

## Case Report

### The Surgical Approach for Adenomyosis in Nulligravid Women: A Case Report

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#### Abstract

**Introduction:** Adenomyosis is a benign uterine condition characterized by endometrial tissue invasion into the myometrium. This often leading to chronic pelvic pain, dysmenorrhea, and infertility. A meta-analysis in 2025 reported prevalence ranging from 5.9% to 46.0%, with a mean of 20.7%. The Osada triple-flap technique was developed to remove adenomyotic tissue while preserving uterine function, serving as an alternative to total hysterectomy. However, intraoperative findings may require surgical adjustment.

**Case Report:** A 32-year-old woman presented with severe abnormal uterine bleeding. Married for 11 years without children, she had a history of recurrent bleeding, multiple hospitalizations, blood transfusions, and myomectomy. Ultrasound showed an enlarged, globular, and heterogenous uterus with asymmetrical thickening. An exploratory laparotomy was performed with the intention of using the Osada technique, but, due to deep cul-de-sac adhesions and suspected malignancy, the surgery proceeded to total abdominal hysterectomy.

**Conclusions:** Adenomyosis significantly affects fertility and uterus-preserving techniques like the Osada method may benefit selected patients. However, in cases with extensive adhesions, distorted anatomy, or suspected malignancy, hysterectomy remains the safest and most definitive treatment.

**Keywords:** Adenomyosis; infertile; abnormal uterine bleeding

### Laporan Kasus: Pendekatan Bedah untuk Adenomyosis pada Perempuan Nulligravida

**Pendahuluan:** Adenomyosis adalah kelainan jinak pada uterus akibat invasi endometrium ke miometrium. Kondisi ini sering menimbulkan nyeri panggul, dismenore, dan infertilitas. Meta-analisis tahun 2025 melaporkan bahwa prevalensi 5,9% hingga 46,0% (rata-rata 20,7%). Teknik Osada triple-flap dikembangkan untuk mengangkat jaringan adenomyosis sambil mempertahankan fungsi uterus, sebagai alternatif histerektomi total. Namun, temuan intra-operatif dapat mengubah rencana tindakan.

**Laporan Kasus:** Perempuan 32 tahun datang dengan perdarahan uterus abnormal berat. Menikah 11 tahun tanpa anak, riwayat perdarahan berulang, beberapa kali rawat inap, transfusi darah, dan miomektomi. Ultrasonografi menunjukkan uterus membesar dan heterogen. Laparotomi eksplorasi direncanakan dengan teknik Osada (triple-flap), namun karena adanya adhesi dalam dan kecurigaan keganasan, dilakukan histerektomi total.

**Kesimpulan:** Adenomyosis dapat mengganggu kesuburan. Metode preservasi uterus seperti metode Osada bermanfaat pada kasus terpilih, tetapi histerektomi total tetap menjadi pilihan paling aman dan efektif pada kasus kompleks dengan adhesi dan kecurigaan keganasan.

**Kata Kunci:** Adenomyosis; infertil; perdarahan uterus abnormal

## Introduction

Adenomyosis is a benign condition characterized by the presence of endometrial tissue within the myometrium, leading to symptoms such as chronic pelvic pain, dysmenorrhea, and infertility. The condition contributes to uterine enlargement and involves both lymphatic and vascular infiltration into the myometrium. The diagnosis of adenomyosis presents significant challenges and relies on a combination of clinical evaluation, imaging studies, and histopathological examination, with the diagnosis typically confirmed through hysterectomy.<sup>1,2</sup> A study in 2020 reported an incidence of 1.03% at 28.9 cases per 10,000 woman-years. The incidence peaked at 30.6 per 10,000 woman-years before gradually declining to 24.4 per 10,000 woman-years. The highest incidence was observed among women aged 41–45 years, reaching 69.1 per 10,000 woman-years.<sup>3</sup> Another study in 2020 estimated the prevalence of adenomyosis to be 0.8%.<sup>4</sup> More recently, a meta-analysis in 2025 reported that the prevalence of adenomyosis ranged from 5.9% to 46.0%, with an overall weighted mean prevalence of 20.7%.<sup>5</sup>

