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Case report

Management of complete vagino-uterine septum in patients seeking fertility: Report of two cases and review of literature

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ABSTRACT

Septate uterus is the most common congenital uterine anomaly encountered clinically. It is associated with infertility and adverse pregnancy outcomes including miscarriages and preterm births. Over the past few decades, the diagnosis and surgical management of uterine septum has changed significantly. Here, we present two women with complete vagino-uterine septum presenting with infertility. The diagnoses of septate uterus were made through three-dimensional ultrasound and magnetic resonance imaging, and they were treated with thin-gauge bipolar electrocautery (VERSAPOINT) through hysteroscopy. A literature review of the approach to managing complete vagino-uterine septum suggests the use of combinatorial imaging modalities such as hysterosalpingography, three-dimensional ultrasonography, and magnetic resonance imaging to accurately define the anatomy. Furthermore, compared with open surgery, treatment through hysteroscopic approaches are associated with shorter operating times and good fertility outcomes. With the use of concomitant laparoscopic and ultrasound monitoring, the incidence of uterine perforation and visceral injury may be reduced. However, referral to a center with such expertise is essential.

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Introduction

Septate uterus, the most common congenital uterine anomaly, arises when there is persistence of the midline partition between the two fused Müllerian ducts during embryological development. Depending on the extent of the failure of reabsorption of this partition, the midline septum may extend from the uterine fundus to before or after the internal cervical orifice (partial or complete uterine septum), to the external cervical orifice (complete uterine septum with septate cervix), or to the upper vagina (complete uterine septum with cervical and vaginal septations; [Figure 1](#)).¹

It has been reported that the incidence of uterine anomalies ranges between 0.5% and 1.9% in the general population,² with a higher incidence of up to 10% found in women presenting with subfertility.^{3,4} The diagnosis and management of patients with complete vagino-uterine septum have been a challenge when

controversy arises in different areas of treatment, particularly in patients seeking fertility treatment. In this paper, we present two women with complete vagino-uterine septum who underwent hysteroscopic metroplasty with resection of the cervical and vaginal septum, and review the evidence behind the modern management of complete vagino-uterine septum.

Case Report

Case 1

Patient A is a 28-year-old woman who was referred to our fertility clinic with primary infertility for 2 years. She attended a health screening a few years previous and had an ultrasound of her pelvis done, which showed a septate uterus. She has a regular menstrual cycle associated with dysmenorrhoea requiring regular simple analgesia, but did not complain of dyspareunia.

On examination, she appeared to have two cervixes and a vaginal septum. Imaging studies through hysterosalpingogram, ultrasound, and magnetic resonance imaging (MRI) confirmed the findings of a septate uterus with a complete septum extending from

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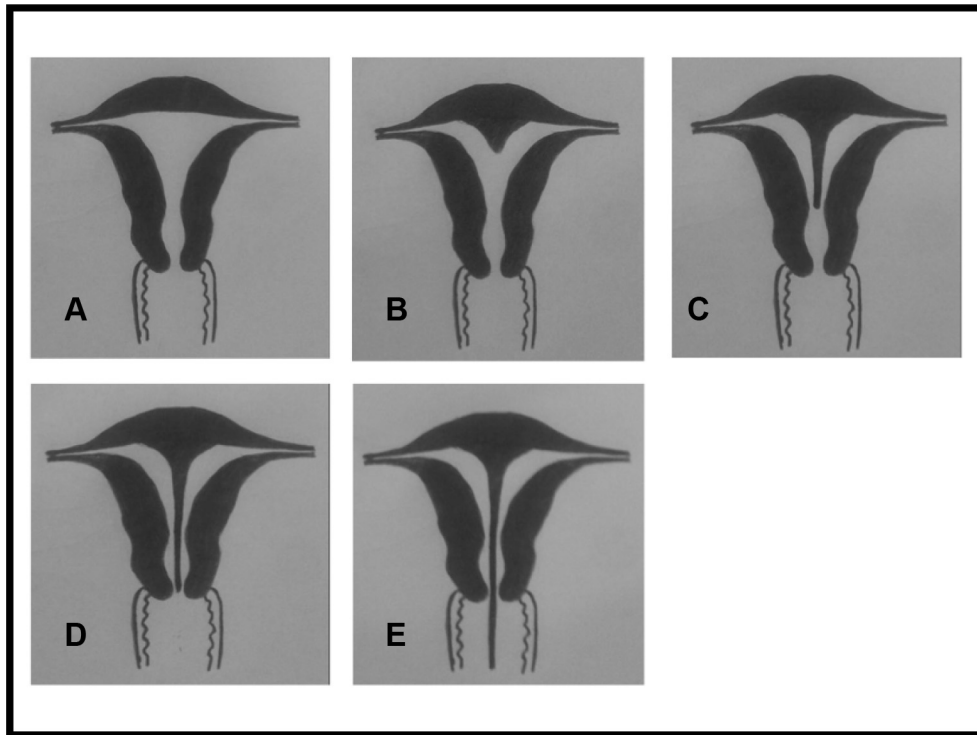


