

# Conservative Fertility-preserving Surgery in a Woman with Huge Adenomyosis using Triple Flap Technique and Mirena: A Case Report

Azam Tarafdari<sup>1</sup>, Fatemeh Keikha<sup>1</sup>, Mahrouz Malek<sup>2</sup>, Azin Ghamari<sup>3</sup>

1. Department of Obstetrics and Gynecology, Imam Khomeini Hospital Complex, Tehran University of Medical Sciences, Tehran, Iran
2. Department of Radiology, Medical Imaging Center, Imam Khomeini Hospital Complex, Tehran University of Medical Sciences, Tehran, Iran
3. Growth and Development Research Center, Children's Medical Center, Tehran University of Medical Sciences, Tehran, Iran



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## Corresponding Information:

Azam Tarafdari,

Department of Obstetrics and Gynecology, Imam Khomeini Hospital Complex, Tehran University of Medical Sciences, Tehran, Iran

Email: tarafdari@sina.tums.ac.ir

## ABSTRACT

**Background and Objective;** Being considered a common benign uterine disorder, Adenomyosis (AD) is defined as the presence of an endometrial gland and stroma within the myometrium. It causes symptoms such as dysmenorrhea, menorrhagia, chronic pelvic pain, and infertility. It has a step-by-step treatment, initiating with medical therapy, conservative surgery, and finally, the hysterectomy. As some of the women with adenomyosis are nulligravid and want to become pregnant later, most patients prefer fertility-preserving surgery.

**Case Report:** This study describes the management of diffuse posterior AD in a 36-year-old virgin female by triple flap technique and levonorgestrel intrauterine device (LNG-IUD) insertion during operation to prevent future recurrence and reduction of patient's symptoms. The application of the triple flap technique and LNG-IUD in this patient was associated with excellent results.

**Conclusion:** The wide excision of the lesion leads to symptom improvement, especially hypermenorrhea and dysmenorrhea; besides, the reconstruction of the uterine wall with this technique increases the risk of the possibility of future pregnancy for our patient.

**Keywords:** Adenomyomectomy, Levonorgestrel intrauterine device, Triple flap technique



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## Introduction

Being considered as a common benign uterine disorder, Adenomyosis (AD) is defined as the presence of endometrial gland and stroma within the myometrium in more than 2.5 mm of depth, which is surrounded by reactive fibrosis, hyperplastic and hypertrophic smooth muscle (1). AD is mostly diagnosed in multiparous women in their 30s or 40s; although, it can occur in nulligravid women, and for cultural and emotional issues, these women are unwilling to hysterectomy. Because it is mostly determined by pathologic examination after hysterectomy, the prevalence of AD is different, ranging between 10% to 66% (2). Based on histological evaluations, AD is classified into the localized (focal) or diffuse; when the whole myometrium or more than half of it, is involved. Its main symptoms include secondary progressive

