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Case Report

Case study of disseminated leiomyomatosis with urological and colorectal involvement and a literature review

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ABSTRACT

Disseminated peritoneal leiomyomatosis (DPL), also known as diffuse peritoneal leiomyomatosis, is a rare disease characterized by a sub peritoneal proliferation of benign nodules, mainly composed of benign smooth muscle cells, macroscopically mimicking peritoneal carcinosis. We report the case of a 60-year-old patient, with hypertension under treatment, being followed for the management of pauci-symptomatic abdominopelvic masses, evolving in a context of conservation of the general state, who underwent hysterectomy with adnexectomy by laparotomy for uterine myoma. Imaging revealed three masses, extending from the perigastric to the pelvic region, with areas of necrosis, moderate peritoneal effusion and lumbo-aortic and primitive iliac lymph nodes with small infracentimetric axes. The management consisted of a complete cytoreduction. Postoperative management was straightforward. LPD is a rare condition, with around 200 cases published in the literature. Its incidence is estimated at around 1/10,000,000, given the generally asymptomatic nature of the disease. The etiopathogenesis of this condition remains poorly elucidated, although several causal theories have been described in the literature, including hormonal, iatrogenic and congenital or hereditary. The management of LPD is not currently standardized, but surgery remains the gold standard. LPD is a rare, benign condition characterized by sub peritoneal proliferation of smooth muscle cells. It occurs most frequently in women of childbearing age, but can also occur in postmenopausal women and men. Treatments for LPD are not standardized, and further studies are required in the near future.

Keywords: Disseminated peritoneal leiomyomatosis, Diagnosis, Case report, Incidence, Surgery, Pathophysiology, Management

INTRODUCTION

Disseminated peritoneal leiomyomatosis (LPD), also known as diffuse peritoneal leiomyomatosis, is a rare disease characterized by a sub peritoneal proliferation of benign nodules composed mainly of benign smooth muscle cells, macroscopically mimicking peritoneal carcinosis.[1] LPD was initially described in 1952 by Willson and Peale and identified as a pathological entity in 1965 by Taubert et al.[2,3] The pathophysiology of LPD is currently poorly understood, and several pathogenic factors have been reported in the literature. The disease usually occurs in women of childbearing age, but can also be encountered in post-menopausal women and men.[4,5]

Preoperative diagnosis of LPD remains a difficult problem, leading to the mistaken belief that the diagnosis is GIST, and the final diagnosis is usually rectified by anatomopathological on the resection specimen.[4] In this

study, we report the case of a patient presenting with large intra-abdominal masses.

CASE REPORT

Patient aged of 60 years, hypertensive on betablocker and Natrixam, operated by laparotomy for uterine fibroid twice (2014 and 2017) at the gynecology department of the Hôpital Militaire d'Instruction Mohamed V in Rabat, having benefited a hysterectomy with adnexectomy. The history of the disease goes back to one year which was marked by the appearance of abdominal distension evolving in a context of conservation of the general state without other associated signs. Clinical examination revealed a palpable mass in the hypogastrium, poorly limited, with no associated adenopathy.

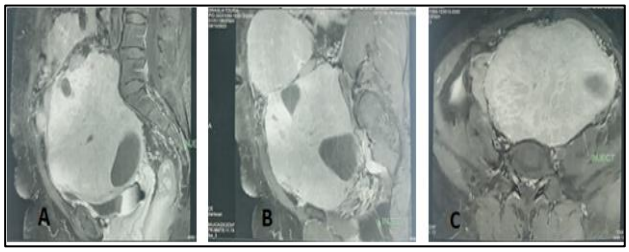

Figure 1: abdominal MRI showing tissue-dense abdominopelvic masses in hypersignal with areas of necrosis; (A) cross-section (B and C) sagittal section.

