Case Report

Uterine Fibroid in a 16-Year-Old Adolescent Managed with a Fertility-Sparing Approach: A Case Report and Review of the Literature

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A B S T R A C T

Background: Uterine fibroids are the most common benign tumor of the uterus in adult women. However, it is exceedingly rare in adolescents.
Case: A 16-year-old girl presented with severe anemia and abnormal uterine bleeding. MRI of the pelvis demonstrated a 9-cm mass within the uterus concerning for leiomyoma or leiomyosarcoma. A fertility preserving myomectomy led to the final diagnosis of a benign uterine fibroid.
Summary and Conclusion: Despite the rare incidence of leiomyomas in adolescents, it is imperative that it is included in the differential diagnosis when young women present with abnormal uterine bleeding. A less common diagnosis to consider is leiomyosarcoma; however, this is histologically diagnosed. Thus, a fertility-preserving myomectomy permits for a diagnostic opportunity to rule out a potential malignancy.
Key Words: Adolescent, Abnormal uterine bleeding, Fibroid, Laparotomy, Myomectomy, Uterine leiomyoma, Leiomyosarcoma

Introduction

Uterine fibroids are the most common benign neoplastic uterine tumor. They occur in approximately 20-30% of women between the ages of 30 and 50 years. However, they are rare in adolescents, with an incidence of less than 1%.[1] Only 25 cases in this age group have been reported (Table 1).[2–26] Uterine leiomyosarcomas are an even rarer occurrence, with only 1 case documented among adolescents. Because of the low sensitivity of pelvic imaging and tumor markers, making a preoperative diagnosis of a leiomyosarcoma can be challenging, and ultimately relies on histological examination after myomectomy or hysterectomy.[27]

Currently, there are limited guidelines regarding treatment approaches to uterine masses in adolescents. Fertility preservation is an important consideration in this population; thus, a myomectomy offers an excellent management approach, permitting an intraoperative frozen section if concerns of malignancy exist preoperatively.

In this case, we present a 16-year-old nulliparous female patient with acute blood loss anemia, abnormal uterine bleeding, and pelvic magnetic resonance imaging (MRI) findings concerning for leiomyosarcoma, whose postoperative histologic diagnosis was a benign uterine fibroid. We also present a review of the literature on the management of leiomyomas in adolescents.

Case

A 16-year-old nulliparous female patient was admitted to our hospital with symptomatic acute blood loss anemia and heavy menstrual bleeding. She had an unremarkable medical, surgical, social, and family history, including no personal or family history of bleeding disorders. Menarche was at age 12 years, and menses occurred monthly with 5 days of bleeding that was increasingly heavy over the past 4 months. On the heaviest days, she would saturate a super-absorbency tampon and super-absorbency pad hourly. She had never been sexually active. This was her second hospitalization within 3 months requiring blood transfusion. Her first admission was at another facility 3 months prior, where she required a blood transfusion and was started on daily triphasic combined oral contraceptive pills (COCs), which did not decrease her menstrual blood loss.

On examination, Tanner staging was appropriate for age. The patient declined a pelvic examination. Her body mass index was 28.1 kg/m^2. She was mildly tachycardic; otherwise vital signs were stable. A pregnancy test was negative. Laboratory analysis showed severe anemia with a hemoglobin of 4.0 g/dL and iron studies consistent with iron-deficiency anemia. Studies for disorders of hemostasis, von Willebrand disease, and thyroid dysfunction were performed, and results were reported normal. Transabdominal pelvic ultrasound demonstrated a uterus measuring 14.5 × 9.9 × 10.0 cm, with a heterogenous area

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Table 1
Case Reports of Leiomyomas in Adolescents

