CASE REPORT

Bilateral ureteric obstruction during in vitro fertilisation therapy in a patient with complex surgically managed Crohn's disease

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SUMMARY

A 37-year-old woman receiving in vitro fertilisation (IVF) treatment presented with lethargy, fevers and anuria. Her background included complex surgically managed Crohn's disease and mild right-sided hydronephrosis (with no evidence of obstruction on MAG3 renogram). On examination, she had a distended abdomen with generalised tenderness. Blood tests revealed an acute kidney injury and confirmed sepsis. She was found to have bilateral ureteric obstruction with worsening of the right-sided hydronephrosis and new-onset left-sided hydronephrosis secondary to bilateral ovarian masses, diagnosed as mild ovarian hyperstimulation syndrome (OHSS). She was initially managed with fluid resuscitation, bilateral nephrostomies, antibiotics and supportive management of mild OHSS. She made a good recovery and was discharged after placement of antegrade stents and removal of nephrostomies. On follow-up, following resolution of ovarian hyperstimulation, she has been diagnosed with a right-sided distal ureteric stricture and is awaiting reconstruction. Her left ureteric stent has been removed.

BACKGROUND

To the best of our knowledge, this is the first case of bilateral ureteric obstruction secondary to mild ovarian hyperstimulation syndrome (OHSS) with a background of Crohn's disease complicated by anuria and sepsis. We examine the significance of Crohn's disease to the pathophysiology of OHSS, as well as making recommendations for practice.

CASE PRESENTATION

A 37-year-old woman was admitted with the symptoms of lethargy, fevers and anuria. On examination, she had a distended abdomen with generalised tenderness. Her background included extensive Crohn's disease for which she had had multiple operations, including an ileocaecal resection with excision of fistulae, and a proctectomy with end colostomy which was complicated by a bladder injury. She had been investigated for an incidental finding of right-sided hydronephrosis with a MAG3 renogram, which showed good drainage with no evidence of obstruction and 44% function on the right. She also suffered from recurrent urinary tract infections (UTIs). She had recently undergone egg collection following hormonal ovarian stimulation as part of in vitro fertilisation (IVF) treatment.

INVESTIGATIONS

Admission blood tests revealed an acute kidney injury (AKI) and raised inflammatory markers. Haematocrit and total serum protein were both within normal limits. An ultrasound scan of her urinary tract demonstrated moderate right-sided and mild left-sided hydronephrosis and adnexal masses of unknown origin; the bladder was empty. Abdominal ascites was not evident. A subsequent CT scan confirmed bilateral hydronephrosis secondary to enlarged hyperstimulated ovaries (figures 1 and 2). Initially, there was concern of moderate/severe ovarian hyperstimulation syndrome (OHSS); the final diagnosis was of mild OHSS with bilateral ureteric obstruction and sepsis.

TREATMENT

Treatment involved intravenous fluids and antibiotics before immediate placement of bilateral nephrostomies. The right drained frank pus (from which *Escherichia coli* was cultured), and the left drained contaminated urine. She was treated with antibiotics for a week before undergoing placement of bilateral antegrade stents and removal of nephrostomies.

OUTCOME AND FOLLOW-UP

Our patient made an uneventful recovery and was discharged after 10 days following resolution of her

Figure 1 CT, coronal section. Multiloculated pelvic lesion causing bilateral hydronephrosis.

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Figure 2 CT, transverse section. Multiloculated pelvic lesion.

Figure 3 Retrograde contrast study, composite. Right: long segment of distal ureteric narrowing. Left: possible short segment of distal ureteric narrowing.

AKI and with normal inflammatory markers. A subsequent pelvic ultrasound showed reduction in size of the ovaries to within normal limits. She underwent a cystoscopy and retrograde contrast studies to ascertain if her stents could be removed; distally, she was found to have a long ureteric narrowing on the right and a possible 1 cm narrowing on the left, and so stents were left in situ (figure 3). She was managed with regular stent changes at her request while she continued with IVF treatment. In light of her high pregnancy risk in combination with failed IVF cycles, she opted for surrogacy; following her final egg collection, her gestational surrogate is pregnant. Subsequent ureteroscopy and retrograde contrast studies suggested no significant stricturing on the left and so the left ureteric stent has been removed. She is currently awaiting right-sided ureteric reconstruction.

