IJCMBS

THE INTERNATIONAL JOURNAL OF CURRENT MEDICAL ANDBIOLOGICAL SCIENCESe-ISSN: 2791-8815

Case Report

Bilateral Ureteral Involvement in Endometriosis - Case Report

Bilateral Üreter Tutulumu Olan Endometriozis: Olgu Sunumu

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Highlights

• Ureteral endometriosis is a rare pathology that, if left untreated, can lead to renal damage.

• It should be considered in the differential diagnosis of resistant ureteral obstructions.

• Surgical intervention is the primary treatment for cases where obstruction develops due to ureteral endometriosis.

Abstract

Ureteral endometriosis is a rare pathology that, if left untreated, can lead to renal damage. In this case, we present a 48-year-old female patient with a rare persistent bilateral ureteral obstruction and hydroureteronephrosis.

Imaging revealed the obstruction but was not helpful in illuminating the etiology. In the patient who underwent exploratory laparotomy, the affected ureter and surrounding fibrotic foci were resected, and ureteroneocystostomy was performed. During follow-up, the patient's clinical condition improved, and no recurrence was observed.

Consequently, ureteral endometriosis is a rare pathology, and imaging may not be helpful in determining the etiology. It should be considered in the differential diagnosis of resistant obstructions.

Keywords: Ureteral endometriosis, upper urinary tract obstruction, obstructive uropathy

ÖZ

Üreteral endometriozis, tedavi edilmediği takdirde böbrek hasarına yol açabilen nadir bir patolojidir. Bu vakada, nadir görülen bilateral ureter obstrüksiyonu ve hidroüreteronefrozu olan 48 yaşında bir kadın hastayı sunuyoruz.

Görüntüleme yöntemleri obstrüksiyonu ortaya çıkardı ancak etiyolojiyi aydınlatmada yardımcı olmadı. Eksploratuvar laparotomi yapılan hastada etkilenen üreter ve çevresindeki fibrotik odaklar rezeke edilerek üreteroneosistostomi uygulandı. Takiplerinde hastanın klinik durumu düzeldi ve nüks gözlenmedi.

Sonuç olarak üreteral endometriozis nadir görülen bir patolojidir ve görüntüleme etiyolojiyi belirlemede yardımcı olmayabilir. Dirençli obstrüksiyonların ayırıcı tanısında düşünülmelidir.

Anahtar Kelimeler: Üreteral endometriozis, üst üriner sistem obstrüksiyonu, obstrüktif üropati

Introduction

Endometriosis is a chronic inflammatory pathology that is seen in approximately 10% of women in the reproductive period and it is characterized by pain and adhesion of the endometrial layer outside the uterus (1). Ureteral involvement of endometriosis, which also has atypical involvements such as gastrointestinal tract, lung, liver, pericard and brain is a rare but serious clinical form. According to research conducted in Europe, the prevalence of urinary tract endometriosis (UTE) varies between 0.3% and 12% among people affected by endometriosis, and between 20% and 52.6% among women with deep endometriosis (2, 3). Extrapelvic endometriosis most commonly affects the urinary system after the gastrointestinal tract, with the bladder being the most affected site in 85% of women with UTE, followed by the ureter at 10%, kidney at 4%, and urethra at 2% (4, 5). When it is located around the ureter, it may cause ureteral obstruction and secondary renal failure. In this case, we present a 48-year-old female patient with a rare persistent bilateral ureteral obstruction and hydroureteronephrosis.

Case Presentation

The patient presented with pain in bilateral flank, abdominal and groin areas, weakness, fatigue, and pollakiuria. Ultrasound (USG) revealed bilateral grade 3 hydroureteronephrosis. Creatinine was 1.15.

In the non-contrast abdominal computed tomography (CT) of the patient, bilateral hydronephrosis was seen, with prominent dilation of approximately 2/3 of both proximal ureters. The distal part was normal on both sides. Subsequently, CT urography was performed with the aim of excluding potential malignancies. The CT urography results demonstrated bilateral ureteral dilation up to the level of the iliac crosses, with normal distal portions. As a result, a diagnostic ureterorenoscopy (URS) was scheduled.

In URS, a circular stenosis was seen in the middle parts of both ureters, and both proximal ureters were dilated. No additional pathology was seen. Double J Stents (DJS) were placed on both sides.

DJS were removed after 75 days. One month later, the patient was admitted to our clinic again with nausea. In the pelvic doppler USG of the patient who had a history of endometriosis; the right ovary size was increased and measured as 55x40 mm. In the right ovary, two cysts were observed measuring 35x25 mm and 20 mm with no vascularization, which may be compatible with the follicle.

