Case Report

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대한영상의학회지

A Rare Angioleiomyoma of the Uterine Cervix: A Case Report with Peculiar MRI Findings

드문 형태의 자궁 경부 혈관근종: 특이한 자기공명영상 소견을 포함한 증례 보고

Yun Chul Hwang, MD, Seo Young Park, PhD*

Department of Radiology, Daegu Fatima Hospital, Daegu, Korea

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***Corresponding author**
Seo Young Park, PhD
Department of Radiology,
Daegu Fatima Hospital,
99 Ayang-ro, Dong-gu,
Daegu 41199, Korea.

Tel 82-53-940-7770
Fax 82-53-954-7417
E-mail uniun0926@gmail.com

ORCID iDs
Yun Chul Hwang
https://orcid.org/0000-0001-8359-4243
Seo Young Park
https://orcid.org/0000-0002-4626-4285

Angioleiomyoma (vascular leiomyoma) of the uterine cervix is an extremely rare type of benign tumor composed of smooth muscle cells and thick-walled blood vessels. Only a few cases of cervical angioleiomyoma have been reported. Here, we present imaging, including ultrasonography, contrast-enhanced CT, MRI, and histopathological findings of a 38-year-old female with an angioleiomyoma of the uterine cervix.

Index terms Angioleiomyoma; Menorrhagia; Magnetic Resonance Imaging

INTRODUCTION

Angioleiomyoma is a rare benign mesenchymal soft tissue tumor (1). It has a higher incidence in the 4th to 6th decade of life and usually originates in lower extremities, head, neck and trunk (1). Therefore, angioleiomyoma in the at female lower genital tract is rare and occasionally found in the uterus, however, cervical angioleiomyoma is an extremely rare type of leiomyoma variant and there has only been a few cases reported thus far.

In this report, we present a case of angioleiomyoma of the uterine cervix because of its extreme rarity and peculiar imaging findings.

CASE REPORT

A 38-year-old female was presented to our gynecological clinic with a recurrent urinary retention and hypermenorrhea. Her vitals were stable. On vaginal examination, a

large protruding mass was seen on the left side wall of the uterine cervix, and a further physical examination revealed that it was within normal limits. For further evaluation of the cervical mass, blood tests including tumor markers and radiological investigations, were performed. The patient's blood indices were as follows: hemoglobin 8.3 g/dL and other indices including carbohydrate antigen 19-9 (CA19-9) and CA125 were in the normal range.

A transvaginal pelvic ultrasonography (US) was performed (Fig. 1A) and a well-demarcated, heterogeneous, large mass measuring 10.6 cm was seen in the pelvic cavity. The mass lesion contained anechoic/hypoechoic areas of cystic portion. Because the left ovary was not seen in US, with a suspicion of a ovarian malignancy, pelvic CT & MRI were performed to better demonstrate the origin and the characterization of the mass (Fig. 1B-D, respectively). An enhanced CT (Toshiba, Aquilion PrimeSP, Tokyo, Japan) scan revealed a large heterogeneous cystic mass with multiple confluent nodular enhancing components. It was located on the left side of the uterine cervix, displacing the uterus right laterally. The MRI (Siemens, Avanto 1.5T, Erlangen, Germany), showed that the mass was located at subepithelial layer of the uterine cervix on sagittal T2-weighted imaging. On Contrast enhanced T1-weighted imaging revealed that there were multiple enhancing soft tissue components which were correlated with T2 low signal intensity portions. The ovaries appeared to be normal. According to the CT and MRI findings, the initial diagnosis of the mass was thought to be a large leiomyosarcoma originating at the uterine cervix. Pelvic mass resection and a total abdominal hysterectomy with a left oophorectomy were performed.

