Case Report

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Sigmoido-ovarian fistula complicating ovarian carcinosarcoma: a case report

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Introduction: Ovarian cancer is the leading cause of death from gynecological cancer. Ovarian carcinosarcomas represent a rare, aggressive entity with a poor prognosis. Spontaneous fistulization of ovarian cancer into the digestive tract is a rare phenomenon.
Presentation of case: A 67-year-old woman with a significant history of cardiac rhythm disorders was consulted for abdominal pain. Examination revealed tachycardia and abdominal guarding. Biology pictured elevated inflammatory markers and low prothrombin time. The abdominal computed tomography scan suggested a perforated sigmoid tumor with a peri-colonic abscess and pneumoperitoneum. She was rushed to the operating theater. Upon exploration, it was an ovarian tumor fistulized to sigmoid with peritonitis. She had an en-bloc resection with a terminal stoma. Control radiological study revealed diffuse lymph node metastasis. She was scheduled for chemotherapy.
Discussion: This complication worsens the prognosis. The fistulous communication in the digestive lumen leads to the overflow of its microbial deposit. The tumor, therefore, becomes superinfected and may result in pelvic peritonitis in case of secondary rupture. On the other hand, the patient is deprived of the benefit of undergoing neoadjuvant chemotherapy, which will decrease the chances of complete macroscopic cytoreduction. Through a literature review, we aim to shed light on this rare entity in order to clarify its pathophysiological consequences and make adequate therapeutic measures.
Conclusion: Fistulization to the large intestine worsens the prognosis of ovarian carcinosarcomas. Surgery is mandatory and should comply with oncological requirements. Adjuvant therapy is mostly needed, although more studies should be conducted to delineate the regimen accurately.

Keywords: carcinosarcoma, case report, digestive, fistula, ovary, tumor

Introduction

Ovarian carcinosarcomas is a fearsome tumor because of its aggressive behavior and lack of guidelines on therapeutic strategies. This situation is further worsened when complications occur, like fistulization to adjacent organs. To our knowledge, this is the first reported case of oophoro-sigmoid fistula amidst carcinosarcoma of the ovary. Through a literature review, we aim to shed light on this rare entity in order to clarify its pathophysiological consequences and make adequate therapeutic measures. This work has been reported in line with the SCARE 2023 criteria[1].

HIGHLIGHTS

- Ovarian carcinosarcomas are rare and aggressive tumors.
- Oopho-digestive fistulization worsens the prognosis.
- Surgery is the backbone of its treatment.
- Following the oncological imperatives ensures better results.
- Reconstruction of the digestive tract should be well thought-out.

Presentation of case

A 67-year-old woman followed for atrial fibrillation and put on acenocoumarol, required surgery for coronary insufficiency, repairing mitral valve replacement and cholecystitis, was referred from an infirmary to our university hospital center for acute abdominal pain evolving for 2 months with anorexia. She had no known allergies or hereditary disease. She denied smoking or alcohol intake. Upon examination, her blood pressure was 100/60 mmHg, she had tachycardia 108 pulse-per-minute, and abdominal guarding was perceived. Her inflammatory markers were slightly elevated: white blood cell = 11 500/ml, C-reactive protein = 25 mg/l. Renal function (creatinin = 60.74 µmol/l) and hemoglobin (11.5 g/dl) were preserved. She had a low prothrombin time due to anticoagulant intake and sepsis (prothrombin time = 18%). Abdominal computed tomography (CT) scan exhibited a 150 mm hydroaeric collection communicating with the sigmoid through a 14 mm parietal defect with pneumoperitoneum, aeroporttie, and enlarged necrotic common iliac

