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Case Report

Laparoscopic resection of giant endometriotic cysts with mesenteric implantation

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ABSTRACT

Endometriosis is defined as the presence of tissue which is histologically similar to the endometrium in locations outside the uterus. It affects women of reproductive age mainly and represents one of the main causes for hysterectomy and infertility amongst women. It has a broad spectrum of symptoms which make for a challenging diagnosis. Extragenital endometriosis affects up to 37% of all patients, and intestinal endometriosis has been observed in up to 12% of women affected by the disease, mainly involving the recto-sigmoid colon, ileocecal region and cecal appendix. Intestinal symptoms such as changes in depositional rhythm, diarrhoea and constipation are frequent and can evolve to acute abdominal obstruction in advanced stages of the disease. Authors present the case of a 51-year-old female that presented to the emergency room with abdominal, colic type, diffuse pain in the left flank, early satiety and postprandial fullness, CT scan revealed the presence of a lobed and septate mesenteric cystic tumor, of approximately 15.5 cm in diameter. Serum oncological markers were found to be within normal parameters. The patient underwent laparoscopic resection of the tumor with trans-operatory study, which ruled malignancy out and confirmed the presence of endometriotic tissue. The patient was discharged 72 hrs after surgery and prescribed anastrozole 1 mg orally every 24 hours. Follow up with Abdominal CT scan was performed 6 and 18 months later, showing no evidence of recurrence; the patient remains asymptomatic 18 months after surgery.

Keywords: Endometriosis, Giant endometriotic cyst, Laparoscopic resection, Mesentery, Mesenteric cystic tumor

INTRODUCTION

Endometriosis was initially described by Rokitansky in 1860 and is defined as the presence of tissue histologically similar to the endometrium in locations outside the uterus, a process that results in a chronic inflammatory reaction. It has a reported prevalence of 8-10% in women of reproductive age, 5-10% in women with chronic pelvic pain and 20-40% in patients with a history of infertility; this condition represents one of the main causes of hysterectomy and public health costs of up to 69.4 billion dollars have been reported in the United States.[1] Symptomatology is usually nonspecific with presence of pelvic pain and dyspareunia in patients of reproductive age being its most common symptom.

It has not been possible to establish an exact theory that unifies the origin of endometriosis, nevertheless, multiple theories have been proposed on the pathogenesis of the disease and can be grouped in general terms as those theories that propose that endometriosis implants originate from uterine endometrium and those that pose the possibility that these implants may come from tissues other than the uterus.[2] The presence of extragenital

endometriosis implants has been reported in 3 to 37% of women affected by this disease; intestinal implantation has been observed in 3 to 12% of the affected patients. The main site of intestinal involvement is the recto-sigmoid colon, observed in 70 to 93% of the cases among several authors. This extragenital location is followed by the ileocecal region, cecal appendix, and other segments of the colon and small intestine in order of frequency.[3] Symptomatology in patients with involvement of the digestive tract is usually the same as that observed in patients with genital endometriosis, however, specific symptoms of the digestive tract are observed, such as rectal pain with perineal irradiation, changes in depositional rhythm, diarrhea and constipation, rectal bleeding which coincides with menstruation, abdominal distension and acute intestinal obstruction in advanced stages.[4]

A multidisciplinary approach, involving surgical and medical management, should be proposed to the patients that present this pathology; the strategy to follow will depend on the patient's clinical presentation and fertility desires. Medical treatment is reserved for cases of satisfied maternity and when there are no serious complications secondary to the disease. In the presence of severe symptomatology, such as intestinal occlusion or ureteral stenosis, the laparoscopic approach is the diagnostic and therapeutic gold standard.[5]

CASE REPORT

Authors present the case of a 51 year old female patient, with no prior history of chronic degenerative diseases, who underwent instrumented uterine curettage and laparoscopic conservative hysterectomy 22 and 3 years prior to admission respectively, who referred abdominal, colic type, diffuse pain predominantly in the left flank, without irradiation, which was sometimes described as being 10/10 in intensity with spontaneous remission, as well as early satiety, postprandial fullness and generalized distension, which was managed by the patient with butylhioscine and paracetamol upon ocurrence. On admission, the patient reported a noticeable increase in abdominal circumference of approximately 1 year, which presented gradually without referring an increase in usual caloric intake.

