CASE REPORTS

Ureteral endometriosis: A case report

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Abstract

A case of endometriosis of ureter in the Department of Gynaecology and Obstetrics of the Third Affiliated Hospital of Inner Mongolia Medical School was collected and analyzed on the basis of diagnosis, physical examination and treatment. The disease was rare due to its atypical clinical symptoms so that it was difficult to diagnose. The paper aims to expand our thinking and improve the gynecologists awareness and the level of diagnosis and treatment of rare diseases through the clinical analysis of the case.

Key Words: Ureter, Endometriosis

1 Medical record

1.1 General information

A 48-year-old female patient was admitted to our hospital at Department of Gynaecology on September 29, 2012 due to progressive menstrual abdominal pain. The patient's menstrual flow was regular and menarche reached at the age of 14, 7/30 days, mild amount and without dysmenorrhea. The last menstrual period was in September 21, 2012. The patient suffered from menstrual abdominal pain, persistent, no fever, vomiting, no malignant, no pain, frequent urination, urgency, dysuria, hematuria, and no discomfort of unknown origin during the three months. B ultrasound examination was performed several times and indicated left ovarian cyst. Past medical history included myomectomy surgery in an affiliated hospital in 2010.

1.2 Physical examination

T 36.8°C, P 70 per min. The patient presented with soft abdomen, tenderness in the left lower abdomen, without rebound pain or muscle tension, percussion pain in the double renal region, or tenderness in the double ureteral route. Gynecological examination results: multiparous type, vaginal smooth, no blood, cervical smooth, no cervical motion tenderness or cervical excitation, retroposition of uterus, normal size, mild quality, good mobility, and light tenderness. A mass of hard quality, unclear boundary, obvious tenderness, poor mobility, about 6 cm $\times$ 6 cm in size was found in the left adnexal region, while no abnormalities were discovered in the right adnexal region.

1.3 Auxiliary examination

Color Doppler ultrasound examination of pelvic: no abnormalities were found in double kidney, ureter, and bladder. The size of the left ovary was enlarged, and the mass of the cystic mass was about 5.0 cm $\times$ 4.1 cm $\times$ 3.6 cm in size. The boundary was clear, with no obvious envelope, and there were dense punctate echo and irregular non echo areas. The left fallopian tube widened about 1.1 cm, and left fallopian tube widening, suggesting left adnexal mass of mixed cystic mass and left ovarian chocolate cyst. All the blood routine, urine routine, four items of blood coag-

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ulation examination, and four items of infectious diseases examination were normal. Serum creatinine was examined to be 66.6 μmol/L, urea nitrogen 1.7 nmol/L.

1.4 Primary diagnosis

Pelvic mass of unknown origin:

(1) Left ovarian chocolate cyst?

(2) Pelvic inflammatory mass?

1.5 Treatment process

After the completion of relevant examination, resection of endometriosis and left ureter exclusion under epidural anesthesia were performed on September 29, 2012. During the operation, the uterus was normal in size, smooth surface, and the left broad ligament mass was about 6.0 cm $\times$ 6.0 cm $\times$ 5.0 cm, hard mass without complete capsule. Double adnexa were normal. The anterior and posterior lobes of the broad ligament were separated, and the whole package was quickly stripped. The rapid diagnosis of the case was uterine fibroids. During the examination, it was found that a 3 cm $\times$ 3 cm size of mass was found in the left lower part of the mass, and it was hard and tightly adhered to the surrounding tissue. The director Zhizhong Liu from the Department of Urology was invited for consultation regarding the diagnosis of the disease and endometriosis of ureter was taken into consideration. Separation of adhesion, dissociation of ureters, and excision of the lesion were performed step by step. There was no dilation and stenosis of the ureter. The ureter should be placed in the peritoneum to avoid postoperative adhesion and stricture. Postoperative pathological findings suggested endometriosis (see Figures 1 and 2). Cefoperazone sodium and metronidazole were used to prevent infection, and the body temperature was normal 3 days after operation. The patient was discharged 5 days after operation.

