

# Extrauterine adenomyomas managed by laparoscopic excision: Three case reports with different theories of origin

Charles B Nagy, Szabolcs Papp, Nesreen Alaa Eldin, Samar M El-Maadawy

## ABSTRACT

**Introduction:** We present three case reports of extrauterine adenomyoma (recto-vaginal/retro-cervical, broad ligament, abdominal). The common presenting symptoms in our patients were pelvic pain, dysmenorrhea, and deep dyspareunia. The cases were successfully treated with laparoscopic excision by a multidisciplinary team of doctors. One patient showed adenomyoma co-existing with endometriosis on histopathological examination of the tissue sample.

**Case Series:** We present 3 cases of extra uterine adenomyomas in 3 different sites, each case representing a different theory of origin and all cases managed laparoscopically with successful outcome without any complications. First case represent the implantation theory following antecedent myomectomy. Second case represents origin of adenomyoma as direct extension from the uterus with background of severe diffuse adenomyosis. Third case represents origin from Müllerian remnants in the recto-vaginal septum with no adenomyosis or obliteration of the pouch Douglas.

**Conclusion:** We propose the theory that adenomyoma which is a form of adenomyosis should be regarded as a form of deep endometriosis involving the uterus rather than a separate entity. We believe that multidisciplinary laparoscopic treatment is the way forward for accurate diagnosis and treatment of adenomyosis in patients requiring to preserve fertility. Future research needs to focus on studying endometriosis behavior and recurrence according to the tissue host to understand the disease and tailor the management according to patient symptoms.

**Keywords:** Endometriosis, Extrauterine adenomyoma, Infertility, Laparoscopy

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## INTRODUCTION

Adenomyoma of the uterus is a circumscribed nodular aggregate of benign endometrial glands surrounded by endometrial stroma with leiomyomatous smooth muscle bordering the endometrial stromal component [1]. Adenomyoma is a localized and focal form of adenomyosis. It is classified as focal, diffuse, and cystic [2]. The prevalence of adenomyosis fluctuates between 5% and 70% [3, 4]. Diagnosis of adenomyosis is made on histological examination of the uterus. Extrauterine

adenomyomas are extremely rare and located outside the uterus [2]. Clinical symptoms of adenomyosis are menorrhagia, pelvic pain, and dysmenorrhea; and risk factors associated are spontaneous and induced abortions, multiparity, endometrial hyperplasia, endometriosis, smoking, and surgical trauma [5, 6]. Current treatment options for symptomatic adenomyosis include hysterectomy, medication, conservative surgery, or minimally invasive techniques including uterine artery embolization [7, 8]. We present three case reports of extrauterine adenomyoma (recto-vaginal/retro-cervical space, broad ligament, and large abdominal) managed by laparoscopic excision.

## CASE SERIES

### Case 1: Combined laparoscopic-vaginal approach for the excision of large extrauterine (recto-vaginal, retro-cervical) adenomyoma

A 33-year-old female patient presented with a long-standing complaint of severe pelvic pain, dysmenorrhea, and deep dyspareunia with irregular heavy periods and lately persistent vaginal bleeding, severe pain in bowel opening, and a sense of incomplete bowel emptying. She had primary infertility for three years. Her blood investigations showed iron deficiency anemia with normal CA-125 levels.

Clinical examination revealed a large mass occupying the space behind the cervix and upper half of the vagina pressing on the rectum, with evidence of bleeding coming from the mass. A colonoscopy showed normal findings of the bowel wall. A pelvic ultrasound scan (Figure 1) confirmed the presence of a large mass behind the cervix attached to the rectum and extending behind the upper part of the vagina, a picture highly suggestive of a deep infiltrating endometriosis nodule. A pelvic MRI of the patient confirmed the above findings and suspicion of malignancy could not be ruled out.

The multidisciplinary team decided to proceed with surgical excision of the mass using the laparoscopic route with the possibility of bowel resection and ureteric stents.

