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Adenomyoma presenting as a primary subserosal pedunculated exophytic mass

Mary Louise Margaret Mamaclay Javier¹, Agnes L. Soriano-Estrella²

Abstract:

Adenomyoma is a benign gynecologic condition affecting women in their late reproductive years. Common clinical presentations include pain, particularly dyspareunia and dysmenorrhea, abnormal uterine bleeding, and infertility. Majority, however, may be asymptomatic. Various presentations of adenomyoma have been written in the literature, but exophytic subserosal growths have rarely been reported. More commonly, it involves the endometrium with invagination into the myometrium. We present a rare case of a primary subserosal pedunculated adenomyoma with no evidence of adenomyosis in a nulligravid premenopausal woman who underwent a uterine-sparing surgery and an extensive colonic resection with colostomy.

Keywords:

Colostomy, exophytic adenomyoma, pedunculated adenomyoma, subserosal adenomyoma, uterine-sparing surgery

Introduction

Adenomyosis is a benign gynecologic condition that is found in about 60% of specimens from women who had undergone hysterectomy for various reasons.^[1] Historically, it has been found to affect women above 40 years old. Common risk factors include increasing age and parity as well as prior cesarean section or uterine surgery.^[1,2]

Focal adenomyosis, a well-circumscribed, predominantly solid mass, is called an adenomyoma. Although such finding is common, there have been very few case reports published about adenomyomas with subserosal exophytic growth. This study highlights a case of subserosal exophytic adenomyoma in a nulligravid who underwent a uterus-sparing surgery and an extensive colonic resection. Moreover,

this study aims to review the common clinical features of adenomyoma exhibiting subserosal exophytic growth as well as compare and contrast its diagnostic features and management options.

Case Report

A gravida 1 para 0 (0-0-1-0) woman in her 30s with no known comorbidity nor past surgery was admitted for elective operation due to a gradually enlarging pelvic mass. Her family history is pertinent for rectal cancer in her mother. Her personal, social, and menstrual history is unremarkable. However, she had a spontaneous abortion at about 12-week age of gestation last August 2019, which did not warrant a curettage.

The patient's history started 5 years before consult when she had an incidental finding of a pelvic mass on whole-abdomen ultrasound. She did not complain of abnormal uterine bleeding, pelvic pressure, weight loss, anorexia, and bladder or bowel changes. A transvaginal ultrasound was

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¹Department of Obstetrics and Gynecology, Philippine General Hospital, University of the Philippines Manila,

²Department of Obstetrics and Gynecology, College of Medicine, Philippine General Hospital, University of the Philippines Manila, Manila, Philippines

Address for correspondence:

Dr. Mary Louise Margaret Mamaclay Javier, Philippine General Hospital, Taft Avenue, Manila, Metro Manila 1000, Philippines. E-mail: mmjavier2@up.edu.ph

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then performed revealing a 5.0 cm × 3.0 cm × 3.0 cm pedunculated subserous myoma uteri. Her attending physician at this point advised close follow-up because surgery was not yet indicated. A follow-up ultrasound [Figure 1] done 2 months before admission showed a well-circumscribed hypoechoic subserous uterine mass measuring 10.7 cm × 6.6 cm × 10.0 cm at the posterior isthmus area (FIGO Grade 7) with a pedicle measuring 3.2 cm long attached to the posterior isthmus area of the uterus. The subendometrial halo was intact, and there were no adnexal masses seen. She was then advised surgical intervention hence admission.

Investigations

On admission, she was ambulatory and had normal and stable vital signs. She was obese but had a normal systemic physical examination. On internal examination, she had normal external genitalia with smooth nulliparous vagina. Her cervix was smooth and firm and was pulled superiorly. Her uterine corpus was enlarged to 14–16-week size. There were no adnexal masses or tenderness. On rectovaginal examination, she had good sphincter tone and intact rectal vault, with the inferior pole of the said posterior isthmus mass palpable at the cul-de-sac. Bilateral parametria were smooth and pliable. Her preoperative laboratory investigation was unremarkable. Preoperative diagnosis was myoma uteri, pedunculated, subserous.

