

Four cases of heterotopia of an intrauterine device embedded in the bladder muscular layer causing cystolithiasis: case report and review of the literature

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Abstract

Family planning is a basic national policy in China. An intrauterine device (IUD) is an important method of achieving family planning owing to its high safety, low cost, and convenient use. The indwelling birth control ring has no obvious adverse effects on the sexual life of women after the operation, and the process is reversible. This ring can be removed for women who want to have children again. There are approximately 100 million women with IUDs worldwide, with approximately 80 million in China. This finding accounts for approximately 40% of women of childbearing age in China. Although an IUD is safe, the invasive operation inevitably leads to complications, such as pelvic infection, abnormal menstruation, and damage to adjacent organs. Among them, ectopic bladder stone formation is a rare complication, but several related cases have been reported. We report four cases of heterotopia of an IUD and cystolithiasis, with diagnosis based on the medical history, clinical manifestations, imaging, and cystoscopic findings. The four patients with ectopic IUDs were treated with cystoscopy combined with laparoscopy (or hysteroscopy). We describe the process of diagnosis and treatment of our patients, and the related literature on an ectopic IUD is reviewed.

Keywords

Cystoscopy, laparoscopy, intrauterine device, cystolithiasis, bladder stone, holmium laser, hysteroscopy

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Introduction

An intrauterine device (IUD) is an effective, safe, economical, and reversible contraceptive device.¹ According to the World Health Organization's 1998 annual report,² the IUD is the main contraceptive measure for Chinese women, which is mostly implemented in rural areas. As a type of foreign body, an IUD often produces an inflammatory reaction. Because of a lack of relevant health knowledge in some women, they cannot be checked and diagnosed in time when corresponding symptoms occur. This leads to the occurrence of some urology-related diseases. At the same time, the operation level of doctors in some low-level hospitals is limited. When the contraceptive ring is placed, it often causes damage to the uterus. This is because the anatomical position of the uterus is adjacent to the bladder, and the bladder is close to the front wall of the uterus when it is filled and forced to contract. When the uterus is damaged, the contraceptive ring can travel to the bladder through the damaged part during contraction of the uterus, and then enter the bladder as a foreign body. Long-term inflammatory stimulation results in formation of bladder stones. Because some patients have symptoms, such as frequent urination, urgency of urination, pain of urination, and discomfort of the lower abdomen, they do not carry out a relevant examination and diagnosis and treatment in time. This situation leads to a delay in the best time for treatment. We report four cases of cystolithiasis caused by an ectopic IUD that ended up in the bladder in our hospital from 2015 to 2017, and discuss the reasonable diagnosis and treatment plan.

Case report

Case 1

A 41-year-old female farmer of Han ethnicity was admitted to the Pangang Group

General Hospital on 30 December 2015 owing to worsened urinary pain 6 months previously. She had a 7-year history of this pain. A urinary routine test showed urinary occult blood 3+, urinary erythrocyte 3+, urinary leukocyte 2+, and nitrite -. During the 7-year history, the patient had persistent dull pain, long and frequent micturition, and urinary urgency of approximately four to six times per day and once per night. These symptoms had greatly increased in the last 6 months and were complicated by stress urinary incontinence. In 1999, she underwent IUD placement in the Huili County People's Hospital. The IUD was an MYCu IUD, model S-32 (Liaoning Aimu Medical Technology Co., Ltd. Anshan City, Liaoning Province, China). The IUD was placed at postpartum and the age of insertion was 25 years. She had never received any operation involving the pelvic cavity, uterus, or bladder (including caesarean section), except for placement of the IUD. During hospitalization in our hospital, cystoscopy showed intravesical migration of the IUD and cystolithiasis, chronic cystitis, and bladder mucosal leukoplakia. Urogenital ultrasound (DW480; Dawei Medical Co., Ltd., Xuzhou City, Jiangsu, China) showed a high echo in the right wall of the bladder, which remained to be determined. A physical examination showed no obvious positive signs in the heart, lungs, abdomen, and urinary system.