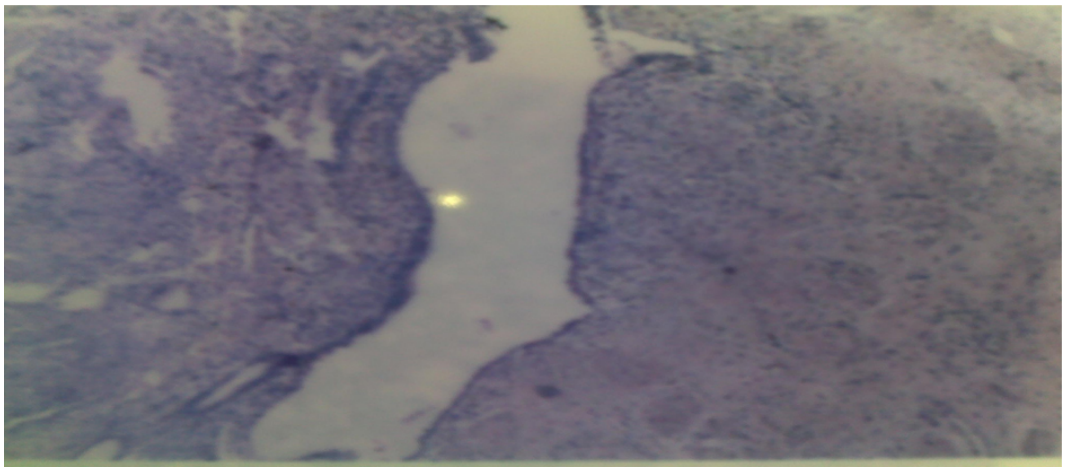

Figure 1: Postoperative pathology 1 of endometriosis

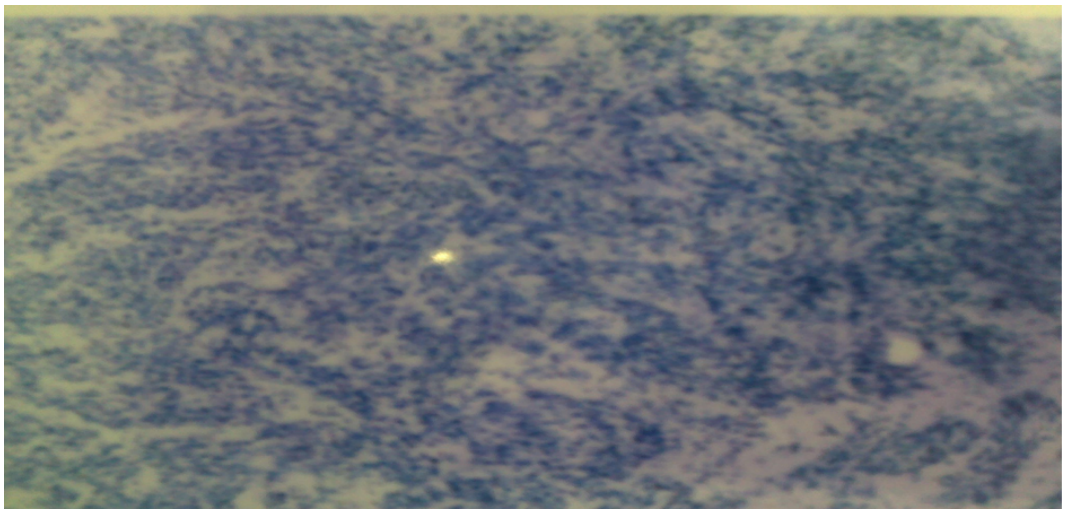

Figure 2: Postoperative pathology 2 of endometriosis

2 Discussion

2.1 Dr. Ping Liu

Dr. Ping Liu is the director of Department of Gynaecology and Obstetrics at the Third Affiliated Hospital of Inner Mongolia Medical University, specializing in gynecologic oncology and minimally invasive techniques.

According to the intraoperative findings, the patient suffered from a kind of deep infiltrating endometriosis. Deep infiltrating endometriosis is a special type of endometriosis with a depth of invasion of more than 5 mm below the peritoneum in this case.[1] The lesions are active and are closely related to pelvic pain,[2] which may invade sacral ligament, vaginal rectum, vagina, rectum, bladder and ureter.

