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Case Report

A case of ovarian serous borderline tumors with normal ovarian findings on preoperative imaging

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ABSTRACT

Ovarian serous borderline tumors (SBTs) are typically diagnosed through imaging techniques such as ultrasound and magnetic resonance imaging (MRI); however, diagnosis can be challenging when the tumor exhibits unusual growth patterns or is of small size. We report the case of a 55-year-old woman in whom bilateral SBTs were discovered during surgery, although preoperative imaging showed no ovarian abnormalities. The patient underwent laparoscopic total hysterectomy and bilateral adnexectomy for uterine fibroids. Preoperative MRI and transvaginal ultrasound did not reveal any ovarian abnormalities, but during laparoscopic surgery, small nodules growing outward were observed on both ovaries. Pathological examination led to the diagnosis of a serous borderline tumor with micropapillary pattern (SBT-MP). This case highlights the limitations of preoperative imaging in detecting small serous borderline tumors with surface papillary growth, as no abnormalities were observed preoperatively despite bilateral ovarian involvement. When an ovarian tumor exhibiting surface growth is identified during surgery, even if it is too small to be detected preoperatively, it is crucial to consider the possibility of a borderline tumor. In such cases, it is essential to perform pathological examination, including intraoperative frozen section analysis, and conduct a thorough macroscopic inspection of the abdominal cavity.

Keywords: Serous borderline tumor, Micropapillary pattern, Surface papillary type, Laparoscopic surgery

INTRODUCTION

Serous borderline ovarian tumors (SBTs) account for 10-15% of all serous ovarian tumors and are clinically considered low-grade malignant tumors.¹ Typical SBTs present as relatively large, unilocular to multilocular cystic tumors with papillary or granular protrusions within the lumen, which can often be easily diagnosed through imaging techniques such as ultrasound or magnetic resonance imaging (MRI).² However, in rare cases, serous borderline tumors may present as solid tumors without forming cystic lesions, displaying an outward growth pattern known as surface papillary serous borderline tumors (SSPBT).³ This type of tumor differs from the typical form and can make preoperative imaging diagnosis challenging. In particular, small serous borderline tumors

with a surface growth pattern may be difficult to detect on imaging, creating a diagnostic challenge and highlighting the importance of careful intraoperative assessment. In the present case, we encountered a serous borderline ovarian tumor that was not identified on preoperative imaging and showed no enlargement apparent of the ovary.

CASE REPORT

The patient was a 55-year-old woman, G3P2, who had not yet reached menopause, but had been experiencing infrequent menstruation for the past few years. There was no significant medical or family history. During a routine health check-up, uterine fibroids were detected, and the patient consulted a previous physician. Although no increase in size was observed during one year of follow-

up, the patient was referred to our hospital for surgical treatment due to a sense of abdominal pressure. At the initial visit, cervical cytology was negative. Transvaginal ultrasound identified an 8.5 cm mass in the uterine body, but the adnexa were not clearly visualized. No ovarian enlargement or ascites was noted. MRI revealed multiple fibroids, the largest measuring 8.3×7.1 cm within the myometrium. Both ovaries appeared normal in size (Figure 1).

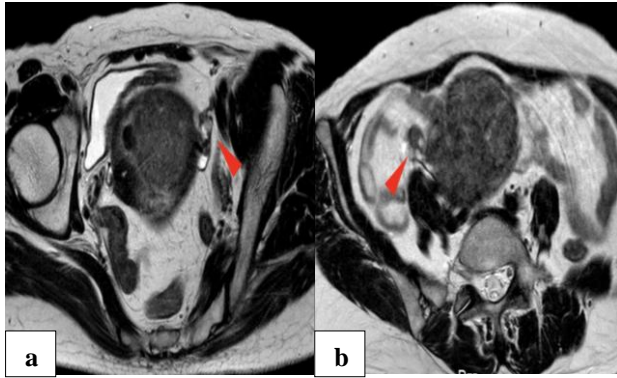


Figure 1: T2-weighted MR images: both ovaries are normal in size, and show low to intermediate signal intensity on T2-weighted images, with no diffusion restriction (a) left ovary, and (b) right ovary.

Preoperative blood tests showed follicle-stimulating hormone (FSH) of 39 mIU/ml and estradiol <10 pg/ml, indicating a state close to menopause. A decision was made not to preserve the ovaries in accordance with the patient's preference. Based on a diagnosis of multiple uterine fibroids, a laparoscopic total hysterectomy and bilateral adnexectomy were performed. During surgery, multiple white nodular lesions were observed on the surface of both ovaries. No other suspicious findings of malignancy, such as ascites or peritoneal involvement, were observed (Figure 2).

