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ORIGINAL RESEARCH

Surgical Management and Outcomes of Unilateral Genital Tract Obstruction with Ipsilateral Renal Anomaly Syndrome: A Retrospective Study

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Purpose: To systematically conduct a retrospective summary of the optimal surgical strategies and techniques for managing various types of unilateral genital tract obstruction with ipsilateral renal anomaly (UGTOIRA) syndrome, and to establish a comprehensive postoperative follow - up protocol.

Methods: The clinical data of patients diagnosed with UGTOIRA syndrome over the past 13 years, encompassing treatment protocols, follow-up assessments, and treatment outcomes, were retrospectively analyzed. Statistical analysis was performed utilizing the SPSS 26.0 statistical software package.

Results: All 68 patients underwent reproductive system surgery, with three additionally receiving urological procedures. The age at surgery ranged from 5 to 52 years, with a median of 15 years (interquartile range, 12–23 years). For Type I, the surgical approach consisted of vaginal oblique septectomy, yielding a 100% clinical cure rate (53/53). Type II-a and II-b were managed with vaginal oblique septectomy and combined vaginal oblique septectomy with ipsilateral cervical plasty, respectively, resulting in an overall 71.4% clinical cure rate (5/7) for Type II. Type III was treated with cervical plasty, achieving a 66.7% clinical cure rate (2/3). Types IV and V were addressed with unilateral hysterectomy and salpingectomy, respectively, both demonstrating 100% clinical cure rates (3/3 and 2/2). The overall cure and improvement rates among the 68 patients were 95.6% (65/68) and 4.4% (3/68), respectively. A one-stage surgical procedure alone was sufficient for cure in 56 cases (82.4%). Significant differences were observed in the distribution of surgical interventions across types and subtypes ($P < 0.001$), as well as in clinical cure rates among different types and subtypes ($P = 0.013$).

Conclusion: The optimal surgical approach, determined through precise classification of UGTOIRA syndrome, ensures a high clinical cure rate, while systematic postoperative follow-up facilitates the early detection and management of recurrent obstruction.

Keywords: UGTOIRA syndrome, uterine malformation, cervical malformation, vaginal malformation, renal agenesis, surgery

Introduction

Unilateral genital tract obstruction and ipsilateral renal agenesis (UGTOIRA) syndrome, also known as Herlyn–Werner–Wunderlich (HWW) syndrome, obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome, or oblique vaginal septum (OVS) syndrome, is a rare congenital urogenital anomaly, a syndrome of unilateral genital tract obstruction with different uterine malformations, and ipsilateral renal agenesis et.[1] The true incidence of UGTOIRA remains unknown. According to the literature, the incidence of obstructive Mullerian duct malformations is estimated to range from 0.1% to 3.8%.[2] Once a unilateral vaginal, cervical or uterine isthmus obstruction is diagnosed, early intervention is strongly recommended. The principal therapeutic approach involves complete excision of the OVS to restore vaginal patency, with timely surgical intervention serving to mitigate potential complications.[3–5] The preferred initial approach is typically single-stage vaginal septum resection combined with vaginoplasty; nevertheless, a two-stage surgical strategy may be more appropriate in cases involving infection or significant anatomical distortion.[6] When the

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OVS is thick, positioned in close proximity to the cervix, and accompanied by minimal or absent hematocolpos in the posterior cavity of the OVS, vaginal septectomy and vaginoplasty may not be technically feasible. In such complex cases, ipsilateral hysterectomy may be indicated.[7,8] Unilateral cervical atresia typically necessitates ipsilateral hysterectomy.[9–11] There is limited literature that systematically explores the surgical management plans for different types and subtypes of UGTOIRA syndrome.

This study presents a retrospective analysis of treatment modalities, follow-up protocols, and clinical outcomes in a case series of 68 patients diagnosed with UGTOIRA syndrome. The primary objective is to synthesize evidence-based surgical insights and best practices.

Materials and Methods

Study Design

This study constitutes a retrospective, descriptive analysis of routinely collected medical record data, encompassing treatment protocols, follow-up assessments, and clinical outcomes.

