

# Confirmation of an internal hydatid bladder fistula using postoperative methylene blue retrograde injection via urinary catheter: a case report

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### **Abstract**

Abdominal hydatidosis resulting in an internal hydatid bladder fistula postoperatively is quite rare and might have serious consequences without timely treatment. A 74-year-old Tibetan woman presented with abdominal distension and was diagnosed with hydatid disease. Cyst contents were removed, and the pericyst was partially resected without contraindication. Furthermore, no internal urinary fistula was found before or during the operation, and the presence of an internal fistula was indicated by methylene blue retrograde injection via urinary catheter after the operation. The use of postoperative methylene blue retrograde injection via urinary catheter is recommended to identify internal hydatid bladder fistula formation.

### **Keywords**

Hydatidosis, postoperative methylene blue retrograde injection, internal hydatid bladder fistula, case report, abdominal distension, cyst

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# Introduction Corresponding author:

Hydatidosis occurs globally, and the tapeworm responsible for this condition parasitizes the liver, abdominal cavity and other organs.<sup>1</sup> It has been reported that Wei Gao, Diagnosis and Treatment Centre for Echinococcosis in Qinghai Province, Qinghai Provincial People's Hospital, No. 2 Gonghe Road, Xining, Qinghai 810000, China.

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hydatid disease affects the kidney, and hydatid components are detected in the urine.<sup>2</sup> However, there has not been a single case in which urinary fistulas were not found before or during the operation but developed after the operation. Based on this case, the use of postoperative methylene blue retrograde injection via urinary catheter is recommended to confirm the formation of an urinary fistula in patients with hydatidosis adjacent to the bladder.

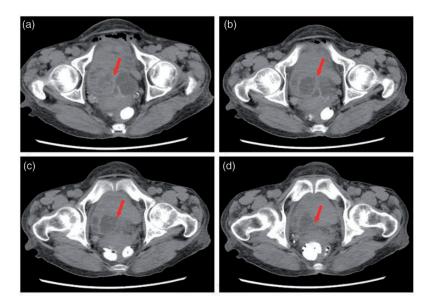
# **Case presentation**

The reporting of this study conforms to CARE guidelines.<sup>3</sup> A 74-year-old Tibetan female patient with a 4-year history of hydatidosis presented with abdominal distension for more than 7 months. Blood cell counts, coagulation factors and liver and renal function were within normal ranges. Urine showed a normal appearance and did not contain any cysts or blood. Abdominal and pelvic computed tomography examinations showed the presence of multiple cystic

hydatid lesions in the liver, abdomen and pelvic cavity (Figure 1). Enzyme-linked immunosorbent assay for hydatid detection showed that the sample was *Echinococcus granulosus*-positive.

The patient received an open surgery. Multiple hydatid lesions were observed in the pelvic cavity during the operation. The lesions were oval with a maximum diameter of approximately 85 mm and contained several daughter cysts. All daughter cysts were removed, and approximately 40% of the outer cyst wall was resected. Part of the pericyst of the cystic lesions penetrated the lateral wall of the bladder and was preserved to decrease the rate of fistula formation. No urine leakage from the residual hydatid cyst walls was observed by the surgeon when clamping the urinary catheter during the operation.

Four hours after the operation, no urine had been released from the urinary catheter, but 900 mL of pale-yellow liquid had drained from the pelvic drainage tube. To confirm the prediction of internal fistula



**Figure 1.** Pelvic computed tomography horizontal scanning in a 74-year-old woman with hydatid disease. The arrows indicate hydatid cysts (a–d).

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formation, we dissolved 20 mg of methylene blue (specifications: 2 mL, 20 mg; batch number: H32024827; Jumpcan Pharmaceutical Group Co., Ltd., Taixing City, China) in 500 mL of 0.9% normal saline, which was then injected retrogradely through the urinary catheter. Methylene blue diluent was detected in the pelvic drainage tube (Figure 2). Additionally, abdomen fluid was sent to a laboratory for confirmation of urine content. The presence of pelvic hydatid cysts that invaded the urinary bladder and induced the formation of an internal fistula in the bladder was considered.

### Discussion

Hydatidosis is prevalent in the Northern Hemisphere, particularly in pastoral areas of Central and Western China. The spleen, lung, brain, abdominal cavity and pericardium are potential sites affected by hydatid disease.<sup>4</sup> In the abdominal cavity, hydatid cysts exhibit exophytic growth, adhere to

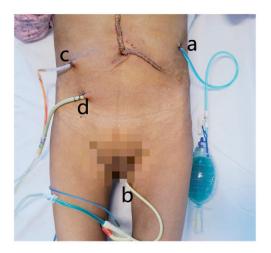


Figure 2. Postoperative methylene blue retrograde injection via urinary catheter. a) methylene blue diluent was detected in the pelvic drainage tube. b) methylene blue diluent was injected retrogradely through the urinary catheter. c and d) right pelvic drainage tube.

and compress the surrounding organs and tissues, including the intestine, bladder, kidney and ureter, and rarely show invasive erosion. This patient had multiple hydatid cysts in the liver and abdominal cavity. The urine volume and colour were normal before the operation, and no obvious abnormality was found during routine urine examination. Preoperative imaging examination could not determine whether a hydatid bladder fistula had formed.

According to the preoperative examinations and surgical findings, the characteristics of hydatid bladder fistula formation are described as follows: 1) Generally, preoperative computed tomography examination may not be used to confirm the formation of internal fistulas if urine examination was normal; 2) Before the operation, the daughter cysts blocked the orifice of the internal fistula, and no urine entered the pelvic cavity; and 3) Owing to the presence of the indwelling urinary catheter during the operation, the surgeon could not observe the flow of urine into the pelvic cavity from the orifice of the fistula.

Methylene blue retrograde injection is used to locate the fistula in complicated cases that are difficult to assess by cystoscopy and imaging.<sup>6,7</sup> Although cystoscopy might be meaningful for the confirmation of fistulas, it is not a therapeutic strategy. Therefore, we did not perform cystoscopy preoperatively. Cystography is another alternative to visualize the fistula and confirm the methylene blue findings. However, methylene blue retrograde injection via urinary catheter is more convenient for clinicians than cystography.

A difficulty of this case was that the internal fistula was blocked by a daughter cyst. We could not find the fistula before the operation or observe the internal floor opening during the operation because of the urinary catheter. After the operation, we confirmed the existence of the bladder fistula using methylene blue retrograde

injection via urinary catheter. In patients with a pelvic hydatid cyst, postoperative methylene blue retrograde injection via urinary catheter provides a straightforward approach to determine the formation of the urinary fistula.

## **Conclusions**

Internal fistula formation as a result of invasive pelvic hydatid cysts in the urinary bladder is not observable intraoperatively or through preoperative imaging. Postoperative methylene blue retrograde injection via urinary catheter is recommended for the detection of pelvic hydatid cysts adjacent to the bladder to confirm the formation of an internal fistula.

# **Ethics statement**

The study was approved by the Ethics Committee of Qinghai Provincial People's Hospital (No. 2021-161). The patient agreed to the use of her imaging results and clinical data for publication and academic research and provided written informed consent.

### **Declaration of conflicting interest**

The authors declare that there is no conflict of interest.

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