

A Bicornuate Uterus with a Unilateral Focal Adenomyosis: A Case Report

Yohanes Iddo Adventa, Anita Rachmawati, Dian Tjahyadi, Mulyanusa Amarullah Ritonga 

Division of Fertility and Reproductive Endocrinology, Department of Obstetrics and Gynecology, Faculty of Medicine, Dr. Hasan Sadikin General Hospital, Padjadjaran University, Bandung, Indonesia

Correspondence: Yohanes Iddo Adventa, Division of Fertility and Reproductive Endocrinology, Department of Obstetrics and Gynecology, Faculty of Medicine, Dr. Hasan Sadikin General Hospital, Padjadjaran University, Bandung, Indonesia, Tel +62 82297669494, Email yohanesiddo@gmail.com

Background: The etiology of adenomyosis remains uncertain, and factors such as pregnancy, postpartum endometritis, cesarean delivery, uterine surgery, and uterine trauma are suspected to cause myometrial disruption that may trigger smooth muscle hyperplasia.

Case: We present a case of a bicornuate uterus with focal adenomyosis confined to the left hemiuterus in a 43-year-old woman who experienced chronic pelvic pain, severe progressive dysmenorrhea, and secondary infertility for three years. She had previously been diagnosed with a bicornuate uterus during two cesarean sections and had a history of curettage, both of which may have contributed to the localized pathology. After thorough counseling regarding management options, a laparotomy total abdominal hysterectomy was performed primarily for pain relief. The postoperative course was uneventful, and the patient was discharged on the third day with complete resolution of pelvic pain, reporting a visual analogue score of 0/10 at follow-up.

Discussion: Previous caesarean sections and curettage are possible in cases of a bicornuate uterus with adenomyosis pathology of hemiuterus. Pain caused by adenomyosis can be managed with laparotomy total abdominal hysterectomy as a gold standard management.

Keywords: adenomyosis, bicornuate uterus, hemiuterine

Introduction

An enlarged uterus, dysmenorrhea, and menorrhagia are common symptoms of adenomyosis, a benign gynecological tumor that accounts for 5–70% of these cases on surgical and autopsy specimens. It is characterized by the presence of ectopic endometrial glands and stroma in the myometrium. The degree and penetration depth of adenomyosis are thought to be connected to the symptoms of the condition.¹ The etiology of adenomyosis is unknown, although pregnancy, postpartum endometritis, cesarean delivery, uterine surgery, and uterine trauma are suspected causes of myometrial disruption that may induce adjacent smooth muscle hyperplasia. However, a clear correlation between pathological findings and the severity of symptoms is absent.^{1,2}

Bicornuate uterus is a congenital incomplete fusion of the bilateral müllerian duct causes the uterine body's right and left cavities to merge above or at the level of the cervix. There have been reports that uterine malformations affect anywhere between 1 in 10 and 1 in 1600 women.¹ The relationship between congenital uterine anomalies and adenomyosis is not commonly documented in the literature. In this unusual instance, unilateral adenomyosis in a bicornuate uterus is described.

Case

A 43-year-old woman, G3P2A1, presented with a 3-year history of dyspareunia and dysmenorrhea. According to her medical records, she had a bicornuate uterus, which was discovered during earlier caesarean section (C-section). Her medical history included two uncomplicated C-sections (Caesarean sections) delivered at term in 2007 and 2011, with baby weights of 2.8 kg and 3.1 kg, respectively. She had a history of curettage due to

