International Journal of Women's Health

Dovepress

open access to scientific and medical research

Open Access Full Text Article

CASE REPORT

Cystic Adenomyoma in Pregnancy: A Case Report

Stephanie Verta, Christine E Brambs, Corina Christmann

Department of Obstetrics and Gynecology, Lucerne Cantonal Hospital, Lucerne, Switzerland

Correspondence: Stephanie Verta, Department of Obstetrics and Gynecology, Lucerne Cantonal Hospital, Lucerne, Switzerland, Tel +41 41 205 28 16, Email stephanie.verta@luks.ch

Abstract: Cystic adenomyomas (CA) are rare. They primarily affect adolescents and young women in their fertile years. Therefore, fertility and pregnancy outcome are of pivotal relevance in this patient collective. Apart from the guidelines of the European Society of Human Reproduction and Embryology (ESHRE) on the management of endometriosis in general, there are no specific treatment recommendations for CA and, as far as our research shows, no data illustrating the behavior of a CA over the course of pregnancy. Thus, we report the case of a 32-year-old 1-gravida, 1-para, preconceptionally diagnosed with a CA by ultrasound. After thoroughly discussing further treatment options, the decision was made to opt for a more conservative approach and not perform surgery before attempting a next pregnancy. The patient conceived spontaneously and sonographic monitoring of the CA throughout pregnancy showed complete regression of the cystic component during the second trimester. A healthy baby was delivered at term by an uncomplicated elective cesarean section. Following a review of the literature and taking into account the course of our case, we propose the feasibility of a conservative, non-surgical approach in women with a CA and the desire to conceive.

Keywords: cystic adenomyoma, endometriosis, conservative management, pregnancy, fertility

Introduction

Uterine adenomyosis is most commonly viewed as a manifestation of endometriosis. It is defined as the presence of ectopic endometrial glands and stromal cells within the myometrium, sometimes accompanied by a hypertrophy of the surrounding myometrium causing a varying enlargement of the uterus.[1–3] According to the archimetrosis hypothesis by Gerhard Leyendecker, hyper- and dysperistalsis, causing tissue lesions, allow for the invagination and infiltration of cells from the basalis of the endometrium into the myometrium and as a result the development of adenomyotic lesions.[4,5]

Depending on its distribution pattern within the myometrium, adenomyosis is subdivided into a diffuse form, which is more common, and a localized or focal form.[2,6,7] The latter comprises, among other things, rare lesions such as solid and cystic adenomyomas (CA).[6,7] While small cystic lesions may also typically occur in diffuse adenomyosis, the diameter of the cyst in these cases is mostly <5mm.[8–11] Larger cystic adenomyotic lesions, on the other hand, are very uncommon.[9,12] First mentioned in 1908, Cullen described the entity of subperitoneal large adenomyomata nodules with irregular cavities filled with chocolate-colored contents.[13] From a histopathologic point of view, these adenomyotic cysts are lined with endometrium (epithelium plus stroma), filled with a viscous, hemorrhagic, chocolate sauce-like content and surrounded by hypertrophic myometrium.[6,7,9–11]

CA of the uterus primarily affects adolescents and young women under the age of 30 years, which is why the term "juvenile cystic adenomyoma" is frequently applied in those cases.[6,9–11,14] However, they can also occur in women > 30 years of age or even after menopause.[6] The leading symptom characteristic of CA, especially in young women, appears to be early onset of severe, often medication-resistant dysmenorrhea, most likely caused by cyclical shedding of the endometrial lining and consecutive bleeding into the cyst.[6,9–11,14] Aside from that, chronic pelvic pain, cyclical lower back pain, dyspareunia, bleeding disorders, and infertility have also been reported.[9,11,15,16] The diagnostic criteria for juvenile CA put forth by Takeuchi et al is the presence of a solitary myometrial cyst ≥ 1 cm embedded in perifocal hypertrophic myometrium, lacking connection to the uterine cavity, in women ≤ 30 years of age suffering from severe dysmenorrhea.[11]

Received: 20 November 2023
Accepted: 29 February 2024
Published: 7 March 2024

Given the fact that the patients affected are close to or even of child-bearing age, fertility, pregnancy, and especially pregnancy outcome are important issues to consider in treatment planning and counseling of these patients. As CAs occur quite rarely, so far no specific recommendations or guidelines exist on therapeutic management, aside from the guidelines of the European Society of Human Reproduction and Embryology (ESHRE) on the management of endometriosis in general.[17] Pregnancies with favorable outcomes following surgery for CA have been reported in literature,[11,16,18] however, insights on how to manage a CA in the immediate context of pregnancy planning hardly exist. To the best of our knowledge, there is no report to present of a conscious decision not to strive for a surgical approach preconceptionally but for a conservative management of the CA in women wishing to conceive.

Thus, we present the case of a 32-year-old Caucasian woman, 1-gravida, 1-para, with a known CA, who conceived spontaneously after consciously deciding against preconceptional surgical removal of the CA and report the clinical aspects as well as the sonographic findings of the CA throughout the course of the pregnancy. The aim of this case report is to show that a conservative, non-surgical management of CAs, in women who desire to conceive, is feasible and to encourage physicians to offer their patients this option, as the surgical approach may entail possible risks for future pregnancies.

Case Presentation

In February 2022, a then 32-year-old 1-gravida, 1-para of Caucasian origin was referred to our endometriosis center for surgical treatment of suspected endometriosis due to endometriosis-associated pain. The suspicion of endometriosis had been based on the alleged finding of an endometrioma in the left ovary. The patient had been experiencing increased dysmenorrhea since February 2021, following the first pregnancy. She did not suffer from any deep dyspareunia, dysuria or dyschezia, hematuria, or hematochezia. Personal history included a spontaneous delivery in July 2020, complicated by postpartum placental retention with the need for manual removal of the placenta and curettage. She had suffered a substantial blood loss of 3500mL and in the end experienced the delivery as traumatizing. Her family history revealed one maternal grandmother with cervical cancer.

During her first consultation in March 2022 detailed transvaginal ultrasound revealed a cystic lesion confined to the uterus, lying within the myometrium of the left uterine wall, with a content of ground glass echogenicity and hyperechoic, possibly calcified white spots lining the inside of the cyst wall (Figure 1A). On color Doppler imaging, no blood flow could be detected within the lesion, underlining its cystic nature (Figure 1B). The cystic lesion measured 33.8×28.7mm and did not display any direct contact with the uterine cavity (Figure 1C). The uterus itself measured 88x48mm in total. Focally, the myometrium surrounding the cystic lesion, facing the surface of the uterus on the left, appeared to be very thin (Figure 1D). Neither ovary showed signs of endometriotic lesions and both were nicely mobile, especially the left ovary seemed to have no contact with the cystic lesion in the uterine wall (Figure 1E). The urinary bladder and rectal wall, the uterosacral ligaments, the pelvic side wall and parametria, as well as the rectovaginal septum were normal and showed no signs of deep endometriosis. Both ureters were functioning normally with no evidence of obstruction. Based on the sonographic findings and the symptoms of the patient, the diagnosis of endometriosis with a cystic adenomyoma was made. Aside from that, sonography also revealed a normally sited, vital pregnancy in the sixth week of gestation (Figure 1F). The patient had discovered, that she was pregnant between the date of referral and her first consultation. Naturally, in light of this turn of events, endometriosis surgery was no longer an issue for discussion.