Study Population

The study cohort consisted of patients diagnosed with UGTOIRA syndrome who underwent surgical treatment at the Department of Obstetrics and Gynecology, Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology, between July 2012 and July 2025. The inclusion criteria were as follows: (1) patients who received surgical intervention for the relief of unilateral reproductive tract obstruction at our institution; (2) completion of preoperative genitourinary system segmental sequential ultrasound screening (SSUS); (3) definitive identification of uterine, cervical, vaginal, bilateral fallopian tube, ovarian, and pelvic abnormalities through laparoscopic exploration or laparotomy, combined with hysteroscopy or gynecological examination, supplemented by pathological diagnosis when available; and (4) attendance of at least one postoperative follow-up visit at our hospital. Patients who did not meet all four criteria concurrently were excluded from the study.

Classification of Female Genital Tract Congenital Anomalies

Clinically diagnosed uterine, cervical, and vaginal malformations were classified in accordance with the ESHRE/ESGE consensus statement on the classification of congenital anomalies of the female genital tract.[12] Furthermore, we have proposed a detailed classification standard for OVS.

Uterine anomalies are classified as follows: U2a: Partially septate uterus; U2b: Completely septate uterus; U3a: Partially bicorporeal uterus; U3b: Completely bicorporeal uterus; U3c: Bicorporeal septate uterus; U4a: Unilateral rudimentary horn with an isolated cavity lacking communication with the contralateral uterus.

Cervical anomalies are classified as follows: C0: Complete absence of ipsilateral cervical development, with an otherwise normal appearance; C1: Septate cervix; C2: Double cervix; C3a: Unilaterally hypoplastic cervix; C3b: Unilateral cervical os atresia; C3c: Partial hypoplasia of the unilateral cervix with a blind-ending structure.

Vaginal anomalies are classified as follows: V0: Complete agenesis of the ipsilateral vagina with normal external appearance; V2/OVS: Presence of an oblique vaginal septum; Low OVS: The lowest point of the oblique septum is located $\leq$3.0 cm from the vaginal orifice; Medium OVS: The lowest point of the oblique septum is situated between 3.0 cm and 6.0 cm from the vaginal orifice; High OVS: The lowest point of the oblique septum is positioned >6.0 cm from the vaginal orifice.

Typing of UGTOIRA Syndrome

The cases were classified into five types according to the anatomical site of obstruction as follows: (1) Type I is characterized by V2 with C1 or C2, which can be further divided into three subtypes: Type I-a (low OVS), Type I-b (medium OVS), and Type I-c (high OVS). (2) Type II is characterized by V2 with C3a or C3b, and it can be divided into two subtypes: Type II-a (C3aV2) and Type II-b (C3bV2). (3) Type III is characterized by C3b with V0. (4) Type IV is characterized by C3c with V0. And finally, (5) type V is characterized by U4a and C0 with V0.

Surgery, Follow-Up, and Clinical Outcomes

The first operation conducted in the Department of Obstetrics and Gynecology at Tongji Hospital, affiliated with Tongji Medical College of Huazhong University of Science and Technology, aimed to address reproductive tract malformations and served as the research index. The date of this procedure was designated as the baseline (time zero) for follow-up, while July 31, 2025—the conclusion of the study's data collection period—was defined as the final follow-up endpoint. The routine clinical follow-up included an inquiry into clinical symptoms, a gynecological examination, a reexamination of gynecological ultrasound, and a reoperation. The data for this retrospective study were extracted from the electronic medical records of the patients. At the conclusion of the follow-up period, the clinical outcomes were evaluated based on the following criteria: (1) symptoms associated with reproductive tract obstruction, (2) the status of surgical margins at the site of OVS and the cervix as assessed by gynecological examination, and (3) ultrasonographic findings indicating the presence or absence of reproductive tract obstruction. The criteria for clinical cure and improvement were the disappearance and remission in relevant symptoms and ultrasonic manifestations of reproductive tract obstruction, respectively.

Statistical Analyses

The statistical software package SPSS26.0 (IBM SPSS Statistics for Windows, 26.0 version, Armonk, NY, USA) was utilized for data analysis. The classified variables are represented as n (%), and the continuous variables are denoted as M (25%, 75%). All continuous variables were rounded to the nearest integer and percentages were rounded to one decimal place. Incidence was compared using the chi-square test. The p-value was reported with three decimal places after the decimal point. Differences were considered statistically significant when the p-value was <0.05.