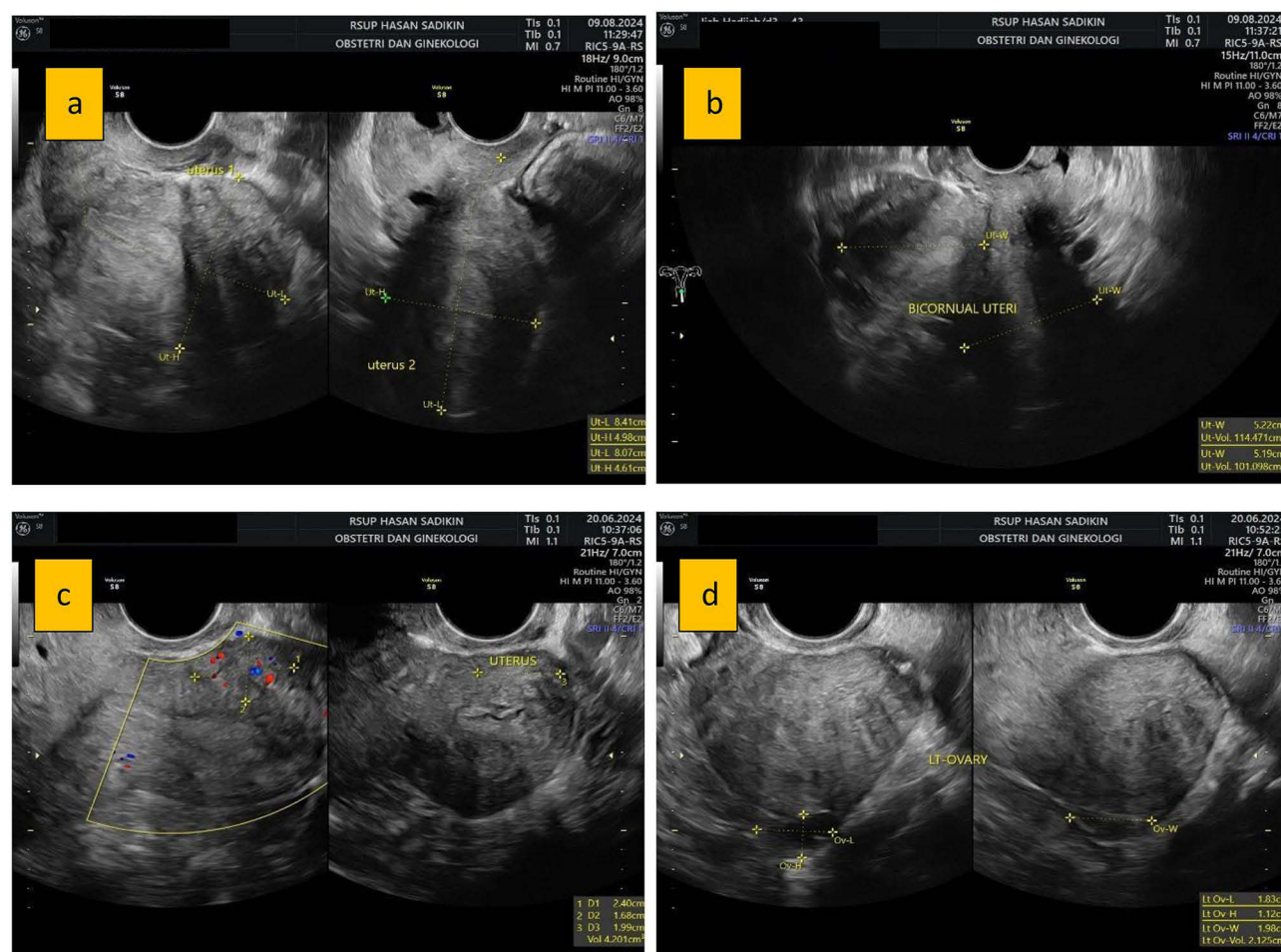


Figure 1 (a and b) transvaginal ultrasonography revealed an anteverted and divided uterus (unicollis bicornuate uterus) measuring 8.41×4.98 x 5.11 cm on the left side with focal adenomyosis and 8.07×4.61 x 5.19 cm on the right side; (c) focal posterior adenomyosis; (d) normal bilateral ovaries.

incomplete abortion in 2011. She was found to have a single cervix on pelvic examination and an unusually wide uterine corpus. Transvaginal ultrasonography revealed an anteverted and divided uterus measuring 8.41×4.98 x 5.11 cm on the left side and 8.07×4.61 x 5.19 cm on the right side. Posterior wall thickening and focally heterogeneous myometrium in the left uterine cavity were also noted as a focal adenomyosis (Figure 1a and b). MRI of the pelvis identified a unicollis bicornuate uterus (class U3b/C0 bicorporeal uterus based on the ESHRE/ESGE classification) with normal renal and ureteric development (Figure 2). There was no dysmenorrhea or diagnosis of adenomyosis identified prior to the curettage or cesarean sections. No surgical signs of endometriosis were observed on either side of the uterus during any of the cesarean procedures.