The patient was treated with Decapeptyl (triptorelin) 3.75 mg GnRh agonist injections for two months prior to surgery to stop menstruation and irregular bleeding. The surgery was conducted jointly by the consultant gynecologist and colorectal surgeon using a novel operative technique of combined vaginal and laparoscopic approach to excise the mass from behind the cervix and upper vagina without the need for bowel resection to avoid the serious effects on the quality of life and fertility. The vagina was reconstructed to cover the large defect after removing the mass (Figure 2A and B). Interestingly, the rectosigmoid bowel wall was not infiltrated in this case so bowel resection was not indicated. Laparoscopy showed



Figure 1: Pelvic ultrasound (Case 1).

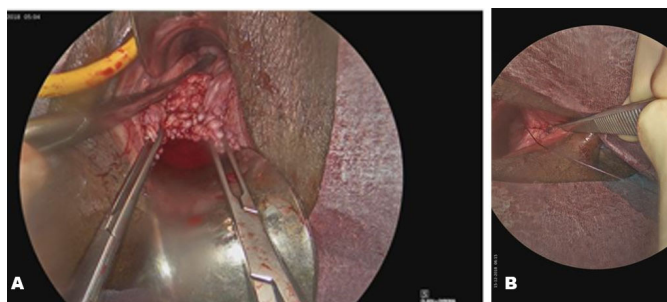


Figure 2 (A and B): Surgical procedure (Case 1).



Figure 3: Actual specimen (Case 1).

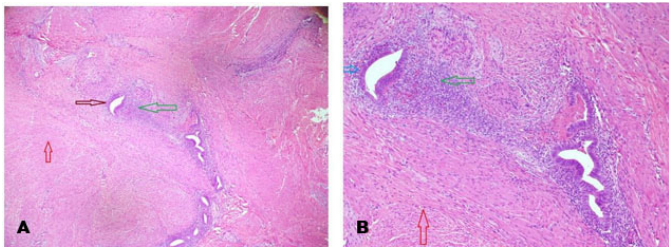


Figure 4 (A and B): Histology slide (Case 1).

normal fallopian tubes suggesting that the patient would be able to get pregnant spontaneously without the need for assisted production techniques.

Histological examination of excision sample (Figure 3) reported recto-vaginal adenomyotic nodule and fragmented tissue with features of endometriosis with no evidence of malignancy (Figure 4A and B).

The patient became pregnant spontaneously one year after the surgery and delivered a healthy baby at 39 weeks by cesarean section.

### Case 2: Rare case of broad ligament extrauterine adenomyoma

A 31-year-old, nulliparous woman [body mass index (BMI) of 33.3] with primary infertility for six years was referred with the finding of a large complex and highly vascular right adnexal mass. She presented with complaints of pelvic pain, dysmenorrhea, and irregular menstruation for the past few months. The patient had a history of laparoscopic myomectomy in 2016 and three failed in vitro fertilization (IVF). Her blood investigation reported an anti-Müllerian hormone level of 7.059 ng/mL.

Pelvic ultrasound (Figure 5A–C) showed a bulky uterus with features of adenomyosis showing a focal adenomyoma at the left anterior uterine wall. Complex

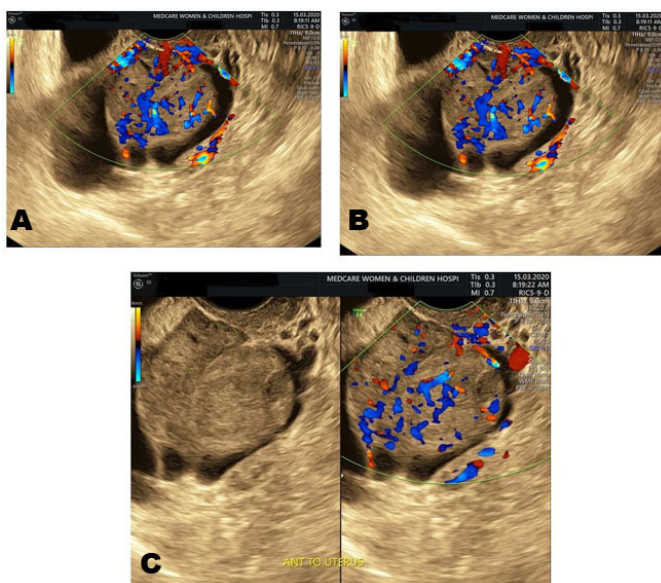


Figure 5 (A–C): Pelvic ultrasound (Case 2).