Figure 1. An illustration showing the different extents of septate uterus. (A) Normal uterus. (B) Partial uterine septum. (C) Complete uterine septum. (D) Complete uterine and cervical septum. (E) Complete uterine, cervical, and vaginal septum.

the uterine fundus into the cervix (Figures 2A and 2B). Renal tract evaluation was normal.

She underwent resection of the vagina septum and hysteroscopic transcervical resection of the cervical and uterine septum with concomitant laparoscopy (Figures 2C–2E). Laparoscopic dye test was also performed and this showed patent fallopian tubes. The vaginal septum was divided with scissors and sutured, and the cervical and uterine septum was resected with a thin-gauge bipolar electrocautery (VERSAPOINT; ETHICON, part of the Johnson & Johnson Family of Companies). The total fluid volume used was 2.8 L and fluid deficit was 200 mL. The operation took 1 hour and 23 minutes. An intrauterine contraceptive device (IUCD) was inserted after the procedure and she was discharged well on the 1st postoperatively day. She was given estradiol valerate 6 mg daily for 4 weeks to encourage endometrial regeneration. The IUCD was removed 3 weeks after the surgery, and she proceeded for intracytoplasmic sperm injection due to male factor infertility 3 months postsurgery.

Case 2

Patient B is a 30-year-old woman who presented with primary infertility of 2 years. She has no significant past medical history and has a regular menstrual cycle. Physical examination revealed a vaginal septum. Transvaginal-ultrasound and MRI of the pelvis showed a septate uterus with a complete septum down to the cervix (Figure 3A) and multiple fibroids of up to 5 cm in size. Her husband's semen analysis showed asthenoteratozoospermia. Infertility work-up for the couple was otherwise unremarkable.

She underwent resection of the vagina septum, hysteroscopic transcervical resection of the cervical and uterine septum, and laparoscopic myomectomy (Figures 3B–3D). The vaginal septum was dissected with scissors and sutured. Cervical and uterine septum was dissected with VERSAPOINT until the level of ostium.

An IUCD was inserted after the procedure and was kept for 10 days postoperatively. Laparoscopic myomectomy was performed after resection of the uterocervical septum. Myoma tissue fragments measuring 9.5 cm × 8.5 cm × 1.5 cm and weighing 50 g in aggregate was removed. The operation took 2 hours and 15 minutes. Estradiol valerate (6 mg daily) was instituted for 4 weeks, and dydrogesterone was given 1 week after surgery.

At 3 months postoperatively, ultrasound sonohysterography was performed and showed a residual septum measuring less than 1 cm (Figure 3E). She was prescribed clomiphene citrate empirically and was encouraged to maintain regular sexual activity. She subsequently conceived with *in vitro* fertilization 1 year postoperatively. Cervical cerclage was inserted prophylactically and a healthy baby weighing 2924 g was delivered at term.

Discussion

The presence of a septate uterus is associated with a reduced clinical pregnancy rate (RR 0.86).⁵ It also increases the risk of first-trimester miscarriage (RR 2.37), second-trimester miscarriage (RR 3.74), preterm birth (RR 2.30), and fetal malpresentation (RR 6.15).⁵ The diagnosis of septate uterus may be confused with uterine didelphys,⁶ especially in cases where the uterine septum is complete, with duplication of the cervix and a longitudinal vagina septum are present. The management of these uterine anomalies are different and thus, getting the correct diagnosis is important in planning for any surgical intervention, and this can be achieved with multimodality imaging through hysterosalpingography, ultrasonography, and MRI. In a study by Patton et al,⁶ 15 out of 16 cases of uterine anomalies were correctly identified with the above combination of investigations.

