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Case Report

Parasitic leiomyoma presenting as an inguinal hernia in a postmenopausal woman

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ARTICLE INFO

Article history:

Received 16 January 2018

Accepted 8 April 2018

Keywords:

Parasitic leiomyoma

Intra-abdominal mass

Inguinal hernia

ABSTRACT

Uterine leiomyomas are one of the most common tumors affecting reproductive-age women. Leiomyomas can present as an intrauterine mass or rarely as an extrauterine tumor. Depending on its location, the diagnosis of extrauterine leiomyoma can be challenging, and multiple imaging modalities may be needed for correct identification and differentiation from malignant entities. We report the case of a 48-year-old-postmenopausal female who presented with a painful left inguinal mass, which was clinically diagnosed as inguinal hernia. Ultrasound, computed tomography, magnetic resonance imaging, and percutaneous biopsy were used to characterize the mass. Surgical resection and histopathological analysis revealed the mass to be a parasitic leiomyoma, a very rare cause of inguinal hernia, especially in a postmenopausal woman.

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1. Introduction

Leiomyomas are common tumors which arise from the smooth muscle cells of the myometrium. They usually present as an intrauterine tumor, but may occasionally be extrauterine. The leiomyomas which evolve to lose their attachment from the uterus and become adherent to other organs (such as the broad ligament, omentum, or retroperitoneal connective tissue) and acquire blood supply from them are called

parasitic leiomyomas [1-3]. Laparoscopic hysterectomy or myomectomy has been reported to increase the risk of development of parasitic leiomyomas, due to unintentional seeding of the fragments during the procedure [3]. Parasitic leiomyomas may cause pain or symptoms from compression of adjacent structures. We present this unique case of a postmenopausal woman who presented with an inguinal swelling at an academic institution, and was diagnosed with parasitic leiomyoma. We discuss the imaging findings and the final diagnosis.

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<https://doi.org/10.1016/j.radcr.2018.04.014>

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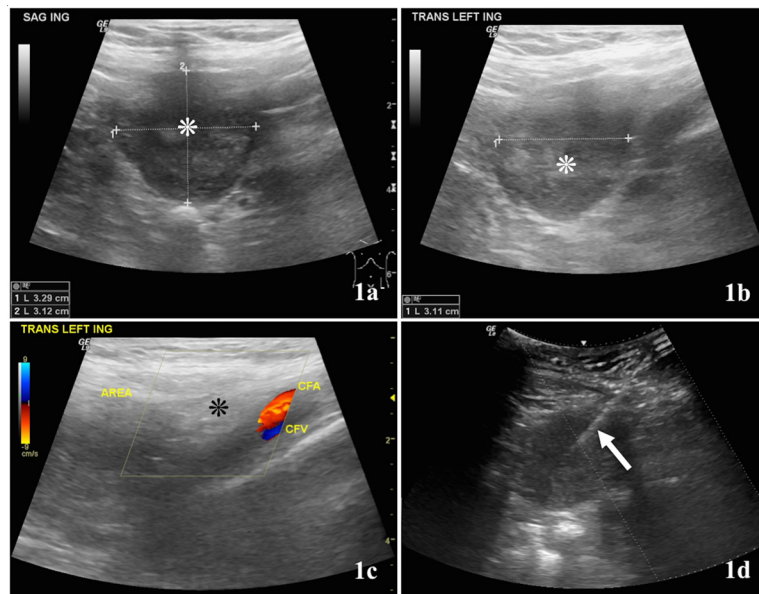


Fig. 1 – Ultrasound of the left lower quadrant of the abdomen. (a) and (b) are sagittal and transaxial US images showing the heterogenous hypoechoic left inguinal mass (asterisk). (c) shows that the mass (asterisk) is superficial to common femoral vessels (red and blue in the color Doppler box). (d) is from the US-guided fine needle aspiration showing the needle (white arrow) inside the mass. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article)

2. Materials and methods

2.1. Patient information and clinical findings

A 48-year-old female, G2(gravida)P2(parity), was seen at the primary care clinic with complaint of a 1-year history of left lower quadrant abdominal pain. She reported having her last menstrual period about 18 months ago. Past medical history was significant for left-sided laparoscopic oophorectomy for an ovarian cyst more than 5 years before this presentation. Patient denied prior hormonal therapy. Initially, patient felt a mass in the inguinal region, only during standing and coughing, and it reduced spontaneously upon lying down. More recently, the mass was constantly present and was associated with deep pain. The pain was worse with coughing and walking. Physical examination showed a firm palpable mass, about 4 cm in size, in the left inguinal region, which was tender and irreducible. The overlying skin was normal and no inguinal adenopathy was present.

