A case of microinvasive squamous cell carcinoma on the mucosal surface of pedunculated submucous leiomyoma protruding through cervix

Sungwook Chun\textsuperscript{1}, Gheeyoung Choe\textsuperscript{2}, Hyejin Yang\textsuperscript{3}, Yong-Beom Kim\textsuperscript{1}, Yong-Tark Jeon\textsuperscript{4}

Departments of Obstetrics and Gynecology\textsuperscript{1} and Pathology\textsuperscript{2}, Seoul National University Bundang Hospital, Seongnam, Korea

Recently, a case of cervical cancer on the mucosal surface of a pedunculated cervical leiomyoma had been reported. We experienced a similar but distinct case of microinvasive squamous cell carcinoma on the mucosal surface of a pedunculated submucous leiomyoma. A 45-year-old Korean woman underwent type I hysterectomy for heavy bleeding from pedunculated submucous leiomyoma. Histopathological examination revealed that the leiomyoma of which mucosa was covered focially by microinvasive squamous cell carcinoma. During the management of pedunculated leiomyoma protruding through cervix, caution must be taken because of, although rare instance, a possible coexisting malignancy.

Key Words: Pedunculated leiomyoma, Microinvasive cervical cancer

INTRODUCTION

Uterine leiomyomas, also known as myomas or fibroids, are the most common solid pelvic tumors in women, occurring in 20\% to 40\% of women during their reproductive years.\textsuperscript{1} They may be subserosal, intramural, or submucosal within the uterus, cervix or broad ligament or on a pedicle. Most of the leiomyomas are asymptomatic, but a few cases can cause variable symptoms. The most common symptoms associated with uterine leiomyomas are abnormal uterine bleeding and pelvic discomfort mostly caused by the mass effect. Profuse bleeding may lead to iron deficiency anemia.

Submucous leiomyomas account for only 5\% of all uterine leiomyomas, but they have clinical significance because they may cause profuse vaginal bleeding and have more potential for sarcomatous change than other types of leiomyomas. Pedunculated submucous leiomyomas rarely prolapse through the cervix.\textsuperscript{2} Recently, a case of invasive squamous cell carcinoma on the surface of a pedunculated cervical leiomyoma has been reported.\textsuperscript{3} We report herein a rare case that showed microinvasive squamous cell carcinoma on the mucosal surface of a pedunculated submucous leiomyoma protruding through cervix after type I hysterectomy due to heavy bleeding.

CASE REPORT

A 45-year-old Korean woman with previously good health presented to the emergency department with a 3-day history of profuse vaginal bleeding. She started menstruation normally on October 16, 2005, but profuse vaginal bleeding started on October 22, 2005 and continued to persist. She received no gynecological examination or a Papancicolaou smear prior to presentation. Gynecological examination revealed an adult’s fist-sized mass protruding through the external os of the cervix; which was round in shape and connected to the intrauterine cavity with a pedicle. Examination also revealed a partially ulcerated, and blood-containing surface that was covered with thin membrane. The size of the mass was $92 \times 80$ mm in
Cervical cancer on the surface of myoma

Fig. 1. Gross morphology of uterus and protruding myoma. Uterus was incised on midline and it shows pedicle attached to the fundal area. Myoma was turned downside up to show cervical canal.

Fig. 2. Gross cut section of the excised mass. The sectioning of the central mass showed the white whirling pattern of myoma and somewhat myxoid appearance. We also detected hemorrhagic focus and necrotic lesion.

Fig. 3. Microinvasive squamous cell carcinoma (depth of invasion <1 mm) on the overlying mucosa of the leiomyoma (H&E, ×200).

discharged on the 8th hospital day.

The uterus was 80 × 40 × 65 mm in size. A round cervical mass of 90 × 75 × 55 mm in diameter was found. The surface of the mass was smooth and covered with thin membrane. A submucous pedunculated leiomyoma was connected to the endometrium with a pedicle and protruded through the cervix (Fig. 1). The gross specimen of the tumor is shown in Fig. 2. Microscopically, the mass was edematous and showed hyaline degeneration. A microinvasive squamous cell carcinoma, whose invasion depth was less than 1 mm, was focally found in the overlying mucosa of the pedunculated mass (Fig. 3). There was no extension of the carcinoma to the vagina, corpus uteri, and parametrium, and both vaginal and parametrial resection margins were clear.

