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Report

Magnetic Resonance Imaging Features of Inflammatory Myofibroblastic Tumor of the Uterus

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We describe the case of a 63-year-old woman with a rare uterine inflammatory myofibroblastic tumor. Magnetic resonance imaging revealed a heterogeneous mass of increased T_2 signal and poor gadolinium enhancement in the enlarged uterine corpus, surrounded by diffusely increased T_2 signal and gadolinium enhancement in the myometrium. Following antibiotic therapy for suspected abscess, the tumor appeared smaller, decreased T_2 signal, and increased gadolinium enhancement on magnetic resonance imaging. Thereafter, the patient requested to undergo total hysterectomy. The predominant pattern of the present tumor may be altered from the myxoid-vascular to compact spindle cell subtype by the medical therapy according to magnetic resonance features reported on extra-uterine inflammatory myofibroblastic tumors, although only the latter subtype was histogically confirmed. We correlate radiologic and pathologic findings in this tumor.

Key words: inflammatory myofibroblastic tumor, magnetic resonance imaging, uterus

Introduction

Inflammatory myofibroblastic tumors (IMTs) are uncommon mesenchymal neoplasms that contain varying amounts of spindle cells, myofibroblasts, plasma cells, and histiocytes. IMTs have been reported in diverse organs, but are overwhelmingly rare in the uterus. We report the magnetic resonance (MR) imaging features of such a tumor.

Case Report

A 63-year-old woman was admitted to our hospital with a several-week history of general fatigue, weight loss, and lower abdominal pain. On admission, she was febrile, and an area of tenderness was palpated in the lower abdomen. Laboratory tests showed slight anemia (hemoglobin 10.8 g/dl), leukocytosis (16,100 /μL), elevated C-reactive protein (10.4 mg/dl), and thrombocytosis (534,000 /μL). MR imaging revealed a heterogeneous mass of increased T_2 signal and poor gadolinium enhancement in the enlarged uterine corpus, surrounded by diffusely increased T_2 signal and gadolinium enhancement in the myometrium (Fig. 1a–b). In addition, ill-defined focal low T_2 signal, indicating adenomyosis, was identified in posterior wall of the corpus. Her symptoms and abnormal laboratory data improved after a week of intravenous antibiotic therapy for suspected abscess. MR imaging after 19 days demonstrated that the mass was smaller, and there were slightly decreased T_2 signal and intense gadolinium enhancement throughout, whereas the remainder of the uterine corpus was almost normalized (Fig. 1c–d). However, the patient requested total hysterectomy and bilateral salpingo-oophorectomy to prevent the recurrent disease.

Grossly, the mass was soft, yellowish, and adhesive (Fig. 2). Microscopically, spindle cells proliferated, and chronic inflammatory infiltrate rich in lymphocytes and foamy macrophages was mixed, with no significant atypia (Fig. 3a). Immunohistochemical studies demonstrated spindle cells to be

