

Diffuse Uterine Leiomyomatosis: A Case Report and Review of the Literature

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Abstract: *Background:* Diffuse uterine leiomyomatosis is a benign disease with a low incidence rate, mainly manifested as menorrhagia and infertility. Due to its clinical manifestations similar to multiple uterine myomas or adenomyosis, it is often misdiagnosed in clinical practice. Research has found that they can be distinguished through histopathological examination. According to existing reported cases, Diffuse uterine leiomyomatosis mainly occurs during the child-bearing period and has a significant impact on fertility. Hysterectomy is recognized as a radical cure, but uterine-sparing treatment methods need to be continuously explored. *Case:* A 41-year-old woman presented with recurrent menorrhagia 5 years after myomectomy. B-ultrasound examination indicated that the uterus was enlarged and the myometrium was full of hypoechoic. Considering the patient's desire to preserve the uterus, we performed laparotomy myomectomy again. *Result:* Hysteroscopic myomectomy has been shown to have a certain therapeutic effect on infertile patients in the early stage of DUL. Laparotomy myomectomy can remove a wider range of lesions and can be combined with gonadotropin releasing hormone agonists or antagonists for pretreatment. Uterine artery embolization is also a commonly administered therapy. High-intensity focused ultrasound ablation has been used with significant success in reducing uterine volume but requires further evaluation for its impact on fertility. *Conclusion:* A DUL patient underwent extensive myomectomy. This study further discussed uterine-sparing treatments, and reviewed existing research reports.

Keywords: Diffuse Uterine Leiomyomatosis, Laparotomy Myomectomy, Uterine-Sparing Treatments

1. Introduction

Diffuse uterine leiomyomatosis (DUL) is a benign disease which primarily manifests as numerous muscle nodules 0.5–3 cm in diameter diffusely growing in the myometrium [1]. Its clinical manifestations include menorrhagia, anemia, lower abdominal pain, dysmenorrhea, infertility, and pressure symptoms. Murray and Glynn were the first to discover and report this disease. It was named Diffuse uterine leiomyomatosis in 1979 [2-3]. Because the symptoms of DUL are similar to multiple uterine myomas or adenomyosis, this disease is often misdiagnosed before surgery and usually requires further clarification of the diagnosis through histopathological examination [4]. With the increase in the case reports of DUL, the awareness of the disease has gradually deepened, and many different management have

been generated. Currently, research suggests that the main method for treating DUL is hysterectomy [5]. Typical myomectomy is not sufficient to treat DUL, and extensive myomectomy is required to achieve satisfactory treatment results. But this surgery increases the probability of perinatal complications such as invasive small intestinal obstruction [6]. Due to the frequent occurrence of DUL in women of childbearing age, the preservation of fertility has become an increasingly important aspect in formulating treatment plans. In this study, We reported a DUL patient who underwent two surgical treatments within 6 years, and reviewed the current management of DUL.

2. Case Report

A 41-year-old woman, G2P1, presented to our hospital in May 2022 with menorrhagia, pelvic compression symptoms,

and anemia for 1 year. She had undergone myomectomy in our hospital in 2016. Pelvic examination revealed a 12-week-size uterus. Transvaginal ultrasonography showed that the size of the uterus was about 9.5*8.9*9.0cm, and the myometrium was almost full of hypoechoic lesions, the larger of which was 4.5*4.4cm, and the endometrial thickness was about 0.6cm (Figure 1). After improving anemia, the patient underwent surgery on May 20, 2022. To assess the intrauterine condition, hysteroscopy was first used to perform intrauterine examination. Under hysteroscopy, the uterine cavity was large, the anterior and posterior walls were almost covered with protrusions of different sizes, the endometrium was smooth, and the opening of the fallopian tubes was visible on both sides. After entering the abdominal cavity, the uterus

was found to be enlarged, with an irregular shape. Dozens of intramural fibroids, 0.5-4.0 cm in diameter, and about 20 uterine submucosal fibroids of varying sizes were identified (Figure 2). The appearance of bilateral ovaries and fallopian tubes was obviously normal. After blunt dissection and complete removal of intramural fibroids and submucosal fibroids, the uterine cavity was first closed by continuous suturing, and then the seromuscular layer of uterus was sutured in the same way. The operation carried out smoothly and the intraoperative blood loss was 300ml. Postoperative pathology revealed leiomyoma with degeneration and necrosis, and focal rich cells (Figure 3A, 3B). The patient has now been discharged and is scheduled to undergo further gonadotropin releasing hormone antagonist (GnRHa) therapy.

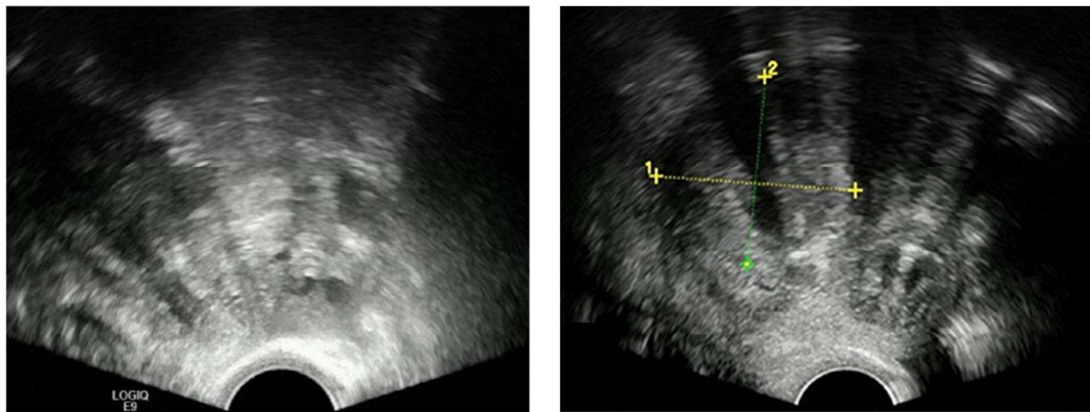


Figure 1. Ultrasound of diffuse leiomyomatosis at presentation.