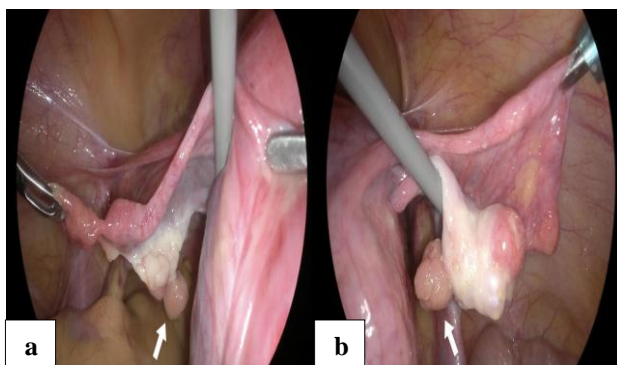


Figure 2: Laparoscopic findings: small solid lesions were found on the surface of both ovaries (a) left ovary, and (b) right ovary.

The uterus with adnexa was carefully retrieved transvaginally to avoid damaging or fragmentation for following pathological evaluation. Histopathological

examination confirmed the presence of benign leiomyomas in the uterus. Tumors measuring 18×8 mm on the left ovary and 10×8 mm on the right ovary were identified, both exhibiting similar histological features. These tumors showed micropapillary structures with surface growth and mild nuclear atypia. Psammomatous calcifications were scattered within the tumor lesions, but no evidence of destructive stromal invasion was observed (Figure 3).

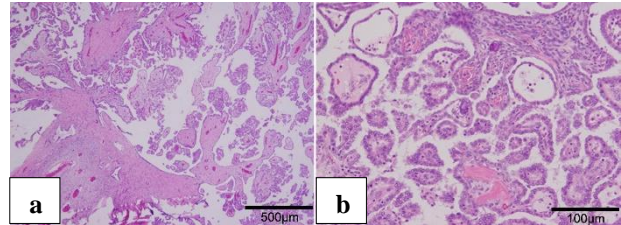


Figure 3: Representative images of H&E staining: the tumors exhibited a micropapillary structure and grew outward, exposing part of the tumor to the peritoneal cavity (a) x40, and (b) x200.

Immunohistochemistry revealed that the tumor cells were positive for PAX-8, WT-1, and estrogen receptor (ER), while p53 was wild-type, and Ki-67 was low at 10%. Based on these findings, a diagnosis of serous borderline tumor with micropapillary pattern (SBT-MP) was made. The patient recovered without complications. At an outpatient visit on postoperative day 15, serum tumor marker levels were normal and CT showed no evidence of metastasis. At 12 months post-surgery, the patient had no recurrence and remains under careful follow-up.

DISCUSSION

Serous epithelial tumors are classified into three patterns based on their growth morphology: cystic, surface papillary, and stromal proliferation types. The surface papillary type is a growth pattern specific to serous tumors, characterized by outward development from the ovarian surface and the formation of papillary nodules. This growth pattern can occur in benign, borderline malignant, and malignant tumors, but it is rare, accounting for approximately 2% of all ovarian tumors.^{1,3}

In the present case, small surface papillary lesions were observed in both ovaries. Although the tumors were small and initially judged to be benign during surgery, subsequent histological and immunohistochemical analysis revealed that the tumors were SBT-MP.

According to previous reports, SSPBT is often depicted as a solid mass, limited to the ovarian surface, with normal ovarian size and shape. Ludovisi et al reported that in five cases of SSPBT, ultrasound imaging revealed solid tumors measuring 4–8 cm, adjacent to the normal ovary.⁴ Furthermore, these tumors may show characteristic findings on MRI. Tanaka et al found that all six cases they studied demonstrated papillary structures with leaf-like

projections, resembling a sea anemone, and a branching pattern on MRI.⁵

The histological feature of this case, SBT-MP is characterized by the presence of continuous areas with micropapillary or cribriform morphology, spanning at least 5 mm, which suggests a tendency for outward growth. SBT-MP represents 5-15% of all SBTs and is considered a prognostically distinct variant of SBTs, differentiating it from the typical form of SBTs.^{6,7}