Because of her intractable dysmenorrhea, the patient underwent laparotomy total abdominal hysterectomy. A bicornuate uterus was removed, and it was discovered that the left cornua were significantly larger than the right cornua (Figure 3). A longitudinal incision showed focal adenomyosis. In contrast, the right uterine corpus had a normal myometrial appearance. Both cornua communicated with the vaginal canal through a unique cervical canal. A detailed histopathological assessment was performed (Figure 4). Our examination confirmed that adenomyosis had affected only the left uterine myometrium; the right uterine myometrium was normal (Figure 4). The patient was discharged on the third postoperative day in a stable condition.

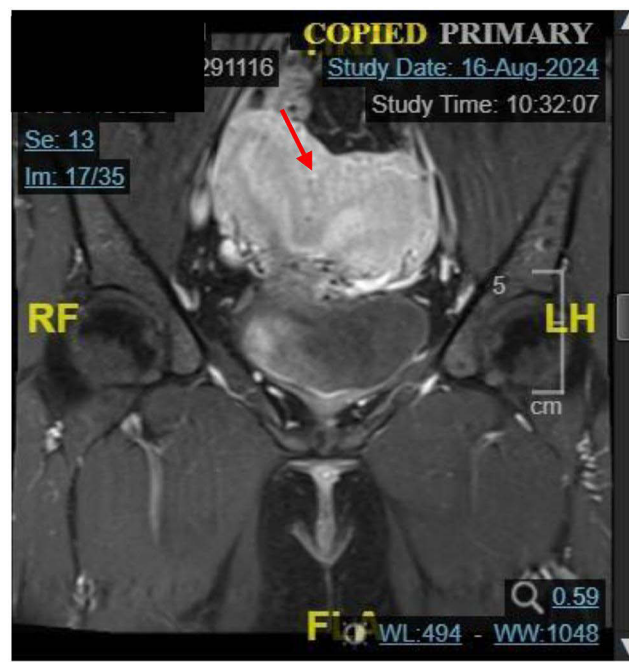


Figure 2 MRI of the pelvis identified a unicornuate uterus (red arrow).

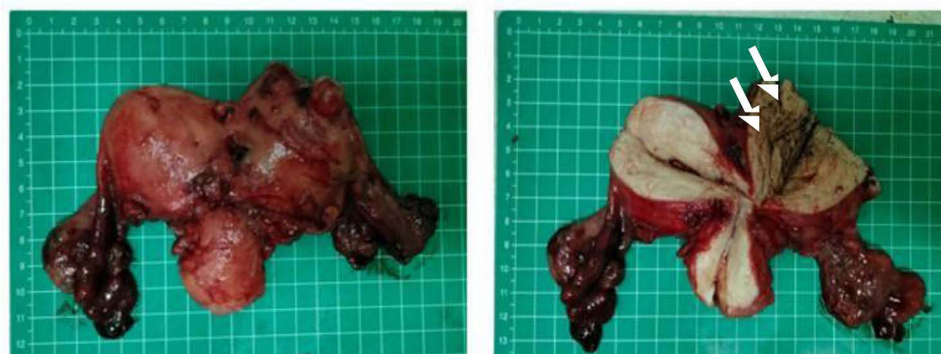


Figure 3 Gross view of unicornuate uterus with adenomyosis (white arrow) in left and a normal right corpus (left picture: before incision, right picture: after incision).