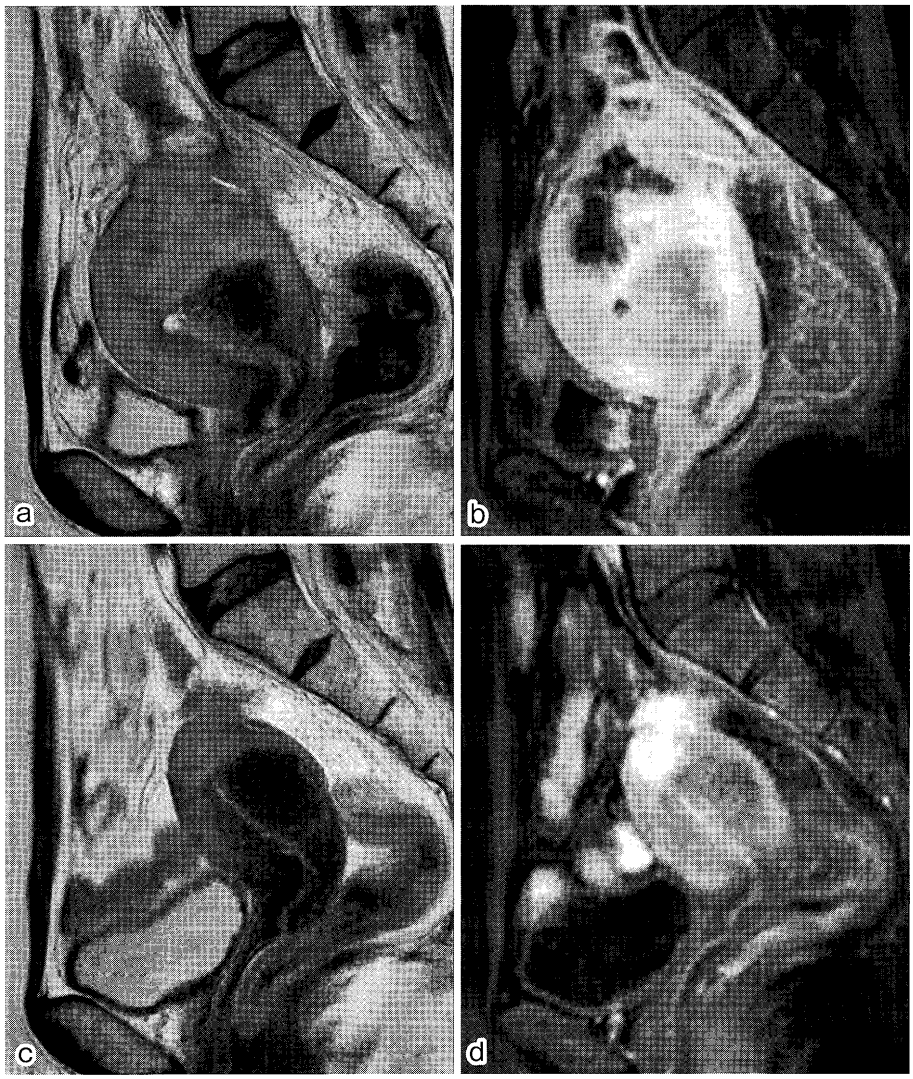

Fig. 1 Magnetic resonance sagittal images of a 63-year-old woman with the uterine inflammatory myofibroblastic tumor (a-b) before and (c-d) after medical treatment.
a, c: T_2-weighted image b, d: T_1-weighted fat-suppressed image after gadolinium administration.
a: Diffused heterogeneously increased intensity in the enlarged uterine corpus; focal low intensity is histologically confirmed adenomyosis in posterior wall of the corpus.
b: Poorly enhanced areas surrounded by the diffusely enhanced corpus.
c: Ill-defined mass at the uterine fundus demonstrating slightly low to intermediate intensity.
d: Homogeneously enhanced mass at the fundus.

positive for smooth-muscle actin and negative for desmin (Fig. 3b). Macrophages were strongly positive for CD68, and inflammatory cells were positive primarily for MIB-1. Consequently, the tumor was diagnosed as uterine IMT, consisting primarily of the compact spindle cell subtype.

Discussion

IMTs are uncommon neoplasms composed of mesenchymal cells that show differentiation toward myofibroblasts or fibroblasts, and such cells are associated with infiltration by inflammatory cells. The pathogenesis of this tumor remains a matter of debate. Some cases have been associated with malignancy or tuberculosis as satellite lesions. The multiplicity of sites that can be involved suggests no particular route of entry or any specific agents. Prior surgery, trauma, or immune disturbances, in addition to infection are included for the possible etiol-