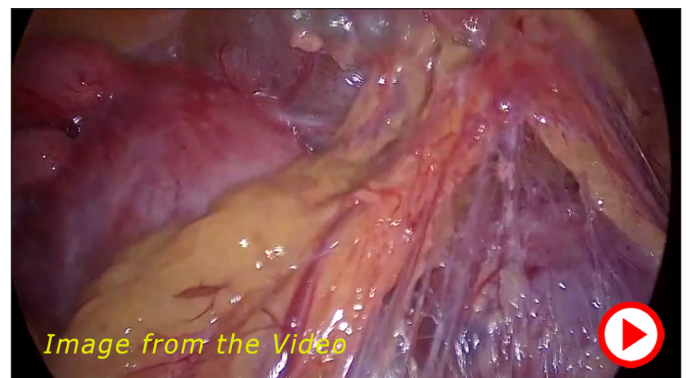
mixed solid and cystic vascular mass lesion at the right adnexal region which appeared to be separable from the right ovary. A right broad ligament adenomyoma along with a small complex right ovarian cyst mostly hemorrhagic in nature was suggested. No signs of deep infiltrating endometriosis were seen.

Laparoscopic extensive adhesiolysis for attached bowel and omentum, followed by selective uterine artery ligation on the right side due to the high vascularity of the tumor was done. The extrauterine adenomyoma was separated from the ovary and the uterus. It was gently dissected off the bladder wall. Because of the superficial infiltration of the bladder wall, the bladder was opened to identify the extent of involvement before the tumor was completely separated and excised. Both ovaries and the rectosigmoid bowel were perfectly normal (Video 1: Surgery). The patient was discharged on 2nd postoperative day with no complications.

The histopathology of the right adnexal mass reported features of adenomyoma with cystic changes, possibly arising from broad ligament and no evidence of malignancy. Omental biopsy histopathology showed mild features of panniculitis.

The patient was treated with gonadotropin-releasing hormone (GnRH) analogue injections postoperatively for three months. The patient had successful IVF and became pregnant six months after the surgery. At 38 weeks of gestation, a healthy baby was delivered by cesarean section.

The patient developed a port site recurrence of adenomyoma about one year after the laparoscopic excision.



**Video 1:** Laparoscopic approach of a case of extrauterine adenomyoma in the right side of the broad ligament attached to and infiltrating the bladder.