Figure 2. Myomectomy findings of diffuse leiomyomatosis.

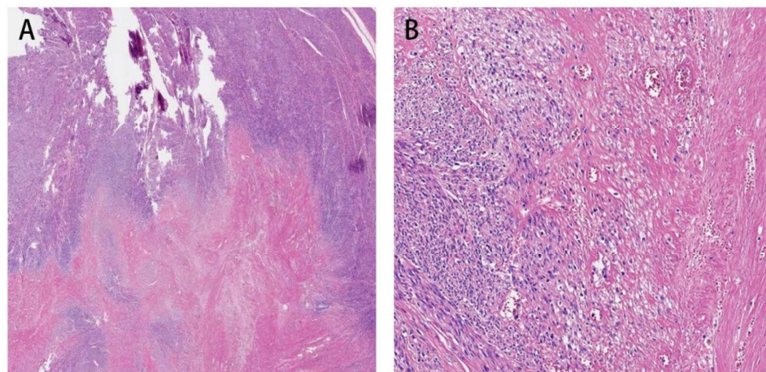


Figure 3. Microscopic features of diffuse leiomyomatosis.

- (A) HE section of low magnification.
(B) HE section of Higher magnification.

3. Discussion

DUL is a disease characterized by symmetrical enlargement of the uterus and almost complete replacement of the myometrium with innumerable smooth muscle nodules, often accompanied by dissemination into the uterine cavity and the formation of submucosal fibroids. The most common clinical symptoms of DUL are menorrhagia, lower abdominal pain, infertility, and bladder or rectal compression [1].

Histologically, the diffusely growing smooth muscle nodules are spindle cells with increased cellularity and vesicular nuclei. Unlike multiple leiomyomata, there is no clear demarcation between nodules growing in the myometrium in DUL. The difference from Endometrial stromal sarcoma is that DUL's leiomyoma cells grow in fascicular growth pattern rather than sheet-like growth. Through X-linked allele inactivation analysis, it can be further confirmed that the different tumoral foci of DUL are derived from different clones of cells. It has therefore been suggested that DUL may be related to multiple leiomyomas [7].

Hysterectomy is generally considered as the first choice for radical cure of DUL. Of note, several cases of patients with DUL showing recurring symptoms within 5 years after myomectomy, a less radical treatment, have been reported [8]. However, since the patients with DUL are young women, many patients seek medical treatment due to infertility. Therefore, the exploration of fertility-preserving treatment options becomes particularly important.

Some studies have demonstrated that hysteroscopic myomectomy has a certain therapeutic effect on infertile patients in the early stage of DUL, which is a favorable option for patients who wish to preserve fertility and conceive in a short period [9-11]. Compared with hysteroscopy, laparotomic myomectomy, although more invasive, can remove a wider range of lesions. Ikuo Konishi suggested that 3-6 months of pretreatment with gonadotropin-releasing hormone (GnRH) agonist or antagonist should be given prior to laparotomic myomectomy, and MRI should be performed again after GnRHa treatment to assess the extent of the lesion. Also in the case he reported, the patient underwent laparotomy myomectomy after GnRHa treatment and ultimately had a good pregnancy outcome [12]. Chen et al. reported the first case of DUL treated by high intensity focused ultrasound (HIFU) ablation [13]. Later, Zhang et al. performed HIFU on 8 patients with DUL, 7 of whom showed a significant reduction in uterine volume at postoperative follow-up, and 1 patient had symptoms of menorrhagia again 8 months after surgery, and received HIFU again [14]. So far, there is no research to prove whether HIFU has an effect on the fertility of DUL patients, and there is no cases of successful pregnancy after HIFU. The therapeutic effect of HIFU still needs to be further comprehensively evaluated. For DUL patients with a strong desire to retain the uterus, uterine artery embolization (UAE) is also commonly administered. Scheurig et al. reported the cases of 6 patients with DUL who received UAE treatment, of whom 1 patient showed an increase in uterine volume at 4 months after surgery,

while the remaining 5 patients were relieved of their clinical symptoms after UAE [15].

In this case, the patient had no fertility requirements, but nevertheless had a strong desire to preserve the uterus. Considering that the patient had a history of myomectomy, and in order to make the greatest effort to remove leiomyomas, we elected to perform extensive myomectomy. During the operation, we removed most of the myometrial lesions as much as possible, while simultaneously resecting the part of the endometrial cavity with a high density of submucosal fibroids. This procedure effectively reduced the volume of the uterus and relieved the patient's symptom of menorrhagia.

4. Conclusion

This study reports a case of DUL treated by laparotomy myomectomy. This case report enriches existing DUL case data and further explores the surgical management of preserving the uterus and improving symptoms. With the development of endoscopy, ultrasound, and interventional technique, the treatment methods for DUL have become more mature, but the long-term efficacy of treatment and their impact on pregnancy outcomes still need to be further studied.

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