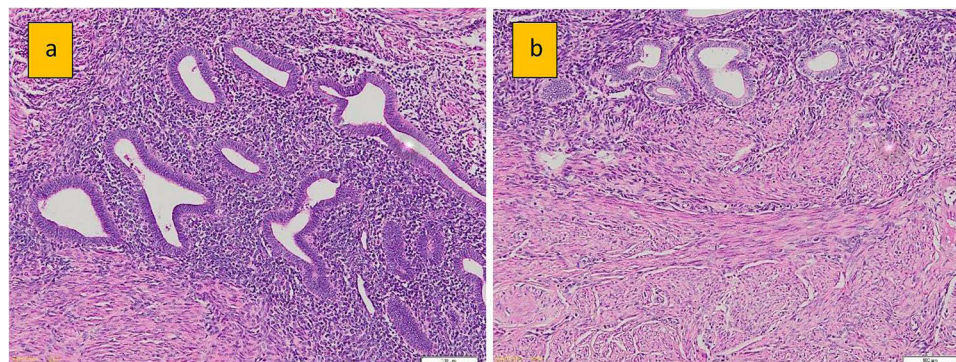


Figure 4 Histology of unicornuate uterus, stained with hematoxylin-eosin, X100, original magnification. (a) Left uterus: endometrial glands and stroma within myometrium; (b) Right uterus: normal myometrium.

Discussion

Since 2005, there has only been a single instance of adenomyosis in a bicornuate uterus in the literature.² The case presented here seems to be an uncommon instance of unilateral adenomyosis of the bicornuate uterus. Uncertainty surrounds the genesis and developmental processes that result in adenomyosis. Most people now agree that the usual border between the myometrium and the basal endometrial layer is disrupted, leading to the downgrowth and invagination of the basalis endometrium into the myometrium, which causes adenomyosis. Direct continuity between the underlying adenomyosis in the myometrium and the basalis endometrium is frequently observed histologically. Although the exact source of this disruption is unknown, potential causes include pregnancy, cesarean delivery, endometritis following childbirth, uterus surgery, and uterine trauma. A number of theories have been developed and examined in an effort to understand the mechanisms of endometriosis.^{3,4}

In this case, histological examination following a hysterectomy was done as a gold standard diagnostic tool. Examination confirmed that adenomyosis had affected only the left uterine myometrium; the right uterine myometrium was normal. Histological examination was the primary method used in the past to diagnose adenomyosis, which frequently understated the condition's importance and prevalence. Advances in imaging technology was performed in this case, including as MRI and transvaginal ultrasound, have greatly improved diagnosis and revealed a wider demography of affected persons, especially younger women of reproductive age.^{5,6} This change has brought attention to adenomyosis's higher than previously thought prevalence, which is thought to impact 15–20% of young women and frequently coexists with uterine fibroids and endometriosis.⁶ However, adenomyosis was not diagnosed using MRI in this case. MRI would likely show characteristic features such as thickened junctional zone exceeding 12 mm, intramyometrial cyst(s), heterogeneous myometrium with hyperintense regions on T2-weighted images, and possibly asymmetrical myometrial bulkiness typical for adenomyosis. Only unicollis bicornuate uterus was accurately diagnosed by MRI. In one study involving 120 consecutive patients referred for hysterectomy, the following sensitivities and specificities were observed: TVUS has sensitivity approximately 65% and specificity around 97.5%; and MRI has sensitivity about 77.5% and specificity: roughly 92.5%. These findings indicate that while both modalities have high specificity, MRI tends to offer slightly higher sensitivity compared to TVUS.⁵ Based on a meta-analytic comparison: Multiple meta-analytical reviews have consolidated data from various studies on this topic. For instance, a meta-analysis reported pooled sensitivity values ranging between MRI at approximately 69% to TVUS at about 75%, with specificities generally high across both modalities but slightly favouring MRI's specificity rate.^{7,8}

Similarly, the prevalence of adenomyosis between 20% and 30% when hysterectomies are done for urogynecological causes. According to certain research, prevalence rates can be significantly greater among women who experience dysmenorrhea and abnormal uterine bleeding, ranging from 30% to 59%. Remarkably, 80% of infertile women with endometriosis also have adenomyosis, with more than 40% of these cases involving deep-infiltrating endometriosis (DIE) lesions. Additionally, 40% of women with endometriosis also have adenomyosis present. Approximately 20% to 25% of women receiving assisted reproductive technologies (ARTs) have adenomyosis.⁶ More awareness is essential for proper care and diagnosis strategies. Adenomyosis is mostly characterized by abnormal uterine bleeding (AUB) and pain, such as pelvic pain and dysmenorrhea.^{6,7} Endometriosis and uterine fibroids are two common accompanying disorders with similar symptomatology, making it difficult to determine which pathology is responsible for a given symptom. Between 30% and 68% of patients with adenomyosis experience dysmenorrhea. Adenomyosis patients frequently have deep dyspareunia, which frequently resembles the dyspareunia of patients with deep-infiltrating endometriosis. Similar to the patient in this instance, the primary symptoms to be managed are the classic symptoms of dyspareunia of adenomyosis and dysmenorrhea. Although the exact cause of the discomfort is unknown, prostaglandins and chronic inflammation may be key factors.⁶

