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A case of ovarian serous cystadenofibroma with scattered lesions in pelvic cavity, like malignant disseminations

Kazuhisa Fujita[1], Chisa Ogawa[1], Takahiro Sako[1], Fumi Utsumi[1], Ken-ichi Inada[2] and Kiyosumi Shibata[1]

[1]*Department of Obstetrics and Gynecology, Fujita Health University, Bantane Hospital, Nagoya, Japan*
[2]*Department of Diagnostic Pathology, Fujita Health University, Bantane Hospital, Nagoya, Japan*

Abstract

Ovarian serous cystadenofibroma is a relatively rare subtype of serous cystadenoma classified as ovarian benign epithelial tumor. We report a rare case of ovarian serous cystadenofibroma with scattered lesions in pelvic cavity, like malignant disseminations. The patient was 22 years old, gravida 0, para 0. In the laparoscopic surgery, numerous hard yellowish-white solid masses of various sizes were present in the bilateral ovaries. Grossly similar masses were scattered in the fimbria of the fallopian tubes, peritoneum, and great omentum. Because the intraoperative rapid histological diagnosis was benign tumor, surgery was completed for only tumor excision. Postoperative histopathological diagnosis is serous cystadenofibroma. Similar pathological findings were noted in the scattered lesions in the peritoneum and great omentum. No malignant or borderline malignant finding was observed. Because of a benign disease, careful treatment taking fertility preservation into consideration is necessary, especially for young patients.

Key words: benign ovarian tumor, dissemination, serous cystadenofibroma.

Introduction

Ovarian serous cystadenofibroma is a relatively rare subtype of serous cystadenoma classified as ovarian benign epithelial tumor. It accounts for approximately 1.7% of benign ovarian tumor cases, develops at 15–65 years old, and the development is bilateral in 30%–50%.[1,2] Abdominal pain and genital bleeding may be present, but it is often discovered as an asymptomatic incidental mass. We report a rare case of ovarian serous cystadenofibroma with scattered lesions in pelvic cavity, like malignant disseminations.

Case Presentation

The patient was 22 years old, gravida 0, para 0, and had no past or familial medical history or life history of note. The patient had been prescribed a low-dose pill for amenorrhea. Four years ago, bilateral ovarian enlargement of 3 cm was noted, which increased to 5 cm 3 years ago. On magnetic resonance imaging (MRI), the bilateral ovaries exhibited a lobulated nodular structure containing many minute cysts (Figure 1(a,b)). The ovarian margin had a thick capsule of low signal intensity on T2-weighted imaging. These MRI findings are known as a "black garland appearance," which is a feature of ovarian fibromatosis. In addition, a 26 × 18–mm cystic mass with a clear boundary exhibiting markedly high signal intensity on T1-weighted imaging and low signal intensity on T2-weighted imaging newly developed in the right ovary (Figure 1(b)). Some minute mural nodules were found in the cystic mass (Figure 1(c)). Because the malignancy could not be ruled out, the patient was

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Correspondence
Kazuhisa Fujita, Department of Obstetrics and Gynecology, Fujita Health University, Bantane Hospital, 3-6-10 Odobashi, Nakagawa-ku, Nagoya 454-8509, Japan.
Email: fujita_kazuhisa@y7.dion.ne.jp