Results

Baseline Characteristics

A total of 68 patients were enrolled in this study. Preoperative imaging diagnostics confirmed that all 68 patients (100%) with unilateral reproductive tract obstruction also demonstrated ipsilateral renal agenesis. Among these cases, one patient presented with a contralateral duplicated kidney. Additionally, four patients (5.9%) exhibited an ipsilateral dysplastic kidney located within the pelvic cavity, accompanied by a dilated and dysplastic ureter. Of these, three cases were associated with ectopic ureteral orifices or fistulous tracts extending into the posterior compartment of the OVS.

All patients underwent surgical intervention involving the reproductive system. Furthermore, three patients received urological management, including excision of the ipsilateral dysplastic pelvic kidney along with resection of the associated ureteral remnant cyst. Surgical intervention was necessitated by complications in 26.5% (18/68) of patients, encompassing endometriosis, pelvic adhesions, and pelvic inflammatory disease.

The median patient age was 15 (12–23) years, with ages ranging from 5 to 52 years. The median interval between diagnosis and surgery was 0 (0,0) months, with intervals ranging from 0 to 12 months. Six patients (6/68, 8.8%) had a history of surgery either in other hospitals or in the pediatric surgery department of our hospital. All patients were followed up for 1 to 144 months, with a median duration of 15 months (10–51 months).

Surgery, Follow-Up, and Clinical Outcomes

Type I (C1V2, C2V2) (n=53)

Among the 53 cases of Type I, three patients had a history of prior surgery. All 53 cases underwent vaginal oblique septectomy, including 20 cases (20/53, 37.8%) classified as Type I-a, 27 cases (27/53, 50.9%) as Type I-b, and 6 cases (6/53, 11.3%) as Type I-c. Among these patients, one underwent uterine septum resection, and another received a combined procedure of hysteroscopic uterine septum resection and cervical septum resection. Additionally, one patient underwent laparoscopic ipsilateral salpingectomy. Three cases were treated with ipsilateral salpingoplasty, and seven cases required surgical intervention for complications. A perimenopausal patient with Type I-a underwent vaginal oblique septectomy combined with total hysterectomy and bilateral salpingectomy. All 53 cases achieved clinical cure, resulting in an overall cure rate of 100% (53/53) for Type I.

Type I-a (Low OVS) (n=20)

Among the 20 patients diagnosed with Type I-a UGTOIRA syndrome, 18 patients (18/20) achieved clinical cure following a single vaginal oblique septum resection procedure, with one perimenopausal patient undergoing concurrent hysterectomy and bilateral salpingectomy during the same operation. One patient with left UGTOIRA syndrome had a history of conservative surgery for a right fallopian tube pregnancy by laparotomy and laparoscopic resection of the right fallopian tube for another right fallopian tube pregnancy before undergoing ultrasound-guided hysteroscopic oblique vaginal septostomy, vaginoplasty, laparoscopic release of pelvic adhesions, left salpingostomy, and resection of a lesion in the left ovary in our gynecology department. Another prepubescent patient, who had previously undergone high ligation of a bilateral inguinal hernia and aspiration of uterine cavity effusion in our department of pediatric surgery, was found during postoperative ultrasound follow-up after oblique vaginal septostomy to have a residual ureter on the same side with ectopic opening into the posterior space of the OVS. The patient subsequently underwent urological management three years and nine months after vaginal oblique septectomy due to vaginal leakage of urine, which involved the removal of an ipsilateral dysplastic pelvic kidney along with the associated ureteral remnant cyst that had opened into the vagina.

Type I-b (Medium OVS) (n=27)

All 27 patients with Type I-b UGTOIRA syndrome underwent vaginal oblique septectomy. Among them, 24 patients (24/27) achieved clinical recovery through a single-stage surgical procedure. Among the remaining three patients: the first patient had a prior history of laparoscopic pelvic adhesion lysis, excision of a left ovarian endometriosis cyst, and left salpingostomy performed at another institution before undergoing vaginal oblique septectomy at our center; the second patient developed vaginal purulent discharge and abscess formation in the ureteral remnant five months postoperatively, necessitating a secondary surgical procedure to remove the ureteral remnant with vaginal communication; the third patient experienced adhesion formation at the septal resection margin two months postoperatively, accompanied by purulent accumulation in the ipsilateral vagina, cervix, uterine cavity, and fallopian tube, necessitating a second surgical procedure to completely resect the residual vaginal septum and the ipsilateral fallopian tube.

