

Xanthogranulomatous cystitis with malakoplakia leading to recurrent spontaneous bladder perforation in a young girl

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ABSTRACT

Recurrent bladder perforation due to xanthogranulomatous cystitis with malakoplakia is rare entity and can lead to spontaneous bladder perforation. A 15 years girl presented with sudden pain abdomen with reduced urine output. Her exploratory laparotomy revealed, perforation of 2 cm at the dome of bladder with unhealthy margins. Excisional bladder biopsy and repair of bladder perforation by 3-0 polyglactin suture was done. The histopathology showed xanthogranulomatous cystitis with malakoplakia. Her records revealed the same histopathology in bladder perforation at age of 9 with lost follow-up till age of 15. Exploratory laparotomy and bladder repair should be done to save the patient.

1. Introduction

The 15-year-old girl was referred from the paediatric department with complaints of recurrent abdominal distension, fever, constipation, and decreased urine output. She had a history of similar complaints 6 years back and was diagnosed with spontaneous bladder perforation due to xanthogranulomatous cystitis and malakoplakia. Open surgical repair was performed at that time.

2. Case presentation

USG and CECT whole abdomen revealed gross ascites without giving any clue about the diagnosis. On voiding cystourethrography, the contrast was seen extravasating into the peritoneal cavity (Fig. 1). A preoperative cystoscopy was performed, revealing single perforation with inflammatory changes in the surrounding mucosa at the dome of the bladder (Fig. 2). Open surgical repair of the bladder was performed under general anesthesia. Margins of the perforation site were freshened up by excising a rim of approximately 2 cm and the bladder was closed using absorbable polyglactin 3-0 sutures. The excised tissue was sent for histopathological examination.

The patient made an uneventful recovery and was discharged following catheter removal after 15 days. The patient is doing well at 12 months of follow-up. The patient is on regular follow-up with periodic urine cultures to rule out persistent bacterial cystitis. Histopathological examination revealed the presence of histiocytes with granular

eosinophilic cytoplasm and foamy macrophages in the lamina propria suggestive of xanthogranulomatous cystitis. Few histiocytes with intracytoplasmic basophilic inclusions were seen suggestive of malakoplakia (Fig. 3).

3. Discussion

Spontaneous bladder perforation is a rare event and recurrences are even rarer. It is usually due to underlying disease which weakens the bladder wall such as a tumor, diverticula, and cystitis.¹ Recurrent bladder perforation has been observed in the neurogenic bladder, diabetes mellitus, alcoholism, pregnancy, and bladder outlet obstruction but has been never been reported in cases of xanthogranulomatous cystitis.^{1,2} Spontaneous bladder perforation presents a diagnostic challenge and diagnosis is often made at exploration.¹ Sudden onset of abdominal distension, ileus, and oliguria should prompt the suspicion of bladder perforation.

Diagnosis is usually confirmed by CT cystography or retrograde cystography.³ In our case, retrograde cystography was performed which was suggestive of bladder perforation while cystoscopy easily confirmed the diagnosis. A retrospective review of the records of the same patient revealed that the previous perforation was also present at the dome of the bladder.

Extraperitoneal perforation of bladder is usually managed conservatively while intraperitoneal bladder perforation of bladder is managed by surgical repair as was done in our case.

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Fig. 1. Retrograde cystography showing extravasation of contrast into the peritoneal cavity.

Xanthogranulomatous cystitis is a chronic inflammatory disorder of the bladder. Etiology is still not clear but various etiological factors have been implemented in the causation of this disorder like a chronic bacterial infection, immunological disorder, abnormal lipid metabolism,

and lipid accumulation in the macrophages.⁴ Surgical resection is usually the curative treatment. Long-term antibiotic treatment may be given.⁵

4. Conclusion

Malakoplakia is a rare histiocytic disease that can occur in all organs but is most commonly found in the urinary bladder. It is rare in children and is more common in males. It is characterized by the presence of single or multiple white-yellow soft raised plaques on the mucosa of the bladder. It is caused by defects in phagocytic or degradative functions of histiocytes in response to gram-negative coliforms (*E. coli* or *Proteus*) that results in a chronic inflammatory state, followed by intracellular deposition of iron and calcium (known as Michaelis-Gutmann bodies).⁵ Clinical presentation and treatment are similar to xanthogranulomatous cystitis. Recurrent spontaneous bladder perforation associated with xanthogranulomatous cystitis has not been reported in the past and this is the first case of xanthogranulomatous cystitis leading to recurrent perforation of the bladder.

Ethics approval and consent to participate

Ethics approval not required and the Written Consent of participation was taken from the parents of patient for publishing a case report.

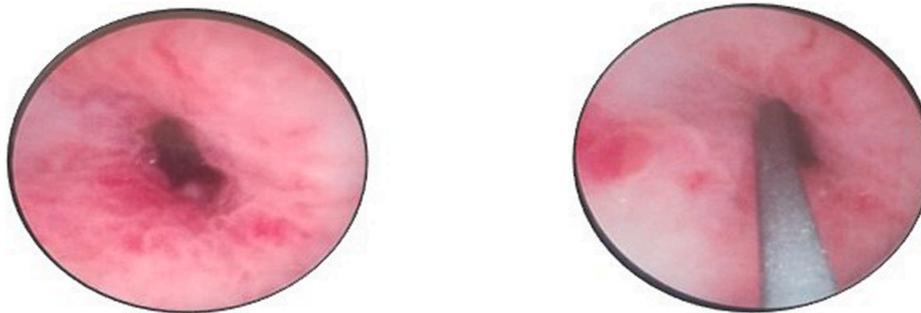


Fig. 2. Cystoscopic views of the perforation at the dome of the bladder.



Fig. 3. Histopathologic examination shows the accumulation of histiocytes with granular eosinophilic cytoplasm and foamy macrophages (A) and few histiocytes showing intracytoplasmic basophilic-inclusions.

Consent of publication

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Author's contribution

Author: Design of Work; the acquisition, analysis; interpretation of data.

Co-Author: Drafted the work or substantively revised it.

Declaration of competing interest

The authors declare that they have no competing interests.

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Not Applicable.

Abbreviations

CT computed tomography

USG- Ultrasound Sonography

CECT High-dose contrast-enhanced computed tomography

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