At histopathological examination, macroscopically, a 9 cm × 9 cm, round shape, well-demarcated mass in the left side muscle layer of the uterine cervix was seen (Fig. 1E). A cut surface of the tumor revealed many focal hemorrhages, which were presumed to be from internal vascular structures. There was no evidence of necrosis in the specimen. Microscopically, the tumor was formed by smooth muscle cells and numerable thick-walled blood vessels with partially patent lumens (Fig. 1F). In immunohistochemical studies, smooth muscle actin (SMA), desmin, CD34, estrogen receptor (ER), progesterone receptor (PR), and Human Melanoma Black 45 (HMB45) studies were performed. Tumor cells expressed SMA, desmin, CD34, ER and PR in a diffused manner while HMB45 was negative. Most of the vessels were stained with a CD34 antibody, which is constitutively expressed on endothelial cells. HMB45 negativity exclude the diagnosis of leiomyolipoma. Based on the results, the tumor was diagnosed as an angioleiomyoma of the cervix. At the two weeks follow-up, there was no evidence of a complication.

This study was approved by the Institutional Review Board of our institution (IRB No. DFE21ORIO100). Informed consent was waived due to the retrospective nature of the study.

DISCUSSION

Angioleiomyoma, also known as vascular leiomyoma, is a rare benign tumor, composed of smooth muscle cells and thick-walled blood vessels. These tumors usually occur in lower extremities, body trunk, head and neck, and retroperitoneum, so it can be a rare clinical presentation in the gynecologic organ.

In an up-to-date search, there are a few previous cases regarding angioleiomyoma in the fe-

Fig. 1. Angioleiomyoma of the uterine cervix in a 38-year-old female, presenting with recurrent urinary retention and hypermenorrhea.
A. The transvaginal pelvic ultrasonography image shows a well-demarcated, heterogeneous, large mass measuring 10.6 cm in the pelvic cavity. The mass includes hypoechoic areas, indicating the cystic portion (arrow).
B. The contrast-enhanced axial CT image shows a well-defined, large cystic mass with multiple confluent nodular enhancing components (arrow) in the pelvic cavity. The fat plane between the cervix (open arrow) and the mass is indistinguishable, implying that the mass may have originated from the cervical wall.
C. T2-weighted sagittal (left) and axial (right) MR images show a heterogeneous cystic mass with a hypointense nodule-like structure originating at the subepithelial layer of the uterine cervix.
D. The T1-weighted axial MR image (left) shows a hypointense mass with some internal high signal intensity areas (arrow), which are enhanced in the fat-suppressed T1-weighted contrast-enhanced MR image (right).