Unfortunately, the pregnancy ended in a miscarriage in the ninth week of gestation with no need for further treatment. In May 2022, as the patient was no longer pregnant, discussion on further procedures concerning the CA and endometriosis-related symptoms was resumed. The key question now was: should surgery on the CA be done before attempting a next pregnancy? At that point, the patient's most urgent concern was to become pregnant again and to have a second child. Pain and other endometriosis-related symptoms were secondary to her. A literature search in the PubMed® database, with no restrictions concerning the year of publication, for the terms "cystic AND adenomyoma AND pregnancy" yielded a total of 8 citations, however none of them reported on the behavior of CA during pregnancy, on which counseling on how to proceed with the CA before the next pregnancy could have been based. Therefore, in an extensive discussion with the patient and her partner, we weighed the risks of preconceptional surgery on the uterus (risk of uterine rupture, intramural ectopic pregnancy, abnormal placentation with the risk of postpartum hemorrhage) against

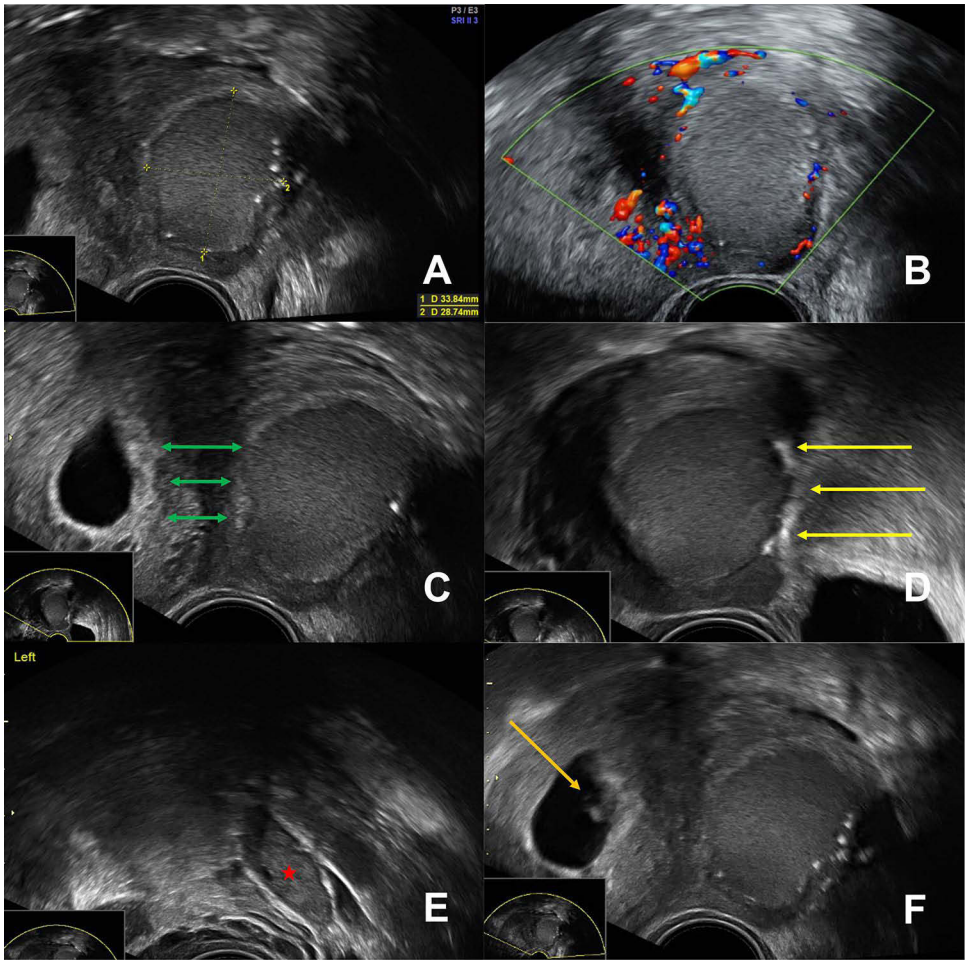

Figure 1 Ultrasound findings of the first consultation in March 2022. (**A**) Cystic adenomyoma within the left uterine wall, filled with a content of ground-glass echogenicity. Hyperechoic, possibly calcified spots line the inside of the cyst wall. The measurement lines show a maximum diameter of 33.8 mm. (**B**) On color Doppler imaging no blood flow can be detected within the cystic adenomyoma, underlining its cystic nature. (**C**) The cystic adenomyoma is located in the left uterine wall and has no contact with the uterine cavity (green double arrows demonstrating the distance between the uterine cavity and the cystic adenomyoma). (**D**) The myometrium in the area of the lateral border of the cystic adenomyoma appears to be very thin (yellow arrows). (**E**) Image of the left ovary (red star) demonstrating that the cyst is not ovarian in origin in the sense of an endometrioma but located in the uterine wall. (**F**) Normally sited, intrauterine pregnancy to the patient's right (Orange arrow). To the patient's left, is the cystic adenomyoma.

the imaginable risks of pregnancy in the presence of a CA (pain-related problems, rupture of the CA, and spontaneous hematoperiteum). Additionally, as the patient was rather oligosymptomatic regarding the CA and endometriosis at the time, surgery for CA would have constituted a preventive measure for problems we did not know would even occur. Together with the patient and her partner, we decided to opt for a conservative approach and to not perform surgery before attempting a next pregnancy.

Fortunately, in September 2022, the patient conceived again spontaneously and a transvaginal ultrasound performed in October 2022 showed a normally sited, vital singleton pregnancy in the sixth week of gestation (Figure 2). The CA displayed the same sonographic features as in previous scans, with no signs of growth. The patient reported on normal pregnancy-related discomfort; no unusual symptoms had appeared.

On a follow-up scan performed in January 2023, in the second trimester at 18 weeks and 4 days of gestation, the CA had almost completely vanished on transvaginal ultrasound. The only clue revealing its existence and location was the presence of the hyperechoic white spots, now compressed to an area of approximately 20x2mm in the left anterior uterine wall, just beneath the uterine surface, close to the vesicouterine fold. The ground glass echogenic content had completely disappeared and, consequently, the space-occupying effect of the CA (Figure 3A-D). Transabdominal ultrasound showed a vital, timely-developed fetus. The placenta displayed normal morphology and was located on the anterior uterine wall.

