Unicornuate uterus and uterus didelphys Indications and techniques for surgical reconstruction: A review

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Summary

Unicornuate uterus and uterus didelphys consist of miscellaneous congenital malformations of the female genital system. These anomalies can cause many gynecological and obstetrical complications including infertility, ectopic pregnancy, recurrent abortions and preterm deliveries. Laparoscopy and hysteroscopy are two helpful operative procedures in establishing an accurate diagnosis and treating effectively. A case of a patient with unicornuate uterus and longitudinal vaginal septum, who presented at our hospital suffering from infertility, is reported.

Key words: Unicornuate uterus; Uterus didelphys; Surgical reconstruction.

Introduction

Unicornuate and didelphys uterus are not often reported in the obstetrical and infertility literature. These types of abnormalities are manifested more often because of their symptoms, caused either by obstruction associated with the rudimentary horn or the presence of a longitudinal vaginal septum of uterus didelphys. Andrews and Jones [1] reported only five cases in their 12-year experience in infertility and obstetrical practice at John Hopkins Hospital. In another trial over a period of 40 years at the same hospital, there were only 29 cases of unicornuate uterus and 25 cases of uterus didelphys [2].

Unicornuate uterus with rudimentary horn results from arrested development of one of the two Mullerian ducts. According to the American Fertility Society [3] the unicornuate uterus group is divided into four subgroups as follows: (IIa) rudimentary horn with cavity communicating to unicornuate uterus, (IIb) with cavity non-communicating, (IIc) with no cavity, (IId) with no horn. Type IIb is the most common and from the clinical point of view, the most important. It is also more susceptible to many obstetric and gynecological complications that can be avoided by the removal of the rudimentary horn. Unicornuate uterus cases with rudimentary horn, in a percentage of 75%-90%, are non-communicating [4].

Didelphys uterus results from complete failure of the Mullerian ducts to fuse in the midline. In general, the failure of duct fusion causes various kinds of uterine anomalies that are classified into seven classes. This classification does not describe most vaginal anomalies. A proposed classification is mentioned in Table 2 [5].

The above-mentioned congenital anomalies are usually associated with contralateral renal agenesis on the affected side, or ipsilateral renal agenesis in cases of obstructed hemivagina in didelphys uterus. Other congenital renal anomalies include hypoplastic, pelvic and horseshoe kidney, combined with ectopic ureter. Didelphys uterus can also be associated with partial vaginal atresia. The obstruction of one hemivagina can cause hematocolpos and hematometra, as well as endometriosis due to retrograde menstruation in the abnormal cavity.

The frequency of unicornuate uterus with rudimentary horn ranges from 1/20,000 to 1/60,000 [6]. The incidence of gestation in a uterine horn ranges from 1/100,000-140,000 or 1/5,000-15,000 of ectopic pregnancies. A twin pregnancy in the residual horn has also been reported and its frequency is estimated as approximately one case in ten million pregnancies [7].

Case Report

A 25-year-old woman with suspected unicornuate uterus presented to our hospital suffering from infertility. She reported a history of an ectopic (right tube) pregnancy which was removed with laparotomy elsewhere. Attentive gynecological examination detected the presence of a longitudinal non-obstructing vaginal septum, while the hysterosalpingography revealed a uterine malformation (Figure 1). Examination of the urinary system revealed normal kidneys and ureters. After patient consent was obtained, we initially performed mini-laparoscopy under general anesthesia (Figure 2), followed by mini-hysteroscopy (1.8 mm) (Figure 3). Then, diagnostic laparoscopy with a 10-mm scope placed intra-umbilically was performed with simultaneous hysteroscopy (5-mm lens-based hysteroscope; Olympus). Laparoscopically (Figure 4) and hysteroscopically (Figure 5) the existence of a unicornuate uterus was confirmed. Endometriosis of Stage I was also observed. The presence of one cervix with a normal cervical canal was detected, while exploration of the uterine cavity showed only one tubal ostium and atrophic endometrium. By utilization of the vaginal valves wide exposure of the vagina was possible and complete excision of the septum was performed using scissors. The operation was completed in 35 minutes and no vaginal suture was needed. Postoperative examination of the vagina two months later showed that the septum was excised completely.

