Case Report

Ovarian Cystadenofibroma: An Innocent Tumor Causing Early Postoperative Small Bowel Obstruction and Perforation Peritonitis

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Abstract

Ovarian serous cystadenofibroma (CAF) is a relatively uncommon variant of benign epithelial tumors of the ovary. It is frequently misdiagnosed as malignant ovarian mass, on both ultrasound (USG) and computed tomography (CT). Although most cases are easily treatable by surgery, some cases can present with life-threatening complications increasing patient morbidity and mortality. The present case report briefs about a 69-year-old female, P4 L4, who presented to the gynecology outpatient department with a complaint of pain in the lower abdomen for 2–3 months. USG and CT were suggestive of a suspicious-looking ovarian mass favoring malignancy. A staging laparotomy with pelvic and para-aortic lymphadenectomy with omental biopsy was done. Although the tumor was benign, extensive surgery, due to the suspicion of malignancy led to the patient developing early postoperative small bowel obstruction, mandating a re-exploration. CAF is a specific type of ovarian tumor that exhibits a combination of benign characteristics. This tumor presents as a partly cystic (containing fluid-filled sacs) and partly solid (composed of fibrous tissue) growth within the ovary, displaying a diverse architectural pattern. Mostly the diagnosis is incidental, on USG done for some other indication. CAF of the ovary needs a very high index of suspicion for diagnosis as these are frequently misdiagnosed as malignant ovarian masses. Although an innocent tumor, extensive surgery done for CAF, under suspicion for malignancy, can sometimes lead to serious complications.

Keywords: *Benign ovarian tumor, cystadenofibroma, early postoperative small bowel obstruction, ovarian tumor*

Submitted: 12-Jan-2024
Revised: 16-Feb-2024
Accepted: 22-Feb-2024
Published: 04-Apr-2024

Introduction

According to the World Health Organization classification, papilloma, cystadenoma, adenofibroma, cystadenofibroma (CAF), metaplastic papillary tumors, and endometrioid polyps fall into the category of benign tumors.[1] CAF is a term used to describe a specific type of ovarian tumor that exhibits a combination of benign characteristics. This tumor presents as a partly cystic (containing fluid-filled sacs) and partly solid (composed of fibrous tissue) growth within the ovary, displaying a diverse architectural pattern. It is believed to have its origins in the Müllerian structures.[2] The cysts in this tumor originate from the ovarian surface epithelium's invaginations, resulting from the natural changes in the ovary's shape during a person's reproductive years. The fibrous component of the tumor is attributed to varying degrees of cortical stromal proliferation and collagenization. Diagnosing CAF can be somewhat subjective and relies on assessing the relative proportions of cystic epithelial tissue to stromal fibrous tissue within the tumor.[3] In addition, the secretory activity of the cystic epithelium influences the tumor's size, shape, and contour. Morphologically, CAFs occupy an intermediate position between simple serous cystadenomas (fluid-filled tumors) and fibromas (tumors predominantly composed of fibrous tissue). They form

Access this article online

Quick Response Code:

Website: https://journals.lww.com/jomh

DOI: 10.4103/jmh.jmh_5_24

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How to cite this article: Mangla M, Ramavath K, Kumar N, Roy S, Srirambhatla A, Palo S. Ovarian cystadenofibroma: An innocent tumor causing early postoperative small bowel obstruction and perforation peritonitis. J Mid-life Health 2024;15:43-7.

a somewhat ambiguous category, which might explain their relatively infrequent occurrence in medical reports.

Case Report

A 69-year-old female, P4 L4, visited the gynecology outpatient department with complaints of pain in the lower abdomen for 2–3 months. The pain was intermittent, dull aching, and diffuse throughout the lower abdomen, not associated with any specific aggravating or relieving factors. The bowel and bladder habits were normal. There was no history of loss of appetite or weight. She had no known medical comorbidities except for a history of gastroesophageal reflux disease, but she was not on any medication. She had undergone a total abdominal hysterectomy 30 years back for a fibroid uterus. The general physical examination revealed no abnormality, except that she was overweight with a body mass index of 28.5 kg/m^2. On per abdomen examination, a solid cystic mass of approximately 10 cm × 10 cm was felt in the lower abdomen, more toward the right side. On per speculum examination, the vault was atrophic. On bimanual examination, the vault was held up well, and the lower pole of the mass could not be reached. The ovarian mass was better felt on per abdomen examination only.