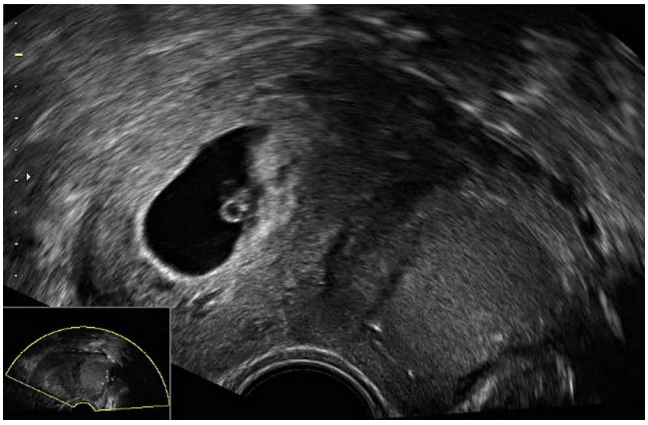

Figure 2 Transvaginal ultrasound in October 2022 shows the normally sited pregnancy within the uterine cavity to the patient's right and, clearly separated from it, the cystic adenomyoma to the patient's left. Its sonographic appearance is unchanged compared to the previous scans.

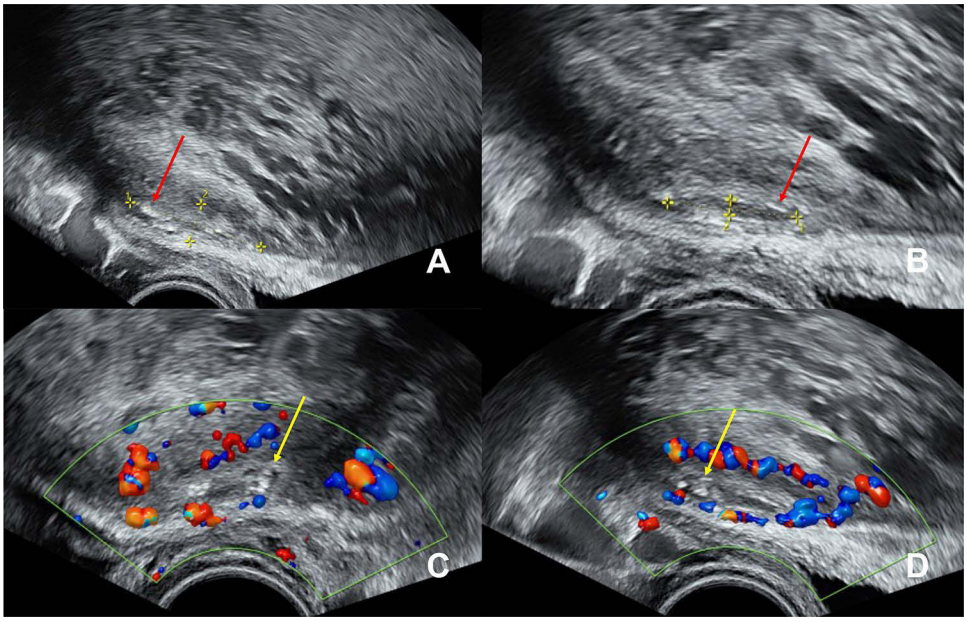

Figure 3 Transvaginal ultrasound in January 2023 during the second trimester of pregnancy, depicting the area of the anterior uterine wall to the left. In **(A)** and **(B)** the cystic adenomyoma is marked by the yellow measurement lines. It is practically only discernible by the marginally located hyperechoic spots (red arrows). The ground-glass content has dissolved. **(C)** and **(D)** show the peripheral blood vessels surrounding the cystic adenomyoma by color Doppler imaging. The hyperechoic spots indicate the former inner cyst wall (yellow arrows).

The CA could not be visualized by means of transabdominal scan. According to the patient, the pregnancy had gone well to date, although she did mention that when walking fast or lifting heavier objects she would experience a pulling sensation in the left lower abdomen, which would pass spontaneously after 30 min of rest. She had not required analgesics for this type of pain and did not feel impaired by it.

Ultrasound performed in the third trimester of pregnancy at 31 weeks and 5 days of gestation was not able to visualize the CA, neither with transabdominal nor with transvaginal scan (Figure 4). Fetal growth, however, was timely and cervical scan displayed sufficient length (40mm). The patient had been experiencing some pain in the area of the symphysis pubis, but the pulling sensation in the left lower abdomen had ceased.

In Mai 2023, at 38 weeks and 5 days of pregnancy, the patient underwent a primary cesarean section due to a traumatic birth experience in the first pregnancy. A healthy baby boy was born with a birth weight of 3730g. Intraoperative inspection of the anterior uterine wall showed no clear signs of the CA and the operation could be carried

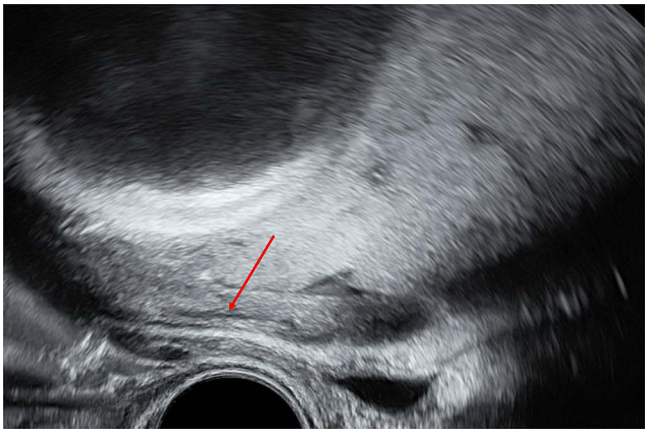

Figure 4 Transvaginal ultrasound in April 2023 during the third trimester of pregnancy, depicting the area of the anterior uterine wall to the left. The bladder, which is closest to the ultrasound probe, is almost empty. Cranial to the bladder is the part of the anterior uterine wall where the cystic adenomyoma was located on the previous scans (red arrow), however, at this point during pregnancy it is no longer visible. The caudal edge of the placenta and the fetal head are also visible.

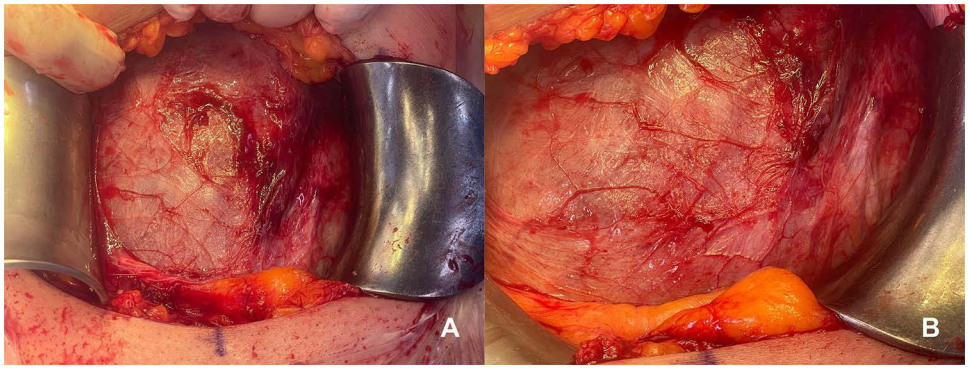

Figure 5 Intraoperative findings in May 2023. **(A)** and **(B)** show the intraoperative view of the anterior uterine wall at the level of hysterotomy during cesarean section. The cystic adenomyoma is not visible.

out without further difficulties or complications such as adhesions or hemorrhage (Figure 5A and B). The post-operative course was uncomplicated. The patient was discharged from the hospital on post-operative day 4.