2.2. Diagnostic assessment

A left lower quadrant ultrasound (US) was performed which demonstrated a well demarcated, hypoechoic, solid mass in the left inguinal region, superficial to the common femoral vessels (Fig. 1). Pelvic ultrasound was also performed which showed a normal uterus with an endometrial stripe of 6 mm, and a normal right ovary. Lymphadenopathy, inguinal hernia, and a soft tissue tumor were considered as possibilities and further evaluation with computed tomography (CT) or Mag-

netic resonance imaging (MRI) was recommended. CT of the abdomen and pelvis (performed with intravenous and oral contrast) showed a solid soft tissue density mass in the left side of pelvis, measuring 4.4×3.3 cm. The mass extends into the left inguinal canal, lateral to the inferior epigastric vessels, and with blood supply from the left broad ligament vessels (Fig. 2). MRI was performed for better intralesional characterization, and demonstrated the well-defined, solid left pelvic mass, hypointense to the uterus on T1-weighted images, no signal drop on fat-saturated images and out of phase images (ruling out macroscopic and microscopic fat respectively), low signal on fat-saturated T2-weighted images with scattered foci of high T2 signal inside the tumor, and with avid contrast enhancement (Fig. 3). Patient was referred to interventional radiology for image-guided biopsy. An US-guided biopsy was performed (Fig. 1) and multiple fine needle and core biopsy samples were obtained. Cytology showed fibrous connective tissue, fibroadipose tissue, and disorganized bundles of smooth muscle. The lesion was reported as possibly leiomyoma, and complete excision was recommended for definitive diagnosis.

2.3. Therapeutic intervention and follow-up

Under general anesthesia, and using a midline laparotomy incision from below the umbilicus to the pubic symphysis, the left inguinal region was accessed. The mass was palpated in the left inguinal canal and then reduced back to the abdominal cavity by manual palpation. There were a few abnormal-appearing lymph nodes near the area of the mass in the left pelvic region and these were dissected out. Other

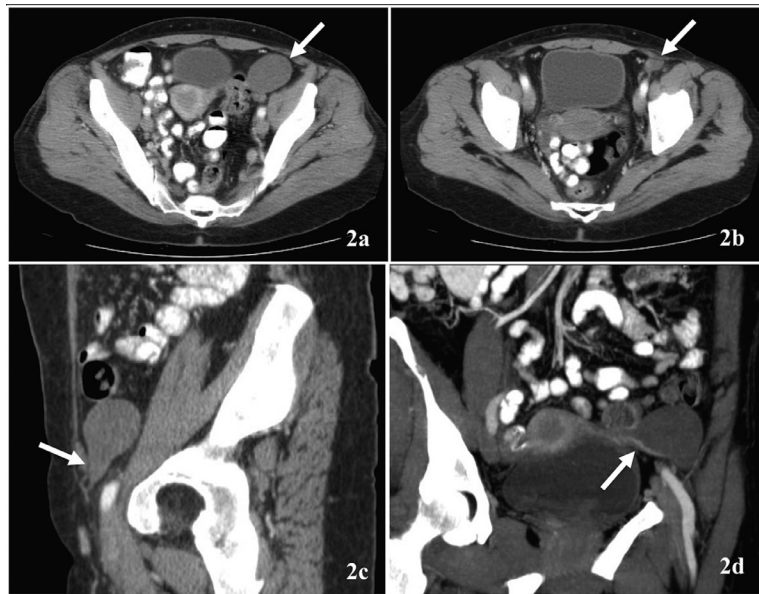


Fig. 2 – CT of the abdomen and pelvis, with intravenous and oral contrast, and in a portal-venous phase. (a) and (b) are transaxial images at the level of the pelvis (in soft tissue windows), and show an oval well-defined, uniform, soft tissue density, enhancing mass (white arrow). (c) is an oblique maximum intensity projection image showing the extension of the mass into the left inguinal canal (white arrow). (d) is an oblique maximum intensity projection image showing a round ligament vessel supplying the mass (white arrow)

