Acellular porcine small intestinal submucosa graft for cervicovaginal reconstruction in eight patients with malformation of the uterine cervix

Jing-Xin Ding, Xiao-Jun Chen, Xu-yin Zhang, Ying Zhang, and Ke-Qin Hua*

Department of Gynecology, The Obstetrics and Gynecology Hospital of Fudan University, 419 Fang-Xie Road, Shanghai 200011, P.R. China

*Correspondence address. Tel: + 86-15921515900; Fax: + 86-21-63455090; E-mail: huakeqin@126.com

Submitted on May 1, 2013; resubmitted on December 15, 2013; accepted on December 20, 2013

STUDY QUESTION: Can surgical reconstruction of the cervix and vagina in patients be achieved using an acellular porcine small intestinal submucosa (SIS) graft?

SUMMARY ANSWER: Our experiences of combined laparoscopic and vaginal cervicovaginal reconstruction using an SIS graft in eight patients were positive, with successful reconstruction and no complications, cervical stenosis or vaginal stenosis.

WHAT IS KNOWN ALREADY: In patients with agenesis and dysgenesis of the uterine cervix and vagina, surgical reconstruction of the internal genitalia is a challenging problem for gynecologists. Hysterectomy with the creation of an artificial vagina was the treatment of choice in the 1990s. Recently, conservative management has been gradually adopted to avoid extirpation of the uterus, including the canalization techniques, the uterovaginal anastomosis and the reconstruction of cervical and vaginal agenesis with some autologous tissues.

STUDY DESIGN, SIZE, DURATION: This prospective observational study from January 2012 to March 2013 included 8 patients aged 10–18 years with malformation of the cervix (1 with cervical agenesis, 1 with a cervical body consisting of a fibrous band and 6 with obstruction of the cervical os) and vagina (4 with complete vaginal aplasia and 4 with a 1–3 cm long vaginal pouch) diagnosed by physical examination and magnetic resonance imaging.

PARTICIPANTS/MATERIALS, SETTING, METHODS: Eight patients underwent combined laparoscopic and vaginal cervicovaginal reconstruction using an SIS graft during the end of menstruation. A T-shaped intrauterine device connected with a 14-French Foley catheter was inserted into the uterine cavity to keep the newly created cervix patent, and then a permanent lower uterine cerclage was placed. Patients were assessed post-operatively at 1, 2, 4, 6, 12 and 15 months, and data on menstruation and the morphology of the neovagina and cervix were recorded.

MAIN RESULTS AND THE ROLE OF CHANCE: The mean ± SD age of the patients was 14.5 ± 2.8 (10–18) years. All patients had a history of cyclic abdominal pain, and the average delay in diagnosis was 4.5 ± 4.0 (0–12) months. One patient had a previous history of unsuccessful attempt at canalization and two post-operative hematometra drainages before referral. The mean operating time was 201 ± 67 (120–330) min, with a mean estimated blood loss of 157 ± 154 (30–500) ml. The first case was converted to laparotomy, and the others were successfully completed. None of the patients had a complication or required blood transfusion. All the patients showed resumption of menstruation. The patients were followed for 8 ± 4 (4–15) months, and no cervical or vaginal stenosis occurred in any of the cases.

LIMITATIONS, REASONS FOR CAUTION: The sample size of this study was small. A larger study that compared this method with previous techniques regarding the complication and success rates would increase the value of the study.

WIDER IMPLICATIONS OF THE FINDINGS: A combined laparoscopic and vaginal cervicovaginal reconstruction with an SIS graft is a potential alternative to the management of congenital agenesis and dysgenesis of uterine cervix and vagina.

STUDY FUNDING/COMPETING INTEREST(S): The work was supported by National Key Clinical Faculty Construction Program of China. No competing interests are declared.