Type I-c (High OVS) (n=6)

None of the six patients with Type I-c UGTOIRA syndrome had a prior history of surgical intervention. All six patients with Type I-c underwent vaginal oblique septectomy, and five (5/6) achieved clinical recovery through a single-stage surgical procedure. The remaining patient (1/6) required secondary surgery to completely excise the residual vaginal septum due to wound site suturing that occurred three months after the initial vaginal oblique septectomy.

Type II (C3aV2, C3bV2) (n=7)

Among the seven patients diagnosed with Type II UGTOIRA syndrome, none of whom had a prior history of surgery, six (6/7) underwent vaginal oblique septectomy. This included all five patients with Type II-a (5/5) and one of the two patients with Type II-b (1/2). Among these six patients, two underwent additional surgical procedures to address concurrent complications. Of the six patients who received surgical intervention, three achieved complete resolution of symptoms, while three demonstrated clinical improvement. The remaining patient, who had Type II-b (1/2), underwent a combined procedure of vaginal oblique septectomy and ipsilateral cervical plasty, resulting in clinical cure. Therefore, the overall clinical cure rate for Type II UGTOIRA syndrome in this cohort was 57.1% (4/7).

Type II-a (C3aV2) (n=5)

All five Type II-a patients underwent vaginal oblique septectomy (four by hysteroscopy), and two patients required surgery for complications. Three cases achieved a cure, and two cases demonstrated clinical improvement.

In one case, the patient underwent transvaginal oblique vaginal septectomy under ultrasonic guidance, along with laparoscopic excision of a left-sided endometriotic cyst, and removal of a salpingohematocele and pelvic hematocele. Three months postoperatively, contracture at the incision site of the OVS as well as hematoma formation posterior to the OVS, cervix, and uterine cavity were identified. The patient had remained free of surgical intervention for 10 years prior to presentation and had a history of infertility.

Another patient underwent three surgical procedures at our hospital, during which her clinical symptoms demonstrated minimal improvement. The patient had experienced dull pain in the lower right quadrant of the abdomen for three years, beginning at the onset of menarche when she was 13 years old. At the age of 16, she was diagnosed with right UGTOIRA syndrome. She subsequently underwent a transvaginal oblique septectomy under ultrasonic guidance, laparoscopic excision of a right ovarian endometriotic cyst, and removal of a right fallopian tube hematocele. Five months postoperatively, ultrasound imaging revealed vaginal oblique septal scar adhesion, minor hematocele formation following OVS and within the uterine cavity, and substantial fluid accumulation within the right fallopian tube. Six months after the initial surgery, she underwent a second procedure: vaginal oblique septal scar resection and vaginoplasty. Six months after the second surgery, follow-up ultrasound revealed recurrence of OVS adhesion, prompting a third surgical intervention: vaginal oblique septal scar resection and vaginoplasty. Postoperatively, ultrasound follow-up revealed residual fluid within the uterine cavity and persistent right lower abdominal pain. The patient was advised to continue with regular follow-up evaluations.

Type II-b (C3bV2) (n=2)

Two cases were classified as Type II-b UGTOIRA syndrome. One patient was clinically cured after a one-stage operation, while the other patient only showed clinical improvement after three operations.

One patient underwent laparoscopic exploration, incision and drainage of the right fimbrial end of the fallopian tube, vaginal oblique septum resection, vaginoplasty, and cervical canal incision via combined vaginal laparotomy. During the four-year follow-up period, the patient did not exhibit any symptoms of menstrual outflow obstruction, and ultrasonography revealed no evidence of hematocolpos or other forms of blood accumulation within the reproductive tract.