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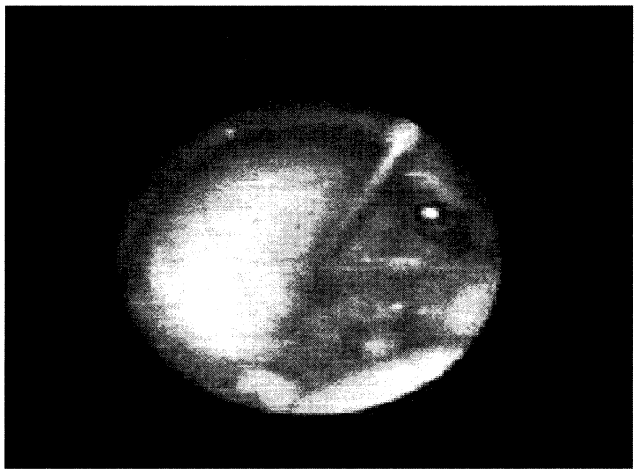

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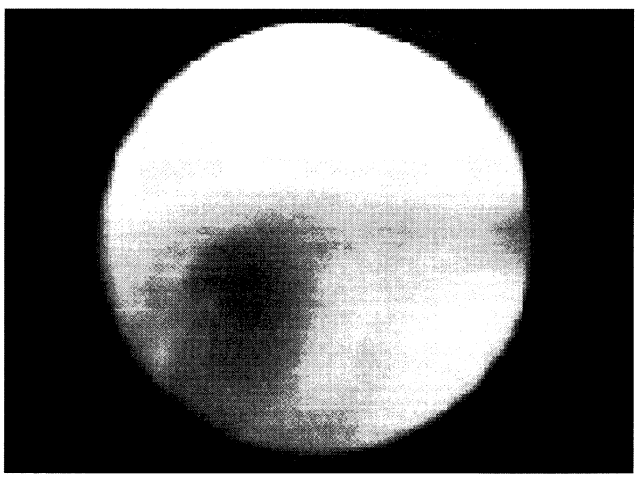

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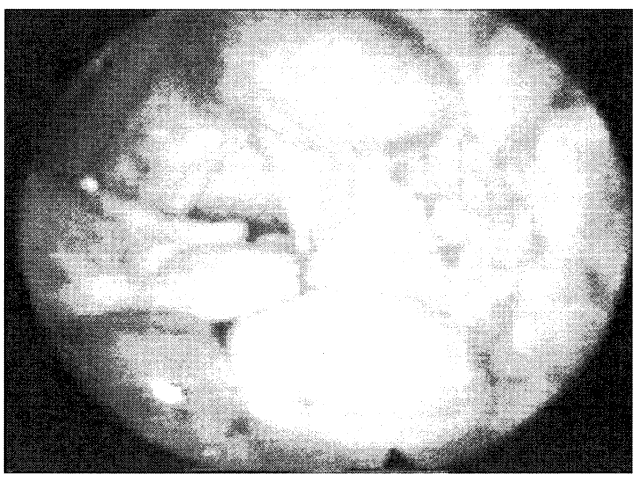

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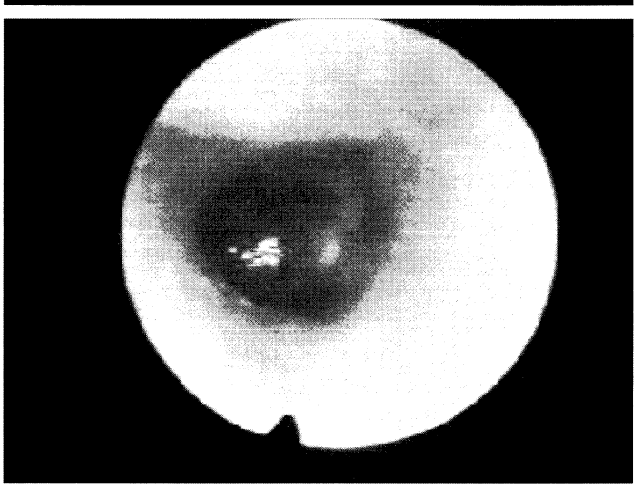

Figure 1. — Hysterosalpingography of an unicornuate uterus.
Figure 2. — Mini-laparoscopy.
Figure 3. — Mini-hysteroscopy.
Figure 4. — Laparoscopy.
Figure 5. — Hysteroscopy.