Key words: cervicovaginal reconstruction / congenital agenesis of cervix / acellular porcine small intestinal submucosa graft / laparoscopy
Introduction

Congenital agenesis and dysgenesis of the uterine cervix and vagina in the presence of a functioning endometrium is a rare Müllerian duct anomaly, and the true incidence is somewhat difficult to determine (Fujimoto et al., 1997). Patients are found to lack a cervix (cervical agenesis) or to have one of three variants of cervical dysgenesis, which are characterized as having (i) an intact cervical body with obstruction of the cervical os, (ii) a cervical body consisting of a fibrous band or (iii) cervical fragmentation (Rock et al., 1995). This entity may be associated with an obstructive phenomenon after menarche that leads to cyclic abdominal pain and pelvic masses. Surgical reconstruction of the internal genitalia, restoration of menses and maintenance of a patent genital tract in patients with this congenital malformation are challenging problems for gynecologists. Hysterectomy with the creation of an artificial vagina was the treatment of choice in the 1990s (Fujimoto et al., 1997). With the rapid development of surgical techniques, conservative management has been gradually adopted to avoid extirpation of the uterus, including the canalization techniques (Fujimoto et al., 1997), the uterovaginal anastomosis (Defarges et al., 2001; Darai et al., 2009) and the reconstruction of cervical and vaginal agenesis with some autologous tissues, such as full-thickness skin grafts (Lee et al., 1999) and bladder mucosa (Bugmann et al., 2002). However, the canalization techniques and the uterovaginal anastomosis were associated with a certain rate of recurrence and complications, and the reconstruction of cervical and vaginal agenesis with autologous tissues might cause some extra trauma to the patients.

In the present study, we report the first use of an acellular porcine small intestinal submucosa (SIS) graft for reconstruction of the cervix and vagina. The SIS graft is one kind of extracellular matrix (ECM)-based collagen material, which is harvested from the submucosal layer of porcine small intestines. The graft is acellular and composed of non-cross-linked collagen (types I, III and V), glycosaminoglycans, proteoglycans and glycoproteins, and produces multiple growth factors (Soiderer et al., 2004). Studies have demonstrated tissue incorporation and neovascularization of an SIS graft in both animal hernias and subcutaneous models (Soiderer et al., 2004; Ayubi et al., 2008).

In this study, we present our successful experiences of combined laparoscopic and vaginal cervicovaginal reconstruction in eight patients with congenital agenesis and dysgenesis of the vagina and uterine cervix by using an SIS graft.

Materials and Methods

Patients

Between January 2012 and March 2013, a total of eight patients with agenesis and dysgenesis of the vagina and cervix underwent cervicovaginal reconstruction in the Obstetrics and Gynecology Hospital of Fudan University, Shanghai, China. The diagnosis was based upon symptoms, gynecologic examination, ultrasonography and magnetic resonance imaging before surgery. The clinical characteristics of the eight patients are listed in Table I. The average ± SD age of the subjects was 14.5 ± 2.8 (range 10–18) years. All patients had a history of cyclic abdominal pain, and the average delay in diagnosis from first symptoms was 4.5 ± 4.0 (0–12) months. One patient had a previous history of unsuccessful attempt at canalization and 2 post-operative hemorrhage drainages before referral. Four patients were given conservative therapy (oral contraceptive pills) for 4–20 months before referral. Four patients had complete vaginal aplasia and the other four patients had a 1–3 cm long vaginal pouch. The patients all had normal secondary sexual characteristics, and

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age (years)</th>
<th>Delay in diagnosis (months)</th>
<th>Previous surgery</th>
<th>Length of vagina before surgery (cm)</th>
<th>Upper genital tract malformations</th>
<th>Urogenital anomaly</th>
<th>Type of cervical malformation</th>
<th>Associated upper genital tract lesions</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>17</td>
<td>6</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>Cervical agenesis</td>
<td>Ovarian endometrioma; pelvic endometriosis; haematosalpinx; pelvic adhesions</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>17</td>
<td>6</td>
<td>0</td>
<td>3</td>
<td>Uterus didelphys</td>
<td>0</td>
<td>Cervical body consisting of a fibrous band</td>
<td>Adenomyosis; pelvic endometriosis</td>
</tr>
<tr>
<td>3</td>
<td>18</td>
<td>12</td>
<td>0</td>
<td>0</td>
<td>Unicorneate uterus</td>
<td>0</td>
<td>Obstruction of the cervical os</td>
<td>Pelvic endometriosis</td>
</tr>
<tr>
<td>4</td>
<td>15</td>
<td>6</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>Obstruction of the cervical os</td>
<td>Pelvic adhesions</td>
</tr>
<tr>
<td>5</td>
<td>13</td>
<td>4</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>Obstruction of the cervical os</td>
<td>Haematosalpinx; pelvic adhesions</td>
</tr>
<tr>
<td>6</td>
<td>14</td>
<td>0</td>
<td>1</td>
<td>3</td>
<td>0</td>
<td>0</td>
<td>Obstruction of the cervical os</td>
<td>Pelvic adhesions</td>
</tr>
<tr>
<td>7</td>
<td>10</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>Horseshoe kidney</td>
<td>Obstruction of the cervical os</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>12</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>Obstruction of the cervical os</td>
<td></td>
</tr>
</tbody>
</table>
the levels of sex steroids and gonadotrophin in plasma confirmed the normal functioning ovaries in all subjects. Associated upper genital tract malformations and pathologies were found in 2 (25%) and 6 (75%) women, respectively. All patients also underwent ultrasound examination of the kidneys or intravenous pyelography, and horseshoe kidney was found in one patient. During surgery, the type of cervical malformation was confirmed, showing that one patient had cervical agenesis, one had a cervical body consisting of a fibrous band and the other six patients had obstruction of the cervical os. Operative time, estimated blood loss, length of hospital stay, length of follow up, the resumption of menstruation and condition of the neovagina and the new cervix were recorded. This study was approved by the Obstetrics and Gynecology Hospital of Fudan University Institutional Review Board and informed consent from the guardians of all the patients was obtained before surgery.