Infertility is a significant concern in adenomyosis, with its presence identified in 8–24% of fertile patients based on transvaginal ultrasound findings. Although the exact mechanism underlying infertility in adenomyosis remains unclear, several contributing factors have been identified. Adenomyosis disrupts the normal architecture of the myometrium, impairing uterine functions critical for implantation and conception. This disruption affects the junctional zone, resulting in abnormal uterine contractility that interferes with embryo implantation. Additionally, altered uterine peristalsis, impaired sperm transport, and changes in endometrial function and receptivity contribute to the decreased

fertility in women with adenomyosis.<sup>4,6,7</sup>

Adenomyosis often presents with nonspecific symptoms, making it difficult to distinguish from other gynecological conditions such as endometriosis or fibroids. Up to 33% of cases may be asymptomatic. Previously thought to occur only in parous women, recent advancements in imaging techniques have led to more accurate diagnoses in infertile patients. Transvaginal sonography (TVS) is currently considered the primary diagnostic tool, identifying characteristic features such as heterogeneous hypoechoic areas in the myometrium, anechoic lacunae or cysts, linear striations extending from the endometrium, poor delineation of the junctional zone, and asymmetric myometrial wall thickening. The confirmation of a diagnosis usually happens when three or more sonographic criteria are present.<sup>6,8</sup>

The surgical approach to adenomyosis is still in a state of flux. Up until now, hysterectomy remains the definitive treatment for adenomyosis; however, several uterine-sparing surgical approaches have been developed in recent decades.<sup>9</sup> Adenomyomectomy, first described by Hyams in 1952, involves excising focal adenomyosis but is technically more challenging than myomectomy due to poorly defined planes between diseased and normal myometrium. Cytoreductive surgery is another option, applicable for both focal and diffuse adenomyosis.<sup>10</sup> The Osada Technique (triple-flap method) offers a specialized approach to radical adenomyosis tissue removal while reconstructing the uterine wall.<sup>11</sup> These procedures offer viable alternatives to hysterectomy, particularly for women desiring fertility preservation. Here, we present a case of an infertile patient with adenomyosis. This report highlights a surgical Approach And Decision-Making On Managing Adenomyosis In The Context Of Infertility.

### Case Presentation

A 32-year-old woman presented with complaints of heavy vaginal bleeding for one day prior to admission. The bleeding was bright red with clots, saturating up to 10 sanitary pads in a day, and came with cramping in the lower abdomen, which intensified during menstruation or episodes of bleeding. Figure 1 illustrates the patient’s medical history. The patient has reported a history of abnormal uterine bleeding since four years ago. A few months later, she underwent a fertility treatment program and was prescribed hormonal therapy; however, the bleeding progressively worsened, prompting her to discontinue the medication. She returned to the hospital one year ago with complaints of increased bleeding requiring transfusion. Over the past year, she reported four hospital admissions for similar complaints—the most recent being seven months ago. At the time, she was prescribed combined estradiol valerate-norgestrel, which she discontinued after two days due to significant pain. She had a history of myomectomy six years ago. The patient has been married twice and

remains nulligravid and nulliparous. Her current medications include tramadol and mefenamic acid. On physical examination, the patient was found to have pale conjunctiva (+/+). The abdominal examination revealed a fixed, smooth-surfaced mass measuring approximately 15x10x15 cm, which was palpable. On pelvic examination, the cervix appeared smooth, with an enlarged uterine corpus, and the fundus had reached the umbilicus level.

Routine blood tests and abdominal ultrasound were performed for diagnosis support. The laboratory results showed severe anemia and hypokalemia (3.22 mmol/L). The ultrasound examination (Figure 2) revealed an enlarged, globular, and inhomogeneous uterus with asymmetrical thickening, measuring 9.86 x 7.49 x 9.25 cm and a calculated volume of 357.68 cm<sup>3</sup>. A hyperechoic lesion with a fan shadowing pattern, measuring 4.51 x 2.57 cm, was identified in the anterior uterine wall. The Color Doppler imaging showed no significant vascularization within the lesion.

