

Benign Ovarian Dermoid: A Rare Cause of Sigmoid-Ovarian Fistula

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Abstract

The case of a young female with lower gastrointestinal symptoms secondary to fistulisation of benign ovarian teratoma into the sigmoid colon is reported and the relevant literature briefly reviewed.

Keywords: Teratoma; Malignancy; Salpingo-oophorectomy; Sigmoidectomy.

Introduction

A gastrointestinal fistula is an abnormal communication of part of the intestine with the skin (external fistula) or adjacent organs (internal). In the vast majority of cases (75 – 85%), they occur as a post-operative complication of surgery on gastrointestinal tract; occasionally, they occur spontaneously, secondary to inflammatory bowel disease, diverticulitis, pancreatitis, appendicitis, radiation, malignancy or trauma [1, 2]. Colonic fistula are uncommon; internal colonic fistula even more so [3]. These usually occur secondary to diverticular disease and usually communicate with the urinary bladder (colovesical) or vagina (colovaginal) [3]. We present the unusual case of a young lady with colo-ovarian fistula secondary to erosion of the colon by a benign ovarian dermoid.

Case report

A 32 year female presented to the OPD with lower abdominal discomfort, loose stools and purulent discharge per rectum for 2-3 months. There was no history of blood in stools, fever, constitutional symptoms or similar episodes in the past. Her abdominal examination was unremarkable, as was digital rectal examination and proctoscopy. She was advised abdominal ultrasound and colonoscopy; ultrasound revealed a 6.4 x 6.1 x 5.3 cms complex cystic mass in the left adnexa with possibility of air fluid level in the mass. There was no other significant finding. Colonoscopy revealed an ulcer at 20 cm from anal verge. This had a clean base and surrounding unhealthy mucosa, biopsy of which revealed inflammatory pathology with no chronicity/malignant activity. Her baseline haematological, biochemical investigations and Serum CEA, CA125 were within normal limits. A computerised tomogram (CT) scan was done,

and this revealed a 7 x 8.2 x 7.3 cms well defined, peripherally enhancing mass in the left adnexa with air and calcific foci and possible communication with the sigmoid colon (**Figure 1**).



Figure 1: CECT scan showing a large mass in the pelvis, with air foci within. The mass is compressing the sigmoid colon.

She was taken up for elective exploratory laparotomy through a midline vertical incision. Intraoperatively, there was a large cyst in the left ovary that was densely adherent to the left fallopian tube, sigmoid colon and urinary bladder. En-bloc left side salpingo-oophorectomy and segmental resection of sigmoid colon was done, with an end to end colo-colic anastomosis and proximal diversion ileostomy. On cutting the sigmoid colon, a fistulous opening with the ovarian cyst was identified (**Figure 2**). Histopathology revealed mature cystic teratoma with fistulisation to the sigmoid colon; there was no evidence of malignancy. The ileostomy was restored after a month, and the patient has remained well over a follow up of almost 18 months.

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Figure 2: Resected specimen showing cut open sigmoid colon with probe inserted into fistulous tract.

Discussion

Ovarian mature cystic teratoma (dermoid) are the most common type of ovarian tumour. These are benign germ cell tumours that are commonly seen between the ages of 20-40

years. These are usually asymptomatic, and are detected on routine pelvic examination. Symptoms occur when complications develop, such as torsion (16%), rupture (1- 4%), malignant transformation (1-2%), infection (1%) or invasion into adjacent viscera [4, 5]. Perforation of a dermoid cyst into the peritoneal cavity or fistulisation into adjacent organs is rare, seen in less than 1% of cases [4, 5]. When fistulisation occurs, it most commonly occurs with the urinary bladder [6]. The lower gastrointestinal tract has traditionally been considered an uncommon site for fistulisation by ovarian dermoid cysts, a literature review by Kizaki et al reported large bowel fistula (transverse colon, sigmoid and rectum) to be more common [5]. They identified 17 cases of ovarian dermoid complicated by fistula formation in literature (from 1938 till 2016), to which we are able add another 9 cases reported from 2016 till date, including the one reported here (**Table 1**) [7-13].

Table 1: Cases of ovarian dermoid with fistulisation reported from 2016 onwards.

Author (Year)	Age	Symptoms	Involved organ	Cause
Kim et al (2017) [7]	17	Lower abdominal pain, sebaceous material in stools, haematochezia	Rectum	Benign
Chan et al (2017) [8]	47	Lower abdominal pain, bleeding per rectum	Sigmoid colon	Benign
Prasad et al (2018) [9]	33	LUTS (urgency, frequency, dysuria)	Urinary bladder and Sigmoid colon	Benign
Chauhan (2018) [10]	55	Abdominal pain, passage of hair and cheesy material in stools	Ileum	Benign
Matsuzono (2019) [11]	25	Diarrhoea, weight loss	Rectum	Benign
Matsuzono (2019) [11]	22	Diarrhoea, weight loss	Sigmoid colon	Benign
Esterson (2019) [12]	59	Subacute intestinal obstruction	Ileum	Malignancy
Camerlo (2020) [13]	68	Lower abdominal pain	Sigmoid colon	Malignancy
Present Case (2022)	32	Lower abdominal discomfort, loose stools and purulent discharge per rectum	Sigmoid colon	Benign

The mechanism of fistulisation is not certain. It is hypothesised that fluid leak due to rupture of the cyst due to partial torsion, malignant change, trauma, infection or chronic pressure leads to dense adhesions between the cyst and the neighbouring organ. Further inflammation and necrosis results into a fistula. Although infiltration by malignant cells from a dermoid having undergone malignant transformation has also been hypothesised as a cause of fistula formation, it is now believed that malignancy is not essential for fistula formation; [5, 6, 10] this is further strengthened by Kizaki’s review in which only 4 (23.75%) had an underlying malignancy; the vast majority (13 cases, 76.5 %) were benign [5]. We feel that in addition to these mechanisms, the narrow, unyielding confines of the pelvis within which these tumours enlarge also contributes by chronic compression of neighbouring viscera (bowel, bladder) with consequent adhesions, ischemic changes and erosion.

Once the fistula forms, the symptoms of the organ involved become apparent [5]. Weight loss, nausea, diarrhoea, abdominal pain, distension, obstruction, bleeding per rectum and dyspareunia have all been reported when the small or large

bowel are involved. When the urinary bladder is involved, the patient may have dysuria, pyuria, haematuria or bladder calculi [5, 6]. Given this wide and varied symptomology, the patient can present to any surgical or medical outpatient; given the rarity of this particular complication, the diagnosis is usually not possible without investigations. Abdominal sonography may detect features such as cystic, solid or complex ovarian mass with high acoustic shadowing due to presence of hair, teeth and fat, high amplitude echoes, ‘tip of iceberg,’ ‘dot-dash’ sign (due to echogenic hair floating within) and fat-fluid or fluid-fluid levels [4]. CT and MRI provide far better sensitivity in diagnosis as well as detecting malignant transformation especially when combined with gadolinium; [4, 14, 15] the presence of air foci, pooling of oral contrast and fluid filled track communicating with bowel and mass should alert the clinician to the possibility of fistulisation [10]. In our patient, the presence of air-fluid level within the cystic lesion alerted us to the possibility of fistulisation with the sigmoid colon, where an ulcer had already been detected on colonoscopy. The treatment is surgical. Care must be taken to rule out malignancy

by pre-operative tumour markers and imaging. Resection of the lesion along with involved intestine and histopathological examination is mandatory.

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