


Ovarian teratoma with bowel fistula managed non-operatively

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ABSTRACT

Background: Mature ovarian cystic teratomas (MOCT) are the most common benign ovarian tumours. They are mainly asymptomatic (60%) but can, in rare cases (<1%), complicate by forming a fistula into adjacent structures such as the bowel. Patients with fistulating ovarian teratomas are typically managed surgically, most often requiring laparotomy.

Case Presentation: We report a case of an 18-year-old woman who presented with vague abdominal and gynaecological symptoms. She expelled a mass with hair and teeth per rectum, which was later confirmed as a MOCT. We managed her conservatively in collaboration with the colorectal and gynaecology teams. At one-year follow-up, she remained asymptomatic with no evidence of a residual fistula.

Conclusion: This case highlights that while most MOCTs with bowel fistulation require surgical management, carefully selected patients may be managed conservatively with close follow-up.

Keywords: Mature ovarian cystic teratomas (MOCT), bowel fistula, case report, non-operative treatment.

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Background

Ovarian teratoma, a germ cell tumour, is the most common benign ovarian tumour [1]. Mature teratomas, synonymously referred to as mature ovarian cystic teratoma (MOCT) or dermoid cysts, account for >95% of all teratomas and 20%–25% of all ovarian tumours [2, 3]. MOCTs contain well-differentiated tissues from all three germ cell layers (ectoderm, mesoderm, and endoderm), macroscopically seen with structures such as teeth, hair, and bones [4].

MOCTs are typically asymptomatic in approximately 60% of cases, and are often discovered incidentally on imaging for unrelated clinical indications. However, symptomatic patients can complain of abdominopelvic pain or masses. Given the vague symptoms, these patients may present to either gynaecologists or general surgeons [2]. This is further complicated when patients develop a fistula and present with unusual clinical symptoms such as passing hair in stool [5].

Fistula formation in MOCTs is a rare complication seen in fewer than 1% of all cases [3]. The underlying pathophysiology is, however, not well studied [6]. In a series of 17 cases, Kizaki et al. reported of fistula formation

involving MOCTs and adjacent organs, such as the bladder and bowel. They identified inflammation (76.5%) as the most common cause, followed by malignant transformation (23.5%). All these cases were managed with laparotomy [3].

Patients with fistulating ovarian teratomas generally require laparotomy for surgical repair. Here, we report a case of an 18-year-old woman who spontaneously passed a MOCT per rectum and was managed conservatively. To the best of our knowledge, this represents the only documented instance of successful non-operative management to date.

Case Presentation

An 18-year-old nulliparous woman presented initially with intermittent right iliac fossa pain and copious yellow vaginal discharge. She had an ultrasound scan done at that time, which revealed the presence of a large lesion of unknown aetiology on the midline and right adnexa. In addition, it was noted that her copper IUCD was incorrectly positioned just above the cervix. She was reviewed by the Gynaecologists, the Copper IUCD was removed, a vaginal swab was taken for microscopy, culture, and

sensitivity (no organisms seen), and follow-up review after MRI scan of the abdomen and pelvis was arranged.

However, three months later, the patient re-presented to the emergency department with left iliac fossa pain, per rectal bleeding, and diarrhoea. Her pain was not associated with nausea, vomiting, or fever. Physical examinations revealed no signs of peritonitis. Blood test showed leucocytosis with (WBC $12.2 \times 10^9/L$; reference 4.0–11.0), thrombocytosis (platelet count $658 \times 10^9/L$; reference 150–400), neutrophilia ($8.5 \times 10^9/L$; reference 1.7–7.5), prolonged PT (13.0 sec; reference 9.0–12.0), elevated CRP (80 mg/L; reference <5) and raised CA 125 (75 KU/L; reference <35). Other tumour markers were within normal ranges, notably β -HCG (<1 IU/L; reference <1), Alpha-fetoprotein (2 KU/L; reference 1–6), and CEA (1 ug/L; reference <4).

Urgent Imaging was requested; however, on the same day, she spontaneously expelled a mass per rectum. Gross morphological inspection demonstrated well-developed tissues, including teeth and hair (Figure 1).

Investigations

CT scan done on her second presentation revealed a sizable pelvic collection with evidence of a large wall defect involving the distal sigmoid/rectosigmoid junction with surrounding active inflammatory change and free fluid (Figure 2). Retrospective review of the scans at the Gynaecology MDT, a month later, identified evidence



Figure 1. Image of gross expelled mass showing teeth and hair

of material within the bowel (teeth) compatible with a dermoid.