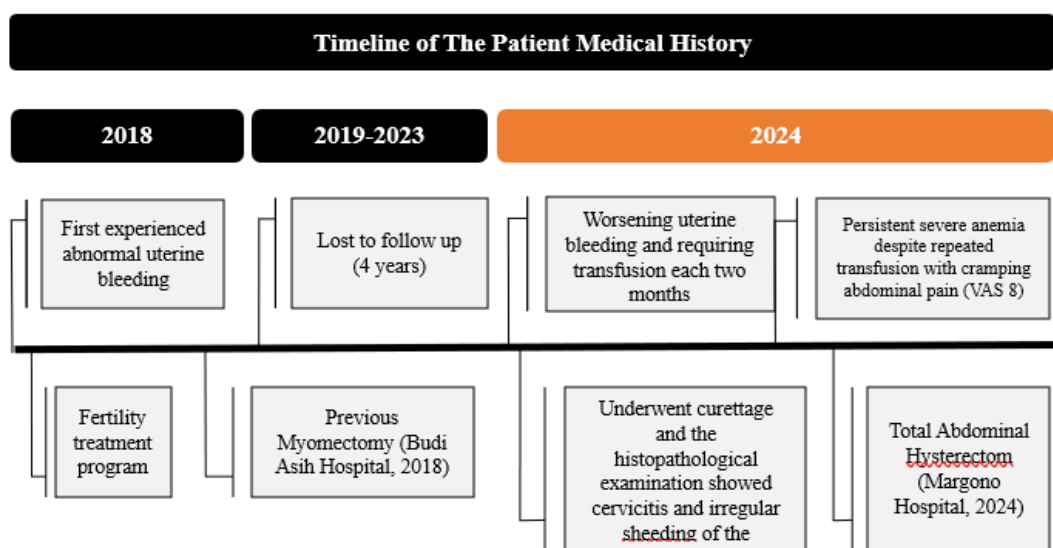
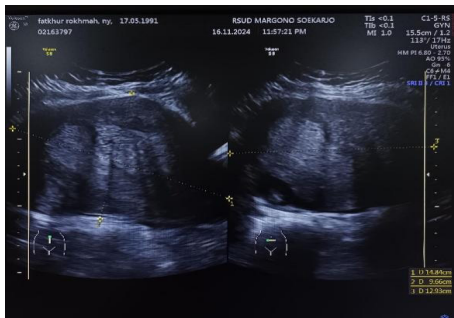


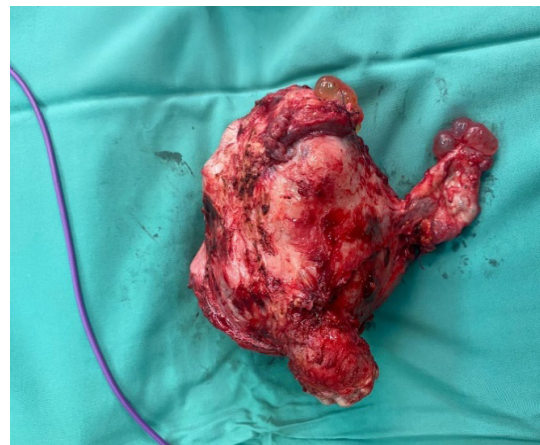
Figure 1 Patient’s Medical History



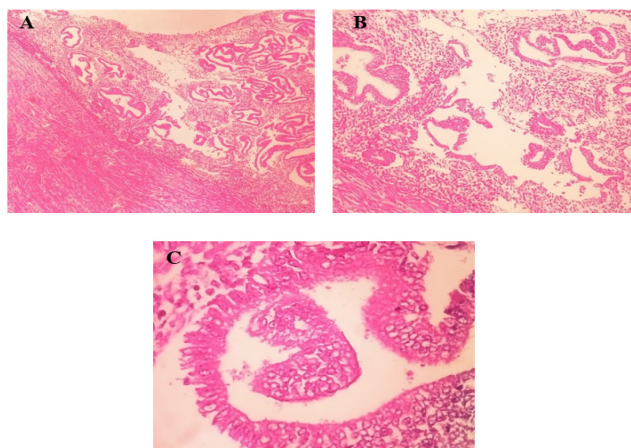
**Figure 2 Abdominal Ultrasound Examination Showing Hyperechoic Lesion with a Fan Shadowing Pattern**