Surgical procedures

Before surgery, the patient received antibiotic prophylaxis (cefuroxime 0.75 g and metronidazole 500 mg i.v. 0.5 h before surgery and repeated every 8 h for 48 h) and a bowel enema. All the patients underwent combined laparoscopic and vaginal cervicovaginal reconstruction at the end of menstruation. The same surgeon (K.H.) and surgical team performed all eight procedures. The procedure was performed under general anesthesia and in a lithotomy position allowing the perineal approaches. An incision was made in the peritoneum over the bladder and dissection of the anterior space between the bladder and the uterus was performed under laparoscopy. Following this, a transverse incision at the vaginal introitus or at the top of the vaginal pouch was made, and a canal was created between the bladder and the rectum using sharp and blunt dissection along the anatomic vaginal route to expose the lower end of the uterus. An incision of 1.5 cm diameter was made on the atretic tissue until the uterine cavity was reached. The same surgeon (K.H.) and surgical team performed all eight procedures. The procedure was successfully completed except that the first case was converted to laparotomy because of difficulties with the laparoscopic technique and lack of experience. Associated procedures performed for the additional upper genital tract pathologies in six patients (75%) were as follows: coagulation of pelvic endometriosis in three cases (37.5%), adhesiolsis in three cases (37.5%), ovarian cystectomy in one case (12.5%) and salpingostomy in two cases (25%).

The perioperative data for the patients are shown in Table II. The operative procedure lasted 201 ± 67 (120–330) min. The estimated blood loss was 157 ± 154 (30–500) ml, with a post-operative hemoglobin decrease of 11 ± 11 (0–27) g/dl. The hospital stay post-surgery was 11 ± 7 (7–14) days. None of the patients had complications or required a blood transfusion. The mean total cost was $5690 ± 1443, depending on cost of the graft, surgical devices and length of hospital stay, but was not correlated with the operating time in China.

Five of the eight patients had positive results in the vaginal discharge culture. The bacteria were Staphylococcus aureus in one patient, Escherichia coli in two patients, Enterobacter cloacae in one patient and Morganella morgani in one patient. Most of these belong to the normal vaginal flora, except for Staphylococcus aureus, which was present due to nosocomial infection. This patient was treated according to the reported sensitivity pattern. There was no serious infection leading to shedding of the graft in any of our patients. All patients completed their post-operative medical visits. The average vaginal length at 1 month was 7.8 ± 1.0 (range 7–10) cm. All patients showed resumption of menstruation. The patients were followed for a mean of 8 ± 4 (4–15) months. During the follow-up, cervical stenosis did not occur in any of the cases, and haematometra was not found on serial ultrasonography. All patients were told to wear the mould for >12 months. Vaginal stenosis did not occur in any of the patients and the neovagina all had good morphological features. Figure 2 shows the normal healing of the vaginal graft and the condition of the new cervix 4 months post-operatively.

Discussion

In this prospective observational study, we presented our successful experiences of combined laparoscopic and vaginal cervicovaginal reconstruction in eight patients with congenital agenesis and dysgenesis of the cervix and sutured to the uterine cavity with separate stitches of 2/0 PDS II delayed absorbable material, and the lower end was sutured to the high vaginal or vestibular mucosa.

