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Case report

Leiomyomatosis peritonealis disseminata in laparoscopic port site and abdomino-pelvic cavity: A case report ^{☆,☆☆}

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ABSTRACT

Leiomyomatosis peritonealis disseminata (LPD) is a rare clinical condition characterized by the development of multiple smooth muscle-like nodules in the peritoneal or abdominal cavity. Here, we report a case of a patient who was diagnosed with LPD after laparoscopic myomectomy with power morcellation. Growing evidence has shown that LPD might develop after using power morcellation for hysterectomy or myomectomy, and this can worsen the prognosis if the spreading tissue contains malignancies, such as leiomyosarcoma. Thus, it is crucial to use laparoscopic morcellation for gynecologic procedures cautiously, and the use of a containment system is even better. If LPD develops without evidence of malignancy, the primary treatment is surgical intervention, and gonadotropin-releasing hormone agonists, aromatase inhibitors, and selective progesterone receptor modulators can be prescribed as adjuvant therapies for recurrent or refractory cases.

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Introduction

Leiomyomatosis peritonealis disseminata (LPD) is a rare benign disease that presents with dissemination and prolif-

eration of peritoneal lesions originating from smooth muscle cells. The etiology and pathophysiology remain unclear. The possible causes could be divided into hormonal, subperitoneal mesenchymal stem cells, metaplasia, genetic, or iatrogenic after morcellation of myoma [1]. Minimally invasive

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laparoscopic surgery for myomectomy or hysterectomy has become predominant for years. Morcellation is frequently used in laparoscopic procedures in which large specimens can be removed through small abdominal excisions. However, an increasing number of studies have demonstrated that there are risks of disseminated tissue spreading in the abdominal cavity, either benign fibroids or malignant sarcoma, which worsens the prognosis. We present a case of iatrogenic LPD after laparoscopic myoma morcellation with the hope that physicians will be persuaded to use power morcellation cautiously.

Case description

A 34-year-old woman presented with a palpable mass in the right lower abdomen. She received laparoscopic myomectomy with power morcellation 7 years ago (Fig. 1). Physical examination revealed 1 tumor approximately 4 cm in diameter in the abdominal wall and another tumor approximately 10 cm in diameter in the right lower quadrant of the abdomen. There were no other associated symptoms. Computed tomography showed 1 solid tumor in the subcutaneous layer of the abdominal wall (arrow, Fig. 2A) and solid tumors in the right paracolic gutter (arrow, Fig. 2B) and in the mesentery of the small bowels (arrowhead, Fig. 2B). Laboratory tests were unremarkable, with normal serum cancer antigen-125 (CA-125) levels.

During the operation, several well-defined tumors (15 cm, 11 cm, 3 cm, and 2 cm in diameter) attached to the peritoneum and mesentery were detected (Fig. 3). Another solid tumor, approximately 4 cm in diameter, was found in the subcutaneous layer of the right abdominal wall. There was 1 subserosal myoma, 3 cm in diameter, in the uterus. The bilateral adnexa were unremarkable. Finally, all lesions were resected without any residual tumor. The postoperative course was smooth, and she was discharged 6 days after the operation.

The histopathologic analysis reported spindle cells arranged in intersecting fascicles, which were immunohistochemically positive for smooth muscle actin and h-caldesmon, in favor of leiomyoma (Fig. 4). Thus, she was diagnosed with iatrogenic LPD, which refers to uterine leiomyoma disseminated in the abdominopelvic cavity after medical interventions. She then received 6 courses of monthly leuprorelin acetate after the operation, and follow-up computed tomography showed no recurrent tumor 9 months after the operation (Fig. 5).

Discussion

LPD is a rare disease characterized by the presence of multiple leiomyoma nodules on the peritoneum, omentum, or mesentery of the bowel. It was first described in 1952, and to date, approximately 200 cases of LPD have been reported [2]. LPD or parasitic myoma have been reported after laparoscopic my-