There is no definite data on the incidence of deep infiltrative endometriosis, approximately accounting for 20% of endometriosis. As a special type of endometriosis, it can be distributed in other parts of the pelvic cavity and pelvic region. Chapron et al.[3] reported deep infiltrating endometriosis lesions located in uterosacral ligament, intestines, vagina, bladder, ureter and omentum, accounted for 52.7%, 22.7%, 16.2%, 6.3%, 2.1% and 1.7%, followed by the distribution of bowel involvement lesions in the rectum, rectum and sigmoid colon junction, sigmoid colon, vermiform appendix, small intestine, cecum and ileocecal, accounted for 65.7%, 17.4%, 6.4%, 4.7%, 4.1%. The typical clinical symptoms of deep infiltrating endometriosis include dysmenorrhea, dyspareunia, chronic pelvic pain, defecation pain and menstrual hematochezia defecation. The tenderness nodules of the posterior vaginal fornix and the posterior part of the uterus can be found by bilanual gynecological examination and vagino-recto-abdominal examination, and the histopathological diagnosis makes the confirmation. The patient was manifested with progressive abdominal pain only without typical clinical symptoms of deep infiltrative endometriosis, and was admitted to the emergency department with lower abdominal pain. The patient required further treatment. Surgical treatment is the first choice for the patient, and surgery is the primary treatment for deep infiltrative endometriosis. The selection of treatment method should be based on patient's age, fertility status, severity of symptoms, extent and extent of involvement. Complete resection of the lesion is important for relieving pain, preventing recurrence and malignant transformation. There are 3 types of surgical procedures. (1) Conservative surgery: separation of adhesions, resection of ectopic lesions, retaining the uterus and ovary, which is suitable for patients with limited lesion range, mild degree, younger or fertility requirement. (2) Semi radical surgery: removal of the uterus, ectopic lesions, at least to retain part of the ovary, which is suitable for childbearing, above 35 years of age, intractable pelvic pain or accompanied by uterine lesions. (3) Radical surgery: hysterectomy and bilateral salpingo, ectopic lesion, was applicable to extensive ectopic lesions and severe hydronephrosis, or hydronephrosis caused by ureteral involvement, or the intestine caused by melena and recurrence. At present, many doctors trend towards laparoscopic surgery, which has the advantages of easy identification of ectopic lesions, less trauma, lighter postoperative pelvic adhesions and faster recovery. However, open surgery should be taken when the condition of extensive distribution of lesions, severe adhesion or involvement of ureter and intestinal tract is met. According to the intraoperative exploration, the diagnosis of ureteral endometriosis was made. Endometriosis of the cervix and ureter is relatively rare, and ureteral endometriosis can eventually lead to hydronephrosis and occult renal failure.[4,5] Endometriosis involving the ureter is often unilateral, involving the distal ureter adjacent to the uterine artery. It is divided into intraluminal type and exogastric type according to the lesion site. Exogastric type accounts for 80% of all the cases, adjacent to endometriosis, involving ureter, ureteral compression, causing fibrosis of the tube wall and surrounding tissue, leading to ureteral stenosis, obstruction, hydronephrosis, impaired renal function. The intraluminal type accounts for 20%, and the lesions are located in the mucosa and lumen of the ureter, which may cause obvious obstruction and periodic hematuria. The two types can exist alone or coexist. The patient in our case belongs to the first type. The pelvic wall endometriosis causes the left ureter wall and the surrounding tissue fibrosis. In the resection of endometriosis lesions, the adhesion lysis of ureter is performed, which effectively avoids the ureteral dilatation and hydronephrosis. Segmental ureteral resection is required for the treatment of ureteral endometriosis, and the ureter end to end anastomosis or ureter reimplantation is performed according to the location and extent of ureteral lesions.[6,7]

To sum up, cases of deep infiltrating endometriosis with ureteral involvement are rare. It is so-called "rare" due to its atypical clinical manifestations. The patient would mistakenly see the doctor in the department of urology due to hydronephrosis especially when the condition is serious, so that early visit to the gynecology department is seldom. The peculiarity of this case lies in the fact that ureteral lesions were found during the operation so that the serial process of removing the lesion, ureter exclusion, separation of adhesions around were performed, effectively prevent patients with possible future ureteral stenosis, renal dysfunction caused by discomfort. At the same time, it retains the reproductive function of patients and improves the quality of life.

2.2 Dr. Zhizhong Liu

Dr. Zhizhong Liu is the director of Department of Urology Surgery at the Third Affiliated Hospital of Inner Mongolia Medical University, specializing in renal transplantation, laparoscopy and minimally invasive techniques.

