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Spontaneous ureteric rupture secondary to an invasive desmoid tumour

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ABSTRACT

INTRODUCTION: Spontaneous ureteric rupture is a rare entity that presents as an extravasation of urine from the ureter without previous surgery, ureteric manipulation and external trauma of the ureter. We report the case of a desmoid tumour presenting as spontaneous ureteric rupture which was managed in our institution.

PRESENTATION OF CASE: A 28 years old healthy male presented with a four day history of generalised abdominal pain secondary to spontaneous right ureteric rupture. Patient was initially managed via insertion of nephrostomy tube and antibiotics. After unsuccessful attempts of retrograde and antegrade ureteric stent insertion, patient was subsequently managed via elective surgical intervention. The excised specimen revealed desmoid tumour as cause of the ureteric rupture.

DISCUSSION: Desmoid tumours are rare benign tumours arising from fascial or musculoaponeurotic structures that do not metastasise, but tend to invade locally. It is often initially managed medically prior to undertaking a definitive surgical intervention. To our knowledge this is the first reported case of ureteric perforation secondary to a desmoid tumour of the mesentery.

CONCLUSION: Spontaneous rupture of the ureter is often misdiagnosed as other conditions. History taking and examination can be unreliable, hence a high level of suspicion and further investigations should be utilised. Once the diagnosis is made, treatment can be individualised based on aetiology.

1. Introduction

Spontaneous ureteric rupture is a rare entity that presents as an extravasation of urine from the ureter without previous surgery, ureteric manipulation and external trauma of the ureter.[1] Historically, there has been a small number of cases reported in the literature usually associated with urolithiasis.[2] Other causes include pelvic cancer, retroperitoneal fibrosis, fluid overload, urinary retention and pregnancy.[3–9] We report the case of a desmoid tumour presenting as spontaneous ureteric rupture which was managed in our institution. To our knowledge this is the first reported case of ureteric perforation secondary to a desmoid tumour of the mesentery.

2. Presentation of case

A 28 years old healthy male with no previous medical history presented with a four day history of generalised abdominal pain. He had no urinary or systemic symptoms. In particular he was afebrile and haemodynamically stable. On examination, there was non-specific generalised abdominal tenderness. Auxiliary blood tests revealed normal renal function and mildly raised liver function tests. Urinalysis, abdominal and chest X-rays were unremarkable. He was discharged from the emergency department with the view to have further outpatient investigation.

The abdominal ultrasound performed 2 weeks later revealed a right sided hydronephrosis. A CT with delayed urogram views confirmed the hydronephrosis and dilated ureter to the level of iliac vessels, where a 5 cm urinoma was apparent (Fig. 1). There was right ureteric disruption at the level of the pelvic brim without associated calculus or mass. Free pelvic fluid with extravasation of contrast was also noted. Patient was referred to urological surgical team who managed patient with intravenous antibiotics and percutaneous nephrostomy tube. The following day patient underwent retrograde study which confirmed the extravasation of contrast at the level of pelvic brim. Retrograde attempts to insert a ureteric stent were unsuccessful (Fig. 2).

Antegrade urogram via percutaneous nephrostomy two weeks later again confirmed contrast extravasation at the same location and two separate attempts of antegrade stenting were both unsuccessful.

After discussion of current status and risks, the patient underwent elective laparotomy with view of ureteric reimplantation.

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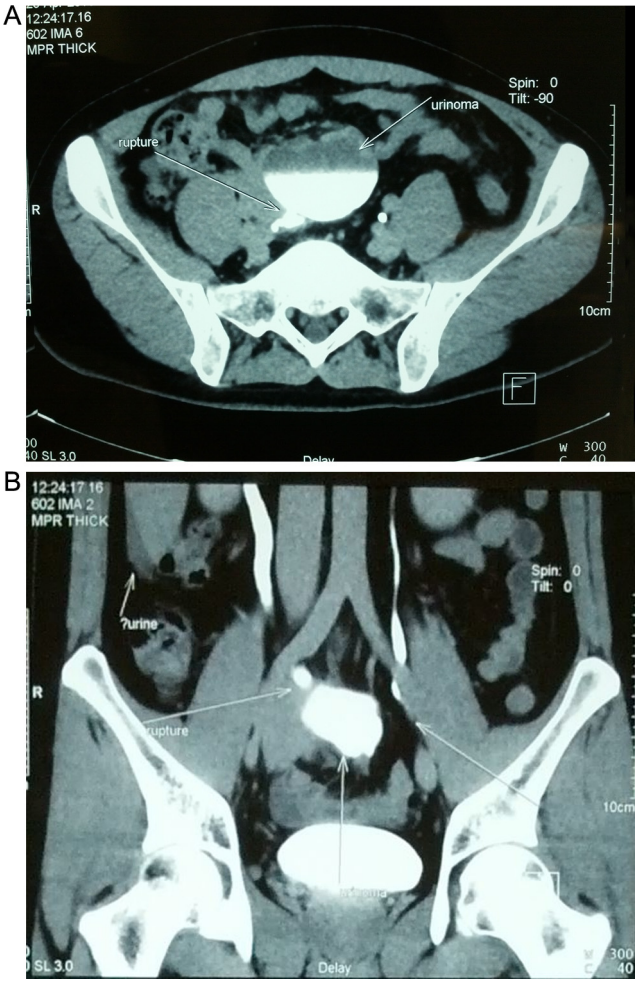

Fig. 1. A CT with delayed urogram views – ureteric rupture at the level of iliac vessels and a 5 cm urinoma associated with contrast extravasation (A: axial views, B: coronal view).

