

Case Report

Ileo-dermoid Fistula: A Rare Presentation of Ovarian Dermoid

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ABSTRACT

Mature cystic teratoma is a benign ovarian tumor that usually presents in reproductive-age females. This tumor usually presents with pain abdomen, bloating, and a lump. Hereby, we describe a case of an ovarian dermoid presented with features of intestinal obstruction secondary to ileo-dermoid fistula formation. A 55-year-old postmenopausal female presented with lower abdominal pain, nausea, vomiting, and the feeling of a lump in the abdomen. On evaluation and imaging, it was diagnosed as a large ovarian dermoid (with malignant transformation) with multiple fistulous communications with ileal loops. The patient was managed by laparotomy, total abdominal hysterectomy with bilateral salpingo-oophorectomy, bowel resection, and anastomosis. The patient was discharged in good condition. The rupture of malignant ovarian dermoid followed by enterodermoid fistula formation and intestinal obstruction is rare. Complete cytoreduction and bowel repair should be considered for optimal results.

KEYWORDS: Ileo-dermoid fistula, intestinal obstruction, malignant transformation, ovarian teratoma

INTRODUCTION

The benign ovarian tumors, mature cystic teratoma, commonly known as ovarian dermoid, account for 15% of ovarian tumors.^[1] This tumor manifests at a young age; 80% of the females on diagnosis are in their reproductive age. Common complications of it are torsion (16%), rupture (1%–4%), and infection (1%).^[2] Malignant transformations of dermoid are uncommon (1%–3%).^[3] Most of the time, malignant transformation manifest in advanced stages, with unfavorable outcomes when symptoms develop due to the invasion of nearby structures and metastasis.

Ovarian malignancy commonly presents with fullness, bloating, feeling of abdominal mass, weight loss, and loss of appetite.^[4] Immediate assessment should be made for intestinal obstruction or perforation risk if a patient presents with unusual symptoms such as vomiting, constipation, fever, and ovarian mass. Here, we discuss a case of ovarian dermoid that presented with pain, fever, and vomiting, which on evaluation, was found secondary to an ileo-dermoid fistula.

CASE REPORT

A 55-year-old postmenopausal female presented to the gynecology department with complaints of pain in the lower abdomen, feeling of a lump in the abdomen, loss of appetite, and weight loss for 3 months which was continuous, dull aching in nature, and increased in intensity from the past 1 week. Furthermore, there was a history of nausea and nonbilious vomiting 2–3 times a day for 1 week and fever for 1 day. On examination, she was conscious and oriented. Her temperature was 102.2°F, pulse was 112 beats/min, and blood pressure was 114/68 mm of mercury in the right arm in the supine position.

On examination, an abdominopelvic mass of 16-week size, firm to hard in consistency with irregular margins, and reduced side-to-side mobility was felt in the lower abdomen. There was mild tenderness in the lower

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abdomen. On per speculum examination, the cervix and vagina were looking healthy. On bimanual examination, the same mass was felt through the right and posterior fornices with restricted mobility. The cervix was felt anterior, with a senile size uterus. There were no palpable lymph nodes.

The ultrasound and contrast-enhanced computed tomography suggested a large heterogeneous mass of about 8 cm × 7 cm × 8 cm in the pelvis, containing fat and calcification with air in the rectouterine space posterior to the uterus and bilateral adnexa [Figure 1]. Two adjacent distal and terminal ileal loops showed fistulous communication with the mass leading to large areas of perforation. The omentum sealed the entire mass. Multiple enlarged ileocolic and mesenteric lymph nodes were seen. Mild inter-bowel fluid collection with pneumoperitoneum was seen in the pelvis. Multiple lytic lesions suggestive of metastases were seen in the lumbar and sacral vertebrae.

