

Sclerosing encapsulating peritonitis and methotrexate

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Introduction

Sclerosing encapsulating peritonitis (SEP) is a rare but a life threatening condition that is difficult to diagnose and is usually limited to the peritoneal dialysis population. We report a rare case of SEP with methotrexate use.

Case report

A 36-year-old female presented with a 6-month history of intermittent obstructive small bowel symptoms and an associated weight loss of 19 kg. Her symptoms began 8 weeks after a short pulse course of methotrexate (each dose of 110 mg) for a molar pregnancy. Other past medical history included an excision of abdominal lipoma, Caesarean section and a fetal death *in utero*.

Outpatient investigations, including ultrasound and gastroscopy, did not find any cause. With worsening symptoms, she was admitted to a hospital. Clinical findings were those of a non-distended abdomen associated with mild epigastric tenderness. Other than a CRP of 65, her blood investigations were unremarkable with a beta-HCG below 2. Abdominal X-ray revealed small bowel dilation with multiple air-fluid levels and collapsed large bowel. Computerised tomography (CT) scan confirmed dilatation of the small bowel with distal collapse of the colon, compatible with a distal small bowel obstruction. A subsequent small bowel follow-through examination established dilated small bowel despite a transit time of only 3 hr.

The patient was managed conservatively with an insertion of nasogastric tube analgesia and antiemetics. Her symptoms manifested on resumption of oral intake, and a surgical opinion was sought.

She underwent a laparotomy and a 3-hr division of adhesions. The intraoperative findings were of severe peritoneal fibrosis and 'cocooning' of the small bowel. The last metre of ileum was found to be compacted into a 10-cm fibrous 'ball'. She spent a further 10 days in hospital with an uncomplicated recovery and resolution of symptoms.

Samples of the peritoneal adhesions were sent for histology and showed features of sclerosing inflammatory peritonitis. Histological sections demonstrated dense fibrous connective tissue, within which bands of amianthoid collagen fibres, admixed with chronic inflammatory cells and reactive fibroblasts.

Discussion

Sclerosing encapsulating peritonitis (SEP) is a poorly understood condition first described in the 1980s. In this condition, excess fibrosis results in an encasement of the small bowel, resulting clinically in obstruction.¹ This process tends to affect the small bowel, although the liver, stomach or duodenum may be rarely involved.

The condition, which usually manifests in young females, can be classified as either idiopathic or secondary depending on the aetiology. The latter may be iatrogenic (beta-blockers) or related to sarcoidosis or systemic lupus erythematosus, and is often described in the context of peritoneal dialysis.² Interestingly, it has been noted that cessation of the aetiological agent does not necessarily imply resolution of the condition.²

Patients often describe symptoms of abdominal pain, anorexia, nausea and weight loss and may clinically have an acute small bowel obstruction. Ascites has been described in the literature and, biochemically, patients may have an elevated CRP.³

There are pitfalls associated with radiological diagnosis. Plain abdominal films may appear normal early in the disorder. The most sensitive modality is ultrasound, which shows thickened bowel walls with a trilaminar appearance along with increased peristalsis. Diagnostic features on CT scan include thickened, adherent bowels with small bowel dilation; however, these changes appear later than ultrasound.³

Methotrexate is an agent that is used in several conditions including psoriasis, rheumatoid arthritis and various neoplasms including gestational trophoblastic disease, leukaemia and breast carcinoma. Its mechanism of action is to inhibit DNA synthesis by preventing the conversion of folic acid to tetrahydrofolic acid. Methotrexate is known to be associated with serosal complications, including pleuritis and pericarditis.⁴ In one study of 168 subjects, all treated with methotrexate for gestational trophoblastic disease (GTD), 42 developed pleuritis as judged by clinical history and absence of radiological signs (normal chest X-ray).⁴ The pathophysiology remains poorly understood.

Unlike methotrexate-induced pleuritis, there has been only one case report of methotrexate-induced peritonitis in the literature, solely based on clinical history and examination alone. Hence, no tissue diagnosis was available nor were there intraoperative findings.

