

Colonic Endometriosis with Extensive Smooth Muscle Metaplasia Simulating Leiomyoma

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Abstract

It is difficult to distinguish colonic endometriosis with smooth muscle metaplasia from a colonic neoplasm. Especially, in a small biopsy specimen, marked smooth muscle proliferation can be misdiagnosed as a spindle cell lesion such as leiomyoma or as a gastrointestinal stromal tumor. We present the case of a woman aged 48 years who had a colonic polyp, raising suspicion of a submucosal tumor. In an endoscopic polypectomy specimen, the lesion showed marked smooth muscle proliferation but in the resection specimen, a nodular mass, composed of endometrial glands, stroma, and a smooth muscle component, was identified. By immunohistochemical staining, smooth muscle cells were positive for estrogen receptor, progesterone receptor, smooth muscle actin, and smooth muscle myosin heavy chain. Endometriosis in the colon can be misinterpreted as a benign or malignant neoplasm. When examining small biopsy specimens, clinicians and pathologists should be aware of the potential of this condition to mimic other intestinal diseases.

Key Words : Colon, Endometriosis, Leiomyoma

Introduction

Endometriosis affects the intestinal tract in 15% to 37% of patients with pelvic endometriosis [1], but is difficult to distinguish from a benign or malignant neoplasm clinically [2-6]. Histologically,

endometriosis shows the endometrial glands and the stroma, and may be accompanied by smooth muscle hyperplasia in 24% of intestinal endometriosis patients [7]. Endometriosis in the intestinal wall can be misdiagnosed as a benign or malignant neoplasm [3], and especially

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endometriosis with marked smooth muscle proliferation (metaplasia) can be misidentified as a spindle cell neoplasm of the intestinal wall. Here, we present a case with a polypoid lesion of the colon, initially interpreted as a spindle cell neoplasm, that was finally diagnosed as colonic endometriosis.

Case Report

A woman aged 48 years visited our hospital for periodical health checks. In a colonoscopic examination, a 1.2 cm-sized submucosal tumor-like lesion was found 20 cm from the anal verge (Fig. 1A). Endoscopic snare polypectomy was performed. On histological examination, the submucosal tumor-like lesion was found to be a spindle cell lesion. The lesion consisted of smooth muscle cells and the resection margin was involved by smooth muscle component. On abdominal CT, a 1.5 cm-sized hyperattenuating lesion was found to remain present in the rectosigmoid area (Fig. 1B). The uterus showed slightly enlarged but no

abnormal shape, and both ovaries were unremarkable. A segmental resection was performed for removal of the smooth muscle proliferative lesion.

On gross examination, an ill-demarcated polypoid lesion ($2.1 \times 1.5 \times 1.0$ cm) was present in the sigmoid colon (Fig. 2). The cut surface showed a lobulating mass in a mucosal extension to the subserosa. The lesion was grayish tan, solid, and fibrotic, with a focal hemorrhagic area. Necrosis was not identified in the lesion.

Microscopically, the lesion was snowman-like shape and had a clear boundary. The lesion consisted of smooth muscle cells with a few endometrial glands and stroma. The smooth muscle component was more than 80% of total volume and endometrial glands and stroma were centrally scattered in smooth muscle component (Fig. 3A). By immunohistochemistry, staining for the estrogen receptor (ER) and progesterone receptor (PR) was positive for endometrial glands, stroma, and the smooth muscle proliferative lesion (Fig. 3B), but negative for colonic mucosa and normal muscle. The smooth muscle proliferative