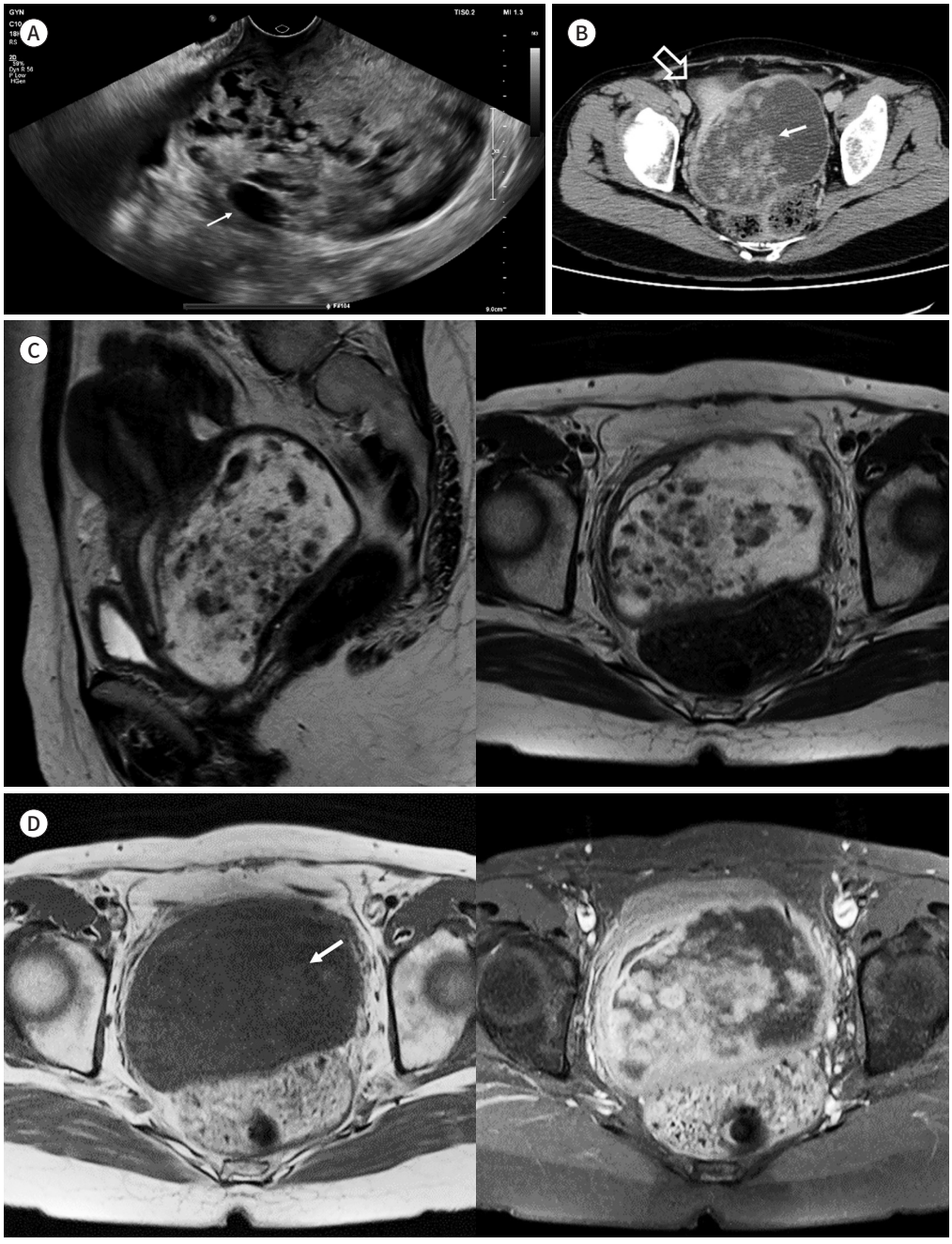
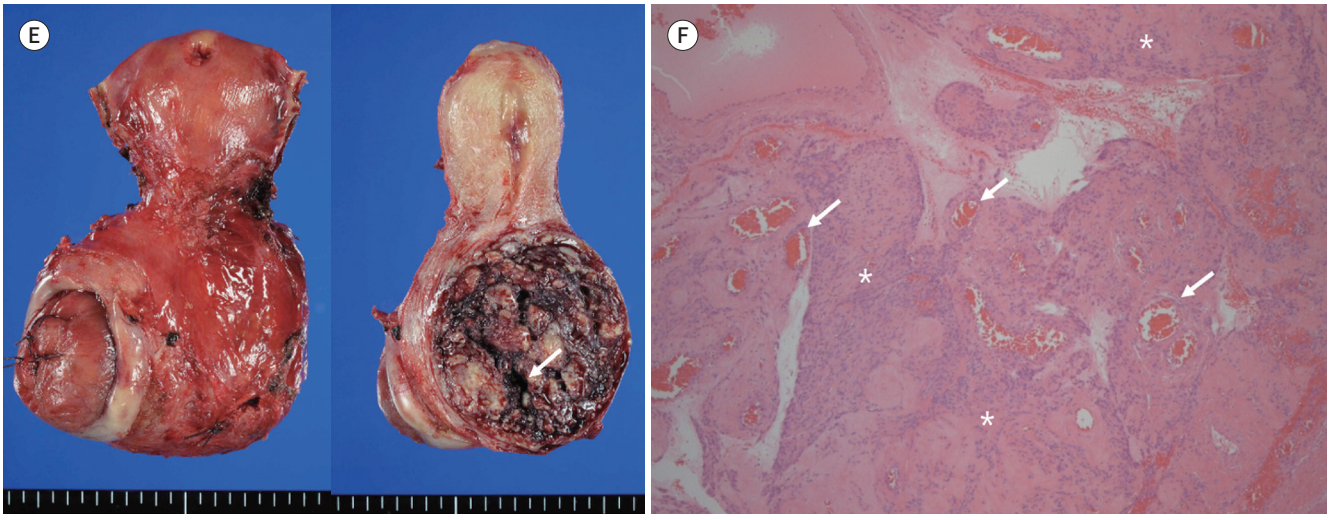