In the early 1980s, it was shown by several groups that metroplasty of septate uterus reduces pregnancy wastage.^{7–9} At that

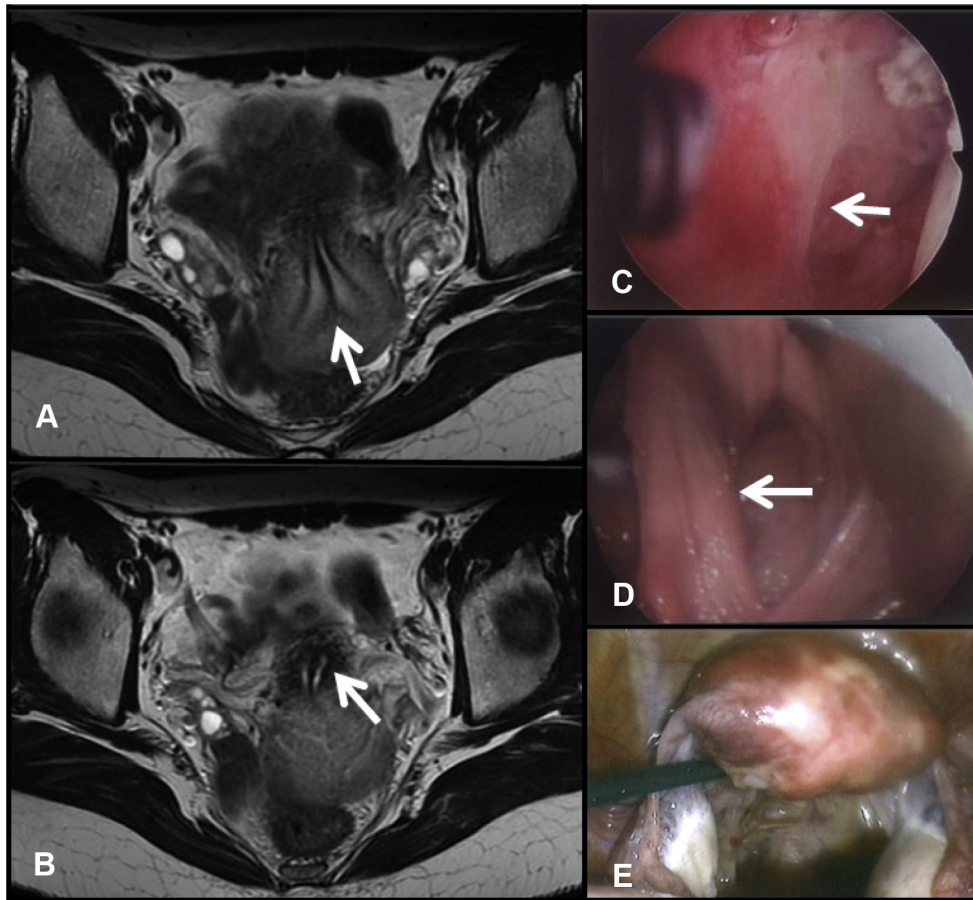


Figure 2. Images of Patient A. (A) Axial T2-weighted magnetic resonance imaging pelvis images showing a complete uterine septum (arrow); (B) a cervical septum (arrow); (C) hysteroscopic view of the uterine septum (arrow); (D) upper vaginal septum (arrow); and (E) laparoscopic view of the pelvis showing a normal uterine fundus.

time, metroplasty was performed transabdominally with the Jones and Tompkins procedure. However, hysteroscopic metroplasty is now the recommended treatment for most uterine septums as there is a higher risk of complications with the abdominal approach.¹⁰ This includes postoperative adhesions, which can potentially lead to infertility and scar rupture in future pregnancies. In addition, laparotomy also results in a longer hospital stay.¹⁰ Even in cases where laparotomy is required, hysteroscopic metroplasty is still the preferred approach to prevent potential subsequent risk especially scar rupture in future pregnancies.

