

Case Report

To cite: Girish B, Bhat VV. Successful Pregnancy Outcome in a Case of Diffuse Uterine Leiomyomatosis: A Journey through Three Surgeries and Thirty-five Fibroids. Pan Asian J Obs Gyn 2019;2(3):1-4.

Received on:

Accepted on:

Successful Pregnancy Outcome in a Case of Diffuse Uterine Leiomyomatosis: A Journey through Three Surgeries and Thirty-five Fibroids

¹Bhavana Girish, ²Vidya V Bhat

¹Fellow of Minimally Invasive Surgery, ²Director

Department of Obstetrics and Gynaecology, Radhakrishna Multispeciality Hospital and IVF Center, Bangalore, Karnataka, India

Access this Article Online
Website: www.pajog.com
Quick Response Code

ABSTRACT

Diffuse uterine leiomyomatosis (DUL) is a rare condition that poses a unique management challenge in women who desire fertility. The need to resect innumerable uterine myomata must be balanced with the desire to retain fertility potential in these women. Hysteroscopic myomectomy is the treatment of choice for DUL as myomas with submucosal or intracavitary components are the main cause of menorrhagia and impaired fertility, and hence resection of these myoma with the aim of achieving a normal-looking endometrial cavity would optimize reproductive outcome. We report the case of a woman with DUL who was managed with hysteroscopic myomectomy in which 17 submucosal myomas were removed followed by a successful pregnancy.

Keywords: Hysteroscopy, myomectomy, infertility.

INTRODUCTION

Diffuse uterine leiomyomatosis (DUL) is a rare condition characterized by the presence of innumerable benign uterine leiomyomata that are 3 cm or less in diameter.¹ The myomas are generally submucosal or intramural in location, ill-defined and thought to be an extreme type of multiple uterine fibroids.² This condition is associated with severe menorrhagia and is most often seen in women of reproductive age group.

There are less than 40 cases of DUL reported in modern literature. Hysterectomy has been suggested as the standard treatment as myomectomy might be unable to achieve a complete clearance and repair of the uniformly involved myometrium.³ Other treatment modalities that have been attempted include hormonal treatment⁴ and uterine artery embolization,⁵ both with limited success. However, the optimal treatment of women with DUL wishing to become pregnant is still a matter of debate.

Several authors have recently proposed hysteroscopic myomectomy as a treatment for DUL in accordance

with the concept that myomas with submucosal or intracavitary components are the main cause of menorrhagia and impaired fertility, and hence resection of these myomata alone with the aim of achieving a normal-looking endometrial cavity would optimize reproductive outcome.⁶ We describe the case of an infertile woman with DUL who had already undergone laparoscopic myomectomy and presented to us with recurrence of fibroids.

CASE REPORT

A 26-year-old, nulliparous woman with infertility and menorrhagia for last 3 years was referred to our Endoscopic centre. She had a history of laparoscopic

Address for Correspondence

Bhavana Girish
Fellow of Minimally Invasive Surgery
Department of Obstetrics and Gynaecology
Radhakrishna Multispeciality Hospital and IVF Center
Bangalore, Karnataka, India
bhavana.sgirish@gmail.com

myomectomy performed 2 years back following which she underwent in-vitro fertilization with embryo transfer which was unsuccessful. Her pelvic examination revealed a diffusely enlarged uterus corresponding to the size of about 12 weeks gestation. A transvaginal scan was performed which showed the presence of innumerable intramural and submucosal fibroids involving all walls of the uterus and maximally measuring around 30 mm in diameter. Her ovaries and other pelvic organs appeared normal. On Magnetic Resonance Imaging, the uterus showed the presence of more than 20 nodular lesions with T2 hypointensity with a confluent appearance, suggestive of leiomyomatosis. The myomata involved body and fundus of the uterus, with the largest present in the anterior wall and resulted in a distortion of the endometrial lining (Fig. 1).

With the provisional diagnosis of DUL, the patient was admitted for pre-operative workup and hysteroscopic



