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Case Report

Endometriosis in the psoas major muscle: a case report

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Abstract: Endometriosis occurs outside the pelvic cavity is uncommon. To our knowledge, until now there is no case like this has been reported. A 49-year-old Chinese woman only with CA125 abnormal was admitted to our clinic. After a series of examinations, a mass in the left psoas major muscle was discovered. Based on the clinical symptoms, serologic markers and imaging features, we still could not rule out the possibility of malignant tumor. Finally, a diagnosis of endometriosis was made based on intraoperative pathological findings. Then she received relevant treatment. Three months later, both the lesion size and CA125 level decreased obviously. Retrospectively analysis of the disease progression, we find that rare cases like this not only enrich the location spectrum of endometriosis, but also help us gain more insight into the pathogenesis of it.

Keywords: Endometriosis, psoas major muscle, image

Introduction

Endometriosis is the presence of functional endometrial glands and stroma outside the uterine cavity; its incidence among childbearing women are estimated ranging from 3% to 10%, nearly 176 million in the world [1-3]. This disease damages female physical and psychological health in terms of pelvic pain and/or infertility [4]. Compelling epidemiological evidence indicates that the implantation of endometrial tissues can occur in any organ, including brain, lung, bowel, abdominal wall, omentum, skin, and bladder [5, 6]. The pelvis, ovaries, and Douglas pouch are the most commonly involved targets [7, 8]. According to the onset location, the disease is generally divided into two categories: the intra-pelvic and extra-pelvic.

Until now, the pathogenesis of endometriosis remains debatable. There are many proposed theories: 1) retrograde menstrual implantation, 2) vascular and lymphatic spread, iatrogenic implantation; 3) metaplastic of the pelvic peritoneal cells; 4) immune system dysfunction and autoantibody formation; and the theory that retrograde menstrual implantation is the most popular one [5, 9-12]. In clinic, the diverse locations, protean clinical manifestations and complicated pathogenesis hinder a clinician from making a correct diagnosis, especially for extra-pelvic cases. As estimated, the delay in diagnosis of endometriosis is about 6.7 years [13]. For the clinician, there is a long way before reaching the right diagnosis. Here, we present a case of endometriosis that occurred in the psoas major with atypical presentations.

Case report

Clinical data

A 49-year-old Chinese female was admitted to hospital, the level of serum Carbohydrate Antigen 125 (CA125) increased (489.58 KU/L, normal range 0-35 KU/L) during her annual checkup (**Table 1**). She had no nausea, fever, headache, dizziness, or weight loss. What special about her medical history was that she had underwent a hysterectomy and left salpingo-oophorectomy for ovary endometriosis and

Table 1. The value of CA125

Date	CA125 (KU/L)
01/20/2015	489.58
04/30/2015	237.40
05/11/2015	214.35
05/29/2015	133.22
07/29/2015	84.31
08/26/2015	14.56
09/22/2015	7.48

Note: CA125 = Carbohydrate Antigen 125.

uterine myoma seven years ago, but we did not get the details of the operation.

Routine examinations

To explore the etiology, the patient underwent a series of examinations and some valuable clues were discovered. The level of CA-199 (164.64 KU/L) and CA-242 (47.7 KU/L) also increased, the carcinoembryonic antigen (CEA) was normal. Then the Positron Emission Tomography-Computed Tomography (PET-CT) showed that there was a soft-tissue-attenuation mass with increased glucose metabolism located in the left side of the psoas major: benign or low-grade malignant tumor was considered. The subsequent lumbar Magnetic Resonance Imaging (MRI) confirmed the presence of the lesion, a miscellaneous intensity mass measuring 4.8×2.5 cm with heterogeneous enhancement in the same location (**Figure 1A**). There was no involvement of the abdominal cavity, no enlarged lymph nodes in the retroperitoneum. A presumed diagnosis that neoplasm originated from the mesenchymal tissue was made.

What a pity, the patient rejected further diagnosis and treatment, and then she discharged from hospital. After three months, the MRI revealed that the size of the mass increased to 7.0×4.2 cm (**Figure 1A**). In addition, the computed tomography angiography (CTA) displayed that the neoplasm encasing the external iliac artery. The abdominal US demonstrated a bright hetero-echoic irregular and ill-defined mass located near the anterior-lateral of the left iliac vessels (**Figure 1B**). The serologic test showed that the level of CA125 was still abnormal (237.40 KU/L, **Table 1**).

As was mentioned, it was a question about how to interpret these findings: the mass was benign or not? Is there relationship between the lesion and history of pelvic surgery? After multidisciplinary consultations, the mass was tentatively interpreted as low-grade malignant tumor, with question about its origins: muscle, nerve tissue, or others. In this situation, did an exploratory laparotomy was indispensable.