Diagnosis

Diagnosis of the above congenital uterine anomalies is usually completed during the examination procedure of a subfertile couple with recurrent pregnancy loss. The presence can also be established incidentally during a laparotomy, laparoscopy or caesarean section.

Considering diagnostic frameworks, intravenous pelvioradiography and ultrasound examination have to be performed, while transvaginal ultrasonography is very useful in the diagnosis of the rudimentary horn.

Unfortunately, the assessment of congenital uterine anomalies demands invasive techniques. Recently a non-interventional method has also been developed. This procedure, called three-dimensional ultrasound suggested by Raga [8] can generate high quality translucent images of the uterine cavity. Other diagnostic procedures are hysterosalpingography, laparoscopy, hysteroscopy and magnetic resonance imaging (MRI). Transvaginal hysterosonographic evaluation is also a new procedure that has been developed by Salle [9]. Ultrasound investigation

Table 1. — *Classification according to the American Fertility Society [3].*

Classification	Anomaly
Class I (Segmental-Mullerian Agenesis-Hypoplasia)	a. vaginal b. cervical c. fundal d. tubal e. combined
Class II (Unicornuate)	a. communicating b. noncommunicating c. no cavity d. no cornu
Class III (Didelphys)	Didelphys
Class IV (Bicornuate)	a. complete b. partial
Class V (Septate)	a. complete b. partial
Class VI (Arcuate)	
Class VII (DES Drug related)	

Table 2. — *Classification of vaginal septa [16].*

Classification	
Class I	*Transverse* a. Obstructing b. Non obstructing
Class II	*Longitudinal* a. Obstructing b. Non obstructing
Class III	*Stenosis/Iatrogenic*

Table 3. — *Symptoms associated with unicornuate and uterus didelphys.*

Habitual abortions
Obstetrical complications (IUGR, premature labour, hemorrhage, malpresentations)
Pelvic pain
Pelvic mass
Irregular intra-uterine hemorrhage
Without symptoms
Dyspareunia
Ectopic pregnancy (in unicornuate uterus)
Hematometra (in unicornuate uterus)
Hypertension during pregnancy (in unicornuate uterus)

Table 4. — *Reproduction fecundity in unicornuate and uterus didelphys [12].*

	Unicornis	Didelphys
Total pregnancies	16	15
Early abortion	6 (37%)	3 (20%)
Ectopic pregnancy	0	1 (6%)
Late abortion (advanced pregnancy)	1 (6%)	1 (6%)
Preterm birth neonates	4 (25%)	8 (54%)
22-28 wks	1	3
28-37 wks	3	5
Term delivery	5 (31%)	3 (20%)
Living children	7 (43%)	6 (40%)

is useful in the assessment of pregnancy in the rudimentary horn of a unicornuate uterus before a dangerous rupture can occur. Sonographically, we can compare the reduced thickness of myometrium in serial ultrasound scans. Gagnon *et al.* [7] reported increased myometrial thickness combined with a reduction of pregnancy symptoms during selected meiosis of one twin presenting in the rudimentary horn. To establish the differential diagnosis between the presence of a blind vaginal pouch in uterus didelphys and common urethral-rectal-vaginal outlet (cloacal anomalies) associated with VACTERL syndrome, (vertebral anal atresia, cardiac, tracheoesophageal, renal and lung) where a complete or incomplete vaginal obstruction has developed and a hydrocolpos is created, further examinations are needed. These examinations are plain-radiography of the lumbosacral spine and MRI in order to exclude other associated spinal cord anomalies or myelodysplasias.