While there is ongoing debate and mixed evidence regarding the exact role of C-section in the development of adenomyosis, several studies suggest a potential association between C-section and adenomyosis, albeit with varying degrees of significance. A case-control study among 189 women with adenomyosis, 25% had undergone a C-section, whereas this figure was 14% among those without adenomyosis, indicating a stronger association (OR 2.08).⁹ An observational data suggested that patients with adenomyosis tended to have longer durations of menstruation both before

and after C-section, implying potential disruptions in uterine tissue integrity and possibly contributing to adenomyosis development.^{10,11} While there is some indication that C-section might be associated with an increased risk of adenomyosis, particularly through mechanical disruption of the uterine lining, the evidence is not conclusive across all studies. Further research is needed to fully understand this relationship, while there is no definitive proof linking C-section directly to adenomyosis, existing literature suggests a plausible connection warranting continued investigation. The possible pathogenesis in this case is in reaction to estrogen and/or inflammation, or in response to a repeated TIAR-induced stress reaction, a cascade of endometrial-myometrial transition (EMT) may occur at endometrium-myometrium interface.¹² Elizabeth Hay introduced the terms mesenchymal-epithelial transition (MET) and epithelial-mesenchymal transition (EMT) almost fifty years ago. An effective reaction to cues that induce epithelial migration (EMT) can facilitate the disintegration of intercellular adhesion complexes and the reduction of apico-basal polarity in epithelial cells. For cells to exit the epithelium and control their capacity for migration, this EMT characteristic is essential. As a result, since there is no basement membrane between the inner myometrium (junctional zone) and the basalis endometrium, occurrences of EMT may occur at EMI.¹³

By invaginating the endometrium basalis into the myometrium, contractions of the myometrium itself and the resulting damage to the endometrial myometrial junction zone (JZ), a highly specialized hormone-responsive layer of the uterine architecture situated in the inner third of the myometrium, may cause adenomyosis.¹⁴ Leyendecker's "invagination theory" was based on the tissue injury and repair (TIAR) idea increased specifically, when the TIAR system is triggered in response to an injury or trauma, local paracrine activity causes the release of increased estrogen, which increases uterine contractility. An enhanced release of estrogens, auto-traumatization, and wound healing all contribute to a vicious cycle that encourages inflammation and, once more, the synthesis of local estradiol. In addition, a novel theory called endometrial myometrial interface disruption, or EMID, has been put forth. According to this theory, which updates the tissue injury and healing theory, uterine procedures that result in EMID may develop "iatrogenic" adenomyosis in later life. EMID can, in fact, be elicited mechanically or thermally (as in electrocoagulation). Both approaches cause tissue damage, which triggers the release of substance P, which functions as an immune modulator. They also trigger the hypothalamic-pituitary-adrenal axis, which increases the release of catecholamines like adrenaline and noradrenaline, potentially lowering cell-mediated immunity. Furthermore, it was shown that EMID, whether it is produced mechanically or thermally, can induce adenomyosis in mice, and that the likelihood of causing adenomyosis seems to be correlated with the severity of the EMID.¹⁵ The EMID hypothesis encompasses hypoxia at the wounding site, bone-marrow-derived stem cell recruitment, epithelial mesenchymal transition, and improved survival of endometrial cells that have been dispersed and displaced as a result of iatrogenic operations.¹²