**Video 1 URL:** <https://www.ijcasereportsandimages.com/archive/article-full-text/101409Z01CN2023#video1>



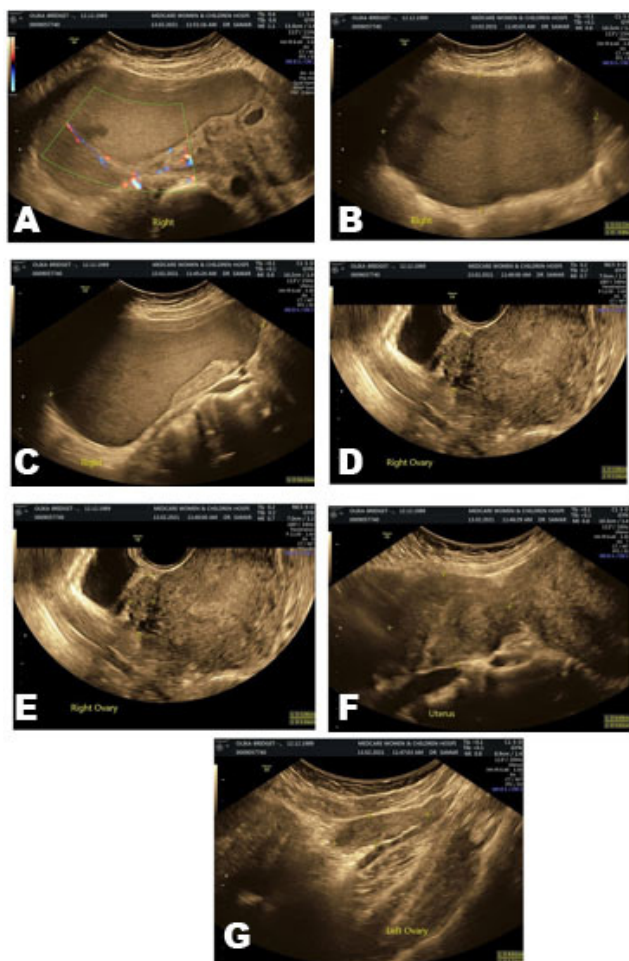


Figure 6 (A–G): Abdomen-pelvic ultrasound (Case 3).

### Case 3: Large extrauterine adenomyoma mistaken as an ovarian endometrioma

A 31-year-old, nulliparous female (weight=74 kg, height=169 cm) was referred with a diagnosis of severe endometriosis and large ovarian endometrioma for a laparoscopic ovarian cystectomy. She had primary infertility for four years. The patient presented with severe dysmenorrhea (10/10), pelvic and abdomen pain, deep dyspareunia, marked abdominal distention, dyspepsia and chronic constipation. She had persistent vaginal bleeding for one month preceded by a history of heavy irregular menstruation for years.

Pelvic and abdominal ultrasound scan (Figure 6A–G) showed bulky fibroid uterus with features of diffuse adenomyosis and multiple fibroids, with a large right adnexal complex cystic lesion which appeared to be separable from both ovaries with the possibility of para-tubal complex cystic mass of unknown etiology or huge extrauterine adenomyoma. No evidence of deep infiltrating endometriosis of the pelvis was seen on the transvaginal ultrasound scan. Magnetic resonance imaging (MRI) of the pelvis showed similar findings.

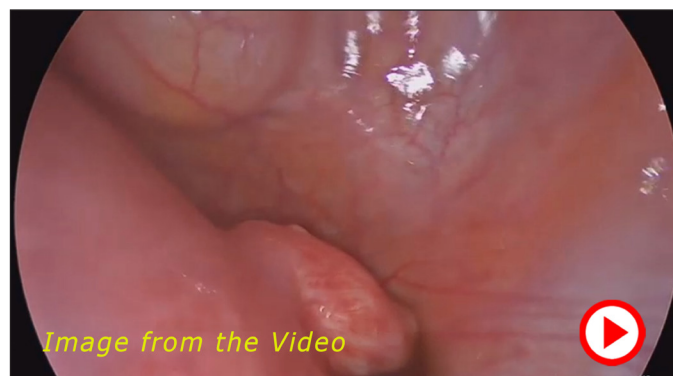
No past history of any surgeries or trauma. Blood investigations showed severe iron deficiency anemia (Hb 8 g/dL).

A multidisciplinary team consisting of a colorectal surgeon, vascular surgeon, urologist, and radiology team decided to do laparoscopic exploration and excision of the adnexal mass.

Laparoscopic exploration showed a huge extrauterine adenomyoma with a pedicle attached to the main uterus collectively measured over 20 cm × 20 cm in diameter rising above the level of the umbilicus with a cystic component full of old blood and the solid component attached to the uterus. After the evacuation of the cystic component, the mass was removed completely with preservation of the right ovarian infundibulopelvic ligament right ovary and right tube (Video 2: Surgery). The left ovary and tube looked healthy. Hysteroscopy and endometrial biopsy were done at the same time.

Histological examination confirmed extra trying adenomyoma with solid component showing smooth muscle bundles separated by well-vascularized connective tissue and cystic component also showing the same appearance and there was no evidence of malignancy or atypia. Endometrial biopsy showed hyperplastic glands with no evidence of cytological atypia or malignancy.