Fig. 1: MRI showing multiple myomas with distorted endometrial cavity

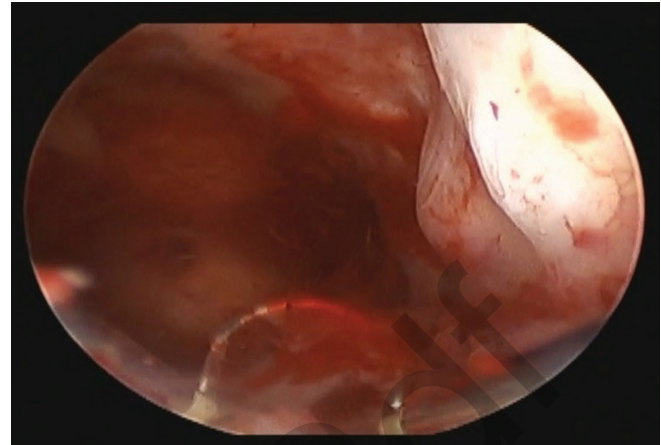


Fig. 2: Hysteroscopy image showing multiple submucosal myomas

myomectomy. She was severely anaemic on admission with a haemoglobin of 6.8 g/dL and was given two units of packed red cell transfusion prior to the procedure. The procedure was performed under spinal anaesthesia. An initial diagnostic hysteroscopy was performed (Fig. 2) followed by hysteroscopic myomectomy using a monopolar resectoscope. Seventeen myomas including FIGO grade 0, 1 and 2 submucous myomas spread throughout the uterine cavity were removed and sent for histopathological examination. We monitored the intravasation of glycine media using a hysteroscope and fluid output measurement. The entire procedure lasted 35 minutes and the maximum safety level of distension media was reached (1000 mL), there was no excessive bleeding post-procedure.

We did not attempt to remove larger G2 and G3 myoma to avoid excessive endocavitary trauma, which would have resulted in the occurrence of intrauterine synechiae. Due to the larger amount of fluid intravasation, the patient was monitored in the intensive care unit overnight following which she was discharged home. She was given one cycle of estradiol valerate 6 mg per day followed by progesterone withdrawal to promote endometrial regeneration. She was also given 2 months of ulipristal acetate post-operatively.

A follow-up ultrasonography performed 2 months after the procedure showed a few intramural fibroids varying between 5 and 30 mm in size away from the uterine cavity and a uniform endometrial stripe. There were few myomas immediately adjacent to and abutting the endometrial cavity. The woman had



Fig. 3: J-shaped uterine incision given during Caesarean section

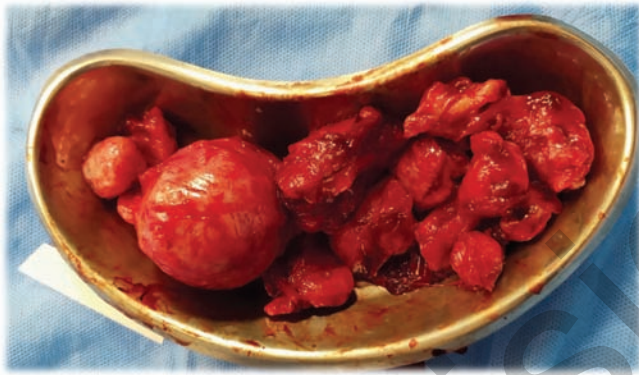


Fig. 4: 18 Fibroids removed during Caesarean myomectomy

regular menstrual cycles with average flow and was encouraged to try for a spontaneous pregnancy. Three months later, the patient spontaneously conceived. She had an uneventful antenatal course after which she developed preterm premature rupture of membranes at 36 weeks and 3 days gestation. She was taken up for an emergency caesarean section due to footling breech presentation and a healthy male baby weighing 2.5 kg was delivered with normal APGAR scores. A J-shaped uterine incision was given due to presence of a 4 × 4 cm myoma at the incision site (Fig. 3) following which caesarean myomectomy was performed with removal of 18 fibroids measuring around 3 × 3 cm, mainly submucosal in location (Fig. 4). She was given 1-unit blood transfusion post-operatively due to a Hb of 8.4 g/dL. She tolerated the procedure well and was discharged 3 days post-procedure.

DISCUSSION

Diffuse uterine leiomyomatosis is characterized by the following:

- Innumerable poorly defined small (0.5–3 cm) myomas
- Total involvement of the myometrium
- Symmetrical enlargement of the uterus

The exact pathogenesis is not known; however, this disease has been thought of as an extreme example of multiple uterine leiomyomata.² Although the underlying histology is similar to leiomyomata, the main consideration during management should be that there are too many nodules for successful removal by myomectomy. Thus, treatment of this condition should be to optimally restore fertility, avoid myometrial compromise and also to avoid incomplete myomectomy.

Although there have not been any well-designed studies about fibroids and fertility, it is believed that the risk of infertility and miscarriage are increased when the endometrial cavity is distorted by leiomyomas.⁷ Since it has been reported recently that pregnancy can be achieved by removal of submucosal myomas alone in patients with DUL,⁶ the resection of intramural myomas may be unnecessary in these patients. Yen et al in their case series of 5 patients with DUL, reported that after clearance of intra-cavitary myomas, conception can occur in the presence of multiple small intramural myomas, with no interference to the ongoing pregnancy and delivery. The typically small size of the multiple intramural myomas in such patients may be the reason that they had little effect on conception and ongoing pregnancy, because it was reported that only intramural myomas of size >4 cm had impact on lowering the pregnancy rate.⁸ They also concluded that conception can occur in an imperfectly treated cavity with a few bands of scar tissue and/or small submucosal myomas in the periphery, and with intramural myomas immediately adjacent to the endometrial lining, as was also noted in our patient. They stated that an early second-look and early intervention could be the most beneficial principle in preventing intrauterine synechiae. From our experience, we have rarely encountered post-operative intrauterine synechiae after hysteroscopic myomectomy due to our routine practice of treatment with cyclical oestrogen and progesterone for 3 months after surgery to promote endometrial regeneration and we do not routinely perform a second-look hysteroscopy in these women.