Authors, Reference	Year of Publication	Age (yr)	Presentation	Myoma Size (cm)	Management	Outcome
Wisot et al[2]	1969	15	AUB	12 × 7 × 6	Abdominal myomectomy	No evidence of disease at 6 mo
Augensen[3]	1981	15	AUB, pelvic mass, urinary retention	10	Abdominal myomectomy	No evidence of disease at 1 yr
De Rooy et al[4]	1986	15	Abdominal pain and mass, hemorrhagic shock	16.5 × 11 × 8.5	Abdominal myomectomy	No evidence of disease at 5 yr
Horejsi et al[5]	1988	15	Not specified	Not specified	Abdominal hysterectomy, BSO	Not specified
Heimer et al[6]	1991	15	Abdominal pain, fatigue	20 × 12	Abdominal myomectomy	No evidence of disease at 6 mo
Morad et al[7]	1993	15	AUB, abdominal mass	7	Abdominal myomectomy	Pregnant after 8 mo, delivered by CD
Fields et al[8]	1996	16	22 wk pregnant, periumbilical pain	Not specified	Expectant management	Not specified
Nguyen Duc et al[9]	2003	15	AUB, abdominal mass, anemia	Not specified	Not specified	Not specified
Bekker et al[10]	2004	15	AUB, abdominal pain	25	Abdominal myomectomy	Not specified
Graspa et al[11]	2006	16	AUB, abdominal pain	18 × 20 × 24	Abdominal myomectomy	No evidence of disease at 6 yr
Diesen et al[12]	2008	14	Dysmenorrhea, abdominal distention	15 × 15 × 10	Abdominal myomectomy	No evidence of disease at 3 mo
Perkins et al[13]	2009	17	Pelvic mass	10	Abdominal myomectomy	Pregnant after unspecified time, delivered at term by CD
Karim et al[14]	2010	16	Pelvic mass, increased abdominal volume	25 × 15 × 10	Abdominal myomectomy	Not specified
Tsili et al[15]	2010	16	Abdominal pain and distension	Multiple, largest 13	Laparoscopic myomectomy	Recurrence of multiple myomas at 2 yr
Khorrami et al[16]	2011	17	AUB, refractory anemia	2.2 × 4.2 × 1.7	Hysteroscopic myomectomy	No evidence of disease at unspecified time interval
Taskin et al[17]	2011	16	Pelvic mass, mass protruding through vagina	4 × 4	Hysteroscopic myomectomy	No evidence of disease at 9 mo
Wright et al[18]	2011	14	AUB, abdominal pain, increased abdominal volume	16	Abdominal myomectomy	Asymptomatic 3 cm myoma at 1-yr follow up
Naiditch et al[19]	2011	15	Pelvic mass, abdominal pain, associated ovarian teratoma	6	Abdominal myomectomy and teratoma resection	Not specified
Perez-Colon et al[20]	2011	15	AUB	5	Not specified	Not specified
Maggiore et al[21]	2013	14	AUB, abdominal and back pain	13 × 10 × 10	Abdominal myomectomy	No evidence of disease at 2 yr
Kayadibi et al[22]	2014	15	Pelvic mass, increased abdominal volume	17 × 20 × 16	Abdominal myomectomy	No evidence of disease at 6 mo
Salehi et al[23]	2016	15	Pelvic pain	17.5 × 14.8 × 4.5	Laparoscopic myomectomy	Not specified
Zigman et al[24]	2018	16	Vaginal discharge, pelvic mass	8	Vaginal myomectomy	Not specified
Morita et al[25]	2019	13	Abdominal distension	11	Laparoscopic myomectomy	No evidence of disease at 18 mo
Kumura et al[26]	2020	12	AUB	2.9 × 2.1 × 2.7	Hysteroscopic myomectomy	Not specified
Murphy et al	2020	16	AUB, anemia, pelvic mass	9	Abdominal myomectomy	No evidence of disease at 5 mo

AUB, abnormal uterine bleeding; BSO, bilateral salpingo-oophorectomy; CD, cesarean delivery.

within the fundus measuring 8.7 cm in diameter. Ovaries were unremarkable bilaterally. Pelvic MRI identified an enlarged uterus with a complex heterogeneous structure measuring 9 cm in diameter that appeared to be compressing but separate from the endometrium (Fig. 1). Modest enhancement at the periphery of the mass was noted. Upon review of the imaging with radiology, the findings were concerning for leiomyoma and possibly leiomyosarcoma. Interventional radiology was consulted for a biopsy of the mass for further evaluation; however, because of elements of central necrosis and peripheral enhancement that were suspicious for leiomyosarcoma, the decision was made to avoid a biopsy to prevent possible peritoneal seeding.

