Calf Endometriosis: A Case Report and Review of Musculoskeletal Involvement

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It is not uncommon for women of childbearing age to have ectopic endometrial tissue (includes endometrial glands and stroma) in the pelvic genital organs. Less often, this versatile condition has been reported in many other unrelated extrapelvic sites. The extreme variability in presentation of endometriosis, in addition to the unusual location of extrapelvic endometriosis, can create a significant diagnostic challenge before definitive histologic examination. Even with high-technology magnetic resonance imaging (MRI) showing several radiopathologic aspects of endometriosis, differential diagnoses with similar radiologic presentations can result in an inaccurate diagnosis if precise attention is not given to helpful clues in the patient's history and clinicoradiologic correlation.

A few cases of endometriosis of the musculoskeletal system have been reported. In the present case report, we describe an endometriosis of the leg, presenting as a soft-tissue mass, and emphasize the importance of patient history and clinicoradiologic correlation for easier diagnosis. The authors have obtained the patient's written informed consent for print and electronic publication of the case report.

Case Report

A woman in her late 20s was referred to our orthopedic clinic because of a painful mass in her left leg. Her vague pain had not been accompanied by any systemic symptoms since its onset 2 years earlier. One year after pain onset, sensation of a gradually growing mass began. History of any related trauma was negative. The patient complained that the size of the mass and the intensity of the pain were fluctuating according to her menstrual cycles and were especially exacerbated during her menstrual period. Several months of nonsteroidal anti-inflammatory drug use and physiotherapy had no effect on the problem. Gynecologic history consisted of a normal vaginal delivery 9 years earlier, no infertility, no abortion, and irregular and painful menstrual cycles since menarche at age 12. Menstrual cycles became regular for 6 years after delivery, with use of oral contraceptives, but the irregularity then returned. Past medical history and family history were nonsignificant.

Physical examination revealed only a soft, mobile, tender, 3×4-cm mass in the posteromedial aspect of the middle third of the left leg. Range of motion of adjacent joints was normal, and there was no sign of skin changes over the mass or of neurovascular involvement.

Laboratory data were in the normal ranges. Plain radiographs did not provide any useful clues, such as soft-tissue calcification or bone involvement. MRI was reviewed by 2 radiologists independently, and different probable diagnoses were suggested. Both radiologists reported a nearly well-circumscribed 35×30-mm mass in the middle portion of the soleus muscle in the middle third of the

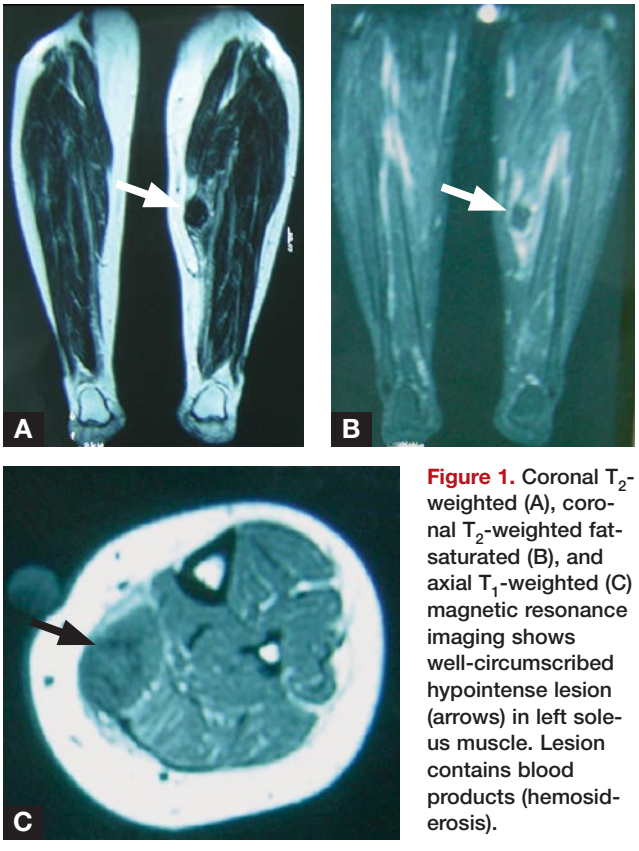

Figure 1. Coronal T_2-weighted (A), coronal T_2-weighted fat-saturated (B), and axial T_1-weighted (C) magnetic resonance imaging shows well-circumscribed hypointense lesion (arrows) in left soleus muscle. Lesion contains blood products (hemosiderosis).