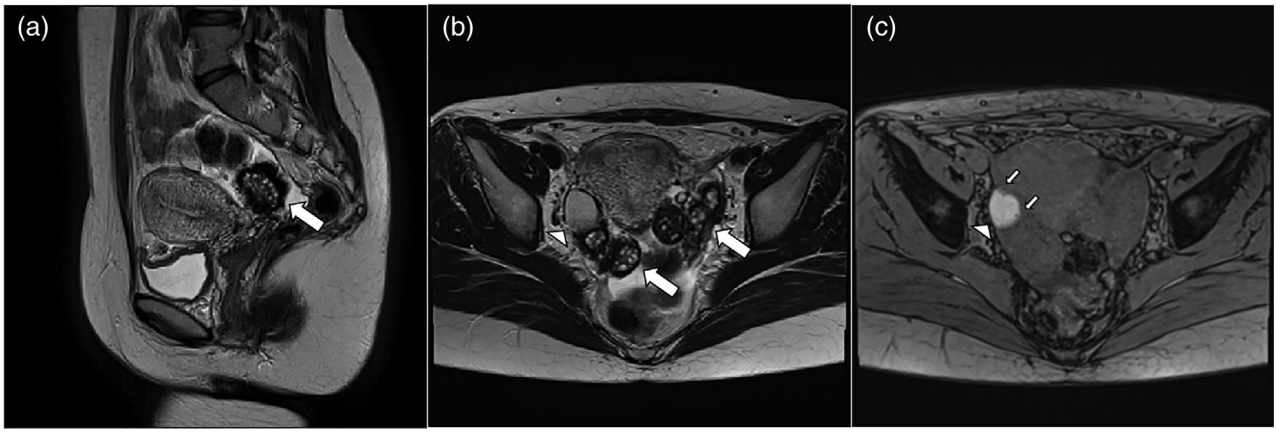

FIGURE 1 (a) Sagittal and (b) coronal views on T2-weighted magnetic resonance imaging (MRI), and (c) coronal view on T1-weighted MRI. The bilateral ovaries exhibited a lobulated nodular structure containing many minute cysts and the margin had a thick capsule with low signal intensity on T2-weighted imaging (large arrows). In addition, a 26 × 18–mm cystic mass with a clear boundary exhibiting markedly high signal intensity on T1-weighted imaging, low signal intensity on T2-weighted imaging newly developed in the right ovary (arrow heads) and minute mural nodules were found in the mass (small arrows)

referred to our hospital for further investigation and treatment. The patient was asymptomatic. All tumor markers were within the standard ranges: CA19-9, 19.5 U/ml; CA125, 34.8 U/ml; HE-4, 27.1 pmol/L. The hormone tests performed 1 month after withdrawal of the pill were as follows: lutenizing hormone, below the detection limit; follicle stimulating hormone, below the detection limit; estradiol, 13.9 pg/ml; progesterone: 0.15 ng/ml. Anti-Mullerian hormone (AMH) was 27.8 ng/ml, being high. Diagnostic laparoscopic surgery was performed after receiving informed consent.

Laparoscopy-assisted ovarian tumor excision was performed with a 3-cm transverse incision made above the pubis. No peritoneal ascites was present. Numerous hard yellowish-white solid masses of various sizes were present in the bilateral ovaries (Figure 2(a,c)). Normal ovarian parenchyma was not grossly confirmed. The right oviduct and great omentum were adhered to the right ovarian tumor, forming a single mass. Grossly similar masses were scattered in the fimbria of the fallopian tubes, peritoneum, and great omentum (Figure 2(b,d)). In the right ovary, a 2-cm cystic lesion was present. The ovarian tumors were pulled out of the body from the skin incision, and manually excised mainly in the pedunculated part. Peritoneal scattered lesions in the oviduct and great omentum were also excised. The right fimbria of the fallopian tube forming a single mass with the tumor was partially excised together. Because the intraoperative rapid histological diagnosis was benign tumor, surgery was completed with only tumor excision. The residual lesions were present.

Postoperative histological diagnosis is serous cystadenofibroma. Macroscopically, the left ovarian tumor is whitish hard solid mass with a size of the quail's egg and an irregular knobby serous surface. In the cut surface, broad-based exophytic papillae on the serosal surface and small cystic lesions in the background of the compact fibrous stroma are recognized. The cyst cavities are filled with watery fluid. Microscopically, the broad exophytic papillae are lined by single-layered, focally ciliated cuboidal or columnar cells (Figure 3(a,b)). These cells have scant pale eosinophilic cytoplasm, oval or rounded nuclei, and show mild nuclear stratification. Nucleoli are not distinct and no cellular atypia or mitoses are noted. In the fibrous stroma, various sized glands or slit-like clefts are scatteredly present. These glands, some of them are cystically dilated, or clefts are also lined by the cells similar to those of the surface one. The inner surface of some glands presents a polypoid appearance similar to the surface papillary projections. Normal follicles with various maturation stages are also found in the stroma. Based on the above findings, we diagnosed the left ovarian tumor as serous papillary adenofibroma. The right ovarian tumor exhibits almost the same features as those of the left except that the cystic changes were less prominent. The omental tumors and peritoneal nodules are small hard whitish