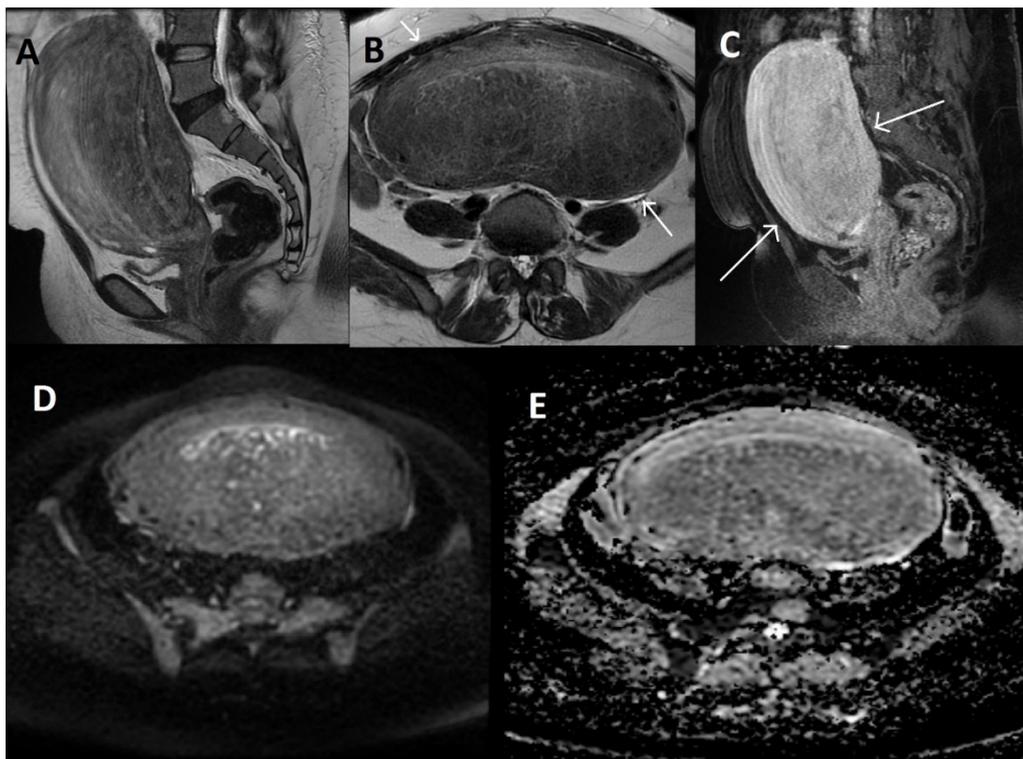
dysmenorrhea, menorrhagia, chronic pelvic pain, and infertility (3). Management of AD comprises long-term hormonal therapy, intrauterine device, gonadotropin-releasing hormone (GnRH) agonists, analgesic, and finally, hysterectomy (4). Hysterectomy is the most common choice, especially in those women with complete family planning; however, the patients' desire for future fertility makes us perform conservative surgeries, which results in uterine preservation. This study aims to describe the management of diffuse AD in a 36-year-old virgin female by triple flap technique and levonorgestrel intrauterine device (LNG-IUD) insertion.

## Case presentation

A 36-year-old virgin female was referred to Imam Khomeini hospital complex in 2020, due to heavy and intermittent vaginal bleeding ten months before our visit. Her past medical history consisted of diabetes mellitus and hypertension, using Amlodipine, Losartan, Metformin, and Glibenclamide. She received oral contraceptive pills, progesterone, and five months of GnRH agonist for the symptoms, which were not useful, and the symptoms were refractory to the conservative medication. Hysterectomy was recommended due to abnormal bleeding unresponsive to medical management, but since she was virgin and nulligravid and willing to future pregnancy, she was referred to our institution for uterine-preserving conservative surgery.

On the physical examination, uterine mass, equivalent to 20-22 weeks of gestation, was obvious. The laboratory tests showed a hemoglobin level of 6.6 g/dL and a platelet level of  $444 \times 10^3$  /mCL. On the abdominal ultrasound study, the uterine measured

200\*180 mm with an endometrial thickness of 4mm, and a posterior uterine mass without obscure boundary was noted. The magnetic resonance imaging (MRI) was in favor of the diffuse posterior AD. MRI was performed with a pelvic coil in 3 tesla MR scanner (GE.healthcare SIGNA) the patient had fasting for about 6 hours intramuscular injection of hyoscine was done 15 minutes before MRI. Routine pelvic MRI protocol (sagittal axial and coronal T2 and axial T1 Images) DWI with b value 0.400,800 and post-contrast images in three plans was done. T2 weighted images showed diffuse enlargement of posterior myometrium of the uterus (low T2 with small bright foci), which appears inhomogeneous on both T1w and T2w images without discrete mass that represents the typical appearance of diffuse adenomyosis and endometrium was thin ([Figure 1a, b](#)). On post-contrast T1 FS images, diffuse heterogeneous enhancement was noted without any separate mass ([Figure 1c](#)). Axial Diffusion-weighted imaging at  $b = 800$  and ADC map showed no restriction. The ADC value was  $1.29 \times 10^{-3}$  mm<sup>2</sup>/s ([Figure 1d, e](#)). Both adnexa, cervix, and other pelvic organs were normal on MRI.



**Figure 1.** Pelvic MRI in a 36 year-old female, showing abnormally enlarged uterus a) sagittal T2 weighted fast spin-echo Images and b) axial T2 weighted Images shows uterus is diffusely enlarged with the fundus at the level of L4 vertebral body. The junctional zone is thick, containing multiple small high signal foci typical for diffuse Adenomyosis. Endometrial thickness was 3mm without intracavitary mass or distortion. c) T1 weighted fat-suppressed images show diffuse heterogeneous enhancement. d and e) DWI and ADC map shows no restriction.