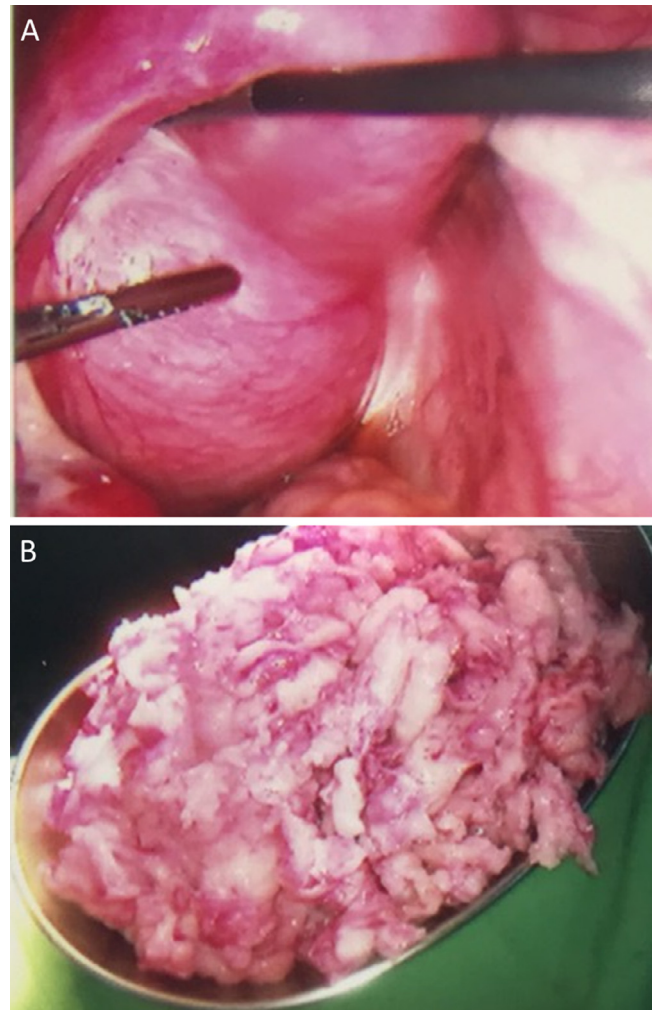


Fig. 1 – The operation pictures of laparoscopic myomectomy with power morcellation 7 years ago. (A) The leiomyoma; (B) Myoma after power morcellation.

omectomy with morcellation [3–5]. The incidence varies between 0.1% and 1% [6,7].

The etiology remains unclear. It is assumed that fragments of the uterus or myoma can be implanted into the peritoneum with subsequent dissemination. In general, the diagnosis of LPD is based on medical history, clinical images, intraoperative observations, and pathological findings. The most common symptoms are abdominal and pelvic pain, a palpable mass in the abdomen, and deep dyspareunia [8]. Rare manifestations, including ascites or associations with endometriosis, have been demonstrated in some studies [9,10]. The mainstay of treatment for LPD is surgery, and most patients have no recurrent tumors after complete surgical resection. GnRH agonists may be added as adjuvant therapy postoperatively. A number of studies have reported that GnRH agonist therapy following surgery will prevent the appearance of new lesions for 5 years [11,12]. Treatment with aromatase inhibitors and selective progesterone receptor modulators (eg, ulipristal acetate) have also been described as effective [13,14]. Although

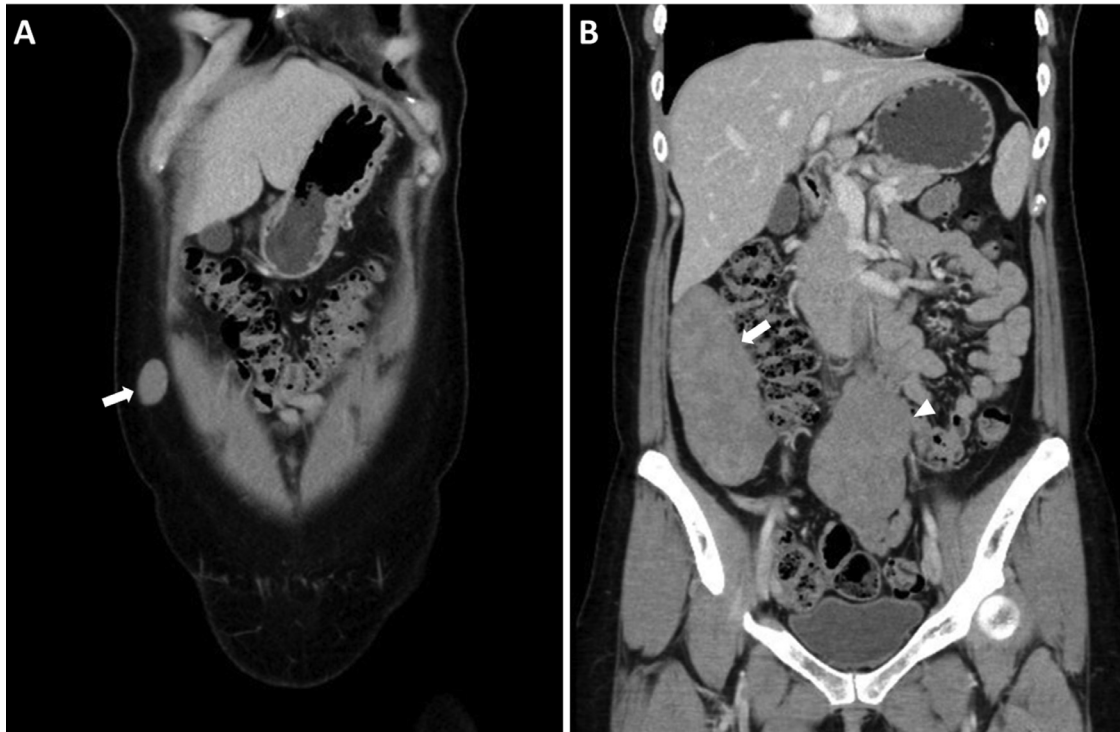


Fig. 2 – The computed tomography showed 1 solid tumor in the subcutaneous layer (arrow, A) and solid tumors in right paracolic gutter (arrow, B) and mesentery of small bowels (arrowhead, B).