On ultrasound (USG), an irregular cystic mass lesion was noted in the right adnexa, measuring 87 mm × 56 mm × 79 mm with thick irregular septations, with the inner wall of the lesion having two irregular and eccentric solid components measuring 29 mm × 16 mm and 45 mm × 10 mm with no perceptible vascularity. All these features favored a lesion of intermediate risk (Ovarian-Adnexal Reporting Data System USG Score of 4) suggestive of ORADS-4 [Figure 1]. The left ovary could not be visualized. For better characterization of the ovarian mass, a contrast-enhanced computed tomography (CT) scan was done which was suggestive of a large lobulated right adnexal cystic lesion with thick internal septations and minimal eccentric soft-tissue density solid component with no significant postcontrast enhancement. The findings were suggestive of a right ovarian complex cyst, likely neoplastic [Figure 2]. Serum CA-19-9 was 1.12 U/mL and inhibin B was 18.8 pg/mL. Lactate dehydrogenase, carcinoembryonic antigen, and alpha-fetoprotein were within normal limits. USG examination of breast and upper and lower gastrointestinal endoscopies was normal.

Suspecting an intermediate to high-risk ovarian lesion, a staging laparotomy was performed through a midline vertical incision. There were minimal ascites, the fluid was sent for cytopathological examination. The bowel was adherent to the anterior abdominal wall. A solid cystic mass of 8 cm × 8 cm × 8 cm was found arising from the right adnexa. The bowel was adhered to the mass and peritoneum at multiple places, for which assessment with adhesiolysis was done. There were no suspicious areas on the omentum, liver, or undersurface of the diaphragm. Small bowel loops were adherent to each other at multiple places, through flimsy adhesions. Pelvic and para-aortic lymphadenectomy with omental biopsy was done. The patient was recovering well, except for occasional episodes of nausea and vomiting. However, on postoperative day 7, she complained of dull aching pain all over the abdomen, abdominal distension, and bilious vomiting, also associated with absolute constipation and three episodes of dark-colored nonprojectile vomiting. On examination, the patient was

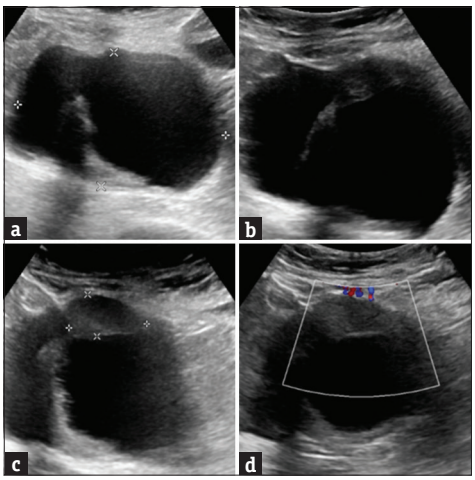

Figure 1: (a and b) Well-defined cystic lesion showing thick internal septation. The inner wall is irregular. (c and d) Eccentric solid component is seen. No vascularity perceivable in the solid component

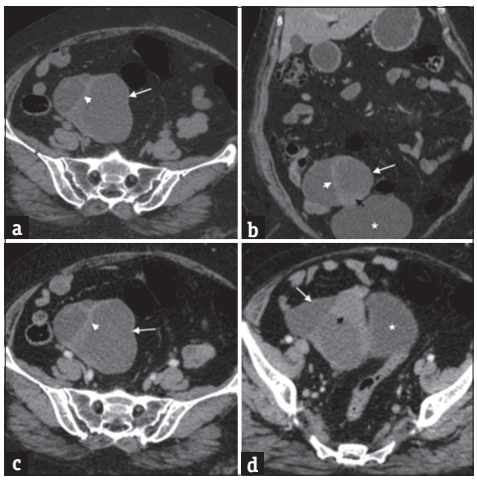

Figure 2: Noncontrast axial (a) and coronal (b) images reveal a thin-walled well-defined cystic lesion (long arrows) arising from the right adnexa extending into the right iliac fossa. Thick internal septations (short arrows) and a smooth eccentric solid component (black arrow) are noted. Axial postintravenous contrast (c and d). The septations (short arrow) and solid components (black arrow) reveal homogenous enhancement. Urinary bladder (*)