DISCUSSION

Submucous leiomyomas are usually asymptomatic, but they can cause symptoms such as abnormal bleeding and other symptoms such as dysuria, urgency, obstruction, and dyspareunia due to the mass effect on the adjacent organs if they grow and protrude through the cervix. Abnormal uterine bleeding occurs in about one third of patients with symptomatic uterine leiomyoma and is the most common indication for myomectomy or hysterectomy according to
the patient's age and the possibility of future pregnancies. Menstrual flow is usually profuse and prolonged. Abnormal uterine bleeding may be associated with any type of the tumor, but it seems that bleeding is more common and more severe in submucous tumors. These submucous tumors may bleed profusely during menstruation and may also bleed between periods due to passive congestion, necrosis, and ulceration of the endometrial and contralateral uterine surfaces of the tumors. In patients with uterine leiomyoma who had profuse menstruation, submucous leiomyomas should be suspected. If submucous leiomyoma is pedunculated, there is usually constant, thin, and blood-tined discharge with menorrhagia, which can lead to profuse vaginal bleeding.2

Recently, the first case of cervical cancer on the mucosal surface of a pedunculated cervical leiomyoma was reported.1 In the case report, the patient initially received radiotherapy based on the result of the colposcopic punch biopsy, and then the pedunculated exophytic mass was excised due to no response to the radiotherapy. Pathologic examination showed a leiomyoma covered with squamous cell carcinoma. It is conceivable that in the case report, the possibility of a coexisting leiomyoma within the bulky tumor mass was overlooked. If they had recognized a large leiomyoma inside the tumor, radiation therapy would not have been performed under the given circumstances.

In our case report, however, the patient initially underwent hysterectomy, which revealed a microinvasive squamous cell carcinoma on the mucosal surface of a pedunculated submucous leiomyoma protruding through the cervix. There would have been the possibility of misdiagnosis of the whole mass as a cancer if a Papanicolaou smear and colposcopic biopsy had been performed. She fortunately avoided unnecessary extensive surgery or radiotherapy. Though hypovolemic symptoms and severe anemia might have influenced the decision to perform an emergency type I hysterectomy, the clinical impression based on physical examination was a protruding and bleeding leiomyoma rather than a cervical cancer, which led to our decision to perform the surgery. Fortunately, the pathologic diagnosis of our case was microinvasive carcinoma. Because type I hysterectomy was sufficient to cure microinvasive cervical cancer, there was no need for additional adjuvant therapies in our case.

We propose the mechanism of carcinoma development on the surface of leiomyoma as follows. A small pedunculated submucous leiomyoma protrudes through the cervix as it grows. After exposure of the overlying mucosa to the environment outside the endometrial cavity, squamous metaplasia may occur on the surface of the tumor with the passage of time. Persistent human papillomavirus infection ensues, and eventually squamous cell carcinoma develops.

In summary, attention should be paid to a cervical mass with bleeding so as not to overlook a possible hidden malignancy and to avoid excessively wide excision in benign tumors.3

REFERENCES

자궁경부를 통해 돌출된 육경성 점막하 자궁근종에서 발견된 미세침윤 편평상피세포암 1예

본당서울대학교병원 성부인과, 병리과
전성욱1 · 최기영2 · 양해진1 · 김용범1 · 전용탁1

자궁근종은 가임기 여성에서 가장 흔하게 생기는 결반 과정 중의 정상, 근육내 또는 점막하 근종으로 나타며, 그 외 자궁경부나 자궁관간막 또는 육경성으로 발생할 수 있다. 최근 육경성 자궁경부 근종의 표면에서 발생한 침윤성 편평상피 세포암이 보고된 적이 있는데, 저자들은 이와 유사하면서도 다른, 과다 절제술에 의해 복식 자궁절제술을 시행한 자궁경부 발이 돌출된 육경성 점막하 자궁근종의 표면에서 발생한 미세 침윤성 편평상피 세포암 1예를 경험하였기에 간단한 문헌 고찰과 함께 보고하는 바이다.

중심단어: 육경성 점막하 자궁근종, 미세 침윤성 편평상피 세포암