On admission to the emergency department, the patient presented vital signs within normality, being conscious, alert, neurologically intact, with discomfort related to dorsal decubitus. Abdominal examination highlighted the presence of a globose abdomen, tense, painful upon deep palpation, with no evidence of peritoneal irritation at the time of admittance, a mass was palpated predominantly in mid-abdominal quadrants, fixed to deep planes, with generalized tympanic sounds in the entirety of the abdomen, with discretely diminished peristalsis, no other abnormal findings were reported on physical examination. Admission laboratory testing showed a hemoglobin of 13 g/ dl, with leukocytes reported at 7.2 mil/ mm^3, neutrophil bands of 5%, glucose of 83, without evidence of biochemical alterations in blood count or blood chemistry. The emergency service requested assessment by the medical gastroenterology service, who in turn requested interconsultation from the general surgery service, requesting complementary imaging studies.

A simple abdominal radiography reported the presence of segmental ileus predominantly in the right iliac fossa, as well as the presence of a radiopaque, ovoid, homogeneous image predominantly occupying medial abdomen and left flank, with edges showing calcium-density, without any other pathological findings. An ultrasound showed the presence of multiple anechoic, round, avascular images, with irregular walls and thick septa ranging from epigastrium to hypogastrium, reaching up to 12.5 cm in its longitudinal axis.

A multi-slice helical CT scan with endovenous contrast was requested due given the ultrasonographic findings; this study reported the presence of a mesenteric cystic tumor, which compressed the left hepatic lobe and was described as being a lobed and septated mass of approximately 15.5 cm in diameter, with calcification of the inferior wall, which extended downwards and displaced the transverse colon in the same direction, as well as the duodenum medially, which was compatible with a probable mesenteric cyst. A panel of oncological markers (carcinoembryonic antigen, 29- 9 carbon antigen and alpha fetus protein) were requested, which were found within normal parameters.

Authors proposed initial laparoscopic management and commented upon the possibility of converting to open surgical management upon transoperative findings. Umbilical access was performed with an open technique and placement of a 12 mm Hasson trocar at the level of the umbilical scar, and capnoperitoneum was insufflated, keeping low intra-abdominal pressures (10mmHg) to avoid hemodynamic alterations secondary to the presence of the cystic mass.

Exploratory laparoscopy was performed using 10 mm 30° lens, demonstrating the presence of a large multilobed mass with root implantation in transverse mesocolon, with displacement of small bowel loops and adhesion to large vessels, pylorus, duodenum and transverse colon, as well as multiple adhesions between bowel loops and tumour, with extension of the mass from the free edge of the liver to the pelvic cavity.

After initial inspection, placement of 5 mm trocars in the anterior axillary line at the level of the right flank and right iliac fossa is carried out; careful lysis of adhesions and separation of the tumor capsule from the loops of the colon and duodenum with cold, sharp dissection in upper, middle and lower left quadrants was then performed. Authors decided to place a 10-mm trocar in the left iliac fossa to improve exposure and manipulation of the tumor,

continuing release of small bowel loops in the midline and the hepatic colic angle.

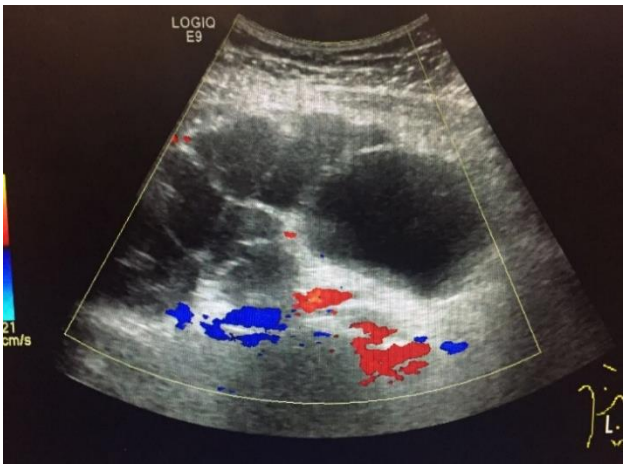

Figure 1: Doppler ultrasound showing multiple anechoic, round, avascular images, with irregular walls and thick septa ranging from epigastrium to hypogastrium, reaching up to 12.5 cm.

