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CLINICAL CASES

Disseminated peritoneal leiomyomatosis. A rare disease with a difficult diagnosis

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Abstract

Background: *Disseminated peritoneal leiomyomatosis is a rare, benign disease characterized by the proliferation of multiple peritoneal and subperitoneal nodules consisting of smooth muscle cells. Diagnosis is difficult due to its similarity to peritoneal carcinomatosis. less There are 200 cases reported in the world literature.* **Clinical case:** *A 29-year-old female with a history of abdominal hysterectomy due to myomatosis 4 years ago, came due to abdominal pain, multiple solid images of different sizes were observed in the tomography, she underwent elective surgery, histological analysis confirmed benign smooth muscle tumors. We present a case operated on in our center.*

Keywords: *Leiomyomatosis peritoneal disseminated. Leiomyoma. Surgery.*

Introduction

Leiomyomatosis peritoneal disseminated (LPD) is a rare, benign disease characterized by the proliferation of multiple peritoneal and subperitoneal nodules consisting of smooth muscle cells[1]. The differential diagnosis of LPD is challenging due to its clinical similarity to peritoneal carcinomatosis or metastatic lesions. and its histological similarity with benign metastatic leiomyoma[2]. The first reported case in the literature was published in 1952 by Wilson and Peale[3]. There are less than 200 cases reported in the world literature, given its rarity there are no established guidelines for its treatment[4]. Some risk factors have been identified for LPD, use of oral contraceptives, metaplasia, genetics, pregnancy, iatrogenic and surgical manipulation[5]. We present a case of LPD intervened in our center.

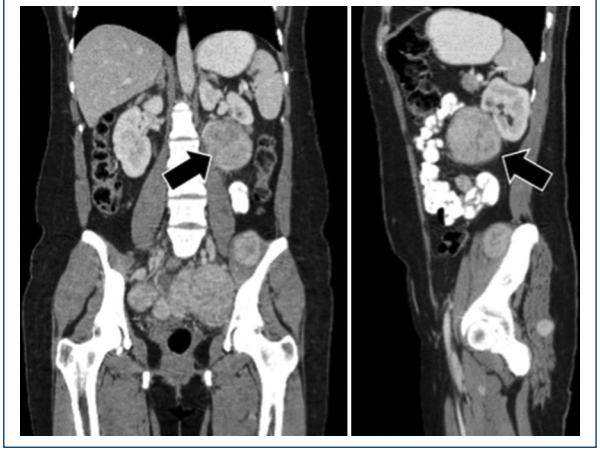

Figure 1. Computed tomography: heterogeneous mass with well-defined borders that displaces the left kidney.

Clinical case

A 29-year-old female with a history of cesarean section and abdominal hysterectomy for uterine myomatosis 4 years ago. He came due to presenting abdominal pain of moderate intensity, located in the hypogastrium with irradiation towards the left pelvic limb, which increased with the passing of days. Ultrasound is performed where multiple images of solid

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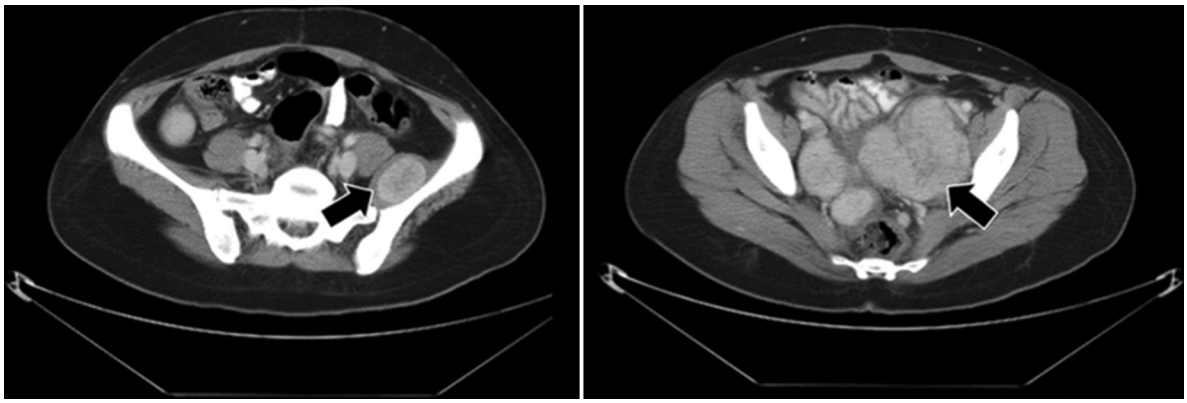

Figure 2. Computed tomography: multiple pelvic heterogeneous nodules.