This patient's successful cesarean section pregnancies are one of the uncommon instances of mullerian abnormalities. Pregnancies with uterine malformations are quite uncommon, and many of them start out without any symptoms. Patients who experience frequent pregnancy losses and fetal malpresentation.^{6,16} Fertility and reproduction issues affect 15% to 25% of women with uterine abnormalities. These individuals usually have higher rates of preterm birth (15–25%), miscarriage (25%), and cervical insufficiency (38%). Malpresentations, aberrant placental and ectopic pregnancies, and poor fetal growth are other consequences. Nonetheless, some pregnancies may be carried to term even though they are less frequent. In fact, other successful pregnancies in patients with a bicornuate uterus exist in the literature. In 2013, a woman diagnosed with bicornuate uterus was able to give birth to a live male infant weighting 2.8 kg. In fact, numerous studies have been done to assess the reproductive performance in women with bicornuate uterus. In a study of 21 patients with bicornuate uterus it was found that the probability of giving birth to a live-born infant in patient with bicornuate uterus with no corrective surgery was 30%, 58% and 79% for the first, second and third pregnancy respectively.¹⁶

The optimum course of action for this patient after a failed oral analgetic treatment, whose need for fertility is complete, is a hysterectomy. In particular, treatment for adenomyosis should be customized for each patient based on their symptoms, such as infertility, pain, or heavy menstrual bleeding. Medical options include non-steroidal anti-inflammatory drugs, tranexamic acid, combined oral contraceptives, levonorgestrel intrauterine system, dienogest, other progestins, or gonadotropin-releasing analogues; interventional options include uterine artery embolization; and surgical options include endometrial ablation, adenomyosis excision, or hysterectomy.¹⁷ The hemiuterus of a bicornuate uterus can also be safely removed, just like in the

case of a didelphys uterus with hemiuterine pathology, using a minimally invasive approach that relieves adenomyosis pain, promotes a speedy recovery, and maintains future fertility.¹⁸

Numerous data points to a strong correlation between the occurrence of adenomyosis and pregnancy, which appears to create a statistically significant predisposition to adenomyosis.⁸ However, it is still unclear whether pregnancy causes or exacerbates adenomyosis, either mechanically or hormonally. A history of a normal pregnancy in the left uterus cavity with uterine damage from curettage was provided for the patient profiled here. More accurate evidence for the idea of pregnancy-related adenomyosis may be obtained by using the right uterine cornua of a bicornuate uterus as a control following a pregnancy in the left cornua.⁸ In conclusion, the subsequent development of adenomyosis in the left cornua lends weight to theories that suggest pregnancy or other acquired factors may be involved in the pathogenesis and development of adenomyosis.

Conclusion

This case demonstrates that adenomyosis may be associated with pregnancy-related or acquired uterine factors, particularly in the context of prior cesarean sections and uterine instrumentation, as shown by the unilateral involvement of only one horn of the bicornuate uterus. In this patient, the later development of focal adenomyosis was accompanied by secondary infertility and severe dysmenorrhea, and total abdominal hysterectomy provided complete symptom relief and improved quality of life. Surgical intervention remains an effective option for patients with refractory symptoms, even though the exact pathophysiology of adenomyosis is still not fully understood.

Informed Consent Statement

Written informed consent was obtained from the patient for the publication of this case report and the accompanying images. All identifying information has been removed to ensure patient confidentiality.

Disclosure

The authors report no conflicts of interest in this work.