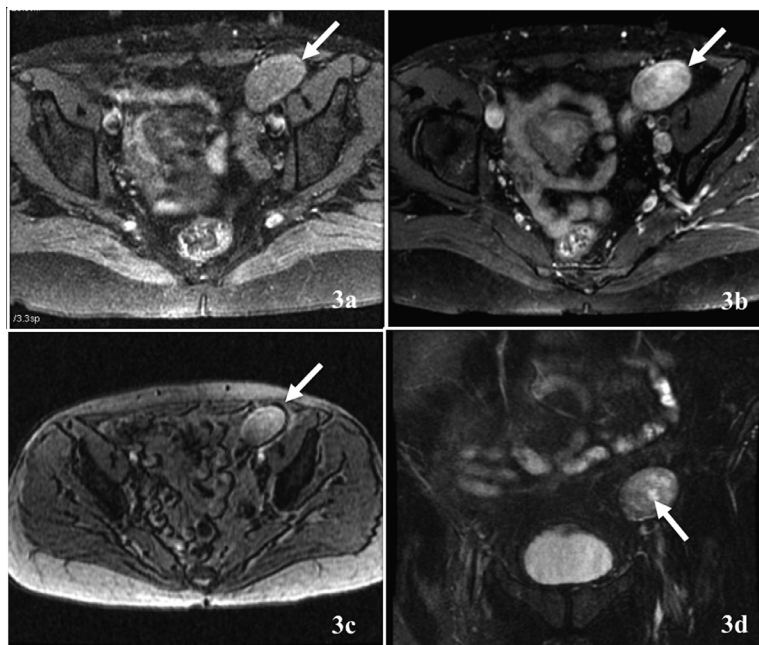


Fig. 3 – MRI of the pelvis. (a) and (b) are fat-saturated T1-weighted axial images without and with intravenous contrast, respectively, showing the intermediate to low signal, heterogeneously enhancing mass in the left side of pelvis (white arrow). (c) is a transaxial T1-weighted out-of-phase image showing no evidence of signal drop (white arrow) in relation to the in-phase image (not shown), to suggest microscopic fat. The fat-saturated T2-weighted coronal image (d) shows an intermediate to low signal in the mass with foci of high signal (white arrow) suggesting degeneration in a fibroid

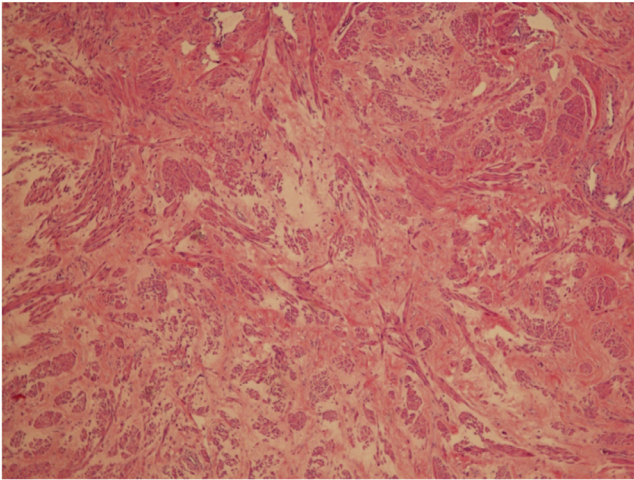


Fig. 4 – Hematoxylin and eosin stain (40x) from the resected inguinal mass showing bundles of smooth muscle cells in a fascicular pattern, separated by connected tissue. Occasional fibrillar cytoplasm and hyaline change is also seen

surrounding structures including the left fallopian tube and the ureter were found to be unremarkable. The mass was excised and sent to pathology. Right oophorectomy was also performed as the right ovary had some suspicious appearance and did not appear as a normal healthy ovary. Patient tolerated the surgery well, recovered uneventfully, and has been asymptomatic since then. Histopathological analysis of the mass showed parasitic leiomyoma (Fig. 4). Reactive changes were seen in the excised lymph node and the right ovary was unremarkable.

3. Discussion

Parasitic leiomyomas are considered to be pedunculated subserosal uterine leiomyomas which have lost attachment to the uterus, and have adhered to and gained blood supply from other adjacent organs. They can be found anywhere in the abdomen and pelvis, and have been reported in the omentum [4,5], the ovaries [6], and in the bowel [7]. Lete et al. published the first systematic review on parasitic leiomyomas in 2016 [3]. In their analysis, the mean age of women at the time of diagnosis was 40 years (ranging from 18-79 years). About 44% had a prior history of myomectomy or hysterectomy, 39% had a history of power morcellation, and 56% had no history of uterine surgery. Several series have described the characteristics of postoperative or iatrogenic parasitic leiomyomas [1-3]. Several other reports have described the occurrence of parasitic leiomyomas in patients without pelvic surgery [4,6]. In our patient, the prior history of ipsilateral laparoscopic oophorectomy probably contributed to the development of parasitic leiomyoma.