On ultrasound follow-up, 4 months after the cesarean section, the CA presented itself as a relatively unremarkable, ill-demarcated, hypoechoic area (Figure 6A) measuring 10.8×6.7×13mm. The characteristic white spots were still visible within and in the periphery of the lesion (Figure 6B). Compared to preoperatively, the CA was now located in the midline of the uterus in the anterior wall, just cranially to the uterotomy. It seemed to extend from the uterine cavity to the ventral uterine surface (Figure 6C). No blood flow could be detected in the center of the lesion, while the surrounding myometrium displayed abundant vascularization (Figure 6D). Overall, the course of the postpartum period had been uneventful. At the time of her last consultation, the patient was asymptomatic, however still under the influence of lactational amenorrhea. The timeline for the entire episode of care outlined above is shown in a diagram (Figure 7).

Discussion

CA, being a rare condition, often poses differential-diagnostic challenges and the inconsistency in nomenclature across literature impairs data collection on symptoms, efficacy of therapeutic measures, as well as fertility outcomes.[6,16,19–21] Consequently, there are no clear treatment recommendations. Possible therapeutic approaches proposed by various case reports and case series throughout literature comprise a conservative, medical therapy by means of analgesics, continuous oral contraceptive pills, or GnRH agonists[10,12,15] as well as a surgical course of action including laparoscopy, laparotomy, and hysteroscopy.[9–11,14,16,18,21,22]

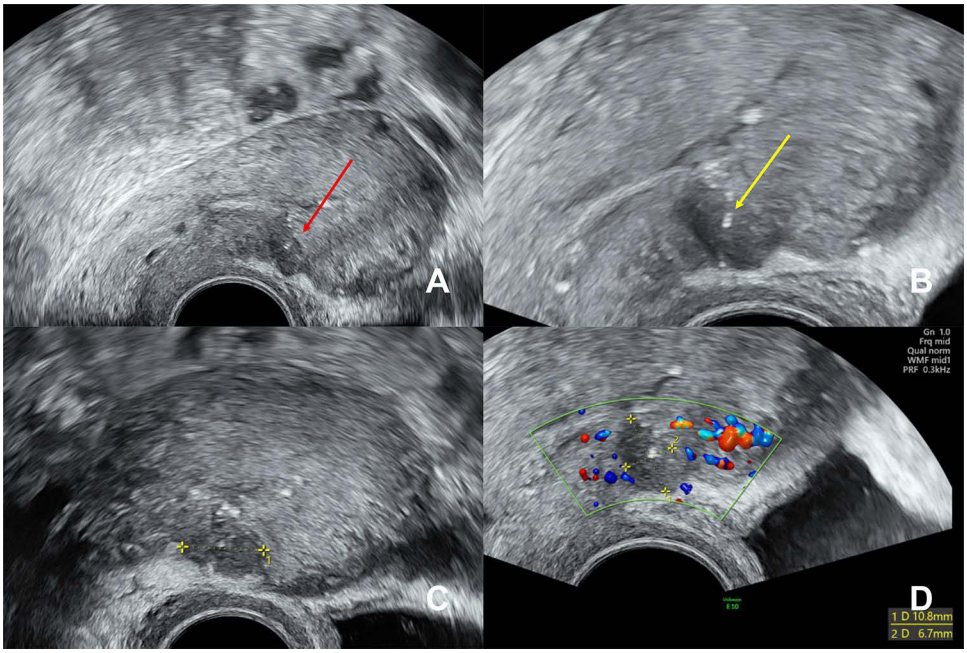

Figure 6 Transvaginal ultrasound in September 2023, four months postpartum. (**A**) Uterus depicted in the sagittal plane. The cystic adenomyoma appears as an unremarkable, ill-demarcated, hypoechoic area (red arrow) in the anterior uterine wall. (**B**) Located within the hypoechoic area are the characteristic white spots (yellow arrow). (**C**) Uterus depicted in the transverse plane. The cystic adenomyoma in the anterior uterine wall extends to the ventral surface of the uterus. (**D**) No vascularization can be detected within the hypoechoic area. The surrounding myometrium is well vascularized. The measurement lines indicate the dimensions of the cystic adenomyoma (10.8x6.7mm).

For analgesia, mostly non-steroidal anti-inflammatory drugs were prescribed, but they often proved ineffective at baseline or the pain became refractory to the analgesic due to worsening of the dysmenorrhea and pelvic pain.[9,11,12,21,22] Menstrual cycle suppression by the (continuous) intake of an oral contraceptive pill has been reported in several cases with either insufficient effect on the AC-related symptoms or recurrence of symptoms after discontinuation of the therapy.[11,14,21]

In the case presented by Branquinho et al, however, the patient experienced significant relief of the pelvic pain by taking a continuous oral contraceptive pill,[12] and Fisseha et al not only reported significant pain relief but also a significant reduction in the size of the lesion, from 2.1 cm to <1 cm, over the course of 2 years of follow-up.[15] Regarding the use of GnRH agonists, Takeda et al could demonstrate its effectiveness in the treatment of abdominal cramps for the duration of the application.[10] Thus, in some cases, treatment with a (continuous) oral contraceptive pill or a GnRH agonist can offer a therapeutic option and should be considered.

With respect to symptom control, the body of literature suggests that the most effective therapeutic approach, especially in the long run, is surgical resection resulting in predominantly significant improvement and often complete resolution of AC-related symptoms such as severe dysmenorrhea and pelvic pain.[9–11,14,16,18,21,22] The largest retrospective study, reporting surgical treatment in 18 cases of CA, was carried out by Kerbage et al. In their cohort almost all the patients suffered from pain (dysmenorrhea, pelvic pain or dyspareunia) prior to surgery, consequently for 12 patients the indication for the surgery was pain. For the remaining 6 patients, indication for surgery was infertility. In the majority of the cases (n=15) the surgical approach for CA treatment was laparoscopic excision, using a technique similar to laparoscopic myomectomy. In 3 cases, the surgery was performed hysteroscopically, either by complete bipolar resection or by opening and draining the AC and coagulating the inner lining. According to Kerbage et al, surgery led to a clear improvement of the symptoms in the majority of the patients. In 1 case surgery did not improve the pain at all and in 2 cases only partial improvement occurred postoperatively.[16] In the study conducted by Takeuchi et al, comprising a total of 9 patients with juvenile CA, all of the enrolled participants complained about dysmenorrhea preoperatively, rating the severity of their pain at 8–10 on the visual analogue scale (VAS). The surgical approach was laparoscopic enucleation of CA in all cases. A statistically significant improvement in pain was observed in all 9 patients postoperatively, with VAS

