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Extrauterine Adenomyoma Invading the Sigmoid Mesocolon in the Pelvic Cavity

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Abstract

Adenomyomas are benign tumors typically located in the uterus, and extrauterine adenomyomas are rare, especially those in the sigmoid mesocolon. Extrauterine adenomyomas resemble the uterine structure and can occur anywhere in the pelvic cavity or abdomen. Here, we present 2 cases of extrauterine adenomyomas invading the sigmoid mesocolon.

Keywords: Extrauterine adenomyoma; Uterine-like masses; Sigmoid mesocolon; Pelvic cavity

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Introduction

Adenomyomas are benign tumors comprising endometriotic tissue and smooth muscle tissue. They are typically found in the uterus [1] and rarely in extrauterine regions such as the ovary [2], broad ligament [3], and small intestine mesentery [4]. The ovary is reportedly the most common site of extrauterine adenomyomas, and those invading the sigmoid mesocolon are extremely rare. Extrauterine adenomyomas are referred to as “uterine-like masses” or “cavitated accessory masses”. Like the characteristic intrauterine adenomyomas, they have a central cavity lined with functional endometriotic tissue and are surrounded by smooth muscle tissue, resembling the structure of the uterus. The differential diagnosis of extrauterine adenomyomas includes endometriosis with prominent smooth muscle hyperplasia or metaplasia, congenital malformation, and smooth muscle tumors with an entrapped endometriotic element. Sometimes, they are considered a malignancy of epithelial or mesenchymal origin [5]. In this report, 2 cases of extrauterine adenomyoma invading the sigmoid mesocolon have been described, and a review of the literature has been presented. Our institutional review board approved this retrospective study and waived patient informed consent.

Case Reports

Case 1

A 29-year-old woman visited a local clinic for lower abdominal pain that had persisted for 5 months. Ultrasonography showed bilateral adnexal masses. She was diagnosed with a tubo-ovarian abscess and received medical treatment for it. However, the mass did not decrease in size. Therefore, the patient was referred to our hospital. Transvaginal sonography showed a mixed echogenic lesion between the right adnexa and posterior cul de sac, and it was diagnosed as a right tubo-ovarian abscess. An abdomino-pelvic axial computed tomography [CT] scan showed an 8.6 cm multilobulated solid cystic mass between the right adnexa and posterior cul de sac (**Figures 1A and B**). The differential diagnosis included a right tubal myoma and an ovarian stromal or germ cell tumor with borderline malignancy. On laparoscopic examination (**Figures 2A and B**), multiple myoma-like lesions were found connected by a fibrotic band in the right adnexa. Four masses of size 5~7 cm were detected; they were adherent to and had invaded the sigmoid colon. The masses were removed and simple closure of the rectal wall was conducted. On pathologic examination, the right adnexal masses were diagnosed as leiomyomas [extrauterine leiomyomas], and those adherent to the sigmoid colon were diagnosed as adenomyomas (**Figures 3A and B**). The patient’s chronic abdominal pain improved after surgery.

Case 2

A 42-year-old woman presenting a palpable mass visited the Department of Obstetrics and Gynecology at CHA University Medical Center. She had dysmenorrhea and chronic pelvic pain. Transvaginal ultrasonography showed multiple myomas of the SS type showing central cystic changes. Pelvic magnetic resonance imaging [MRI] showed multiple myomas with similar

