CASE REPORT

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Simultaneous Colonic Obstruction and Hydroureteronephrosis due to Mesenteric Fibromatosis

Sung Hoon Jung*, Chang Nyol Paik*, Ji Han Jung†, Kang-Moon Lee*, Woo Chul Chung*, and Jin-Mo Yang*

*Departments of *Internal Medicine and †Pathology, College Medicine, The Catholic University of Korea, Seoul, Korea*

Mesenteric fibromatosis (MF) is a rare benign mesenchymal lesion that can occur throughout the gastrointestinal tract, especially small bowel. Its biological behavior is intermediate between benign fibrous tissue proliferation and malignant fibrosarcoma. In previously reported cases of MF, we could find colonic obstruction or ureter obstruction, but simultaneous involvement of colon and ureter was not able to be seen. We described a patient that presented with colonic obstruction and hydroureteronephrosis due to MF at sigmoid colon which mimicked submucosal tumor such as gastrointestinal tumor. This case resulted in a positive positron emission tomography scan suggesting malignant neoplasm, but β-catenin positivity on immunohistochemical staining separated MF from gastrointestinal stromal tumor and sclerosing mesenteritis. The clinical course of the patient was improved after surgical resection. **(Gut and Liver 2009;3:215-217)**

Key Words: Mesenteric fibromatosis; Colonic obstruction; Hydroureteronephrosis

INTRODUCTION

Mesenteric fibromatosis (MF) is a rare, locally invasive, nonmetastasizing tumor. It can involve any site of gastrointestinal tract, but the most common site is the mesentery of the small bowel. Its biological behavior is intermediate between benign fibrous tissue proliferation and fibrosarcoma. Sometimes, it is often confused with submucosal tumor or malignant neoplasm of gastrointestinal tract, especially gastrointestinal stromal tumors (GISTs). We described a case of MF which induced obstruction of colon and ureter simultaneously and resulted in a positive positron emission tomography (PET) scan.

CASE REPORT

A 49-year-old woman was admitted for constipation and left lower quadrant abdominal pain for seven days. She had undergone a hysterectomy to treat uterine myoma, nine years ago. Physical examination revealed heperactive bowel sound and palpable mass at left lower quadrant of abdomen. Initial complete blood cell counts and blood chemistry results were as follows: white blood cell count, 8.2×10^9/L (normal, 4.5×10^9/L-10.5×10^9/L); hemoglobin, 12.7 g/dL; platelet count, 264×10^9/L; blood urea nitrogen, 24.4 mg/dL; creatinine, 1.1 mg/dL. The result of electrolytes and urinalysis were unremarkable. Sigmoidoscopy (Fig. 1) and barium enema (Fig. 2) showed near-complete obstructive mass at sigmoid colon covered with normal mucosa. Abdominal CT scan disclosed not only an about 5.6×5.2 cm sized well circumscribed, locally infiltrative soft tissue mass in left pelvic cavity, but also marked left hydroureteronephrosis and atrophic change of left kidney (Fig. 3), suggestive of longstanding ureteral obstruction. PET scan revealed the corresponding pelvic mass with focal increased activity with a maximum standardized uptake value (SUVmax) of 4.1 (Fig. 4). The patient underwent colon segmental resection with left oophorectomy and salpingectomy. The mass was measured 7.5×6.8 cm and involvement of resected margin was absent. Light microscopy shows ordely arrangement of uniform fibroblasts associated with moderate amouts of collagen (Fig. 5). Immunohistochemistry disclosed that

Correspondence to: Chang Nyol Paik
Department of Internal Medicine, College of Medicine, The Catholic University of Korea, St. Vincent's Hospital, 93-6, Ji-dong, Paldal-gu, Suwon 442-723, Korea
Tel: +82-31-249-7137, Fax: +82-31-253-8898, E-mail: cmcu@catholic.ac.kr
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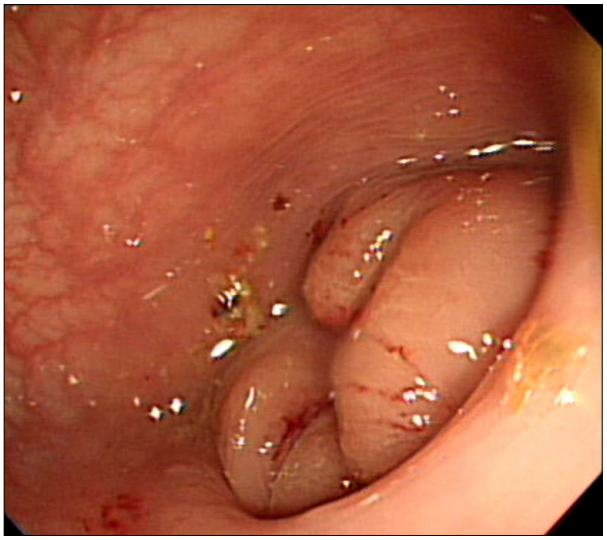

