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CASE REPORT

Disseminated peritoneal leiomyomatosis with malignant transformation involving right ureter: A case report

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Abstract

BACKGROUND

Disseminated peritoneal leiomyomatosis (DPL) with myxoid leiomyosarcoma is a rare variant of leiomysosarcoma, and hematuria as a presenting symptom has never been reported. Through this case report, we emphasize the investigation of the etiology, clinical presentation, diagnosis, treatment, and prognosis of DPL with malignant changes mimicking metastatic urinary tract cancer and to help develop further clinical management.

CASE SUMMARY

We describe a case of DPL with malignant transformation involving the right ureter after laparoscopic hysterectomy. An exploratory laparotomy was performed and all visible nodules were surgically removed. DPL with focal malignant transformation to myxoid leiomyosarcoma was confirmed based on pathology results.

CONCLUSION

Professionals who preoperatively diagnose DPL with malignant change to myxoid leiomyosarcoma involving the genitourinary tract should consider symptoms of abdominal pain, hematuria, and imaging of disseminated pelvic tumors in women, especially those with prior history of laparoscopic hysterectomy. Early complete removal of all tumors is the cornerstone to prevent DPL from malignant changes.

Key Words: Disseminated peritoneal leiomyomatosis; Leiomyosarcoma; Laparoscopic

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Core Tip: Disseminated peritoneal leiomyomatosis (DPL) is a rare disease characterized by the presence of multiple nodules composed of smooth muscle cells located in both peritoneal and extraperitoneal spaces of the abdomen. Malignant changes in DPL correspond to a rare variant of leiomyosarcoma characterized by aggressive behavior. We describe a case of DPL with malignant transformation involving the right ureter after laparoscopic hysterectomy, mimicking urothelial carcinoma with peritoneal carcinomatosis. The aim of our case report is to investigate the etiology, clinical presentation, diagnosis, treatment, and prognosis of DPL and to help develop further clinical management of this disease.

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INTRODUCTION

Disseminated peritoneal leiomyomatosis (DPL) is a rare disease characterized by the presence of multiple nodules composed of smooth muscle cells located in both the peritoneal and extraperitoneal spaces of the abdomen[1]. This disease is usually observed in women of reproductive age. To date, hundreds of cases have been reported[2,3]. However, malignant changes in DPL with myxoid leiomyosarcoma are rare, and hematuria as a presenting symptom has never been reported[4,5]. Herein, we present a case of DPL with malignant transformation involving the right ureter after laparoscopic hysterectomy. The aim of our case report is to investigate the etiology, clinical presentation, diagnosis, treatment, and prognosis of DPL with malignant changes mimicking metastatic urinary tract cancer and to help develop further clinical management.

CASE PRESENTATION

Chief complaints

A 72-year-old woman presented with gross hematuria one month before visiting our hospital.

History of present illness

This patient also noted intermittent abdominal cramping pain for half a year. The patient reported no urinary urgency, dysuria, flank pain, or fever.

History of past illness

The patient had undergone laparoscopic hysterectomy for uterine leiomyoma at another institution 2 years ago prior to her visit. Prior medical histories of hypertension, diabetes mellitus, and gout were noted.

Personal and family history

The patient had no relevant personal or family history.

Physical examination

Physical examination revealed multiple painful hard subcutaneous nodules in the lower abdomen.