Carrington *et al.* [10] compared the MRI findings with the surgical findings of uterine congenital anomalies and postulated the superiority of MRI over other diagnostic methods. MRI can outline the external uterine perimeter, tissue characteristics of the septum and the type of uterine cavity and is also recommended in the assessment of congenital renal malformations.

Symptoms

The symptoms associated with unicornuate uterus and uterus didelphys are summarized in Table 3.

The etiology of the obstetric complications is not well defined. The predominant theory is the reduced volume of the uterine cavity. Many different trials have been reported in an attempt of explain why some women with

Table 5. — *Laparoscopic treatment of unicornuate uterus with rudimentary horn [22].*

Author	Year	Symptoms	Attachment to the uterus	GhRH agonist	Removal
Canis	1990	Dysmenorrhea	Not specified	3 months	Low transverse abdominal incision (2 cm)
Mais	1994	Dysmenorrhea	Fibrous band	6 months	Endoscopic stampler-morcellator
Nezhat	1994	Pelvic pain	Yes	None	Simultaneous hysteroscopy
Falcone	1995	Dysmenorrhea	Yes	None	Enlarging 10-mm trocar site
Amara	1997	Pelvic pain	Yes	None	Morcelation - Infraumbilical incision
Perrotin	1999	Painful pelvic mass	Fibrous band	None	Through appendicectomy scar (2 cm)

Table 6. — *Experience of University of Cleveland (1983-1998) in the treatment of uterus didelphys [27]..*

Symptoms	Surgical procedure	Diagnosis	Outcome
Dysmenorrhea-Pelvic mass	Excision of longitudinal vaginal septum	Didelphys uterus-longitudinal vaginal septum	Resolution of symptoms
Malodorous vaginal discharge-Salpingitis	Excision of longitudinal vaginal septum-Laparoscopy	Left incomplete elongated vaginal septum	Resolution of symptoms
Dysmenorrhea	Excision of L-longitudinal vaginal septum, L-Hemisterectomy	Elongated vaginal septum-Didelphys-Endometriosis	Full term pregnancy
Lower abdominal pain-pelvic pain	Excision of longitudinal vaginal septum-Laparoscopy	Complete R long vaginal septum-Didelphys- Hydrosalpinx	Full term pregnancy
Pelvic pain	R-Fimbrioplasty-Laparoscopy, Resection of longitudinal vaginal septum	Didelphys-Hematosalpinx & Hematocolpos-longitudinal vaginal septum-Adhesions	Resolution of symptoms

congenital uterine anomalies lose their reproductive fecundity, while others with the same problem experience normal pregnancies. Possible reasons considered are the increased intra-uterine pressure associated with cervical incompetence, insufficient endometrial blood flow due to the presence of a septum, local vascular insufficiency and insufficient blood flow to the uterus. Immunological factors should also be excluded.

It is generally accepted that congenital uterine malformations are associated with preterm labour and miscarriage in late pregnancy [11]. Indeed, an analysis by Raga *et al.* [12] showed that 21.6% of 342 pregnancies ended in early miscarriage suggesting that congenital uterine malformations create not only space problems but also local defects interrupting normal fetal growth after implantation (Table 4). It seems logical to assume that embryo implantation on or close to the septum is associated with an abnormal obstetrical outcome and early abortion.

Hematometra is rare in the rudimentary horn of the unicornuate uterus, as the horn usually has no functional endometrium.

Patients with unicornuate uterus present with a greater frequency of endometriosis than other patients who present different types of congenital uterine anomalies [13]. The possible explanation is the presence of functional endometrium in the rudimentary horn, which does not communicate with the uterus.

