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Pseudo-Meigs Syndrome Due to Subserosal Leiomyoma Diagnosed by MR Imaging: Case Report[1]

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We report a case of pseudo-Meigs syndrome due to a large subserosal leiomyoma in a patient with a high serum carcinogenic antigen 125 level. Initial clinical examination suggested disseminated malignant disease though the typical signal characteristics of leiomyoma, seen at MR imaging, led to the diagnosis of pseudo-Meigs syndrome.

Index words : Leiomyoma
Magnetic resonance (MR)

The clinical appearance of benign neoplasms of gynecologic origin is sometimes similar to that of disseminated malignant neoplasms (1). This is usually the case in patients with Meigs or pseudo-Meigs syndrome. Meigs syndrome was defined in 1954 as a fibroma or fibroma-like tumor associated with hydrothorax and ascites, which resolved on removal of the tumor. Pseudo-Meigs syndrome has an identical clinical constellation but the tumor types which occur were not initially described by Meigs (1 - 3). Among such cases, uterine leiomyomas with associated ascites and hydrothorax are particularly rare and although serum levels of carcinogenic antigen (CA)-125 are as elevated in these patients as in malignant disease, reports have stated that the effusions disappeared spontaneously after removal of the leiomyoma, and the patient was cured (3 - 5). We report a case of pseudo-Meigs syndrome due to a large subserosal leiomyoma in a patient with a high serum CA-125 level. Clinical examination at first suggested disseminated malignant disease, but MR imaging showed that the mass had the typical appearance of uterine leiomyoma, with ascites and pleural effusion but no evidence of malignancy.

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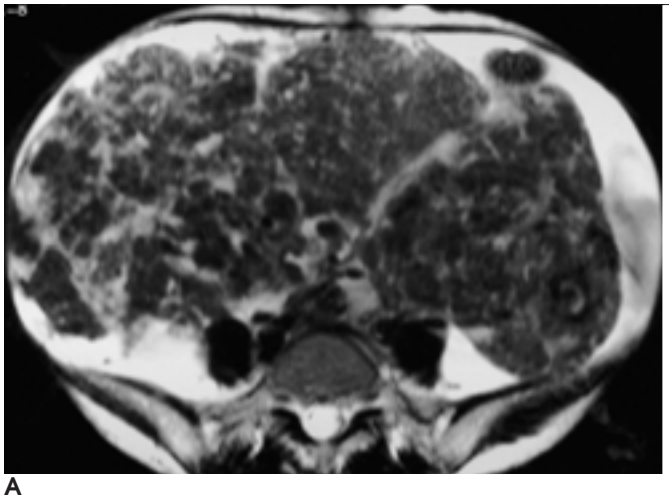
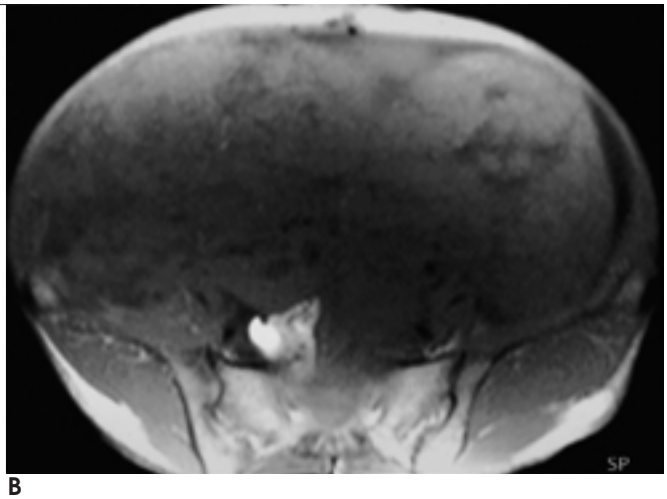
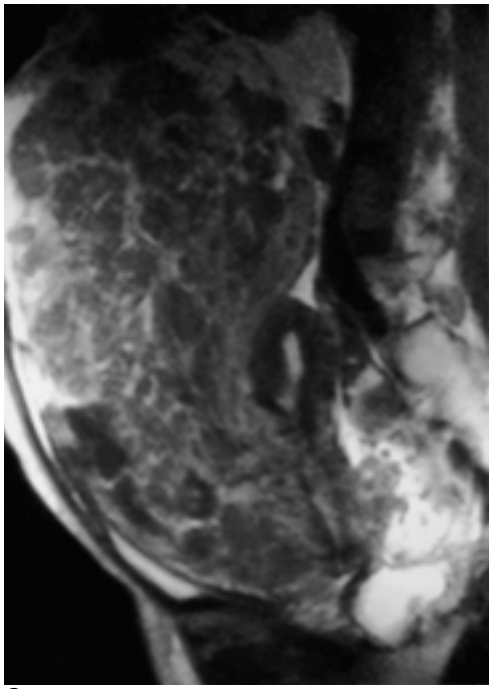
Case Report

