



Trauma and reconstruction

## A case report of ureteral endometriosis with severe hydronephrosis

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### ABSTRACT

Ureteral endometriosis accounts for about 5 % of urologic endometriosis, with a peak incidence in the age group of 30–35 years, usually with unilateral onset. Because of its insidious onset, often combined with asymptomatic loss of renal function, and the ineffectiveness of medications, surgery is the gold standard of treatment. Thus, here we report a case of ureteral endometriosis with severe hydronephrosis to raise awareness of this condition.

### 1. Introduction

Endometriosis (EMs) is a common disease among women of reproductive age. It is classified into ovarian, superficial, and deep infiltrating endometriosis (DIE). Ureteral endometriosis (UE) is a type of DIE and is relatively rare. The incidence of UE in women of reproductive age is approximately 1 %.<sup>1,2</sup> According to research studies, when endometriosis involves the urinary system, it most commonly affects the bladder (approximately 85 %), followed by the ureters (approximately 10 %). Involvement of the kidneys and urethra is extremely rare.<sup>3</sup> UE is typically unilateral, with 64 % of affected females showing a tendency towards the left side. This may be related to the presence of the sigmoid colon, which reduces fluid movement within the left pelvic cavity, thereby facilitating adhesion and growth of the endometrium during the menstrual cycle.<sup>1,4</sup> UE is more difficult to diagnose, with up to 50 % of patients being asymptomatic and often detected with impaired renal function due to more severe hydronephrosis.<sup>5</sup>

### 2. Case report

The patient is a 51-year-old woman who was admitted to the hospital after a three-week history of right-sided hydronephrosis detected during a medical examination. She does not experience any discomfort. Eight years ago, she underwent hysteroscopic surgery for endometrial polyps outside the hospital and has no history of endometriosis. She got married at twenty-two and had a vaginal delivery of a baby girl, whose menstrual cycles regularly. The examination suggests that both renal regions are not full, the right renal region has light percussion pain, and the right upper ureteral point has light pressure pain. Further investigations

including CTU, KUB and IVP revealed an upper middle stenosis about 1.5cm in the right ureter and severe hydronephrosis of the right kidney. ( Fig. ABC ) Cr:80μmol/L, eGFR:69.88mL/min. The patient underwent a retrograde right ureteral balloon dilation and stent placement procedure. During the procedure, ureteroscopy revealed luminal narrowing. The ureteral stent was removed after 2 months of treatment. However, 1 week later, the patient presented with discomfort in the right lumbar region and underwent a follow-up examination. It was found that the right kidney still had some degree of hydronephrosis, although it was slightly improved compared to before. Consequently, the patient underwent a repeat procedure of ureteral balloon dilation and stent placement for the ureteral stricture. The stent was removed after 2 months, but 1 week later, the symptoms reoccurred. Ultimately, under general anesthesia, the patient underwent ureterolysis of the mid-segment of the right ureter and Ureteral stenectomy + end-to-end anastomosis. The double J-tube was removed four months after surgery. Postoperative pathology: endometriosis of the right ureter. ( Fig. DEF ) During the six-month postoperative follow-up, the patient's hydronephrosis resolved, and she reported no pain or discomfort in the lumbar region.

### 3. Discussion

UE commonly involves the distal one-third of the ureter, specifically the upper 3–4 cm above the junction of the ureter and the bladder.<sup>6</sup> The pathogenesis of UE is currently not well understood. The implantation theory is the most widely accepted explanation, which proposes that endometrial fragments containing menstrual blood, enter the abdominal cavity through retrograde flow via the fallopian tubes or during uterine

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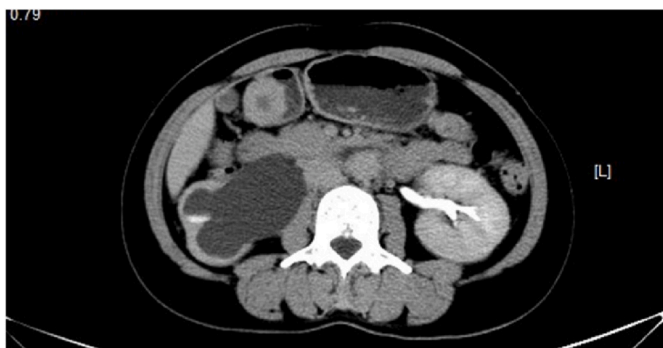
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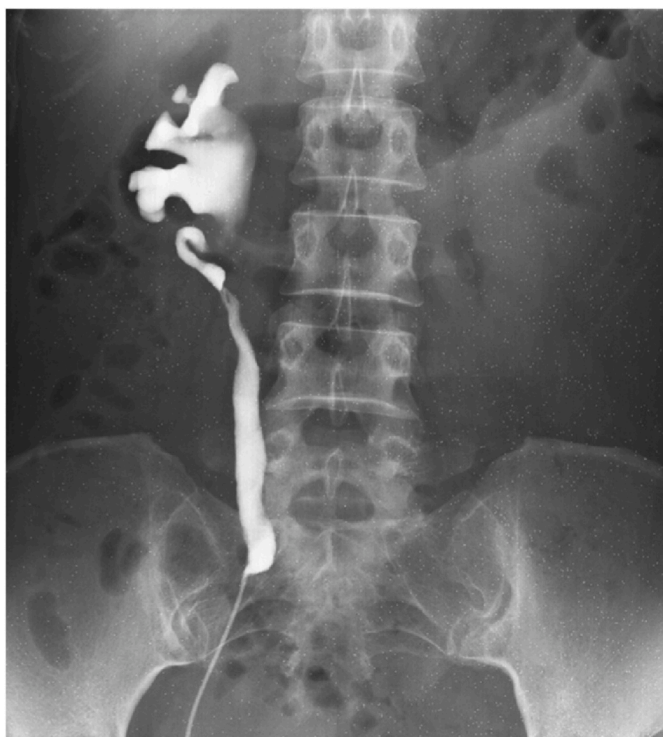
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A. (Heavy hydronephrosis of the right kidney with cortical thinning).