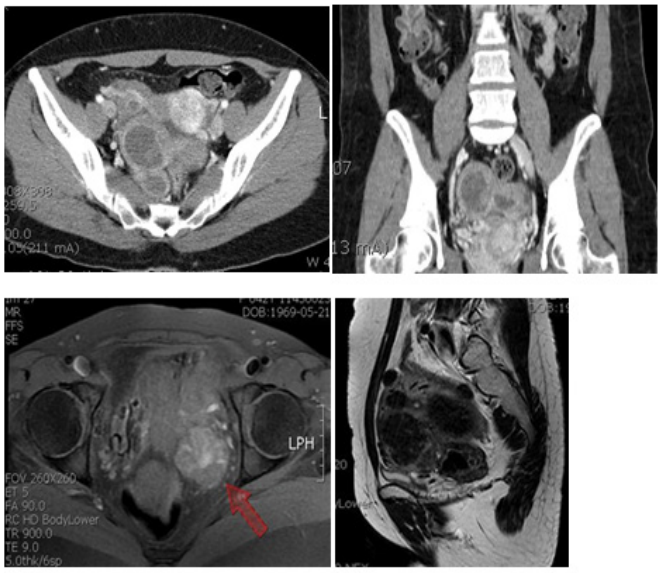

Figure 1 Case 1. Contrast-enhanced CT scan showing an 8.6-cm multilobulated solid cystic mass. The mass was well defined and had a cystic portion (A: transverse view, B: coronal view). Case 2. C: Transverse T1-weighted MR images showing a mass of about 4.4 × 4.7 × 3.6 cm (red arrow) with multifocal high signal intensity in the left posterior aspect of the lower uterine body. D: Sagittal T2-weighted image showing the heterogeneous signal intensity of a cystic lesion (yellow arrow).

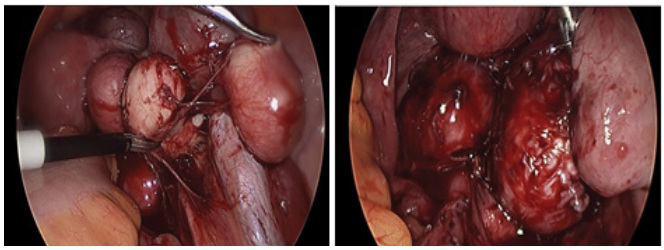

Figure 2 Case 1. A: Laparoscopic view of multiple myoma-like lesions in the right adnexa. These lesions were not connected to the uterus. B: Multiple masses, 5–7 cm in size, adherent to the sigmoid colon. The masses had a cystic cavity containing thick, dark chocolate-colored material.

characteristics within the uterus and a 5 cm cystic mass in the posterior cul de sac of the posterior aspect of the lower uterine body. MRI showed multifocal T1 high-signal-intensity portions and multifocal nonenhancing portions in the mass (**Figures 1C and D**). Multiple myomas and an SS-type myoma with red degeneration in the posterior aspect of the lower uterine body were diagnosed. The differential diagnosis included leiomyosarcoma. Intraoperatively, the uterus was found to be grossly enlarged. The 5 cm mass that had invaded the sigmoid colon was located in the cul de sac between the uterus and sigmoid colon. The right ovary seemed enlarged because of the 3 × 4 cm ovarian cyst. The inner surface of the cystic wall of the mass invading the sigmoid colon was rough, and it contained a dark chocolate-colored fluid. After the mass was removed, the anterior wall of the sigmoid colon showed a longitudinal tear of about 4–5 cm. Total abdominal hysterectomy and primary repair of the sigmoid colon were performed, along with right ovarian cystectomy. On histopathological examination, the adenomyomas and leiomyomas were found to be localized in the uterine myometrium. Further, the sigmoid colon mass was diagnosed as an adenomyoma (**Figures 3C and D**), and the right ovarian cyst was diagnosed as an endometriotic cyst. The patient's chronic pelvic pain subsided after the operation.

Discussion

About 30 cases of extrauterine adenomyomas at various sites have been reported [4], the most common site being the ovary. The cases presented here are those of rectovaginal adenomyomas invading the sigmoid colon, which is a very rare site of extrauterine adenomyomas. The gross finding of a uterine-like mass varied between the cases, as did the size of the masses. However, in both cases, the internal component including solid and cystic areas was detected, and functional endometrial glands and stroma within a muscular walled structure resembling a uterus were found on microscopic examination [5]. Extrauterine adenomyomas need to be distinguished from several other conditions. In particular, they are often considered malignancies of epithelial or mesenchymal origin because in imaging studies like CT and MRI, they show solid and cystic portions, which are