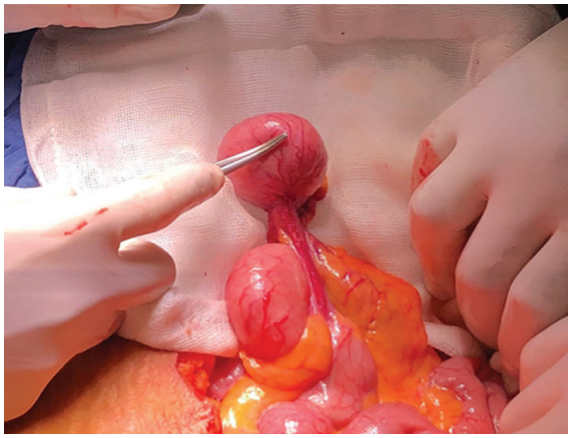

Figure 3. Intraoperative image showing representative pedunculated nodular formation.

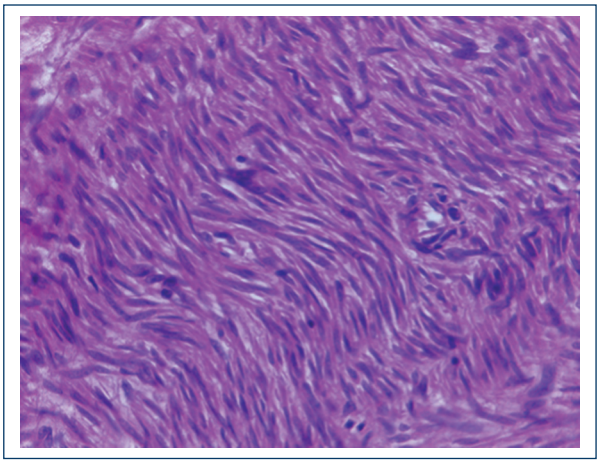

Figure 4. View at 40X, spindle nuclei, fine chromatin, no mitosis, no atypia.

characteristics in the pelvic cavity are identified. In the simple and contrasted abdominopelvic tomography, a left retroperitoneal image of 6.4 x 7.4 x 7.2 cm with a volume of 178.3 cc was observed. displacing the kidney on the same side (Fig. 1), multiple solid images (approximately ten) of different sizes were found in the pelvis, the largest being 7.4 x 8.0 x 7.6 cm with intense enhancement with contrast medium (Fig. 2). The patient underwent elective surgery, during surgery multiple peritoneal and retroperitoneal nodules were found in the right obturator fossa, left iliac muscle, left renal pole, pelvic cavity and right paracolic frame (Fig. 3). They were completely resected carefully by the surgeons, hemostasis was performed with electrocoagulation and sutures (Vicryl 2-0), with satisfactory postoperative evolution.

Histological analysis of the surgical specimens confirmed the diagnosis. The histological structure of the tissue consisted of benign smooth muscle cell tumors, without mitotic figures or cellular atypia, without necrosis (Fig. 4). Immunohistochemistry: overexpression of estrogen (Fig. 5) and progesterone (Fig. 6) receptors. Follow-up was continued for 1 year, being asymptomatic and without evidence of ultrasound disease.

Discussion

LPD is an extremely rare clinical condition. Two main theories of the etiology and pathophysiology of LPD have been reported: a hormonal theory with mesenchymal stem cell metaplasia and an iatrogenic origin after surgery. According to the hormonal theory, LPD is supposed to result from the metaplastic change of mesenchymal stem cells with exposure to high levels of female

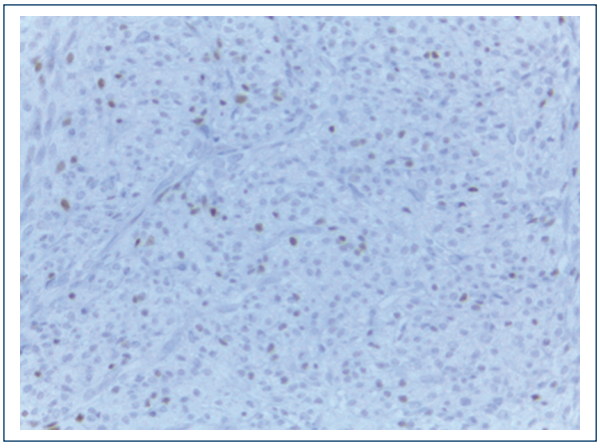

Figure 5. Immunohistochemistry, estrogen receptors with nuclear intensity 1.