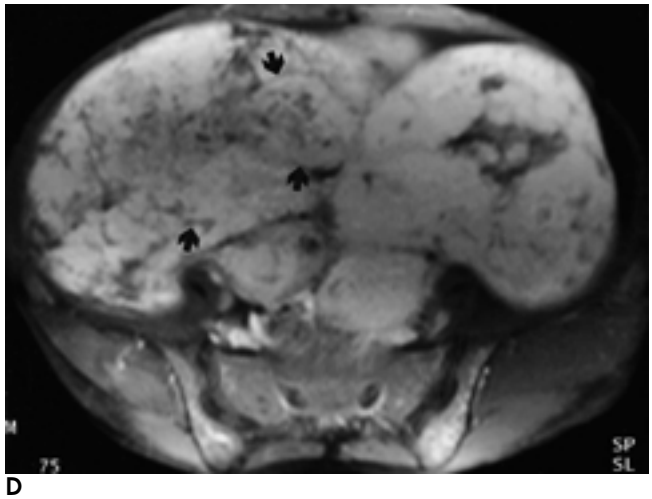
A 38-year-old woman presented with a low abdominal mass, first apparent several months earlier. Four years prior to this she had undergone right salpingo-oorphorectomy due to an ovarian cyst. Pelvic examination revealed a hard, mobile, tender mass about 20cm in diameter and occupying the pelvis and lower abdomen. Lung auscultation and plain X-ray examination of the thorax indicated that right-side pleural effusion was efficient, and except for a CA-125 level of 520 (normal range 0 - 35U/ml) U/ml, laboratory tumor markers were within normal limits. The patient underwent pleural aspiration, together with abdominal paracentesis, and the aspirated yellowish pleural fluid contained inflammatory cells and some red cells. Microbiological culture was negative. The ascitic fluid contained some macrophages and lymphocytes.

Transvaginal sonography showed that a large bilobulated mass with irregular margins surrounded the uterus. Free peritoneal fluid was seen, while the left adnexa showed no abnormalities. Magnetic resonance (MR) images obtained using a 1.5 T unit (Vision Plus, Siemens, Erlangen, Germany) depicted a large, hetero-









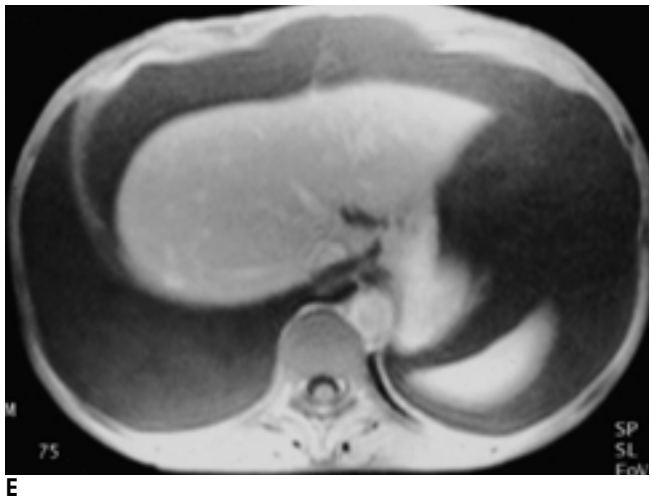


Fig. 1. A 38-year-old woman with pseudo-Meigs' syndrome due to subserosal leiomyoma.

A, B. Axial T2-weighted (TR/TE, 4275/138) (**A**) and corresponding T1-weighted (147/4.8) (**B**) turbo spin-echo MR image show a large, heterogeneous pelvic mass. Most of the mass is of low to intermediate signal intensity on the T2-weighted image (**A**) and isointense to muscle, an appearance suggestive of a leiomyoma.

C. Sagittal T2-weighted turbo spin-echo MR image (4.4/90) demonstrates continuity of the mass with the uterine fundus.

D. Contrast-enhanced fat-suppressed T1-weighted gradient-echo MR image (147/4.8) shows diffuse contrast enhancement of subserosal leiomyoma. Multiple branching linear structures of high signal intensity on T2-weighted image (**A, C**), low signal intensity on T1-weighted image (**B**) and no enhancement on the contrast-enhanced image (**D**) represent edema (arrows).

