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Parasitic Leiomyoma with Lymphatic Dilatation in Trocar Port-Site of Abdominal Wall: A Case Report 트로카 삽입 부위에 생긴 림프관 확장을 동반한 기생 평활근종: 중례 보고

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Uterine leiomyoma is the most common benign pelvic tumor in female and being symptomatic is an indication for surgical removal. As laparoscopic surgery has been developed, some cases related to parasitic leiomyomas in the port site have been reported. A 40-year-old female who a history of previous laparoscopic surgery to remove uterine myoma 2 years ago visited in outpatient clinic of general surgery with palpable mass in left lower abdomen. Contrast enhanced abdomen CT and pelvis MRI were done to evaluate the mass. It was diagnosed parasitic leiomyoma in pathologic study after surgical removal and parasitic leiomyoma should be considered when patient visited presenting abdominal mass with the history of laparoscopic myomectomy.

Index terms Leiomyoma, Uterine; Postoperative Complications; Morcellation

INTRODUCTION

Uterine leiomyomas are the benign tumors that can occur in the women of reproductive age. It is the most common benign tumor and is surgically removed if symptomatic. The term of parasitic leiomyoma was first described in 1909 (1) and it was defined as abnormal variant of free-lying leiomyoma for some reasons from the uterus with blood supply from neighboring structures (2).

As laparoscopic surgery began to develop, some case reports of parasitic leiomyoma due to

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This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (https://creativecommons.org/ licenses/by-nc/4.0) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited. surgical fragment were increased (2).

Here, we report the rare case of a patient with parasitic leiomyoma with lymphatic dilatation in Trocar port site of lower abdominal wall who have a history of previous laparoscopic myomectomy.

CASE REPORT

A 40-year-old female visited in the outpatient clinic of general surgery with palpable mass in left lower abdomen. She had a history of previous laparoscopic surgery to remove uterine myoma 2 years ago. On physical examination, there was a large palpable mass without tenderness in left lower abdomen just below previous Trocar port insertion site. Laboratory test showed mild elevation of aspartate aminotransferase (49 U/L, normal range: 19–48 U/L) and the others were unremarkable.

CT (SOMATOM Perspective, Siemens, Erlangen, Germany) scan of abdomen revealed a welldemarcated large bilobulated mass with heterogeneous enhancement. It was located just below previous port insertion site in left lower abdominal wall through subcutaneous fat, left rectus abdominis muscle, and peritoneum (Fig. 1A). For further evaluation, contrast-enhanced pelvis MRI (Skyra, Siemens) was done. On T2-weighted imaging revealed that there was a 9.5 cm \times 7.9 cm \times 8.0 cm well demarcated mass with suspicious internal cystic component at left lower abdomen. Contrast-enhanced T1-weighted imaging showed heterogeneous enhancement excluding cystic portion (Fig. 1B). Diffusion weighted image (DWI) and apparent diffusion coefficient image showed multifocal diffusion restricted areas (Fig. 1C).

Because the patient had a history of previous laparoscopic myomectomy, initially it was suspected as parasitic leiomyoma at port site with internal cystic change. But malignant tumor of smooth muscle cells such as leiomyosarcoma with secondary degeneration was also suspected due to some restricted diffusion areas and heterogeneous enhancement.

Surgical resection was done and it was diagnosed leiomyoma with lymphatic dilatation in pathologic study (Fig. 1D-F). In operation field, there was no leakage of cystic lesion of the mass and it was removed intactly. In pathologic examination, there was no fat nor cystic component. Surgical resection of the mass was done successfully and the patient discharged healthily 2 days after operation.

Written infromed consent was obtained from the patient for the use of data for research purposes.

DISCUSSION

Uterine leiomyoma is the most common benign pelvic tumor for women. It occurs in about 25% of reproductive age and also founded above 80% of surgically removed uterus (2).