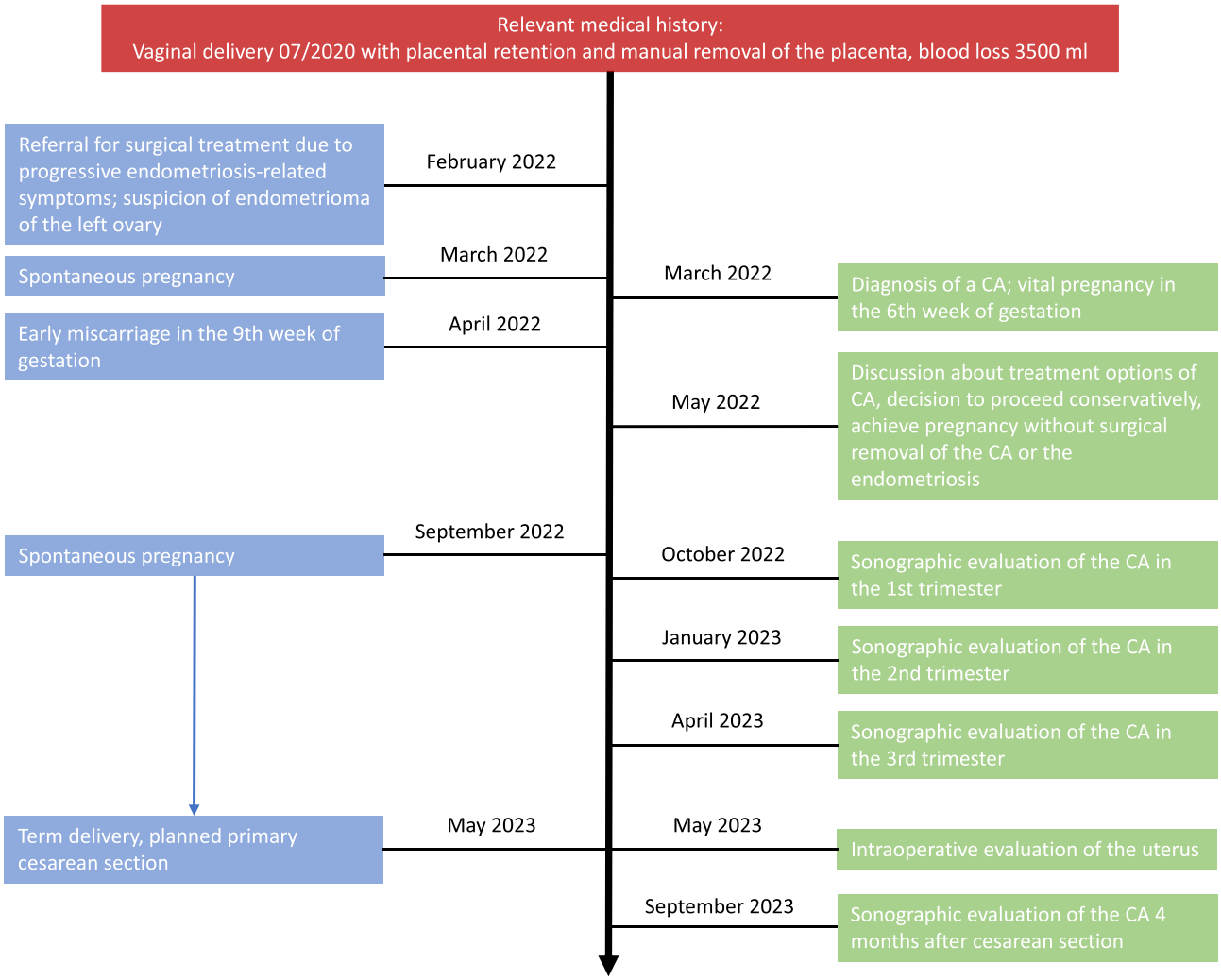

Figure 7 Diagram of the timeline for the episode of care.

ratings between 1 and 3.[11] A smaller case series by Kriplani et al on 4 patients with juvenile CA was also able to demonstrate a significant improvement in dysmenorrhea, at least during the first menstrual cycle after surgery, following laparoscopic CA resection.[14] Takeda et al described 2 cases of juvenile CA with severe early-onset dysmenorrhea. Laparoscopic excision was performed, in one case following repeat GnRH agonist administration, leading to the absence of dysmenorrhea following surgery.[10] Of the 2 cases of juvenile CA reported by Dadhwal et al, both suffering from severe dysmenorrhea and irregular pain in the lower abdomen, one was treated by laparotomy, the other by laparoscopic resection. Postoperatively, the patient who received the Pfannenstiel laparotomy experienced a complete resolution of the dysmenorrhea and pelvic pain (insufficient follow-up information was provided in the other case).[18]

Nonetheless, even if the current available data advocates that the most effective therapeutic approach is surgery, an important issue that has not been discussed sufficiently is the question whether freedom from symptoms over a longer period of time after surgery is primarily achieved by hormonal therapy often administered postoperatively.[9,10,16,18] If that were the case, then the argument that initial hormonal treatment leading to menstrual cycle suppression is only effective during the time of intake is pointless, as it results in the same measure: continuation of the suppression of the menstrual cycle by hormonal treatment, with or without surgery.

The majority of the surgical interventions were carried out minimally invasive by laparoscopy. Isolated cases of open excision by laparotomy have been reported, with equal resolution of dysmenorrhea and pelvic pain, even positive pregnancy outcomes following surgery.[18] Regardless, in this day and age, minimal invasive surgery should be offered,

especially to this young patient collective, without exception. The hysteroscopic approach will most likely remain a rarity as, according to the definition criteria of juvenile CAs by Takeuchi et al, there is typically no connection of the CA to the uterine cavity.[11]

It can be concluded from the data described above that in the majority of cases the indication for surgical management of the CA was pain, which is in accordance with the recommendations formulated by ESHRE regarding surgical treatment: "It is recommended to offer surgery as one of the options to reduce endometriosis-associated pain".[17]

The question as to whether preconceptional surgical intervention offers an actual benefit with regard to fertility and pregnancy outcome has not been sufficiently addressed by literature to date. And considering that the affected women are mainly of childbearing age, it would be important to do so.

In the case series by Kerbage et al 6 of the 18 patients underwent surgical resection of the CA for infertility. In the postoperative period 8 patients desired to become pregnant, of which 5 required assisted reproductive therapy (ART), 1 an insemination, and 4 in vitro-fertilization (IVF), resulting in a need for IVF in 50% of the women wanting to become pregnant after CA surgery. Finally, 7 patients delivered a total of 12 babies following CA surgery. How many of these were the result of IVF is not specified.[16] In their case series including 9 patients, Takeuchi et al reported 2 patients out of 3 with childbearing desire who conceived after surgery. In one case, the patient became pregnant and delivered vaginally at term twice within 2 years of surgery. In the other case, the patient conceived 7 years after the surgery and had an elective cesarean section at term.[11] Of the 2 cases of juvenile CA Dadhwal et al reported, one became pregnant and delivered a healthy newborn 3 years after surgery.[18]

Although, as seen above, successful pregnancies have been reported following surgical intervention for CA, the available data is insufficient to clearly demonstrate the benefit of an invasive surgical approach in the case of CA in terms of fertility and pregnancy outcome, and there are no data whatsoever on possible pregnancy complications following resection of CAs. Thus, it is of utmost importance to address the issue of possible complications and risks for future pregnancies caused by surgery involving incision and opening of the myometrium. To estimate said risks, the available data on myomectomy have to be consulted, as there are great parallels on the level of surgical procedure for the resection of both CA and myoma and a more extensive body of literature on the topic of pregnancy complications after myomectomy.

