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Extrauterine leiomyomata presenting with sepsis requiring hemicolectomy

Leiomiomas extrauterinos com sepse que exigem hemicolectomia

Relato de Caso

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Abstract

Extrauterine leiomyomas are rare, benign, and may arise in any anatomic sites. Their unusual growth pattern may even mimic malignancy and can result in a clinical dilemma. Occasionally, uterine leiomyomas become adherent to surrounding structures. They also develop an auxiliary blood supply, and lose their original attachment to the uterus, thus becoming 'parasitic'. Parasitic myomas may also be iatrogenically created after uterine fibroid surgery, particularly if morcellation is used. This report presented two cases of parasitic myomas with sepsis, both requiring right hemicolectomy. It reviewed the pertinent literature.

Resumo

Leiomiomas extrauterinos são raros e benignos e podem surgir em qualquer local anatômico. O padrão de crescimento não-usual destas lesões pode até mesmo imitar malignidade e resultar em dilema clínico. Ocasionalmente, os leiomiomas uterinos aderem-se às estruturas circunvizinhas, desenvolvem suprimento sanguíneo auxiliar e perdem sua ligação original ao útero, tornando-se assim "parasíticos". Os miomas parasíticos podem também ser criados iatrogenicamente após cirurgia fibroide uterina, especialmente se for utilizada morcelação. Este relato apresentou dois casos de miomas parasíticos com sepse, ambos exigindo hemicolectomia à direita, bem como revisão da literatura pertinente.

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Introduction

Uterine leiomyomas affect 20 to 30% of women older than 35 years, and their incidence may be on the rise since many women are postponing their reproductive careers¹. Extrauterine leiomyomas are rare, benign, and may arise in any anatomic sites. Their unusual growth pattern may even mimic malignancy and can result in a clinical dilemma². Occasionally, uterine leiomyomas become adherent to surrounding structures. They also develop an auxiliary blood supply, and lose their original attachment to the uterus, thus becoming 'parasitic.' Parasitic myomas may also be iatrogenically created after uterine fibroid surgery, particularly if morcellation is used. With the surge in laparoscopic gynecological procedures, especially laparoscopic myomectomy, the potential for developing parasitic myomas is being recognized more frequently^{3,4}. We presented two cases of parasitic myomas with sepsis, both requiring right hemicolectomy. The pertinent literature was also reviewed.

Case report 1

A 28-year-old female presented to the gynecologist at a peripheral hospital with a history of miscarriage two weeks prior to the presentation. She complained of feeling unwell, with fever, and generalized abdominal pain with nausea. On clinical examination, the abdomen was tender with a palpable mass on the right side. An ultrasound scan of her abdomen and pelvis demonstrated a 17×12 cm right sided complex abdominal mass, which was reported to be possibly of ovarian origin.

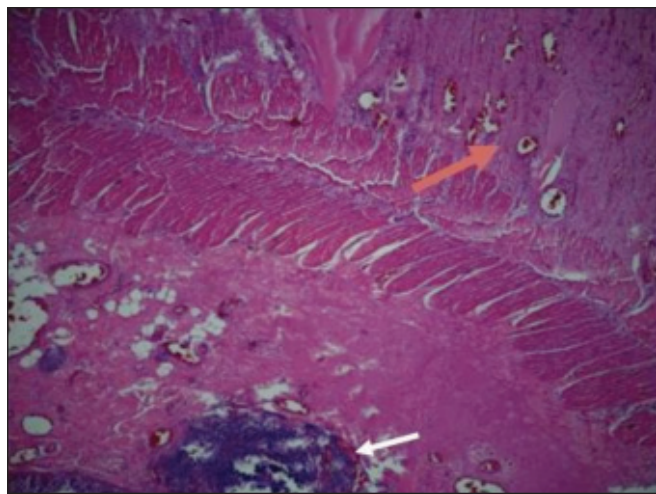


Figure 1. Bowel mucosa and smooth muscle cells (white arrow) adjacent to whirled smooth muscle cell typical of uterine fibroids (red arrow).

An initial exploratory laparotomy in the peripheral hospital found 300 mL of foul smelling pus in the peritoneal cavity, although there was a reported inability to find the source of the pus. The pelvic organs appeared normal except for a fibroid uterus.

The patient was then referred to a tertiary care facility. On presentation, she appeared ill with pyrexia and tachycardia. The abdomen was diffusely tender with a palpable mass on the right hypochondrium. Hematology showed leukocytosis (WBC count of $18 \times 10^3/\text{dL}$) and mild anemia (hemoglobin of 9 g/dL).