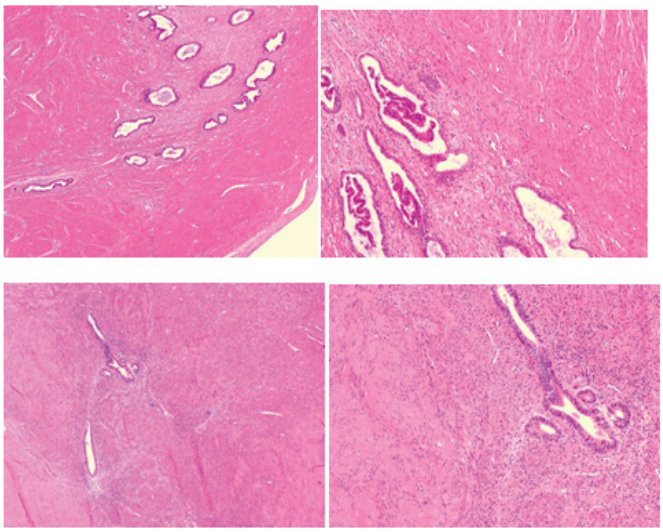

Figure 3 Case 1. A: Histologically examined section showing the endometriotic lining of the cystic cavity (hematoxylin and eosin [H&E] staining; original magnification, ×40). B: At a higher magnification, the endometrial epithelium and stroma with the gland surrounded by myomatous smooth muscle can be seen (H&E staining; original magnification, ×100). Case 2. C: Histologically examined section showing the endometriotic lining of the cystic cavity (H&E staining; original magnification, ×40). D: At a higher magnification, the endometrial epithelium and stroma with the gland surrounded by myomatous smooth muscle can be seen (H &E staining; original magnification, ×100).

characteristic of such malignancies [6,7] The contrast-enhanced CT scan in case 1 in the present study showed a multilobulated solid cystic mass between the right adnexa and posterior cul de sac. We considered it to be an ovarian tumor such as a stromal or germ cell tumor with borderline malignancy. In case 2, MRI showed a mass with multifocal T1 high-signal-intensity regions in the left posterior aspect of the lower uterine body. Differential diagnosis included SS-type myoma with red degeneration and leiomyosarcoma. In another case report, the authors reported that on preoperative CT, the mass was diagnosed as a gastrointestinal stroma tumor with cystic degeneration, but the results of postoperative histologic findings showed that it was an adenomyoma [8]. Thus, a well-defined mass with central cystic changes detected on imaging studies in the pelvic cavity in women is probably indicative of a uterine-like mass. Extrauterine adenomyomas seem to be associated with endometriomas and adenomyosis [9]. In fact, the patient in case 2 in the present study had a right ovarian cyst, which was diagnosed as an endometrioma on pathologic examination. Three theories have been proposed with regard to the etiological factors of extrauterine adenomyomas. The first concerns the metaplastic transformation of the subcoelomic mesenchyme [10]. The subcoelomic mesenchyme lies underneath the mesothelial surface of the peritoneum, and peritoneal mesothelial cells are considered to be the origin of metaplastic cells. According to the abovementioned theory, uterine-like masses might originate from the peritoneal mesothelium. The subcoelomic mesenchyme is thought to be multipotent and may proliferate in response to estrogen hormone stimulation, and estrogen is considered important in uterine-like mass development, affecting both endometrial tissue and smooth muscle proliferation [11]. This theory is supported by the effectiveness of gonadotropin-releasing hormone agonist therapy [11] and unopposed estrogen therapy [12] in the treatment of uterine-like masses. Second, the congenital anomaly theory [13] is based on the fact that this disease is often associated with congenital genital tract abnormalities. The uterine-like mass may result from either a mullerian duct fusion defect or partial duplication of the mullerian system. The third theory is the heterotopia theory [14], which indicates that these masses are neither a congenital anomaly nor a metaplasia. However, it seems to be consistent with the metaplastic theory. It is most likely that the rectovaginal adenomyomas in our cases were derived from the subcoelomic mesenchyme, since no mullerian fusion defect was detected. A previous study suggested that adenomyomas of the rectovaginal septum are associated with metaplasia of the mullerian rests, and that extrauterine adenomas of the rectovaginal septum must be differentiated from other rectal neoplasms, particularly cancer [15]. In summary, extrauterine adenomyomas are extremely rare. On imaging evaluation, they are sometimes diagnosed as malignancies, endometriosis, or ovarian leiomyoma because they have solid and cystic portions. Although their pathogenesis is unclear, metaplasia has been suggested, as in the cases presented here. The current report may aid in the diagnosis of such adenomyomas in the future.

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