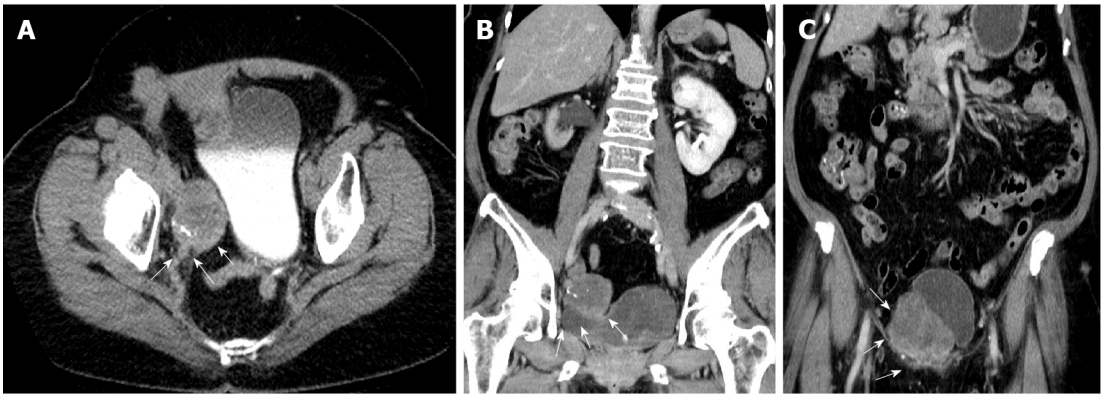

Figure 1 Abdominal computed tomography demonstrated that the tumor extended from the right distal third ureter to the ureterovesical junction. A: White arrowhead in the axial view; B and C: Coronal view.

Laboratory examinations

Laboratory examination revealed an elevated leukocyte count of 15109/mL, hemoglobin count of 12.9 g/dL, and C-reactive protein count of 3.72 mg/dL. Urine cytology, urinalysis, blood coagulation, kidney function, and liver function were all within normal range.

Imaging examinations

Abdominal computed tomography (CT) suggested urothelial carcinoma of the right lower third ureter with hydronephrosis and multiple seeding lesions at the anterior abdominal wall, subcutaneous fat, and bilateral inguinal areas (Figure 1).

FINAL DIAGNOSIS

Percutaneous ultrasound-guided biopsy of the most superficial lesion in the right lower quadrant of the abdomen was performed first. The tumor cells showed smooth muscle cell differentiation, which was compatible with leiomyoma as evidenced by pathology results.

TREATMENT

Because of persistent lower abdominal pain, the patient requested all tumors to be removed. An exploratory laparotomy was conducted with a lower midline incision, and multiple tumors of different sizes were found attached to the rectus muscle, bilateral inguinal areas, right ureter, and sigmoid colon (Figure 2). All nodules were meticulously dissected and resected with margins. Segmental resection of the right ureter, ureteroneocystostomy, partial resection of the sigmoid colon wall with primary closure, and transverse colostomy were also performed. The postoperative convalescence was uneventful. Final pathology revealed DPL with focal malignant transformation to myxoid leiomyosarcoma. Microscopically, the tumor was composed of malignant spindle cells with moderate to abundant eosinophilic cytoplasm arranged in interlacing fascicles (Figure 3).

OUTCOME AND FOLLOW-UP

Abdominal discomfort and pain improved significantly postoperatively. The transverse colostomy was closed after 3 mo. Adjuvant systemic chemotherapy was recommended, with periodic follow-up imaging; however, the patient opted for active surveillance only. The patient was doing well without evidence of recurrence 24 mo after the operation.