The tumor dome was cut using a harmonic scalpel, obtaining approximately 2300 cc of ocher colored, heterogeneous in consistency, old hematic fluid which was carefully aspirated without contamination of the abdominal cavity and then sent together with a fragment of the capsule for trans-operative histopathological study; once the tumor dome was approached, it was dissected throughout its circumference using a harmonic scalpel and sharp dissection, observing the presence of an internal tumor capsule with multiple cystic lesions in caudal direction, with adherence of the posterior wall of said lesions to vena cava, aorta and bile duct.

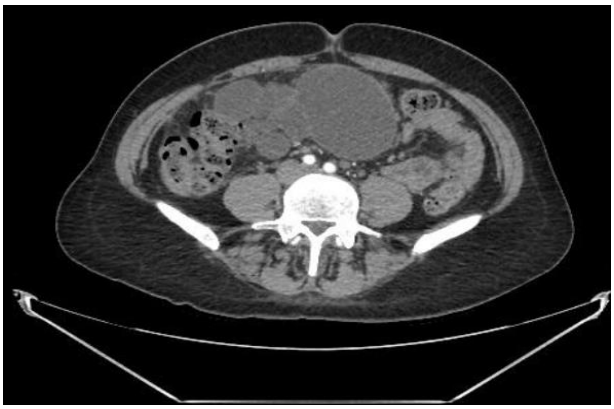

Figure 2: Abdominal CT scan showing the presence of a multilobed mesenteric cystic tumor (endometriotic implants), of approximately 15.5 cm in diameter (axial plane).

Once malignancy was ruled out through trans- operative histopathological diagnosis and probable endometriosis was diagnosed, authors decided to continue careful dissection of this new segment of endometriotic implants, without resecting posterior wall of the lesion due to its intimate relationship to structures with a high risk of incidental lesion (large vessels, bile duct, duodenum). Once the anterior capsular resection was completed, it was retrieved using 4 laparoscopic bags (endobag) through the 10 mm incision in the left iliac fossa, the tissue obtained was sent for definitive histopathological study, confirming histopathological diagnosis of "cystic endometriosis, without evidence of malignancy in the tissue analyzed".

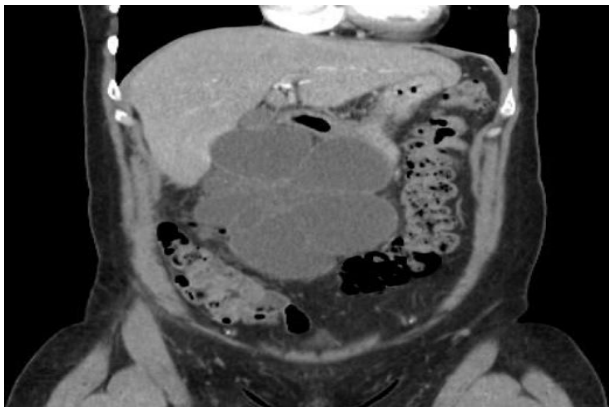

Figure 3: Abdominal CT scan showing the presence of a multilobed mesenteric cystic tumor (endometriotic implants), of approximately 15.5 cm in diameter (coronal plane).