References

1. Su HY, Chen CH, Gao HW, Liu JY. A bicornuate uterus with a unilateral cornual adenomyosis. *Obstetrics Gynecol.* 2005;105(5 II):1191–1193. doi:10.1097/01.AOG.0000147838.04119.bd
2. Kong WY, Zhao SR, Deng K, Zhang Q, Liu W, Yan L. Effects of bicornuate uterus on pregnancy and obstetric outcomes of in vitro fertilization / intracytoplasmic sperm injection. *Eur J Obstetrics Gynecol Reprod Biol.* 2021;258:132–138. doi:10.1016/j.ejogrb.2020.12.046
3. Grimbizis GF, Gordts S, Di Spiezio Sardo A, et al. The ESHRE/ESGE consensus on the classification of female genital tract congenital anomalies. *Hum Reprod.* 2013;28(8):2032–2044. doi:10.1093/humrep/det098
4. Pfeifer SM, Attaran M, Goldstein J, et al. ASRM müllerian anomalies classification 2021. *Fertil Steril.* 2021;116(5):1238–1252. doi:10.1016/j.fertnstert.2021.09.025
5. Bazot M, Cortez A, Darai E, et al. Ultrasonography compared with magnetic resonance imaging for the diagnosis of adenomyosis: correlation with histopathology. *Hum Reprod.* 2001;16(11):2427–2433. doi:10.1093/humrep/16.11.2427
6. Kolovos G, Dedes I, Imboden S, Mueller M. Adenomyosis—A call for awareness, early detection, and effective treatment strategies: a narrative review. *Healthcare.* 2024;12(16):1641.
7. Alcázar JL, Vara J, Usandizaga C, Ajossa S, Pascual MÁ, Guerriero S. Transvaginal ultrasound versus magnetic resonance imaging for diagnosing adenomyosis: a systematic review and head-to-head meta-analysis. *Int J Gynecol Obstet.* 2023;161(2):397–405. doi:10.1002/ijgo.14609
8. Venkatesh M, Awal SS. Hemiuterus with functional non-communicating horn in a young female. *Egypt J Radiol Nucl Med.* 2021;52(1):1–3. doi:10.1186/s43055-021-00507-z
9. Riggs JC, Lim EK, Liang D, Bullwinkel R. Cesarean section as a risk factor for the development of adenomyosis uteri. *J Reprod Med.* 2014;59(1–2):20–24.
10. Chen H, Wang W, Wang H, Wang X. Association between the occurrence of adenomyosis and the clinical outcomes of vaginal repair of cesarean section scar defects: an observational study. *BMC Pregnancy Childbirth.* 2022;22(1):1–11. doi:10.1186/s12884-022-04529-x
11. Timmermans M, Nisolle M, Brichant G, et al. Impact of adenomyosis and endometriosis on chronic pelvic pain after niche repair. *J Clin Med.* 2023;12(10):3484. doi:10.3390/jcm12103484
12. Rossi M, Vannuccini S, Capezzuoli T, et al. Mechanisms and pathogenesis of adenomyosis. *Curr Obstetrics Gynecol Reports.* 2022;11(2):95–102. doi:10.1007/s13669-022-00326-7
13. Khan KN, Fujishita A, Mori T. Pathogenesis of human adenomyosis: current understanding and its association with infertility. *J Clin Med.* 2022;11(14):4057. doi:10.3390/jcm11144057
14. Zhai J, Vannuccini S, Petraglia F, Giudice LC. Adenomyosis: mechanisms and pathogenesis. *Semin Reprod Med.* 2020;38(2–3):129–143. doi:10.1055/s-0040-1716687

15. Hao M, Liu X, Guo SW. Adenomyosis in mice resulting from mechanically or thermally induced endometrial–myometrial interface disruption and its possible prevention. *Reprod Biomed Online*. 2020;41(5):925–942. doi:10.1016/j.rbmo.2020.07.023
16. Ikhuorah T, Oboh D, Abramowitz C, Musheyev Y. Bicornuate uterus: a rare case of a viable full term pregnancy in the right uterine horn. *Radiol Case Rep*. 2023;18(6):2107–2111. doi:10.1016/j.radcr.2023.03.014
17. Rao Y, Chen M, Li Y. A new treatment for Robert’s uterus with adenomyosis: a case report and review of the literature. *BMC Womens Health*. 2024;24(1):1–6. doi:10.1186/s12905-024-03286-7
18. Bindra V, Reddy CA, Swetha P, Madhavi N. A case of didelphys uterus: tackling the double trouble adenomyosis and infertility. *Fertil Steril*. 2022;118(4):808–809. doi:10.1016/j.fertnstert.2022.06.027

International Medical Case Reports Journal

Publish your work in this journal

The International Medical Case Reports Journal is an international, peer-reviewed open-access journal publishing original case reports from all medical specialties. Previously unpublished medical posters are also accepted relating to any area of clinical or preclinical science. Submissions should not normally exceed 2,000 words or 4 published pages including figures, diagrams and references. The manuscript management system is completely online and includes a very quick and fair peer-review system, which is all easy to use. Visit <http://www.dovepress.com/testimonials.php> to read real quotes from published authors.

Submit your manuscript here: <https://www.dovepress.com/international-medical-case-reports-journal-journal>

Dovepress
Taylor & Francis Group