Case Report

Obstet Gynecol Sci 2018;61(3):425-429
https://doi.org/10.5468/ogs.2018.61.3.425
pISSN 2287-8572 · eISSN 2287-8580

Obstetrics & Gynecology Science

# Successfully removed uterine angioleiomyoma by robot-assisted laparoscopic myomectomy

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Angioleiomyoma is a rare type of leiomyoma variant and there are a few cases reported to date. Herein, we present a case of angioleiomyoma in a 36-year-old woman with lower abdominal pain, initially diagnosed by degenerated uterine leiomyoma. The transvaginal ultrasonogram showed an ovoid-shaped heterogeneously hyperechoic lesion in left cornual site of uterus and pelvic magnetic resonance image showed an about 5.1 cm sized heterogenous T2 intermediate high mass with poor enhancement. The patient underwent a robot-assisted laparoscopic myomectomy, and final histopathologic diagnosis revealed uterine angioleiomyoma. This case is the first case of angioleiomyoma resected by robotic surgery. The patient is on follow up for over 1 year and shows no evidence of recurrence until now.

Keywords: Angiomyoma; Uterine myomectomy; Robotic surgical procedures

## Introduction

Angioleiomyoma, known as vascular leiomyoma, is a rare variant of leiomyoma composed of smooth muscle cells and thickened vessels [1]. Angioleiomyoma usually occurs in the subcutaneous tissue, most often in the lower extremities, head and neck region [1], and can be rarely found in the female genital tract including uterine corpus, uterine cervix, ovary or broad ligament [2-8].

Preoperative diagnosis may be difficult because these lesions have similar clinical and radiological findings with those of conventional leiomyoma. Therefore, a definite diagnosis can be made only after histopathologic examination [9]. The treatment of choice for angioleiomyoma is complete surgical excision such as angiomyomectomy or simple hysterectomy depends on the patient's desire to preserve fertility [10].

We recently experienced a case of successfully removed angioleiomyoma by robot-assisted laparoscopic myomectomy in woman who wanted future fertility. This case is the first case of angioleiomyoma resected by robotic surgery, and we report this case with a brief literature review.

## Case report

A 36-year-old multiparous woman visited Seoul St. Mary's Hospital complaining of lower abdominal pain. She had been diagnosed with uterine myoma by transvaginal ultrasonography 2 years ago, and regularly visited hospital for monitoring it. When she was initially diagnosed with uterine myoma, she had no specific symptom caused by the uterine myoma. The size of uterine myoma was 3.5 cm 2 years ago, but it was increased to 4.5 cm when the patient visited our hospital with lower abdominal pain. The pelvic ultrasonography revealed 4.5 cm sized uterine myoma. The mass was hyperechoic, and,