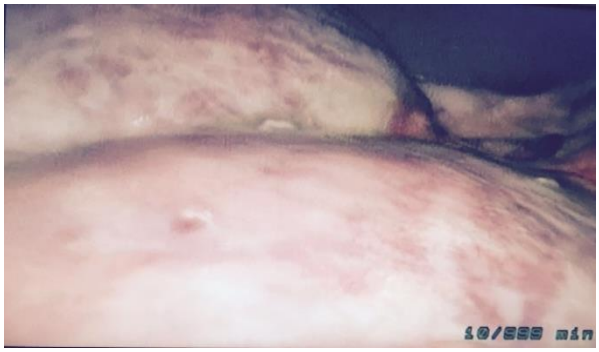

Figure 4: Laparoscopic view of giant endometriotic cysts with mesenteric implantation.

Figure 5: Follow-up abdominal CT Scan 6 months after surgery demonstrating no evidence of endometriotic implants.

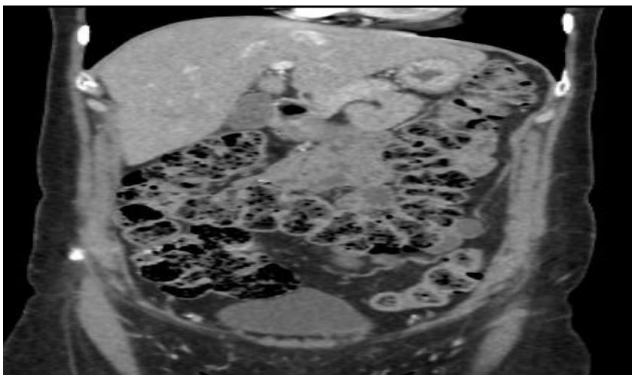

Figure 6: Follow-up abdominal CT Scan 6 months after surgery demonstrating no evidence of endometriotic implants (coronal plane).

Figure 7: Follow-up abdominal CT Scan 18 months after surgery demonstrating no evidence of endometriotic implants.

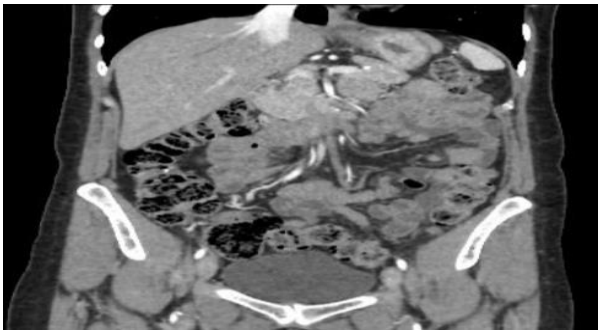

Figure 8: Follow-up abdominal CT Scan 18 months after surgery demonstrating no evidence of endometriotic implants (coronal plane).

The patient had an adequate postoperative evolution and was offered liquid diet within 36 hours of the surgical procedure, progressing the diet that same day and being discharged 72 hours after surgery, with adequate toleration to feeding, ambulation and hemodynamic stability.

The patient underwent postoperative medical management with anastrozole 1 mg orally every 24 hours. An abdominal multi-slice CT scan was requested for postoperative control 6 months and 18 months after the procedure, without evidence of recurrence of the tumor; the patient remains asymptomatic, without an increase in the abdominal perimeter, abdominal distension or pain 18 months after surgery.

DISCUSSION

Endometriosis is considered a chronic disease whose diagnosis is complex and, in most cases, late. There is a broad spectrum in the clinical presentation of the disease, with predominantly nonspecific symptoms and high complexity in the diagnosis by imaging studies, however, a history of chronic pelvic pain, dysmenorrhea, dyspareunia and infertility in women of reproductive age, should increase clinical suspicion. Diagnostic delay of up to 8-10 years on average has been reported; laparoscopy with biopsy and histopathological analysis of the lesions, evidencing the presence of ectopic endometrial glands and stroma in the sample is currently considered to be the gold standard in diagnosis. The most common locations of the disease are the rectosigmoid area (50-90%), the rectovaginal septum (13%), small intestine (7%) cecal appendix (3-18%) blind (4%) and Meckel's diverticulum (with 2 cases reported in the current literature); no reports have been found in the literature on the mesenteric implantation of endometriotic cysts in current literature. At the level of the small bowel, the terminal ileum tends to be more prone to lesions due to regional proximity to the fallopian tube and the ovary.[6]