Since the computed tomography (CT) scan was not available at that time, based on the signs of peritonism, a decision was taken to perform an exploratory laparotomy. Intra-operatively, 100 mL of frank pus was found originating from the right paracolic gutter. There were significant adhesions noted in the abdomen making dissection extremely difficult. A large solid mass 15×13 cm was found closely adherent to distal ileum and right colon. A right hemicolectomy and primary ileocolic anastomosis were performed with an 'en bloc' excision of the tumor with the specimen. There was no communication with the uterus.

Post-operatively, sepsis was controlled with intravenous antibiotics. The patient tolerated oral fluid intake on the third postoperative day. Her recovery was uneventful, except for an uncomplicated grade 1 superficial surgical site infection.

Histology revealed ischemic changes in a parasitic (serosal) leiomyoma with areas of localized serositis (Figure 1). Adjacent ileum and colon showed no inflammatory bowel disease, dysplasia, or neoplasia.

Case report 2

A 30-year old female had a laparoscopic myomectomy for multiple large fibroids. The procedure was quite eventful. There was prolonged operating time (eight hours), heavy blood loss, and a significant persistent post-operative ileus (eight days). The patient presented to our hospital 11 days after the procedure and had complaints of generalized abdominal pain and high-grade fever, with chills and rigors.

On examination, the patient was tachycardiac (110/minutes) and had an elevated temperature (38.6°C). Her abdominal examination revealed generalized tenderness with peritonism and a tender right upper quadrant mass. The patient also had a leukocytosis (WBC count of 16,000) along with a left axis shift. A CT scan revealed free fluid in the pelvis and an enhancing mass in the right upper quadrant, suggestive of intra-abdominal abscess as there was air within it (Figure 2).

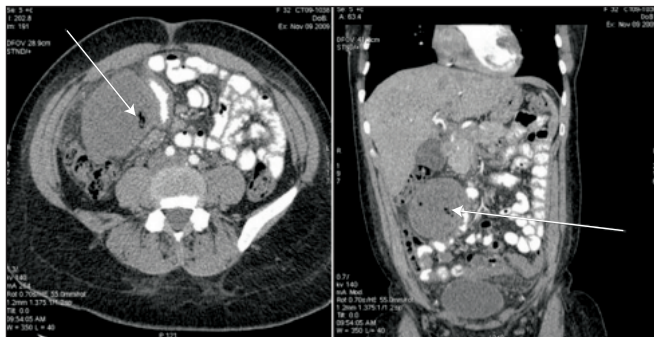


Figure 2. Computed tomography scan showing mass in the right upper quadrant with intramural air (arrow).

A diagnostic laparoscopy revealed approximately 100 mL of pus in the pelvis and a mobile mass in the right upper quadrant, which was intimately related to the hepatic flexure of the colon and several loops of small bowel. Separation was not possible due to serosal tears.

A conversion to upper midline laparotomy was done, and a right hemicolectomy was performed with a primary ileocolic anastomosis and 'en bloc' resection of the mass with the specimen. Grossly after separation of the mass (now recognized to be a fibroid) from the bowel, there was significant friability and bleeding from the serosal surface (Figure 3).

Postoperatively, the patient's sepsis resolved with intravenous antibiotics and she had an uneventful recovery. Histology confirmed ischemic changes in a leiomyoma. Adjacent ileum and colon showed no inflammatory bowel disease, pathogens, dysplasia, or neoplasia.

Discussion

Uterine leiomyomas are benign tumors of smooth muscle origin with protean symptomatology, and they are the most common gynecological tumors in women of reproductive age. Very rarely, benign uterine leiomyomas display bizarre growth patterns with associated extrauterine benign-appearing smooth muscle tumors, which are similar to those found in a uterine fibroid. Leiomyomas occur infrequently outside the uterus. Although they are histologically benign, extrauterine leiomyomas may mimic malignant tumors at imaging and may present a diagnostic challenge. The clinical symptoms and imaging features depend on the location of the lesion and on its growth pattern. These tumors may have arisen from uterus and underwent separation. The spectrum of smooth-muscle tumors arising from the uterus ranges from benign leiomyoma to malignant leiomyosarcoma, but also includes a variety of lesions



Figure 3. Parasitic fibroid (red arrow) intimately related to the mesenteric border of the right colon/hepatic flexure.

with growth patterns that extend outside the uterus, including parasitic leiomyoma, intravenous leiomyomatosis, disseminated peritoneal leiomyomatosis, and benign metastasizing leiomyoma. These benign tumors resemble typical uterine leiomyomas at both gross and microscopic levels².