On post-operative Day 7, the vaginal mould was removed and the condition of the neovagina and cervix was assessed. A culture of the vaginal discharge was prepared at the same time. After vaginal irrigation with 10% povidone-iodine, a condom-covered plastic mould was placed in the vagina. Patients were instructed to wear the vaginal mould for 12 months in succession and then to wear the mould overnight every 3 days. Patients were assessed post-operatively at 1, 2, 4, 6, 12 and 15 months, and the menstrual pattern and the morphological results of the neovagina were recorded.

Statistical analysis

Descriptive statistics are used. Data are expressed as the mean ± SD and minimum and maximum values.

Results

The procedures were successfully completed except that the first case was converted to laparotomy because of difficulties with the laparoscopic technique and lack of experience. Associated procedures performed for the additional upper genital tract pathologies in six patients (75%) were as follows: coagulation of pelvic endometriosis in three cases (37.5%), adhesiolsis in three cases (37.5%), ovarian cystectomy in one case (12.5%) and salpingostomy in two cases (25%).

The perioperative data for the patients are shown in Table II. The operative procedure lasted 201 ± 67 (120–330) min. The estimated blood loss was 157 ± 154 (30–500) ml, with a post-operative hemoglobin decrease of 11 ± 11 (0–27) g/dl. The hospital stay post-surgery was 11 ± 7 (7–14) days. None of the patients had complications or required a blood transfusion. The mean total cost was $5690 ± 1443, depending on cost of the graft, surgical devices and length of hospital stay, but was not correlated with the operating time in China.

Five of the eight patients had positive results in the vaginal discharge culture. The bacteria were Staphylococcus aureus in one patient, Escherichia coli in two patients, Enterobacter cloacae in one patient and Morganella morgani in one patient. Most of these belong to the normal vaginal flora, except for Staphylococcus aureus, which was present due to nosocomial infection. This patient was treated according to the reported sensitivity pattern. There was no serious infection leading to shedding of the graft in any of our patients. All patients completed their post-operative medical visits. The average vaginal length at 1 month was 7.8 ± 1.0 (range 7–10) cm. All patients showed resumption of menstruation. The patients were followed for a mean of 8 ± 4 (4–15) months. During the follow-up, cervical stenosis did not occur in any of the cases, and haematometra was not found on serial ultrasonography. All patients were told to wear the mould for >12 months. Vaginal stenosis did not occur in any of the patients and the neovagina all had good morphological features. Figure 2 shows the normal healing of the vaginal graft and the condition of the new cervix 4 months post-operatively.
vagina and uterine cervix by using an SIS graft. The results were satisfying compared with previous results. There were no complications, and no cervical or vaginal stenosis. Congenital agenesis and dysgenesis of the uterine cervix and vagina is an extremely rare malformation and its management remains a challenging problem for gynecologists. The preservation of fertility is difficult in these patients. Cervical drilling has been reported to be associated with deadly peritonitis (Maciulla et al., 1978). Niver et al. (1980) studied three cases of congenital atresia of the uterine cervix and vagina in 1980. All three cases ended with total hysterectomy because of uncontrollable ascending infection from the neovagina. Thus, it was concluded that total hysterectomy with the McIndoe procedure is the appropriate treatment for these patients.

Recently, the conservative management has been gradually adopted to preserve the uterus. In the early attempts of canalization techniques, there was a high risk (40–60%) of secondary stenosis of the cervix (Fujimoto et al., 1997).

Uterovaginal anastomosis was preferred to canalization techniques, as it was associated with a much lower rate of recurrence and complications (Deffarges et al., 2001; Gurbuz et al., 2005; Kriplani et al., 2012). Kriplani et al. (2012) reported laparoscopic-assisted uterovaginal anastomosis in 14 patients with cervical agenesis and partial or complete vaginal aplasia. One patient (7.1%) had a genital infection and re-stenosis, and underwent hysterectomy. Deffarges et al. (2001) reported 18 cases that underwent uterovaginal anastomosis, and secondary stenosis occurred in 1 patient (5.6%) who finally required canalization.