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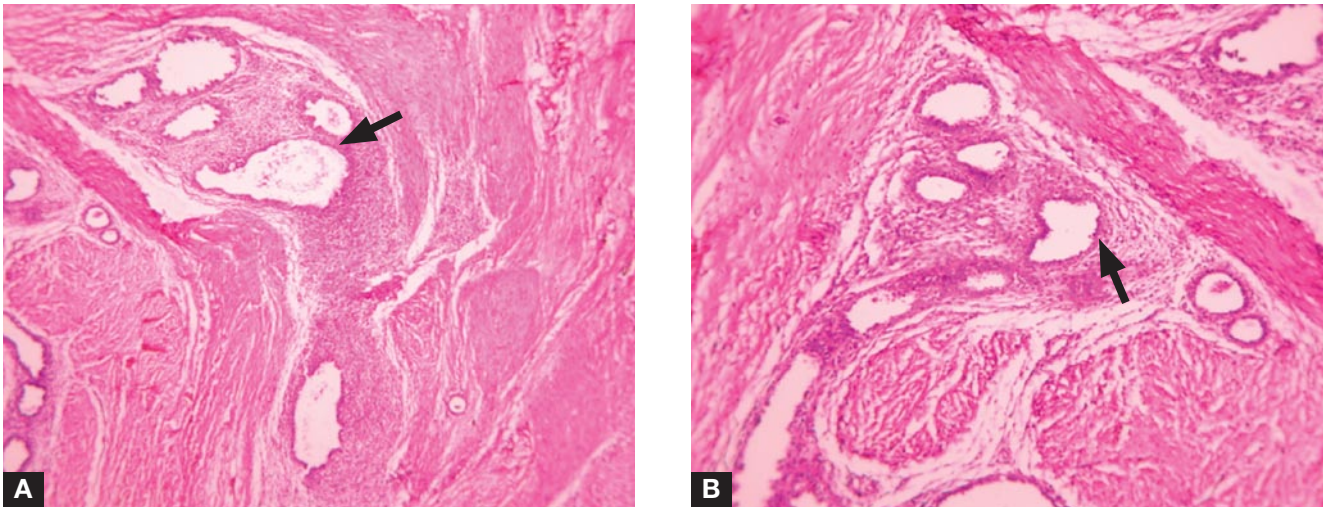

Figure 2. In lesion, glands (arrows) among stroma are lined with normal epithelium. Hematoxylin-eosin stain, original magnification (A) ×10 and (B) ×40.

left leg. The majority of the mass was hypointense on T_1- and T_2-weighted sequences, and no extension to bone or neurovascular structures was detected. Fat-suppressed images showed the hypointense mass with a surrounding hyperintense area of edema (Figure 1). One of the radiologists related these MRI findings to a desmoid tumor or nodular fasciitis, and the other, who might have paid more attention to the patient's history, thought the mass might contain hemosiderin or a large proportion of fibrous tissue and therefore suggested endometrioma, hematoma (both of which correlated with the patient's history), desmoid tumor, malignant fibrous histiocytoma, and fibrosarcoma as differential diagnoses.

Through a posteromedial approach, excisional biopsy was performed. The mass had no obvious capsule and was located under the medial head of the gastrocnemius in the soleus muscle. It was detached from the intact fascia and removed along with a small margin of normal muscle.

Grossly, the cut of ill-defined mass showed a tan-white trabecular structure firmly connected to the surrounding muscle. Microscopic examination revealed a fibrotic background and differently sized glandular and cystic structures lined with flat to cylindrical epithelium. Stroma contained a few spindle-shaped cells surrounding these glands. In addition, hemosiderin deposits were seen in several areas (Figure 2). Given the presence of endometrial glands, stroma, and hemosiderin deposits, the ward of Pathology considered endometriosis to be the definitive diagnosis.

At the follow-up visit, 1 week after removal of the mass, the patient was symptom-free. Subsequently, she reported no similar symptoms during her menstrual periods.

Discussion

Endometriosis, classically defined as the presence of functional endometrial glands and stroma outside the uterine cavity and musculature, commonly occurs in pelvic genital organs, especially the ovary, and primarily affects women of reproductive age (mean age at diagnosis, 25-29 years).[1,2] As accurate diagnosis of the most common pelvic type requires laparoscopy (the gold standard for definitive diagnosis), determination of actual prevalence of this condition is difficult. Therefore, subclinical disease is probably frequent.[1,3] The overall incidence of endometriosis is estimated to be 5% to 10% in women of childbearing age,[1,4,5] but most studies have found only a small proportion of cases in extrapelvic sites.[1,5-11] Known risk factors for developing endometriosis include positive family history, short menstrual cycles with prolonged flow, and obstruction of normal menstrual flow.[1] Endometriosis is not exclusive to women of reproductive age; it is found in premenarche and postmenopausal females and even in men who have received high-dose estrogens for prostatic carcinoma.[12-16]

Case reports and some retrospective series have described endometriosis in most organ systems, including the gastrointestinal tract, urinary tract, pleura and parenchyma of pulmonary system, abdominal wall, external genital tract, musculoskeletal system, and central and peripheral nervous system.[4,6,16-18] Because of extremely variable and nonspecific presentations, such as pelvic pain, menstrual-related pain, and infertility, even pelvic endometriosis can be sufficiently enigmatic, so when this versatile condition occurs in unusual sites with more nonspecific presentations, it is a significant diagnostic challenge, or it is recognized only retrospectively on the basis of histopathologic examination.

