

URETERAL ENDOMETRIOSIS: A CASE REPORT AND A REVIEW OF THE JAPANESE LITERATURE

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A 42-year-old woman was referred to our hospital because of abdominal fullness and a large abdominal mass. Computed tomography (CT) demonstrated bilateral ovarian tumors, uterine myoma and left hydronephrosis. On excretory urography the left kidney was not visualized and retrograde pyelography (RP) revealed left hydronephrosis and a filling defect in the left lower ureter. Based on the diagnoses of endometriosis of bilateral ovaries, uterine myoma and a left ureteral tumor, abdominal total hysterectomy, right salpingo-oophorectomy and partial ureterectomy were performed. Pathologically, in the uterus, both leiomyoma and adenomyosis, and endometriosis of the right ovary and ureter were diagnosed. Medication with buserelin acetate was started.

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Key words: Endometriosis, Ureter

INTRODUCTION

Endometriosis is a common disease, affecting as many as 1 in 15 women of reproductive age¹⁾, but cases occurring in the urinary tract are rare, especially in the ureter. We report our experience of a case of ureteral endometriosis in which retrograde pyelography showed a filling defect in the left ureter that was suspected of malignancy. A brief review of ureteral endometriosis in the Japanese literature is presented.

CASE REPORT

A 42-year-old Japanese woman was admitted to our hospital with complaints of abdominal fullness and a palpable mass in the lower abdomen in November 1999. Physical examination revealed a mass that was round and firm, but not tender, in the suprapubic area. Urinalysis showed microscopic hematuria with 10-19 red blood cells in a high-power field. Urine cytology was negative for malignancy. Initial blood counts and blood chemistry, including the LDH level, were entirely normal. Serum tumor markers were within normal ranges except for the elevated CA125 level of 56 U/ml (normal range <50 U/ml).

Transvaginal ultrasonography demonstrated multiple iso-dense areas in the uterine muscle and swelling of the right ovary. Abdominal ultrasound showed left hydronephrosis with no renal parenchyma. Excretory urography showed a left non-functioning kidney. An enhanced CT scan revealed multiple hypodense lesions in the uterine muscle (Fig. 1A), and swelling of bilateral ovaries. Furthermore, it revealed severe hydronephrosis in the left kidney (Fig. 1B). Retrograde pyelography showed a filling defect in the lower third of the left

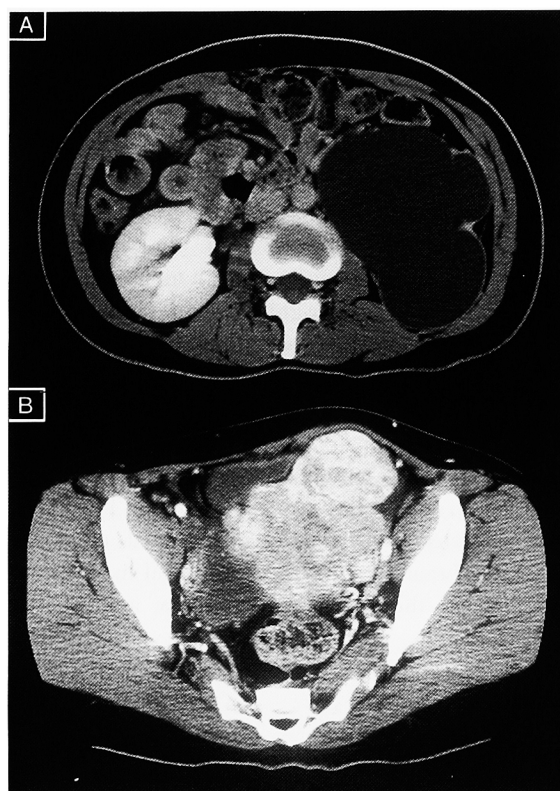


Fig. 1. Enhanced CT scans. A: The abdominal CT scan shows severe hydronephrosis in the left kidney. B: The pelvic CT shows multiple hypodense lesions in the uterine muscle, and a nodular mass adhering anterior to the uterus.

ureter that was 8 cm from the ureteral orifice (Fig. 2). From these findings, the diagnoses of uterine myoma, endometriosis of bilateral ovaries and ureteral tumor were made.

