Combination of hysteroscopic resection of endomyometrium with insertion of Mirena: An alternative treatment for adenomyosis

Akmal Zulayla Mohd Zahid, BM, Noorneza Abdul Rahman, MBBS, Zatul Akmar Ahmad, MBBS

Department of Obstetrics and Gynaecology; Faculty of Medicine, Universiti Teknologi MARA, Sg Buloh Campus, 47000 Sg Buloh, Selangor, Malaysia

SUMMARY
Adenomyosis is a common gynaecological condition encountered in clinical practice. Treatment of adenomyosis can present a great challenge to gynaecologists as women often become resistant to hormonal treatment eventually needing surgical intervention. Hysterectomy has long been the definitive treatment for adenomyosis. However, with women currently being diagnosed at an earlier age and still have not completed their family, there is an increasing demand for effective intervention with uterine conservation. We report here two cases of patients who had undergone a combination of hysteroscopic resection of the endomyometrium combined with Mirena insertion with successful outcome.

INTRODUCTION
Adenomyosis is a common gynaecological condition encountered in clinical practice. It is characterized by endometrial glands and endometrial stroma and a variable degree of smooth muscle hyperplasia within the myometrium. Adenomyosis can be classified based on the degree of invasion of the disease: 1) diffuse adenomyosis, in which the foci of ectopic endometrial mucosa are scattered throughout the uterine musculature, 2) focal adenomyosis, in which the affected area is markedly restricted and embedded within the myometrium, and 3) exomyometrial types, which can take the forms of polypoid adenomyomas, adenomyomas of the endocervical type, and retroperitoneal adenomyomas. Patients can be asymptomatic or present with abnormal uterine bleeding, dysmenorrhoea, chronic pelvic pain and subfertility. Historically, diagnosis of adenomyosis is usually obtained from histological examination of specimens following hysterectomy or excision of the adenomyosis (adenomyomectomy). However, the advancement of transvaginal ultrasound scan (TVS) and magnetic resonance imaging (MRI), has improved its diagnosis pre-operatively.

Treatment of adenomyosis can be challenging especially in patients who wish to preserve their uterus. A step-up strategy is often used, starting with conservative treatment with hormonal or non-hormonal medication followed by uterine-sparing surgical techniques (surgical removal of adenomyotic tissue) and hysterectomy in older women with a resistant disease or those who have completed their family. However, not all women are willing to have hysterectomy despite their older age and having completed their family.

Mirena or levonorgestrel-releasing intrauterine device is an effective treatment for adenomyosis, reducing menstrual bleeding and relieving dysmenorrhoea. Özdeğimenci et al demonstrated significant and comparable improvements in haemoglobin levels in adenomyosis-related menorrhagia and also showed superior effects on patients psychological and social life when comparing Mirena to hysterectomy. Maia et al established that the use of Mirena following endometrial resection in patients with adenomyosis was associated with higher amenorrhoea rates and lower rates of dysmenorrhoea than the group of patients who had Mirena inserted alone. We report two cases where the combination of hysteroscopic surgery and insertion of Mirena were used for treatment of adenomyosis successfully.

CASE REPORT
Case 1
A 48-year-old para 2, complained of 3 years’ history of worsening prolonged heavy menstrual bleeding of up to 10 days and severe dysmenorrhoea. She took mefenamic acid and tranexamic acid for a year, then continuous Norethisterone for 2 years. Despite the hormonal treatment, she still developed recurrent anaemia requiring prolonged iron supplementation. Bimanual examination revealed a 12-week sized uterus which felt bulky anteriorly. Ultrasound scan (USS) pelvis revealed 4x5cm adenomyosis in the anterior wall of uterus distorting the uterine cavity. Magnetic Resonance Imaging (MRI) of her pelvis confirmed the USS finding. She opted for the insertion of Mirena, which was inserted easily in the outpatient clinic. The Mirena spontaneously expelled a week after. She was not keen on hysterectomy despite completing her family. Decision was made to perform hysteroscopic resection of endometrium with reinsertion of Mirena. The patient was given one dose of Lucrin 3.75mg 1 month before the procedure.