Ectopic pregnancy is also common in unicornuate uterus. In a published trial by Heinomen [14], 34 women with unicornuate uterus achieved 93 pregnancies. Twenty of these pregancies were ectopic (22%). The accumulated percentage of ectopic pregnancy in all attended gestations at that same hospital for the same time period was 2.2%. Increased frequency of ectopic pregnancy has not been reported in other congenital uterine anomalies (1-2%). Pregnancy in the rudimentary horn or in its tube may occur due to sperm migration through the abdominal cavity. When rupture occurs, usually in the 10^{th}-15^{th} week of gestation, an emergency situation arises with internal hemorrhage and threat to the woman's life. In the same study three cases of placenta accreta are also mentioned [14]. This probably occurred because the myometrial wall was extremely thin and the embryo implantation in the non-functional endometrium was in the majority of cases pathological. The same study reported increased cases of pregnancy-induced hypertension in patients where unicornuate uterus was combined with unilateral renal agenesis.

The pregnancy rate in the residual cornu, as mentioned, is extremely rare [7]; 80-85% of pregnancies grew in a non-communicating rudimentary horn. In these cases, pregnancy occurs due to intraperitoneal sperm migration or migration of inseminated ovum and this aspect is enhanced by the fact that the corpus luteum is present on the opposite side. In contrast to tubal ectopic pregnancies, rudimentary horn gestation is often not detected except during the second trimester. The duration of pregnancy depends on the muscular wall thickness as well as on the myometrial efficiency to hypertrophy and dilatation. The average gestational period is 21 weeks, but it can range from 5 to 35 weeks. In some patients diagnosis is postulated after rupture and intraperitoneal hemorrhage. Maternal mortality reaches 5% and embryo loss 95%, even if, in rare cases, embryo survival has been reported.

In uterus didelphys many patients present with abdominal pain which starts immediately after menarche due to the development of hematacolpos. The right side of the vagina is affected twice as often as the left side [15]. Large fistulas allow the drainage of menstrual blood from the obstructed part of the vagina and therefore may delay the onset of symptoms.

The longitudinal vaginal septum composes 12% of vaginal malformations. The patient continues to bleed despite tampon placement and experiences dyspareunia. It is associated with uterine developmental anomalies and especially with didelphys uterus. From an obstetrical point of view, this kind of anomaly can cause dystocia and spotting after labour [16].

A transverse vaginal septum is postulated clinically by failure of tampon use, or by septal injury that occurs during sexual intercourse. In rare cases, fever may emerge or symptoms of ascending pelvic inflammation. If the transverse septum is complete, then it causes amenorrhea and pain during puberty. Endometriosis is the sequelae of vaginal obstruction. In puberty, this clinical entity presents

with hydrocolpos and in addition hydroureter and hydronephrosis. While in rare cases the tumor presses the inferior vena cava causing cardiorespiratory insufficiency.

Surgical reconstruction techniques of unicornuate uterus & uterus didelphys

Uterine developmental anomalies are managed by different surgical techniques which have replaced laparotomy with perfect results in regard to reproductive fecundity.

It is generally accepted that the non-communicating rudimentary horn of unicornuate uterus with functional endometrium has to be removed [14]. Opinions are divided when it is communicating, or does not have a functional endometrium. It is not well proven if the pregnancy course has a better outcome when the rudimentary horn has been removed. Other trials support the view that resection of the horn has better results in uterine cavity dilatation during pregnancy [17]. However many authors suggest that the rudimentary horn must be removed for prevention of complications such as ectopic pregnancy, endometriosis, torsion and malignancies [18, 19].

Therapeutic treatment options of unicornuate uterus can be summarized as follows:

1) Laparoscopic removal of rudimentary horn.

2) Rudimentary horn resection and removal by laparotomy.

3) Laparoscopic removal of rudimentary horn and adhesiolysis.

4) Laparoscopic removal of rudimentary horn and removal of endometriomas.

5) Selected reduction in twin pregnancy of the embryo within the rudimentary horn under ultrasound guidance [7].

6) Juxtaposition of a remaining fallopian tube to the contralateral ovary, after previous surgical procedures (oophorectomy) in unicornuate uterus [20].

7) Hysteroscopic re-communication between unicornuate uterus and rudimentary horn cavity with resectoscope [21].

8) In co-existence of unicornuate uterus and transverse septum, septum resection is performed as well as laparoscopy for endometriosis.

