



ADENOMYOMECTOMY IN A YOUNG FEMALE WITH ADENOMYOSIS WITH FAILED MEDICAL MANAGEMENT: A CASE REPORT

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ABSTRACT

Adenomyosis is a benign gynaecological condition characterised by the presence of endometrial glands and stroma within the myometrium at least 1 high power field (2.5mm) from the basal layer of the endometrium. Adenomyosis is usually seen in parous women in 4th -5th decade. Women with adenomyosis may be asymptomatic or present with symptoms of heavy menstrual bleeding (HMB), dysmenorrhea, dyspareunia and subfertility. In severe cases it has an impact on not only physical health but deteriorates the mental health and quality of life of the patient. Various treatment modalities are available for the disease. I report a case of 32 year nulligravida with HMB and dysmenorrhea. Careful history taking, examination and imaging revealed uterine adenomyosis. Initially patient was started on Gonadotropin releasing hormone (GnRH) agonist therapy for 6 months but the symptoms were not relieved. Adenomyomectomy using triple flap procedure was then performed. Patient was followed up till 1 year post surgery and showed improvement in symptoms. The aim of this case report is to provide an overview of various treatment options that can be employed in the effective management of adenomyosis on a case to case basis

KEYWORDS

adenomyosis, heavy menstrual bleeding, adenomyomectomy, triple flap procedure.

INTRODUCTION

Adenomyosis also called as 'endometriosis externa' is a benign gynaecological condition which affects 1% of the female population (1). The exact etiology and pathogenic mechanism of adenomyosis is not known. The most widely accepted theory suggests the downward invagination of endometrial basal layer into the endometrium as submucosa is absent at endometrial myometrial interface (2). Other theories include de novo development of adenomyosis from embryological misplaced pluripotent müllerian remnant, invagination of the basal layer proceeding along the intramyometrial lymphatic system, or origination from bone marrow stem cells that are displaced through the vasculature (3). It is classified into diffuse or focal adenomyosis depending upon the location, extent of the disease and histological features (4). The treatment is instituted taking into consideration factors like age, severity of the disease and whether or not the patient is desirous of future child bearing. If the severity of symptoms is less medical management with non steroidal anti inflammatory drugs (NSAID's) and hormones (combined oral contraceptive pills, progestogens, GnRH agonists, levonorgestrel intrauterine system and aromatase inhibitors) is done to relieve symptoms. However, if symptoms are not relieved by medical management then hysterectomy is the mainstay of treatment. Although women who have not completed child bearing conservative surgical management in the form of adenomyomectomy remains the preferred treatment modality.

Case Report

A 32 year nulligravida presented to the outpatient department of a tertiary care hospital with the chief complaint of heavy menstrual bleeding and congestive dysmenorrhea for the past 2 years. Her previous menstrual cycles lasted for 3-4 days with regular moderate flow with a cycle length of 28±2 days. For the past 2 years she was experiencing HMB along with passage of clots that lasted for 5-6 days with history of change of 3 pads/day. On examination her blood pressure was 114/76mm of Hg and Pulse Rate was 80/min. Her BMI was 27.5 (overweight). Her per abdominal examination was normal. On per speculum examination, cervix and vagina were healthy. On per vaginal examination uterus was enlarged~10-12 weeks size, globular, regular in contour, non-tender, firm, mobile, adnexa was nil palpable. Ultrasonographic findings showed enlarged uterus ~10*6.5*4.5cm with intramyometrial cysts and irregularly defined endomyometrial junctional zone suggestive of adenomyosis. Since the patient was young nulligravida and wanted to preserve fertility medical management was initially started with mafenamic acid 500mg thrice daily for 5 days during menstruation. After 3 months of treatment patient was again evaluated but there was no improvement in symptoms. As the patient was trying to conceive combined oral contraceptives could not be given. So she started on intramuscular injection of leuprolide acetate (GnRH agonist) 3.75 mg/ monthly for 6 months. She again started having the same complaints 3 months after stopping the injections. Patient was admitted in the hospital in view of severe anaemia (Hb-6g%) and was transfused 3 units of whole blood. After having failed attempts with medical management, patient was

planned for adenomyomectomy (triple flap procedure). The procedure consisted of radical excision of adenomyotic tissue with subsequent triple-flap reconstruction of the uterus. The enlarged uterus was bisected in the midline from the fundus till the upper border of cervix all the way down till the endometrial cavity was reached. All the adenomyotic tissue was excised from the surrounding myometrium (figure 1a) leaving 1cm margin of tissue above the endometrium and a 1 cm margin of tissue below the serosal surface. The endometrial lining was then reconstructed using 3-0 vicryl suture (figure 1b). On one side of the bisected uterus the myometrium and serosa was reconstructed in anteroposterior plane using multiple interrupted 2-0 vicryl suture (figure 1c). Then the contralateral side was brought over the reconstructed first side to cover the seromuscular suture line (figure 1d). Complete haemostasis was achieved. She was discharged on 4th postoperative day in stable condition and advised to follow up after 6 weeks. She was advised not to conceive for 1 year and was counselled regarding the risk of uterine rupture in subsequent pregnancy as well as the importance of early booking and regular antenatal natal visits. On follow up the patient reported significant improvement in her symptoms.