Endometriosis is more common in premenopausal women

aged 15-44 years, with a statistical incidence of 10%-20%.[8] Ectopic endometrial or stromal lesions are common in the pelvic cavity, such as the ovary, broad ligament, pelvic tissue, Douglas pouch, and fundus ligament. Endometriosis in the urinary system is rare, and only 1.2%-3.9% patients have clinical symptoms. Therefore, there is a lack of large clinical case reports.[9] In recent years, more than 10 cases have been received in our department, and all of them were accompanied by different degrees of lower ureteral stenosis, upper segment expansion and hydronephrosis. The patient was only manifested as progressive abdominal pain, without no obvious dysmenorrhea, dyspareunia, or no urinary symptoms, no pain, no frequent urination, urgency, dysuria, hematuria and discomfort, so it is difficult to make the diagnosis before surgery. For the diagnosis of endometriosis in the urinary system, imaging examination plays an important role in determining the clinical diagnosis and preoperative evaluation. Therefore, regular routine ultrasound examination of urinary system should be performed in endometriosis patients, especially during menstruation with back pain, pelvic mass, and urinary tract involvement for early detection as well as early treatment of hydronephrosis. However, it is difficult to diagnose endometriosis simply by B-mode ultrasonography because of the lack of specificity in B-mode ultrasonography of endometriosis.[10] For patients with hydronephrosis diagnosed by B ultrasound, it is feasible to perform intravenous urography. Typical cases are manifested as lower ureteral stenosis and tortuosity, stenosis above the ureteral dilatation and hydronephrosis. When intravenous urography (IVU) showed no image, retrograde urography (RUG) or MRI examination should be performed to further clarify the location of the obstruction and the extent of hydronephrosis. It is generally believed that MRI is the best diagnostic method for the diagnosis of endometriosis.[11]

Endoscopic technique plays an increasingly important role in the diagnosis and treatment of urinary endometriosis. Cystoscopy and ureteroscopy can not only be used directly to observe endometriosis lesions, but also helpful to biopsy and pathological diagnosis. To date, laparoscopy is an effective method for the diagnosis and treatment of endometriosis.[12]

2.3 Dr. Xiaoli Kang

Dr. Xiaoli Kang is the associate director of the Third Affiliated Hospital of Inner Mongolia Medical University, specializing in gynecological endocrinology.

According to the intraoperative findings and postoperative pathology, the patient was diagnosed with endometriosis of the urinary system. For the treatment of endometriosis in the urinary system, drug therapy is considered to relieve symptoms only. Surgery is an effective and long-term method to ensure a good prognosis.[13] The surgery was divided into conservative surgery and radical surgery. Conservative surgery refers to resection of the lesion and invasion of the organs and tissues, radical resection on the basis of conservative treatment of the uterus and double accessories. As the disease occurs in reproductive age patients, some patients have fertility requirements, and it is theoretically possible to relapse after radical mastectomy. Therefore, radical resection is limited to the patients with pelvic lesions involving the ovary and the sacral ligament. No matter what kind of operation is performed, the resection of the lesion should be thorough, and the resection range is equal to the scope of tumor resection, to prevent residual lesions recurrence. The treatment should be individualized according to patient age, fertility requirements, the severity of renal damage and range, and corresponding treatment plan could be adopted. The diagnosis of renal function depends on early diagnosis and prompt treatment. The curative effect and prognosis of the disease are good, and the recurrence rate of the disease is different in literatures, ranging from 8% to 62%.[14]

Scioscia et al.[15] reported 199 cases of ureteral endometriosis in 6 groups with 12-41 months follow-up visits, and the average recurrence rate was 8%. The cause of recurrence may be related to incomplete resection of lesions. Endometriosis is a benign lesion in histology, but its biological behavior tends to be malignant, such as local infiltration or distant metastasis, invasion and destruction of surrounding tissues and organs, which has malignant potential.[16] The malignant transformation rate was 0.7%-10.0%,[14] mainly in the ovary. There were also reports of malignant transformation of the bladder endometriosis.[13] The case of ureteral endometriosis may lead to lower ureteral stenosis and hydronephrosis if left untreated. Therefore, timely and thorough treatment should be carried out once the diagnosis of urinary endometriosis is made.

2.4 Dr. Hairong Guo

Dr. Hairong Guo is the associate director of the Third Affiliated Hospital of Inner Mongolia Medical University, specializing in gynecological endoscopy.

