Case Report

Endometriosis of the Paralumbar Muscles: A Case Report and Literature Review

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Abstract

The objective of this study is to report a case of deep endometriosis of the paralumbar muscles (psoas, multifidus, and erector spinae) and review existing literature on its management. A 34-year-old female with a history of endometriosis was seen for infertility. Paralumbar muscle masses seen on computed tomography (CT) scan were sampled, confirming endometriosis. Gonadotropin-releasing hormone agonist was given for 2 months. The patient was primed for assisted reproduction. A literature review was conducted to provide an understanding of paralumbar muscle endometriosis. To our knowledge, this is the first reported case of multifidus and erector spinae muscle endometriosis and fifth case of psoas muscle endometriosis. Because the available information is scarce, data from the existing literature on deep endometriosis may aid in the diagnosis and management. Magnetic resonance imaging and CT scan are essential imaging techniques to map lesions. Excision seems prudent, but the approach should be individualized depending on the patient's presentation and her preferences.

Keywords: Back muscles, endometriosis, erector spinae, multifidus, paraspinal muscles, psoas muscle

Introduction

Endometriosis is a common benign, inflammatory, estrogendependent condition with a prevalence that varies from 1% to 11.5% of women.^[1] One of its three distinct expressions is deep endometriosis. Described as "deeply infiltrating endometriosis" or "adenomyosis externa," it is characterized histopathologically as endometrial glands and stroma penetrating adjacent fibromuscular tissue, obliterating and substituting connective tissue septa.^[2]

This article aims to report a case of deep endometriosis with endometriotic cysts of the paralumbar muscles – psoas, multifidus, and erector spinae muscles – and review the existing literature on its diagnosis and management.

CASE REPORT

A 34-year-old married, nulligravid, regularly menstruating Taiwanese woman presented to the gynecology outpatient

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clinic for infertility. She underwent oophorocystectomy in 2012 for severe pelvic endometriosis and was given gonadotropin-releasing hormone (GnRH) agonists for 6 months. She was subsequently lost to follow-up. Over the past year, she experienced dysmenorrhea of 5/10 severity, left flank soreness, urinary frequency, vomiting, dyschezia, and diarrhea associated with menstruation. A gastroenterologist performed biopsy of a mucosal lesion seen on colonoscopy, which confirmed endometriosis.

On pelvic examination, a nodular posterior vaginal fornix, slightly enlarged uterus, and a 7 cm × 7 cm left adnexal mass were palpated. CA-125 was 568.2 U/mL. Abdominal computed tomography (CT) scan revealed a left endometrioma, bilateral

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hydrosalpinges, and mild hydronephrosis. At the level of L4–L5, a 4-cm enhancing cystic lesion was seen at the area of and displacing the left psoas muscle. Another 4-cm enhancing cystic mass was found encroaching on the left multifidus and erector spinae muscles [Figure 1]. Retroperitoneal endometriosis was considered. Abscesses could not be ruled out.

In view of the patient's desire for an immediate pregnancy, a complete extirpative surgery was excluded. On laparoscopy, both adnexa and rectum were plastered to the posterior uterine wall, occluding view of the *cul-de-sac*. Chocolate-colored fluid leaked from intraoperative rupture of the cystic ovaries. The patient underwent lysis of adhesions, bilateral oophorocystectomy, and bilateral proximal tubal occlusion. Double J ureteral stenting and cystoscopy revealed patent ureteral orifices and a submucosal nodule at the trigone. Postoperative CT-guided aspiration of retroperitoneal cysts was performed.

The histopathology report confirmed endometriosis [Figure 2]. Postoperatively, the patient was given monthly GnRH agonist subcutaneous injections. After 2 months, a repeat abdominal CT scan showed regression in the size of endometriotic lesions with the resolution of hydronephrosis. The ureteral stents were subsequently removed. Repeat CA-125 result was 39 U/mL. She is currently being primed for *in vitro* fertilization.

METHODS

PubMed/MEDLINE, Cochrane Library, EMBASE, and Ovid Discovery were screened for the search terms, "psoas muscle," "multifidus," "erector spinae," and "endometriosis" from inception until May 2019. Studies of endometriosis not involving any of the aforementioned muscles were excluded. Articles were assessed by two independent reviewers (MCT and YJC). Data such as author, date of publication, parity, clinical presentation, imaging, and interventions were then tabulated. The study was reviewed and approved by the Human Investigational Review Board of Taipei Veterans General Hospital (VGH) (VGH IRB: 2017-10-012AC).

RESULTS

The method generated 13 articles, from which four relevant case reports were culled for qualitative analysis. All were

cases of psoas muscle endometriosis. No study describing endometriosis of the multifidus or erector spinae muscles was found. The extracted data are summarized in Table 1.

