Leiomyomatosis peritonealis disseminata (LPD) five years after laparoscopic myomectomy: a case report and review of the literature

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***Abstract*— Leiomyomatosis peritonealis disseminata (LPD) is an extremely rare smooth muscle benign tumor in women's reproductive age. It is characterized by the dissemination of multiple small nodules on the peritoneal surface, which are chiefly comprised of smooth muscle. To date, there are only around 100 cases previously reported in the literature. In the present study, we described the case of a patient who developed leiomyomatosis peritonealis disseminata five years after a laparoscopic myomectomy, consisting of multiple nodules located on the surface of the sigmoid colon and ileum mesenterium, parietal peritoneum of trocar puncture hole, ovarian, bladder and Douglas pouch.**

***Index Terms*— laparoscopic, LPD, myomectomy, tumor**

I. INTRODUCTION

Leiomyomatosis peritonealis disseminata (LPD) is a rare smooth muscle tumor, which is first described by Willson and Peale in 1952, and later named as LPD by Taubert *et al* in 1965 [1]-[2].This disease is characterized by the dissemination of multiple pelvic and abdominal nodules throughout the omental and peritoneal surfaces. The etiology of LPD is unknown, including several theories, such as the genetic, hormonal and iatrogenic theories [3].Since this disease is rarely reported, we herein present the case of a patient who developed LPD following laparoscopic myomectomy and myoma morcellation, which was associated with pelvic and abdominal dissemination.

II. CASE REPORT

A 46-year-old multiparous woman was admitted to the Gynecology of Huiya Hospital of The First Affiliated Hospital, Sun Yat-Sen University with irregular vagina bleeding and pelvic lumps. The patient had a history of laparoscopic myomectomy and myoma morcellation in 2011, and it was confirmed leiomyoma by the pathology after surgery. After checking the clinical notes, we found that the patient underwent laparoscopic removal of multiple, intramural myoma of 4-6 cm that was morcellated during removal. During the first 3 years after surgery, the patient remained asymptomatic and nothing abnormal was found by transvaginal ultrasonography. However, a pelvic ultrasound scan showed small uterine fibroids during the health examination in 2014. The patient still remained asymptomatic. But uterine myomas increased. In the past 2 months this year, the patient suffered from irregular vagina bleeding. A pelvic ultrasound scan showed multiple uterine fibroids and multiple pelvic lumps with unclear nature. The serum cancer antigen 125 (CA-125) level was 36.4 U/ml (normal range, 0-35 U/ml). Other standard tumor markers were within normal limits: carcinoembryonic antigen (CEA) 0.62 ng/ml; cancer antigen 72-4 (CA72-4) 6.52 U/ml; carbohydrate antigen 19-9 (CA 19-9) 19.68 U/ml; alpha-fetoprotein (AFP) 4.04 ng/ml. Moreover, a contrast enhanced MRI scans in another hospital revealed multiple pelvic lumps, which were thought about to be multiple subserous myomas, but not excluded ovarian tumor. Thus, the patient was admitted to our hospital and scheduled to undergo abdominal laparotomy. Before surgery, the surgical procedure (type of incision, risks, possible complications…) was explained by our consultant. During this procedure, multiplegrey red nodules of different size were identified, involving the sigmoid colon and ileum mesenterium, parietal peritoneum of trocar puncture hole, ovarian, bladder and Douglas pouch (size between 2 and 5 cm) (Fig.1, 2). Intraoperative frozen section pathology of mass lesions revealed benign smooth muscle tumors. Since the patient was 46 years old and she was not planning to have more children, she subsequently underwent abdominal total hysterectomy, bilateral salpingo-oophorectomy and excision of other disseminated nodules. The final histopathologic examination was confirmed with leiomyomatosis peritonealis disseminata. There were not any intraoperative or postoperative complications and the patient was discharged smoothly after surgery. Two months later, the patient was asymptomatic with CA125 level less than 35U/ml.