A combined gynecology and gastrosurgery team performed an exploratory laparotomy. Intraoperatively, 200 ml of foul-smelling ascitic fluid mixed with the fecal matter was drained and sent for culture. The right-sided ruptured dermoid cyst was seen impacted in the pouch of Douglas and adhered to the bowel [Figure 2a and b]. The cyst wall was necrotic, friable, and irregular, with a hairball within. Bowel perforation was present at two sites in the adjacent ileum. A 1.5 cm × 1 cm perforation

was noted in the ileum 50 cm proximal to the ileocecal (IC) junction. Another 0.8 cm × 0.5 cm perforation was seen 10 cm proximal to the IC junction. Multiple enlarged mesenteric lymph nodes were seen.

Bowel resection followed by primary anastomosis of the ileum and ascending colon was done with total abdominal hysterectomy and bilateral salpingo-oophorectomy [Figure 2c and d]. Abdominal wash was done, and an intraperitoneal drain was placed. Postoperatively, the patient was kept on total parenteral nutrition for 5 days and intravenous antibiotics. The patient had a surgical site infection for which resuturing was done on postoperative day 16.

The histopathology showed a cyst wall lined by stratified squamous epithelium with an invasive tumor composed of lobules and nests of atypical squamoid cells exhibiting moderate nuclear atypia [Figure 3]. The mature ectodermal component, mesodermal components, and endodermal components were seen. Ovarian surface and right-sided fallopian tube tumor involvement were seen. Section from the myometrium showed surface ulceration and tumor cell invasion in the myometrium. Sections from both the perforations in the bowel show the presence of an invasive tumor with cut margins of the bowel free from the tumor invasion. Ileocolic and mesenteric lymph nodes showed invasion. Features suggest squamous cell carcinoma arising from mature cystic teratoma involving the right fallopian tube, uterus, bowel loop, and lymph nodes. The tumor's pathologic stage classification (pTNM, AJCC 8th Edition) was pT2aN1bM1b.

The patient underwent six cycles of platinum-based chemotherapy after the surgery but died later due to a recurrence of malignancy.

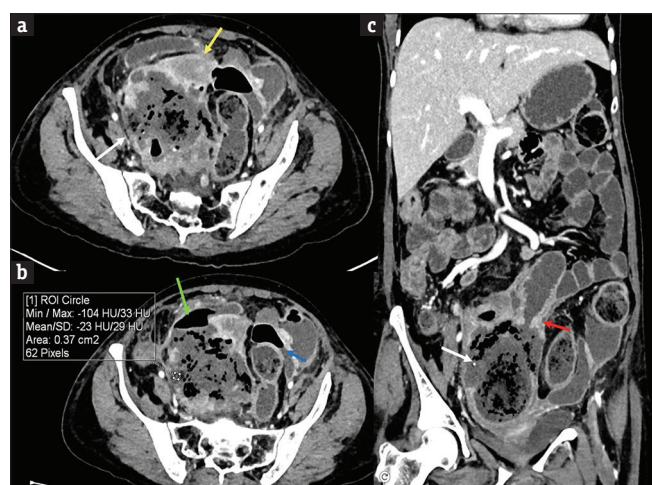


Figure 1: CECT of the abdomen. (a and b) Axial images show a large irregular, predominantly cystic mass with peripheral heterogeneously enhancing solid areas in the right adnexal region (white arrow) posterior to the uterus (yellow arrow). The mass shows areas of fat attenuation (indicated in ROI in B) along with multiple areas of mottled air foci, air-fluid level (green arrow), and extraluminal air (blue arrow). (c) The coronal oblique image shows foci of calcification (white arrow) within the mass. It depicts a direct fistulous communication between an ileal loop and the dermoid cyst (ileo-dermoid fistula - red arrow) with proximally dilated small bowel loops. CECT: Contrast-enhanced computed tomography, ROI: Region of interest