Fedele et al. (2008) reported laparoscopically assisted uterovestibular anastomosis in 12 consecutive patients with uterine cervix atresia and vaginal aplasia. The neovagina was created by the traction exerted on the uterine body by the cardinal ligaments and ovarian vessels that had been stretched caudally by the anastomosis, with no need for vaginal molding after surgery. It was reported that all of the women experienced regular menstrual cycles after the operation, and that the mean length of the neovagina at 6-month follow-up evaluation was 6 cm. However, 9 of the 12 patients had a vaginal length of >2 cm (2–6 cm) before surgery, implying that the stretching effect on the length of the neovagina is limited, and that there might be some cases with an unsatisfactory vaginal length. Thus, in patients with a vagina <3 cm long, the use of exogenous material to reconstruct the neovagina might be recommended.

New materials and methods have been used to reconstruct the cervix and vagina, such as full-thickness skin grafts (Lee et al., 1999), bladder mucosa (Bugmann et al., 2002), prefabricated bilateral pudendal thigh flaps (Gürlek et al., 2008) and free microvascular transfer of the appendix and ascending colon (Hou et al., 2008). However, the use of the bowel or bladder mucosa is associated with operative morbidity. The presence of a scar, whether it is in a cosmetically acceptable location or not, remains one of the most compelling reasons for the decline in the number of patients who undergo the procedure with pudendal thigh flaps. A neovagina created with skin grafts maintains a keratinized epithelium with sebaceous glands and hairs many years after surgery, which is an obvious disadvantage (Barberini et al., 1992).

Heterologous tissue, such as amniotic membrane, has also been used to reconstruct the neovagina. Chakravarty et al. (2000) reported reconstructive surgery for congenital cervicovaginal atresia in 18 patients. Amniotic membrane was used to line the neovagina and a cervical stent was used to prevent cervical stenosis. However, the amniotic membrane was not sterile with the possibility of transmitting disease. In addition, the amniotic membrane was not resistant to infection, which possibly could have induced shedding of the membrane.

SIS is derived from the submucosal layer of pig small intestine that has been mechanically separated from the adjoining intestinal layers. The SIS is de-cellularized, biocompatible and does not produce an immunologic

### Table II Perioperative data for the patients who underwent combined laparoscopic and vaginal cervicovaginal reconstruction by using acellular porcine SIS graft.

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Mean ± SD or n (range or percentage)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Operating time (min)</td>
<td>201 ± 67 (120–330)</td>
</tr>
<tr>
<td>Estimated blood loss (ml)</td>
<td>157 ± 154 (30–500)</td>
</tr>
<tr>
<td>Hemoglobin change (g/dl)</td>
<td>11 ± 11 (0–27)</td>
</tr>
<tr>
<td>Hospital stay post-surgery (days)</td>
<td>11 ± 2 (7–14)</td>
</tr>
<tr>
<td>Complications (n)</td>
<td>0</td>
</tr>
<tr>
<td>Transfusion (n)</td>
<td>0</td>
</tr>
<tr>
<td>Conversion to laparotomy</td>
<td>1</td>
</tr>
<tr>
<td>Associated procedures for upper genital tract lesions</td>
<td></td>
</tr>
<tr>
<td>Ovarian cystectomy</td>
<td>1 (12.5%)</td>
</tr>
<tr>
<td>Coagulation of pelvic endometriosis</td>
<td>3 (37.5%)</td>
</tr>
<tr>
<td>Salpingostomy</td>
<td>2 (25%)</td>
</tr>
<tr>
<td>Adhesiolysis</td>
<td>3 (37.5%)</td>
</tr>
<tr>
<td>None</td>
<td>2 (25%)</td>
</tr>
<tr>
<td>Total cost ($)</td>
<td>5690 ± 1443</td>
</tr>
<tr>
<td>Median vaginal length at 1 month</td>
<td>7.8 ± 1.0 (7–10)</td>
</tr>
<tr>
<td>Follow-up (months)</td>
<td>8 ± 4 (4–15)</td>
</tr>
<tr>
<td>Resumption of menstruation</td>
<td>8 (100%)</td>
</tr>
<tr>
<td>Vaginal stenosis</td>
<td>0</td>
</tr>
<tr>
<td>Stricture of the cervix</td>
<td>0</td>
</tr>
</tbody>
</table>

Figure 2 Appearance of the neovagina and cervix under colposcopy at 4 months after surgery. Original magnification × 7.
Intestinal submucosa graft for cervicovaginal defects