The most severe and potentially fatal complication of previous myomectomy is uterine rupture during pregnancy. The overall risk is small, comparable to the risk after a cesarean section (0.2–1%).[23–27] Incidences reported in literature vary and typically range from 0.2% to 1%.[26,28–35] Some studies, however, have reported considerably higher rates up to 3.7%[36] and 4.94%.[37] Uterine ruptures after preconceptional myomectomy mostly occur during the early or mid-third trimester of pregnancy and typically before onset of labor, much less frequently during labor, as opposed to uterine ruptures occurring after cesarean section.[26,34,36,38–40] Surgery-related factors thought to increase the risk of uterine rupture during pregnancy are as follows: use of electrocautery,[34,36,37,41–43] single-layer or non-multilayer suture,[36,37,41,42] and especially non-full-thickness suture, creating space for the development of a postoperative hematoma.[33,34,37]

According to current literature, a history of myomectomy furthermore increases the risk of preterm delivery, ranging from 3.1% to 35%,[28,35,44,45] of having a cesarean section at birth,[28,31,35,38] and of suffering from increased blood loss during delivery.[28,35,46] Additionally, preconceptional myomectomy is associated with a significantly higher risk of placenta accreta compared to a cohort without previous myomectomy, 1.5% vs 0.5%.[28,29] Breaching the endometrial cavity during laparoscopic myomectomy thereby significantly increases the risk of placenta accreta compared to the cases where there is no breach, 24% vs 5.2%, respectively.[47]

Finally, a very interesting study conducted by Kinugasa-Taniguchi et al in 2011 compared obstetric outcomes of women with a myoma present and women with a history of myomectomy and was able to show that outcomes were better in those pregnancies with a myoma present in terms of cesarean section rate, preterm delivery rate, and blood loss. Thus, the authors advocate for a conservative management of myomas, in particular if they are asymptomatic in women with a childbearing desire.[48]

Over the past few years, it has become more and more evident that endometriosis and adenomyosis are associated with adverse pregnancy outcomes including increased miscarriage rate, gestational hypertension, pre-eclampsia, placenta previa, preterm delivery, fetal malpresentation, pre- and post-partum hemorrhage, low birth

weight (LBW), small for gestational age (SGA) and overall reduced live birth rates.[49–53] Especially women with deep infiltrating endometriosis (DIE) are at risk for severe complications in the course of pregnancy. Bowel perforation, predominantly at the level of the rectum and sigmoid, and spontaneous hematoperitoneum from decidualized endometriotic lesions have been reported in connection with this patient collective.[49,53–55] However, even if the prevalence of these potentially life-threatening events is unknown, they are very rare and the relative risk correspondingly very low.[53,55]

Mooney et al carried out a review of literature to address the question whether pre-pregnancy surgery for endometriosis leads to an improvement in obstetric outcomes and found that there was insufficient data to estimate the impact of endometriosis surgery on pregnancy outcomes. However, the studies available suggest that pre-pregnancy surgery might even worsen obstetric outcomes in terms of increased risks of placenta praevia, caesarean delivery, obstetric hemorrhage, gestational hypertensive conditions, and preterm delivery.[52]

At this point in time, the evidence to hand does not support prophylactic pre-pregnancy surgery for endometriosis in order to avoid adverse pregnancy outcomes related to this condition.

The data available show clearly that we need to exercise more restraint with regard to surgical treatment of benign diseases for any other indication than symptoms, first and foremost pain, as data does not support prophylactic intervention in terms of obstetrical outcome. Especially in diseases such as endometriosis including CAs, the intention to do good has the potential to do harm.

Conclusion

CAs are rare and affect primarily women of childbearing age. Thus, considerations of fertility, pregnancy, and especially pregnancy outcome are of crucial relevance. The lack of sufficient data and evidence, however, makes conscientious preconceptional counseling of women with CA all the more difficult. As long as it cannot be shown that there is a clear benefit of preconceptional surgical resection of a CA with regard to fertility and obstetrical outcome, it is essential to consider very carefully the possible risks that surgery to the uterus can have on future pregnancies and not imprudently or automatically opt for surgery in the case of CA diagnosis, especially not without exhausting conservative, medical therapeutic options.

We herein propose the feasibility of a conservative, non-surgical management preconceptionally in women with a cystic adenomyoma and the desire to conceive. Patient counseling on management of CAs, especially in women of childbearing age, must include the possibility of a conservative, non-surgical approach; however, more data must be collected to be able to offer sound advice to patients wishing to conceive.

Abbreviations

ART, assisted reproductive therapy; CA, cystic adenomyoma; DIE, deep infiltrating endometriosis; ESHRE, European Society of Human Reproduction and Embryology; IVF, in vitro fertilization; LBW, low birth weight; SGA, small for gestational age; VAS, visual analogue scale.

Data Sharing Statement

The original data used for this article (complete patient history as well as ultrasound and intraoperative images) are available from the corresponding author upon reasonable request.

Ethics Approval and Consent to Participate

Institutional approval for the publication of the case details was not required.

Consent for Publication

Written informed consent was obtained from the patient for publication of her case as well as the accompanying images. A copy of the written consent is available from the corresponding author upon reasonable request.

Acknowledgments

We would like to thank Carolyn Nabholz for English language corrections.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising, or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Disclosure

The authors report no conflicts of interest in this work.