Heavy menstrual bleeding resolved within 1 day of starting a monophasic COC taper (norethindrone 1 mg/ethinyl estradiol 35 μg TID for 3 days, BID for 2 days, and daily for 1 week). Once anemia resolved with blood transfusion and iron supplementation, various treatment approaches were discussed on an outpatient basis. With the goal of fertility preservation, the decision was made to proceed with a myomectomy via laparotomy with intraoperative frozen section. Gynecology oncology was on standby. Intraoperative findings included a 12-cm anterior

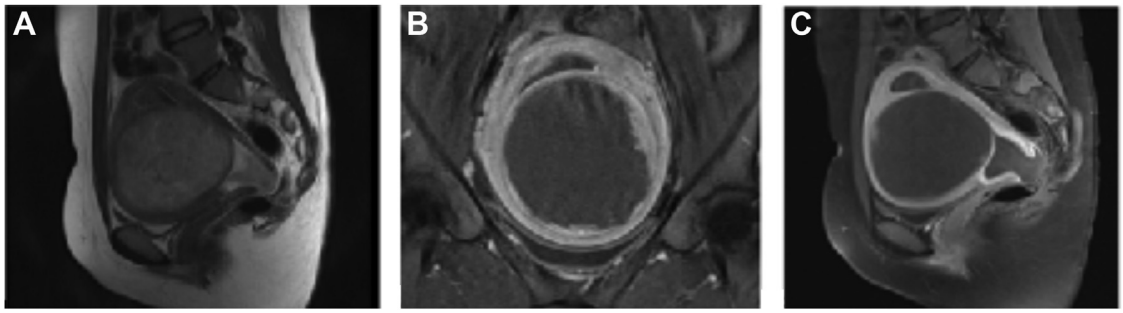

Fig. 1. (A) T2-weighted sagittal magnetic resonance image of pelvis, demonstrating uterine mass with central necrosis. Coronal (B) and sagittal (C) T1-weighted magnetic resonance image of the pelvis, with heterogeneous signal and ring enhancement along periphery.

intramural mass with a submucosal component (Fig. 2). Fallopian tubes and ovaries appeared normal bilaterally. Regarding technique, dilute vasopressin was injected into the anterior uterine wall prior to uterine incision to aid in hemostasis. The mass was dissected bluntly from the myometrium and shelled out intact; care was taken to avoid structural disruption of the mass. The entire specimen was sent for frozen pathology, which demonstrated an infarcted leiomyoma. Estimated blood loss was 50 mL. Final pathology results demonstrated a benign leiomyoma with extensive infarction and focal dystrophic calcification, weighing 418 g.

The patient was discharged home on postoperative day 1 after meeting postoperative milestones. As she was no longer anemic and the source of her abnormal uterine bleeding (AUB) was removed, COCs were discontinued to evaluate her natural menstrual cycles. Because of the size and location of the uterine incision, the patient was extensively counseled that she would require a cesarean delivery as the mode of delivery for future pregnancies to reduce the risk of uterine rupture.

At a 5-month follow up visit, she had lighter regular menses and denied dysmenorrhea. Follow-up pelvic ultrasound demonstrated a normal-appearing uterus without masses.

Summary and Conclusion

In this report, we describe a rare and infrequently diagnosed case of a uterine fibroid in an adolescent who presented with AUB and symptomatic anemia. AUB in this age group is most commonly due to anovulation secondary to an immature hypothalamic–pituitary–ovarian axis.[28] Although structural causes are less common and malignancy is extremely rare, the differential diagnosis remains similar to that in adult women. Uterine fibroids are found in up to one-third of adult women with AUB, compared to less than 1% of adolescent women.[1] Furthermore, a gynecological mass in an adolescent is more likely to be of ovarian origin or a Mullerian anomaly. Uterine sarcomas are exceedingly rare among adult women, representing 3% of all uterine cancers, the most common being leiomyosarcoma.[27] They often present with symptoms similar to benign fibroids, including heavy menstrual bleeding, pain, a pelvic mass (more rapidly growing in the case of sarcomas), and pressure symptoms dependent on the size and location of the mass.[1,27] Our patient described a change in her menstrual flow in the preceding 4 months, possibly suggesting a more recent and rapid growth of the mass.

Ultrasound imaging is the preferred method for investigating pelvic structures. Transabdominal ultrasound is preferred for the adolescent population, particularly if they are sexually inactive, compared to a transvaginal ultrasound, which is preferred for adults.[26] Although sonography remains first line, it can be limited in soft tissue characterization and evaluation of an enlarged uterus. Alternatively, MRI can provide further characterization of uterine abnormalities. However, differentiating between leiomyoma and leiomyosarcoma remains challenging, as no

