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Letter to the Editor

A case of paraovarian serous cystadenofibroma that showed a malignant potential in image inspection



Introduction

A serous cystadenofibroma is a relatively rare ovarian epithelial tumor.¹ Only one case of paraovarian serous cystadenofibroma has been reported.² The appearance of a cystadenofibroma on imaging is often complicated; cystic- to solid-appearing masses may be visualized, and it often resembles a malignant tumor.^{3–5} We herein present a case of paraovarian serous cystadenofibroma that could be treated by laparoscopy. Written informed consent was obtained from the patient for publication of this case. The local Institutional Review Board exempted our case from the need for approval.

Case Report

A 25-year-old woman was referred to our hospital for a paraovarian tumor with a small solid component.

Her height was 158 cm, and she weighed 73 kg (body mass index: 29.2 kg/m²). The uterus was normal and the bilateral adnexa were not palpable. The tumor markers were all in the normal range (CA125: 13 U/mL; CA19-9: 18 U/mL; and Carcinoembryogenic antigen (CEA): 1.3 ng/mL). Transvaginal ultrasonography (USG, Figure 1A) and contrast-enhanced pelvic magnetic resonance imaging (MRI, Figure 1B) revealed a 30-mm cystic mass near the left ovary with a thickened wall and a small solid component, but there was no contrast enhancement or high diffusion weighted imaging signal on MRI. The appearance was not clearly malignant, and we finally diagnosed a paraovarian cyst. Three months later, the size of the tumor reached 40 mm. We performed 18F-fluorodeoxyglucose positron emission tomography/computed tomography (FDG-PET/CT). This showed a weak positive uptake of FDG [maximum standardized uptake value (SUVmax) = 3.5] at the thickened wall segment (Figure 1C). It was difficult to exclude the possibility of a malignant or borderline malignant tumor. However, this tumor appeared to be paraovarian, so we performed laparoscopic tumor excision.

Laparoscopy revealed an approximately 40-mm left paraovarian tumor with a small nodule on the surface (Figure 2A). The tumor was adherent to the left tube, ovary, and mesentery. The tumor was removed with the left fallopian tube without rupture after exfoliation.

On macroscopic examination, there were small papillary structures on the surface and inside the tumor (Figure 2B). Microscopic

examination revealed a unilocular cyst lined by a single layer of tall, columnar, ciliated cells. The stroma contained spindly fibroblasts. There was no atypia or architectural complexity. Accordingly, the tumor was finally diagnosed as a paraovarian serous cystadenofibroma (Figure 2C).

Discussion

Ovarian cystadenofibroma is a surface epithelial tumor that contains fibrous components. It is classified into serous, endometrioid, mucinous, clear cell, and mixed categories, according to the epithelial cell type. The serous type is the most common (75%). The susceptible age for this tumor is between 40 years and 50 years, and it is rare at younger ages.^{1,2}

It is often diagnosed preoperatively as a malignancy because of a solid component or irregular thick septa. Preoperative USG appearance of ovarian cystadenofibroma has been reported.^{3,4} Cystadenofibroma is cystic and thin walled, and contains simple cysts with solid nodules. Septation appears in 30.4%, and solid nodules in 56.5–80%^{3,4} of the cases. The most frequent appearance is a unilocular, complex cystic tumor (43.4%). The usefulness of color Doppler sonography to evaluate the vascularity of the cyst wall, septum, or solid component has been reported.³

The diagnosis of cystadenofibroma using CT has also been reported.¹ Sixteen cases of cystadenofibroma that were diagnosed pathologically after surgery had all been diagnosed as malignant or borderline malignant tumors by preoperative examination using CT, because of a solid component or wall thickening.¹

In this case, we could not exclude the possibility of malignancy or borderline malignancy by USG, CT, and MRI. Thus, we performed FDG-PET/CT. Recently, the usefulness of FDG-PET/CT for differentiating ovarian cancer from borderline or benign tumors has been reported.^{6–8} The mean SUVmax of borderline malignant tumors was 3.6, and 70% of them showed positive uptake elsewhere in the tumor; focally increased SUVmax (>5) in a tumor was considered positive for malignancy.⁸ In this case, SUVmax of the tumor was not as strong, so we considered this tumor to be nonmalignant based on FDG-PET/CT findings.

In these circumstances, preoperative differential diagnosis to exclude malignancy or borderline malignancy is difficult with cystadenofibromas. Preoperative diagnosis of an ovarian tumor, particularly in a young woman, should be performed carefully to preserve fertility, and ongoing data collection with combined diagnostic imaging and histopathology is important.

Conflicts of interest: The authors have no conflicts of interest relevant to this article.