Endometriosis is a common gynecological disease in women of childbearing age. The lesions are mostly confined to the ovary, pelvic peritoneum, rectovaginal septum, and sacral ligament. Due to less typical symptoms, endometriosis is often misdiagnosed or missed diagnosed. At present, the relevant literature is limited to cases or medical records series reports. For the treatment of ureteral endometriosis, there is still lack of evidence-based medicine to evaluate the effective treatment. The specific treatment of ureteral endometriosis depends on the patients' age, fertility requirements, the extent of the lesion, the degree of ureteral involvement and renal function. The principles of treatment are to remove the lesion, relieve the obstruction, protect the renal function, prevent the recurrence, and reduce the re-

lated complications as much as possible.[17]

The patient still needs drug treatment after surgery. Drugs for the treatment of ureteral endometriosis include progesterone, nemestran, danazol, gonadotropin-releasing hormone agonist (GnRHa), 3 months for 1 cycle.[18] Danocrine and GnRHa were the most frequently used according to literature, the dosage was 200-800 mg/d for Danocrine,[19] subcutaneous injection of GnRHa was taken every 28 days. There was no significant difference between the two treatments.[20] It is generally believed that the effect of simple drug treatment for endometriosis is limited. It is even necessary to monitor closely the progress of ureteral obstruction during the course of drug treatment for patients with mild ureteral involvement or with no impairment of renal function. Once the drug is stopped, there is still a recurrence of the disease, such as ureteral obstruction and impaired renal function, which may need further surgery treatment. In addition, ureteral endometriosis often has fibrosis and carries poor response to drug therapy. Therefore, the current drugs are mainly used for preoperative reduction of tissue edema, narrowing lesions, providing favorable condition for surgical resection,[21] or for postoperative adjuvant therapy to reduce recurrence.[22]

According to the condition of the patient, further drug treatment is needed after surgery, which can effectively reduce the recurrence, and further prevent hydronephrosis caused by ureteral stricture.

2.5 Dr. Nan Sun

Dr. Nan Sun is residents of Gynecology and Obstetrics at the Third Affiliated Hospital of Inner Mongolia Medical University, specializing in gynecological endocrinology.

The patient was diagnosed as ureteral endometriosis. Despite endometriosis in the urinary system is rare, however, it often leads to loss of renal function or is misdiagnosed as ureteral tumor, resulting in renal ureter resection. Approximately 1% of the patients may have endometrial infiltration with pelvic endometriosis at the same time. The incidence of endometriosis in the urinary tract involving bladder is about 84%, ureter 10%, kidney 4%, urethra 2%.[23] According to different studies, ureteral endometriosis incidence rate is 0.01%-1%,[24] although the incidence is very low, but it is not the main cause of the symptoms of kidney function loss.[25]

Pathologically, ureteral endometriosis is often divided into two types: intraluminal type (25%) and exogastric type (75%). The intraluminal type refers to the ectopia of the endometrium causing the thickening of the ureter wall and the hyperplasia of the muscular layer. Rare cases can also be seen as ectopic endometrium, causing polypoid growth of the ureter mucosa, protruding to the lumen. The exogastric type refers to the ectopic infiltration of ureteral serosa and peripheral tissue, causing peripheral inflammatory reaction and fibrosis leading to distortion and stenosis.[26] According to the intraoperative findings, the patient was diagnosed as the second type.

At present, it is generally believed that endometriosis may be associated with ectopic blood circulation, hematogenous spread of lymphoid, iatrogenic dissemination and immune abnormalities. Ureteral endometriosis is believed to be caused by lymphocyte metastasis. The exogastric type is caused by the residual of embryonic cells and the reflux of blood. According to the classification, endometrial invasion of the reproductive system leads to external oppression and stenosis of the ureteral lumen, resulting in loss of renal function in the exogastric type. The intraluminal type is often caused by the metastasis of lymph and vein,[27] which is manifested as ureteral obstruction or hematuria associated with menstruation,[28] and is often misdiagnosed as malignant tumor of ureter. The patients had medical history of uterine fibroids enucleation so that the endometrium caused by pelvic surgery could not be excluded. Therefore, in the process of gynecological surgery, we shall avoid endometriosis.

Conflicts of Interest Disclosure

The authors have no conflicts of interest related to this article.

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