The patient underwent holmium laser lithotripsy through a cystoscope under epidural anesthesia on 3 January 2016. After the bladder stone was broken, a pair of forceps was placed and the end of the IUD was clamped. The pair of forceps was then pulled outside from the bladder muscle layer. There was slight bleeding, clear vision, good filling of the bladder, and no obvious bladder tears. A #18 three-chamber catheter (Bard Sdn. Bhd., Kulim, Kedah, Malaysia) was inserted postoperatively and the bladder was continuously irrigated

with saline. The patient developed abdominal distension and pain along with mild fever (highest body temperature was 38.6°C). Postoperative pelvic computed tomography (CT) (64-slice spiral CT; General Electric Co., Boston, MA, USA) showed effusion of the pelvis and the peritoneum. After medical discussion, pelvic effusion occurred because of formation of urinary leakage in the bladder. Laparoscopic removal of a perivesical hematoma and repair of bladder rupture were performed under general anesthesia on 5 January 2016. The perivesical hematoma was approximately 3 × 3 cm during the operation. After removing the hematoma and the surrounding necrotic tissue, the bladder defect was approximately 1 cm at the hematoma site. The bladder defect was sutured with 2-0 absorbable suture, and the bladder fistula and pelvic drainage tube remained. The pelvic drainage tube was removed 1 week after the operation. Three weeks later, the bladder fistula was pulled out, and the patient was discharged on 27 January 2016 without a urinary fistula.

Case 2

A 39-year-old female laborer of Han ethnicity was admitted to the Pangang Group General Hospital on 21 February 2017 because of diuretic distension pain for 1 year. She had an MYCu IUD, model L-36 (Liaoning Aimu Medical Technology Co., Ltd.) The IUD was placed at postpartum and the age of insertion was 23 years. She had never received any operation involving the pelvic cavity, uterus, or bladder (including caesarean section), except for placement of the IUD. Abdominal CT (64-slice spiral CT; General Electric Co.) showed a metallic shadow in the bladder and the uterine lacuna. Cystoscopy (19.5 Fr; Wolf Co., Saarbrücken, Germany) showed bladder stones as foreign bodies and chronic cystitis. A urinary routine test showed urinary

occult blood 3+, urinary erythrocyte 3+, urinary leukocyte 3+, and nitrite +. A physical examination showed obvious tenderness of the suprapubic bladder area, the urethra was red and swollen, and there was no bleeding, suppuration, ulcers, or other changes.

Because of severe inflammation of the bladder, holmium laser lithotripsy was performed to treat the bladder stones after obtaining consent from the patient and her family members. The IUD rings were removed in two stages. The patient was discharged from hospital on 7 May 2016. Because of the patient's decision, the IUD rings were not taken in phase 2 in the Gynecology Department of our hospital after discharge. On 22 November 2016, cystoscopy was performed in the Gynecology Department of our hospital and recurrence of a stone (approximately 2 × 2 cm) was found. After urological consultation, holmium laser lithotripsy and laparoscopic ring removal were performed under general anesthesia on 25 November 2016. The IUD was removed from the bladder serosa by performing laparoscopy. The bladder was sutured with 3-0 absorbable suture. A bladder water injection test (Melan solution) was negative. After the operation, a #18 three-chamber catheter was retained. This catheter was pulled out and the patient was discharged without urinary fistula after 1 week.

Case 3

A 30-year-old woman of Han ethnicity with intermittent hematuria visible to the naked eye for longer than 1 month visited the gynecological clinic of the Pangang Group General Hospital on 19 February 2017. She had an MYCu IUD, model M-34 (Liaoning Aimu Medical Technology Co., Ltd.). The IUD was placed at postpartum and the age of insertion was 22 years. She had never received any operation involving the pelvic cavity, uterus, or bladder (including

caesarean section), except for placement of the IUD. Transvaginal ultrasonography (DW480; Dawei Medical Co., Ltd.) showed a strong light band in the cervix, dark cystic fluid in the cervix of up to 4×3 mm, and pelvic effusion. She was scheduled to have hysteroscopy for loop removal in the Obstetrics and Gynecology Department (Figure 1). On 20 February 2017, hysteroscopy (7 Fr; Wolf Co.) showed no contraceptive device in the uterus. Pelvic CT (64-slice spiral CT; General Electric Co.) showed an abnormal metal dense shadow between the anterior wall of the uterus and the bladder, an ectopic contraceptive ring, a small amount of pelvic effusion, and a cyst in the right adnexal area. Cystoscopy showed stones in the posterior wall of the bladder (approximately 2 cm) that were suspended from the bladder wall and visible IUD metal rings.