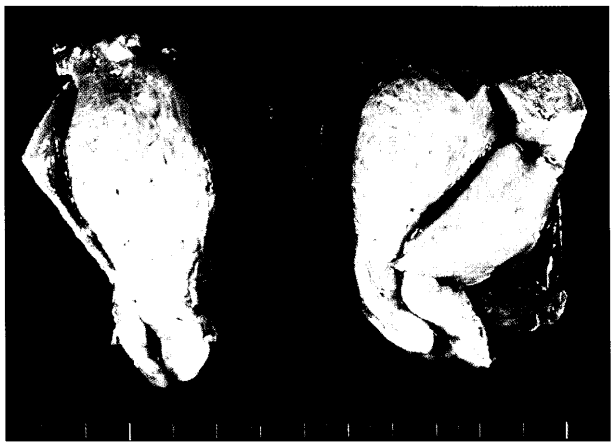

Fig. 2 Gross photograph of the resected uterus
A soft and yellowish mass protrudes at the uterine fundus.

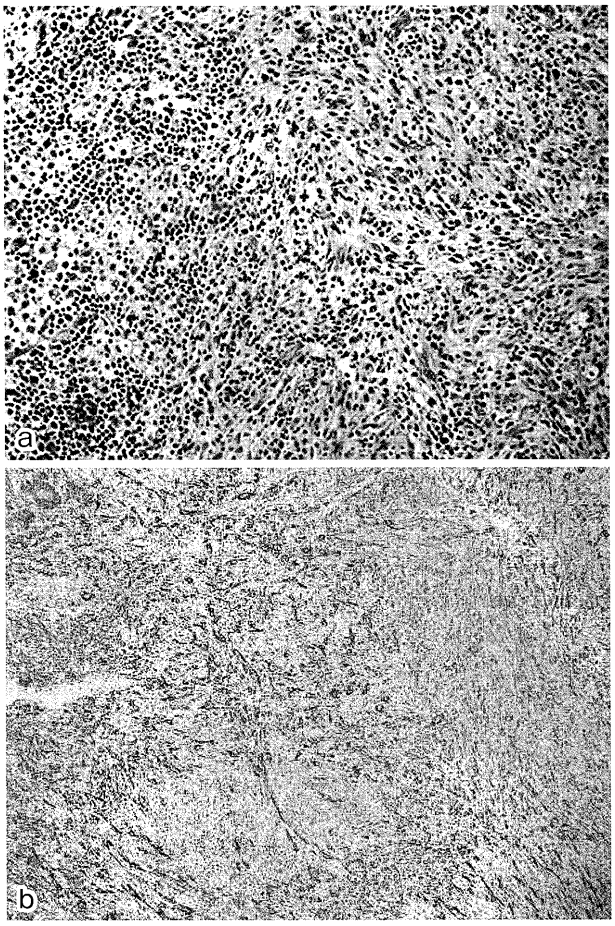

Fig. 3 Surgical photomicrograph of the uterine inflammatory myofibroblastic tumor
a: hematoxylin-eosin stain, b: immunohistochemical smooth-muscle actin stain
A predominance of the compact spindle cell subtype, positive for smooth-muscle actin.

ogy[1]. The clinical onset may be insidious or rapid and is accompanied by constitutional symptoms of fever, weight loss, and a variety of laboratory abnormalities, which include acute-phase reaction, thrombocytosis, anemia, and elevated sedimentation ratio. These symptoms, also recognized in our patient, probably result from an overproduction of interleukin-6 in approximately 15% to 30% of cases[2)3)]. Although the majority of IMTs are benign, adequate surgical removal is required because local recurrence is common. Furthermore, though cases of metastasis or major vascular invasion have been described, so have patients whose tumors disappeared both spontaneously and after medical treatment[4]. Originally described in the lung and previously referred to as inflammatory pseudotumor, IMTs are known to arise in various organs[5]. Uterine localization is, however, overwhelmingly rare, and to our knowledge, only 11 cases have been reported. Only one report describes the MR characteristics of uterine IMT, and the tumor in that report was depicted with prominent myxoid change so that the unique features for such a neoplasm were obscured by marked degeneration[6]. Ours is practically the first report regarding the MR imaging characteristics of uterine IMT.

