

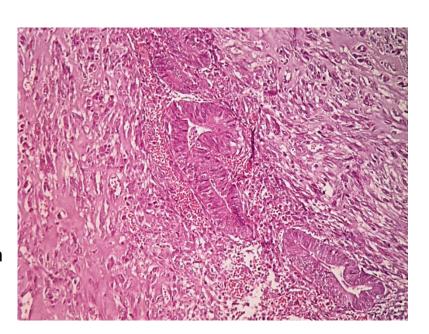
A huge pedunculated polypoidal subserous adenomyoma, case report

 $El Sharkawy S_a$, Abdelzaher E_b

a Obstetrics and gynecology department ,Faculty of medicine , Alexandria university , Alexandria , Egypt

_b pathohology department ,Faculty of medicine , Alexandria university , Alexandria , Egypt.

For farther inquiries, please contact: samirsuzan6@gmail.com



background

Adenomyosis is traditionally defined as a tumor-like benign growth of endometrial glands and stroma into the myometrium, surrounded by hypertrophic and hyperplastic myometrial cells. This probably happens by invagination of the basalis layer of the endometrium facilitated by antiapoptotic activity and associated with relative hyper-oestrogenic environments. (1)

Adenomyosis is classified into two main types; 1) diffuse Adenomyosis which causes symmetrical enlargement of the uterus and; 2) focal type (adenomyoma) which causes asymmetrical uterine enlargement. Rarely, focal adenomyomas are presented as polypoidal form which protrude outside the uterus or inside its cavity. (2)

Mostly, adenomyotic cases are presented with menorrhagia, dysmenorrhoea and chronic pelvic pain, and are usually associated with endometrial hyperplasia, endometriosis and fibromyomas. Different modalities for diagnosis of extrauterine masses are available; like Transvaginal ultrasound, computerized tomography (CT) and magnetic resonance imaging (MRI), but definite diagnosis polypoidal Adenomyosis cannot confirmed be preoperatively. (3)

case

In 2010, the 28 years old Miss R. R. complained of vague pelvic pain associated with urinary symptoms (dysuria, frequency and urgency). She visited her urologist how referred her to our clinic for checking. She performed an abdominal ultrasound and was diagnosed as having a subserous fibroid (5×6 cm) which compress the urinary bladder causing its irritation. She refused any operative interference at that time for fear of postoperative adhesions as she was not married or had children yet. She was advised to follow up the fibroid every year by ultrasound and to take symptomatic treatment for her complains.

For the next 8 years, she performed multiple ultrasound examinations that revealed gradual increase in the size of the fibroid (7×5.5 cm in 2013, 9.4×8.1 cm in 2017 and 10×8 cm in 2018). It was described as subserous type and extended up to the level of umbilicus, another two interstitial fibroids developed over time with an endometrioid cyst in the right ovary (7×5 cm in maximum dimensions). The patient was so afraid that she might have to perform hysterectomy and would not be able to get pregnant or to have kids in the future, to the extent that she started psychotherapy for obsessive compulsive disorder (OCD) in 2013.

In 2018, her pelvic pain reached unbearable level, associated with dysmenorrhea, dysuria, dyscasia and metrorrhagia. She decided to take the risk and go to the operative room to remove the tumors and the uterus if necessary.

She was operated on 24th of March 2018, with a lower transverse abdominal (pfannenstial) incision and under general anesthesia. Her preoperative hemoglobin level was 13.9 gram/dl and had normal kidney function tests. There was a huge subserous mass attached to the uterine fundus with a narrow pedicle, its size was around 15× 8×6.5 cm, firm in consistency, smooth lobulated pale surface with filmy omental adhesions lying over. Dilated veins and multiple small hemorrhagic cysts were scattered on its surface looking like sarcoma. The mass was removed with the associated multiple interstitial myomas and a right ovarian cystectomy was done for the endometrioma.

Histo-pathological examination of the large mass revealed that it was an adenomyomatous proliferation made up of bland cystically dilated endometrial glands and stroma surrounded by fascicles of hyperplastic smooth muscle cells. Thanks to God, No evidence of malignancy was detected. Other associated specimens were endometriotic cyst and multiple leiomyomas. On the basis of operative, gross pathological and histo-pathological features, this lesion was considered as an extremely rare type of Exophytic adenomyomas.

Postoperative period was smooth. Later, the patient was advised to take combined oral contraceptive pills on regular bases to treat endometriosis related pain and prevent recurrence of endometriomas. Few months after being operated, Miss R. R. was relieved from chronic pelvic pain and did not need farther treatments for OCD too.

discussion

Subserosal adenomyomas are extremely rare lesions that almost always are misdiagnosed as different masses like fibroids or ovarian cancers. (4) Exophytic adenomyomas has been recognized for the first time by Sir Mazur et al. in 1981 and regarded it as a benign lesion. (5) cystic Adenomyomas were diagnosed in 24% of hysterectomy specimens, but lesions were less than 5 mm in maximum diameter (6), while larger lesions are rare. (7-11) After reviewing literature, the largest two polypoidal adenomyomas reported were around 10 cm in maximum diameter. (4,12) According to our knowledge, this is the largest subserous polypoidal adenomyoma reported up till now, with its enormous size (15× 8×6.5 cm). We suppose that the unique history of the patient (who put up with severe pelvic pain for almost a decade) is the main factor that offered enough time for the mass to reach such a huge size.