An abdominopelvic CT scan was performed and found peritoneal and abdominopelvic masses extending from the perigastric region to the pelvic level, with bumpy contours, heterogeneous tissue density and areas of necrosis, enhancing after injection of PDC. Their dimensions were mass number 115×144×144 mm, mass number, 114×9×2×85 mm and mass number 144×153×146 mm. Imaging also revealed a moderately large intra-peritoneal effusion and millimetric primitive lumbo-aortic and iliac lymph node formations.

A complementary abdominopelvic MRI was performed, showing voluminous intraperitoneal masses of abdominopelvic topography, with a bosselated outline, presenting in hyper-signal, strongly enhancing after injection of paramagnetic contrast medium, with the presence of areas of necrosis and measuring respectively: 153×144×151 mm, 132×117×108 mm and 101×79×73 mm in diameter, associated with a moderate intraperitoneal effusion (Figure 1). After preoperative preparation, the patient underwent complete cytoreduction by median laparotomy.

Intraoperative exploration after adhesiolysis showed three masses (01 pelvic mass measuring 20×30 cm in contact with the bladder dome, 02 masses in the left lower abdomen measuring 20×20 and 15×15 cm in intimate contact with the sigmoid colon, proximal bowel and mesentery), and moderate ascites. A sigmoid resection with end-to-end anastomosis and resection and closure of the bladder dome with two hemi-surjects. Drainage was ensured by two thoracic-type drains, one at the level of the left parieto-colic gutter and the other in contact with the bladder (Figure 2). Postoperative recovery was straightforward, with discharge on day 8.

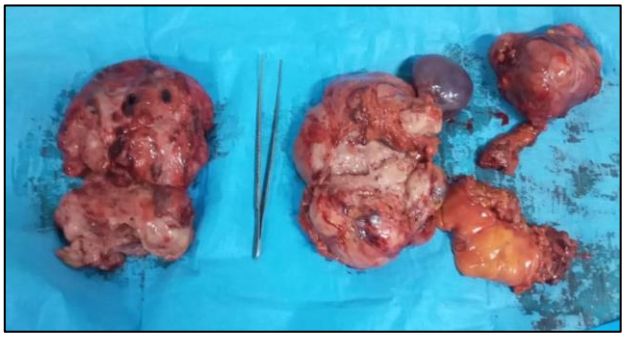

Figure 2: Surgical specimen showing 03 irregular 15 cm long masses invading the sigmoid and bladder dome.

DISCUSSION

Several etiopathogenic theories have been put forward in the literature, including iatrogenic, hormonal and hereditary. The main pathophysiological hypothesis is peritoneal implantation of uterine smooth muscle cells following surgical treatment of uterine myomas.[6] Approximately 200 cases have been published in the literature since its first description in 1952 by Wilson, but the true prevalence of the disease remains unclear.[7-9] After, myomectomy, the occurrence of disseminated peritoneal leiomyomatosis remains rare, estimated at around 0.1% in the series by Sizzi et al.[10]

Its incidence is 1/10,000,000, given the asymptomatic nature of the disease, therefore LPD remains underdiagnosed.[8,9] Although the disease occurs in women of childbearing age, cases of LPD have been described in postmenopausal women, as well as in men.[5,11,12] The etiology of LPD remains poorly elucidated, but there is a correlation between the occurrence of LPD and exposure to high levels of estrogenic hormones, whether endogenous or exogenous (contraceptive pill), as well as a history of uterine myoma surgery (myomectomy, hysterectomy) and endometriosis.[12-14]