DISCUSSION

Deep endometriosis occurs in 14.4% of women with endometriosis and is characteristically multifocal. [7] Commonly affected areas include uterosacral ligaments, vaginal fornix, rectovaginal septum, parametria, pouch of Douglas, rectum, rectosigmoid, bladder, ureter, and vesicouterine fold. Infrequently, endometriotic lesions may implant on other anatomic sites such as the abdominal wall, umbilicus, inguinal region, round ligament, thorax, and appendix. [7]

Paralumbar muscle endometriosis is extremely rare in a commonly occurring condition that is endometriosis. Only four cases of psoas muscle endometriosis have been documented. No multifidus or erector spinae muscle involvement has been previously reported.

The clinical presentation of endometriosis infiltrating the psoas muscle may be described as catamenial and is reported as back, hip, or leg pain with or without signs of lower extremity radiculopathy. Related symptoms include dysmenorrhea, dyspareunia, and chronic pelvic pain. As with other forms of endometriosis, CA-125 levels may be increased. The index case presented with pelvic pain, gastrointestinal (GI), and urinary symptoms such as frequency and flank soreness, with nodularity of the posterior vaginal fornix. These and a history of surgery for endometriosis led to a diagnosis of deep endometriosis. The findings are consistent with studies that identify both GI and urinary tract systems as the most frequent sites of extragenital endometriosis. The rectum, sigmoid, and bladder are affected in majority of cases.^[8]

Diagnosis

According to Redwine, ovarian cysts often coexisted with deeper pelvic and intestinal disease.^[9] This observation and the presence of bladder lesions indicate the possibility of multiple organ involvement. Imaging techniques are thus critical in mapping lesions. Transvaginal ultrasound (TVS) is the diagnostic test of choice for screening deep endometriosis involving the rectosigmoid and rectocervical areas because of its higher sensitivity and specificity compared to magnetic resonance imaging (MRI) and manual pelvic examination.^[10]

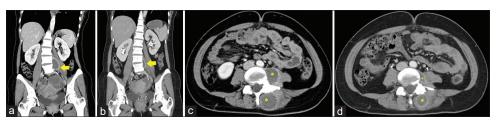


Figure 1: Axial view of pretreatment (a) and posttreatment (b) lesions (arrows) and coronal view of pretreatment (c) and posttreatment (d) cystic lesions of the paralumbar muscles (*)

Study	Age	Parity	Presentation	Imaging findings	Surgery/OP	Medical treatment and outcome
Bhat 2007 ^[3]	39	2	Right hip pain radiating to thigh; pain/difficulty in moving hip joint	CT scan: Multiloculated cystic lesion within the iliopsoas muscle	Laparotomy, excision of iliopsoas muscle cyst OP: Solitary lesion on right iliopsoas muscle	Cyclical hormones for 3 months with symptomatic relief
Andrade 2015 ^[4]	38	2	History of endometriotic cyst excision; dysmenorrhea,	Vaginal ultrasound: Thickened rectosigmoid wall	Guided biopsy; laparoscopy, incomplete resection of psoas endometriosis	GnRH agonist for 3 months preoperatively; GnRH postoperatively
			dyspareunia, dyschezia, chronic pelvic pain with radiation to the anterior left thigh associated with menstruation; free rectovaginal septum	MRI: Heterogeneous solid mass of 5 cm, between the left iliacus, psoas muscles, and descending colon	Laparoscopy, femoral nerve neurolysis, excision of endometriotic tissues, total hysterectomy with bilateral salpingo-oophorectomy OP: Retroperitoneal nodule on left; sigmoid loop adhesion	None after second laparoscopy With clinical improvement after second laparoscopy
Tao 2016 ^[5]	49		Elevated CA-125 levels during annual examination History of endometriosis Underwent TAH-LSO 7 years prior	PET-CT: Soft-tissue attenuation mass in the left side of the psoas major MRI: Miscellaneous intensity mass measuring 4.8 cm × 2.5 cm with heterogeneous enhancement Repeat MRI: 7.0 cm × 4.2 cm mass	Laparotomy, biopsy OP: 10 cm lesion at the left psoas major; adhesions	GnRH agonists for more than 3 months with a resultant decrease in CA-125 levels
Zhao 2018 ^[6]	28		Lower left abdominal and back pain and swelling	MRI: Small cyst in left uterine wall; 10 cm mass near left retroperitoneal iliac vessel and left ureter	Ultrasound-guided percutaneous drainage Laparoscopy, resection of psoas muscle endometriosis Op: Psoas muscle mass; dense adhesion between uterus and sigmoid colon; adhesion between part of omentum, sigmoid, and left pelvic side wall	Oral contraceptives for 3-6 months with no recurrence

PET: Positron-emission tomography, CT: Computed tomography, MRI: Magnetic resonance imaging, GnRH: Gonadotropin-releasing hormone, TAH-LSO: total abdominal hysterectomy, left salpingo-oophorectomy, OP: Operative findings