Fig. 1. Sigmoidoscopy showed that the sigmoid colon was almost totally obstructed by a mass covered with normal mucosa.

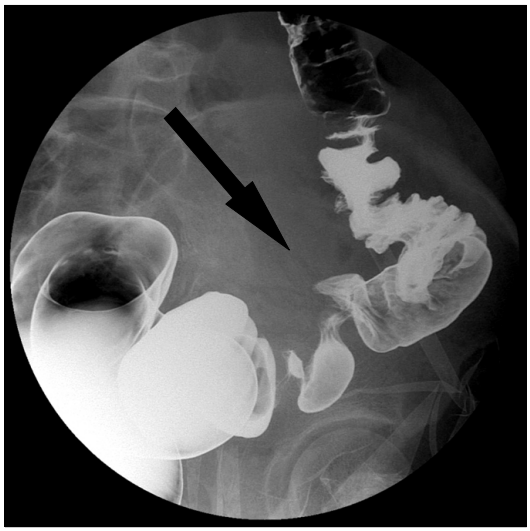

Fig. 2. A barium enema demonstrated the almost total obstruction of the sigmoid colon (arrow).

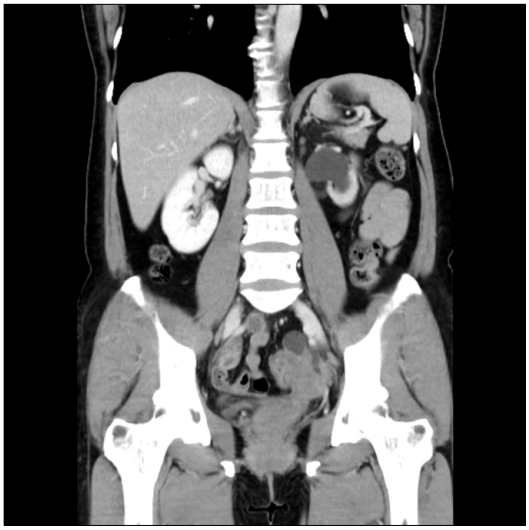

Fig. 3. Abdominal CT scanning disclosed a 5.6×5.2-cm, well-circumscribed, locally infiltrative soft-tissue mass in the left pelvic cavity, with left hydroureteronephrosis and atrophic changes in the left kidney.

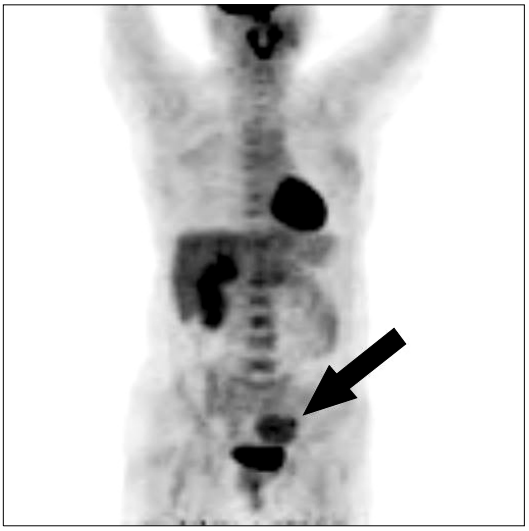

Fig. 4. A PET scan revealed a mass in the left pelvic cavity with focally increased activity (arrow).

the lesion were negative for CD34 and *C-kit*, whereas were positive for actin, vimentin. Especially immunohistochemical stain for β-catenin shows strong nuclear positivity (Fig. 6). The final pathologic diagnosis was MF. The patient was currently followed up in our outpatient department and was asymptomatic.