9) In co-existence of unicornuate uterus and longitudinal septum, septum resection is performed as well as laparoscopy for endometriosis.

1st surgical option: Laparoscopic resection of rudimentary horn.

Initial management consists of ligation of the round ligament after the ureteric course has been well defined. Then, removal of the ipsilateral tube without disruption of the blood supply to the ovary is followed by the ligation of the utero-ovarian ligament and uterine artery. In the international literature various techniques have been reported regarding procedure tools: scissors, bipolar diathermy, staplers, etc. When the rudimentary horn is firmly attached to the uterus, then ligation of the uterine artery is performed at its ascending course close to the rudimentary horn.

The most difficult part of the surgical procedure is when the rudimentary horn is firmly attached to the unicornuate uterus. It is quite vascular and caution is needed in order to avoid deep dissection into the myometrium of the remaining unicornuate uterus. At the end of the procedure a few sutures might be necessary in order to reapproximate the myometrium. The specimen can be removed either by using a morcellator, or by surgical extension of a suprapubic trocar incision of 10 mm, or through the pouch of Douglas.

Since the first laparoscopic removal of a rudimentary horn reported by Canis *et al.* [18], this kind of surgical procedure has become choice therapy for such Mullerian dysgenesis. The international literature presents nine cases of laparoscopic removal of remnant mullerian ducts, some of which are presented in Table 5 [22].

It is obvious that these operations need adequate equipment and experienced surgeons because of the increased risk of complications such as ureter damage, especially when endometriosis, or duplicated ureter are present. When endometriosis is present gonadotropin releasing hormone agonist treatment is recommended pre-surgically [18].

2nd surgical option: Laparoscopic removal of the rudimentary horn, endometriosis therapy and dissection of the perpendicular or transverse vaginal septum.

In this case, the previously suggested therapy for the rudimentary horn is followed along with a laparoscopic procedure for endometriosis and removal of the transverse or longitudinal vaginal septum.

a) Surgical therapy of transverse vaginal septum

Initially a Foley catheter is inserted in the bladder for verification of its anatomic position and then a needle is inserted inside the protruding part of the hematocolpos in order to drain the old blood. After that, the septum is excised with cautery or scissors until there is access to the cavity behind the septum. The whole thickness of the septum is grasped with an Allis forcep and is removed up to the mucinous membrane. Then, the mucinous membrane is sutured above and below the removed septum with absorbable sutures. A dilator is left in situ for six weeks. If the septum is high and quite thick then the removal is difficult. In such cases, various other approaches are needed in order to have clear and well-defined borders, such as finger application in the rectum, cervical palpation, laparoscopic evaluation, etc.

In rare cases, when the margins are not clear, it is necessary to perform a laparotomy. In this condition, a guide is inserted, penetrates the fundus of the uterine body, comes through the cervix and then comes out through the septum.

b) Surgical treatment of longitudinal vaginal septum

Initially a Foley's catheter is applied and then a needle is inserted inside the dilated vagina in order to drain the hematocolpos. The resectoscopic electrode is then inserted inside the obstructed vagina following the same direction of the needle. Then the septum is removed and the vaginal mucinous membrane seated under the septum is sutured in the same way as previously described for the

transverse septum. Failure of reconstructing the membrane may cause re-appearance of a new incomplete septum.

3rd surgical option: Hysteroscopic removal under ultrasound guidance [24].

Excision of the vaginal septum can be accomplished hysteroscopically [23]. In the international literature, hysteroscopic removal has been described under ultrasound guidance. The traditional techniques of hematocolopos drainage using small scissors or scalpels for septum removal and then anastomosis of the vaginal mucosa, is often difficult to perform in young women because of the small size of the vagina and the frequently upper location of the septum.

Hysteroscopically, under general anesthesia a 7.6 mm resectoscope is inserted into the vagina, without disruption of the hymen. The vagina is distended with continuous irrigation of sorbitol and mannitol at a ratio of 5:1. The bag of fluid is placed 100 cm above the patient's body. Under ultrasound guidance the distinction between the dilated vagina with the infused liquid and the haematocolpos becomes possible. Then the resectoscope is directed towards the septum. Under ultrasound guidance an incision is performed with the use of a straight and angled loop.