The procedure was carried out using the bipolar resectoscope (Olympus Inc., Germany) equipped with a 3-mm deep and 5-mm wide loop (Olympus Inc., Germany) with isotonic saline as distension media under general anaesthesia. The cervix
Case Report

was dilated to 8mm to allow the introduction of the bipolar resectoscope. A large focal adenomyoma was found occupying the entire surface of the anterior uterine wall protruding into the uterine cavity. The endometrium over the posterior uterine wall was normal in appearance, and the uterine cavity length measurement was 10cm. The focal anterior adenomyosis was resected using the loop diathermy. The rest of the endometrium was ablated using rollerball diathermy, and Mirena was inserted. Histopathology result revealed endometrium in the proliferative phase and confirmed the presence of adenomyosis. Six months following the procedure, she reported 5 days of light bleeding during menstruation with no dysmenorrhoea symptoms.

Case 2

A 44-year-old para 4, complained of prolonged heavy menses lasting of up to 14 days with no dysmenorrhea. The patient had no palpable masses per abdomen. Bimanual examination revealed an anteverted uterus at 10 weeks size. The transvaginal scan revealed a retroverted bulky uterus with focal posterior adenomyosis of 2.3 cm x 2.4 cm. Endometrium was not distorted. Pipelle sampling showed a secretory phase. She opted for insertion of Mirena, but it was removed after one week as the Mirena was displaced into the endocervical canal causing discomfort. Outpatient hysteroscopy was performed at the same time which revealed limited uterine distension most likely due to the protrusion of adenomyosis into the cavity. She was given Depo Provera injection temporarily to control her symptoms but started having prolonged heavy menses again following discontinuation of the treatment. As a result, she developed anaemia with haemoglobin of 7.5 g/dl and was given parenteral iron treatment. She was offered the option of hysteroscopic resection of endometrium or total laparoscopic hysterectomy. She opted for the former and was given 1 dose of subcutaneous Lucrin 3.75mg 1 month before the operation.

The procedure was carried out using the bipolar resectoscope (Olympus Inc., Germany) with isotonic saline as distension media under general anaesthesia. The uterine cavity appeared small initially due to mucosal fold elevation/protrusion of the posterior wall into the uterus due to the adenomyosis. During the resection process, there was scattered cluster of glands and locules of bleeding areas seen within the myometrium. The base was ablated with the roller ball to achieve haemostasis. At the end of the procedure, the uterine cavity was restored, and the Mirena was inserted. The procedure was uncomplicated, and she was discharged home the next day. Histopathological findings of the specimen were consistent with adenomyosis. She was very satisfied with the result, and at 6 months follow-up appointment, and she was having light, regular menses lasting for 7 days.

DISCUSSION

Treatment of adenomyosis can present as a significant challenge to gynaecologists. Patients often become resistant to non-hormonal and hormonal medication after a while. Those with resistant adenomyosis will usually end up with hysterectomy once they get older or when they have completed their family. However, uterine-sparing surgery is becoming more popular as patient demographics are getting younger and with increasing wishes to preserve their uterus. Uterine-sparing surgery aims to control symptoms of adenomyosis and offer an optimal uterine environment for conception and pregnancy. The latter is not really an issue for both our cases as they have long completed their family. However, they still wished to avoid major surgery like hysterectomy unless necessary.

Uterine-sparing surgery includes complete or partial excision of the adenomyotic lesion. Complete excision of adenomyosis or adenomyomectomy is typically performed for focal adenomyosis. Partial excision or cytoreductive surgery is usually associated with diffuse adenomyosis, where attempting to remove all adenomyotic tissue can lead to “functional hysterectomy” and increased complication. A systematic review and meta-analysis by Mikos et al in 2019 found that complete excision of adenomyosis was associated with improvement of pain, menorrhagia, and reduction of uterine volume by a factor of 6.2, 2.3, 9, and 2.3 respectively. Partial excision of adenomyosis was associated with improvement in pain, menorrhagia, and uterine volume reduction by a factor of 5.9, 3.0, and 2.9, respectively. Most of the uterine-sparing surgery are performed via laparotomy or laparoscopy. There are very few publications on uterine-sparing surgery by hysteroscopy.