At laparotomy, a large pelvic inflammatory mass was found arising from the uterus and involving

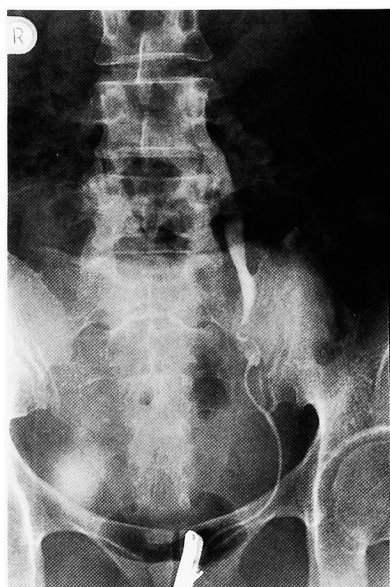


Fig. 2. Retrograde pyelography revealed a filling defect in the lower third of ureter.

both ovaries. A 65 mm right chocolate ovarian cyst was found. The left lower ureter, left ovary and sigmoid colon adhered to each other with fibrous connective tissue, partially obstructing the left ureter at the midpelvic level. Concurrently, a polypoid tumor was found in the ureter proximal to the adhering portion. Resection of the mass was undertaken conjointly with a consultant gynecologist, and total hysterectomy, right salpingo-oophorectomy and partial ureterectomy with ligation of the distal end of the ureter were performed. There had been no agreement about nephroureterectomy with the patient. Pathologically, the uterus with both leiomyoma and adenomyosis, endometriosis of the right ovary and ureter were diagnosed. The small mass detected by RP was constructed of intact transitional mucosa, but the submucosal tissues were replaced by the endometrial stroma and benign

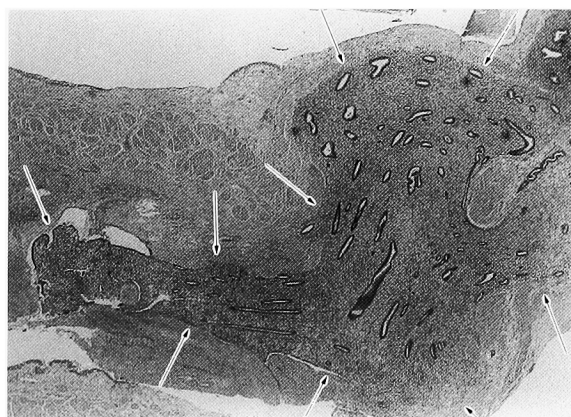


Fig. 3. Endometriosis, with glands and surrounding stroma within wall of ureter. Note endometrial tissues and stroma (arrows) in muscularis and submucosa (H & E stain, ×40).

proliferative endometrial glands (Fig. 3). No malignant cells were seen in any specimen, including the left ureter. As a result, the left hydronephrosis was concluded to be caused by the intrinsic type of ureteral endometriosis. Postoperatively, a GnRH analogue (buserelin acetate) was started.

DISCUSSION

Endometriosis is the aberrant growth of endometrial tissue at ectopic sites, being found in 15 to 20% of gynecological laparotomies²⁾, but urinary tract involvement occurs in only 1.2% of affected premenopausal women³⁾, the bladder, ureter and kidney being affected at a ratio of 40:5:1⁴⁾

The etiology of endometriosis currently falls under either the embryonic, metaplastic, or migratory theories⁴⁾. The migratory theory prevails today.

The first case of ureteral endometriosis in Japan was reported by Hirota in 1971⁵⁾. Excluding those cases in which the primary involvement was in the bladder with secondary to ureterovesical junction obstruction, we reviewed 104 cases published in Japan prior to 1999, and added the present case to make a total of 105.

Ureteral endometriosis usually occurs in women between menarche and menopause but may also appear postmenopausally⁶⁾. In the present series of 105 cases, the patients' ages ranged from 18 to 56, the average being 39.1 years. In the majority of our series it occurred between ages 31 and 45 (68.3%), with the greatest number in the period between 41 and 45 (29.7%). Endometrioma usually obstructs the ureter by a gradual process of extrinsic fibrous compression⁷⁾, and diagnosis is later than that of adenomyosis.

Patients usually presented with menstrual dysfunction, flank, lumbar or lower abdominal pain, or gross hematuria (Table 1). The pain was associated with menses in only 4.0% of the cases. In 38.5% of the cases with gross hematuria, the ureteral involvement was intrinsic. In the group with

Table 1. Symptoms in 105 patients with ureteral endometriosis

Symptoms	Number of patients (percent)
Pain	68 (68.7%)
Flank, lumbar	51
Abdominal	13
With menses	4
Menstrual disorders	13 (13.1%)
Bladder symptoms	7 (7.1%)
Chills and fever	10 (10.1%)
Hematuria	13 (13.1%)
Negative	12 (12.1%)
Not stated	6
Total	105

negative symptoms, 83.3% were referred to the clinics because of the existence of hydronephrosis. Several series have noted a high incidence of previous pelvic surgery (60 to 70%) in patients with endometriosis of the urinary tract^{4,8)} Previous gynecological operations were reported in 27 of the 83 cases (32.5%), including total hysterectomy or oophorectomy (15 cases) and induced abortion (12 cases).