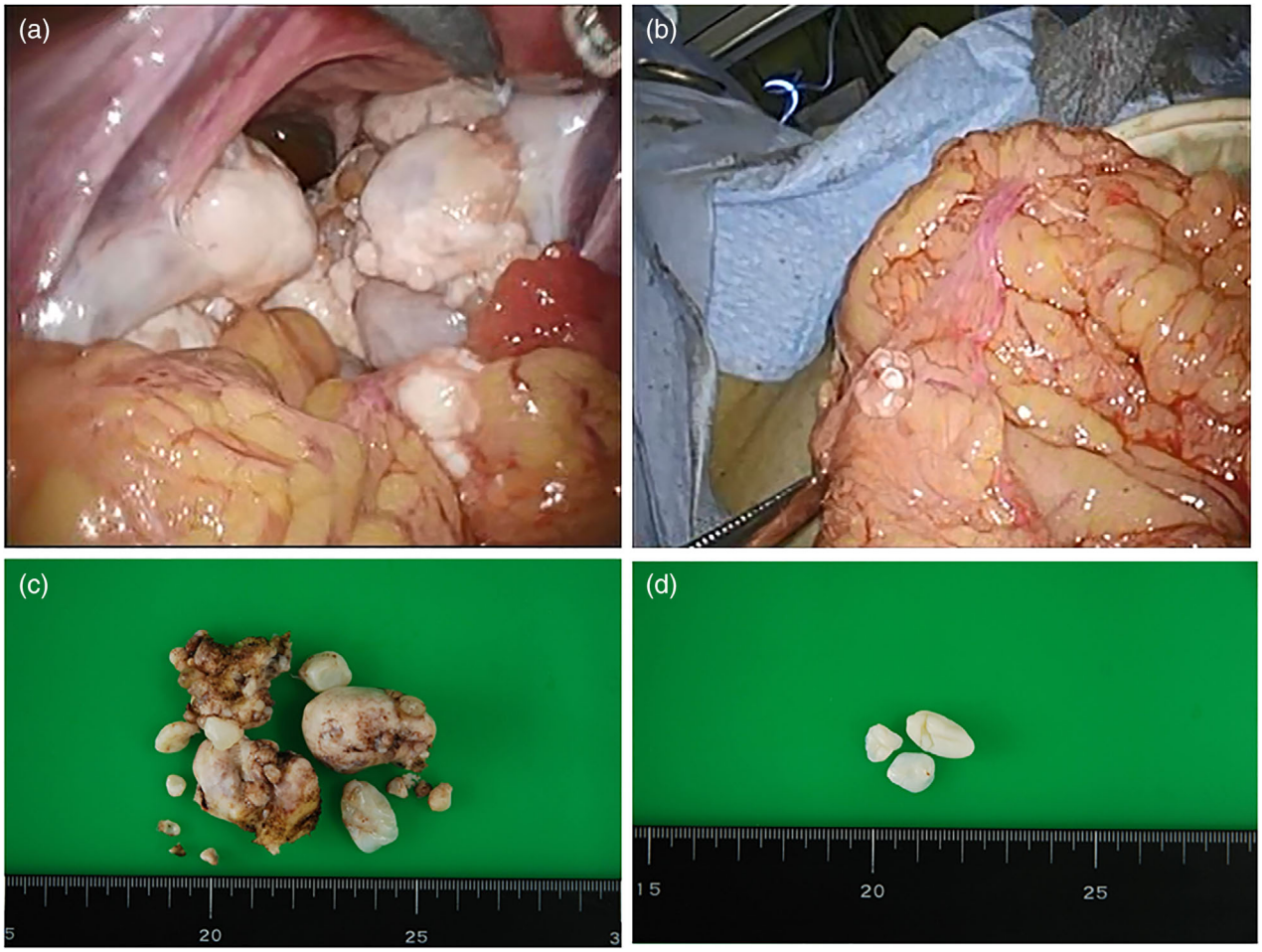

FIGURE 2 Intraoperative findings. Numerous hard yellowish-white solid tumors of varying sizes were present in the bilateral ovaries and normal ovarian parenchyma was not macroscopically confirmed (a and c). The right oviduct and great omentum were adhered to the right ovarian tumor, forming a mass. Scattered tumors were noted in the fimbria of the fallopian tube, peritoneum, and great omentum (b and d)

masses composed of cystic spaces and compact stroma with focal calcification. The surface of them has broad-based papillae and the lining cells are also the same as those of the bilateral ovaries (Figure 3(c,d)). In immunohistological staining, Vimentin was positive in most of the epithelium and stroma; WT-1 was positive in most of the epithelium; CD10 was almost negative (Figure 3(e,f,g). These immunohistological findings were consistent with serous cystadenofibroma. Almost similar immunohistochemical characteristics were observed in both peritoneal scattered lesions in the oviduct and great omentum. No interstitial infiltration was observed (Figure 3(c,d,h,i,j)).