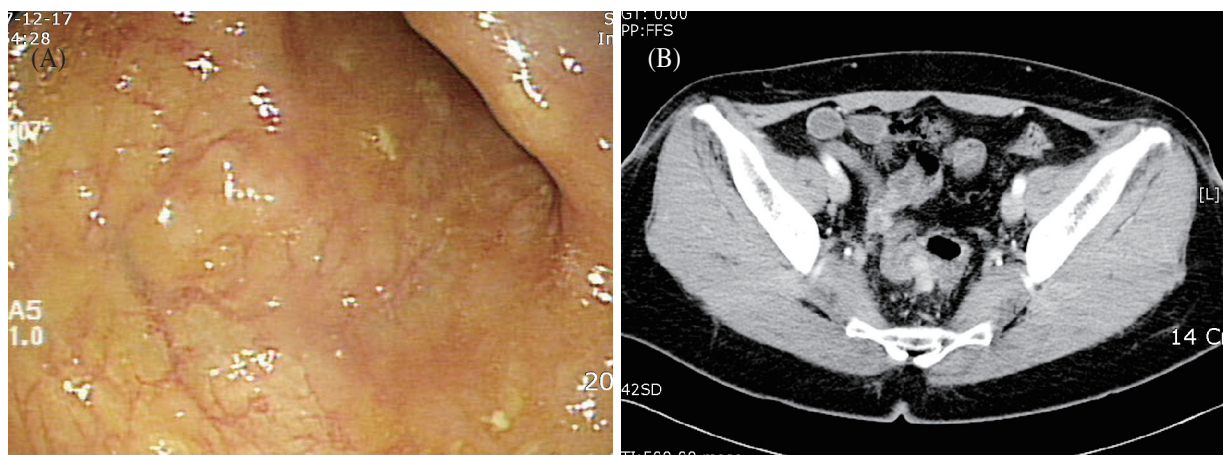


Fig. 1. (A) A 1.2 cm-sized submucosal tumor-like lesion was present in colonoscopic examination. (B) A 1.5 cm sized hyperattenuating lesion is remained in rectosigmoid area (Abdominal CT examination).

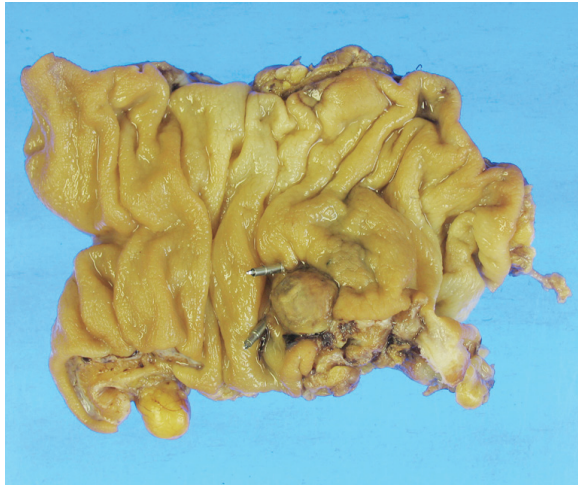


Fig. 2. A 1.5 cm-sized polypoid lesion was present in the colonic mucosa.

lesion stained positive for smooth muscle actin (SMA), desmin, and smooth muscle myosin heavy chain (SMMHC) (Fig. 2B). Immunohistochemical staining for C-kit, CD34, and S-100 proteins was negative, and staining for CD10 was positive on the endometrial stroma. The smooth muscle

proliferative lesion was distinguished from indigenous normal muscle, and showed hypercellularity, moderate nuclear atypia, and occasional mitoses.

The patient has had no evidence of post-operative complication and no evidence of recurrence for 6 months after surgery.

Discussion

Endometriosis with predominant smooth muscle proliferation or smooth muscle metaplasia can be misinterpreted as submucosal tumors on endoscopic examination [2,5,7-10]. On radiologic examination, endometriosis with smooth muscle metaplasia can appear as a submucosal spindle cell tumor of the intestinal wall, including a gastrointestinal stromal tumor or leiomyoma [5,7].