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and lumbo-aortic lymph nodes measuring 2.5 cm (Fig. 1). The diagnosis of the perforated sigmoid tumor with an abscess was made. Upon these findings, she was rushed to the operating theater, after a short resuscitation including prothrombin complex concentrate intake. She was approached via a midline incision. Purulent effusion was aspirated. Operative findings were purulent peritonitis and a huge ovarian tumor invading the sigmoid colon (Fig. 2). Purulent effusion was aspirated with a bacteriological sample. She had an en-bloc resection of the left ovary and sigmoid, and with the creation of the left iliac fossa stoma as a Hartmann procedure, the right ovary was removed. This decision was made regarding the hemodynamic instability during surgery, requiring noradrenalin intake, and peritonitis prohibiting digestive anastomosis. The operating timing was about 3 h, and a lengthy period was dedicated initially to restore a good hemodynamic status. The estimated blood loss was 250 ml. Postoperative follow-up was uneventful; she was discharged on the 10th postoperative day, after being put back on oral anticoagulants. The histopathological examination revealed on gross appearance a $17 \times 11 \times 8$ cm unilocular cystic ovarian tumor. The cut section showed a 6 cm whitish, necrotic, and hemorrhagic intra-cystic tissular component. Microscopy showed mixed carcinomatous and sarcomatous cell proliferation. The epithelial contingent included glandular structures and rows of cylindrical cells with basophilic cytoplasm of moderate abundance, dense irregular elongated nuclei with high-grade atypia (Fig. 3). The sarcomatous contingent consisted of spindle-shaped cells of variable size, with ovoid nuclei and numerous mitoses (Fig. 4). Cancer cells have spread to the surface of sigmoid delineating a punctiform perforation with infiltration of pleomorphic inflammatory cells. Histological results were consistent with a carcinosarcoma, staged TNM (8th edition) pT2b with the following phenotype: PAX8 + and WT1. On a multidisciplinary consultation meeting, a second stage surgery to perform appendicectomy, omentectomy, and lymph node removal was decided. The patient was since lost to follow-up, returning 4 months later to our outpatient department with back pain. CT scan control revealed multiple peri-aortic and latero-aortic, inter-aorto-cave and latero-cave lymph nodes with iliac lymph nodes compressing the left ureter with ipsilateral cortical narrowing. She was scheduled for palliative chemotherapy.

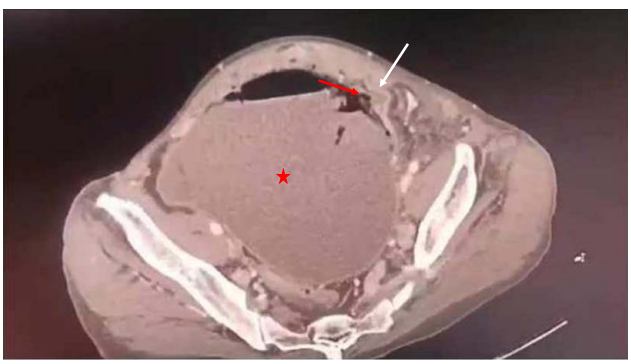

Figure 1. Abdominal CT slices exhibiting the huge intra-abdominal abscess (red star) with the collapsed sigmoid (white arrow) in contact and multiple air bubbles. The wall of sigmoid is discontinued (red arrow), suggesting its rupture. CT, computed tomography.

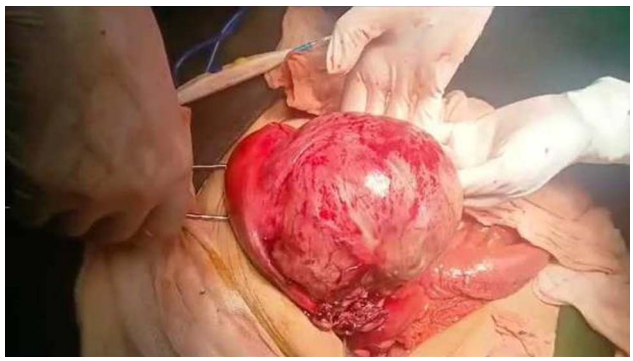

Figure 2. Intraoperative view of the enlarged left ovary invading the sigmoid.

Discussion

Ovarian cancer is the leading cause of death from gynecological cancer[2]. Seventy percent of patients are diagnosed at an advanced stage[2]. Ovarian carcinosarcomas represent a rare, aggressive entity with a poor prognosis. In a study by the National Institute of Cancerology in Mexico, eight cases of carcinosarcoma popped up in a time-lapse of 4 years and thus represented 4.1% of all the malignant primary tumors of the ovary[3].