References

1. Bergeron C, Amant F, Ferenczy A. Pathology and physiopathology of adenomyosis. *Best Pract Res Clin Obstet Gynaecol*. 2006;20(4):511–521. doi:10.1016/j.bpobgyn.2006.01.016
2. Harmsen MJ, Van den Bosch T, de Leeuw RA, et al. Consensus on revised definitions of Morphological Uterus Sonographic Assessment (MUSA) features of adenomyosis: results of modified Delphi procedure. *Ultrasound Obstet Gynecol*. 2022;60(1):118–131. doi:10.1002/uog.24786
3. Munro MG. Classification and Reporting Systems for Adenomyosis. *J Minim Invasive Gynecol*. 2020;27(2):296–308. doi:10.1016/j.jmig.2019.11.013
4. Leyendecker G. Endometriosis: a dysfunction and disease of the archimetra. *Hum Reprod Update*. 1998;4(5):752–762. doi:10.1093/humupd/4.5.752
5. Leyendecker G, Wildt L. A new concept of endometriosis and adenomyosis: tissue injury and repair (TIAR). *Horm Mol Biol Clin Investig*. 2011;5(2). doi:10.1515/HMBCI.2011.002
6. Brosens I, Gordts S, Habiba M, Benagiano G. Uterine Cystic Adenomyosis: a Disease of Younger Women. *J Pediatr Adolesc Gynecol*. 2015;28(6):420–426. doi:10.1016/j.jpag.2014.05.008
7. Habiba M, Benagiano G. Classifying Adenomyosis: progress and Challenges. *Int J Environ Res Public Health*. 2021;18(23):12386. doi:10.3390/ijerph182312386
8. Slezak P, Tillinger KG. The Incidence and Clinical Importance of Hysterogenic Evidence of Cavities in the Uterine Wall. *Radiology*. 1976;118(3):581–586. doi:10.1148/118.3.581
9. Cucinella G, Billone V, Pitruzzella I, Lo Monte AI, Palumbo VD, Perino A. Adenomyotic Cyst in a 25-Year-Old Woman: case Report. *J Minim Invasive Gynecol*. 2013;20(6):894–898. doi:10.1016/j.jmig.2013.04.022
10. Takeda A, Sakai K, Mitsui T, Nakamura H. Laparoscopic management of juvenile cystic adenomyoma of the uterus: report of two cases and review of the literature. *Journal of Minimally Invasive Gynecology*. 2007;14(3):370–374. doi:10.1016/j.jmig.2007.01.005
11. Takeuchi H, Kitade M, Kikuchi I, Kumakiri J, Kuroda K, Jinushi M. Diagnosis, laparoscopic management, and histopathologic findings of juvenile cystic adenomyoma: a review of nine cases. *Fertil Steril*. 2010;94(3):862–868. doi:10.1016/j.fertnstert.2009.05.010
12. Branquinho MM, Marques AL, Leite HB, Silva IS. Juvenile cystic adenomyoma. *Case Rep*. 2012;2012(nov16 1):bcr2012007006–bcr2012007006. doi:10.1136/bcr-2012-007006
13. Cullen TS. Adenomyoma of the uterus. *J Am Med Assoc*. 1908;L(2):107. doi:10.1001/jama.1908.25310280023002d
14. Kriplani A, Mahey R, Agarwal N, Bhatla N, Yadav R, Singh MK. Laparoscopic Management of Juvenile Cystic Adenomyoma: four Cases. *J Minim Invasive Gynecol*. 2011;18(3):343–348. doi:10.1016/j.jmig.2011.02.001
15. Fisseha S, Smith YR, Kumetz LM, Mueller GC, Hussain H, Quint EH. Cystic myometrial lesion in the uterus of an adolescent girl. *Fertil Steril*. 2006;86(3):716–718. doi:10.1016/j.fertnstert.2006.03.019
16. Kerbage Y, Dericquebourg S, Collinet P, Verpillat P, Giraudet G, Rubod C. Cystic adenomyoma surgery. *J Gynecol Obstet Hum Reprod*. 2022;51(3):102313. doi:10.1016/j.jogoh.2022.102313
17. Becker CM, Bokor A, Heikinheimo O, et al. ESHRE guideline: endometriosis. *Hum Reprod Open*. 2022;2022(2):hoac009. doi:10.1093/hropen/hoac009
18. Dadhwal V, Sharma A, Khoiwal K. Juvenile Cystic Adenomyoma Mimicking a Uterine Anomaly: a Report of Two Cases. *Eurasian J Med*. 2017;49(1):59–61. doi:10.5152/eurasianjmed.2017.17028
19. Acién P, Acién M, Fernández F, José Mayol M, Aranda I. The Cavitated Accessory Uterine Mass: a Müllerian Anomaly in Women With an Otherwise Normal Uterus. *Obstet Gynecol*. 2010;116(5):1101–1109. doi:10.1097/AOG.0b013e3181f7e735
20. Acien P, Bataller A, Fernandez F, Acien MI, Rodriguez JM, Mayol MJ. New cases of accessory and cavitated uterine masses (ACUM): a significant cause of severe dysmenorrhea and recurrent pelvic pain in young women. *Hum Reprod*. 2012;27(3):683–694. doi:10.1093/humrep/der471
21. Bedaiwy MA, Henry DN, Elguero S, Pickett S, Greenfield M. Accessory and Cavitated Uterine Mass with Functional Endometrium in an Adolescent: diagnosis and Laparoscopic Excision Technique. *J Pediatr Adolesc Gynecol*. 2013;26(4):e89–e91. doi:10.1016/j.jpag.2012.11.003
22. Koukoura O, Kapsalaki E, Daponte A, Pistofidis G. Laparoscopic treatment of a large uterine cystic adenomyosis in a young patient. *BMJ Case Rep*. 2015;bcr2015210358. doi:10.1136/bcr-2015-210358
23. Gyamfi-Bannerman C, Gilbert S, Landon MB, et al. Risk of Uterine Rupture and Placenta Accreta With Prior Uterine Surgery Outside of the Lower Segment. *Obstet Gynecol*. 2012;120(6):1332–1337. doi:10.1097/AOG.0b013e318273695b