In the present case, the smooth muscle component was predominant, representing more than 80% of the endometrial mass. Only the

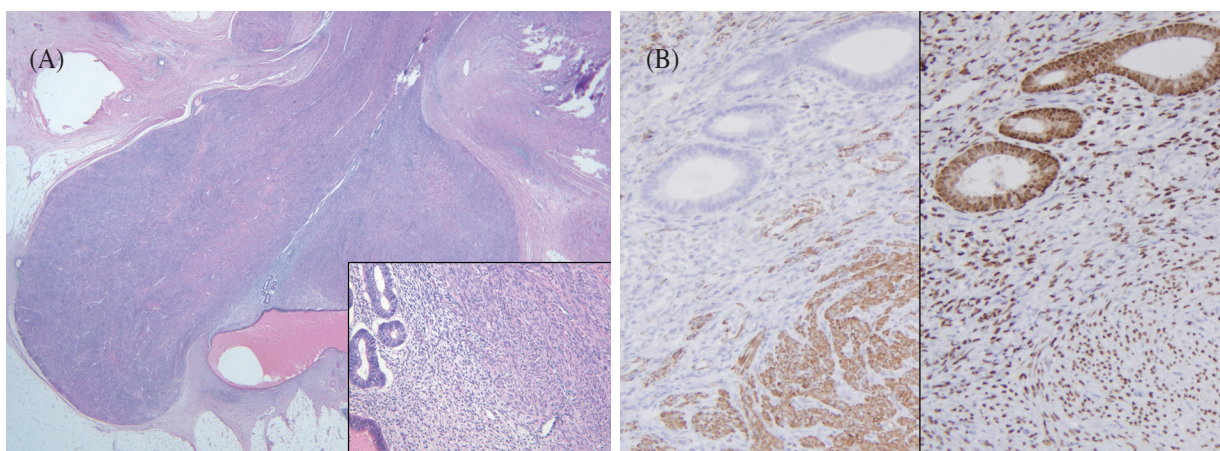


Fig. 3. (A) Some scattered endometrial gland and stromal tissue was identified in the indicated smooth muscle proliferative lesion (H&E, $\times 40$ and $\times 200$). (B) Endometrial glands, stroma, and smooth muscle cells were positive for estrogen receptor (left, Immunohistochemical stain for estrogen receptor, $\times 200$). The smooth muscle cells were positive, but endometrial gland and stromal cells were negative for smooth muscle myosin heavy chain (right, Immunohistochemical stain for smooth muscle actin, $\times 200$).

smooth muscle component was present in a small fragment obtained by endoscopic biopsy and resulted in misinterpretation of the condition as a spindle cell neoplasia that might be either a gastrointestinal stromal tumor or leiomyoma. If a few fragments of endometrial glands consisting relatively short columnar epithelium with prominent nucleoli or no mucinous components were identified, the biopsy lesion could be considered endometriosis with smooth muscle metaplasia of large intestine.

On immunohistochemistry, endometriosis components, such as endometrium and myometrium, are positive for ER and PR [8,9]. SMA, SMMHC, and desmin are expressed in the smooth muscle component, and ER and PR are also present [7].

Differential diagnoses include intravenous leiomyomatosis, intestinal leiomyoma with endometriosis, leiomyosarcoma and synovial sarcoma. In intravenous leiomyomatosis, the margin is irregular, and lacks an endometrial components. Intestinal leiomyoma with endometriosis does not show expression of either ER or PR in the smooth muscle component. PR expression can be present in leiomyoma with specific site, such as sinonasal area. However, both of ER and PR expression was not identified in leiomyoma except uterine origin [11]. Leiomyosarcoma have hypercellular lesion with marked cellular atypia and also lacks an endometrial components. Synovial sarcoma may have similar histologic features. Especially in biphasic type, epithelial and spindle cell components are similar to endometrial glands and smooth muscle component. However, synovial sarcoma does not show expression of SMA, SMMHC and desmin. In the present case, the endometrial glands and stroma were scattered in a nodular mass of smooth muscle, which could be distinguished from normal

indigenous muscle. Therefore, the smooth muscle fraction was an endometriosis component and the condition was consistent with smooth muscle metaplasia.

Endometriosis is commonly misinterpreted as a benign or malignant neoplasia because the condition can present with variable proportions of components [12]. Especially, endometriosis with marked smooth muscle proliferation or metaplasia can be misdiagnosed as spindle cell neoplasia in a small biopsy specimen. Such misdiagnosis can lead to overtreatment such as wide resection and chemotherapy. Therefore, a tumor-like lesion in the female pelvic or abdominal cavity requires careful examination and treatment.

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