Postoperatively the patient was treated with GnRH analogue monthly injections for three months. The patient was not planning for pregnancy.



**Video 2:** Laparoscopic approach of a case of huge extrauterine adenomyoma arising from and connected to the uterus with large amount of old blood collected inside, resembling large endometrioma on the right side.

**Video 2 URL:** <https://www.ijcasereportsandimages.com/archive/article-full-text/101409Z01CN2023#video2>



## DISCUSSION

Adenomyosis coexists with other benign disorders, such as endometriosis (70%), leiomyomas (50%), endometrial hyperplasia (35%), and endometrial polyps (2%) [9, 10]. Cases 1 and 3 had coexisting endometriosis and Case 2 had undergone myomectomy.

All three patients had infertility. Recent studies show that adenomyosis negatively affects in vitro fertilization, pregnancy, and the live birth rate, as well as increases the risk of miscarriage and risk of obstetric complications [4].

Pistofidis et al. have described adenomyosis classification based on intraoperative and histopathology findings as diffuse, sclerotic, nodular, and cystic. It reported that in the study all cases of cystic and nodular adenomyosis were treated by laparoscopic excision of the lesion. 89% of patients with sclerotic adenomyosis were treated with wide laparoscopic excision of the abnormal tissue and 81% of patients with diffuse adenomyosis were treated with laparoscopic hysterectomy [11].

In our cases, Case 1 had large nodular recto-cervical, recto-vaginal extrauterine adenomyoma, which was excised by a multidisciplinary team with a novel operative technique of combined vaginal and laparoscopic approach. One-year post-surgery the patient had a spontaneous conception and delivered a healthy full-term fetus.

In Case 2, the patient had a solid-cystic broad ligament adenomyoma managed surgically with laparoscopic excision. Selective uterine artery ligation was done before excision of the tumor due to high vascularity. Post-surgery, the patient was treated with GnRH analogue injections. Six months after the procedure patient had a successful IVF and became pregnant.

Case 3 had a large cystic-solid adenomyoma mistaken as an ovarian endometrioma. Despite the enormous size (20 cm × 20 cm) of adenomyoma, she was successfully treated with laparoscopic excision preserving the uterus.

There are many theories about the pathogenesis of adenomyosis. One theory suggests that metaplastic changes of intra-myometrial embryonic pluripotent Müllerian remnants in the adult uterine wall can possibly lead to the establishment of de novo ectopic endometrial tissue within the myometrial wall, creating adenomyotic lesions [12, 13]. This theory can be applied to Case 3 which presented an example of extrauterine adenomyoma that is probably arising from metaplastic head and Müllerian remnants as there was no antecedent surgery and its logic to be explained by the retrograde menstruation surgery.

Case 2 developed recurrence of adenomyoma one year after the laparoscopic excision. A study by Zhu et al. concluded that the postoperative drug (GnRH agonist with oral contraceptives) use may be beneficial to reduce the recurrence of adenomyosis, especially for adenomyosis with endometriosis [14].

Szubert et al. in the review of adenomyosis as a risk factor for myometrial or endometrial neoplasms

concluded that adenomyosis may be a potential risk factor for myometrial or endometrial neoplasms [15].

We strongly recommend that adenomyosis nodules or hysterectomy specimens of the adenomyotic uterus should be removed using in-bag morcellation to avoid significant morbidity due to risk of dissemination and recurrence as reported by Belmarez et al. [16] and also encountered in our Case 2.

In Case 1, the finding of large isolated recto-vaginal adenomyoma without infiltrating the rectum highlights the utmost importance of accurate preoperative radiological evaluation and clinical examination in planning the suitable surgical procedure for the individual patient. As in our new pro-forma for ultrasound mapping, we highlighted 3 separate types of recto-vaginal deep endometriosis, our case was a clear example of isolated recto-vaginal septum without rectal wall involvement. El-Maadawy et al. have also highlighted the importance of ultrasound mapping to tailor an appropriate surgical approach to enhance the patient quality of life and fertility, ensuring radical excision of the disease and minimizing operative and postoperative complications in deep infiltrating endometriosis [17].