After being described by Carl von Rokitansky's, Thomas Stephen Cullen and Johannes Pfannenstiel, classified endometriosis according to its anatomical location; Acosta et al. presented a modification to this classification including a distinction between internal, external and extragenital endometriosis. In 1979, the American Fertility Society (AFS) changed some aspects of this classification, thus creating the AFS score, which was modified again in 1985 and finally in 1997, giving way to the American Society for Reproductive Medicine revised score in 1997. While the revised classification of AFS continues to be the most commonly used, it does not take into account deeply infiltrating endometriosis or involvement of other organs, so the ENZIAN classification was created, same that acts as a supplement to the classification of the AFS. This score takes into account the involvement of retroperitoneal structures, other organs and deeply infiltrating endometriosis, making mention of the involved organ. This classification also divides affectation according to three compartments and the severity thereof according to the size of the lesion described. The combination of both classifications has proven very useful in the classification and description of extragenital endometriosis.[7,8]

The course of action in the treatment of endometriosis depends on different factors which need be taken into account, being the presence of serious complications (such as bowel perforation, ureteral stenosis and bowel

occlusion) the main indication for surgical approach of the disease, followed by magnitude of the symptomatology and impact on the quality of life reported by the patient. Among the available medical treatment options are the hormonal treatment schemes, which decrease estrogen synthesis, decrease bleeding and cause a state of atrophy of endometriosis implants; gonadotropin-releasing hormone (GnRH) receptor antagonists, cause a down-regulation of the release of follicle-stimulating hormone (FSH) and luteinizing hormone (LH), thus suppressing ovulation; Aromatase inhibitors, such as letrozole, anastrozole and exemestane, decrease conversion of androstenedione and testosterone to estrone and estradiol, respectively, by suppressing the aromatase enzyme. Selective progesterone receptor modulators (SPRM), which improve the symptomatology by suppressing ovulation as well as endometrial proliferation. Selective estrogen receptor modulators (SERM) such as raloxifene are in clinical trials to determine their efficacy in the treatment of endometriosis by binding to estrogen receptors and their anti-estrogen or estrogen-dependent tissue activity. There is also non-hormonal medical treatment, such as dopamine receptor 2 agonists, which inhibit angiogenesis and decrease the size of lesions, or statins, which have demonstrated inhibition of angiogenesis mechanisms and cell proliferation observed in the development of tissue similar to endometrium. There are also other naturally occurring substances such as resveratrol, which have protective and therapeutic effects against endometriosis and which lead to a decrease in inflammatory response, apoptosis, oxidative stress, angiogenesis and inhibition of adhesion and invasion of endometrial lesions.[9-12]

The main surgical treatment modalities are ablation and excision; The surgical management of deeply infiltrating endometriosis (DIE) has proved to be a unique challenge due to the structures that are normally involved, such as the retrocervical region, the urinary tract, the rectum, the vagina, among others. Excision treatment of DIE improves pain symptoms and reduces the recurrence of severe symptoms (bleeding, intestinal occlusion). Estimated recurrence of an endometrioma usually varies between 6% and 17% after surgery; hysterectomy in conjunction with excision of endometrial implants laparoscopically using an early discharge protocol (fast track) is the optimal treatment for pain and severe symptomatology of the disease, with the lowest rates of reoperation being observed in those patients who also underwent a bilateral oophorectomy.[13-15]

CONCLUSION

Endometriosis is a common disease in women of reproductive age, being one of the main causes of infertility in this group worldwide, with pain being the main symptom reported. Therapeutic approach of the disease is dependent on the depth of the infiltration of endometrial implants and the symptomatology that the patient refers to. In case of deeply infiltrating endometriosis, interdisciplinary management with hysterectomy, bilateral oophorectomy and laparoscopic excision of endometrial implants with postoperative hormonal suppression has been shown to reduce the pain associated with the disease as well as the recurrence rate, so it should be considered in patients whose symptoms alter the quality of life or which manifest serious extragenital complications.

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