Fasih et al. have described in detail the imaging patterns of extrauterine leiomyomas [8]. At US they have a whorled

appearance, with variable echogenicity, depending on calcification, fibrosis, and degeneration. Differential may include ovarian or tubal masses, broad ligament cysts, or leiomyomas. MRI is extremely useful in differentiating ovarian and tubal masses and cysts from parasitic leiomyomas. Typical leiomyomas demonstrate low to intermediate signal on T1-weighted images, and low signal intensity on T2-weighted images [8, 9]. Focal high T2 signal may be seen with myxoid degeneration and necrosis. A cobblestone appearance has been described on both T1- and T2-weighted images with hyaline degeneration. Contrast enhancement is seen on both CT and MRI. US-guided biopsy may help in definitive diagnosis [8, 9]. Dynamic contrast enhanced MRI is the modality of choice for sonographically indeterminate adnexal masses. Low T2 signal is characteristic of benign fibrous tumor (such as an ovarian fibroma) or muscular tumors (such as uterine leiomyoma). Contrast enhanced T1-weighted images are helpful in indeterminate cases to differentiate between a solid benign vs a malignant tumor [10]. Our case is limited by availability of only the T2 fat-saturated images (Fig. 3c).

Round ligament leiomyomas are a related entity. Although again uncommon, they are believed to arise from the smooth muscle cells in the round ligament. They are typically diagnosed in premenopausal middle-aged women and can be found anywhere along the entire length of the round ligament, from the uterus to the labia majora, passing through the inguinal canal [11-13]. These tumors have not been reported to be associated with prior myomectomy or hysterectomy. They can present as an inguinal mass and may even incarcerate [11-13]. Imaging is helpful in differentiating these from bowel containing inguinal hernias and other inguinal canal pathologies such as lipoma, liposarcoma, hematoma, abscess, neurofibroma, desmoid tumor, uterine fibroid, endometriosis, pseudomyxoma peritonei, femoral artery aneurysm, lymphadenitis, saphena magna thrombophlebitis, dermoid, and epidermoid cysts [14]. The imaging characteristics of round ligament leiomyomas are similar to uterine and other extrauterine (or parasitic) leiomyomas [8,14]. In the presented case, the temporal association of the development of the mass after ipsilateral laparoscopic oophorectomy differentiated it as a parasitic leiomyoma rather than a round ligament leiomyoma.

Parasitic leiomyomas are most commonly present with abdominal pain and the patient may also have symptoms from compression of other surrounding structures such as increase in urinary frequency, constipation, and dyspareunia. Sometimes, parasitic leiomyomas are asymptomatic and are found incidentally on imaging or during other surgical procedures [3]. In our patient, who was postmenopausal, we speculate that prior left-sided laparoscopic oophorectomy may have played a role in the development of the left-sided parasitic leiomyoma. The patient presented with classical symptoms of an inguinal hernia which over time became irreducible and caused pain. The imaging characteristics were consistent with a degenerating leiomyoma and an US-guided fine needle aspiration made the preliminary diagnosis. The unique features of our case include a growing leiomyoma, in a postmenopausal woman, which presented as an inguinal hernia, and probably developed secondary to prior laparoscopic oophorectomy. The limitations include use of both CT and MRI for charac-

terization, and availability of only the T2 fat-saturated images. Our search found only one other report in the literature of a parasitic leiomyoma as a cause of an inguinal mass [15].

Treatment of a parasitic leiomyoma is indicated if the patient has symptoms. Surgical resection is the treatment of choice for symptomatic parasitic leiomyomas [3]. In some cases, the parasitic leiomyoma may be asymptomatic, or found incidentally, or may not grow on follow-up imaging. In such cases, watchful waiting and imaging follow-up may be indicated. Postresection prognosis is good and the chances of recurrence are low. In our patient, the definitive diagnosis was made after surgical resection, which was well tolerated.

4. Conclusions

Parasitic leiomyomas are rare extrauterine leiomyomas which can be found anywhere in the abdomen or pelvis. Prior hysterectomy, myomectomy, and morcellation procedures increase the risk of the development of parasitic leiomyomas. They usually present with pain and other symptoms from compression of adjacent structures. Although similar in presentation and imaging characteristics, parasitic leiomyomas are distinct from round ligament leiomyomas. Similar to round ligament leiomyomas, a parasitic leiomyoma may also present as an inguinal hernia. Surgical resection is the treatment of choice for symptomatic tumors.

Conflicts of Interest

None.

Disclosure

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Consent

Written consent was taken from the patient for publication of this report.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2018.04.014.

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