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Our report of operative findings of sclerosing encapsulating peritonitis with confirmed histology after methotrexate treatment adds weight to causal relation between methotrexate and sclerosing encapsulating peritonitis.

References

- 1 Cohen O, Abrahamzon J, Ben-Ari J, Frajewisky V, Eldar S. Sclerosing encapsulating peritonitis: Primary and secondary forms. *J Clin Gastroenterol.* 1996; 22: 54–57.
- 2 Laloo S, Krishna D, Maharajh J. Abdominal cocoon associated with tuberculous pelvic inflammatory disease. *Br J Radiol.* 2004; 75: 174–176.
- 3 Perks FJ, Mutchison JT, Gibson P, Jackson SHL. Imaging finding in sclerosing encapsulating peritonitis. *J Royal College Physicians Edin.* 2004; 34: 116–119.
- 4 Sharma S, Jagdev S, Coleman R, Hancock B, Lorigon PC. Serosal complications of single agent low dose methotrexate used in gestational trophoblastic disease: First reported case of methotrexate induced peritonitis. *Br J Cancer* 1999; 81: 1037–1041.

Incarcerated uterine procidentia and vesical calculi: A case report

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Introduction

In the English language scientific literature, there have been five previous case reports over the past 47 years of uterine procidentia which were incarcerated secondary to bladder calculi. We present the sixth case of this uncommon association.

Case report

A 76-year-old Balinese widow, gravida VIII, para VIII, presented with a 9-month history of a mass protruding from the vagina and a 1-month history of dysuria.

On admission to hospital, the patient was afebrile; BMI = 25, blood pressure 180/90 mmHg, chest, breast and abdominal examination normal.

Vaginal examination revealed an irreducible complete uterine procidentia. During an attempted reduction, there was constant urinary leakage. The passage of a metallic urinary catheter per urethra resulted in the tell-tale sound of 'impact on stone' consistent with the presence of intravesical calculi.

Preoperative investigations included haemoglobin 133 g/L, white cell count 7177 per mL and normal liver and renal function tests. Urinary culture confirmed *Escherichia coli* infection sensitive to cephalosporin. Chest X-ray and electrocardiogram confirmed the patient's fitness for anaesthesia.

The patient proceeded to vaginal cystolithotomy, hysterectomy and colpocleisis under spinal anaesthesia.

A longitudinal incision was made through the prolapsed anterior vaginal wall and the bladder was dissected free. A 3.5-cm-longitudinal incision was made into the anterior wall of the bladder, and three bladder calculi, each 5 cm in diameter, were extruded through the incision. A two-layer repair of the bladder incision was performed and, thereafter,

a vaginal hysterectomy with colpocleisis was completed. The patient was catheterised with a 14-gauge silastic catheter postoperatively. The estimated blood loss was 350 mL. Intraoperative intravenous cephalosporin and metronidazole were administered and continued for 5 days postoperatively.

The urinary catheter was removed on the tenth postoperative day and the patient was discharged home well. A postoperative follow up 2 months later confirmed her uneventful recovery.

Discussion

Incarcerated uterine procidentia has become a rarely reported condition although less so in developing countries. This clinical entity should be dealt with as a gynaecological emergency because of the potential urinary obstruction.¹

While bladder calculi remain the most common cause of incarceration, other causes include gross prolapse associated with multiparity, trauma, intestinal obstruction and cervical cancer.^{2–6}

Older reports¹ quoted a 30% mortality rate with death from septicaemia, urinary complications, ischaemia of the uterus and bowel strangulation; however, with contemporary surgery, excellent results have been achieved.

While most reported cases have undergone vaginal cystolithotomy, it has been with an acknowledged greater chance of vesicovaginal fistula, depending on the state of the tissues at the time of operation.⁶ Neider *et al.*⁷ suggested that suprapubic cystolithotomy may be the preferable route for removal of the calculi as it could allow a more complete evaluation of

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