The postoperative course was uneventful and the patient was discharged on the fourth day. She is being followed up every 3 months with outpatient transvaginal ultrasound and tumor markers (CA-125 and CA19-9) test. She has not had ovarian enlargement or increased tumor markers for 18 months after surgery.

Discussion

In the serous cystadenofibroma, interstitial fibrous components are dominant and cystic components are present in a mixture at varying ratios. Therefore, on T2-weighted MRI, small cysts exhibiting punctate high intensities are scattered centering on a low-intensity solid mass, so-called a "black sponge-like appearance."[3]

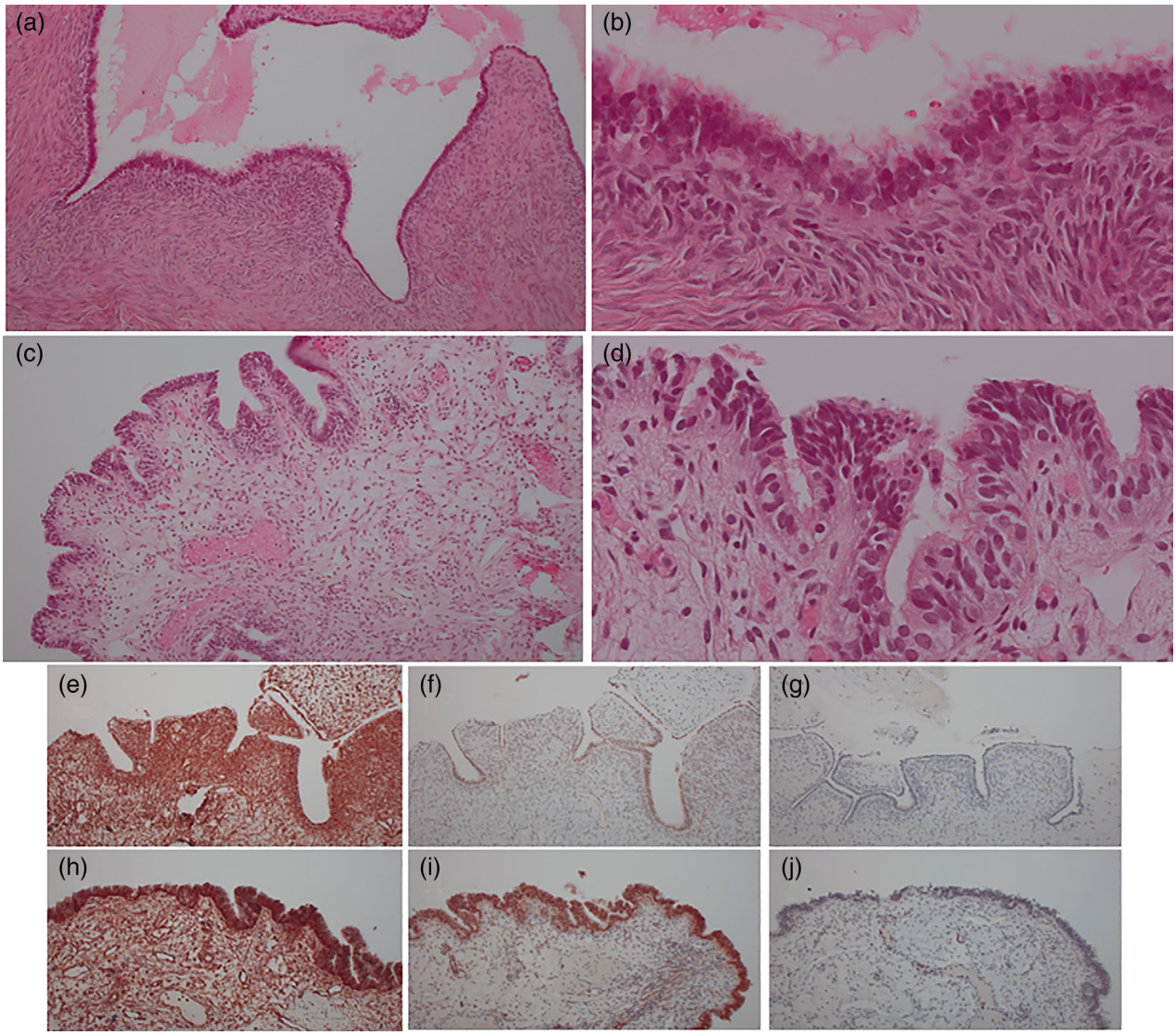

FIGURE 3 Histological features of serous cystadenofibroma. (a) Low magnification of the left ovarian tumor (hematoxylin and eosin [H&E] stain, ×10), (b) high magnification of the same tumor (H&E stain, ×40), (c) low magnification of the tumor in the great omentum (H&E stain, ×10), and (d) high magnification of the same tumor (H&E stain, ×40). Immunohistological staining in left ovarian tumor by (e) vimentin, ×10, (f) WT-1, ×10, and (g) CD-10, ×10. Immunohistological staining in great omental lesion by (h) vimentin, ×10, (i) WT-1, ×10, and (j) CD-10, ×10. The interstitium with noticeable fiber components protruded on the papilla in an extraverted direction and the surface layer was covered by ciliated cuboidal or columnar epithelium, resembling oviductal epithelium. The nuclear atypia was not observed in both ovarian tumors and peritoneal lesions. Ductal structures of varying sizes similarly lined by epithelium were also found in the interstitium. Histopathological diagnosis is serous cystadenofibroma. In immunohistochemical staining, Vimentin was positive in most of the epithelium and stroma; WT-1 was positive in most of the epithelium; CD10 was almost negative. These findings were consistent with serous cystadenofibroma. Peritoneal lesions and great omental lesions have almost similar immunohistochemical characteristics

Differentiation between benign and malignant is often difficult on preoperative imaging. Thus a few cases misdiagnosed as malignant tumor have been reported.[1–7] In this case, it was more difficult to diagnose, because the intraoperative macroscopic findings have many peritoneal lesions, like disseminations of malignant tumor.