Another patient underwent three surgical procedures over an eight-year period, ultimately resulting in the removal of the uterus and fallopian tubes on the affected side. The patient presented with severe pain in the lower left quadrant of the abdomen following menarche at 11 years of age. She was diagnosed with left-sided UGTOIRA syndrome during an emergency visit to our hospital. She promptly underwent vaginal oblique septectomy and left cervical atresia resection via vaginal laparotomy. However, dysmenorrhea persisted after the surgical intervention. Eight years later, the patient was readmitted due to persistent vaginal and anal pain lasting three days during menstruation. Ultrasonography revealed closure of the left cervical external os, significant hematocolpos and hematometra in the left cervical canal and uterine cavity, left-sided adenomyosis, thickening of the left fallopian tube, and an endometriotic cyst in the left ovary. Gynecological examination indicated stenosis of the surgical scar at the OVS incision site. The patient subsequently underwent a second laparoscopic procedure, which included left ovarian cystectomy, ovarioplasty, lysis of pelvic adhesions, ultrasound-guided cervical dilation, excision of vaginal scar tissue, and fallopian tube drainage. Menstruation resumed seven months postoperatively, characterized by reduced menstrual flow and diminished lower abdominal pain. The third hospital admission was necessitated by persistent lower abdominal pain lasting more than one month. Ultrasonography revealed a marked accumulation of blood within the left cervical canal, uterine cavity, and fallopian tube. Gynecological examination confirmed obstruction of the OVS scar. Subsequently, the patient underwent a third surgical intervention, which included transabdominal extrafascial left hysterectomy, left salpingectomy, left ovarian suspension, and lysis of pelvic adhesions. Two years post-surgery, the patient presented to our hospital with complaints of nonspecific lower abdominal discomfort. Ultrasonography revealed the presence of an encapsulated fluid collection measuring 8.0 * 4.0 * 4.0 cm^3, located posterior to the uterus within the pelvic cavity.

Type III (C3bV0) (n=3)

Three cases were categorized as Type III. Among these, two patients (2/3) had no prior surgical history, while one patient (1/3) had undergone two previous surgeries. All patients received concurrent ipsilateral cervical plasty and surgical management of complications, with one case additionally requiring Strassman metroplasty. The overall cure rate for Type III cases was 100% (3/3).

The initial patient presented to our hospital with primary complaints of dysmenorrhea and postmenstrual spotting. She underwent comprehensive gynecological surgical intervention, including laparoscopic exploration with excision of a right salpingo-hematocele and pelvic hematocele, diagnostic hysteroscopy, as well as ultrasound-guided transvaginal

right lateral cervical incision and subsequent cervicoplasty. Postoperative follow-up at the 10-month mark revealed no evidence of residual hematoceles within the reproductive tract on ultrasonographic examination, and the patient remained asymptomatic with no clinical manifestations.

The second patient presented to our hospital with a primary complaint of dysmenorrhea. She underwent comprehensive surgical intervention, including laparoscopy, laparotomy, Strassman metroplasty, left ovarian cystectomy, left ovarian reconstructive surgery, hysteroscopy, and drainage of the left cervical blind pouch. Postoperative follow-up assessments at one, three, and six months demonstrated normal uterine cavity morphology on ultrasonographic evaluation, with no evidence of intracavitary hematoceles or clinically significant symptoms.

The third patient, presenting with a large pelvic mass, previously underwent transabdominal left ovarian cystectomy with adhesiolysis at an outside institution one year prior. Postoperative histopathological analysis confirmed the diagnosis of an endometriotic cyst in the left ovary. Following surgery, the patient developed recurrent abdominal pain and persistent pelvic masses. Two months postoperatively, an ultrasound-guided aspiration of an abdominal mass was performed at the same institution, yielding a brownish fluid. The patient subsequently presented to our facility half a month later and was diagnosed with left unilateral gonadotropin-independent theca cell tumor with insulin resistance and androgen excess (UGTOIRA) syndrome. Surgical intervention at our center included laparoscopic pelvic adhesiolysis, left ovarian cyst excision, left salpingostomy, diagnostic hysteroscopy with methylene blue instillation, and cervical reconstruction. At the nine-month follow-up, ultrasonography revealed a small intracavitary hemorrhagic focus in the left uterine cavity, which resolved completely by the subsequent four-month assessment, with no residual hematocele detected on ultrasound evaluation.

Type IV (C3cV0) (n=3)

The three cases were categorized as Type IV. Among them, one case (1/3) had a surgical history of three procedures, another case (1/3) had undergone one prior surgery, and the remaining case had no prior surgical intervention. All patients underwent concurrent ipsilateral hysterectomy and salpingectomy in conjunction with surgical management of associated complications, with all achieving clinical remission. The overall cure rate for Type IV cases was 100% (3/3).