Received: 2017.05.30. Revised: 2017.07.05. Accepted: 2017.07.24.
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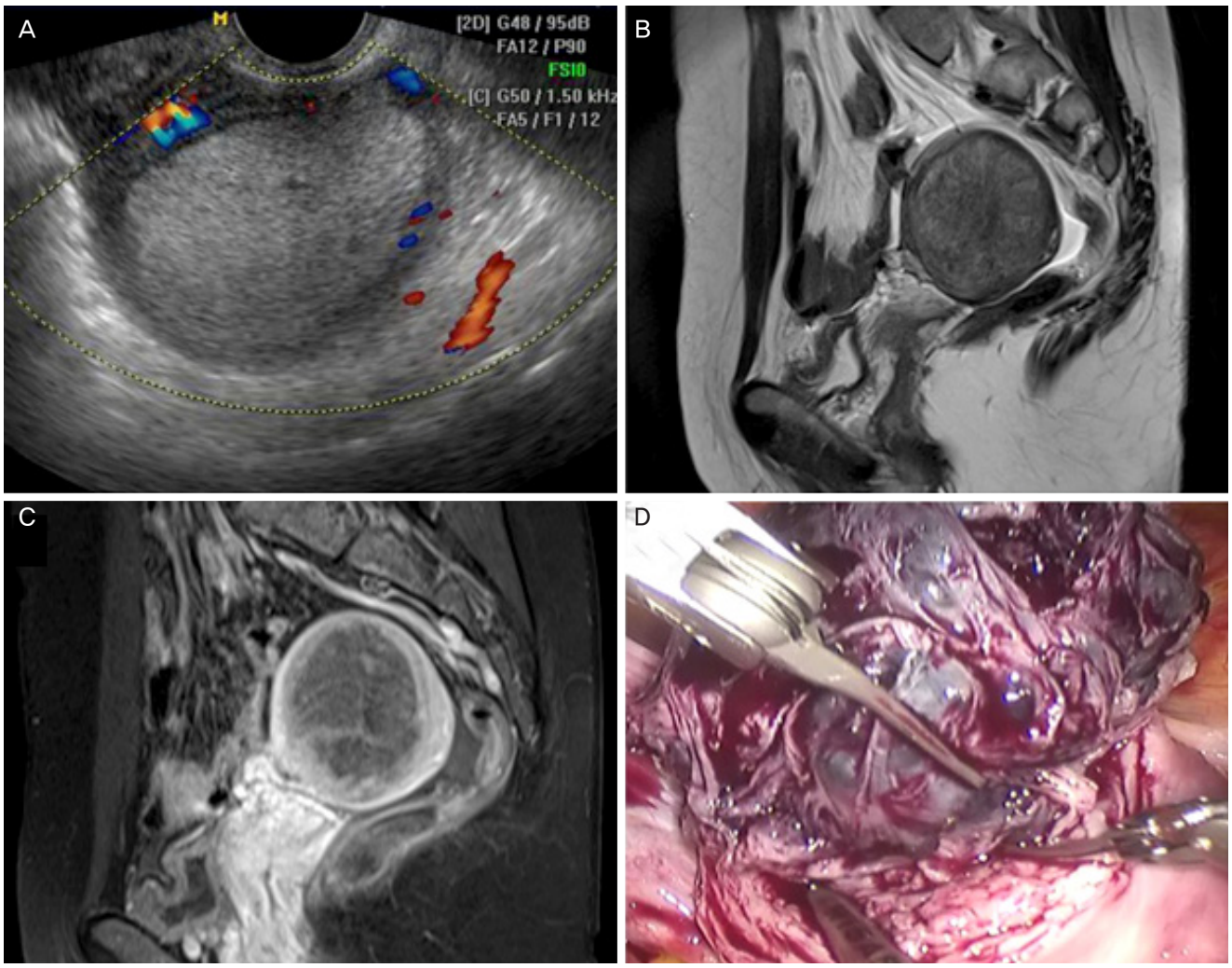


**Fig. 1.** (A) Transvaginal ultrasonography showing a 4.5 cm sized hyperechoic mass with vascular structures around the mass. (B) Sagittal view on pelvic magnetic resonance image showing 5.1 cm sized heterogeneous, intermediate to high signal intensity in T2-weighted image and (C) poor enhancement in T1-weighted post-gadolinium image. (D) Intra-operative findings showing about 5 cm sized, blood-filled multicystic tumor mass in the myometrium.

upon application of color Doppler, demonstrated vascular structures only around the mass (Fig. 1A). Pelvic magnetic resonance image (MRI) showed 4.7 cm sized mass on left side of the uterus. It was heterogeneous, intermediate to high signal intensity in T2-weighted image (Fig. 1B) with poor enhancement in T1-weighted post-gadolinium image (Fig. 1C), and a few intratumoral high signal intensity foci in T1-weighted image. And there was no diffusion restriction in pelvic MRI. Because of the heterogeneity of the mass, we considered it as a degenerated uterine myoma such as hemorrhagic change or red degeneration. Serum cancer antigen 125 level was 9.28 U/mL. All the laboratory tests were within normal limits.