Holmium laser lithotripsy was performed under general anesthesia on 25

February 2017 after urological consultation. During the operation, approximately half of the IUD circular metal rings were located in the bladder at the bottom of the stones. Foreign body forceps and vigorous lithotripsy forceps failed to pull out the IUD. Because of the long duration of having the IUD and serious adhesion between the IUD and its surroundings, removal of the laparoscopic ring was performed after consenting with the family members. When gynecologists performed laparoscopy to separate pelvic adhesions, they found that the IUD could not be separated from the adventitia of the bladder, and extraction of the IUD was unsuccessful (Figure 2). This might have been due to the enlarged IUD at the end of the bladder, and therefore, it could not be removed. Urological surgeons used a holmium laser to break the middle part of the IUD under guidance of a cystoscope, and then used foreign body forceps to remove part of the

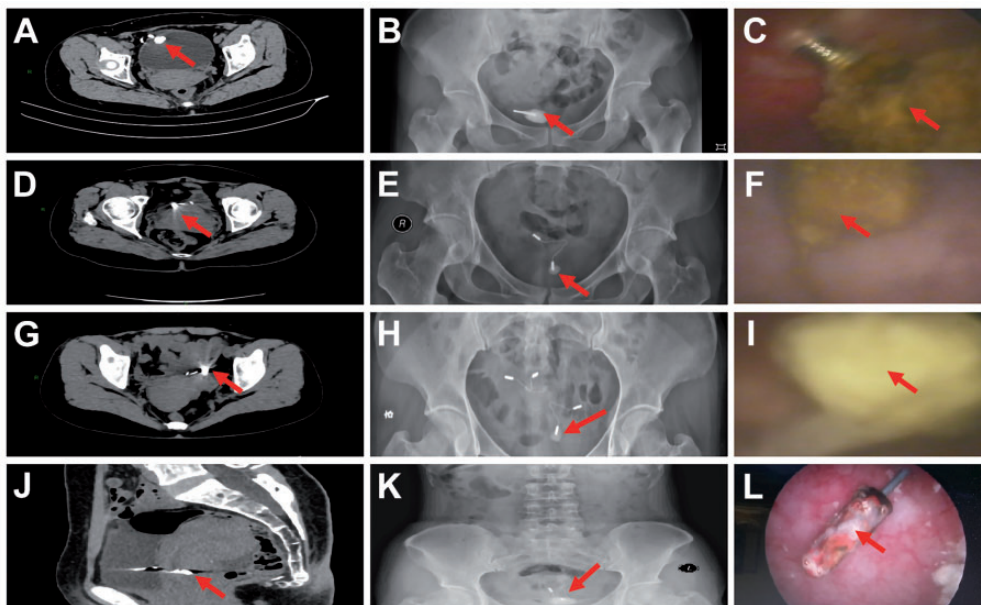


Figure 1. Transvaginal ultrasonography. The red arrow indicates penetration of the intrauterine device ectopically into the uterus.

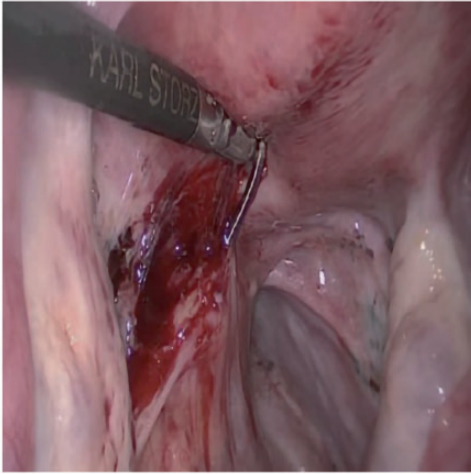


Figure 2. Photograph showing that the intrauterine device is embedded in the deep muscular layer of the bladder (laparoscopic view).