Donnez et al. suggested that uterocervical adenomyosis could be the cause of deep endometriotic nodules, as is also the case for deep anterior endometriosis, called bladder adenomyotic nodules [18].

We propose the theory that adenomyosis should be regarded as a form of deep endometriosis involving the uterus rather than a separate entity, due to the very close histopathological similarity.

Saunders et al. reported that we may make more progress in developing patient-focused treatments if we stop considering endometriosis as a single “disease” with a diagnosis based solely on the presence of a lesion(s) resembling endometrium. The disease model is problematic, not only because of the poor correlation between numbers/location of lesions and pain symptoms, but also because it is estimated that up to 50% of asymptomatic fertile women presenting for other surgical procedures may have lesions [19].

We believe that the risk of recurrence of deep endometriosis depends not only on the nature of the disease but more importantly on the host tissue whether it is myometrium, bowel, ovarian, peritoneal, or extra-pelvic. This concept will significantly facilitate future research and the reproducibility and accuracy of results as we compare type-specific deep endometriosis, i.e., bowel, uterine, ovarian, and so on, rather than comparing stages that include more than one type of endometriosis in the same stage.

In the presented case reports multidisciplinary team consisting of an experienced gynecologist, colorectal surgeon, urologist, and radiology specialist decided on the treatment plan resulting in a short hospital stay, uneventful recovery, and preservation of the uterus.

## CONCLUSION

We propose the theory that adenomyoma, which is a form of adenomyosis should be regarded as a form of deep endometriosis involving the uterus rather than a separate entity. Future research needs to focus on studying endometriosis behavior and recurrence according to the tissue host rather than the staging systems, as we believe that accurate endometriosis typing and comparing tissue-specific characteristics of endometriosis is the way forward to understand the disease and tailor the management according to patient symptoms.

We believe that multidisciplinary laparoscopic treatment involving an experienced gynecologist, colorectal surgeon, urologist, and radiology specialist is the way forward for accurate diagnosis and treatment of adenomyosis in patients wanting to preserve fertility.

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## Author Contributions

Charles B Nagy – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Szabolcs Papp – Acquisition of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Nesreen Alaa Eldin – Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy

or integrity of any part of the work are appropriately investigated and resolved

Samar M El-Maadawy – Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

**Guarantor of Submission**

The corresponding author is the guarantor of submission.

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**Conflict of Interest**

Authors declare no conflict of interest.

**Data Availability**

All relevant data are within the paper and its Supporting Information files.

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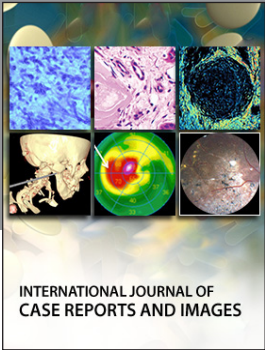
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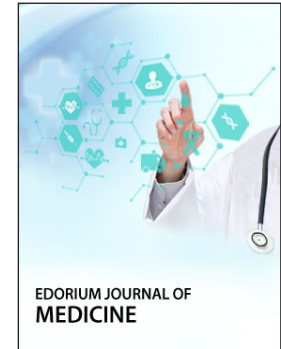
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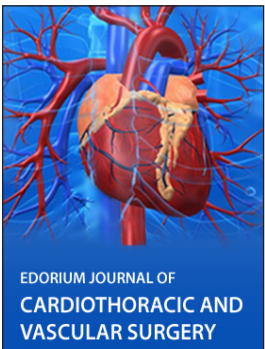
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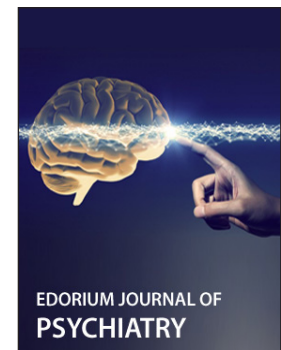
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