DISCUSSION

MF is a rare, benign fibrous lesion found in the bowel mesentery or the retroperitoneum. The mesentery of the small bowel is the most common site.[1] MF tend to be locally invasive and to recur locally, but do not metastasize.[1,2] It can occur spontaneously or after surgical trauma and also is associated with hormonal therapy, familiar polyposis, or Gardner's syndrome.[1,3] The diagnosis of MF is based on clinical suspicion, which depends on the location or local effect of tumor. The role of imaging is to define the degree of extension to local structure and tumor relationship to neurovascular structure.[4,5] Pathologic confirmation was made by microscopic examination and immunohistochemistry. However, GISTs are potentially misdiagnosed as MF, because of their features such as large size, infiltration of adjacent structures and mitotic

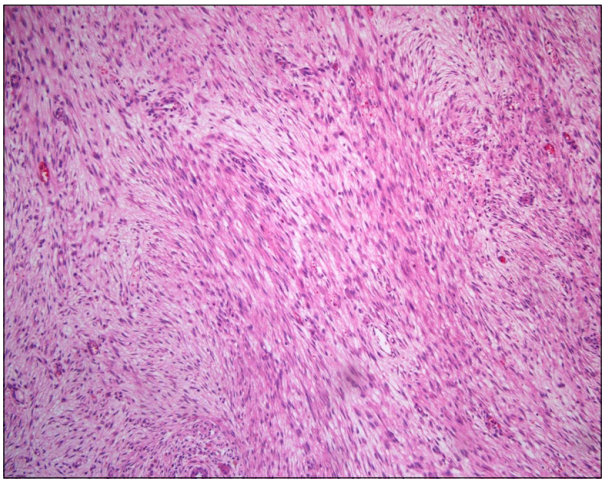

Fig. 5. Light micrograph showing the orderly, uniform arrangement of fibroblasts, which are associated with moderate amounts of collagen (×100).

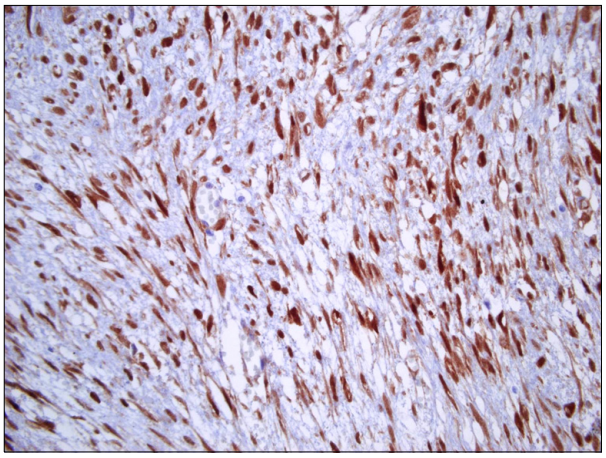

Fig. 6. Immunohistochemical stain for β-catenin; strong nuclear positivity can be observed (×400).

activity.[6] Their diagnostic discrimination is essential because of their very different biological behaviors and their therapeutic strategies.[7] The absence of CD34 and S100 expression supports the fibromatous nature of the lesion and may be helpful in discriminating MF from GIST.[8] In addition, recently a report showed that β-catenin separates MF from GIST and sclerosing mesenteritis.[9] Our case showed that the lesion were negative for CD34, whereas positive for β-catenin, which were compatible with fibromatosis. The preferred treatment of MF is wide local surgical excision with a margin of uninvolved tissue.[1,3] In the current case, unusually, the mass involved sigmoid colon and ureter at once and result in more aggressive features such as colonic obstruction with local invasion and hydroureteronephrosis. PET scan showed hot uptake in corresponding mass, considered as submucosal tumor of malignant neoplasm such as GIST. The operation was performed and the pathologic result was MF. We emphasize three points about MF through this case. First, however rare, MF is considered as one of the causes of stenosis of the colon in patients with a history of pelvic surgery. Second, PET scan is not helpful for differential diagnosis of other malignant neoplasm from this disease, which is correspond with recent report that MF has a positive interpretation of fluorodeoxyglucose PET scan.[10] Third, β-catenin is useful to differentiate between MF and GIST.

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