Parasitic leiomyomas are presented as peritoneal benign smooth-muscle masses separate from the uterus. A parasitic leiomyoma likely originates as a pedunculated subserosal leiomyoma that twists and torts from its uterine pedicle. It becomes free in the peritoneal cavity and it survives by recruiting neovascularization from adjacent structures. In the era of laparoscopy, where many procedures are done with morcellation techniques to remove the specimen, it is not difficult to fathom the possible genesis of this rare condition^{3,4}. In the cases presented, one of the patients had a true parasitic myoma, while the other one may have had a retained myoma, which had already started to recruit a blood supply and may have presented later on as a parasitic myoma.

Parasitic leiomyomas, though infrequent, are most commonly located in the broad ligament, pelvic peritoneum, cul-de-sac, and omentum². In both cases, these parasitic tumors seem to have a predilection for the recesses of the right side of the abdomen. In the case of the latter, the fibroids are usually placed in the right side of the abdomen, in order to allow an easier localization when they are ready to be morcellated and removed. One of the fibroids was left inside the abdomen inadvertently. Even so, in the early phase of the laparoscopic myomectomy, it was not unusual to leave the excised fibroid in the abdomen allowing for self-digestion.

The literature is limited in its description of parasitic leiomyoma since this is a rarely diagnosed condition. It is likely that this condition may be under-diagnosed since many of these tumors, if present, may remain asymptomatic for a number of years and may only

become evident on routine scanning for other causes. The two cases presented may be the first description to our knowledge of parasitic leiomyomas presenting as generalized abdominal sepsis.

Kho and Nezhat⁴ have one of the largest series in the literature of parasitic myomas, they attempted to determine their cause, associations, and risk factors. However, in this retrospective review, they were only able to identify 12 cases of parasitic myomas from 2000 to 2008. Laparoscopy was used as a diagnostic tool, which confirmed the presence of intra/retroperitoneal myomas distinct from the uterus in the 12 cases. Ten of the 12 patients had prior abdominal surgery. Eight had prior morcellation procedures; six performed laparoscopically, two performed by laparotomy. This clearly suggests that prior surgery for uterine fibroid was a risk factor for the development of parasitic leiomyoma⁴.

Within the last 15 years, laparoscopic myomectomy has gained popularity and has shown clear advantages over traditional open techniques. It is usually indicated in patients with uterine fibroids numbering less than three to four with a maximum size of 8 to 9 cm⁵. However, these indications have been called into question, and it has been shown that in the hands of experienced laparoscopic surgeon, size, number, and location do not matter⁶. Despite the controversies regarding the indication, laparoscopic myomectomy has been shown to be a feasible option that boasts a conversion rate as low as 1 to 3%, fertility rates after laparoscopic myomectomy of more than 50% and successful myomectomy rate of more than 80%⁵. As surgeons' dexterity with the procedure improved and the associated standardization of the technique accomplished, operative intervention time has become acceptable and blood loss became less important. In addition, laparoscopic myomectomy is also associated with all of the advantages of laparoscopy with respect to postoperative adhesion, decreased postoperative pain, short hospital stay, and early return to normal function⁷.

Despite the optimism regarding laparoscopic myomectomy and its superior safety profile, there are a number of case reports in the literature describing parasitic myomas after laparoscopic myomectomy. One of the early cases was described by Hutchins and Reinoehl⁸, in 1998, in which a 6 cm fibroid was lost at the time of a laparoscopic assisted supracervical hysterectomy and resulted in severe abdominal pain. The fibroid was eventually removed by laparotomy. In this case, the fibroid was located in the

right upper quadrant of the abdomen at laparotomy similar to Case 2⁸.

Cucinella et al.⁹ recently reported, on their series of parasitic myomas, that they identified four cases over a three-year period. Electric morcellators were used in 423 cases and the overall prevalence of developing parasitic myoma was reported at 0.9 and 1.2% for those who had laparoscopic myomectomies⁹. There are also cases in the literature in which parasitic myomas have been found attached to the bowel and anterior abdominal wall after laparoscopic myomectomy^{10,11}. There were no cases in the literature in which parasitic myomas were the primary cause or associated with generalized abdominal sepsis.