Furthermore, an MRI pelvis was also conducted three days after the CT-scan, which showed a moderate volume of partially loculated pelvic free fluid and a 6.4 x 5.9 cm heterogeneous, gas-containing central pelvic collection communicating with a wide fistulous defect within the anterior wall of the mid sigmoid colon, which appeared to be left Tubo-ovarian in origin (Figure 3).

Histopathology analysis two weeks later confirmed the presence of teeth, hair, sebaceous glands, and concluded a diagnosis of mature ovarian teratoma.

Differential Diagnosis

Our differential diagnoses evolved as the patient's symptoms changed. At the first presentation, pelvic inflammatory disease with a tubo-ovarian abscess was considered, given the vaginal discharge and ultrasound findings of a malpositioned intrauterine device. Following the development of left iliac fossa pain and rectal bleeding, colorectal pathology such as colitis or inflammatory bowel disease was entertained. Ultimately, the passage of tissue per rectum, together with cross-sectional imaging and histology, confirmed the diagnosis of a fistulating ovarian teratoma.

Treatment

The patient was not keen on any invasive treatment or investigations, which guided our choice of management. She was managed conservatively in collaboration with the Colorectal team and discharged following a 5-day inpatient stay. She remained asymptomatic and returned back to regular activity within two months.

Outcome and Follow-up

Follow-up MRI scans at one year demonstrated no definitive evidence of a residual fistula. Although she initially declined endoscopy, she subsequently consented to flexible sigmoidoscopy for further evaluation, which at 21 months identified scarring at the site of the previous sigmoid fistula with no evident fistula opening (Figure 4). The patient was reassured.

Discussion

Ovarian teratoma with bowel fistulation is a rare condition, described in both paediatric and elderly patients, with the youngest documented case reported in a 9-year-old girl and the oldest in an 85-year-old woman [7, 8]. The first reported case in literature was in 1965 [9]. Our patient was an otherwise healthy young woman, under 20 years of age, who presented with vague abdominal and gynaecological symptoms. Importantly, this case is distinctive in documenting successful conservative management, with no apparent complications observed at 21 months follow-up.