Because she wanted to preserve her uterus for having babies in future, we planned to perform a robot-assisted laparoscopic myomectomy. After an inspection of the operative field using the basic robotic system, we found 5 cm sized uterine myoma on the left anterior wall of the uterus. For hemostasis, diluted vasopressin (vasopressin 10 IU in 100 mL of normal saline) was injected into the serosa and myometrium surrounding the myoma. Using curved monopolar scissors, we performed a transverse incision over the vertex of the myoma. After serosal incision, we found blood-filled multicystic tumor mass in the myometrium which was seemed to have an unclear margin between the mass and normal myo-

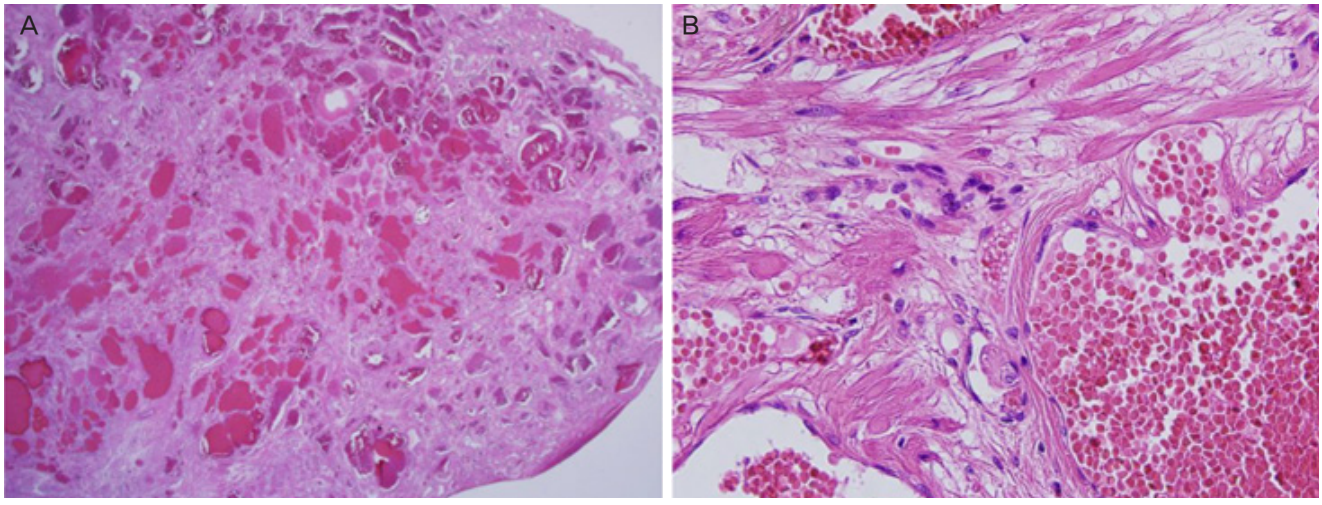


**Fig. 2.** (A) Microscopic findings showing thickened smooth muscle fibers and blood vessels (hematoxylin & eosin, ×40). (B) No pleomorphism or mitotic figure is identified in spindle smooth muscle cells and endometrial cells (hematoxylin & eosin, ×100).

metrium (Fig. 1D). Fortunately, the mass was well-separated from the myometrium, and we performed myomectomy and repaired remaining myometrium with a 2-0 polydioxanone suture. After completion of the 2-layered myometrial sutures, we repaired the serosal layer with a baseball suture. Morcellation of the tumor mass was conducted under conventional laparoscopy. The patient had an uneventful recovery and was discharged on post-operative day 2.

Microscopic findings showed thickened smooth muscle fibers and blood vessels (Fig. 2A). No pleomorphism or mitotic figure was identified in spindle smooth muscle cells and endometrial cells (Fig. 2B). The final histopathologic diagnosis was uterine angioleiomyoma.