A host of different hysteroscopic approaches with various instruments have been described for the treatment of uterine septum, including cold scissors, unipolar resectoscopes, thin-gauge bipolar electrocautery (VERSAPOINT), and laser devices. [Table 1](#) provides an overview of individual studies on the operative time and pregnancy outcomes of different hysteroscopic methods used for metroplasty. Two prospective studies, one randomized study, and another cohort study, comparing unipolar resectoscopes and VERSAPOINT demonstrated comparable reproductive outcomes but shorter operating times and lower complication rates for the VERSAPOINT group.^{11,12} Other studies comparing unipolar resectoscopes and cold scissors were inconclusive as to which is more superior.^{13,14} Laser resection has been shown to be effective,^{15–17} although not surprisingly, the use of scissors for resection was found to be the simplest, fastest, and least expensive method for correction of uterine septum.¹⁷ In 2007, Bettocchi et al¹⁸ reported that office hysteroscopic metroplasty was successfully performed using 5Fr scissors through an office hysteroscope with an outer

sheath measuring 4 mm in diameter without any analgesia or anaesthesia. Although the introduction of minihysteroscopes avoids cervical dilatation and reduces uterine injury, and the safety of bipolar surgery in reducing electrolyte imbalance and thermal injury, more evidence based data is required in order for it to be considered as the new gold standard in surgical treatment of uterine septums.¹⁹ The use of monopolar resectoscopes requires an electrolyte-free distending medium such as hypotonic glycine, glucose, sorbitol, or mannitol which requires strict real-time monitoring of systemic absorption of the fluid media in order avoid fluid overload, hyponatremia, and even death.²⁰ The use of bipolar energy devices such as the VERSAPOINT, allows the use of isotonic saline as a distention media, thus reducing the risks of hypernatremia.

In general, indications for metroplasty include repeated pregnancy loss and otherwise unexplained cause of infertility. This is supported by a pooled analysis of 18 studies including 1501 women on the outcome of hysteroscopic metroplasty, which showed an overall pregnancy rate of 60.1% and live birth rate of 45.0%. This analysis included women with recurrent miscarriages, late miscarriages, primary infertility, secondary infertility, and preterm delivery.¹⁶ More recent studies have also confirmed that hysteroscopic resection of the uterine septum is a safe procedure which can improve pregnancy outcomes.^{21–26} [Tables 2 and 3](#) illustrate the pregnancy outcomes of women with septate uterus before and after hysteroscopic metroplasty. Other relative indications for metroplasty include infertility requiring assisted reproductive technologies.¹ Several retrospective matched-control studies have reported