4th surgical option: Selected reduction in twin pregnancy under ultrasound guidance [7].

Because this condition jeopardized the life of both the mother and the fetuses, the authors decided on conservative therapy with selective termination. A surgical procedure with possible partial hysterectomy was excluded because of the greater risk of haemorrhage, abortion and preterm labour.

Selective termination (fetocide) took place in the rudimentary horn followed by amniocentesis of the remaining twin. The pregnancy continued normally until 36 weeks and then a cesarean section was performed due to intrauterine growth retardation. A male infant of 2,600 g was born.

5th surgical option: Remaining fallopian tube juxtaposition to the contralateral ovary.

Kennard *et al.* [20] described a case of a woman who was missing an ovary from the one side and the contralateral tube because of previous operations.

In that case, after laparotomy, the remaining fallopian tube was juxtaposed close to the ovary by suturing it to the posterior uterine wall with a single 4.0 Dexon suture. The reason why the authors preferred this method was that even though intraperitoneal migration of the ovum from the remaining ovary to the opposite tube could allow pregnancy, the likelihood of this event is unknown.

6th surgical option: Hysteroscopic treatment of unicornuate uterus with rudimentary horn [21].

Nogueira et al selected this method because classical surgical therapy, consisting of removal of the rudimentary horn leads to the formation of adhesions, endometriosis and subfertility problems. Furthermore, the problem of restricted dimensions of the cavity of the unicornuate uterus still exists. This method is applied in unicornuate uterus with a non-communicating rudimentary horn. (Class IIA). This operation consists of the opening of the arched wall using a hook-shaped electrode, thus permitting communication between the two cavities and drainage of the hematometra. There was a complete relief of symptoms, and the following diagnostic hysteroscopy one month after surgery showed a single cavity.

The earliest case of uterus didelphys with unilateral hematocolpos was reported by Wilson in 1925 [25].

If uterus didelphys and perpendicular vaginal septum coexist, the septum must be removed in any case of obstruction, dyspareunia or subfertility, because sexual intercourse may occur in the part of the vagina contralateral to the ovulation side.

Surgical therapy of uterus didelphys consists of resection of the vaginal septum in a single procedure, as incising the septum first and resecting two or three months later as previously recommended could lead to subsequent pyocolpos following spontaneous closure. Simultaneously with septum resection, laparoscopic treatment of coexisting endometriosis and adhesiolysis can be accomplished. In some cases, in spite of septum resection, delayed surgery could lead to hemi-hysterectomy due to continuous retrograde menstruation causing hematometra, hematosalpinx and pelvic adhesions.

In the past, some authors suggested prophylactic cervical cerclage even though cervical incompetence was undefined [17], but the consensus is that cerclage is not necessary if cervical incompetence has not been confirmed. Infrequently, plastic surgery may be needed such as the Strassman metroplasty for didelphys uterus and concerns mainly the uterine fundus [26].

Therapeutic options of uterus didelphys reconstruction are summarized as follows:

– Vaginal septum resection using classical instruments and laparoscopically treated endometriosis therapy.

– Vaginal septum resection using resectoscope under ultrasound guidance.

– Rarely, the Strassman technique for reconstruction of didelphys uterus.

– Laparoscopic hemi-hysterectomy.

– Laparoscopically assisted vaginal metroplasty [27].

The experience of the University of Cleveland in the treatment of uterus didelphys is mentioned in Table 6 [28].

Regarding uterine malformations, in the Obstetrical and Gynecological Department of the University Hospital of Ioannina, reconstruction of bicornuate uterus is also accomplished preferring Strassman's technique modified by Kaskarelis. Until today, this operation has been applied in 18 cases with excellent outcomes.

Initially a vertical incision on the uterus is done in the middle incision of the septum. Then an intrauterine device is applied and uterine suturing is accomplished with two continuous sutures of figure of eight. Finally, if two cervices exist fixation is completed vaginally [29].

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