Endometriosis of the musculoskeletal system, though rare, has been reported in the pubis, buttocks (gluteus minimus muscle), thigh, knee, shoulder, elbow, forearm, hand, sciatic and obturator nerves, and femoral vein at the saphenous opening.[19-34] Most cases of extremity endometriosis occur in the groin or anteromedial aspect of the thigh,[24,29] probably by progression of pelvic endometriosis down the round ligament and into the inguinal canal; however, confirmation of pelvic endometriosis has not consistently been made.[9] In all cases, diagnosis was based on histologic appearance of tissue samples, and patients had local pain as the chief complaint. The presenting symptom of mus-

culoskeletal types, interestingly, has always been a type of cyclic pain that may be the most helpful clue for considering endometriosis as one of the most probable diagnoses.[4]

Endometriosis is confirmed histologically with identification of 2 of 3 features: endometrial glands, stroma, and hemosiderin pigment.[35] It is unclear whether both glands and stroma are required for the pathologic definition of the disease.[1]

The 4 theories for pathogenesis of endometriosis are metastatic, metaplastic (metaplastic differentiation of serosal surface and müllerian remnant tissue), induction (combination of first 2), and altered immunity. Each theory has its weak and strong points justifying aspects of this incompletely understood condition. Although more studies are needed, endometriosis can use all these mechanisms for development.[1] Up to 90% of women have bloody peritoneal fluid during the premenstrual period (retrograde menstruation), and the capacity of cast-off endometrial cells to remain viable and capable of implantation has been partially demonstrated.[1,36] On the basis of metastatic theory (the most probable pathogenesis), uterine endometrium can be transplanted to ectopic locations by many mechanisms, including lymphatic, vascular, and iatrogenic dissemination and retrograde menstruation.[1]

Endometriosis has a wide spectrum of pathology, from implants composed of variable levels of inflammation, hemorrhage (at different stages), and fibrosis to endometriotic cysts (endometriomas) with a thick, fibrotic wall caused by repeated cyclic hemorrhage within a deep implant. Implants and cysts may change in appearance during the menstrual cycle, becoming more swollen and congested with menses. The amount of pigment (hemosiderin, hemofuscin) in the implant appears to increase with age of lesion.[37]

MRI can show various histopathologic aspects of endometriosis by several types of signal-intensity change on different image sequences; however, most previous studies have concentrated on pelvic endometriosis. Depending on lesion age, cystic or solid mass, and associated blood products, MRI appearance varies significantly. Characteristically, it is homogeneously hyperintense on T_1-weighted sequences with relatively low signal intensity on T_2-weighted sequences, ranging from faint, dependent layering to complete signal void, reflecting the concentration of blood products. Alternatively, low signal intensity on T_1- and T_2-weighted images can result from hemosiderin-laden macrophages combined with the fibrous nature of the cyst wall. Some lesions are heterogeneous in signal intensity because the blood products are in various stages of degradation after multiple episodes of bleeding. MRI characteristics of solid endometriosis have been described as low to intermediate in signal intensity, with punctuate regions of high signal intensity on T_1-weighted images, uniform low signal intensity on T_2-weighted images, and enhancement corresponding to the abundant fibrous tissue seen in these lesions on histologic examination.[37,38]

Most soft-tissue masses have high signal intensity on T_2-weighted images. Soft-tissue masses with low signal intensity on T_2-weighted images include neurofibroma, cicatricial fibroma, malignant fibrous histiocytoma, aggressive fibromatosis, and calcified masses (myositis ossificans, extraskeletal osteosarcoma, or chondroblastoma, and synovial sarcoma).[39] Endometriosis can be added to this list, as it can present with low signal on T_2-weighted MRI images. In our case, low signal intensity on T_1- and T_2-weighted MRI sequences is in accord with the histologic findings of fibrotic background, blood products, and hypocellularity.

Conclusions

Because of varying MRI presentations of endometriosis and a long list of differential diagnoses with similar imaging changes in musculoskeletal forms, reliance on MRI characteristics can only be misleading. However, patients having menstrual-related symptoms and signs in all previous reports of musculoskeletal endometriosis and in our case might indicate that careful and comprehensive history taking and physical examination constitute the most reliable tool for correct diagnosis. As development of endometriosis is probable in women, particularly women of childbearing age, it is judicious to consider this diagnosis for each mass found in a woman and to look for the clues of cyclic changes of symptoms during menstrual period and abnormality of menstrual cycle.

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