Cystitis Cystica Causing Bilateral Ureteric Obstruction
J Nariculam, A Patel, D Murphy, A Rane

Abstract
Cystitis cystica is a rare benign proliferative lesion of the bladder, caused by hyperplasia of the bladder submucosa, usually as a response to chronic irritation. It is considered to be premalignant, leading to adenocarcinoma of the bladder. Though cystitis cystica occurs mainly at the bladder neck and trigone region of the bladder, we report a case of cystitis cystica causing bilateral ureteric obstruction and loin pain.

CASE REPORT
A 46 year old gentleman presented to the urology clinic with abdominal and loin pain. An ultrasound scan revealed a left-sided hydroureronephrosis. Following this, an IVU was performed, which demonstrated an irregular filling defect at the left VUJ.

A rigid cystoscopy showed an inflamed bladder neck and trigone. Multiple bullae were seen and neither ureteric orifices were visible.

Multiple biopsies were taken of the abnormal mucosa and bullae. Histopathology of these specimens confirmed cystitis cystica et glandularis with (globlet cell) metaplasia.

The gentleman continued to have left sided loin pain. A repeat IVU continued to show marked hydronephrosis with clubbing of the calyces on the left side with hydrourerette. The IVU also demonstrated right hydronephrosis and hydrourerette without calyceal clubbing. The patient however was asymptomatic on this side.

A DTPA scan was also performed to rule out any non-functioning kidneys. This showed divided function of 48% for the right kidney and 52% for the left kidney with the left kidney showing early PUJ obstruction.

With continuing pain in the left loin, a percutaneous nephrostomy followed a week later by antegrade stent was placed in the left ureter as retrograde stenting was deemed difficult due to the inability to locate the ureteric orifices.

A week following antegrade stent insertion, the patient was readmitted to hospital with fever, rigors and left loin pain. He was given broad spectrum antibiotics and e-coli were cultured from the urine specimen. We assumed that this episode of gram negative sepsis was solely due to stent insertion of the left ureter. He settled with antibiotics and was discharged home. A routine check rigid cystoscopy and biopsy three months following stent insertion showed an unchanged bladder with cystitis cystica still present. His urine at this point was sterile. It was decided at the multidisciplinary team meeting to perform a repeat rigid cystoscopy in a further six months. Over the next six months, the patient didn't complain of any stent related pain. He also didn't develop any further urinary tract infections.

The rigid cystoscopy performed six months later demonstrated a much improved bladder. No cystitis cystica nodules were seen, the ureteric orifices were visible and patent. No further bladder biopsies were taken. The JJ stent in the left ureter was removed. The patient was discharged home and placed on bladder surveillance in the form of flexible cystoscopy.

DISCUSSION
Cystitis cystica is a rare benign hyperplastic condition of the urinary bladder. It is associated with chronic urothelial irritation. A chronic irritative factor is said to be involved and this condition reflects mobilisation of the humoral immune defence response by various agents. It usually occurs in the elderly or occasionally in children.

In its minor form, it has the same clinical features as cystitis, but its major form may be mistaken for bladder tumour on endoscopy. On cystoscopy, cystitis cystica nodules are usually found located on the trigone and bladder neck.
region. Cystoscopic biopsy is mandatory, as the diagnosis is histological. It appears histologically as submucosal nests of columnar epithelial cells surrounding a central liquefied region of columnar degeneration. Cystitis cystica glandularis is essentially cystitis cystica that has undergone metaplasia to form glandular tissue. It differs from cystitis cystica only in the nature of the epithelia.

Cystitis cystica can be considered a premalignant disease of the urinary bladder. It is perceived that these glands undergo dysplasia and can progress to adenocarcinoma of the bladder. Previous published case reports have confirmed this pathological sequence of events.

In the absence of obvious lesion, treatment is usually medical, based on the eradication of the irritative factors. Surgery is required in case of complications of the disease like recurrent haematuria or features associated with bladder outlet obstruction secondary to the pseudoneoplastic growth.

We have reported a case of cystitis cystica et glandularis causing bilateral ureteric obstruction and loin pain. To our knowledge, there are no cases of cystitis cystica causing bilateral ureteric obstruction and loin pain in adults in the literature. There are however many reported cases describing unilateral ureteritis cystica causing unilateral ureteric obstruction.

The patient described is now free of cystitis cystica and the obstruction resolved. However, as cystitis cystica can predispose to bladder malignancy and that its clinical course is unclear, this patient will require long-term surveillance in the form of cystoscopic examination.

References

Author Information

Joseph Nariculam, MRCS
East Surrey Hospital

Amit Patel, MRCS
East Surrey Hospital

Declan Murphy, FRCS
East Surrey Hospital

Abhay Rane, FRCS Urol
East Surrey Hospital