Parasitic myomas are rare occurrences, which can prove to be a diagnostic dilemma and may even be present in the acute setting or even mimic a malignancy. There are increasing reports of this condition being recognized after laparoscopic myomectomy especially in those cases where tumors are morcellated for removal. It is likely that primary parasitic myomas will continue to be rare occurrences, however, parasitic myomas, which are iatrogenically caused, may be diagnosed more commonly in this the laparoscopic era.

More parasitic myomas may be iatrogenically created after surgery, particularly surgeries using morcellation techniques, and within the recent literature there is the emergence of iatrogenic parasitic leiomyomas as a new class of myomas¹². With increasing rates of laparoscopic procedures, surgeons should become aware of the potential for iatrogenic parasitic myoma formation and take the necessary intraoperative precautions to minimize their occurrences.

Some authors recommend a thorough inspection and washing of the abdominal-pelvic cavity at the end of the surgery, in an attempt to prevent this rare complication and in patients with a history in which morcellation techniques were used in a previous operation and who have now presented with abdominal masses, iatrogenic parasitic myomas should be considered in the differential diagnosis^{9,13}. The literature on parasitic myoma is sparse and to our knowledge there have not been any reported cases where their primary mode of presentation was abdominal sepsis. This article is intended to add to the existing literature on parasitic myomas and to highlight that the incidence of this rare condition could be on the rise in the laparoscopic era. The article also highlighted that this condition can manifest itself acutely with abdominal sepsis as its primary mode of presentation.

References

1. Van Katwijk C, Peeters LLH. Clinical aspects of pregnancy after the age of 35 years: a review of the literature. *Hum Reprod Update*. 1998;4(2):185-94.
2. Fasih N, Prasad Shanbhogue AK, Macdonald DB, Fraser-Hill MA, Papadatos D, Kielar AZ, et al. Leiomyomas beyond the uterus: unusual locations, rare manifestations. *Radiographics*. 2008;28(7):1931-48.
3. Hameed N, Ali MA. Recent trends in laparoscopic myomectomy. *J Ayub Med Coll Abbottabad*. 2004;16(1):58-63.
4. Kho KA, Nezhat C. Parasitic myomas. *Obstet Gynecol*. 2009;114(3):611-5.
5. Malartic C, Morel O, Akerman G, Tulpin L, Clément D, Barranger E. Laparoscopic myomectomy in 2007: state of the art. *J Gynecol Obstet Biol Reprod (Paris)*. 2007;36(6):567-76.
6. Sinha R, Hegde A, Mahajan C, Dubey N, Sundaram M. Laparoscopic myomectomy: do size, number, and location of the myomas form limiting factors for laparoscopic myomectomy? *J Minim Invasive Gynecol*. 2008;15(3):292-300.
7. Sizzi O, Rossetti A, Malzoni M, Minelli L, La Grotta F, Soranna L, et al. Italian multicenter study on complications of laparoscopic myomectomy. *J Minim Invasive Gynecol*. 2007;14(4):453-62.
8. Hutchins FL Jr, Reinhoehl EM. Retained myoma after laparoscopic supracervical hysterectomy with morcellation. *J Am Assoc Gynecol Laparosc*. 1998;5(3):293-5.
9. Cucinella G, Granese R, Calagna G, Somigliana E, Perino A. Parasitic myomas after laparoscopic surgery: an emerging complication in the use of morcellator? Description of four cases. *Fertil Steril*. 2011;96(2):e90-6.
10. Pezzuto A, Serboli G, Ceccaroni M, Ferrari B, Nardelli GB, Minelli LL. Two case reports of bowel leiomyomas and review of literature. *Gynecol Endocrinol*. 2010;26(12):894-6.
11. Moon HS, Koo JS, Park SH, Park GS, Choi JG, Kim SG. Parasitic leiomyoma in the abdominal wall after laparoscopic myomectomy. *Fertil Steril*. 2008;90(4):1201.e1-2.
12. Nezhat C, Kho K. Iatrogenic myomas: new class of myomas? *J Minim Invasive Gynecol*. 2010;17(5):544-50.
13. Larraín D, Rabischong B, Khoo CK, Botchorishvili R, Canis M, Mage G. "Iatrogenic" parasitic myomas: unusual late complication of laparoscopic morcellation procedures. *J Minim Invasive Gynecol*. 2010;17(6):719-24.