Treatment

The patient underwent an exploratory laparotomy and myomectomy under regional anesthesia. Intraoperatively, there was a 15.0 cm × 10.0 cm × 8.0 cm smooth, lobulated, pedunculated tumor, with its pedicle measuring 3.0 cm × 0.5 cm attached to the posterior aspect of the uterus with dense adhesions to the middle extending to the upper mesorectum [Figure 2]. An intraoperative referral to colorectal surgery was

done due to dense adhesions of the uterine mass to the mesorectum. To adequately resect the mass, the surgical team did a Hartmann's procedure with its distal margin at mid-rectum and proximal margin at the distal sigmoid. The end stoma was exteriorized at the left lower quadrant of the patient's abdomen. Postoperative diagnosis was a uterine fibroid tumor with extension to the mesorectum. Cut section revealed a creamy, tan, trabeculated mass with cystic spaces filled with blood [Figure 3]. Histopathology showed a pedunculated subserous adenomyoma with hyaline degeneration and adhesions to the intestines but without evidence of vascular invasion [Figure 4].

Outcome and follow-up

The patient had an uneventful postoperative status and was discharged immediately. She was due for colostomy reversal last April 2020; however, due to the COVID-19 pandemic, this procedure was put on hold.

Discussion

Adenomyosis has been increasingly diagnosed among younger women using various imaging modalities. Histopathology, as a gold standard in diagnosis, reveals the presence of heterotopic endometrial glands and stroma in the uterine myometrium.^[1,2] Adenomyoma refers to a well-circumscribed predominantly solid mass of endometrial tissue interspersed within the myometrium.^[3,4]

The histogenesis of adenomyosis reveals the invagination of the basal layer of the endometrium into the myometrium.^[1,3] Procedures that may cause trauma to the endometrial-myometrial junction or the junctional zone such as cesarean section or uterine surgery or conditions such as increasing parity are considered to be significant risk factors.^[3] Such irregularities can be

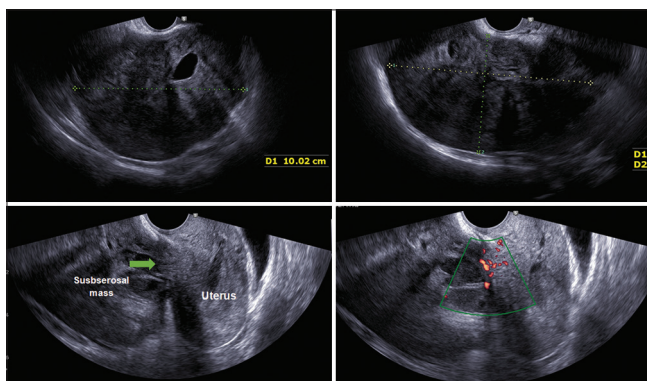


Figure 1: Transvaginal ultrasound findings. (Left to right, upper row) anteroposterior view of the posterior myometrium. Transverse view of the posterior myometrium. (Left to right, lower row) pedicle (arrow) of the subserosal uterine mass attached to the posterior isthmus area of the uterus. Color flow mapping of the pedicle showing minimal vascularity (color score = 1)

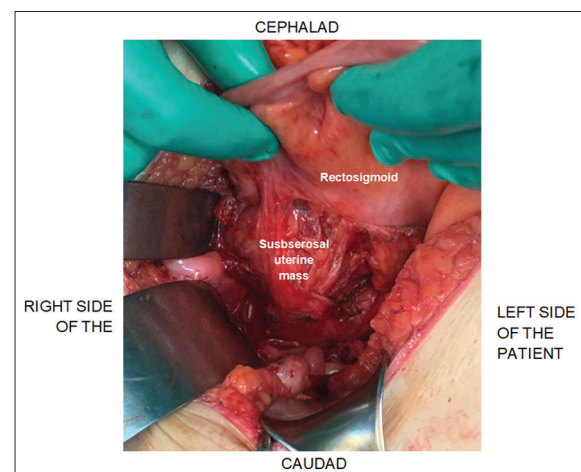


Figure 2: Subserosal exophytic mass that is closely adherent to the posteroinferior border of the rectosigmoid colon