febrile and uncomfortable but well-oriented to time, place, and person with no signs of dehydration. Initial vitals included a blood pressure of 130/90 mmHg, a regular pulse of 90 beats/min, and a respiratory rate of 22 breaths/min. On inspection of the abdomen, she had diffuse abdominal distention, with guarding and rigidity. On palpation, she had mild diffuse tenderness all over the abdomen, more on the right iliac fossa region, which was radiating toward the left iliac fossa. On auscultation, sluggish gut sounds were heard. No lymph nodes were palpable. A nasogastric tube was inserted, which drained 1200 mL of dark-colored feculent fluid. Blood investigations revealed thrombocytopenia and neutrophilic leukocytosis with a total leukocyte count (TLC) of 23 × 109/L. The abdominal USG report showed dilated bowel loops and increased bowel gases with previous wound site fluid collection in the intermuscular plane of the anterior abdominal wall in the right lumbar and iliac region. Abdominal X-ray showed multiple air-fluid levels with no gas under the diaphragm. The abdominal CT scans revealed dilated bowel loops, mesenteric thickening, inter-bowel loop adhesions, and free gas under the diaphragm, with around 6 cm × 4 cm × 3 cm of pelvic fossa fluid collection [Figure 3]. An emergency re-exploratory laparotomy was undertaken. There was around 200 mL of fecal-purulent fluid present in the peritoneal cavity extending toward the anterior abdominal wall. The muscles in the right anterolateral abdominal wall in the lumbar and iliac region were necrosed. Rectus sheath and muscle were also necrosed. There was a foul smell with around 100 mL of purulent collection. Small bowel loops were dilated and adhered to the anterior abdominal wall. Dense intersmall bowel adhesions were present. One 6–8 cm length of the mid-ileal segment is presented as a closed loop [Figure 4]. The 10 cm long distal ileal loop, around 10 cm proximal to the ileocecal junction was gangrenous and paper thinned out with thickened mesentery. The entire small bowel was dilated and edematous and ischemic spots were present over the bowel wall. After thorough peritoneal lavage, resection of the gangrenous segment of the distal ileal loop along with end ileostomy was done. A pelvic drain was inserted.

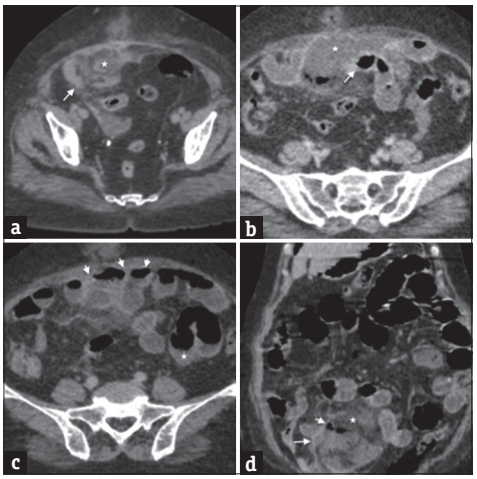

Figure 3: Postoperative computed tomography of abdomen and pelvis. (a and b) Axial images reveal collapsed distal ileal loops (long arrows) adherent to a contained abscess with adjacent inflammation (*) just deep to the surgical scar. The zone of transition in the distal ileal loop is seen in b (arrow). (c) Dilated ileal loops in close approximation to the anterior abdominal wall (arrowheads). Dilated small bowel loops (*). (d) Coronal image shows free air (short arrow), intraperitoneal contained inflammation (*) with adherent bowel loops (long arrow)

The patient was kept on ventilator support on synchronized intermittent mandatory ventilation mode for 2 days in the postoperative intensive care unit with inotropic support. Postoperative TLC was 25,000 cells/mm^3. Gradually, patients improved clinically and could be weaned off from ventilator support on postoperative day 3. Intraoperative main wound site pus culture sensitivity showed *Klebsiella pneumonia* sensitive to amikacin. The patient was allowed orally from postoperative day 3. The stoma functioned well and ileostomy closure was done after a proper nutritional assessment of the patient after 6 weeks.