Two cases presented with right-sided anomalies, while one case demonstrated a left-sided anomaly. Intraoperatively, all three cases exhibited partial hypoplasia of the ipsilateral cervix, manifesting as a blind-ending structure with an otherwise intact vagina, in conjunction with a complete bicornuate uterus. Among the two cases without communication, minimal hemorrhage was observed within the ipsilateral cervical canal, moderate intracavitary bleeding within the corresponding uterine cavity, and significant hemoperitoneum within the ipsilateral fallopian tube.

The first patient presented with a nine-month history of intermittent right lower quadrant abdominal pain and a seven-month history of a pelvic mass localized to the right adnexal region. Prior to admission at our institution, she had undergone three surgical interventions at a local hospital within a seven-month period: (1) an electric vacuum aspiration of the left uterus, (2) laparoscopic drainage and right salpingostomy, and (3) a comprehensive laparoscopic procedure including pelvic adhesiolysis, right salpingostomy, hysteroscopy, cystoscopy, and vaginal exploration. Despite these interventions, her right lower quadrant abdominal pain persisted.Upon evaluation at our hospital, a gynecological examination was performed using a sterile speculum and catheter. Findings included a smooth left cervix with an 8-cm uterine cavity depth. A rudimentary cervical remnant was noted on the right vaginal wall, appearing disconnected from the main cervical structure. The patient subsequently underwent a transabdominal surgical procedure comprising right hysterectomy, right salpingectomy, and lysis of pelvic adhesions. Postoperative recovery was uneventful, with no reported abdominal discomfort.

The second patient has presented with dysmenorrhea persisting for two years since menarche. One year prior to presentation, she underwent transabdominal exploration and cystectomy of a left ovarian cyst at our institution. Postoperative histopathological examination confirmed the diagnosis of an endometriotic cyst. Despite surgical intervention, her dysmenorrhea persisted. Subsequently, she received further gynecological management, which included laparoscopic exploration, hysteroscopy, total laparoscopic left hysterectomy, left salpingectomy, partial small bowel resection, lysis of intestinal adhesions, and pelvic adhesiolysis. During the follow-up period, the patient reported complete resolution of dysmenorrheic symptoms.

The third patient presented with a fistula at the site of partial cervical fusion, accompanied by minimal ipsilateral cervical canal hemorrhage. She was asymptomatic and sought medical attention for infertility evaluation. Surgical intervention included laparoscopy, right-sided hysterectomy, left cervicoplasty, right ovarian cystectomy, hysteroscopy with endometrial polypectomy, and fallopian tube patency testing. Histopathological examination confirmed the right ovarian cyst as an endometrioma. Despite surgical management, the patient did not achieve pregnancy during the subsequent one-year follow-up period.

Type V (U4aC0V0) (n=2)

Only two cases were categorized as Type V. Both patients had no prior surgical history and presented with primary complaints of dysmenorrhea. Ultrasonographic evaluation in the first case revealed significant intracavitary and fallopian tube hematoma secondary to obstruction, with no communication to the contralateral uterine cavity. The second case demonstrated marked narrowing of the lower uterine segment on the right side, with anomalous communication to the left internal cervical os. Both patients underwent laparoscopic resection of the obstructed uterine horn along with ipsilateral salpingectomy. Follow-up assessments confirmed complete resolution of dysmenorrhea in both cases, yielding a 100% clinical cure rate (2/2) for Type V classification.

Comprehensive Surgical Path

A total of 60 patients (60/68, 88.2%) underwent vaginal oblique septectomy, comprising 17 cases (17/60, 28.3%) performed with visual guidance and 43 cases (43/60, 71.7%) utilizing hysteroscopic assistance. Among the 68 patients, 20 (20/68, 29.4%) received laparoscopic surgery, 45 (45/68, 66.2%) underwent hysteroscopic procedures, and 7 (7/68, 10.3%) were treated via laparotomy. Transabdominal ultrasound guidance was employed in 56 cases (56/68, 82.4%).

