

Ileal Endometriosis: a Rare Cause of Enterocutaneous Fistula

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Clinical Case

The patient is a 52-year-old woman with a remote history of right-sided nephrectomy and prior cesarean section who presented to the emergency department with 2 months of purulent, cutaneous drainage from her right lower abdomen (Fig. 1). She had been previously seen by her primary care physician for the aforementioned complaint and treated with 2 weeks of antibiotics to no resolution of drainage. On presentation, patient denied any constitutional symptoms of infection, changes to bowel habits, or unintentional weight loss. She was afebrile with no leukocytosis, hemodynamically stable, and exhibited only mild abdominal tenderness. Family history was not significant for pertinent oncologic or inflammatory bowel disease. Her last menstrual period was 2 years ago.

Computed tomography of the abdomen and pelvis demonstrated a cystic lesion within the right anterior hemipelvis measuring of approximately $7 \times 7 \times 5$ cm (Fig. 2a, b). The terminal ileum was observed to pass along the cystic lesion with evidence of oral contrast extrusion from the terminal ileum into the cyst. In addition, an adjacent collection of gas

and fluid was observed anterior to the bladder with cutaneous fistulization through the right lower abdominal wall. At that time, concomitant adnexal neoplasia or abscess could not be ruled out.

Given concern for a contained bowel perforation without clear etiologies, patient was taken to the operating room for exploratory laparotomy. Intraoperatively, the cystic mass was severely adherent to the terminal ileum as well as the pelvic floor. The right ovary was confirmed normal. A segment of the terminal ileum and the cystic mass were removed en bloc and an abscess cavity in the space of Retzius was entered and appropriately drained. The pathology report demonstrated that the cyst wall contains endometrial glands surrounded by smooth muscles, consistent with ileal endometriosis (Fig. 3a, b). No evidence of neoplastic changes was identified. The patient was discharged on postoperative day 6 without postoperative complications.

Discussion

Endometriosis has an established notoriety in masquerading as other diseases as it can involve extra-uterine organs, including the large bowel, rectum, small bowel, and appendix. Although it is relatively uncommon, previous case series report up to 5.4% incidence of gastrointestinal involvement, including a large series of 1573 patients with an intraoperative diagnosis of endometriosis.¹ Symptoms may correlate with location and extent of disease; for example, patients with recto-sigmoid involvement may present with constipation, blood per rectum, and diarrhea. Involvement of the appendix may present with appendicitis if the implants obstruct or compress the appendiceal lumen. Concurrently, the terminal ileum can also be involved with a variety of sequela, including obstruction, stricture or inflammation, intussusception, and volvulus. More severe cases have been described as contained or frank perforations with or without bleeding of the distal ileum.

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Fig. 1 Enterocutaneous fistula in the right lower abdomen

While spontaneous enterocutaneous fistula can be caused by a variety of disorders including inflammatory bowel disease, neoplasm, tuberculosis, and other rare conditions, to our

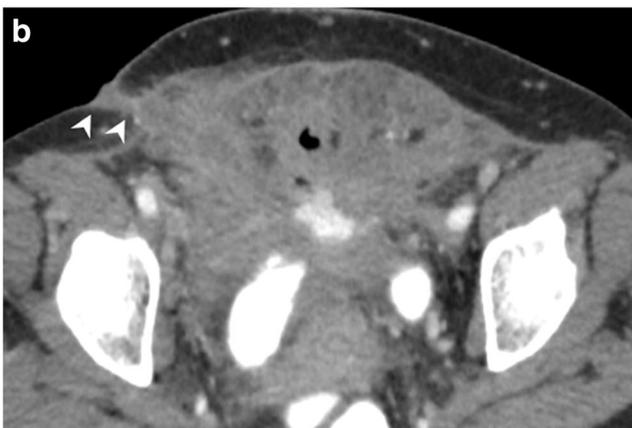
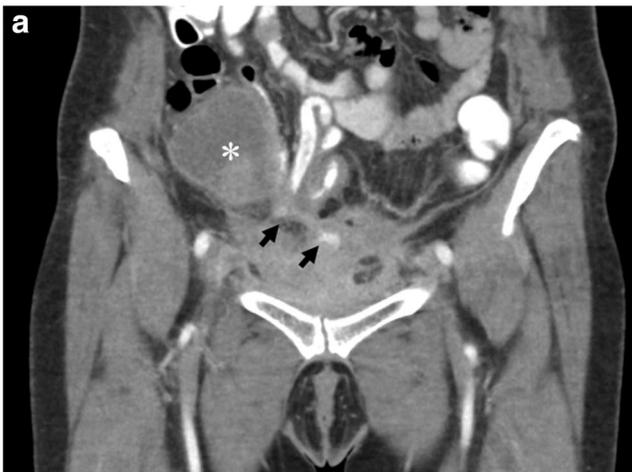