The histopathologic examination was done by a pathologist with more than 10 years of experience in gynecologic pathology. The ascitic fluid was negative for malignant cells. On gross examination, the ovarian cyst measured 10 cm × 8 cm × 6.5 cm. The capsule was intact. The cyst wall showed tiny papillary excrescences and a single solid area with a whitish-whorled cut surface. Sections from the cyst wall showed lining by a single layer of cuboidal cells, with focal papillary formations and underlying areas showing dense fibrous tissue and hyalinization. There was no evidence of

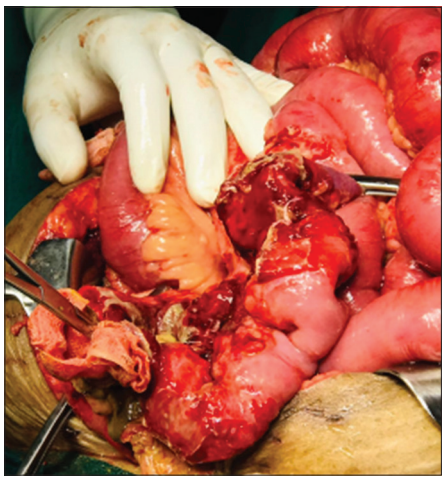

Figure 4: Intraoperative image during re-exploration, showing dense inter-bowel loop adhesions, gangrenous segment of small bowel, and site of perforation

nuclear atypia, stratification, or invasion. Omental and peritoneal biopsy showed no significant pathology. The diagnosis was confirmed as serous CAF of the right ovary [Figure 5]. The histopathology of the resected bowel was suggestive of nonspecific mild active enterocolitis, with submucosal lymphoid follicle hyperplasia and dense transmural mixed inflammatory infiltrate of neutrophils, lymphoplasmacytic cells, histiocytes, occasional multi-nucleate giant cells, and extravasated red blood cells.

Discussion

Ovarian serous CAF is a relatively uncommon variant of serous cystadenoma categorized as a benign epithelial tumor of the ovary. It represents about 1.7% of all cases of benign ovarian tumors, typically emerging between the ages of 15 and 65.[4] Notably, this tumor displays bilateral development in approximately 30% to 50% of cases.[5] It belongs to the category of surface epithelial tumors and is composed predominantly of benign-appearing stroma derived from the ovarian stroma. This stromal component is thought to be identical to a fibroma.[6] The amount of fibrous stroma can vary in different tumors, with some studies indicating that solid nodules are present in 33% to 80% of cases.[7]

Although the most common variety is serous, as in our case, they may also be of endometroid, clear-cell, or mucinous type. These tumors can be classified into benign, borderline, and malignant according to the degree of cellular differentiation.[8] These tumors can vary in size and may or may not cause symptoms. Mostly, the diagnosis is incidental, on USG done for some other indication. Some common symptoms if present could include pelvic pain, abdominal discomfort, or changes in menstrual patterns. Rarely, they have even been reported to present with clinical features of abnormal uterine bleeding or feminization due to hyperestrogenism induced by the tumor.[9,10]