The SIS graft is also readily available and is completely biodegradable. It has been used in a variety of applications including body wall repair (Badylak et al., 2001, 2002), vascular grafts (Lantz et al., 1990), dural replacement (Cobb et al., 1996), urinary bladder augmentation (Kropp et al., 1995), perineal hernia repair (Stoll et al., 2002) and others (Chen and Badylak, 2001, Rosen et al., 2002). This sterile, freeze-dried, non-permanent, acellular matrix graft made from the pig jejunum appears to promote the rapid ingrowth of surrounding tissue (Jankowski et al., 2004). Therefore, epithelialization of the new cervix and the neovagina is allowed to occur. The procedure and outcome of cervical reconstruction and vaginoplasty using an SIS was favorable in this study. None of the patients had complications or required a blood transfusion. The patients were followed for 8 ± 4 (4 – 15) months, and the morphologic results of the neovagina and new cervix were good. The vaginal length was 7.8 ± 1.0 (range 7 – 10) cm, meaning all the patients had a satisfying vaginal length.

The vagina is colonized by various micro-organisms. Therefore, graft infections are one of the most challenging issues in these surgeries. The SIS graft, one of the biological xenografts, was used in this study to create the anatomical cervix and to maintain the patency of the genital tract. The biological xenograft, including SIS graft, a component of which was the ECM, has been shown to be resistant to infection in contaminated cases, both in the animal models and clinical reports (Patton et al., 2007; Hiles et al., 2009). Early revascularization of the graft is thought to enhance resistance to infection and contamination (Menon et al., 2003). In our study, although some bacteria were found in the neovagina, there was no serious infection leading to shedding of the graft in any of our patients, and the neovagina and the new cervix both showed good morphology and function. These results are consistent with the previous studies that have demonstrated the safety of the use of the biological xenograft in the contaminated areas (Patton et al., 2007; Hiles et al., 2009).

In the cervicovaginal reconstruction in these patients, the technique that kept the neocervix patent by resisting the compressive forces of the uterine musculature was also very important. In the past, we used a 14-French Foley catheter which was retained in the uterine cavity. However, we found there was high defluxion rate, and the long catheter was inconvenient for the patient. In this study, a T-shaped IUD connected to a 14-French Foley catheter was inserted into the uterine cavity to keep the newly created cervix patent, and to allow the growth of epithelium lining the newly constructed reproductive tract. The IUD was taken out 4 – 6 months later with satisfying results. Menstruation resumed in all the patients, and cervical stenosis did not occur in any of the cases.

Among the eight patients in our study, one patient had complete cervical aplasia, one had a cervical body consisting of a fibrous band and the other six patients had obstruction of the cervical os. According to previous reports (Rock et al., 1995), the prognosis of surgery depends on the type of cervical malformation. The patients with cervical agenesis have a poorer prognosis than those with cervical dysgenesis. Thus, it is important to find the best material to line the endocervical canal and to have a good cervical stent. In the patient with complete cervical aplasia in this study, the SIS graft was carefully sewn around the uterine catheter and the lower uterine segment during the surgery to allow epithelialization of the endocervical canal. The uterine catheter was left in place for 6 months to allow the formation of a permanent fistulous tract. The result was satisfying. Menstruation remained and the cervical canal was patent after the T-shaped IUD connected to the catheter was taken out.

Limitations of the study are the small sample size and the lack of a control group, both of which are difficult to include in a study of a very rare condition.

In summary, abnormalities in Mullerian development may result in various urogenital anomalies. Hysterectomy can be avoided and the possibility for future fertility can be preserved in those patients with agenesis of the vagina and cervix and with a functional uterine endometrium. A combined laparoscopic and vaginal cervicovaginal reconstruction using an SIS graft is a potential alternative to the management of congenital agenesis and dysgenesis of uterine cervix and vagina.

Authors’ roles

K.H. designed the study, operated the patients and reviewed the manuscript. J.D. co-operated the patients, reviewed the reported cases, followed the patients, made the tables and figures and wrote the paper. X.C. designed the T-shaped IUD connected to the catheter, co-operated the patients, reviewed the literature and reviewed the manuscript. X. Zh. and Y. Zh. co-operated the patients, co-analysed the results and reviewed the manuscript.

Funding

The work was supported by National Key Clinical Faculty Construction Program of China.

Conflict of interest

The authors made no disclosures.

References