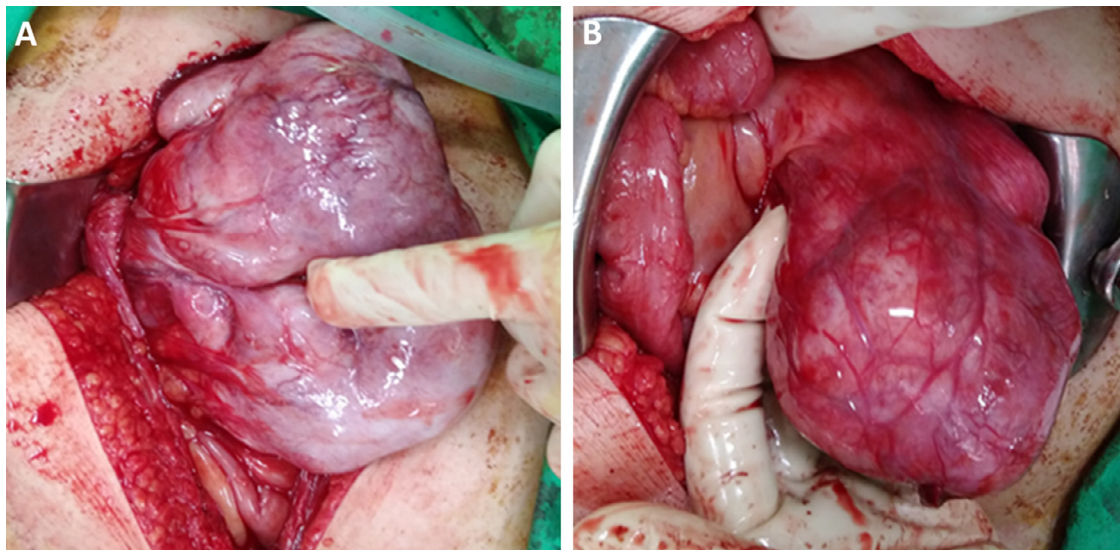


Fig. 3 – The operation pictures of the laparotomy. The well-defined tumors attached to the peritoneum (A) and mesentery (B).

their prognosis is very good, reoperation for diagnosis and treatment is still stressful for these patients. For rare and advanced cases, 1 study reported that objective tumor response was achieved and sustained for 1 year with systemic chemotherapy [15].

In addition, special attention should be given to the risk of malignancy appearance with possible resultant upstaging of the disease and worsened prognosis. Protective measures should be taken to prevent spreading of morcellated fragments of leiomyoma in endoscopic surgery.

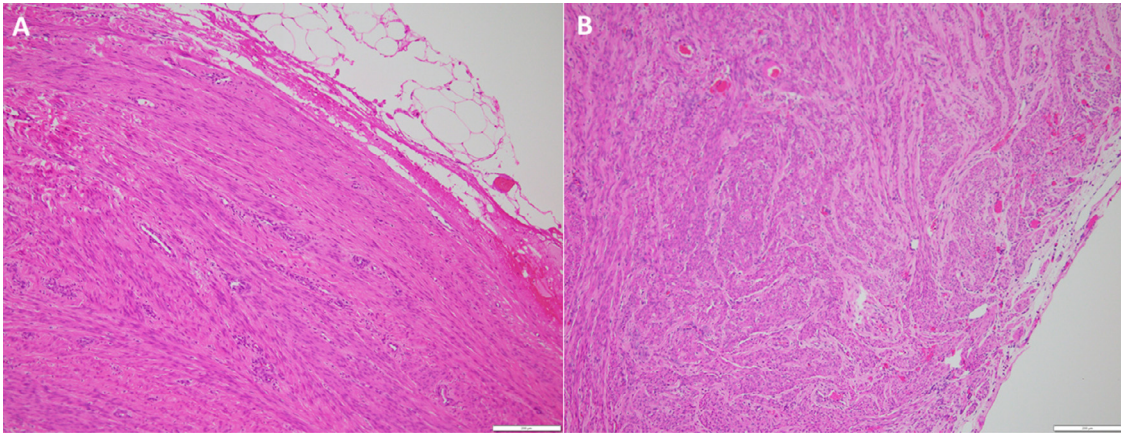


Fig. 4 – The pathology reported smooth muscle neoplasm in favor of leiomyoma in the port-site tumor (A) and abdominal tumor (B) (H&E stain, 100x).



Fig. 5 – The follow-up computed tomography showed no recurrent tumor in 9 months later.

Patient consent

We had got the patient consent. We used non-identifiable images in the manuscript.

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