The majority of endometrial lesions were located in the lower third of the ureter (98 cases) and all the others in the middle third (5 cases). Ureteral endometriosis is usually unilateral^{2,4,8)} In only 12.6% of cases did bilateral involvement occur. Of 103 case reports in which the side of the involvement was indicated, 51 occurred on the right side, 39 on the left side, and 13 were bilateral.

Endometriosis involving the ureter has been separated into two types: 1) intrinsic and 2) extrinsic⁹⁾ Of a total of 79 classified cases, 20 intrinsic (25.3%) and 59 extrinsic (74.7%) ones have appeared in the literature.

It seems to be very difficult to make the correct diagnosis before exploration or medical therapies. Only 30 cases (37.5%) could be diagnosed as ureteral endometriosis, although 23.8% of them were considered to be ureteral tumors, 15.0% ureteral stenoses with unknown causes, and 3.8% invasion of ovarian tumors. Kerr reported that more intravenous urography should be ordered in patients with previous gynecologic surgeries¹⁰⁾.

The specific mode of treatment should result from a combined urologic and gynecologic evaluation aimed to improve fertility, relieve obstruction and preserve renal function^{3,6)}. Numerous modalities have been utilized to treat ureteral endometriosis, including

surgical castration alone or in combination with ureterolysis, segmental ureterectomy, as shown in Table 2. An alternative to castration, consisting of medical hormone manipulation with estrogen-progesterone combination, danazol or a GnRH analogue, was administered to 52.5% of the patients. Of 95 case reports in which the renal function was indicated, 15.8% lost renal function on the affected side. There are also instances when a kidney beyond salvage is best treated by nephroureterectomy (14.9%) or no surgery at all. However, since the decision becomes extremely difficult for a young nullipara desirous of maintaining her childbearing potential, more conservative treatment modalities have been utilized⁷⁾. Non-surgical therapies were documented in 24.8% of our series.

In summary, ureteral obstruction caused by endometriosis is uncommon. It is, however, an important complication that imposes a 16% chance for permanent loss of renal function on the affected side. When confronted with a patient exhibiting symptoms of endometriosis, particularly those symptoms related to the genitourinary tract, diagnosis of urinary tract involvement demands suspicion and careful pelvic examination. The ideal course of treatment should be individualized, taking into account the risks of surgery versus the limitations of medical therapy. The standard management is surgical. The clinical response of the endometriosis to hormonal therapy is excellent, but drugs are unlikely to relieve endometriotic ureteric obstruction once dense fibrosis has occurred.

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Table 2. Types of treatment for 105 patients with ureteral endometriosis

Treatment	Number of patients (percent)
Kidney preservation	63 (62.4%)
partial resection with ureteroureterostomy	19
ureterocystoneostomy	23
ureteral lysis	13
nephrostomy	8
Non-surgical therapy	25 (24.8%)
double-J stent insertion	13
hormonal therapy alone	12
Nephroureterectomy	15 (14.9%)
reasons: non-functioning kidney	7
suspicion of ureteral tumor	7
unknown	1
Combined with gynecological operation	37 (36.6%)
Hormonal therapy administration	53 (52.5%)
Not stated	4
Total	105

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和文抄録

尿管エンドメトリオーシスの1例（本邦報告例105例の臨床的検討）

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田 沼 康

42歳，女性．腹部膨満感にて当院受診．検尿上，顕微鏡学的血尿を認め，血中 CA125 は軽度高値を示した．腹部 CT 検査では左水腎症と子宮に隣接する巨大腫瘍を認め，逆行性腎盂造影では左下部尿管に陰影欠損を認めた．子宮筋腫および左尿管腫瘍の診断から右附属器および子宮摘除術と尿管部分切除術を施行した．組織学的に左尿管腫瘍は粘膜下に存在する外性子

宮内膜症であった．1971年以来本邦では104例が報告され，術前診断は37.5％と，悪性腫瘍との鑑別が困難な症例も少なくない．一方で腎保存術も62.4％で施行され，術後腎機能も良好な症例もみられる．尿路も視野に入れた各種検査と妊孕性を考慮した治療法の選択が望まれる．

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