The magnetic resonance (MR) urography (Figure 1) did not reveal any additional pathological findings or malignancy-related involvement distal to the dilated portion of the ureter.

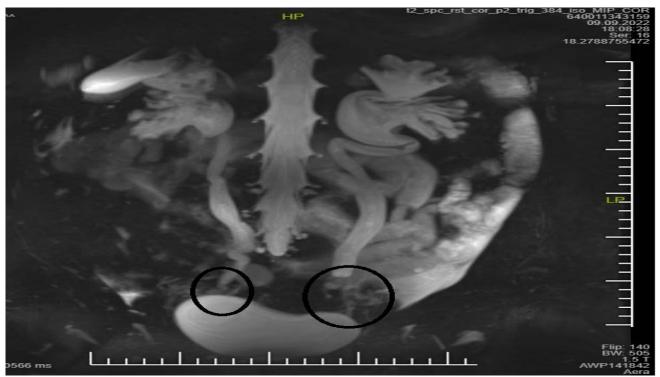


Figure 1.T2 MRI UROGRAPHY sequence image showing bilateral hydro-uretero-nephrosis secondary to ureteral stenosis. (*The stenotic segments have been marked*)

On MR urography, a cystic lesion was observed in the right adnexal region extending to the level of the culde-sac, measuring 47x27 mm at its widest point with T1A hypointense and T2A hyperintense features. The lesion contained septations but did not show any post-contrast pathological enhancement. Additionally, a T1A hyperintense and T2A hypointense lesion, measuring approximately 2 cm, was observed superior to the aforementioned lesion, which was consistent with a hemorrhagic cyst - endometrioma and did not show contrast enhancement in the post-contrast series.

Exploration was planned on the patient according to the data in the hand.

The patient was positioned in the supine position under general anesthesia. The abdomen was reached with a midline incision under the umbilicus. Two fixed fibrotic lymph nodes with a retracting appearance surrounding both ureters in a circular form were removed. The fibrotic parts of both ureters were removed and ureteroneocystostomy was performed using the Boari-flap and psoas-hitch method. DJS were placed in both ureters.

Pathology Result: Diffuse endometriosis and fibrosis were observed in the surrounding tissues of both ureteral segments that were removed. (Figure 2)

Follow-up: After 75 days, both DJS were removed. The patient did not have any symptoms or complaints during the 2-month follow-up. Our patient has been followed for 8 months without any problem.

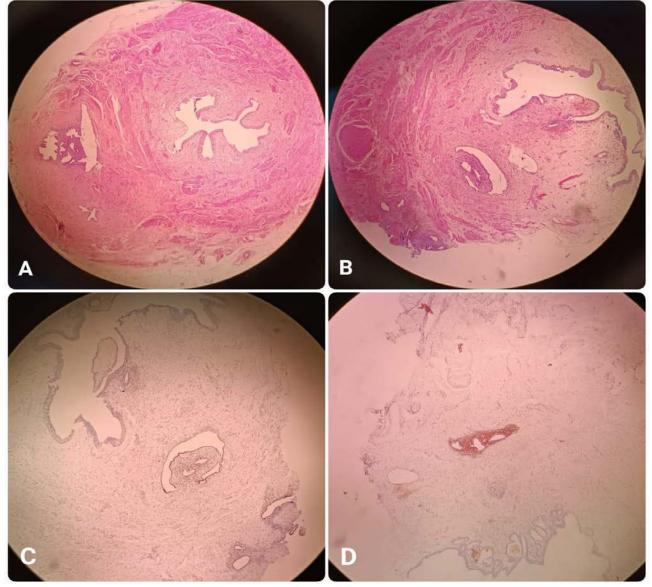


Figure2. Endometriosis foci in surrounding tissue adjacent to the ureteral segments. A-B: An endometriosis focus located on the left and ureteral tissue on the right side. (H&E; x 40).C: Endometrial epithelium staining positive with anti-estrogen antibody(x40).D: Endometrial stroma staining positive with anti-CD10 antibody (x40)

Discussion

The incidence of genitourinary system involvement in endometriosis is 1-2% (6). The most commonly involved organ in the urinary system is the bladder while ureter, kidneys and urethra may also be involved (7). Ureteral involvement can be histologically classified into two groups based on the type of involvement, namely extrinsic and intrinsic involvement (7). Extrinsic involvement refers to the presence of endometriosis in the periurethral tissue, while intrinsic involvement refers to the occurrence of endometriosis in the muscularis propria, lamina propria, and ureter lumen.