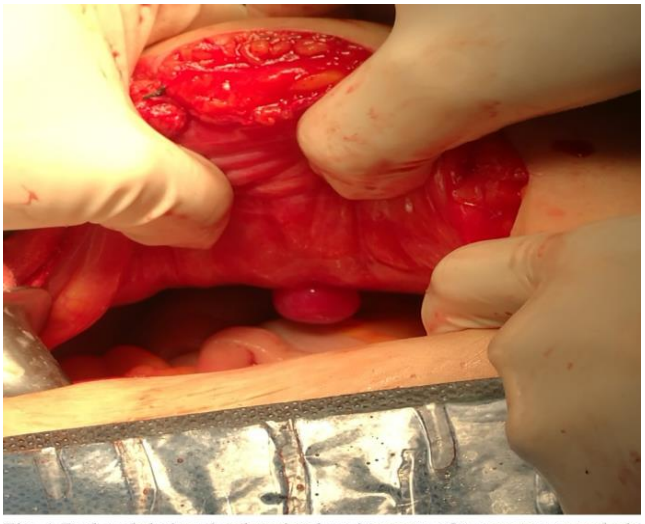

Fig. 1 Red nodule involved parietal peritoneum of trocar puncture hole

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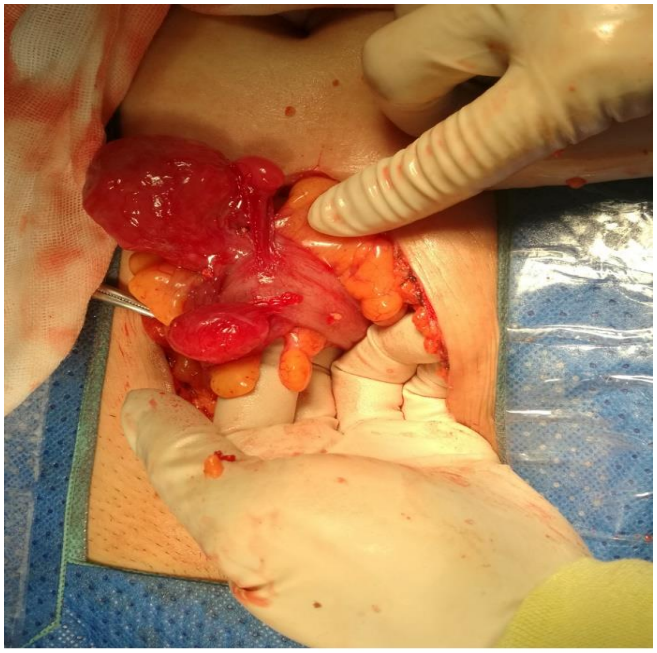

Fig. 2 Red nodules involved the sigmoid colon and ileum mesenterium

III. .DISCUSSION

LPD is a rare smooth muscle tumor described firstly in 1952, characterized by dissemination of multiple pelvic and abdominal smooth muscle nodules which appear grossly malignant but histological benign [1]. The etiology of LPD remains largely unknown, including several theories, such as the genetic, subperitoneal mesenchymal stem cell metaplasia, hormonal and iatrogenic theories (including myoma morcellation during laparoscopic surgery) [3]. Patients with LPD may present non-specific symptoms, including irregular vagina bleeding, abdomen pain, gastrointestinal bleeding and peritonitis (LPD implants in the bowel wall). It's difficult to be diagnosed LPD preoperative through imaging. The intraoperativefrozen section examination may help with the diagnosis, but the final diagnosis still relies on pathological examination.

Although the specific pathogenesis for the LPD is not clear, it is now accepted that it may come from iatrogenic fibroids parasitic [4]. As it is known, during laparoscopic myomectomy, the resection of myoma is shattered into small piecesby pulverize firstly and then the debris tissues are removed by manipulating hole. Small fragments of fibroid tissue in the process of rotary cutting may be dispersed and left throughout the abdominal cavity, and then get blood oxygen supply and growth through the formation of new blood vessels between adjacent tissues. At present, several studies had suggested that LPD after laparoscopic myomectomy was associated with myoma morcellation [5]–[7].In our case, the patient had a history of laparoscopic uterine fibroids eliminate. In combination with patient's history, intraoperative situation and pathological diagnosis, we highlighted that the development of LPD was associated with myoma morcellation during procedure. Therefore, we should check the various parts of the basin of the abdominal cavity carefully as far as possible, remove residual fragments of the myoma and give a large amount of abdominal cavity flushing again and again; these methods should be able to play a role of prevention of this complication. In addition, the morcellation of fibroids or uterus in a sealed bag may also have the potential to prevent the rare morcellation-related complications [8]. At the same time, some scholars suggested that after laparoscopic surgery recurrent abdominal pain or unidentified pelvic mass, iatrogenic fibroids, cultivation of ectopic foci should be vigilant the occurrence of LPD.

In summary, LPD is a rare disease and is difficult to be diagnosed. The etiology of LPD is still unknown. The use of laparoscopic power morcellation, which duo to fragments of the myoma tissue, may be a cause of the development of LPD. However, our case suggested that in order to prevent the development of LPD, the morcellated fragments of the myoma tissue should be removed clearly and be not left in the abdominal cavity during procedure.

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