IUD in the bladder. Gynecologists also removed another larger part of the IUD in the abdominal cavity through laparoscopy. No major bleeding was found in the bladder and abdominal cavity after extraction. No leakage of the bladder was found during a bladder water injection (Melan solution) test after the operation. After 1 week of an indwelling catheter, the catheter was pulled out and the patient was discharged.

Case 4

A 47-year-old female teacher of Han ethnicity was admitted to Pangang General Hospital on 1 June 2017 because of dysuria for 3 months and hematuria for 2 weeks. She had an MYCu IUD, model M-34 (Liaoning Aimu Medical Technology Co., Ltd.). The IUD was placed at postpartum, and the age of insertion was 25 years. She had never received any operation involving the pelvic cavity, uterus, or bladder (including caesarean section), except for placement of the IUD. Abdominal ultrasound (DW480; Dawei Medical Co., Ltd.) showed bladder stones. Abdominal plain



Figure 3. Imaging data (computed tomography [A, D, G, and J], X-ray [B, E, H, and K], and cystoscopy [C, F, I, and L]) of the four cases. (A–C) case 1, (D–F) case 2, (G–I) case 3, and (J–L) case 4. The red arrows indicate stones from the ectopic IUD rings in the bladder.

film (BJI-2; Xianwei Photoelectric Technology Co., Ltd., Shanghai, China) showed IUD implantation and bladder stones. The patient refused to undergo cystoscopy for her own reasons. Hysteroscopy was performed under general anesthesia on 3 June 2017. Intraoperative extraction of the IUD ring failed. On 9 June 2017, the patient underwent holmium laser lithotripsy through a cystoscope under epidural anesthesia (Figure 3). During the operation, the stones were broken up by a holmium laser and the IUD ring in the bladder was broken in the middle. The ends of the IUD were removed from the urethra and the vagina. After the operation, a #18 three-chamber balloon catheter was retained and then removed after 1 week. No bladder leakage occurred, and the patient was discharged from the hospital on 16 June 2017.

Discussion

Indwelling IUDs are considered as a relatively safe, effective, simple, economical and reversible method of contraception.¹ For 40% of women³ of childbearing age, an IUD has become the contraceptive of choice. Common adverse reactions and complications associated with IUD implantation include bleeding, uterine perforation and ectopic pregnancy, infection, dysmenorrhea, abnormal menstrual volume, infectious abortion, and endometrial tumors.⁴ The risk associated with ectopic placement of an IUD in the bladder and stone formation is approximately 1% to 3%.^{5,6} However, in recent years, the number of cases of retention of IUDs and abnormal placement has been increasing, causing physical and mental pain to patients.^{7,8} The relevant departments and medical workers should understand and pay attention to this issue.

A retrospective analysis of 33 cases of ectopic IUD by Wu et al.⁹ showed partial insertion of an IUD into the muscular layer in 21 cases. Of these, one woman never underwent reexamination after 13 years of IUD placement, and only 2 patients had a partial ectopic bladder among the 12 cases of uterine perforation cases. One of these women developed bladder stones associated with an IUD after ectopic bladder formation. Goldbach et al.¹⁰ reported that the incidence of an ectopic M₁Cu II functional IUD was higher than that of other IUDs, such as the T-type, copper-type, O-type, and uterine cavity type. Additionally, the uterine cavity type of IUD had the lowest incidence. Sun¹¹ and other scholars believe that the M₁Cu II IUD is similar to a “V” type and has a sharp lateral wall. If the operation is not properly performed, abnormal torsion and insertion of the sharp part of the IUD easily occurs. Fan and Tang¹² investigated 98 patients with insertion after placement of an IUD. They found that

90.82% of patients who had failed to remove an IUD had the M₁Cu II functional IUD, 63 had placement during lactation, and 8 had placement performed immediately after induced abortion. IUDs of this type are partially impacted on one or both sides of the wall, and the degree of the impacted muscular layer is relatively shallow. Abnormal placement of the IUD into the bladder and cases of bladder stone formation are rare, and their mechanism(s) is still unclear. These problems may be related to the uterus itself, the proficiency of the operator, the indwelling time of the IUD, low estrogen levels, and other factors.^{8,13}