Early identification and treatment of this rare entity is crucial, given the potential risk of clinical progression to renal failure resulting from delayed diagnosis and treatment due to nonspecific symptoms and partial obstruction. Clinicians' awareness of the diagnosis and treatment of ureteral involvement can reduce associated morbidities. In the present case, the diagnosis was made after 9 months from the onset of symptoms.

Regarding the clinical manifestation of endometriosis, the symptoms are dependent on the location and extent of the endometrial tissue, and primarily comprise chronic pelvic pain, dysmenorrhea, deep dyspareunia, cyclical bowel complaints, fatigue, and infertility. In a relevant study, urinary symptoms were reported in only 15.9% of the patients. In our case, the patient's symptoms at admission were characterized by bilateral flank pain radiating to the abdomen and groin, as well as pollakiuria.

Imaging has an important place in the diagnosis of the disease. USG and IV urography are cheap and safe methods, they may be preferred in the first place. CT and MR urography are important because they are guiding for surgery and evaluating other possible diagnoses. In particular, magnetic resonance imaging (MRI) can detect even small endometrial foci (8). But in our case, it could not help the diagnosis. However, if clinical suspicion persists, endometriosis should still be considered.

Imaging plays a crucial role in the diagnosis of endometriosis. The initial assessment is performed USG and intravenous urography due to their cost effectivity and safety. However, CT and MR urography hold significance in guiding surgical intervention and evaluating other potential diagnoses. Of these, MRI is particularly useful at detecting smaller endometrial lesions. Nevertheless, in our case, it was not effective in aiding the diagnosis. Endometriosis should still be considered as a possible diagnosis in case of ongoing clinical suspicion.

Silloe et al. used the degree of contact between the ureter and endometriosis lesions in a study investigating the efficacy of MRI in distinguishing whether ureteral involvement is intrinsic or extrinsic, and they evaluated other MRI findings such as periureteral fat signal changes, the presence of parametrial nodules, and external ureteral compression of the lesion in contact with the ureter (9). In the same study, they compared MRI and surgery to distinguish between intrinsic and extrinsic involvement and concluded that MRI was more sensitive (91%-82%) but less specific (59-67%). In our case, MRI did not assist in diagnosis.

Treatment of ureteral endometriosis is primarily consists of resolvement of obstruction, prevention of renal injury and preservation of function.

One of the treatment alternatives for endometriosis is medical therapy which includes progestogens and GNRH agonists. On the other hand, this therapy which is indicated for early stage disease has an incomplete response and high recurrence rates after the discontinuation of therapy (10). Medical treatment should never delay surgical treatment in a condition such as hydronephrosis which may cause kidney injury (11). We planned surgical treatment on our case as soon as possible due to the bilateral hydroureteronephrosis and high creatinine levels.

In the surgical approach, which is a more permanent and definitive solution in the treatment of obstruction, the options are ureterolysis, ureteroureteral anastomosis and ureteroneocystostomy. The decision of which method will be used is determined according to the patient. Ureterolysis is indicated as the first choice in mild extrinsic cases. However, in case of intrinsic involvement uretero-ureteral anastomosis can be considered in limited involvement with the possibility of preserving the ureter (7). But high recurrence rates have been reported in this technique (12). The other option, ureteroneocystostomy, is particularly indicated in cases where the ureterovesical junction is involved and the uninvolved part of the distal end of the ureter is 1 cm or less (13). In our case, we removed the tissue surrounding the ureter in a circular manner and the fibrotic ureter at the lower end of both ureters. Then we performed ureteroneocystostomy.

Conclusion

As a result, ureteral endometriosis is a rare disease that is difficult to diagnose because of its non-specific symptoms. Suspecting the disease in the presence of associated symptoms is important in diagnosis. MRI is the gold standard for diagnosis and surgery is the most effective treatment option. Increasing awareness of this disease which can lead to kidney loss if its diagnosis is missed has a key role in achieving better results.

Acknowledgements: none

Ethical Approval: none

Author Contributions: Concept: HT Literature Review: EA, AT Design: MK Data acquisition: ANG Analysis and interpretation: MG Writing manuscript: MK Critical revision of manuscript: HT

Conflict of Interest: The author(s) do not have any potential conflict of interest regarding the research, authorship and/or publication of this article.

Financial Disclosure: This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

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