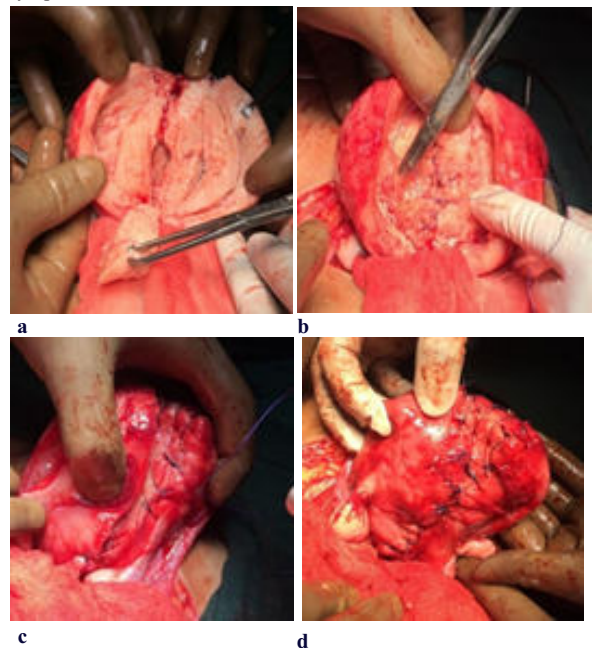


FIGURE 1:
a) excision of adenomyotic tissue b) endometrial reconstruction c) reconstruction of one side of bisected uterus in anteroposterior plane d) contralateral side brought over the reconstructed first side.

DISCUSSION

Adenomyosis is a benign uterine disorder usually seen in parous women in 4th-5th decade. Over the last decade due to advancement in imaging modalities it is now also seen in young reproductive age women (5). This case also reports its presence in young 32 year nulligravida patient. The physio-pathological mechanisms, involving sex steroid hormone aberrations, inflammation, fibrosis and neuroangiogenesis, are not fully understood (6). Women may present with infertility/subfertility, HMB, dysmenorrhea, dyspareunia although one third of the women remain asymptomatic (7). The chief complaints of the patient in the present case were HMB and dysmenorrhea. Trans vaginal ultrasonography is the first line imaging technique for the diagnosis as it is readily available, inexpensive with a sensitivity of 84% and specificity of 64% (8). Recently a standardised reporting system of ultrasound findings has been developed for the diagnosis of adenomyosis called as the MUSA (Morphological Uterus Sonographic Assessment) criteria. It includes 8 criteria and the diagnosis is made if ≥ 2 criteria are fulfilled (9). The following are the criteria: a) asymmetrical thickening of uterine walls, b) intramyometrial cysts c) hyperechoic islands d) fan-shaped shadowing of the myometrium, e) myometrial echogenic subendometrial lines and buds f) translesional vascularity g) irregular junctional zone h) interrupted junctional zone. In this case there was presence of intramyometrial cysts and irregular junctional zone. Adenomyosis has a negative impact on women's quality of life in a high percentage of cases because of AUB and pain requiring a lifelong management plan through medical or surgical treatment (10). As far as the treatment is concerned various medical hormonal and non-hormonal drugs are used off label to provide symptomatic management. GnRHa have been used for the treatment of adenomyoma related diseases, resulting in a reduction in uterine size and symptomatic improvement. However, this effect is often transient and most patients will experience a recurrence of their symptoms (11). We had the same experience with GnRH agonist therapy in the present case when the symptoms reappeared 3 months after stopping the treatment. If medical methods fail to provide relief then surgical methods are chosen. Women desirous of future childbearing may opt for conservative surgeries. Conservative surgical options include endometrial ablation, hysteroscopic endometrial and adenomyoma resection, adenomyectomy, high-intensity focused ultrasonography and uterine artery embolization. However, robust evidence supporting conservative surgical treatments of adenomyosis is still lacking (12). Adenomyectomy has been reported to improve symptoms. The rationale for adenomyectomy includes that of cytoreduction, debulking to relieve the mechanical disturbances, and correcting physiological disturbances that may impair sperm transport (13). Focal adenomyosis lesion can be treated laparoscopically. However, diffuse adenomyosis must be treated by laparotomy. Since in this patient diffuse adenomyosis was present decision for laparotomy was taken. Various surgical methods of adenomyectomy have come up since the first case report by Hyama in 1952 (14). In this patient we chose triple flap method because it is based on a completely new idea that differs from standard surgical methods. The method involves reconstruction of a defect in the uterine wall using the remaining normal uterine muscle. This method has better reproductive outcome and lower risk of uterine rupture in subsequent pregnancy (15). In the present day scenario since the disease burden due to adenomyosis has increased significantly this case report highlights the various aspects in its management and treatment.

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