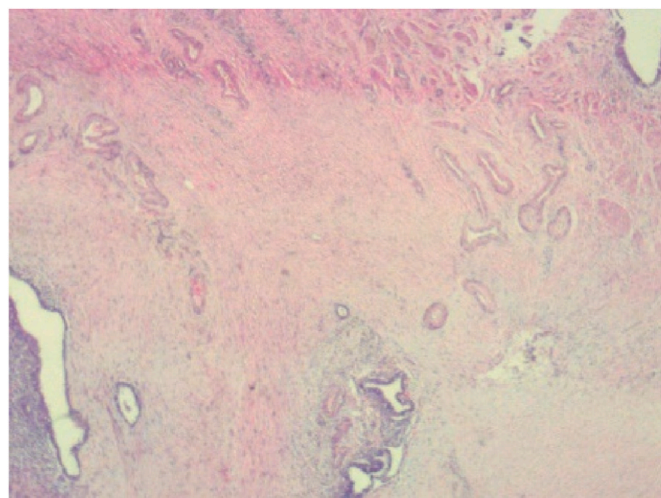
B. (Marked narrowing of the lower and middle ureter, with a little contrast passing through).

surgery. These fragments then implant in various locations such as the ovaries, uterosacral ligaments, intestinal wall, and bladder. It is believed that the distant organs and tissues such as the kidneys and ureters are primarily affected by the spread of endometrial tissue through blood vessels or lymphatic channels, leading to the development of the disease.<sup>7</sup>

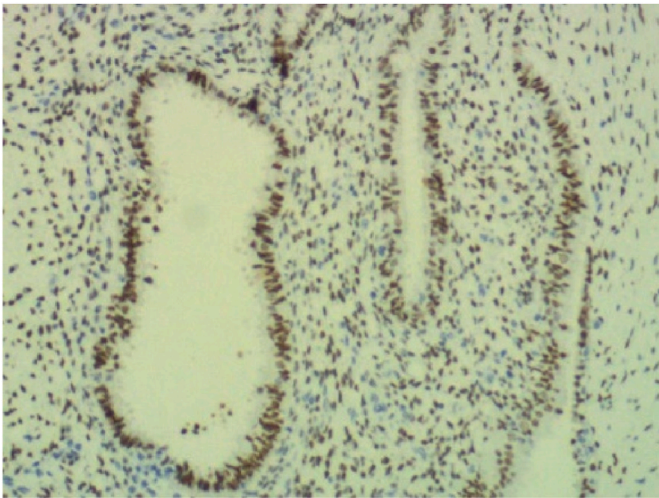
Currently, the gold standard for diagnosis is still through pathological tissue biopsy. As part of preoperative evaluation, transvaginal ultrasonography (TVS) is considered a first-line imaging modality for diagnosing DIE.<sup>8</sup> Furthermore, studies have shown that pelvic MRI and TVS have good diagnostic efficacy for UE. The results indicate that TVS and MRI have a sensitivity of 97 % and 87 %, respectively, and both have a specificity of 100 %.<sup>9</sup> This indicates that pelvic MRI provides additional options for the diagnosis of UE, especially for patients who have not had sexual intercourse or cannot tolerate the pain associated with TVS. It is also considered the best non-invasive method for assessing the location of endometriosis and distinguishing endometriosis from cancer.<sup>10</sup> Furthermore, since most UE patients have hydronephrosis, which can lead to occult renal failure, a combination of examinations such as



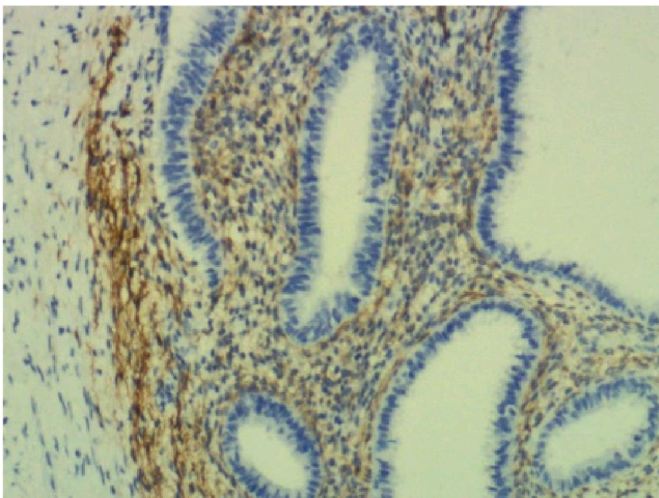
C. (3D reconstruction suggests that the right kidney is not visualized).