The International Federation of Gynecology and Obstetrics (FIGO) classified uterine leiomyoma into eight subtypes according to their positional relationship with myometrium (3). Especially, parasitic leiomyoma is a rare subtype of pedunculated leiomyomas, lie free from uterus. They may originate from fragment of leiomyoma that have detached from the uterus and are supplied blood from adjacent organs (4). The first report of parasitic leiomyoma was reported by Kelly and Cullen in 1909 as parasitic leiomyoma of the urinary bladder (1). However, with recent increase in laparoscopic surgery, a new type of parasitic leiomyoma has emerged. Large leiomyoma needs to be morcellated in order to be retrieved from the abdominal cavity. During morcellation, some fragment of

Fig. 1. A 40-year-old female with parasitic leiomyoma in the trocar insertion site of the abdominal wall with lymphatic dilatation.

A. Contrast-enhanced CT scan of the abdomen. Axial (left) and coronal (right) images show a 9.5 cm \times 7.9 cm \times 8.0 cm sized, well-demarcated bilobulated mass with heterogeneous enhancement in the left lower abdominal wall.

B. Contrast-enhanced T1 image (left) shows heterogeneous enhancement of the mass (arrows) excluding cystic portion. Axial T2 (right) weighted imaging shows dumbbell-shaped well-demarcated mass with internal cystic component (*) at the left lower abdomen.

C. Axial diffusion weighted imaging (left)/apparent diffusion coefficient (right) image shows multifocal diffusion restricted areas of the mass (arrows).

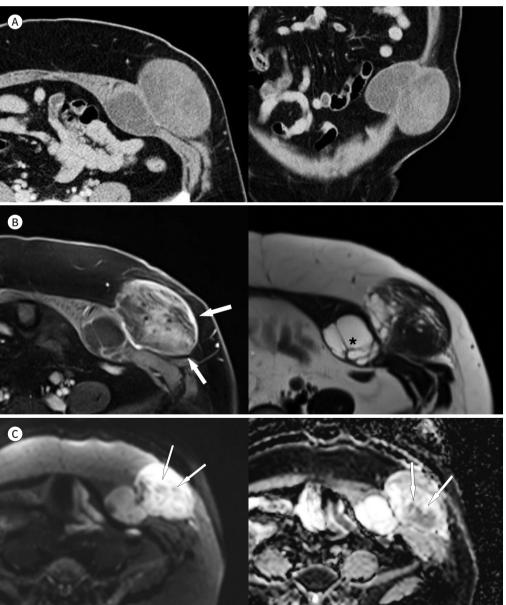
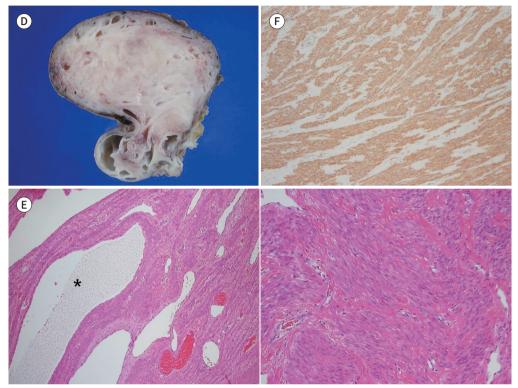


Fig. 1. A 40-year-old female with parasitic leiomyoma in the trocar insertion site of the abdominal wall with lymphatic dilatation (Continued).

D. The gross specimen photograph shows a 7.5 cm \times 7.0 cm \times 6.5 cm sized, grayish, white mass with interlacing bundles of fibers.

E. Photomicrography shows lymphatic dilatation (*, left) (hematoxylin and eosin stain, \times 20) and spindle cell with long and blunt-end (right) (hematoxylin and eosin stain, \times 100).

F. In an immunohistochemical stain image (\times 100) smooth muscle actin is positive.



leiomyoma may be inadvertently left in the abdominal cavity and subsequently implanted (5). The incidence of parasitic leiomyoma due to power morcellation is about 0.2%–1.25% and median diagnosis interval was 48 months (6).