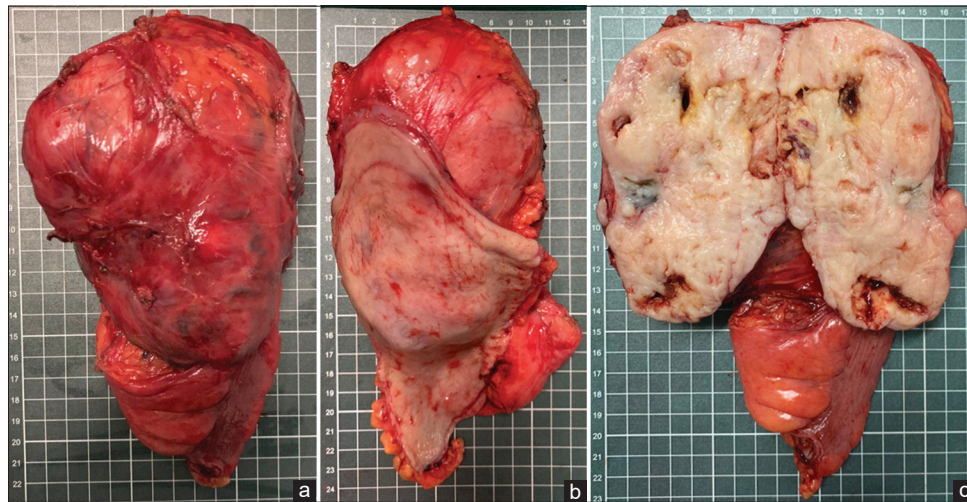


Figure 3: Anterior (a) and posterior (b) views of the subserosal exophytic mass and its cut section (c) revealing a creamy tan, trabeculated with areas of cystic spaces containing blood

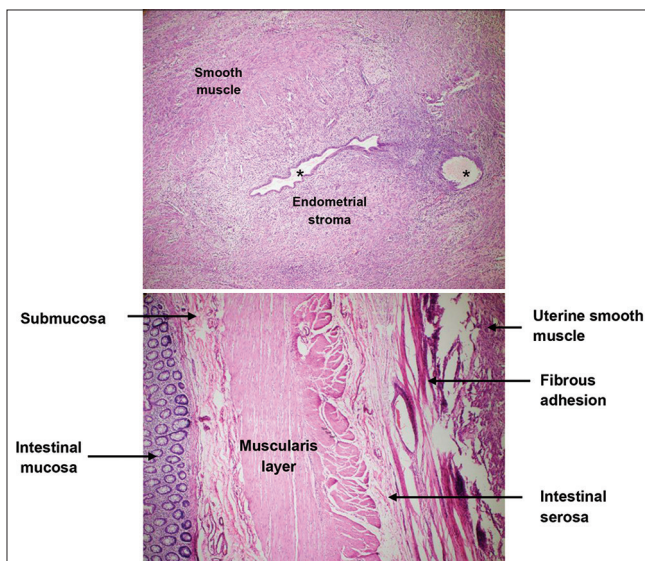


Figure 4: Final histopathologic diagnosis consistent with pedunculated subserous adenomyoma with hyaline degeneration showing (top) endometrial glands (*) and stroma surrounded by myometrial smooth muscle with (bottom) adhesions to the intestines. Intestinal glands are well represented in the leftmost side of this photomicrograph while the rightmost side represents the myometrial tissue of the mass

seen using imaging modalities such as transvaginal ultrasonography which has a sensitivity of 53% up to 89% and a specificity of 50%–89%. Magnetic resonance imaging (MRI) has been found to be more sensitive and specific at 88%–93% and 66%–91%, respectively.^[1] The diagnosis of adenomyosis pointing to such zonal irregularity is also found in asymptomatic women.^[3] Furthermore, many women have been found to have ectopic endometrial glands and stroma but with intact junctional zone. Studies have suggested the role of hematogenous spread, immunologic mechanisms, and multipotential perivascular cells in the pathogenesis of adenomyosis.^[3]