In the clinic, most patients with an IUD are treated for urinary tract infections or hematuria.⁴ All of our four patients had a history of urinary tract infection or intermittent hematuria visible by the naked eye before admission to our hospital. Previous studies have pointed out that female patients should be vigilant regarding the presence of foreign bodies in the urinary system.¹⁴ This is especially the case in patients with a history of IUD implantation when they suffer from repeated infections or symptoms of urinary tract irritation that are treated ineffectively. The diagnosis of an IUD embedded in the muscular wall of the bladder causing cystolithiasis depends on imaging examinations, such as B-mode ultrasonography, and kidney–ureter–bladder for preliminary screening. However, cystoscopy is required to make a definite diagnosis. Additionally, hysteroscopy and CT might also be necessary. The combination of these two techniques can preliminarily determine the location of an IUD and determine whether the other end of the IUD is located in the uterus or abdominal cavity.¹³

In the present study, the IUD was not found by hysteroscopy in the four patients. CT, cystoscopy, and intraoperative exploration showed that part of the IUD was located in the abdominal cavity and part of it

was in the bladder, which was in accordance with other reports.¹⁵ Three of these patients had the IUD for a long time. Studies in China and other countries have shown that the time of IUD implantation is positively correlated with ectopia.¹⁶ If an IUD remains in the body for too long, there might be serious adhesion between the IUD and the surrounding tissues. This leads to the possibility that the IUD cannot be removed by using foreign body forceps under cystoscopy. If the IUD can be easily removed from the bladder, it may lead to formation of hematoma around the bladder, as observed in case 1. This in turn causes infection or even formation of a localized abscess, resulting in poor healing of the bladder and the possibility of occurrence of urinary fistula. Therefore, patients with a long duration of an IUD require removal under laparoscopy combined with cystoscopy. Most of the patients in China use copper-containing IUDs, which enlarge at both ends after a long time of indwelling (similar to that of the MCu IUD, see Figure 4 for details). If only the stones in the bladder are broken, the enlarged end of the IUD that contains copper in the bladder cannot pass through the muscle layer of the bladder, leading to failure of removal. Therefore, use of a holmium laser is necessary to break the IUD from the middle and remove the two ends of the IUD separately. The integrity of the IUD is confirmed after removal. This method can minimize loss of bladder and avoid occurrence of urinary fistula.^{4,17,18}

Comparison of our four cases with the previous literature suggests that patients with an ectopic IUD resulting in bladder calculi should undergo preoperative cystoscopy + CT + hysteroscopy (or transvaginal ultrasonography) for determining the location of the IUD and facilitating formulation of surgical plans.¹⁹ If the majority of IUDs are located in the bladder, then cystoscopy can be used to remove the ring, and



Figure 4. MCu intrauterine device. The MCu intrauterine device is V-shaped, and its arms are sharper than those in the body after corrosion, and these arms can penetrate the uterine and bladder walls.

if the majority of IUDs are located in the abdominal cavity, then cystoscopy combined with laparoscopy assists in removing the ring. Using the surgical scheme of Cases 2 and 4, bladder trauma can be minimized by removing the IUD separately.^{20,21} Checking the IUD after the operation to determine its integrity is necessary because the IUD is often distorted during the process of retention. Therefore, the absolute integrity of the IUD cannot be guaranteed. Some residues of the IUD might remain in the bladder muscle layer after the operation.²² In this case, we can distort, stretch, and fold the IUD repeatedly *in vitro* after removal. If the IUD is not fractured, the risk of an IUD being present *in vivo* is low.¹

Conclusions

For patients with an ectopic contraceptive ring in the bladder muscular layer and bladder calculi, cystoscopy combined with laparoscopy (or hysteroscopy) remains helpful. A holmium laser can be used to interrupt the IUD during the operation, and the two parts of the IUD can be removed by cystoscopy from the urethra and by laparoscopy by puncture with a trocar. The curative

effect of this method is superior compared with other surgical methods.

Ethics statement

The patients provided signed informed consent for publication. The case report was approved by the Medical Ethics Committee of Pangang Group General Hospital (Approval number: 2019 (Examine) No. 2).


Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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