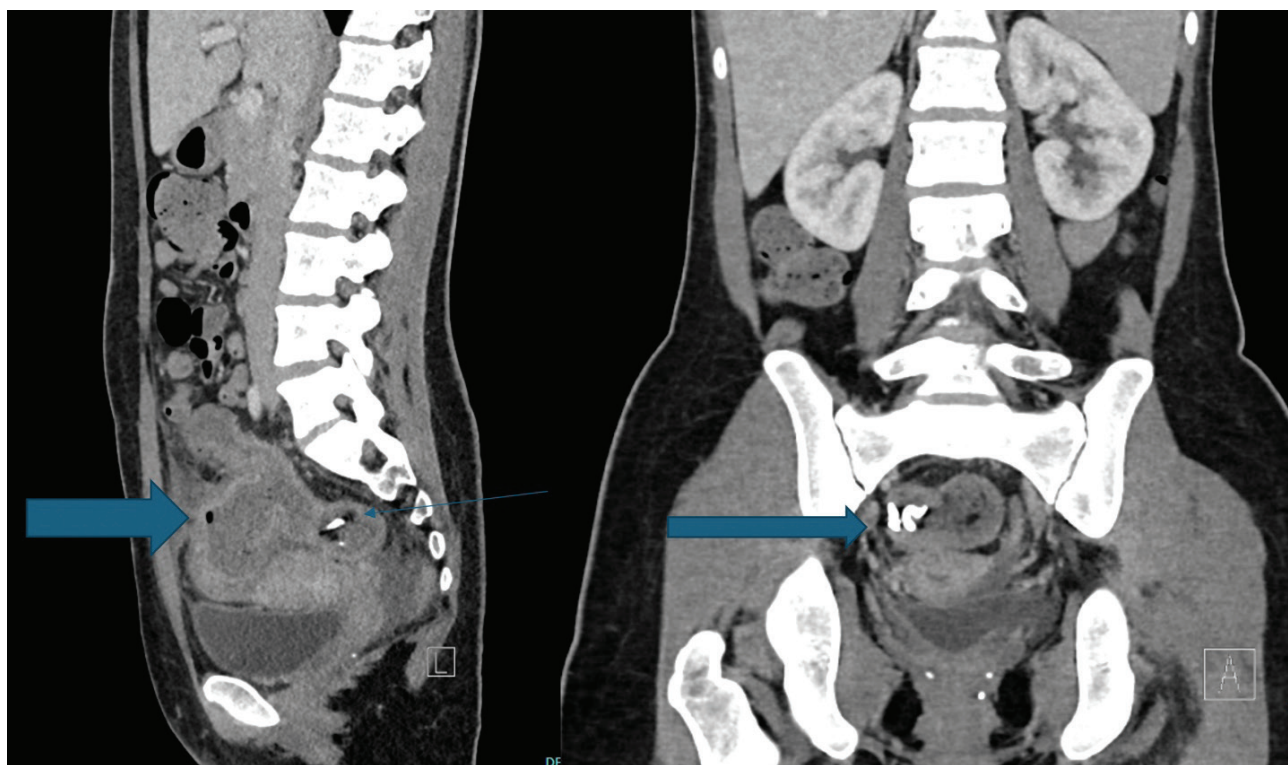


Figure 2. CT-scan – Dermoid within the sigmoid colon. Sagittal View: Small Blue Arrow demonstrates Dermoid in Rectum; Large Blue Arrow shows fistula in sigmoid colon. Coronal View: Large Blue arrow highlights the Dermoid cyst.

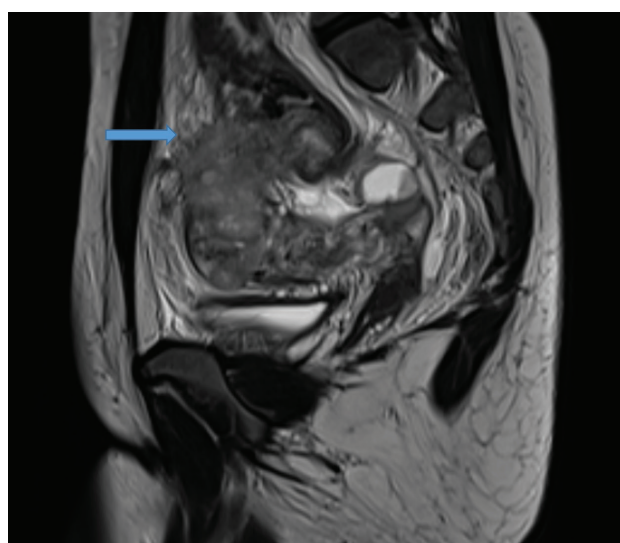


Figure 3. MRI T2 Sagittal sequence- Dermoid passed, but fistula into the sigmoid evident as shown by the blue arrow.

Approximately 80% of females diagnosed with ovarian teratomas are of reproductive age [10]. Abdominal pain is one of the most frequently reported presenting symptoms [11]. Other manifestations reported include gastrointestinal symptoms such as rectal bleeding, passage of hair with stool, diarrhoea, and tenesmus [12]. Genitourinary symptoms may be found in cases involving the urinary system. These include urinary frequency, dysuria, and haematuria [5]. Unfortunately, the vague and heterogeneous nature of these symptoms makes diagnosis challenging. Our patient

initially presented with right iliac fossa pain, which later localised on the left side, underscoring the potential for misdiagnosis, particularly if confounding factors and red herrings such as a misplaced IUCD are present. Radiological imaging and histology are essential in confirming the diagnosis.

Choice of imaging depends on local availability. Ultrasound is usually considered first line due to ease of access and minimal invasiveness [13]. CT scan has higher sensitivity (93%-98%) compared to ultrasound, and provides detailed cross-sectional images, enabling identification of characteristic features and relationship to surrounding viscera [13, 14]. Additionally, MRI offers superior soft tissue resolution, detecting features potentially missed on CT [13]. In our case report, an emergency CT revealed a distal sigmoid colon wall fistula defect and pelvic collection. This was subsequently confirmed on the MRI of the pelvis. Although initially missed, the fistulated teratoma was retrospectively seen on review of the CT scan at an MDT meeting. Endoscopy, although invasive, enables direct visualisation and tissue diagnosis. Flexible sigmoidoscopy was later performed despite initial resistance from our patient. Overall, multimodal imaging is necessary to enhance diagnostic accuracy and identify complications.

Ovarian teratomas that fistulate into the bowel present a complex clinical scenario. The majority of published case reports involve surgical treatment with laparotomy or laparoscopy. This is possibly due to the extent of