Clinically, SBT-MP is characterized by a higher incidence of bilateral involvement, outward growth, and peritoneal implants compared to conventional SBTs. However, there are no significant MRI findings that can distinguish between the two types.⁸

In the current case, no abnormalities were detected in the ovarian morphology or size on preoperative imaging, and the tumor was not identified by ultrasound or MRI due to its small size. The tumor was only discovered during surgery as small nodules with outward growth. The difficulty in detecting the ovarian tumors preoperatively may have been due to several factors. First, transvaginal ultrasound may have been limited by the presence of a large uterine fibroid, obstructing direct visualization of the adnexa. Second, MRI, while useful for detecting larger cystic lesions, may not have had enough contrast resolution to identify small surface papillary tumors, particularly in the absence of significant ovarian enlargement or peritoneal involvement. This case highlights the limitations of preoperative imaging. Since the discovery of borderline malignant tumors in ovaries of normal size is rare, SSPBT, which often present with normal ovarian morphology, are likely to be more difficult to diagnose early compared to typical SBTs. As demonstrated in this case, when a small ovarian surface lesion is unexpectedly identified intraoperatively, SSPBT should be considered. In such cases, intraoperative frozen section analysis is recommended to guide immediate surgical decision-making. Furthermore, although no postoperative adjuvant therapy was required in this case, long-term follow-up remains essential due to the potential for recurrence, particularly in micropapillary variants.

The sensitivity of intraoperative rapid diagnosis in distinguishing serous borderline tumors from benign tumors has been reported to range from 48% to 79%, which is lower than the diagnostic sensitivity of general intraoperative frozen section (IFS) analysis (89.8%).^{9,10} Recent studies have shown that combining DNA microarray technology with machine learning models targeting SNTN or AOX1 can increase the accuracy of rapid diagnosis of serous borderline tumors to 97.3%.⁹

SSPBT tends to have a higher incidence of bilateral onset, peritoneal implants, presence of tumor cells in peritoneal lavage fluid, and increased serum CA125 levels compared to typical SBTs. However, no significant differences in

recurrence rates or disease-free survival (DFS) have been observed.^{5,11}

The recurrence rate of SBTs is approximately 4.9%, which is relatively low. However, patients undergoing fertility-sparing surgery (FSS) have a higher risk of recurrence.¹² Among patients with advanced-stage (stage II/III) SBTs, 44–58% of those who underwent FSS experienced recurrence.^{13,14} Moreover, the presence of invasive implants increases the risk of recurrence by 2–4 times.¹⁵ SBT-MP has a higher frequency of invasive implants and a greater risk of recurrence, with DFS dropping to 75.9% after 3 years.^{16,17} Furthermore, SBT-MP is known to recur as low-grade serous carcinoma (LGSC). In particular, in advanced-stage (stage II/III) SBTs with micropapillary pattern have been reported to carry a higher risk of progression to LGSC than those without it.¹⁸

These findings suggest that both preoperative imaging for staging and complete tumor resection, along with a thorough assessment of disease spread within the abdominal cavity, are crucial for accurately evaluating SBTs, particularly micropapillary SBTs. This approach will significantly contribute to improving patient prognosis.

In this case, even after reviewing surgical footage retrospectively, no clear signs of ascites or peritoneal implants were observed. However, pathological findings revealed that the tumor was exposed to the peritoneal cavity, indicating that adequate exploration of the peritoneal cavity for staging was not performed. Fortunately, the patient has remained free of recurrence for 12 months postoperatively. Nevertheless, even in surgeries not aimed at treating ovarian tumors, when an ovarian tumor showing outward growth is discovered intraoperatively, it is important to consider the possibility of a borderline malignant or more malignant tumor, even if it is too small to be identified preoperatively. In such cases, assessing for extra-ovarian disease intraoperatively, performing rapid intraoperative diagnosis, and conducting cytological examination of ascitic fluid or peritoneal lavage are critical.

CONCLUSION

This case highlights the importance of considering SBTs in cases where small, outward-growing ovarian tumors are detected intraoperatively, even if preoperative imaging shows no abnormalities. Despite a normal ovarian appearance on preoperative imaging, the presence of small surface tumors requires careful intraoperative assessment. Given the limitations of current imaging techniques, further research is needed to develop better diagnostic methods, including the potential role of advanced MRI techniques and intraoperative fluorescence imaging, to improve the detection of small SBTs and enhance patient management. Careful intraoperative assessment and timely pathological evaluation remain essential for the successful management of SBTs.

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