To investigate the patterns of clinical presentation, diagnosis, and management of subserosal adenomyoma, various databases such as PubMed, EBSCOhost, SCOPUS, and Ovid were used to search for articles using the word string “subserosal polypoid adenomyoma,” OR “subserosal adenomyoma,” OR “pedunculated adenomyoma,” OR “exophytic adenomyoma.” Excluding duplicate articles, the search revealed 19 articles published from 1996 to 2020. Nine articles were excluded because they pertained to other topics. Of the ten remaining articles, there were five case reports, one case series, and four retrospective reviews about imaging and molecular biologic features of adenomyoma.

All five case reports [Table 1] involved six patients aged 32–52 years with a mean age of 43 years who were all symptomatic.^[5–9] Common symptoms were vaginal bleeding, severe recurrent dysmenorrhea, and chronic pelvic pain. Unusual presentations involved sudden onset of severe lower abdominal pain and vulvar pruritus.^[6–9] Moreover, patients who were reported had no identifiable risk factors.^[1,2,5–9] Since some subserosal masses may continue to grow and outgrow its blood supply, they may develop pedicles and extend into adjacent organs like the rectum.

Our index patient had an incidental finding of a subserous mass on imaging that was preoperatively diagnosed as a myoma. This finding is usual. In fact, two out of the six reported cases had a histopathologic diagnosis of subserosal adenomyoma that were mistakenly diagnosed as myoma on imaging.^[5,7] Some may also mimic ovarian tumors based on location and elevated serum levels of cancer antigen 125.^[8,9]

Laparoscopic resection with morcellation is a common management option.^[5–7] However, more

Table 1: Summary of case reports

Year of publication and author	Patient profile and presenting symptom	Diagnostics	Preoperative diagnosis	Management	Histopathologic diagnosis
Jung <i>et al.</i> (2002) ^[9]	Case 1: 43 years old with 1-year history of menorrhagia and dysmenorrhea Case 2: 52 years old with vulvar pruritus	Case 1: Transvaginal ultrasound: Submucosal leiomyoma in the posterior wall of the lower uterine segment Case 2: Transvaginal ultrasound: Mass on the left side of the pelvis	Case 1: Submucosal myoma Case 2: Ovarian tumor	Case 1: Total abdominal hysterectomy Case 2: Total abdominal hysterectomy with left salpingo-oophorectomy	Case 1: Adenomyosis Case 2: Adenomyosis; left endometriotic cyst; subserosal adenomyoma with adipose metaplasia
Sakai and Matsukuma (2002) ^[6]	48 years old G2P0 (0-0-2-0) with severe lower abdominal pain	Transvaginal ultrasound: Solid and cystic mass adjacent to the uterus MRI: Well-circumscribed solid mass containing multicystic low-intensity areas contiguous with the right side of the uterine wall CA 125 was elevated at 221 U/mL	Ovarian cancer	Total hysterectomy with bilateral salpingo-oophorectomy	Cystic adenomyoma with epithelial metaplasia; intramural leiomyoma and adenomyosis; no pelvic endometriosis, bilateral ovaries and fallopian tubes were unremarkable
Takeda <i>et al.</i> (2013) ^[6]	32 years old nulligravid with sudden onset lower abdominal pain	Pelvic MRI: Heterogenous mass with mural and subserosal growth in the posterior uterine wall; small left endometriotic cyst; single adenomyomatous nodule with extensive exophytic subserosal growth; intact junctional zone CA 125 was elevated at 202.9 U/mL	Adenomyoma over segmental adenomyosis; adenomyoma with mural and exophytic subserosal growth	Laparoscopic excision, morcellation, intercede application, and postoperative daily 150 µg desogestrel and 30 µg ethinyl estradiol	Adenomyoma
Elshafie <i>et al.</i> (2013) ^[5]	46 years old with vaginal bleeding	Pelvic ultrasound: Multiple leiomyomata	Myoma uteri	Laparoscopic myomectomy then subsequent total abdominal hysterectomy with bilateral salpingo-oophorectomy and bilateral pelvic lymphadenectomy	Fibroids; adenomyoma; mild and focal moderate cytological atypia with sparse mitotic figures suggesting transformation to a low-grade Mullerian adenosarcoma; no evidence of adenomyosis
Calgana <i>et al.</i> (2015) ^[7]	39 years old with severe recurrent dysmenorrhea with chronic pelvic pain	Transvaginal ultrasound and pelvic MRI: Subserosal myoma CA 125 was elevated at 122.4 U/mL	Myoma uteri adenomyosis	Laparoscopy with radical resection of the pedunculated subserosal masses; morcellation	Pedunculated cystic adenomyoma