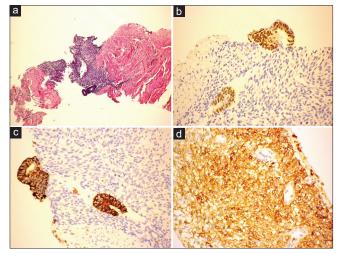


Figure 2: Histological and immunohistochemical studies confirming paralumbar muscle endometriosis. (A) Hematoxylin and eosin stain. Endometrial glands and stroma are seen interspersed in muscle, x4. (B) Paired box 8 immunostain. Ectopic endometrial gland nuclei stain positive for PAX8, x20. (C) Cytokeratin immunostain. Endometrial glands stain positive for CK while stromal cells stain negative, x20. (D) Cluster designation 10 immunostain. Ectopic endometrial stroma stain negative for CD10 while endometrial glands stain negative, x20

However, this modality has limited sensitivity in detecting lesions at more posterior compartments. In one study, TVS detected rectosigmoid thickening but not psoas muscle involvement.^[5] Therefore, it is less useful in mapping all endometriotic lesions prior to surgery.

MRI, CT scan, and a combination of positron-emission tomography-CT scan plus MRI have been used to diagnose retroperitoneal endometriosis. In the index case, the absence of nerve involvement or a palpable mass on physical examination precluded the clinical diagnosis of paralumbar muscle endometriosis. Likewise, pelvic, GI, or urinary tract symptoms may have confounded any pain from the growing retroperitoneal masses. This could explain why the lesions were discovered only incidentally on CT scan.

In this regard, the use of appropriate imaging techniques is crucial in diagnosing patients exhibiting signs and symptoms of multifocal endometriosis. MRI and CT scan are both useful in these situations, with MRI having a higher sensitivity and specificity in highlighting soft tissues in the abdominopelvic cavity and detecting multiple organ involvement.^[11] With

the five reported cases of paralumbar muscle endometriosis, three were reliably identified through MRI, whereas two from CT scan. However, due to the limited number of cases, the superiority of MRI over CT scan in detecting posterior muscular involvement remains inconclusive.

On imaging, psoas muscle endometriosis is characterized as an enhancing, multiloculated, heterogeneous, cystic or solid, retroperitoneal mass measuring between 4 and 10 cm in the greatest dimension. It is worth mentioning that in the case presented, hydronephrosis was seen on initial scan with resolution on subsequent imaging concurrent with tumor size reduction. This and the absence of ureteral endometriosis on laparoscopy are consistent with the compression of the left renal pelvis by a displaced left psoas muscle.

Treatment

Endometriosis resection is generally regarded as the treatment of choice for deep endometriosis. [9]. However, complete extirpation of endometriotic lesions may not be preferred for all patients with endometriosis, including those with infertility.

There are only a few studies about infertility and surgery for deep endometriosis. No definitive recommendations exist, and some studies are conflicting. [12,13] The presence of infertility in deep endometriosis does not prove causality, but there appears to be a strong link between infertility and the presence of distinct endometriotic lesions in deep endometriosis. [13] These include superficial endometriotic implants, pelvic adhesions, endometriotic cysts, and adenomyosis. In contrast, deep endometriotic lesions are not exposed to the peritoneal cavity and are thus not implicated in the local inflammatory process in endometriotic lesions strongly associated with infertility. This lack of causality is insufficient to support surgery for infertility in patients with deep endometriosis. [13]

In view of the foregoing, the role of surgery in patients with deep endometriosis, particularly paralumbar muscle endometriosis, and infertility, remains to be elucidated. For this reason, it is important to make a comprehensive assessment of affected women and determine their individual needs based on their symptoms and desires for future fertility.

For patients opting medical treatment alone, hormonal therapy is essential in controlling symptoms. Pain is prevalent and is managed through menstrual cycle suppression. A systematic review on endometriosis identified that levonorgestrel intrauterine system, danazol, and GnRH agonists were all effective in achieving the desired effect. [14] A combination of GnRH agonist therapy and assisted reproductive techniques may also be effective in bolstering pregnancy rates. [12]

It is important to note, however, that there is a risk of recurrence for medical treatment without exeresis.^[14] This must be considered when opting medical therapy, especially in women

undergoing assisted reproduction. Long-term treatment plans should be discussed with the patient and entails working with a multidisciplinary care team.

In summary, this case report and literature review shows a paucity of data on paralumbar muscle endometriosis. At present, there are very few studies on psoas muscle endometriosis. To our knowledge, this is the first reported case of multifidus and erector spinae muscle endometriosis. Because of the limited information available, existing literature on the multifocal deep endometriosis may aid in the diagnosis and management of paralumbar muscle endometriosis. A comprehensive patient assessment is necessary. TVS is limited to the assessment of pelvic and abdominal cavities and may not detect posterior abdominal wall lesions. The use of appropriate imaging techniques, such as MRI and CT scan, is thus essential to map lesions. Excision seems prudent in most cases, but the approach should be individualized depending on a patient's presentation and her preferences, with special consideration of those desiring future fertility.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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