As our patient also had severe menorrhagia, we considered that the myomas should be removed as completely as possible. However, as she had already undergone a previous laparoscopic myomectomy with removal of the larger intramural myomas, we decided to only remove submucosal myomas protruding into the uterine cavity by hysteroscopic surgery. Previous case reports have also successfully used medical management with GnRH-analogues in women with DUL.⁴ We decided against this approach as in our experience, pre-operative administration of GnRH-analogues results in shrinkage of submucous fibroids leading to incomplete resection and resulting in higher recurrence rates with poorer pregnancy outcome. To prevent recurrence, we used ulipristal acetate for 2 months in our patient. Our report confirms studies elsewhere in which the hysteroscopic removal of submucosal myomas had good long-term results in controlling menorrhagia and avoiding hysterectomy. The strength of this treatment is the preservation of the uterus with vaginal approach, the avoidance of extensive myometrial wounding with a complete recovery of menorrhagia, and a satisfactory outcome of fertility.

Pregnancy in women with DUL is associated with increased incidence of obstetric complications including miscarriage, preterm delivery, malpresentations, caesarean section and intrapartum haemorrhage. There is a theoretical risk of adherent placenta following hysteroscopic myomectomy, although this is an extremely rare occurrence.⁹ Literature also has mentioned the risk of placental abruption due to faulty placentation over a myoma,¹⁰ but this too did not occur in our patient, although her intramural myomas appeared quite close to the endometrial lining. Our patient, who had extensive inner-myometrial surgery and the coexistence of multiple intramural myomas, had no problems with uterine integrity during pregnancy, labour, or postpartum contraction, and she did not show any signs of uterine rupture, uterine atony, or postpartum haemorrhage. There have been no adverse neonatal outcomes reported in previous studies, so the prognosis of pregnancy in women with DUL appears to be favourable based on past reports.

CONCLUSION

Patients desirous of pregnancy, with DUL with previous surgeries should be managed by hysteroscopic surgery

with the aim of restoring the endometrial cavity. This might be an incomplete surgery but this can deal with the current problems of infertility and menstrual complaints in patients desirous of pregnancy.

Source of Support

Nil

Conflict of Interest

There are no conflicts of interest.

Financial Disclosure

Nil

REFERENCES

1. Fedele L, Zamberletti D, Carinelli S, Motta T, Candiani GB. Diffuse uterine leiomyomatosis. *Acta Eur Fertil.* 1982;13:125-31.
2. Baschinsky DY, Isa A, Niemann TH, Prior TW, Lucas JG, Frankel WL. Diffuse leiomyomatosis of the uterus: a case report with clonality analysis. *Hum Pathol.* 2000;31:1429-32.
3. Lapan B, Solomon L. Diffuse leiomyomatosis of the uterus precluding myomectomy. *Obstet Gynecol.* 1979;53(3 Suppl):82S-84S.
4. Fedele L, Bianchi S, Zanconato G, Carinelli S, Berlanda N. Conservative treatment of diffuse uterine leiomyomatosis. *Fertil Steril.* 2004;82:450-3.
5. Kido A, Monma C, Togashi K, Ueda H, Itoh K, Fujii S, et al. Uterine arterial embolization for the treatment of diffuse leiomyomatosis. *J Vasc Interv Radiol.* 2003;14:643-7.
6. Shimizu Y, Yomo H, Kita N, Takahashi K. Successful pregnancy after gonadotropin-releasing hormone analogue and hysteroscopic myomectomy in a woman with diffuse uterine leiomyomatosis. *Arch Gynecol Obstet.* 2009;280(1):145-7.
7. Pritts EA. Fibroids and infertility: a systematic review of the evidence. *Obstet Gynecol Surv.* 2001;56:483-91.
8. Oliveira FG, Abdelmassih VG, Diamond MP, Dozortsev D, Melo NR, Abdelmassih R. Impact of subserosal and intramural uterine fibroids that do not distort the endometrial cavity on the outcome of in vitro fertilization-intracytoplasmic sperm injection. *Fertil Steril.* 2004;81:582-7.
9. Capella-Allouc S, Morsad F, Rongieres-Bertrand C, Taylor S, Fernandez H. Hysteroscopic treatment of severe Asherman's syndrome and subsequent fertility. *Hum Reprod.* 1999;14:1230-3.
10. Cooper NP, Okolo S. Fibroids in pregnancy—common but poorly understood. *Obstet Gynecol Surv.* 2005;60:132-8.