MRI: Magnetic resonance imaging, CA: Cancer antigen

radical procedures such as total hysterectomy with bilateral salpingo-oophorectomy and bilateral pelvic lymphadenectomy may be done on suspicious-looking masses.^[5] Based on the histopathologic findings, such masses may reveal cellular atypia with sparse mitotic figures pointing to a transformation into a low-grade Mullerian adenosarcoma.^[5] This finding suggests that patients may eventually develop a malignant sarcomatous lesion from a benign tumor. Indeed, subserosal masses may mimic a lot of related diseases. Thus, a careful preoperative evaluation, sensitive and specific imaging, and application of

relevant biomarkers are all important. Although adenomyoma is a complex disease and ascertaining the diagnosis is difficult without histopathology, it is usual that diagnosis may only be arrived at intraoperatively.

Our index patient was preoperatively diagnosed with subserosal myoma uteri based on ultrasound. Knowing that MRI has a better sensitivity and specificity, the diagnosis of adenomyoma may have been considered if it were done to our patient. In a review of seven cases of adenomyoma, two cases of subserosal adenomyoma did not have a concomitant adenomyosis based on MRI.^[9]

Although solid portions may either be homogenous or heterogenous, MRI with high-intensity signal on T1-weighted images should include a differential diagnosis of adenomyoma, whether adenomyosis is concomitantly present or not.^[10,11]

Because asymptomatic premenopausal women are reluctant about radical surgical procedures, uterus-sparing operative techniques have been reviewed.^[4] Complete versus partial excision of adenomyoma had comparable results based on dysmenorrhea reduction (82% vs. 81.8%), menorrhagia control (68.8% vs. 50%), and pregnancy rate (60.5% vs. 46.9%), respectively.

Although the uterus of our index patient had been spared, she underwent an extensive colonic resection resulting in a colostomy. While histopathology revealed no evidence of vascular invasion to the rectosigmoid, further dissection of the subserosal uterine mass away from the bowels would likely pose technical difficulty which may lead to further injury. Such masses may present a diagnostic dilemma and more extensive evaluation may only be done intraoperatively.

Learning points/take-home messages:

- Exophytic subserosal growths of adenomyoma have rarely been reported
- A uterine-sparing surgery may be done for premenopausal women who wish to preserve fertility
- A possible colonic resection may be warranted for pelvic masses with exophytic subserosal growths
- Careful surgical planning must be done in order to counsel patients regarding extensive surgeries.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Authorship contributions

Mary Louise Margaret M. Javier - Involved in the conceptualization, methodology, resources, data curation, writing –original draft, review and editing.

Agnes L. Soriano-Estrella - Involved in the conceptualization, writing –review and editing, supervision.

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Conflicts of interest

There are no conflicts of interest.

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