Three basic histologic patterns of IMTs have been recognized, those with (a) myxoid, vascular, and inflammatory areas; with (b) compact spindle cells intermingled with inflammatory cells; and with (c) dense plate-like collagen[7]. Although IMTs have no definitive characteristics radiologically, the unique predominant histologic pattern may influence the MR imaging characteristics of these tumors. This may help explain the change in MR appearance of our patient's tumor after antibiotic therapy. T_2 signal intensity has been reported to depend on the quantity of cellular material relative to fibrous tissue and may decrease as cells and water in the mass are reduced[4]. Seol and colleagues described that the area showing abundant inflammatory cells in vascular stroma could be hyperintensity on T_2-weighted images; in contrast, the paucicellular area in collagenous stroma might be isointensity or low signal intensity on the images[8]. Furthermore, marked fibrosis could be responsible for

hypovascularity[9]. Horger and colleagues reported that a tumor of the predominantly compact spindle-cell pattern showed relatively low T_2 signal and high gadolinium enhancement, whereas a tumor of the myxoid-vascular subtype demonstrated high T_2 signal and less gadolinium enhancement[10]. In our case, the MR features before medical treatment resembled those of a tumor of the myxoid-vascular subtype that was reported by Horger's group. However, inflammation improved with antibiotic therapy, and MR findings after this treatment were similar to those of the compact spindle cell subtype. Thereafter, our patient underwent hysterectomy, and surgical pathology confirmed uterine IMT of the compact spindle cell subtype. Accordingly, MR interpretation after medical treatment correlated well with this histological conclusion. There is evidence that IMTs evolve toward the dense collagen type, with a decrease in cellular content and an increase in fibrosis[4)9)]. Therefore, the predominant pattern of our case in MR findings may be radiologically altered from the myxoid-vascular to compact spindle cell subtype by medical therapy, although the former subtype was not histologically confirmed.

Conclusion

Although the MR characteristics of rare uterine IMTs have been scarcely investigated, improvement of our patient's tumor and its MR appearance following medical therapy correlate well with previously reported radiologic and pathologic findings. Thus, knowledge of such MR characteristics is useful for proper patient management as well as accurate diagnosis.

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子宮に発生した炎症性筋線維芽細胞性腫瘍のMRI所見

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炎症性筋線維芽細胞性腫瘍は，筋線維芽細胞や線維芽細胞に分化する細胞を含み，炎症細胞の浸潤が加わった稀な腫瘍である．様々な臓器に発生しうるが，子宮における報告は極めて稀である．今回我々は，組織学的に診断された子宮炎症性筋線維芽細胞性腫瘍の症例を経験したのでMRI所見を中心に報告する．

症例は63歳女性．全身倦怠感，発熱，体重減少および下腹部痛を主訴に精査目的にて当院産婦人科に入院となった．MRIで腫大した子宮体部にT2強調像で不均一な信号上昇を示す造影効果の低い腫瘤を認め，周囲の子宮体部筋層は瀰漫性に高信号を示し，強い造影効果を呈し，膿瘍形成が疑われた．抗生剤治療により臨床症状の改善を認めた．再度MRIが施行され，子宮体部腫大は改善，子宮体部腫瘍も縮小し，T2強調像ではやや信号が低下し，ほぼ均一で強い造影効果を示すように所見の変化を認めた．しかし，患者の強い希望により，再発予防目的で子宮全摘術および両側子宮付属器切除術が施行された．病理組織学的にはcompact spindle cell subtypeの子宮炎症性筋線維芽細胞性腫瘍と診断された．

子宮炎症性筋線維芽細胞性腫瘍は良性病変であるものの再発の可能性が高く，転移や血管浸潤も報告されており，外科的切除の適応がある．子宮病変の報告は11例しかなく，中でもMRIにおける画像所見を報告したものは1例のみである．抗生剤治療に反応する特徴的な臨床経過とMRIを中心とした画像診断により本症の術前診断の可能性が示唆される．