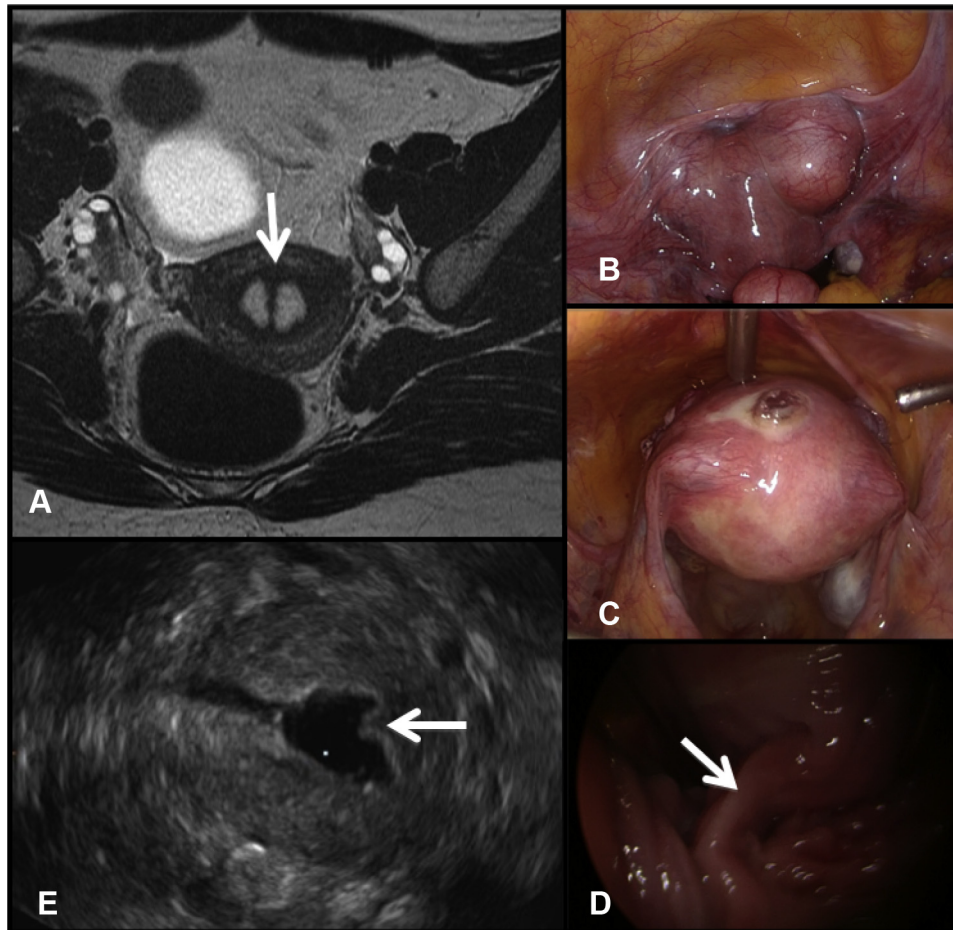


Figure 3. Images of Patient B. (A) Axial T2-weighted magnetic resonance imaging pelvis image showing a septate uterus. (B) Laparoscopic view of the pelvis showing multiple fibroids. (C) Laparoscopic view of the pelvis after myomectomies showing a normal uterine fundus. (D) Upper vaginal septum. (E) Three-dimensional sonographic image showing a small residual septum 3 months after resection.

Table 1
Operation time and pregnancy outcomes of different hysteroscopic metroplasties.

Study	Patient No.	Instruments used	Operation time (min)	Outcomes		
				Pregnancy rate (%)	Delivery rate (%)	Abortion rate (%)
Colacurci et al, 2007 ¹¹	160	Monopolar resectoscope	23.4	70.0	81.6	18.4
		VERSAPOINT	16.9	76.9	84.0	16.0
		Differences	$p < 0.05$	NS		
Litta et al, 2008 ¹²	63	Monopolar resectoscope	20.5	81.2	61.5	23.1
		VERSAPOINT	15.4	79.3	60.9	26.1
		Differences	$p < 0.05$	NS		
Cararach et al, 1994 ¹³	70	Monopolar resectoscope	10	67.9	—	25.0
		Scissors	30	88.2	—	20.0
		Differences	N/A	$p < 0.05$	—	NS
Vercellini et al, 1993 ¹⁴	23	Monopolar resectoscope	22.0	—		
		Scissors	17.0	—		
		Differences	NS			
Yang et al, 2006 ¹⁵	45	Neodymium: YAG laser	—	71.0	—	—
Choe & Baggish, 1992 ¹⁶	14	Neodymium: YAG laser	—	68.4	52.6	—
Candiani et al, 1991 ¹⁷	21	Argon laser	30	—		
		Scissors	19	—		
		Differences	57% longer in laser group ($p = 0.001$)	—		

N/A = not available; NS = not significant.

improvement in pregnancy rate and live birth rate and reduction in abortion rate in patients who underwent *in vitro* fertilization, or intracytoplasmic sperm injection after hysteroscopic resection of uterine septum.^{27–29} As our patients are planned for assisted reproductive technologies, metroplasty was performed.

In cases where a cervical septum is present, there is debate as to whether the cervical septum should be preserved. Some have chosen to preserve the cervical septum with the concern that the unification of the cervix may result in cervical incompetence and profuse bleeding intra-operatively.³⁰ Others prefer to unify the

Table 2
Literature on pregnancy, live births, and abortion rates of women with uterine septum before and after hysteroscopic metroplasty.

Study	Uterine anomaly	Patient characteristic	Metroplasty status	Patient No.	Pregnancy rate (%)	Live birth rate (%)	Abortion rate (%)
Tonguc et al, 2011 ²⁶	PUS, CUS	Primary infertility	Not done	25	20.0	8.0	60.0
			Done	102	43.1	35.3	11.4
			Differences	—	$p = 0.03$	$p = 0.008$	$p = 0.02$
Ban-Frangež et al, 2009 ²⁸	PUS, CUS	IVF/ICSI	Not done (control) ^a	31	—	—	80.6 (21.0) $p < 0.001$
			Done (control) ^a	106	—	—	29.2 (19.8) NS
Tomazević et al, 2010 ²⁹	PUS, CUS	IVF/ICSI	Not done	289	16.0 (39.0)	2.5 (33.9) $p = 0.001$	77.1 (16.7)
			(control) ^a	—	$p < 0.03$	—	$p < 0.001$
			Done (control) ^a	538	34.7 (39.7) NS	26.2 (32.1) NS	29.2 (18.4) NS

CUS = complete uterine septum; ICSI = intracytoplasmic sperm injection; IVF = *in vitro* fertilization; NS = not significant; PUS = partial uterine septum.

^a Control = normal uterus.

Table 3
Literature on pregnancy rates and live birth rates of women with uterine septum after hysteroscopic metroplasty.