Xia et al in 2017 performed hysteroscopic excision of endometriometrial lesion in 51 women and demonstrated significant improvement in menorrhagia and dysmenorrhoea with low recurrence rate during the 2-year follow-up. Out of 51 women, 7 (14%) required another surgery due to unsatisfactory outcome. All 7 of these patients were diagnosed with diffuse-type adenomyosis involving all layers of the myometrium. Thus a complete resection of the deep ectopic myometrium is difficult without significant risk of perforation or damaging the myometrium integrity. Therefore, hysteroscopic excision of endometriometrial is probably more suitable for focal adenomyosis near the inner muscular layer of the myometrium.

Maia et al performed endometrial resection in 95 women with adenomyosis and randomised the women into two groups, the first group had Mirena inserted immediately following the procedure and the control group received no further treatment. The Mirena group had a significantly higher amenorrhoea and no dysmenorrhoea rates compared to the control group. None of the women in the Mirena group required a second surgical procedure to control persistent uterine bleeding and pain compared to 19% of the women in the control group. This was explained by the possibility of regeneration of the remnants of the deeply embedded glands within myometrium which was not removed during resection. The presence of Mirena within the uterine cavity releases continuous progesterone which suppresses of endometrial proliferation. The other advantage of the combination procedure is that the deeply embedded endometrial glands within the myometrium theoretically are not exposed to high levels of levonorgestrel secreted by the Mirena within the uterine cavity, especially when the endometrium is intact. Resecting the endometrium will
A n alternative treatment for adenomyosis

potentially remove the barrier to diffusion of progestin into underlying myometrium. It will deliver higher doses of levonorgestrel directly to the ectopic glands deeply embedded the myometrium. Therefore, combining the endomyometrial resection and Mirena insertion will provide an effective treatment for adenomyosis as demonstrated in both of our cases.

Both our patients opted to have Mirena insertion for their treatment of adenomyosis. Unfortunately, both had problems with expulsion and displacement of the Mirena. For the first case, initial scan at the clinic clearly demonstrated protrusion of the adenomyosis which distorted the uterine cavity. For the second case, initial scan at the clinic demonstrated no distortion of the endometrial cavity by the focal adenomyosis. However, outpatient hysteroscopy performed revealed a smaller uterine cavity which did not distend well possibly due to the mucosal elevation or protrusion from the adenomyosis. Therefore, patients having problem with Mirena expulsion or displacement in the presence of adenomyosis, should have hysteroscopy performed to investigate the uterine cavity. Both our patients were keen to preserve their uterus therefore, hysteroscopic surgery was performed with the aim to reduce the protrusion of adenomyotic tissue into the uterine cavity which will reduce uterine contractility and minimize the risk of Mirena expulsion.

We did not perform the procedures under ultrasound guidance as we were not aiming for complete excision of the adenomyosis. However, for patients refusing to have Mirena insertion following endometrial resection, the procedure can be done under ultrasound guidance to ensure safe removal of as much as adenomyotic tissue as possible. Maia et al reported that all their patients with adenomyosis who received Mirena after endometrial resection developed amenorrhoea. This was not demonstrated in both of our cases as we only resected the affected part of the endomyometrium and not the whole endometrium as was described in their study.

CONCLUSION

Hysteroscopic resection of focal superficial adenomyosis can be offered for cases following Mirena expulsion such as demonstrated in both our cases. Hysteroscopic resection is a much less invasive procedure compared to hysterectomy. Combining hysteroscopic surgery with insertion of Mirena will give significantly better outcomes in terms of reduction of menstrual bleeding, dysmenorrhoea and lower hysterectomy rate. Therefore, it can also be routinely offered to patients with adenomyosis, particularly the diffuse type. Hysteroscopic resection with Mirena insertion should be considered for patients with a strong desire to preserve their uterus.

CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

REFERENCES