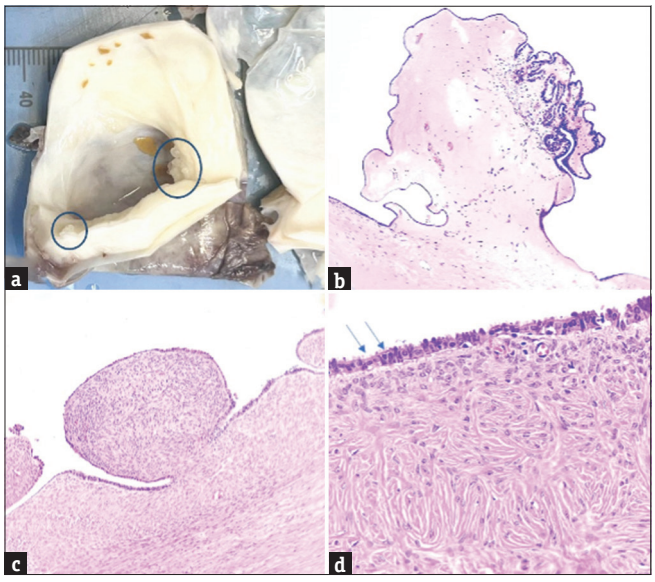

Figure 5: (a) Cut surface of the ovarian mass displaying multiloculated cyst with varying wall thickness and papillary excrescences (circled) at places, (b and c) Cyst wall with polypoidal projections lined by a single layer of flattened to columnar cells and showing prominent fibroblastic stromal component with varying degrees of cellularity (×10, hematoxylin and eosin), (d) Higher magnification showing single layer of tall, columnar, ciliated cells (arrow) resembling normal tubal epithelium with underlying stroma containing bland spindled fibroblasts in a collagenized background (×40, hematoxylin and eosin)

CAF of the ovary is frequently misdiagnosed as malignant ovarian mass, on both USG and CT. The CT characteristics of primary ovarian CAF have rarely been discussed in medical literature. In a study by Cho *et al.*,[11] they highlighted challenges in accurately diagnosing ovarian CAFs preoperatively through CT scans. According to their findings, all 12 cases of ovarian CAFs were initially misidentified on CT as malignant ovarian tumors. In the present case, the diagnosis was that of a suspicious ovarian mass with a high probability of neoplasia. In contrast to CT or ultrasonography, magnetic resonance imaging (MRI) can provide distinctive diagnostic insights for ovarian CAF by revealing dense fibrous stromal proliferation alongside scattered small cystic glandular structures in T2-weighted images.[12] This ability to identify these specific features aids in distinguishing between benign and malignant tumors in the ovaries.[12] Outwater *et al.* documented that the solid component of this tumor exhibits a low signal intensity on T2-weighted MRI images, resembling that of skeletal muscle. This is attributed to the presence of fibrous tissue within the solid portion.[13] Furthermore, Takeuchi *et al.* highlighted that distinct small or miniature cystic areas within this solid component are distinctive features of CAF. These areas manifest as a unique black sponge-like appearance when observed on T2-weighted images.[14] Jung *et al.* reported diffuse or partial thickening of the cyst wall with dark signal intensity in multilocular cystic masses as a characteristic feature of CAF, further suggesting that a prominent solid component with a higher T2-signal intensity and strong enhancement are the typical findings of cystadenocarcinofibroma.[15]

Treatment for ovarian CAFs typically involves surgical removal, especially if they are causing significant symptoms, growing rapidly, or if there is any suspicion of malignancy (cancer). In most cases, these tumors do not become cancerous, but a surgical biopsy may be performed to confirm their benign nature. The prognosis is good with appropriate treatment. The complications for the same are rare such as rupture of the cystic fluid or torsion. However, in some cases, even an innocent

tumor such as CAF can lead to serious complications, just like in the present case.

The authors considered it important to report this case, to highlight the fact, that CAF, can be a mimicker of malignancy on imaging. The extensive surgery done in the present case, considering the tumor to be intermediate to high risk for malignancy, led to postoperative complications. The tumor in the present case was associated with multiple flimsy adhesions in the loops of the small bowel, which in the postoperative period could have further progressed, leading to early postoperative small bowel obstruction and perforation eventually mandating surgical exploration. The present case is reported to highlight the fact that even benign ovarian masses can lead to serious complications, increasing morbidity for the patient. The case also highlights the difficulty the clinicians face in such cases, in deciding whether conservative or surgical management would be more appropriate for such cases of early postoperative small bowel obstruction, and when to switch from conservative to surgical. It should be kept in mind that after surpassing the 2-week threshold, these adhesions transform into dense, highly vascularized structures, establishing an unfavorable environment, that heightens the probability of bowel damage when revisiting the area, therefore, early surgical intervention should be favored.[16]

Conclusion

CAF of the ovary needs a very high index of suspicion for diagnosis as these are frequently misdiagnosed as malignant ovarian masses. Although an innocent tumor, extensive surgery done for CAF, under suspicion for malignancy, can sometimes lead to serious complications.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and that due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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