D. HE stain (20x magnification), ureteral mucosal uroepithelium in the upper right corner, endometriotic foci in the lower and lower left corner.



E. Endometrial glandular epithelial cells and some endometrial mesenchymal cells are ER positive, supporting endometrial origin.



F. Endometrial mesenchymal stromal cells positive for CD10, supporting endometrial origin

Key words deep infiltrating endometriosis (DIE) Ureteral endometriosis (UE) ureterocystostomy hydronephrosis.

KUB + IVP, CTU, contrast-enhanced CT, renal scans, and urinary system ultrasound are required for accurate determination of the location and nature of the condition.<sup>11</sup>

UE can be subdivided into two categories based on the location of the endothelial ectasia, with approximately 20 % presenting with intrinsic UE and 80 % presenting with ureteral extrinsic UE.<sup>12</sup> Extrinsic UE is caused by the invasion of endometrial tissue into the outer layer of the ureter or surrounding connective tissues. In contrast, intrinsic UE can be defined as the involvement of endometrial tissue in the intrinsic muscle layer, submucosal layer, or lumen of the ureter, which have slight differences of treatment. Combining oral contraceptive pills and progestogens is a first-line treatment for pain associated with UE. However, it cannot relieve strictures or obstructions; it can only alleviate pain. Additionally, surrounding tissues of the fibrotic lesions derived from UE typically do not respond to hormone therapy. Surgical intervention is particularly necessary when UE causes ureteral obstruction, especially to preserve the renal system.<sup>12,13</sup>

Currently, common surgical treatment options for UE include: ureteral endarterectomy, ureterolysis, segmental resection and end-to-end anastomosis, ureterocystostomy. Laser ureteral endometriosis incision

is performed to treat intrinsic UE. However, 60 % of cases experience disease recurrence. In later stages, balloon dilation and stent implantation may be required. Due to the high recurrence rate and ureteral strictures, ureterolysis has limited effectiveness in patients with intrinsic UE.<sup>14,15</sup> For extrinsic UE, ureterolysis is initially considered, especially for lesions smaller than 3cm. However, there is still a risk of recurrent adhesion-related strictures, which may require a second surgery. Ultimately, segmental resection of the ureter with end-to-end anastomosis or reimplantation may be necessary, with the surgical difficulty higher compared to the initial procedure.<sup>16</sup> A systematic review reported that out of 668 cases of UE, 579 cases (86.7 %) considered simple ureterolysis to be sufficient.<sup>17</sup> However, there are also reports indicating that the recurrence rate of hydronephrosis after ureterolysis can be as high as 12 %, while the recurrence rate of hydronephrosis after ureterovesical anastomosis is 3.1 %.<sup>18</sup> Another study reported that out of 27 patients who underwent ureterocystostomy for UE, 24 cases (80.0 %) had no severe hydronephrosis after the surgery. One patient experienced recurrence of hydronephrosis (3.3 %), but the degree of hydronephrosis significantly improved postoperatively ( $P < 0.001$ ). Although the number of cases is limited, ureterocystostomy remains a promising treatment option for UE.<sup>19</sup> The latest research suggests that indocyanine green can be used to observe the blood perfusion after ureterolysis in patients with UE, which can greatly enhance the safety of the surgery.<sup>20</sup> Considering the overall condition of this patient, it is deemed to be a case of intrinsic UE. The placement of a ureteral stent and dilation alone yielded poor results. Ultimately, segmental resection of the narrowed portion of the ureter with end-to-end anastomosis was performed, and the surgical outcome was favorable.

#### 4. Conclusion

UE is a relatively rare disease that presents diagnostic challenges, but it carries a high risk of potential kidney failure. It not only necessitates regular check-ups for women in the high-risk age group but also requires careful consideration of appropriate treatment options. For intrinsic UE, both laser ureteral endometriosis incision and ureterolysis yield poor results, often necessitating segmental resection of the narrowed portion. On the other hand, the treatment of extrinsic UE mainly relies on ureterolysis and segmental resection with anastomosis, resulting in a better prognosis. In cases where severe hydronephrosis has led to unilateral renal function loss, nephrectomy may be considered as a treatment option. The choice of surgical approach should be based on factors such as the degree of hydronephrosis, length and location of the narrowed segment, and patient preferences, which need to be comprehensively considered.

#### CRedit authorship contribution statement

**Ji Li:** Conceptualization, Data curation. **Jing Bai:** Formal analysis. **Haifeng Wang:** Writing – review & editing. **Bo Chen:** Conceptualization, Writing – original draft.

#### Declaration of competing interest

All authors have no conflict of interest.

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