They are often declared at an advanced stage, as highlighted by the French cohort study, including 12 referent centers for rare gynecological malignancies, where the vast majority of women already had an advanced-stage ovarian tumor (FIGO $\geq$ III)[4]. Despite the fact that the majority of these tumors (86%) are successfully cytoreduced to a residual disease <1 cm and

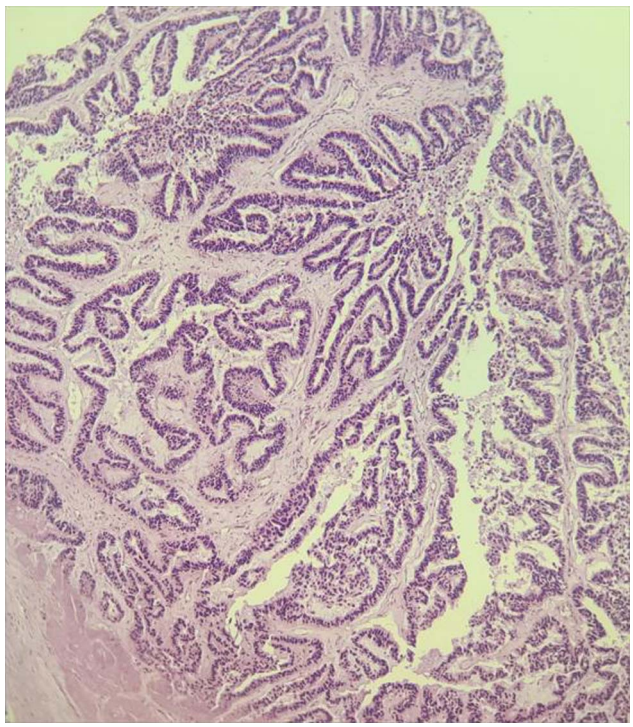

Figure 3. Carcinomatous contingent made up of tubulovillous structures stained with hematoxylin and eosin stain, $\times 10$.

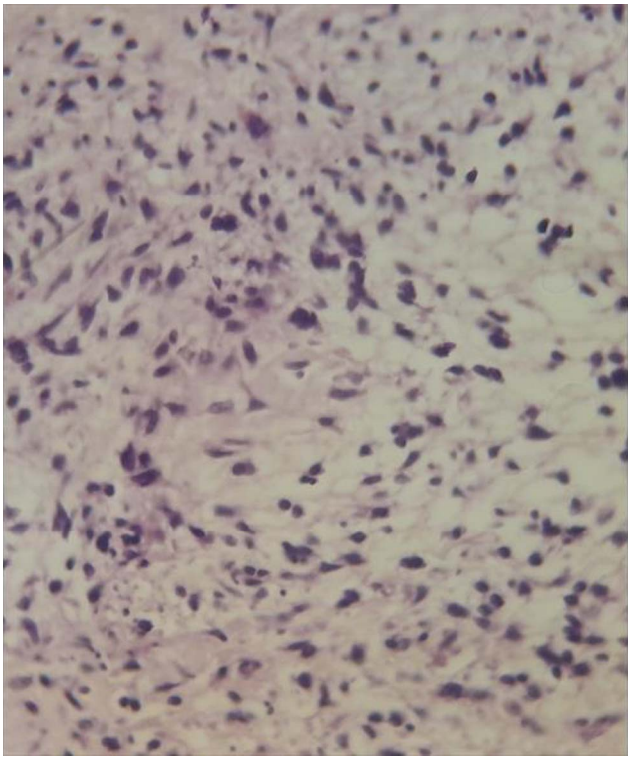

Figure 4. Sarcomatous contingent of elongated cells with atypical hyperchromatic nuclei stained with hematoxylin and eosin stain, ×40.

accompanied by adjuvant chemotherapy, oncological results are disappointing[5]. They have poorer survival compared to high-grade serous carcinoma, as confirmed by a recent cohort study from the Edinburgh Ovarian Cancer Database of 82 ovarian carcinosarcomas and 362 high-grade serous ovarian carcinoma in a comparative cohort[6]. This implies that the majority of women (92.8%) will require postoperative chemotherapy[4]. Despite efforts to improve management, recurrence-free survival is only 15.1 months, and overall median survival is 37.1 months[4].

Their clinical presentation is very similar to that of ovarian and uterine carcinomas, with no particular specificity. In a Japanese comparative study of MR imaging published in 2021, it was found that carcinosarcomas formed larger masses, and the stained-glass appearance, hemorrhage, and necrosis were more frequently observed than in serous carcinoma[7].

Surgery is the backbone treatment because it shows improved survival in patients treated with optimal cytoreductive surgery with residual tumors <2 cm reaching 30 months (vs. 5 months, $P = 0.042$)[8].