E. Contrast-enhanced fat-suppressed T1-weighted gradient-echo MR image (147/4.8) at the level of the upper abdomen discloses massive ascites and right pleural effusion.

geneous pelvic mass. This was mostly of low signal intensity at T2-weighted turbo spin-echo imaging (TR/TE, 4275/138) and of intermediate signal intensity identical to that of muscle at T1-weighted FLASH imaging (147/4.8), an appearance suggestive of leiomyoma (Figs. 1A, B). Sagittal T2-weighted HASTE (4.4/90) demonstrated that the mass and the lower uterine body were continous (Fig. 1C). The mass contained multiple branching linear structures, presenting as high signal intensity at T2-weighted imaging and low signal intensity at T1-weighted imaging. After gadolinium injection, diffuse enhancement, to the same extent as that of adjacent myometrium was observed (Fig. 1D), though the branching linear structures were not enhanced. Massive ascites and right hydrothorax was identified, but there was no evidence of loculation, pleural or peritoneal thickening, or nodularity (Fig. 1E).

Exploratory laparotomy revealed 300 ml of serous ascites and a large, bilobate, pedunculated mass, smooth, firm and pale gray in appearance, protruding from the uterine corpus. The patient underwent total hysterectomy and left salpingo-oorphorectomy. There was no palpable pelvic or periaortic adenopathy, and grossly the liver, diaphragm, bowel, and omentum were free of disease. The mass measured about 22×14 cm, and sectioning revealed that its cut surface was whorled. Hyaline degeneration had given rise to weeping. The final diagnosis reached on the basis of the low mitotic rate, minimal cytologic atypia, and lack of coagulative tumor cell necrosis, was leiomyoma with hyaline degeneration and interstitial edema.

The patient's postoperative course was uncomplicated. Repeated chest radiographs, obtained four months later, indicated a complete absence of pleural effusion.

Discussion

Meigs syndrome was first reported in a patient with ovarian fibroma accompanied by ascites and pleural effusion, which resolved after resection of the tumor. However, some later cases of the syndrome occurred in patients with ovarian tumors other than fibroma, or tumors of the uterus and fallopian tubes, and the condition was then referred to as pseudo-Meigs syndrome (2). The majority of such cases have involved pathologic conditions affecting the ovaries, or rarely, the uterus (3). Seventeen such reports document uterine leiomyomas as the cause of pseudo-Meigs syndrome.

The etiology of ascites is subject to much debate and still remains unclear (4). A plausible theory is that pressure on the lymphatic vessels in the tumor itself causes the escape of fluid through lymphatic surface. A discrepancy between the arterial blood supply to a large mass of tumor tissue and its venous and lymphatic drainage may also lead to stromal edema and transudation of fluid into the peritoneal space (5).

CA-125 is an antigenic determinant on a high molecular-weighted glycogen that is expressed by epithelial ovarian tumors and others of normal or abnormal Mullerian origin (2). It has been demonstrated that increased serum elevation of CA-125 levels in patients with Meigs or pseudo-Meigs syndrome is caused by mesothelial expression of CA-125 (8).

MR imaging is currently considered as the most accurate imaging technique for the detection and localization of leiomyomas (6). Because of its ability to clearly demonstrate individual tumors, the modality has been shown to be more sensitive than US, with which accurate assessment of an enlarged, myomatous uterus (> 140 cm^3) is not consistently possible because of the limited field of view (6). At T2-weighted imaging, leiomyomas typically demonstrate distinct low signal intensity relative to that of the myometrium, and at T1-weighted imaging, intermediate signal intensity. These characteristic intensities are attributed to extensive hyalinization, which occurs in more than 60% of uterine leiomyomas (7), though various histopathologic patterns of degeneration, some of which alter the MR imaging appearance, may be apparent (8). In our case, although the leiomyoma was huge, its signal intensity was typical of a leiomyoma with hyaline degeneration. The high signal intensity linear branching structures seen at T2-weighted imaging represent edema, the presence of which affects the signal intensity of leiomyomas and may antedate hyalinization and cystic degeneration (8).

For patients with a huge pelvic mass and elevated CA-125 levels, a diagnosis of malignancy is an inevitable possibility, and when ascites and pleural effusion are present, the possibility of metastatic disease will always be considered. In these cases, the typical signal characteristics of leiomyoma at MR imaging suggest a possible diagnosis of pseudo-Meigs syndrome.

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자기공명영상으로 진단된 장막하 자궁근종에 의한 Pseudo-Meigs 증후군: 증례 보고[1]

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저자들은 골반 종양 환자에서 동반된 복수와 흉수 그리고 혈청 CA-125의 수치의 증가로 임상적으로 악성양상을 보였으나, 자기공명영상으로 자궁근종의 특징적인 신호강도를 보여 진단 가능하였던 pseudo-Meigs 증후군을 보고하고자 한다.