24. Justus Hofmeyr G, Say L, Metin Gülmezoglu A. SYSTEMATIC REVIEW: WHO systematic review of maternal mortality and morbidity: the prevalence of uterine rupture: SYSTEMATIC REVIEW. *BJOG Int J Obstet Gynaecol*. 2005;112(9):1221–1228. doi:10.1111/j.1471-0528.2005.00725.x
25. Sentilhes L, Vayssière C, Beucher G, et al. Delivery for women with a previous cesarean: guidelines for clinical practice from the French College of Gynecologists and Obstetricians (CNGOF). *Eur J Obstet Gynecol Reprod Biol*. 2013;170(1):25–32. doi:10.1016/j.ejogrb.2013.05.015
26. Claeys J, Hellendoorn I, Hamerlynck T, Bosteels J, Weyers S. The risk of uterine rupture after myomectomy: a systematic review of the literature and meta-analysis. *Gynecol Surg*. 2014;11(3):197–206. doi:10.1007/s10397-014-0842-8
27. Landon MB, Hauth JC, Leveno KJ, et al. Maternal and Perinatal Outcomes Associated with a Trial of Labor after Prior Cesarean Delivery. *N Engl J Med*. 2004;351(25):2581–2589. doi:10.1056/NEJMoa040405
28. Milazzo GN, Catalano A, Badia V, Mallozzi M, Caserta D. Myoma and myomectomy: poor evidence concern in pregnancy: myoma and myomectomy in pregnancy. *J Obstet Gynaecol Res*. 2017;43(12):1789–1804. doi:10.1111/jog.13437
29. Komatsu H, Taniguchi F, Harada T. Impact of myomectomy on the obstetric complications: a large cohort study in Japan. *Int J Gynecol Obstet*. 2023;ijgo.14767. doi:10.1002/ijgo.14767
30. Koo YJ, Lee JK, Lee YK, et al. Pregnancy Outcomes and Risk Factors for Uterine Rupture After Laparoscopic Myomectomy: a Single-Center Experience and Literature Review. *J Minim Invasive Gynecol*. 2015;22(6):1022–1028. doi:10.1016/j.jmig.2015.05.016
31. Ordás P, Spagnolo E, Fernández LGL, et al. Comparison of surgical and obstetric outcomes in women with uterine leiomyomas after laparoscopic vs. abdominal myomectomy: a single-center cohort study. *Front Surg*. 2022;9:997078. doi:10.3389/fsurg.2022.997078
32. Spong CY, Landon MB, Gilbert S, et al. Risk of Uterine Rupture and Adverse Perinatal Outcome at Term After Cesarean Delivery. *Obstet Gynecol*. 2007;110(4):801–807. doi:10.1097/01.AOG.0000284622.71222.b2
33. Sizzi O, Rossetti A, Malzoni M, et al. Italian multicenter study on complications of laparoscopic myomectomy. *J Minim Invasive Gynecol*. 2007;14(4):453–462. doi:10.1016/j.jmig.2007.01.013
34. Dubuisson JB. Pregnancy outcome and deliveries following laparoscopic myomectomy. *Hum Reprod*. 2000;15(4):869–873. doi:10.1093/humrep/15.4.869
35. Lee SJ, Ko HS, Na S, et al. Nationwide population-based cohort study of adverse obstetric outcomes in pregnancies with myoma or following myomectomy: retrospective cohort study. *BMC Pregnancy Childbirth*. 2020;20(1):716. doi:10.1186/s12884-020-03406-9
36. Kim MS, Uhm YK, Kim JY, Jee BC, Kim YB. Obstetric outcomes after uterine myomectomy: laparoscopic versus laparotomic approach. *Obstet Gynecol Sci*. 2013;56(6):375. doi:10.5468/ogs.2013.56.6.375
37. Cui TY, Fei LT, Mei DY. Pregnancy outcomes following different surgical approaches of myomectomy: pregnancy outcomes after TAM and LM. *J Obstet Gynaecol Res*. 2015;41(3):350–357. doi:10.1111/jog.12532
38. Gambacorti-Passerini Z, Gimovsky AC, Locatelli A, Berghella V. Trial of labor after myomectomy and uterine rupture: a systematic review. *Acta Obstet Gynecol Scand*. 2016;95(7):724–734. doi:10.1111/aogs.12920
39. Margueritte F, Adam C, Fauconnier A, Gauthier T. Time to conceive after myomectomy: should we advise a minimum time interval? A systematic review. *Reprod Biomed Online*. 2021;43(3):543–552. doi:10.1016/j.rbmo.2021.05.016
40. Kim HS, Oh SY, Choi SJ, et al. Uterine rupture in pregnancies following myomectomy: a multicenter case series. *Obstet Gynecol Sci*. 2016;59(6):454. doi:10.5468/ogs.2016.59.6.454
41. Parker WH, Einarsson J, Istre O, Dubuisson JB. Risk Factors for Uterine Rupture after Laparoscopic Myomectomy. *J Minim Invasive Gynecol*. 2010;17(5):551–554. doi:10.1016/j.jmig.2010.04.015
42. Wu X, Jiang W, Xu H, Ye X, Xu C. Characteristics of uterine rupture after laparoscopic surgery of the uterus: clinical analysis of 10 cases and literature review. *J Int Med Res*. 2018;46(9):3630–3639. doi:10.1177/0300060518776769
43. Cobellis L, Pecori E, Cobellis G. Comparison of intramural myomectomy scar after laparotomy or laparoscopy. *Int J Gynecol Obstet*. 2004;84(1):87–88. doi:10.1016/S0020-7292(03)00301-1
44. Fagherazzi S, Borgato S, Bertin M, Vitagliano A, Tommasi L, Conte L. Pregnancy outcome after laparoscopic myomectomy. *Clin Exp Obstet Gynecol*. 2014;41(4):375–379.
45. Mahalingam M, Hu M, Schointuch M, et al. Uterine myomas: effect of prior myomectomy on pregnancy outcomes. *J Matern Fetal Neonatal Med*. 2022;35(25):8492–8497. doi:10.1080/14767058.2021.1984424
46. Gimovsky AC, Frangieh M, Phillips J, et al. Perinatal outcomes of women undergoing cesarean delivery after prior myomectomy. *J Matern Fetal Neonatal Med*. 2020;33(13):2153–2158. doi:10.1080/14767058.2018.1542680
47. Wada S, Fukushi Y, Ono Y, Ota H, Tsuzuki Y, Yamada H. Influence of uterine cavity breach in laparoscopic myomectomy on the risk of obstetric complications. *Gynecol Minim Invasive Ther*. 2022;11(4):221. doi:10.4103/gmit.gmit_146_21
48. Kinugasa-Taniguchi Y, Ueda Y, Hara-Ohyagi C, Enomoto T, Kanagawa T, Kimura T. Impaired delivery outcomes in pregnancies following myomectomy compared to myoma-complicated pregnancies. *J Reprod Med*. 2011;56(3–4):567.
49. Vercellini P, Viganò P, Bandini V, Buggio L, Berlanda N, Somigliana E. Association of endometriosis and adenomyosis with pregnancy and infertility. *Fertil Steril*. 2023;119(5):727–740. doi:10.1016/j.fertnstert.2023.03.018
50. Nirgianakis K, Gasparri ML, Radan AP, et al. Obstetric complications after laparoscopic excision of posterior deep infiltrating endometriosis: a case–control study. *Fertil Steril*. 2018;110(3):459–466. doi:10.1016/j.fertnstert.2018.04.036
51. Shi J, Dai Y, Zhang J, Li X, Jia S, Leng J. Pregnancy outcomes in women with infertility and coexisting endometriosis and adenomyosis after laparoscopic surgery: a long-term retrospective follow-up study. *BMC Pregnancy Childbirth*. 2021;21(1):383. doi:10.1186/s12884-021-03851-0
52. Mooney SS, Ross V, Stern C, Rogers PAW, Healey M. Obstetric Outcome After Surgical Treatment of Endometriosis: a Review of the Literature. *Front Reprod Health*. 2021;3:750750. doi:10.3389/frph.2021.750750
53. Mittelstadt S, Stäbler A, Kolb M, et al. Acute endometriosis-related sigmoid perforation in pregnancy- case report. *BMC Pregnancy Childbirth*. 2022;22(1):647. doi:10.1186/s12884-022-04973-9
54. Pisanu A. Rectal perforation from endometriosis in pregnancy: case report and literature review. *World J Gastroenterol*. 2010;16(5):648. doi:10.3748/wjg.v16.i5.648
55. Setúbal A, Sidiropoulou Z, Torgal M, Casal E, Lourenço C, Koninckx P. Bowel complications of deep endometriosis during pregnancy or in vitro fertilization. *Fertil Steril*. 2014;101(2):442–446. doi:10.1016/j.fertnstert.2013.11.001