Enterogenital fistulas occupy the dominant cause of gynecological fistulas nearly 55%, according to a retrospective Norwegian cohort study of 1627 women treated for gynecological fistula[9]. Formation of a fistula to the sigmoid colon is the most common site of enterogynecologic fistula, accounting for 20% of all involved organs[10]. Fistulae usually occur after previous surgery and chemotherapy, in the setting of relapsed disease rather than as an initial presentation[11]. Spontaneous fistulization of ovarian cancer into the digestive tract is a different and rare phenomenon. Half of the cases were secondary to the malignant tumor[12]. However, the fistulous communication can be caused by a benign dermoid cyst of the ovary, in which spontaneous rupture is due to torsion, infection, trauma, or chronic pressure during labor[13]. In fact, no infiltration of malignant cells was detected around the fistula in a reported case[13].

This complication worsens the prognosis. The fistulous communication in the digestive lumen leads to the overflow of its microbial deposit. The tumor, therefore, becomes superinfected and may result in pelvic peritonitis in case of secondary rupture or by diffusion, as reported in our case. This complication occurs insidiously as it mimics acute appendicitis in a reported case[14]. Furthermore, the initial diagnosis is misleading, as reported in our case, where radiological exploration revealed an intra-abdominal abscess secondary to the sigmoid fistula. This is explained by the scarcity of this entity. Diagnosis is based on an abdominal CT scan revealing obvious thickening of the tumor wall and an air-fluid level within the tumor, suggesting that the ovarian tumor might have communication to the digestive tract[13].

On the other hand, the patient is deprived of the benefit of undergoing neoadjuvant chemotherapy, which will decrease the chances of complete macroscopic cytoreduction and metachronous metastasis following surgery, as exhibited by our case.

Surgery is not an easy matter. In fact, carcinological resection is associated with prolonged duration of surgery, increased perioperative blood loss, the necessity of postoperative transfusion, admission to intensive care units and prolonged hospital stay[15].

Some authors have described the spontaneous closure of colo-ovarian fistula, such as Imamura, who reported a case of a spontaneous resolution of a colo-ovarian fistula. However, this patient had a recrudescence of her symptoms after 46 years from the index event; therefore, she required surgery[16]. In fact, Shai *et al.*[17]. are the only authors who described conservative treatment and demonstrated that in selected patients in which no concrete evidence of infection was described, conservative management with antibiotics and delayed surgery can be considered for a fistula occurring in the setting of treatment-naïve high-grade serous ovarian carcinoma. This approach may result in a favorable surgical outcome. Indeed, fistulization does not always imply a tumor invasion. The chronic inflammation inflicted by the iterative twisting attacks or chronic pressure causes this fistulous communication[13]. However, considering the high probability of cancerous invasion causing fistulization, en-bloc resection is imperative in order not to leave a residual neoplastic niche.

Except for associated peritonitis when the anastomosis is prohibited, reconstructive of the digestive tract is a subject of concern because of nonprepared patients and non-optimized frail patients, thus explaining high fistula formation in the setting of optimal cytoreduction on elective condition of advanced ovarian cancer (11%), particularly in colorectal peritoneal carcinosis implants (6.4%)[18]. When deciding whether or not to perform anastomosis, the surgeon should assess the risk for fistula. In fact, peritoneal cancer index >20, more than two visceral resections, and multiple digestive resections are more prone to this complication[18]. A primary anastomosis is required in favorable conditions. In the case of an uncompensated hemodynamic state or peritonitis secondary to perforation or diffusion, transformation into a terminal stoma is the rule, as explained in our case.

Adjuvant therapies are essential to control tumoral seeding secondary to its perforation. Its management is similar to that of epithelial ovarian cancers, which are high-grade serous ovarian cancers[19]. Deemed to be a FIGO IIB tumor, the therapeutic sequence of management is based on either initial surgery followed by chemotherapy or neoadjuvant chemotherapy followed by surgery[20].

Conclusion

Fistulization to the large intestine worsens the prognosis of ovarian carcinosarcomas. Surgery is mandatory and should comply with oncological requirements. Reconstruction of digestive continuity should take into consideration the patient's characteristics, especially his hemodynamic status and intraoperative findings. Adjuvant therapy is mostly needed, although more studies should be conducted to delineate the regimen accurately.

Ethical approval

This case report was accepted for publication by our institutional ethics committee.

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent form is available for review by the editor-in-chief of this journal upon request.

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Not applicable.

Author contribution

All authors have contributed equally to the work.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

Guarantor

Rakia Siala.

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