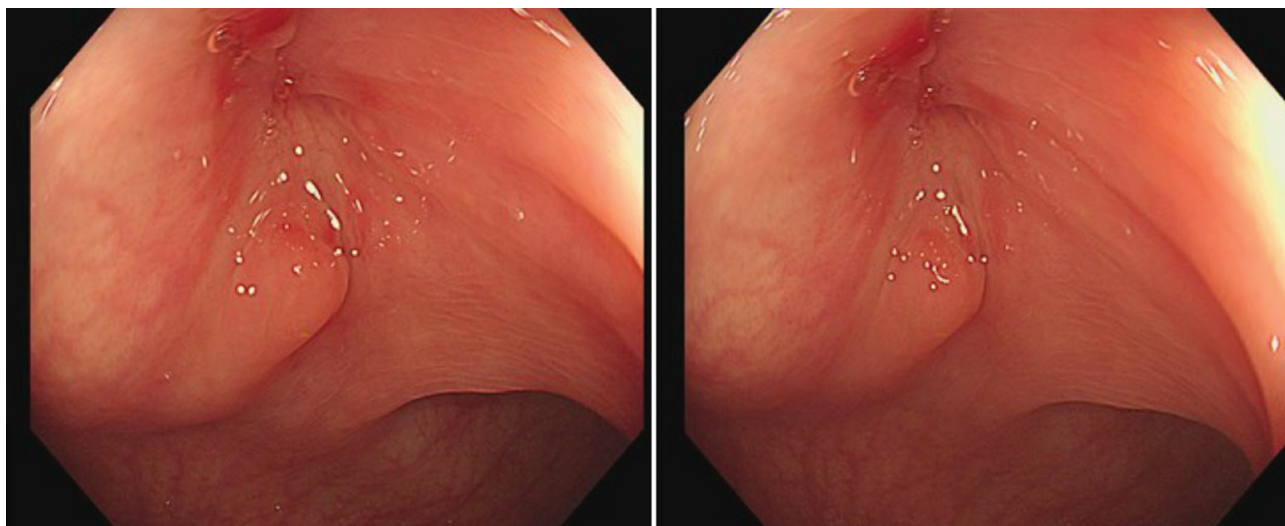


Figure 4. Distal Sigmoid Flexible sigmoidoscopy.

pathology, the need for direct visualisation, and histological assessment [3]. To the best of our knowledge, this case report is the only documented instance of conservative management for fistulating disease. Standard surgical management involves excision of the teratoma, repair or resection of affected bowel or adjacent structures, and commonly unilateral or bilateral salpingo-oophorectomy. Therefore, young patients should be counselled on the risk of loss of fertility. Our patient was 18 years old at the time of presentation and was not keen on invasive treatment. Additionally, our patient fully expelled the dermoid in whole, as later confirmed by MRI imaging. Conservative treatment, however, carries risks such as the malignant transformation into squamous cell carcinoma, which is associated with poor prognosis [8, 10]. For this reason, close follow-up with imaging was instituted to detect any complications early. A multidisciplinary approach involving surgeons, radiologists, gynaecologists, and other relevant specialists is essential to optimise clinical outcomes.

Limitations

Patient follow-up was limited to 21 months. Management was influenced by patient preference, and the rarity of the condition limits comparative analysis.

Conclusion

This case highlights that while most MOCTs with bowel fistulation require surgical management, carefully selected patients may be managed conservatively with close follow-up. This points to the importance of individualised multidisciplinary care, taking into consideration patient factors such as age and patient wants. A rigorous systematic review is recommended to identify a wider range of cases and management options.

Although most MOCTs with bowel fistulation require surgery, this case demonstrates that carefully selected

patients may be managed conservatively with close surveillance. Individualised multidisciplinary care, considering patient age and preferences, is essential. A systematic review is warranted to provide a comprehensive evaluation of the available literature on this condition.

What Is New?

- Ovarian teratoma with bowel fistulation is a rare complication, reported in women ranging in age from as young as 9 years to 85 years old.
- Clinical presentation can be vague, with overlapping gastrointestinal and gynaecological symptoms, which might delay timely diagnosis.
- Passage of tissue containing hair or teeth per rectum is pathognomonic of a mature ovarian cystic teratoma and should raise the index of suspicion for fistulating disease.
- Cross-sectional imaging and histopathology are essential for confirming the diagnosis and defining the extent of fistulation.
- Cases reported in the literature have been managed surgically; to the best of our knowledge, this is the first case demonstrating conservative management.

List of Abbreviations:

CEA	Carcinoembryonic Antigen
HCG	Human Chorionic Gonadotropin
MOCT	Mature Ovarian Cystic Teratoma
PT	Prothrombin Time

Conflict of interests

The authors declare that there is no conflict of interest regarding the publication of this article.

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Consent for publication

Informed written consent was given by the patient for the publication of this case report and accompanying images. All patient identifiers were removed to ensure anonymity.

Ethical approval

The authors assessed the case in line with the UK National Research Ethics requirements. Formal Ethical approval is not required at our institution to publish an anonymous case report.

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Summary of the case

1	Patient (age, gender)	18 years, female
2	Final diagnosis	Mature ovarian teratoma with bowel fistula
3	Symptoms	Right iliac fossa pain, yellow vaginal discharge, later left iliac fossa pain, per rectal bleeding, diarrhoea
4	Medications	N/A
5	Clinical Procedure	Conservative management – no surgery performed
6	Specialty	General Surgery, Gynaecology