Parasitic leiomyoma has a non-specific clinical symptoms and it depends on the site of tumor recurrence. The common sites include the pelvic cavity, small intestines, rectum, vaginal stump, and laparoscopic port site (7).

Parasitic leiomyoma is benign smooth muscle tumor, but if it has detached from uterus and is found in the pelvic cavity or laparoscopic port site, differential diagnosis is required to exclude malignancy. Physical examination and measurement of tumor markers such as carbohydrate antigen 125, α-fetoprotein, or carbohydrate antigen 19–9 may be helpful, though they may be falsely elevated (2). Therefore US and MRI are very helpful for diagnosis. Typically, non-degenerated leiomyoma is shown hypointense on T2- and isointense on T1 weighted imaging. But if degeneration is accompanied, various signal intensity with heterogeneous enhancement can be shown and in that case, differentiation from leiomyosarcoma is essential (8). In our case, the tumor presented with a well circumscribed dumbell shaped mass with internal T2 high signal intensity portion and shows several restricted diffusion areas on high B value DWI. So, initially, we thought leiomyosarcoma with internal cystic change should be in-

cluded for differential diagnosis, but, later it is confirmed to leiomyoma with cystic dilatation of lymphatics and internal vessels on pathologic findings. By Tamai et al. (9) leiomyoma without degeneration sometimes has a T2 high signal intensity rim, which corresponding to a pseudocapsule of dilatated lymphatics, vein or edema.

In order to decrease developing parasitic leiomyoma, various alternatives have been proposed to reduce the risk of parasitic leiomyoma that may occur after power morcellation. Using containment bag or by cutting of large specimen to remove through colpotomy or minilaparotomy are alternative methods (10), but the most important thing is essentially careful to check intraabdominal cavity after morcellation to avoid remnant myoma fragment.

In conclusion, parasitic leiomyomas in Trocar insertion site are one of the rare late complication of laparoscopic myomectomy. As laparoscopic surgery began to develop, it replaces conventional surgery that need to wide skin incision and mass excision. The morcellation of the mass is essential course to take out the whole mass from the small port site in laparoscopic surgery. In this course, fragmented tissues may become dropped and continue to form parasitic myomas. Because of these risk factors, in order to reduce the incidence of parasitic leiomyoma, it is recommended that shredding should be done in a surgical bag.

In summary, this study suggest parasitic leiomyoma should be considered when patient visited presenting abdominal mass with the history of laparoscopic myomectomy and malignant tumors of smooth muscle cells also should be ruled out.

Author Contributions

Conceptualization, P.S.Y.; data curation, all authors; investigation, all authors; validation, P.S.Y.; visualization, all authors; writing—original draft, all authors; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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트로카 삽입 부위에 생긴 림프관 확장을 동반한 기생 평활근종: 증례 보고

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자궁 근종은 여성에서 가장 흔한 양성 골반 강 내 종양이며 증상이 있을 경우 수술적 제거를 하게 된다. 내시경 수술이 발달하게 되면서 포트 삽입 부위의 기생 평활 근종들이 보고되기 시작하였다. 40세의 여자 환자가 왼쪽 아랫배에서 만져지는 종괴를 주소로 외래로 내원하였 고 2년 전 내시경 근종 제거를 받은 과거력이 있었다. 조영증강 CT와 MRI를 시행하여 해당 종괴에 대한 평가를 시행하였으며 수술적 제거를 시행하였다. 병리 검사에서 해당 종괴는 기 생 평활 근종으로 최종 진단되었는데 이는 이전 내시경 근종 제거술을 시행한 환자에서 수술 부위에 만져지는 종괴를 주소로 내원하였을 때 반드시 기생 평활 근종을 감별 진단으로 고려 하여야 함을 시사한다.

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