Benign tumors should not have "disseminations." In this case, there is no malignant or borderline malignant finding on both ovarian tumors and peritoneal lesions. Because of the lack of interstitial infiltration in the peritoneal lesions, it was different from the dissemination of malignant tumor. We think that those peritoneal lesions were something like implants, which observed in the parasitic uterine myoma.[8] Ovarian tumor masses that fell in the abdominal cavity might have induced blood vessels through certain unknown mechanisms, and engrafted on the surfaces of the great omentum or peritoneum.

As cases similar to peritoneal lesions in this patient, there are some case reports of complications of ovarian adenofibroma and endosalpingiosis or peritoneal serous cancer.[9,10] Endosalpingiosis is characterized by the ectopic presence of benign glands lined by oviductal-type epithelium. There is a rare case report of endosalpingiosis forming mass lesions resembling neoplasms.[11] Because the peritoneal mesothelium shares a common origin with the ovaries and fallopian tubes, ovarian tumor-like lesions may occur in the peritoneum. Histologic differentiation between endosalpingiosis and ovarian serous tumor, dissemination of malignancy/borderline malignancy is very difficult. Careful differentiation is necessary to avoid unnecessary postoperative treatment of benign lesions. This case report might be helpful in making a decision.

Amenorrhea was observed in this patient, but no case of ovarian serous cystadenofibroma complicated by amenorrhea has been reported. Ovulation disorder might have developed due to the hardened ovarian surface by solid tumor components. It may be cause of a high AMH level and amenorrhea. It is necessary to carefully follow the course regarding fertility.

Ovarian serous cystadenofibroma is a benign tumor, and the prognosis is favorable. Excess surgical stress should be avoided. This patient was treated by only tumor excision to preserve fertility and the residual lesions can be macroscopically noted. Careful course observation is necessary because there is a risk of enlargement of tumors.

Conclusion

We experienced a rare case of ovarian serous cystadenofibroma with scattered lesions in pelvic cavity, like malignant disseminations. It is difficult to differentiate between benign and malignant on preoperative imaging examination, and intraoperative macroscopic findings. However, it is a benign disease and should be carefully handled taking fertility preservation into consideration, especially for young patients.

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Conflict of Interest

None declared.

Author Contributions

All authors revised the manuscript, approved the manuscript to be published, and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Data Availability Statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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