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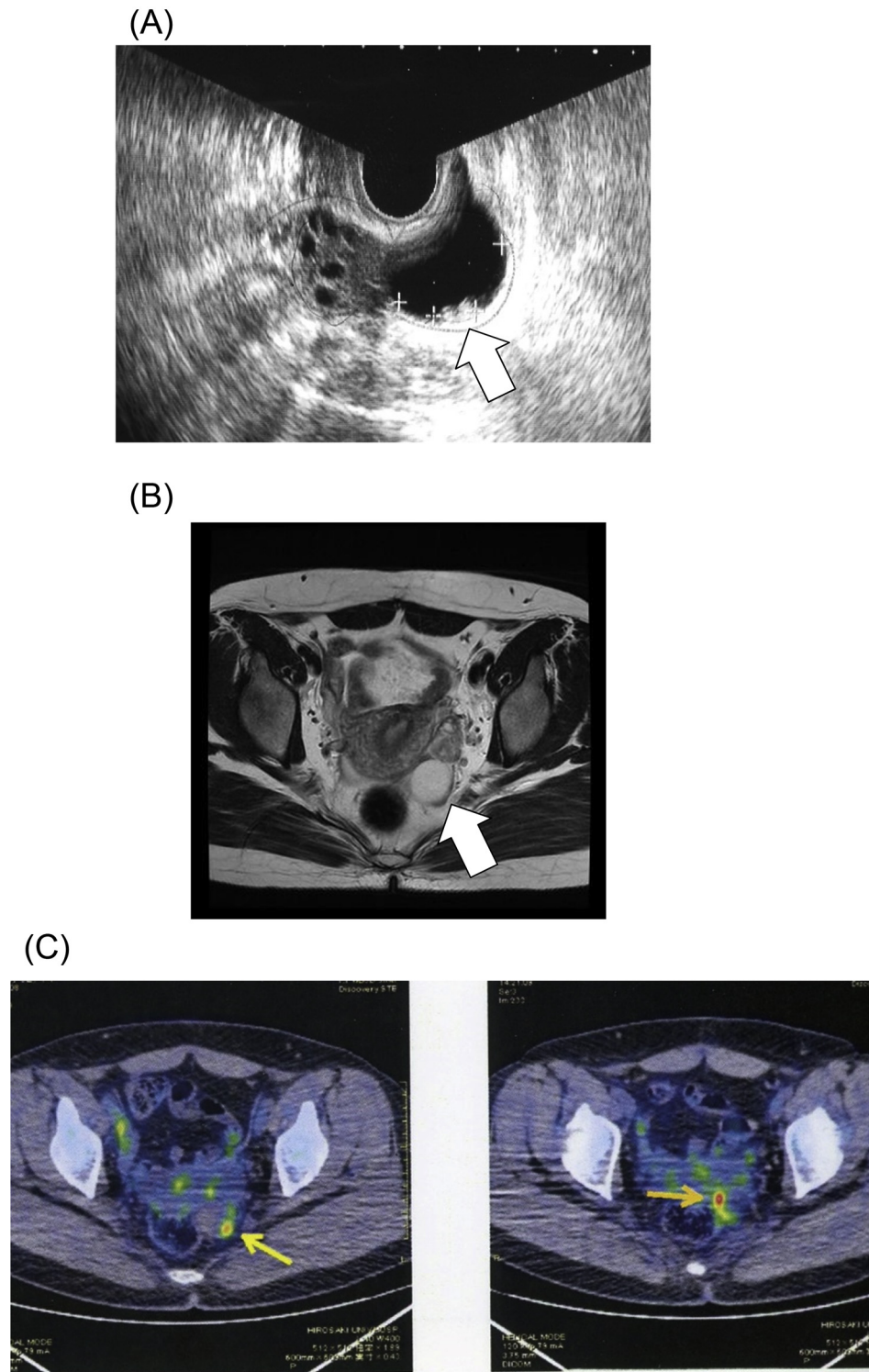
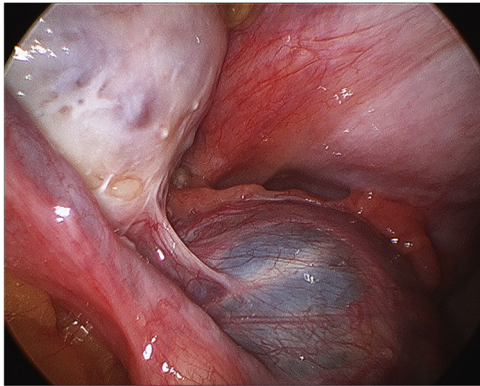


Figure 1. Preoperative images of paraovarian cystadenofibroma. (A) A small cyst is located next to the left ovary and the white arrow shows the solid part of the tumor, by transvaginal ultrasonography. (B) Contrast-enhanced pelvic MRI shows a simple cystic mass with a thick wall (white arrow) next to the left ovary. (C) FDG-PET/CT shows a weak positive uptake of FDG (yellow arrow) at the left adnexal lesion. CT = computed tomography; FDG-PET = 18F-fluorodeoxyglucose positron emission tomography; MRI = magnetic resonance imaging.

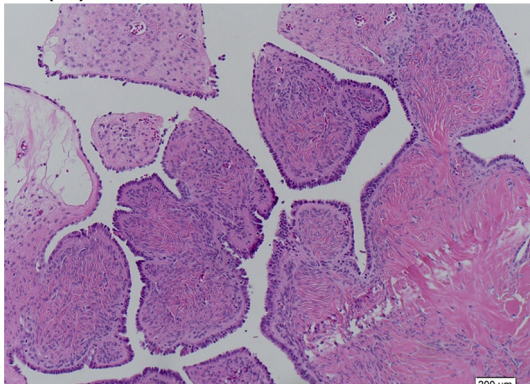
(A)



(B)



(C)



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Figure 2. Operative and postoperative findings of paraovarian cystadenofibroma. (A) The tumor was diagnosed as a paraovarian tumor and the left ovary was normal on laparoscopy. (B) Macroscopic examination revealed small papillary structures on the surface and inside the tumor. (C) Microscopic diagnosis was paraoophoric serous cystadenofibroma (unilocular cyst lined by a single layer of tall, columnar, ciliated cells without atypia and architectural complexity and spindly fibroblasts in the stroma).