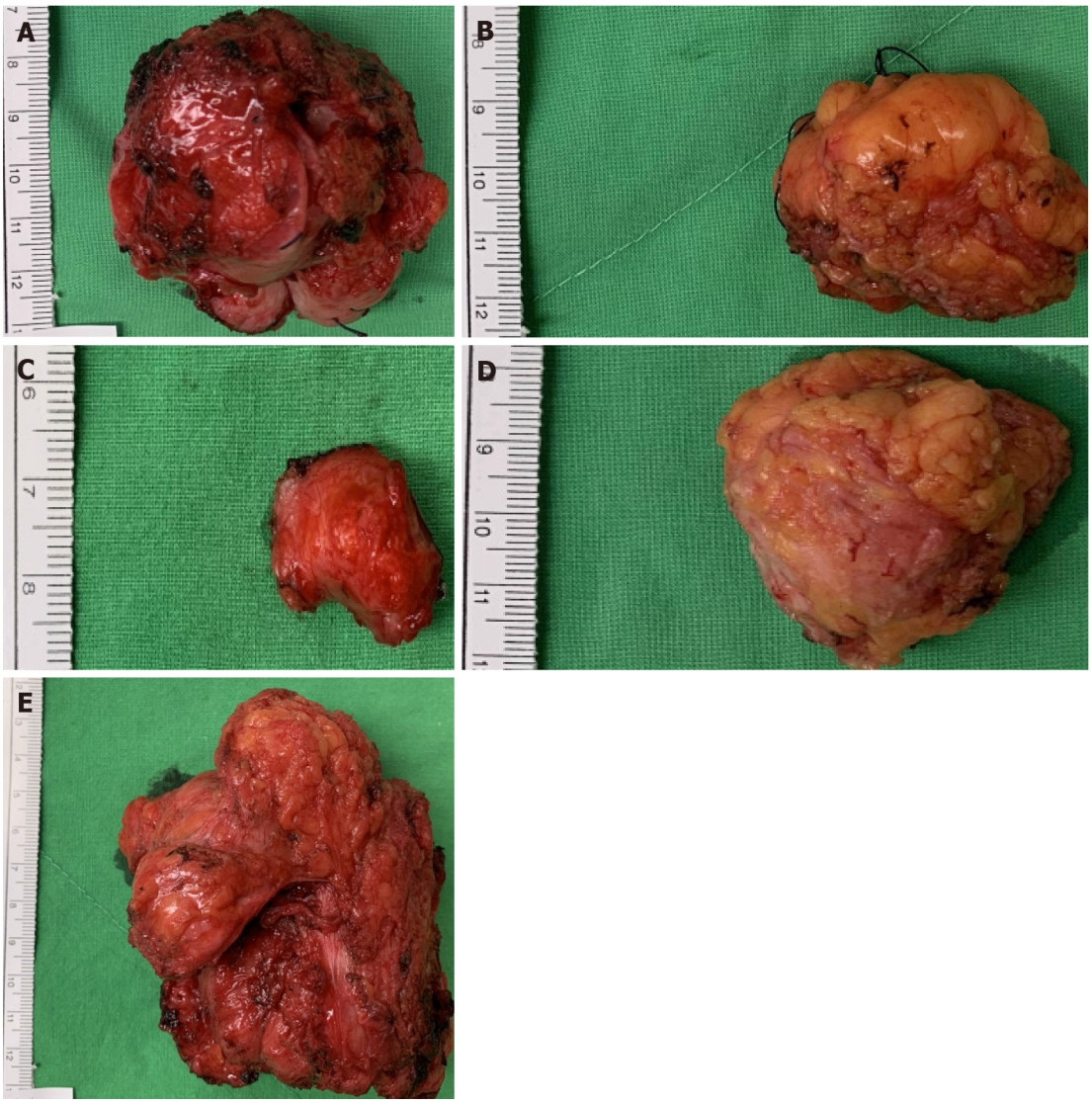

Figure 2 Intraoperative images of firm gray-whitish tumors. A: Located at right distal ureter; B: Located at right inguinal area; C: Located at left inguinal area; D: Located at abdominal wall; E: Located at sigmoid colon.

DISCUSSION

The etiology and pathophysiology of DPL are not yet well-established. Most reported cases are related to a history of laparoscopic hysterectomy or uterine myomectomy. Iatrogenic contamination after morcellation of myoma during laparoscopic surgery is considered to be a possible cause of DPL[1]. In the current case, the patient underwent laparoscopic hysterectomy 2 years ago; the use of a power morcellator may enhance potential for tumor implantation and dissemination[6,7].

Most patients with DPL are asymptomatic. In these patients, DPL is found incidentally through imaging. Several non-specific symptoms, including abdominal pain, distension, menostaxis, and bleeding from the rectum or vagina have been reported[7-9]. In the present case, the patient reported abdominal pain for one month, which is the most common manifestation of DPL. In addition, she first complained of gross hematuria and was referred to our urology outpatient department. To the best of our knowledge, this is the first report of DPL with hematuria as an initial presentation that could mimic urothelial cancer with peritoneal carcinomatosis[10,11]. Preoperative diagnosis of DPL is challenging, and only histopathologic examination can discriminate DPL from peritoneal metastatic malignancies or benign metastasizing leiomyoma[9,12]. Hence, we performed percutaneous ultrasound-guided biopsy of the abdominal wall lesion to delineate the nature of these tumors, and pathology showed a smooth muscle tumor compatible with leiomyoma.