Fig. 2 Computed tomography of the abdomen and pelvis. **a** Contrast leak in the fistula tract between a cystic lesion (*) adjacent to the terminal ileum and abscess cavity in the space of Retzius (arrows), **b** fistula opening in the right groin (arrowheads)

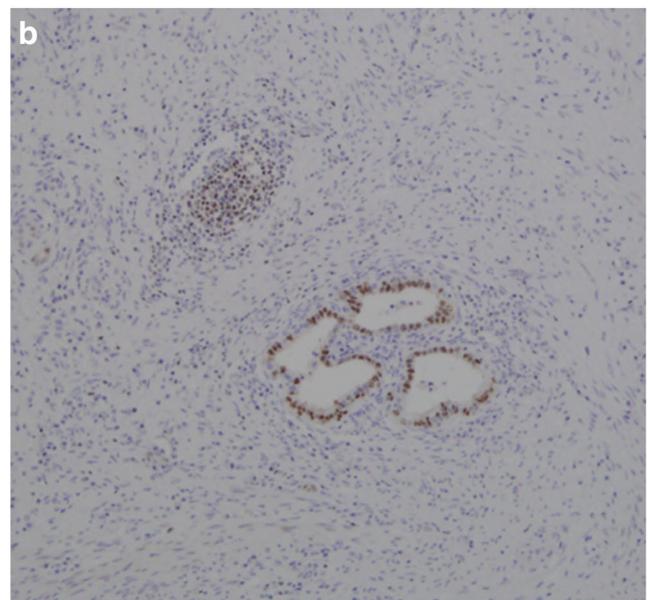
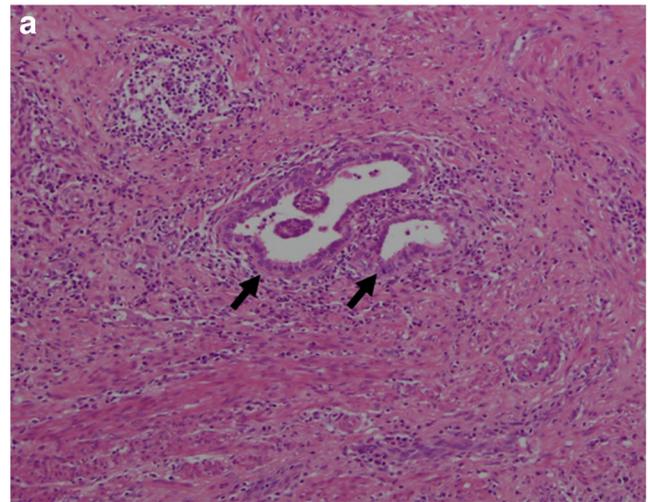


Fig. 3 Histological findings of the cyst wall. **a** Endometrial glands (arrows) surrounded by smooth muscles, **b** PAX8 stain positive for endometrial cells

knowledge, this is the first case report with spontaneous enterocutaneous fistula caused by ileal endometriosis. The standard work-up for enterocutaneous fistula should be explored, including a thorough history and physical focusing on exacerbating factors, such as foreign body, distal obstruction, prior radiation or inflammatory process, neoplasm, and epithelization of the tract. Given the varied manifestations of endometriosis, the preoperative diagnosis of ileal endometriosis can be challenging despite this extensive work-up, thus is often confirmed by pathologic examination of the surgical specimen. Recent reports have described this diagnostic dilemma in a case with endometriosis of the terminal ileum mimicking Crohn’s disease.² In addition, neoplastic transformation of gastrointestinal endometriosis has been reported, which can be influenced by long-standing inflammation and

increased growth or turnover rates.³ Thus, malignancy should always be considered in the differential diagnosis for these patients, particularly postmenopausal patients, presenting with an indolent history of disease without clear etiology.

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Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflicts of interest.

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