Comprehensive Surgical Methods

In all 68 cases, the initial gynecological procedure performed at our institution was vaginal oblique septectomy, which was conducted in 60 cases (60/68, 88.2%). Among these 60 patients, concurrent surgical interventions included: hysteroscopic cervicoplasty in one case, hysteroscopic septum resection in one case, combined hysteroscopic resection of both the uterine and cervical septa in one case, laparoscopic ipsilateral salpingoplasty in four cases, laparoscopic ipsilateral salpingectomy in one case, total hysterectomy with bilateral salpingectomy in a 52-year-old patient, and excision of an ipsilateral ureteral remnant cyst (with vaginal vault fistulation) in one case.

Postoperatively, four patients (4/60, 6.7%) required secondary surgical procedures: residual vaginal septum and ipsilateral fallopian tube excision (n=1), isolated residual vaginal septum excision (n=1), dysplastic kidney and pelvic ureteral remnant resection (n=1), and ureteral remnant excision (n=1). Additionally, two patients (2/60, 3.3%) underwent a total of three surgical interventions: in one case, residual vaginal septum resection was performed in both the second and third surgeries, while in the other, residual vaginal septum excision occurred during the second surgery, followed by ipsilateral hysterectomy and salpingectomy in the third.

The remaining 8 patients (8/68, 11.8%) did not receive vaginal oblique septectomy. Specifically, two patients underwent ipsilateral cervical reconstruction (one concurrently with hysteroscopic uterine septum resection, and the other with concurrent ipsilateral salpingectomy), one patient was treated with combined cervicoplasty and Strassman metroplasty, and five patients underwent ipsilateral hysterectomy with salpingectomy.

Additionally, 21 patients (21/68, 30.9%) received surgical management for associated complications, including endometriosis, pelvic adhesions, and pelvic inflammatory disease.

Overall Clinical Outcomes

In the cohort of 68 patients, the overall cure and improvement rates were 95.6% (65/68) and 4.4% (3/68), respectively. Among patients without prior surgical history, the primary (one-stage) surgical intervention performed by the Department of Obstetrics and Gynecology at our institution achieved a cure rate of 82.4% (56/68). Detailed procedural information for patients achieving cure through primary surgery alone is presented in Table 1. The surgical history, types of procedures performed, and clinical outcomes of the 12 patients (12/68, 17.6%) with prior relevant surgical interventions, requiring reoperations, or who did not achieve primary cure through initial one-stage surgery are detailed in Tables 2 and 3.

Table 1 Description of the Operations of Patients with Different Types and Subtypes of Unilateral Genital Tract Obstruction with Ipsilateral Renal Anomaly Syndrome Who Were Cured by a One-Stage Operation Only (n=56, 82.4%)

Type (n, Percentage Within Type)	Subtype (n, Percentage within Subtype)	Operations
Type I (vaginal obstruction) (n=47,88.7%)	Type I-a (n=18,90.0%) Type I-b (n=24,88.9%) Type I-c (n=5,83.3%)	With the exception of one perimenopausal patient who additionally requested concurrent hysterectomy, all other patients underwent simultaneous vaginal oblique septectomy along with surgical management of endometriosis, pelvic inflammation, and adhesions.
Type II (cervicovaginal obstruction) (n=4,57.1%)	Type II-a (n=3,60.0%) Type II-b (n=1,50.0%)	All patients underwent concurrent vaginal oblique septectomy along with surgical treatment for endometriosis, pelvic inflammatory disease, and adhesions, with one case additionally receiving ipsilateral cervical plasty.
Type III (cervical obstruction) (n=2,66.7%)		Both patients underwent concurrent ipsilateral cervical plasty along with surgical intervention for endometriosis, inflammatory conditions, and adhesions, with one patient additionally receiving a Strassman metroplasty.
Type IV (unilateral partial cervical aplasia) (n=1,33.3%)		Ipsilateral hysterectomy and salpingo-oophorectomy performed concurrently with surgical intervention for endometriosis, pelvic inflammatory disease, and adhesiolysis.
Type V (unilateral rudimentary horn with cavity and without communicating with contralateral uterus) (n=2,100%)		Concurrent ipsilateral hysterectomy and salpingectomy with surgical management of endometriosis, inflammatory lesions, and adhesive disease.