The patient was diagnosed with heavy menstrual bleeding secondary to adenomyosis (AUB-A) and severe anemia. Initial management was focused on stabilizing the patient’s condition, starting with the transfusion of four units of packed red blood cells, targeting a hemoglobin level of >10 g/dL. Once stabilized, the patient was programmed to undergo an exploratory laparotomy, and the triple-flap technique was planned. During the procedure, the patient exhibited enlarged left and cystic right ovaries measuring 5x5x3 cm and 4x3x3 cm, along with uterine enlargement that was accompanied by adhesions to the urinary bladder, rectosigmoid colon, Douglas cavity,

rectum, and the omentum. An adhesiolysis has been attempted. A surgical staging was done. The procedure was concluded with total abdominal hysterectomy, based on the clinical judgment of the surgeon, as there was no clear dissection plane and suspicion of uterus malignancy. During further exploratory, enlargement of the right pelvic lymph node was found, sizing 2x1.5x1.5 cm; thus, bilateral pelvic lymphadenectomy was needed. A histopathologic analysis was ordered. The procedure was concluded in 1 hour and 20 minutes of operating time with an estimated blood loss of 700ml; no active bleeding was observed during evaluation.



**Figure 3 Gross Appearance of The Uterus**



**Figure 4 Biopsy Specimen Demonstrated Edematous and Hyperemic Bundles of Myometrium Interspersed with Island of Endometrial Tissue. (A) 4x Magnification (B) 10x Magnification (C) 40x Magnification Post-Total Hysterectomy**

## Discussion

Adenomyosis is characterized by the infiltration of endometrial tissue into the myometrium, often presenting with diffuse growth patterns and poorly defined boundaries between normal and affected tissue. Historically, the diagnosis was only confirmed at histology after hysterectomy. Recent advancements in imaging techniques have significantly improved the accuracy of detecting adenomyosis. Symptoms associated with adenomyosis include abnormal uterine bleeding, dysmenorrhea, chronic pelvic pain, and dyspareunia.<sup>12</sup> In infertile women with abnormal ultrasound findings, adenomyosis should be considered during preoperative assessment to guide appropriate management.<sup>9,13</sup> Medical treatments for adenomyosis always follow the principles of the management of endometriosis, which are usually aimed at reducing endogenous estrogen production or promoting endometrial differentiation through the use of progestins. Although, the treatment approach varies depending on the patient's age, clinical symptoms, comorbidities, and desire for future fertility. The medical management of adenomyosis consists of nonsteroidal anti-inflammatory drugs (NSAIDs) and suppressive medical therapy such as oral contraceptives, progestins, levonorgestrel-releasing intrauterine devices, and GnRH agonists. The goal of therapy is important for adenomyosis cases and can include symptom relief, and possibly increased fertility. Lesion resorption is not the therapeutic goals; lesions survive any drug at any dose, for any period of use, and come back after treatment discontinuation.<sup>14</sup> While hysterectomy and endometrial ablation are considered definitive treatments for severe adenomyosis, conservative options should be prioritized in patients seeking fertility.<sup>15,16</sup> In the case presented, the patient experienced heavy vaginal bleeding that resulted in a hemoglobin

level of 4 g/dL and reported severe lower abdominal pain (VAS 8) despite the received medication. The patient has been attempted for COC; however, her symptoms tend to worsen after 2 days of use. Thus, a surgical approach was recommended for this patient, as it is considered to be necessary in cases where symptoms are medication resistant.

The Osada Method, also known as the Triple-Flap Technique, represents an advanced surgical approach to adenomyomectomy, markedly different from conventional methods. This technique allows for the complete excision of adenomyosis tissue while preserving uterine integrity, making it suitable for women desiring future pregnancies. The procedure begins with a small transverse incision to access the peritoneal cavity. Following the exteriorization of the uterus, a rubber tourniquet is applied around the proximal cervix to minimize intraoperative bleeding. The adenomyotic tissue is meticulously excised, maintaining a 1 cm margin from both the endometrium and serosa. The endometrial cavity is opened to aid surgical guidance, enabling precise excision while avoiding injury to the fallopian tubes and adjacent structures.<sup>11,17</sup>