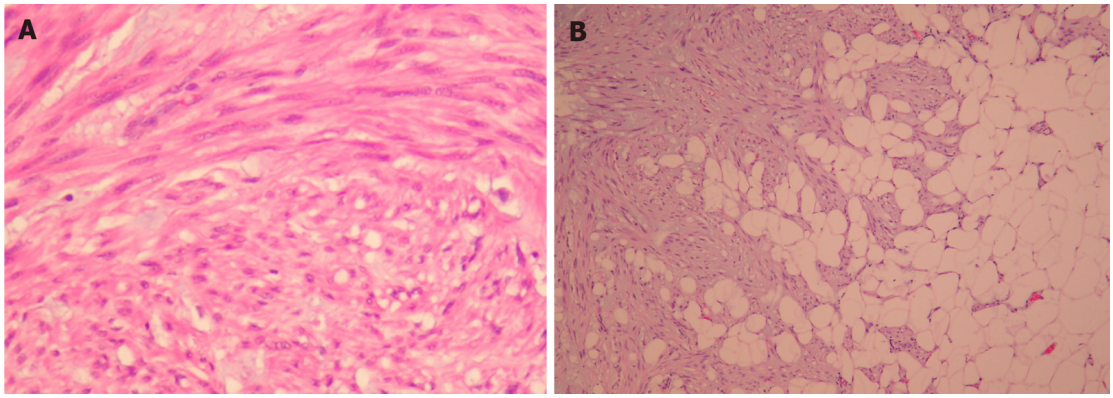

Figure 3 Histological appearance of disseminated peritoneal leiomyomatosis tumor. A: The tumor has hypercellular areas with focal myxoid matrix, composed of spindled-shape neoplastic cells arranged in interlacing fascicles and storiform growth pattern (original magnification, 200 ×); B: Myxoid matrix infiltrated into adipose tissue (original magnification, 100 ×).

DPL is histologically benign but can transform into a malignant leiomyosarcoma. The duration between the initial diagnosis of DPL and malignant changes varies from 1 mo to 8 years[4,13]. This duration can be under- or overestimated because malignant change may occur focally and insidiously, which makes histological sampling difficult. In the current case, the duration of malignant transformation was speculated to be less than 2 years according to the patient's operative history. Nevertheless, the focal tumor specimen involving the right ureter revealed a smooth muscle tumor with infiltrative borders, rich myxoid matrix, spindled neoplastic cells arranged in interlacing fascicles, mitotic activity up to 4 mitoses in 10 high-power fields, and foci of tumor necrosis. These findings are compatible with DPL with focal malignant transformation to myxoid leiomyosarcoma.

Standard treatment for DPL is debated. Since most DPLs are found in women of reproductive age, conservative treatment should be considered. Treatment of DPL includes a variety of treatments, such as active surveillance, hormone therapy, debulking surgery, chemotherapy, and radiation therapy, while surgical removal remains the mainstay because of its malignant potential[13,14]. In our case, no adjuvant chemotherapy or radiotherapy was administered.

CONCLUSION

In conclusion, we present a case of DPL with focal malignant transformation involving the right ureter, mimicking urothelial carcinoma with peritoneal carcinomatosis. Preoperative diagnosis of malignancy is usually challenging. DPL with malignant change to myxoid leiomyosarcoma involving the genitourinary tract should be weighed against differential diagnoses in women presenting with abdominal pain and hematuria with imaging of disseminated pelvic tumors, especially those with prior history of laparoscopic hysterectomy. Early complete surgical resection of all tumors is the most important factor in preventing malignant transformation of DPL, even though it has a relatively favorable outcome.

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