Study	Study design	Patient No.	Uterine anomaly	Patient characteristics	Pregnancy rate (%)	Live birth rate (%)
Nouri et al, 2010 ²¹	Systematic review	1587	PUS, CUS	Recurrent & late miscarriages, primary & secondary infertility, preterm delivery	60.0	45.0
Bendifallah et al, 2013 ²²	Observational retrospective study	128	PUS, CUS	Primary infertility & recurrent miscarriages	60.9	54.7
Bakas et al, 2012 ²³	Prospective observational study	68	PUS, CUS	Unexplained primary infertility	44.0	36.8
Dalal et al, 2012 ²⁴	Unknown	72	PUS, CUS	Unexplained primary infertility	45.8	—
Paradisi et al, 2011 ²⁵	Retrospective comparative single-center study	246	PUS	Unexplained infertility & recurrent miscarriages	61.3	64.9
Tonguc et al, 2011 ²⁶	Retrospective study	102	PUS, CUS	Primary infertility	43.1	35.3

CUS = complete uterine septum; PUS = partial uterine septum.

cervix to improve the safety and ease of performing hysteroscopic metroplasty.^{31,32} Moreover, a prospective observational study on 10 women with complete uterine septum who underwent incision of the cervical septum with Metzenbaum scissors showed that none of the women experienced significant bleeding, cervical incompetence, second trimester miscarriage, or preterm delivery.³² In fact, it has been associated with lower caesarean section rates, although the plausible mechanism remains unknown.³¹ The cervical septum was resected in our patients without any excessive bleeding intraoperatively. As for the vagina septum, resection was readily achieved. This is usually done for symptom relief such as dyspareunia.

Postoperatively, our patients had IUCDs inserted and were prescribed exogenous estrogen therapy. The underlying rationale was to keep the denuded areas from adhering together, whereas estrogen therapy is given to promote endometrial growth over the raw areas. However, a few small studies reported that there were no differences between those who were treated with IUCDs, balloon catheters, or estrogen therapy, and those who had no treatment postoperatively in terms of intrauterine adhesion formation and pregnancy outcomes, suggesting that such measures are not necessary.^{33–37} As the number of study patients in these studies is small, further studies are required in this area.

Hysteroscopic metroplasty has been associated with short- and long-term complications, which commonly include perforation, bleeding, cervical lacerations, formation of intrauterine synechiae, increased risk of uterine rupture, and adherent placenta in subsequent pregnancy. In a meta-analysis by Valle and Ekpo,¹ a surgical complication rate of ~1.5% was reported out of 2167 cases analyzed. Several cases of uterine rupture have been reported and all described some complications during metroplasty including excessive excision and uterine perforation especially when using monopolar electrocautery.¹ This can occur even prior to the onset of labor, and as early as 19 weeks of gestation.³⁸ In our patients, hysteroscopic metroplasty was performed with laparoscopic guidance. This allows a direct vision of the limits of resection, and to

displace any loops of bowel lying next to the uterus. An alternative to laparoscopic monitoring is with the use of real-time ultrasonography.^{39–40}

Kormányos et al⁴¹ reported a complete uterine septum resection rate of 62%, where a normal uterine architecture was detected on follow-up. This study also showed a significant higher delivery and term pregnancy rate in the group where uterine resection was complete as compared with the group with remnant septums. In addition, after resection of the remnant septum, this group of women showed increased pregnancy, delivery, and term pregnancy rates. Fedele et al⁴² further showed that a residual septum of between 0.5 cm and 1.0 cm does not appear to worsen the reproductive prognosis when compared with a residual septum of < 0.5 cm.⁴² There is therefore controversy on the management of residual septums. The most crucial part of hysteroscopic metroplasty would be achieving complete uterine septum resection without causing any perforation or leaving any residual septum.

Conclusion

Over the past decades, the evolution of minimally invasive techniques such as hysteroscopic metroplasty has enabled the management of uterine septums to be safer and less morbid compared with the traditional transabdominal approach. With studies demonstrating good pregnancy outcomes and a generally low morbidity after hysteroscopic metroplasty, this procedure should be considered in patients with septate uterus seeking for fertility. The use of a thin-gauge bipolar electrocautery (VERSA-POINT) has been shown to shorten operating times and reduce intraoperative complications while maintaining good pregnancy outcomes. With the use of concomitant laparoscopic or ultrasound monitoring, the incidence of uterine perforation or visceral damage may also be reduced.

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