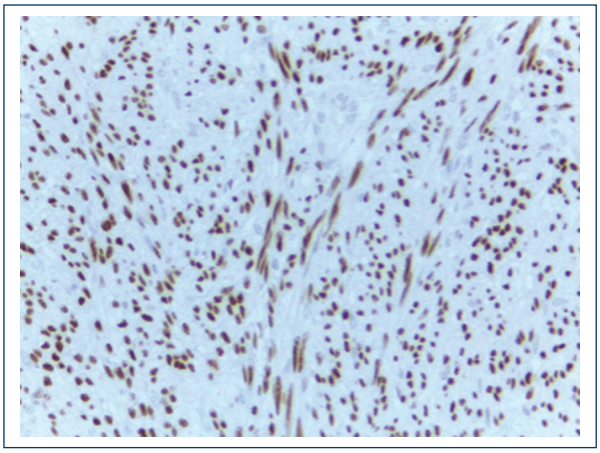

Figure 6. Immunohistochemistry, nuclear intensity 2 progesterone receptors.

steroids[6]. Recent publications have highlighted a link between surgical history such as hysterectomy for myomatosis or the laparoscopic uterine fibroid fragmentation technique and the development of LPD, due to the potential for tumor implantation and dissemination[7]. More specifically, LPD can present years after myomectomy or hysterectomy[8]. In our case, the history of hysterectomy due to myomatosis without adequate containment systems could have played a role in the pathogenesis of LPD.

The preoperative diagnosis of LPD can be challenging due to its clinical manifestations and nonspecific radiological features. Most patients with LPD remain asymptomatic. Symptoms, if any, are nonspecific and include abdominal pain and discomfort, bloating, or abdominal masses that may lead to intestinal obstruction[9]. In the case presented, the patient presented abdominal pain due to compression.

Ultrasound, computerized axial tomography and nuclear magnetic resonance have been described as useful elements in preoperative diagnosis. The typical images are solid nodules with regular contours and variable size scattered on the peritoneal surfaces, which can be confused with peritoneal carcinomatosis or gastrointestinal tumors due to similar image characteristics[10]. The definitive diagnosis is histological, and confirms in our case that the nodules are formed mainly by muscle fibers and fibroblasts, without atypia or necrosis. The differential diagnosis includes leiomyosarcoma, mesothelioma, tuberculosis, lymphoma and peritoneal carcinomatosis[11]. Intraoperatively, LPD presents as multiple round nodules, ranging in size from several millimeters to centimeters, and can be detected on any peritoneal surface or omentum in the abdominal cavity, small or large intestine, mesentery, and retroperitoneum[12].

There are few data on the most appropriate treatment for LPD. Recently, determining therapy according to the patient's age, symptoms, and desire to have children has been proposed. For women with reproductive desire, hormone therapy with gonadotropin-releasing hormone injection, aromatase inhibitor, or selective progesterone receptor modulator is usually the first-line treatment option. This approach is also preferred in the prevention of postoperative recurrence[13-14]. For women without reproductive desire, the best alternative may be a more extensive surgical procedure with total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, myomectomy, and excision of the nodes[15].

In this case, given the size of the nodules, the exacerbation of the symptoms and the patient's clinical worsening, it was decided to perform surgical treatment.

Conclusion

LPD is a rare clinical condition, it is mainly associated with a history of minimally invasive uterine myomectomy or hysterectomy for myomatosis. The LPD should prefer the differential diagnoses of women with disseminated intra-abdominal or pelvic tumors, especially those with a history of gynecologic surgery. The case presented is of interest because it represents an infrequent entity that can unequivocally simulate peritoneal carcinomatosis. Surgery remains the main therapeutic weapon in symptomatic cases. Surgeons' knowledge

of this rare condition is essential to establish a correct diagnosis and ensure proper treatment.

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Conflicts of interest

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Ethical disclosures

Protection of people and animals. The authors declare that no experiments have been performed on humans or animals for this research.

Data confidentiality. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors declare that no patient data appear in this article.

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