Following excision, the uterine wall defect is reconstructed using the triple-flap method. This procedure involves creating overlapping flaps of healthy myometrium to restore structural integrity and prevent hemorrhage. The first flap is sutured with interrupted 2-0 vicryl stitches, and the contralateral uterine wall is folded over the suture line to prevent direct alignment of suture planes. This careful reconstruction results in an outer layer composed of serosa and myometrium and an inner layer of normal myometrium and endometrium. The tourniquet is removed only after repairs are complete, as the layered flaps effectively control bleeding.<sup>11,17</sup>

Clinical studies have confirmed the

effectiveness of the Osada Method, with most patients achieving restored uterine blood flow within six months and a high rate of successful pregnancies. Among women desiring conception, 74.2% became pregnant, and 51.6% successfully delivered healthy infants through elective cesarean section, with no reported cases of uterine rupture. The observed low recurrence rate (3.5%) further supports this technique as a durable, fertility-preserving option for diffuse and nodular adenomyosis.<sup>17</sup> This technique represents a more radical approach for extensive diffuse adenomyosis, prompting the usage of this procedure for the patient in the beginning.<sup>12</sup>

Patients with adenomyosis frequently present with concurrent uterine pathologies like fibroids and endometriosis. In this case, the uterus resembled reported sizes of >1000 grams in many severe cases, making conservative excision impractical. Considering the longer operation time needed using conservative excision, especially when the patient had severe anemia and history of multiple transfusions, the complexity of the reconstructive surgery could lead to increased intraoperative blood loss. This consideration led the clinician to perform a total hysterectomy (taking into notes that both the patient and the patient's guardian already consented before the procedure).<sup>18</sup>

The presence of extensive adhesions in the cul-de-sac, including uterine, bladder, rectosigmoid, and appendical attachments, indicates a condition commonly referred to as "frozen pelvis", a scenario wherein anatomical planes are severely distorted. Such adhesions complicate minimally invasive or conservative approaches due to increased risk of injury and bleeding, particularly in areas with adherent bowels, and the inability to guarantee clear surgical margins. Multiple reports have identified that adhesions to the uterus frequently occur in most nonpregnant women in procedures performed at laparotomy and laparoscopy. The impact of

tissue injury is reflected in the occurrence of oxidative stress, generating reactive oxygen and reactive nitrogen species, which are more likely in the presence of an anaerobic state. This pathogenesis might be one of the underlying factors to the adhesion found in this patient, as a history of myomectomy was reported, although no precise report of an antiadhesion adjuvant received post previous myomectomy; thus, it remains in the unknown.

The surgical approach to adenomyosis remains in flux, as the intraoperative outcome remains uncertain. This study presents a case of severe adenomyosis to illustrate the clinical decision-making process between total hysterectomy and the fertility-preserving Osada Method. The procedure of total hysterectomy, as performed in this case, offers rapid control of bleeding and definitive treatment by removing the uterus and is appropriate for patients with severe disease or those not prioritizing fertility. In contrast, the Osada Method might provide a conservative alternative, enabling excision of diseased tissue with successful fertility outcomes and low recurrence. Given the patient's age, clinical history, and the extent of the disease, fertility preservation was not a primary concern in this setting; hence, the decision for total hysterectomy was made based on clinical considerations, treatment goals, and needs. Moving forward, for similar patients desiring to preserve fertility, the Osada Method might still be considered as a viable surgical option.

## Conclusion

This case highlights the importance of individualized surgical decision-making in the management of advanced adenomyosis with coexisting pelvic pathology, especially in patients with nulliparity and nulligravid background. Although uterine-preserving techniques such as the Osada triple-flap

method are beneficial for selected patients with focal disease and a desire for fertility, they are contraindicated in cases with extensive adhesions, distorted anatomy, and suspicion of malignancy. The total abdominal hysterectomy allowed for effective removal, hemostatic control, and proper oncologic assessment if needed. Despite the evolution of conservative options, total hysterectomy continues to be regarded as the safest and most effective treatment for complex cases of adenomyosis, as well as those with suspected malignancy.

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