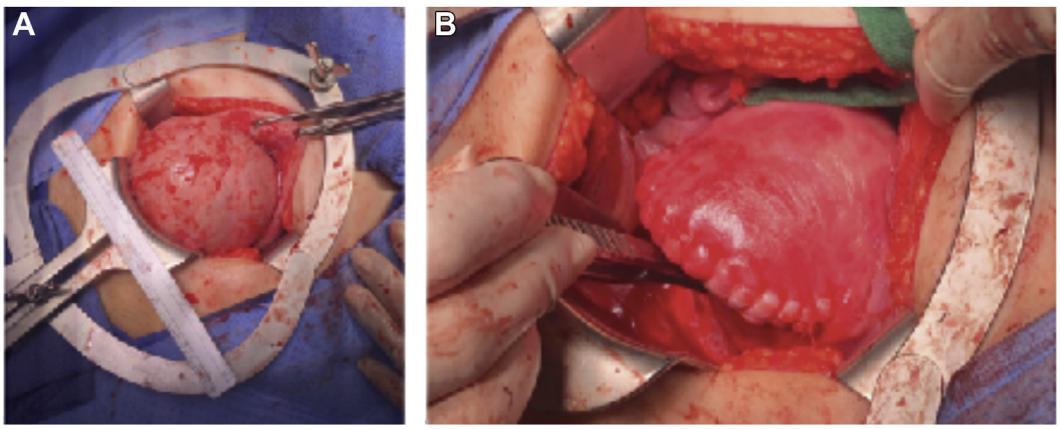

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Fig. 2. (A) Uterine leiomyoma dissected bluntly from myometrium. (B) Fibroid shelled out intact. Uterine closure was performed with excellent hemostasis.

pathognomonic imaging criteria have been reported.[29] It has been suggested that features of malignancy on MRI are the presence of irregular margins, necrosis, and rapid growth.[30] Additionally there are no tumor markers specific to leiomyosarcomas.[27]

As there are no definite guidelines for the management of symptomatic uterine fibroids for adolescents, adult treatment options, including medical and surgical therapies, are extrapolated for use in younger patients. Medical management, such as contraceptive steroids, selective progesterone receptor modulators, gonadotropin-releasing hormone agonists, and aromatase inhibitors, have not been formally studied in the adolescent population. The use of COCs for managing fibroid growth is limited[31]; studies have demonstrated that estrogen and progesterone can improve bleeding symptoms and decrease the risk of developing clinically significant leiomyomas.[26] Surgical management includes uterine artery embolization, myomectomy, or hysterectomy. In our case, we prescribed COCs to aid in minimizing menstrual bleeding and in allowing time to recover from severe anemia prior to proceeding with an abdominal myomectomy. A fertility-sparing myomectomy is an appropriate approach for adolescents with large symptomatic myomas, even if imaging and history are concerning for leiomyosarcoma. This provides an opportunity for intraoperative frozen section to histologically assess for malignancy, rather than proceeding with a hysterectomy based on a presumptive diagnosis. Frozen section in the analysis of gynecologic specimens has a generalized specificity of 96%[32]; more specifically, the frozen section analysis of leiomyomas have low false-negative rates and negligible false-positive rates.[33] A limiting factor of frozen section is ensuring sampling of target tissue and obtaining an adequate volume of tissue[33]; therefore, we sent the entire specimen to provide an adequate sample for the most accurate diagnosis.

Among the previously reported cases as summarized in Table 1, myomectomy was also the most common surgical treatment choice.[1] It can be approached via hysteroscopy, laparoscopy, or laparotomy, depending on the location, number, and size of the fibroids. In our case, we proceeded with a laparotomy because of the size of the uterine fibroid and our concern about avoiding morcellation with the possibility that it was a leiomyosarcoma, as morcellation of sarcomas can be associated with disease spread and decreased survival rates.[33] Counseling of patients regarding uterine fibroid management should include future fertility desires, recurrence following surgery, the importance of antenatal visits during pregnancy, and the possible need for cesarean delivery.

Fibroids in adolescents have been documented as young as age 12 years,[26] and the most common presenting complaint among young women is AUB. Although the most common surgical approach to management in the literature is abdominal myomectomy, there are 3 cases that describe a laparoscopic approach. Of all 26 case reports, follow-up ranged from 3 to 18 months, and only 1 case describes a recurrence of leiomyomas at 18-month follow-up. Interestingly, there is 1 case report of leiomyosarcoma, in a 16-year-old patient, that presented as a recurrent mass after an abdominal myomectomy for a benign leiomyoma 6 months prior.[34] This also highlights the importance of follow-up to evaluate for recurrence.

In conclusion, we present a 16-year-old girl who presented with abnormal uterine bleeding and a uterine mass concerning for possible leiomyosarcoma, resulting in the diagnosis of a uterine fibroid after a fertility-sparing myomectomy. Although leiomyomas are rare in such an age group, pelvic ultrasound should be considered in adolescents with abnormal uterine bleeding refractory to medical management.

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