## Discussion

Angioleiomyoma is a rare benign mesenchymal neoplasm consisting of smooth muscle cells and thickened vessels, which usually arise from the lower extremities, head and neck region [1]. It has also occurred in unusual locations in female genital tracts such as uterus, retroperitoneum, broad ligament, and ovary [4,5,7-9]. According to Garg and Mohanty [9], uterine angioleiomyoma is an extremely rare tumor as only 16 cases have been reported to date. Uterine angioleiomyoma usually occurs in the middle-aged women of the fourth to sixth decades, and is commonly presented with menorrhagia, abdominal pain or palpable abdominal mass. High vascularity of the lesion also increases the risk of spontaneous rupture of angioleiomyoma with severe intraabdominal bleeding [11].

Uterine angnioleimyoma is commonly presented as a well-circumscribed solitary mass arising from uterine corpus. However, the preoperative diagnosis of uterine angioleiomyoma is extremely difficult because it has no specific radiologic finding differentiating it from conventional uterine leiomyomas in ultrasonogram and pelvic MRI. Also, microscopic diagnosis using immunohistochemical markers is essential for differential diagnosis from other tumors such as endometrial stromal nodule, angiomyofibroblastoma of perivascular epitheloid cell tumor [12]. The histological characteristics of angioleiomyoma is composed of interlacing fascicles of spindled smooth muscle cells around abundant thick-walled blood vessels, in contrast to the usual leiomyomas in which the density of the vascular network is similar to or less than the normal myometrium. Most of these lesions have rare mitotic figures, pleomorphism, or necrosis [12].

The treatment of choice for angioleiomyoma is complete surgical excision such as angiomyomectomy or simple hysterectomy depending on the patient's desire to preserve fertility. No case of recurrence of uterine angioleiomyoma after surgical resection has been reported until now.

In the reviewed cases, most patients underwent hysterectomy with or without salpingo-oophorectomy because the patients were middle-aged women who did not want to preserve their ferility. To the best of our knowledge, we found

only 3 cases of angioleiomyomectomy in relatively young women [13-15] and only one case reported laparoscopic excision of the tumor [13]. The present case is the first case of robot-assisted-laparoscopic angioleiomyomectomy being reported in the literature.

When myomectomy is performed with robot-assisted laparoscopy, more appropriate and clear visual field of operation is achieved through a 3D-HD screen compared to conventional laparoscopy. Especially, robot-assisted laparoscopic myomectomy is beneficial in severely degenerated myoma or variant tumor as shown in this case, which tends to have unclear margin between the mass and normal myometrium. Actually, the resected angioleimyoma in this case was friable, multicystic and irregular-shaped, while conventional leiomyoma appears as round, circumscribed, and solid nodules. Because the mass was too friable to allow a firm grip with a screw or tenaculum and had irregular margin grossly between the lesion and normal myometrium, the complete excision of the mass might be difficult by conventional laparoscopy.

In conclusion, the uterine angioleiomyoma is a rare benign variant of uterine leiomyoma composed of smooth muscle cells and thickened vessels. Preoperative diagnosis is very difficult and a definite diagnosis can be made only after histopathologic examination. The complete surgical excision such as angiomyomectomy or simple hysterectomy is an effective treatment with a satisfactory outcome, and Robot-assisted laparoscopic angioleiomyomectomy also could be a good alternative method for the patient who wants future fertility.

## Acknowledgements

This research was supported by Basic Science Research Program through the National Research Foundation of Korea (NRF) funded by the Ministry of Science, ICT & Future Planning (2012R1A1A3020083, 2017R1D1A1B03028045). In addition, we would like to thank Jeeyune Park, Ka-young Kim and Hye-Sung Hwang for English proofreading.

## Conflict of interest

No potential conflict of interest relevant to this article was reported.

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