Adenomyomectomy with triple flap technique was performed, as previously described by Osada in 2011 (5). The surgery was performed following the transverse suprapubic incision and placing the rubber

tube around the proximal od cervix. The AD was visible after bisection of the uterus from the serosa surface of the fundus in midline and in the sagittal plane and the AD tissue was resected from the

surrounding myometrium by grasping by Martin Forceps. The uterus was then reconstructed by the triple flap technique. An LNG-IUD (Mirena, Bayer Co., Germany) was applied intraoperatively in uterine cavity for the prevention of future AD recurrence. She was administered with four units of packed red blood cells intraoperatively. During the postoperative period, no signs and symptoms of fever and hemorrhage were noted. In the 6-month follow-up visit, the uterine size was decreased significantly (the uterine size was equivalent to 12 weeks of gestation), the hemoglobin level was alleviated (hemoglobin level of 12.5 g/dL), and menstrual cycles was normal without any abnormal bleeding.

## Discussion

The treatment of AD is a step-by-step strategy, starting with conservative management of symptoms with medication, followed by conservative surgery, and finally, hysterectomy. The main limitation of the medication is the potential recurrence of the symptoms, as the medications cause the AD regress and not eradicate. In severe cases refractory to medical treatment, conservative surgery has been considered, especially in patients who want to preserve fertility. The goal of conservative surgery is to restore normal anatomy and physiology of the uterus, preserve fertility, eliminate severe symptoms, and improve the patient's quality of life. The triple flap technique was first described by Osada in 2011 (5); this technique was performed on 104 patients with high efficacy in symptom elimination, fertility preservation, and low rate of relapse (5). Few studies have been done to evaluate the efficacy of this technique in patients with diffuse AD. Another study on 113 women operated with this technique indicated that the blood flow becomes normal within six months, and from 62 women who became pregnant, 32 normal babies were delivered by cesarean section. No uterine rupture was reported, and recurrence of AD occurred for four patients (6). This technique was applied by Kim *et al.* for a 44-year-old woman with huge AD and pelvic adhesion (7). In 2020, Kaplunov *et al.* used this technique for 41 women with diffuse AD (8); AD was associated with hystero myoma in 51.2% of patients. They concluded that this technique is a good alternative technique for hysterectomy in these patients (8). Similar to our patient, Tskhay *et al.* in 2019, this technique is studied in 26 women with AD; they reported that in the 18-months follow-up period, the menstruation became normal and the AD signs and symptoms disappeared in all patients, three patients became pregnant spontaneously, and two of them delivered full-term babies following the cesarean section (9); in contrast, our patient had no delivery post-operatively because she was virgin. Wiweko *et al.* in 2016, studied 40 women with AD treated by AD resection (with/without Osada's technique) and who were underwent hysterectomy (10). They concluded that conservative surgery is efficient for symptom relief and

preserving fertility (10). Several operative and postoperative complications could be considered for this technique; as AD has an unclear border with normal myometrium, the small vessels in the mass could result in remarkable hemorrhage, which leads to blood transfusion intra and postoperatively and even converts to hysterectomy, in our patient severe hemorrhage did not occur; four units of packed red blood cells were transfused; additionally, according to the extent of the lesion, uterine rupture in the following pregnancy could occur. However, as seen in our patient, the flap techniques did not cause extra morbidities, including the occurrence of the hematoma, dehiscence of the uterine scar, or adhesions (11). The LNG-IUD releases progesterone into the uterus, which is effective for five years; it downregulates the uterine estrogen and progesterone receptors, which brings about the endometrial proliferative antagonism (3). This could help to alleviate the symptoms in women with AD and prevent AD relapse in the future.

## Conclusion

The application of the triple flap technique and LNG-IUD in this patient was associated with excellent results; the wide excision of the lesion leads to symptom improvement, especially hypermenorrhea and dysmenorrhea; besides, the reconstruction of the uterine wall with this technique increases the risk of the possibility of future pregnancy for our patient. Besides, complications such as hemorrhage did not occur. For the prevention of future AD recurrence, LNG-IUD was administered. The 6-months follow-up period was satisfying, the size of the uterine was decreased, and the symptoms were disappeared

## Acknowledgments

None

## Conflict of Interest

The authors have no conflicts of interest relevant to this article

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