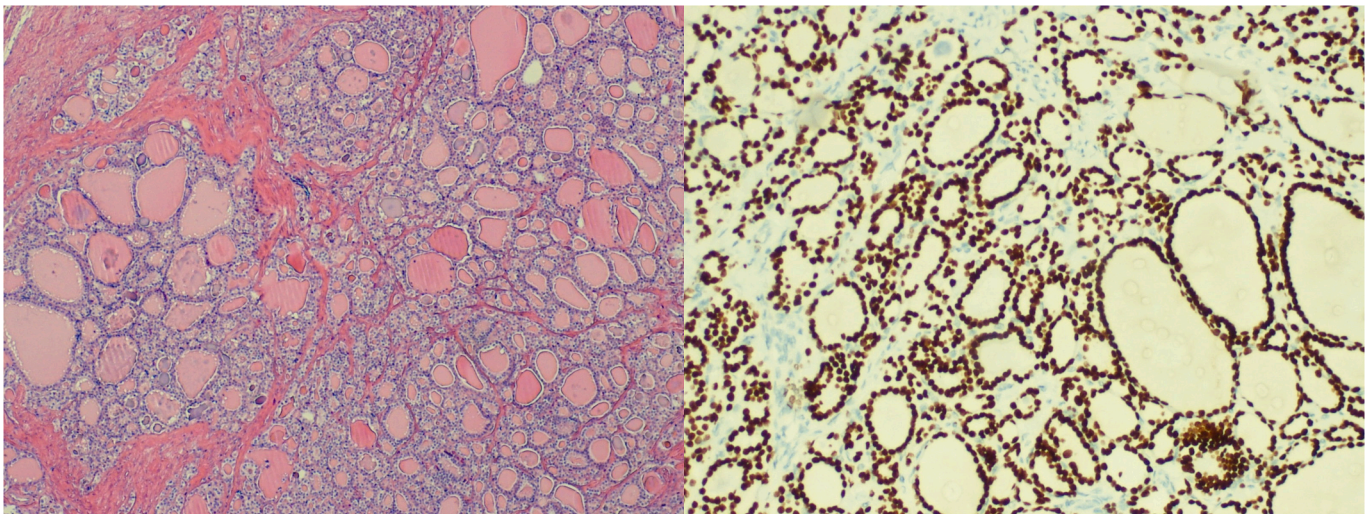
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**Fig. 1.** Hematoxylin and eosin stain medium power 100 $\times$  (left panel), and calretinin immunohistochemistry low power 40 $\times$  (right panel), bowel peritoneum well-differentiated papillary mesothelioma.



**Fig. 2.** Hematoxylin and eosin stain low power 40 $\times$  (left panel), and thyroid transcription factor-1 immunohistochemistry medium power 100 $\times$  (right panel) struma ovarii.

At laparoscopy, the left ovary had a smooth and bulky appearance. The rectovaginal pouch contained a small volume of green fluid. An isolated 3 cm pink sea-anemone-like structure attached to the sigmoid peritoneum was found incidentally, adjacent to the left ovarian surface, but seemingly unrelated. This was excised with a margin of peritoneum for histologic evaluation. The remainder of the pelvic and abdominal organs and peritoneum appeared normal. Hysteroscopy was unremarkable other than for a small endometrial polyp, which was excised.

Microscopically, the peritoneal lesion had well-defined papillary structures, and benign papillary mesothelial proliferation consistent with WDPM (Fig. 1). There were no cytological atypia or infiltrative features. Immunohistochemical staining demonstrated calretinin and CK 5/6 positivity within the mesothelial lining (Fig. 1).

The excised ovary demonstrated a struma ovarii consisting of benign thyroid tissue, mature fibroadipose connective tissue, and a variety of epithelia (Fig. 2). The fallopian tube was normal. Cytological evaluation of the ascites found mesothelial cells and chronic inflammatory exudate. Ascitic microbiology was unremarkable.

Retrospective review of pre-operative imaging was unable to visualize the WDPM.

At 6-week post-operative follow-up, the patient reported an uncomplicated recovery and resolution of the pelvic pain. The incidental finding of unifocal WDPM was discussed, along with the uncertainty of the natural history of the disease reported in the literature. A shared decision was made not to perform routine follow-up, but instead reassess in the event of pelvic or abdominal symptoms. A post-operative staging magnetic resonance imaging scan and transvaginal pelvic ultrasound scan showed no evidence of residual disease.

Written consent was provided for publication of this case report.

### 3. Discussion

Understanding of WDPM is evolving, but remains immature, with the largest series of 75 cases only recently reported [1]. Follow-up of WDPM has been suggested due to a risk of recurrence and malignant transformation, though this rare outcome has been reported only 7 to 15 years following primary treatment, and the effectiveness of any follow-up methodology is undemonstrated [1,5,9]. Furthermore, the 3 cm WDPM in this case was not visible on ultrasound or CT, so an expectation that scheduled follow-up imaging might be effective does not appear to



have foundation. There is a paucity of description of radiologically detected isolated WDPM in the literature.

Post-treatment surveillance should have the ability to detect recurrence and impact survival outcomes. There appears to be no evidence that surveillance following treatment of WDPM achieves these requirements. Furthermore, there are no reports of non-invasive unifocal WDPM recurring following excision biopsy [5,6]. Any theoretical benefit must be balanced against physical and psychological patient harms, and resource overuse. Physical harm associated with investigation, without corresponding mortality benefit, has been demonstrated by study of ovarian cancer screening, and associated anxiety is unquantified [10].

Benign struma ovarii require no extended follow-up [8]. There are no prior reports of struma ovarii with WDPM, nor a suggested relationship. Their co-existence in this case is considered to be an incidental finding.

The authors conclude this to be a case of benign, fully resected, unifocal disease, with no indication for scheduled follow-up. The role of post-operative cross-sectional imaging, to detect additional peritoneal disease, in the context of otherwise normal laparoscopic findings, is unknown, and in this case it did not detect any abnormality. Given that WDPM is usually diagnosed in the young and healthy, any unknown benefit of follow-up must be balanced against anxiety, harm, cost, and low risk of recurrence. Management must therefore be individualized, holistic, and planned together with a well-informed patient.

#### Contributors

Sam Holford was directly involved in the patient's care, contributed to the case report's conception and design, and to drafting and editing of the manuscript.

William Viner contributed to the case report's conception and design, critically revised the manuscript for important intellectual content, and was directly involved in the patient's care.

James Hunt contributed to the drafting of the manuscript, and was directly involved in the preparation and interpretation of the patient's histology.

All authors approved the final draft of the report.

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#### Patient consent

Written informed consent was obtained from the patient for publication of this case report.

#### Provenance and peer review

This article was not commissioned and was peer reviewed.

#### Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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