Fig. 1. Angioleiomyoma of the uterine cervix in a 38-year-old female, presenting with recurrent urinary retention and hypermenorrhea.
E. The gross appearance of the specimen (right) shows a round, well-demarcated mass on the left side of the cervix. The cut surface of the specimen (left) shows that the tumor is in the muscle layer of the cervix. On the cut surface of the tumor, muscle components and many hemorrhagic components (arrow), which are presumed to be from internal vascular structures, are seen. There is no evidence of necrosis in the specimen.
F. Microscopically (hematoxylin and eosin stain, × 40), the tumor shows numerous thick-walled vessels (arrows) with smooth muscle cells (asterisks).



male genital organ, with less than 20 cases located in uterine corpus. Uterine cervix, ovary and broad ligament are extremely unusual organs of tumor involvement (2).

The etiology of angioleiomyoma at the female genital organ is not clear, but it is thought to originate from minor trauma, chronic venous obstruction or sex hormone stimulation (3). Most of these tumors are similar with uterine leiomyoma by histology and positive hormone receptors (4). Pathologically, it has no features of atypia, mitosis, pleomorphism or necrosis (5). They seem to have a good prognosis, with a low potential for local tumor recurrence.

Despite this tumor's benignity, several cases were reported with serious complications accompanying menorrhagia leading to severe anemia or consumptive coagulopathy (6). Usually, this tumor presents with abnormal vaginal bleeding, low abdominal pain or even urinary symptoms because of its mass effect on the urinary bladder.

Radiologic appearance of angioleiomyoma is nonspecific; even larger sized tumors can undergo cystic degeneration, in which it can be also be confused with ovarian malignancy (7). Pathologic confirmation is the only final diagnosis.

In our case, the tumor presented with a well defined, large heterogeneous enhancing solid and cystic mass on an enhanced CT scan. On an MRI, it shows a well circumscribed cystic mass, which is confirmed to have a cystic degeneration on pathologic finding, with variable internal enhancing soft tissue components and some high signal intensity portions on T1-weighted imaging.

So far, typical imaging findings of angioleiomyoma are a well-defined single, unilocular mass or a single unilocular necrotic mass with an internal solid and laminated configuration. However, in view of the inability to differentiate it from other pathologies, such as leiomyosarcoma, endometrial stromal tumor or angiomyofibroblastoma, histological and immunohisto-

chemical analysis are helpful in differentiating angioleiomyoma from other tumors (8).

Mesenchymal tumors arising in the uterine cervix is rare and are mostly leiomyoma (9). However, like our case, an angioleiomyoma in uterine cervix can arise, so it should be part of the differential diagnosis of uterine and cervical masses.

The definite treatment of choice is removal of the tumor completely, either angiomyomectomy or in a total hysterectomy. In our patient, a total hysterectomy with a left salpingoophorectomy was performed. The clinical outcome between the total hysterectomy and tumor resection alone with the preservation of the uterus have not yet been studied yet and requires further investigation. No recurrence has been reported in any cases (10).

In conclusion, angioleiomyoma is an extremely rare benign tumor arising in the uterine cervix. It should be kept in mind by radiologists, clinicians and pathologists to recognize this uncommon benign tumor and differentiate it from other malignant neoplasm.

Author Contributions

Conceptualization, P.S.Y.; data curation, P.S.Y.; project administration, 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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드문 형태의 자궁 경부 혈관근종: 특이한 자기공명영상 소견을 포함한 증례 보고

황윤철 · 박서영*

자궁 경부 혈관근종은 평활근 세포와 두꺼운 벽의 혈관으로 구성되어 있는 극히 드문 양성 종양이다. 현재 자궁 경부 혈관근종에 대한 증례 보고는 거의 없는 것으로 알려져 있다. 저자들은 38세 여성의 증례를 통하여 자궁 경부 혈관근종의 초음파, 조영증강 전산화단층촬영, 자기공명영상을 포함한 영상학적, 그리고 병리학적 소견에 대하여 보고하고자 한다.

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