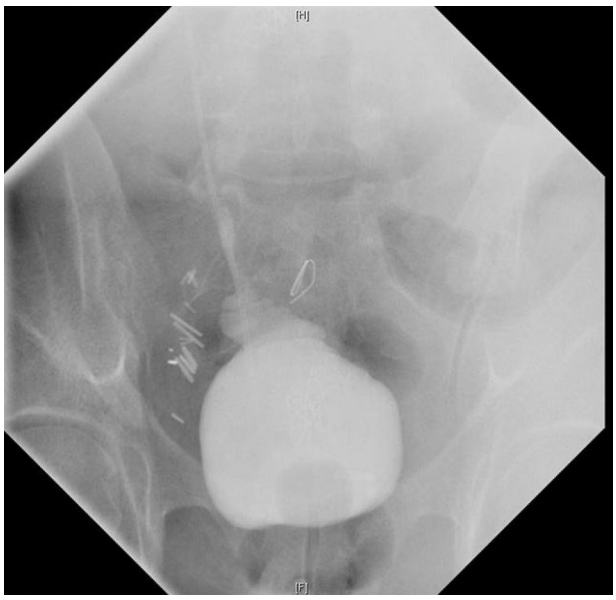

Fig. 3. Postoperative retrograde micturating cystourethrogram.

Intraoperative findings revealed a mesenteric mass involving the ureter, terminal ileum and caecum. The intra-operative frozen section showed dense fibrous tissue and chronic inflammation. Due to poor quality of tissue and suspicious nature of the mesenteric lesion, an ileocolic resection was performed with functional end to end enterocolic anastomosis with the assistance of general surgical colleagues. Ureter was divided above the fibrous lesion and implanted to bladder via a Boari flap and psoas hitch. Patient had an uneventful post-operative recovery. Post-operative cystogram showed no leak and the ureteric stent was removed at six weeks post operatively (Fig. 3). Follow up MR abdomen showed no further mesenteric mass lesions intra-abdominally.

The final histopathology confirmed the diagnosis of mesenteric desmoid-type fibromatosis. The macroscopic and microscopic examination revealed a 55 mm × 45 mm × 40 mm irregular mass centred in the mesentery which invaded and destroyed the muscle layer in the small bowel wall and extended into the submucosa (Fig. 4). The ureter was entrapped and completely encircled by the mass which appeared to focally penetrate and destroy the ureteric muscle. Immunohistochemical staining results of the mass is shown in Table 1.

Patient was well at 6 months post-surgery on follow up. Outpatient colonoscopy and geneticist referral showed no abnormalities. The patient was found to not have any familial disorders such as Familial Adenomatous Polyposis (FAP) or Gardner's syndrome.

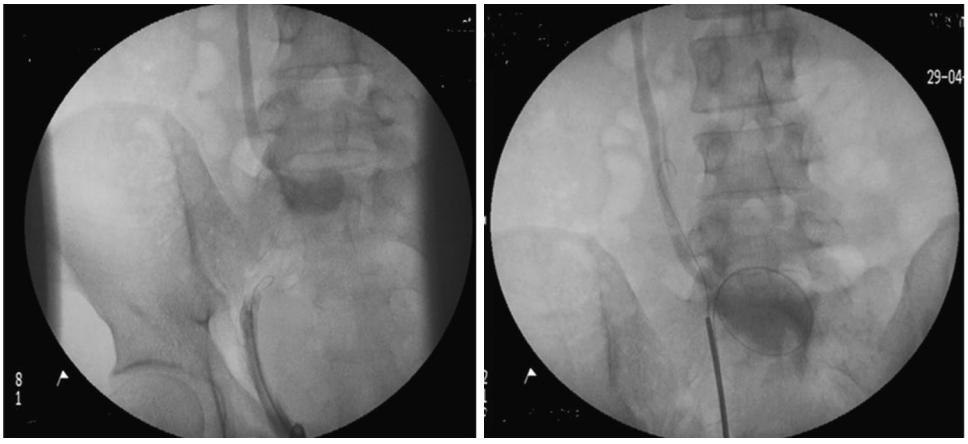

Fig. 2. Intraoperative retrograde urogram – ureteric disruption and extravasation of the contrast.