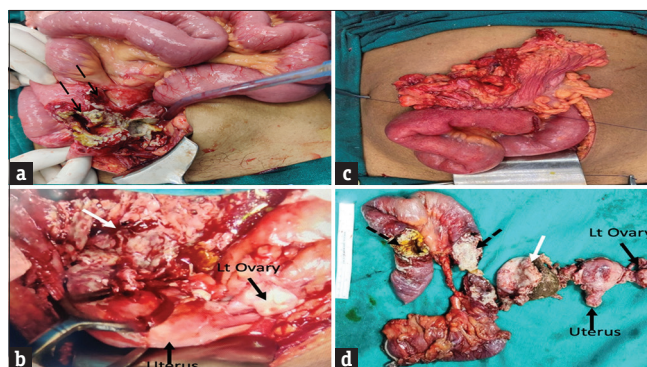


Figure 2: (a) Intraoperative images showing bowel loops with perforation (black dotted arrow). (b) Uterus with both tubes and ovaries. Right ovary showing ruptured ovarian dermoid (white arrow). (c) Resection and anastomosis of bowel loops. (d) Operative specimen of the uterus with both tubes and ovaries. Right ovary showing ruptured ovarian dermoid with a hairball (white arrow). The resected bowel loops show perforation (black dotted arrow)

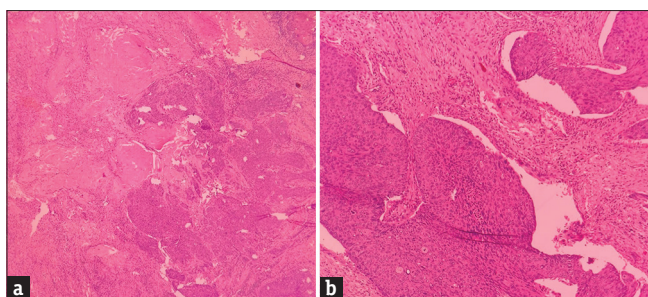


Figure 3: (a) Photomicrograph at $\times 10$ showing normal ovarian stroma (left side) with the presence of tumor nests of squamous cell carcinoma (right side); (H and E), (b) Photomicrograph at $\times 40$ showing tumor nests of squamous cell carcinoma (H and E)

DISCUSSION

Due to its rarity, very less is known about the malignant transformation of the ovarian dermoid. The management is planned according to its histological type.^[5] Histologically, malignant transformation of dermoid is squamous cell carcinoma in 80% of cases.^[5] Other types are adenocarcinoma, sarcoma, carcinoid, and struma ovary.

The bladder is the most common site for dermoid fistulae.^[6] Small and large bowel perforations are noted rarely. The bowel spread is usually by an invasion of serosa, and the disease rarely invades mucosa to form a fistula.^[7] The plausible causes for bowel invasion are ischemia and gangrene secondary to chemical peritonitis, infection, chronic pressure during labor, trauma, torsion, rupture, and malignant invasion.^[8]

The patient with such fistulas presents with pain, rectal bleeding, and the passage of dermoid content in the stool such as teeth and hairs.^[9] This case presented with uncommon features such as fever and vomiting. Conservative management with nil per orally followed by intravenous fluids and antibiotics can help in some fistula with benign dermoid.^[7] However, complete cytoreduction and management of the fistula during surgery are advisable in malignant dermoid.

The prognosis remains poor due to the advanced stage. A large study by the Taiwanese Society of Cancer on malignant transformation of dermoid concluded that early diagnosis and complete cytoreduction with additional concurrent/adjuvant chemotherapy is essential for better survival.^[5] This study also found that the age, size of the tumor, and the presence of solid components in a dermoid are important risk factors for malignant transformation. In our case study, the patient received chemotherapy but long-term survival was poor.

Esterson *et al.* in 2019 in his review, found that, including his case, there were only 7 cases of mature cystic teratoma that had documented intestinal obstruction, 29 cases had reported entero-dermoid fistula formation, and only 2 cases had malignant transformation with

fistula presenting as intestinal obstruction.^[2] This case report also shows similar mature cystic teratoma with malignant transformation and ileal fistula formation leading to intestinal obstruction and peritonitis.

In conclusion, if a patient with ovarian mass presents with vomiting and fever, it should be given immediate attention. The rupture of the dermoid followed by enterodermoid formation is a rare entity. Complete cytoreduction and bowel repair should be considered for optimal management.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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