Laparoscopic hysterectomy surgery with tumor morcellation can potentially increase implantation and dissemination of tumor cells intraperitoneally.[15,16] Cases of autosomal dominant family grouping with variable penetrance have recently been described in the literature.[12] Diagnosis of LPD is a challenge, as the symptoms are not very specific. The majority of patients are pauci or asymptomatic, in which case they include abdominal pain or discomfort, abdominal distension or even intestinal obstruction.[1,12] Cases of LPD associated with endometriosis and ascites have been reported in the literature, and cases of LPD associated with ascites and

adenopathies have also been described. In our case, sub-centimetric lymph node formations at iliac and lombo-aortic level were identified.[17,18] Preoperative detection of LPD using imaging procedures such as abdominal ultrasound, CT or MRI can be beneficial, but they are of little help in the differential diagnosis of malignancies. Therefore, preoperative biopsy and histopathology analysis are essential for the diagnosis of LPD.[1] Microscopically, LPD is generally characterized by smooth muscle cells in interdigitated fascicles with or without mitotic figures (19), and they are devoid of nuclear polymorphism, hyperchromasia, cellular necrosis and cellular atypia unlike leiomyosarcoma.[1,13] Intraoperatively, LPD presents as multiple round nodules, ranging in size from several millimeters to several centimeters, that can be detected on any peritoneal or omental surface of the abdominal cavity, small or large intestine, mesentery and retroperitoneum.[12,14]

Prior to initiating a therapeutic approach, imaging should be performed to define the size of the lesion and look for possible malignancy.[7] LPD imaging findings include several complex, firm, solid peritoneal lesions that resemble typical peritoneal lesions.[6] Contrast-enhanced CT shows firm, hypo-dense, well-defined nodules with significant, uniform enlargement, while the surrounding peritoneum is usually normal. However, in cases of necrosis, degeneration or endometrial implantation, enhancement is heterogeneous, simulating peritoneal carcinosis.[20,21] On magnetic resonance imaging, they appear as well-defined solid masses, hypointense on T1-weighted images, hypointense, isointense or hyperintense on T2-weighted images depending on the water content of the tumor and consistent, uniform enhancement on dynamic contrast enhancement images. LPD with malignant conversion shows necrosis and bleeding in the tumor masses.[22]

When LPD or uterine leimyosarcoma is accompanied by hyperintense T2-weighted MRI images and diffusion restriction, an unusual histological type of leiomyoma with high cellularity should be sought.[23] In our case, tissue-dense hypersignal T2 masses with areas of necrosis were described on abdominal-pelvic MRI (Figure 1). At the moment, there are no standard guidelines for the management of DPL, although surgery remains the mainstay of treatment. However, due to its minimally invasive advantage, laparoscopy should be the first choice for surgical diagnosis and treatment.[4] Recently, it has been proposed that treatment should be individualized according to the patient's age, reproductive needs and comorbidities.[14]

For patients with fertility needs, focal resection is feasible, and gonadotropin agonists or aromatase inhibitors are used postoperatively. Indeed, for women with a desire to procreate, hormone therapy is probably the best approach, but if surgery is necessary, it should consist of mass removal and omentectomy followed by hormone replacement therapy.[24,25] In women without childbearing potential or postmenopausal women, a more extensive surgical approach with total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy and tumour volume reduction may be the best alternative.[26] In our case, we opted for surgical treatment with complete cytoreduction, and found no need to supplement with hormonal therapy. Although LPD is a rare benign disease with a good prognosis, degeneration of LPD into a malignant tumor has been reported in the literature. In addition, the simultaneous identification of LPD and leiomyosarcoma has been reported, although it is unclear whether malignancy has occurred due to transformation of LPD tumours.[27,28]

CONCLUSION

Disseminated peritoneal leiomyomatosis (DPL) is an uncommon condition characterized by a sub peritoneal proliferation of benign nodules, mainly composed of benign smooth muscle cells, macroscopically mimicking peritoneal carcinosis. The pathophysiology remains poorly understood at present, and several pathogenic factors have been reported in the literature. The disease usually affects women of childbearing age, but can also be seen in post-menopausal women and men. The preoperative diagnosis of LPD remains a difficult embarrassment for the practitioner, the final diagnosis being rectified by anatomopathology on the resection specimen. The treatment of LPD is not standardized, and further studies are needed in the near future.

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