During the operation, we could see a mass measuring 10×10×8 cm in the left psoas major, with the peritoneum, iliac vessels and ureter adhesion. Considering the mass with rich blood supply and the adhesion of adjacent tissues, a decision that just did a biopsy rather than completely removed it was made. Then the hematoxylin and eosin (HE) staining showed endometriosis in the mass. While ectopic glands and stroma expressing cytokeratin (CK++) and cluster of Differentiation 10 (CD10+++) (**Figure 2**). The patient was diagnosed as endometriosis pathologically.

Treatment and follow up

We followed up the patient after she received GnRHa (Leuprorelin Acetate Microspheres) for more than three months. Effective treatment was indicated: the mass in size (4.8×2.1 cm) and the level of CA125 (84.31 KU/L, **Table 1**) decreased remarkably (**Figure 3A**). Moreover, we used Susceptibility Weighted Imaging (SWI) and Diffusion Weighted Imaging (DWI) to find some more valuable clues: multiple punctate signal voids within the mass on SWI; hyperintense on DWI, with no restriction of Brownian movement on corresponding Apparent Diffusion Coeffecient (ADC) map (**Figure 3B**). These findings just reflected some features of endometriosis. During our last two follow-ups, we saw that the level of CA125 have already reduced to normal (7.48 KU/L, **Table 1**). In the end, the treatment effects, laboratory tests, and imaging manifestations were in line with former pathological diagnosis. The patient was finally diagnosed as endometriosis without any doubt.

Discussion

As is known to all, endometriosis can occur in liver, cesarean section scar, rectus abdominis muscle, appendix, umbilical cord and so on [10, 14-16]. For this case, the onset location was very rare; the clinical symptoms were atypical and without classical cyclic pelvic pain; the radiology showed a solid mass mimicking a

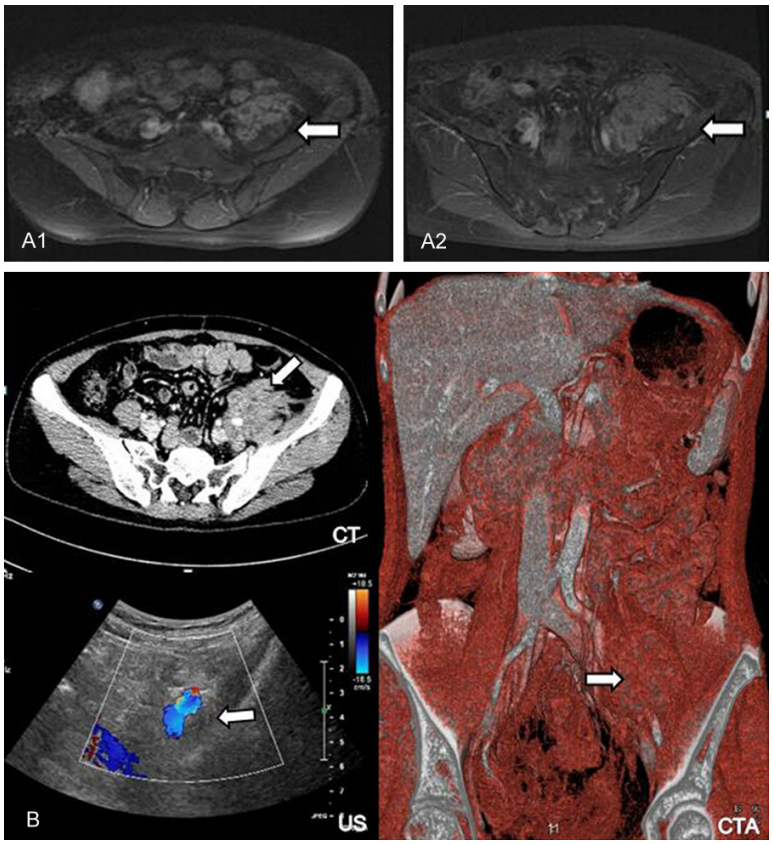

Figure 1. Imaging manifestations. A1. Axial post-contrast T1-weighted image shows a heterogeneous enhancement mass in the left psoas major. A2. Three months later the mass increases in size. B. The mass encases the left external iliac artery on axial CT, US and coronal CTA. White arrow shows the lesion.

neoplasm. All of these misled the doctor to make a false conclusion.

However, we still could find some subtle clues that imply or support the truth: the abnormality of the CA125 and the history of operation. The current study reveals that CA125 has high sensitivity in the detection of endometriosis, especially for late-stage ones [17, 18]. In clinic, the abnormality of CA125 in women can strongly suggest the possibility of endometriosis or epithelial ovarian cancer. If she just had a history of hysterectomy or intra-pelvic endometriosis, we should firstly exclude endometriosis before making any other diagnoses. For this case, we prefer to believe that the lesion was developed by iatrogenic cause, implanted the spread tissue during the surgery.