DISCUSSION

Although enlarged ovaries are a normal side-effect of IVF treatment, OHSS is a potential complication. Exogenous

Table 1 Classification of severity of OHSS by symptom

Degree of OHSS	Possible clinical symptoms
Mild	<ul style="list-style-type: none"> ▶ Abdominal bloating ▶ Mild abdominal pain ▶ Ovarian size usually <8 cm
Moderate	<ul style="list-style-type: none"> ▶ Nausea±vomiting ▶ Ultrasound evidence of ascites ▶ Ovarian size usually 8–12 cm
Severe	<ul style="list-style-type: none"> ▶ Oliguria ▶ Haemoconcentration haematocrit >45% ▶ Hypoproteinaemia ▶ Ovarian size usually >12 cm
Critical	<ul style="list-style-type: none"> ▶ Haematocrit >55% ▶ White cell count >25 000/mL ▶ Oligo/anuria ▶ Thromboembolism ▶ Acute respiratory distress syndrome

Adapted from RCOG. The management of ovarian hyperstimulation syndrome. Green-top Guideline No. 5. 2016.¹

OHSS, ovarian hyperstimulation syndrome; RCOG, Royal College of Obstetricians and Gynaecologists.

gonadotropin can hyperstimulate the ovaries, which release various vasoactive peptides in response (including vascular endothelial growth factor). This causes an increase in vascular permeability, resulting in an increased ovarian size and a shift of fluid extravascularly to third space compartments. The most severe forms of OHSS have an estimated incidence of 3–8% and can be life-threatening. Clinical features vary by severity, with more severe forms characterised by abdominal pain, ascites, nausea and vomiting and renal failure (table 1).¹

There are six previously reported cases of ureteric obstruction secondary to varying degrees of OHSS. In two patients, the obstruction was unilateral,^{2 3} and in four, bilateral.^{4–7} We present the first case of bilateral ureteric obstruction secondary to mild OHSS with a background of Crohn's disease complicated by anuria and sepsis.

Our patient had extensive Crohn's disease requiring multiple operations and had previously suffered gastrointestinal obstruction secondary to adhesions. One other case of bilateral ureteric obstruction secondary to OHSS describes a background of Crohn's disease.⁴ Other cases describe patients with abdominal compartment syndrome secondary to OHSS with previous bowel resections due to Crohn's disease,⁸ and with hydroureter in a transplanted kidney secondary to OHSS.² It has been postulated that adhesions may take up any potential space that the hyperstimulated ovaries might expand into;⁴ in addition, we would add that adhesions may limit the elasticity of abdominal structures, preventing them from mobilising out of the path of the enlarging ovaries. It seems probable that these factors combine to cause an increase in abdominal compartment pressure and, hence, the likelihood of compression of local structures, including the ureters.

The incidence of ureteric strictures in those with Crohn's disease is contested but is thought to be between 0.3% and 25%. They occur due to local reaction when bowel adjacent to the ureters undergoes inflammation, fistulation, local abscess formation and fibrosis.⁹ In our patient, the finding of a right-sided ureteric stricture is almost certainly related to her Crohn's disease and previous surgeries. As her ovaries enlarged, this seems likely to have acted as a risk factor for development of ureteric obstruction, with resultant sepsis.

Learning points

- ▶ Ovarian hyperstimulation syndrome (OHSS) is a potentially life-threatening complication of in vitro fertilisation treatment.
- ▶ Patients with pre-existing adhesional disease (secondary to either inflammatory conditions such as IBD or previous surgery) may be predisposed to ureteric obstruction should OHSS develop.
- ▶ Patients with Crohn's disease can develop ureteric strictures, which may also predispose to ureteric obstruction and sepsis should OHSS develop.
- ▶ In this patient group, renal function should be monitored regularly, and there should be a low threshold for obtaining renal imaging.
- ▶ Pregnancy in these patients is likely to be higher risk; multidisciplinary care from fertility, obstetrics, urology and gastroenterology is needed.

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Competing interests None declared.

Patient consent Obtained.

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