Thanks to the frequent use of MRI, a relatively new hypothesis classified Adenomyosis into intrinsic and extrinsic types. The intrinsic (intra-myometrial) type which develops from invagination of the basal endometrium ⁽¹³⁾, while the other extrinsic (extra-uterine) type develops from invasion of pelvic endometriotic lesions into the uterus causing disruption of the uterine serosa, this type mostly occurs in the posterior uterine wall and frequently associated with pelvic endometriosis. ⁽¹⁴⁾

It is noteworthy to know that the risk of endometrial carcinoma in patients having atypical polypoidal adenomyomas is almost 8.8%, which is much higher than the overall risk of cancers in endometrial polyps (0.8%).⁽¹⁵⁾ These lesions should not be considered benign except after careful histo-pathological examination and any residual or recurrent polypoidal adenomyoma should be considered a risk for developing cancer, and be managed accordingly.

Conclusion

Polypoidal subserous adenomyoma is a whole different type of Adenomyosis that needs more efforts to understand its pathogenesis. Although it is a less common finding, it occurs sometimes and should be included in the differential diagnosis of pelvi-abdominal masses in women. Gynecologists should be aware that these lesions may reach such big sizes, and they carry a considerable risk for malignant change.

References

(1) Bergeron C, Amant F, Ferenczy A. Pathology and physiopathology of adenomyosis. Best Pract Res Clin Obstet Gynaecol 2006;20:511–21.
(2) Benagiano G, Brosens I. History of adenomyosis. Best Pract Res Clin Obstet

Gynaecol 2006;20:449–63.
(3) Paul, P.G. et al. Extrauterine adenomyoma: a review of the literature. Eur J

Obstet Gynecol Reprod Biol. 2018;22:130-6

(4) Sakai Y., Matsukuma S. Large cystic uterine adenomyoma showing marked

epithelial metaplasiaand exophytic polypoid growth. Arch Gynecol Obstet.

2003;269:74–6 DOI 10.1007/s00404-003-0538-1
(5) Mazur MT. Atypical polypoid adenomyomas of the endometrium. Am. J. Surg.

Pathol. 1981; 5:473–82.
(6) Slezak P, Tillinger KG. The incidence and clinical importance of hysterographic evidence of cavities in the uterine wall. Radiology, 1976:118:581–6

evidence of cavities in the uterine wall. Radiology. 1976;118:581–6 (7) Chung YP, Lin HH, Sheu BC, Mao TL, Chang DY, Huang SC. Adenomyosis in the broad ligament and tamoxifen: report of a case. J Obstet Gynecol Res. 1997;23:69–73

(8) Dobashi Y, Fiedler PN, Carcangiu ML. Polypoid cystic adenomyosis of the uterus: report of a case. Int I Gynecol Pathol. 1992:11:240–3

uterus: report of a case. Int J Gynecol Pathol. 1992;11:240–3
(9) Gershon CE, Omer AZ, Mariam S, Hartmut EB. Giant adenomyotic cyst of the

uterus. Br J Obstet Gynecol. 1993;100:596–8 (10) Iribarne C, Plaza J, De la Fuente P, Garrido C, Garzon A,Olaizola JI.

Intramyometrial cystic adenomyosis. J Clin Ultrasound. 1994;22:348–50 (11) Keating S, Quenville NF, Korn GW, Clement PB. Ruptured adenomyotic cyst of

the uterus—a case report. Arch Gynecol. 1986;237:169–73 (12) Silvestro C., Francesca M., Giada F., Elisa R., Luigi F. Multiple extrauterine adenomyomas and uterus-like masses: case reports and review of the literature. Fert Stert. 2009;91(5)1956.e9-1956.e11 doi:10.1016/j.fertnstert.2009.01.076 (13) Garcia L, Isaacson K. Adenomyosis: review of the literature. J Minim Invasive

Gynecol. 2011;18:428–37. (14) Kishi Y, Suginami H, Kuramori R, Yabuta M, Suginami R, Taniguchi F. Four subtypes of adenomyosis assessed by magnetic resonance imaging and their specification. Am J Obstet Gynecol. 2012;207:114.e1–114.e7.

(15) Savelli L, De Iaco P, Santini D et al. Histopathologic features and risk factors for benignity, hyperplasia, and cancer in endometrial polyps. Am. J. Obstet.

- Gynecol. 2003;188:927–31.

 1. Journal Article, Name of Journal
- Journal Article, Name of Journal
- 3. Journal Article, Name of Journal