Besides what mentioned, we should admit that radiology especially MRI is useful in differentiating endometriosis from other diseases. The SWI confirmed the hemosiderin and deoxy-hemoglobin deposited in the mass as signal voids. In addition, the appearances on DWI may be helpful to distinguish benign from malignant tumors [19]; additionally, adhesions are reported as an extremely common and important complication of the endometriosis, here the unclear fat space around the mass may just support the idea [5, 19]. Therefore, we could not deny the fact that MRI is a valuable and reliable tool for assisting us in making a correct diagnosis. We hope that in future there are more specific MRI signs of endometriosis will be discovered.

In conclusion, making a quick and accurate diagnosis of endometriosis is a tough problem in clinics, especially for extra-pelvic ones. Just as the guideline says, the diagnosis of endometriosis should be based on medical history; laboratory and imaging information; while the laparoscopy and histology is still the gold standard [4]. So when we make a diagnosis about masses in women at the reproductive age, the endometriosis should be considered in the wide range of differential diagnoses, especially for patients with known endometriosis or those that have undergone pelvic surgery/cesarean before. Finally, rare cases like this not only enrich the location spectrum of endometriosis, but also help us gain more insight into the pathogenesis of it, and we will be more experienced when facing similar cases in the future.

Disclosure of conflict of interest

None.

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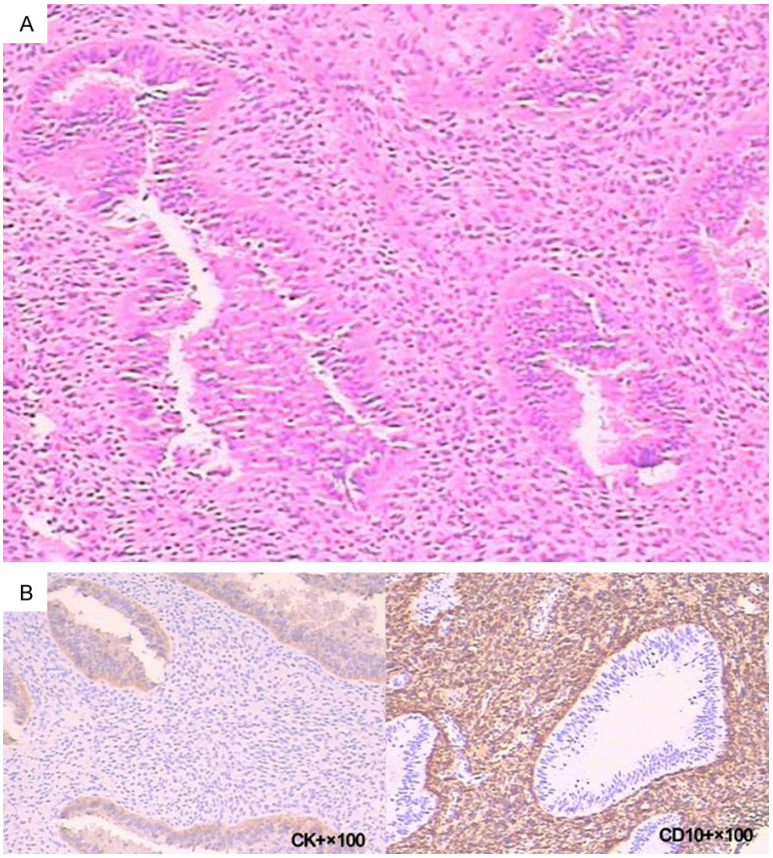

Figure 2. Pathological examination results. A. Endometriosis in left psoas major on HE staining. B. Ectopic glands and stroma express CK and CD10.

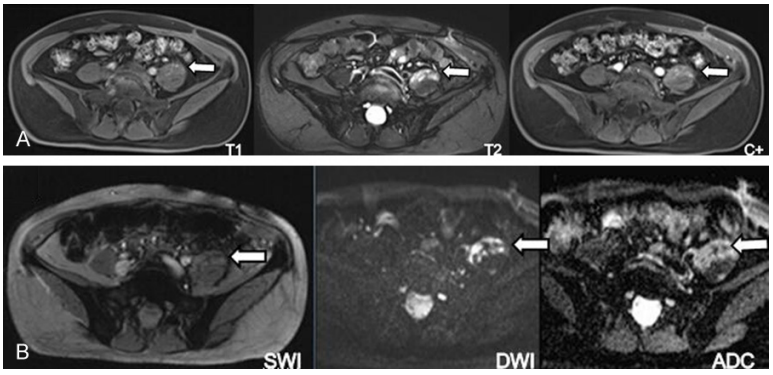

Figure 3. MRI follow-up. A. The mass is iso-and low signal intensity on axial T1-weighted image; hyperintense on T2-weighted image; with heterogeneous enhancement after contrast administration. B. The SWI shows multiple punctate signal voids within the mass, and the contents of it are hyperintense on DWI and bright on ADC. White arrow shows the lesion.

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