Table 2 Description of the Operations and Clinical Outcomes of Patients with Different Types and Subtypes of Unilateral Genital Tract Obstruction with Ipsilateral Renal Anomaly Syndrome Who Had a History of Related Surgeries (n=6, 8.8%)

No.	Type and Subtype	• Surgical History • Operation • Reoperation	Follow-Up Duration (months)	Clinical Outcome
1	Type I-a	• One surgical operation history: "Laparoscopic high ligation of bilateral indirect inguinal hernia sacs, accompanied by abdominal exploration and colposcopy". • Transvaginal partial resection of oblique vaginal septum. • One reoperation: "Resection of the remaining ureteral cyst and the dysplastic kidney in the ipsilateral pelvis".	45	Cure
2	Type I-a	• Historical record of two surgical procedures: "Conservative laparotomic surgery for right tubal pregnancy" and "Laparoscopic right salpingectomy for recurrent right tubal pregnancy". • Hysteroscopic oblique vaginal septum resection with concurrent vaginoplasty, laparoscopic pelvic adhesiolysis, left salpingostomy, and left ovarian lesion excision.	6	Cure
3	Type I-b	• One surgical operation history: "Laparoscopic pelvic adhesiolysis with excision of a left ovarian endometrioma and left salpingostomy". • Transvaginal oblique septectomy.	12	Cure
4	Type III	• Historical record of two surgical procedures: "Transabdominal left ovarian cystectomy with cyst wall denudation and concurrent intestinal adhesion lysis" and "Ultrasound-guided percutaneous abdominal mass aspiration". • Laparoscopic pelvic adhesions lysis with left ovarian cyst excision, left salpingostomy, hysteroscopy methylene blue drainage and cervical plasty.	12	Improvement
5	Type IV	• Historical record of three surgical procedures: "Left uterine electric suction abortion", "Laparoscopic right salpingostomy drainage", and "Comprehensive laparoscopic procedure including pelvic adhesiolysis, right salpingostomy, hysteroscopy, cystoscopy, and vaginal exploration". • Transabdominal right hysterectomy, right salpingectomy and pelvic adhesions lysis.	6	Cure
6	Type IV	• One surgical operation history: "Transabdominal exploration with denudation of left ovarian cyst". • Laparoscopic exploration, hysteroscopy, transabdominal left hysterectomy, left salpingectomy, partial resection of small intestine, separation of intestinal adhesion and separation of pelvic adhesion.	12	Cure

Table 3 Description of the Operations Performed on Patients with Different Types and Subtypes of Unilateral Genital Tract Obstruction with Ipsilateral Renal Anomaly Syndrome Who Underwent Reoperations or Did Not Achieve a Cure Through One-Stage Surgery (n=6, 8.8%)

No.	Type and Subtype	• Operation • Reoperation	Follow-Up Duration (months)	Clinical Outcome
1	Type I-b	• Transvaginal resection of the oblique septum. • Robot-assisted laparoscopic excision of right pelvic ureteral remnant cyst with adhesiolysis.	35	Cure
2	Type I-b	• Transvaginal oblique septectomy with ultrasonic monitoring with postoperative indwelling balloon catheter for a duration of one month. • One reoperation: "Complete vaginal oblique septectomy, vaginoplasty, laparoscopic ipsilateral salpingectomy, and pelvic adhesiolysis".	15	Cure
3	Type I-c	• Transvaginal oblique septectomy. • One reoperation: "Complete vaginal oblique septectomy with postoperative indwelling balloon catheter for a duration of one month."	6	Cure
4	Type II-a	• Transvaginal oblique septectomy under ultrasonic guidance, laparoscopic excision of left ovarian endometrioma, and evacuation of salpingo-hematocele and pelvic hematocele. • No reoperation.	120	Improvement
5	Type II-a	• Transvaginal oblique septectomy under ultrasonic guidance with concurrent laparoscopic excision of a right ovarian endometriotic cyst and right fallopian tube hematocele. • Two reoperations were performed: "Vaginal oblique septal scar resection and vaginoplasty" and "Vaginal oblique septal scar resection and vaginoplasty", both for a closed vaginal oblique septal scar.	96	Improvement
6	Type II-b	• Combined vaginal laparotomy-assisted transvaginal oblique septectomy with left cervical atresia resection. • Two reoperative surgical procedures were performed to address a closed vaginal oblique septal scar: "Laparoscopic left ovarian cystectomy with ovarioplasty, pelvic adhesiolysis, ultrasound-guided cervical dilation, vaginal scar excision, and fallopian tube drainage" and "Transabdominal extrafascial left hysterectomy with left salpingectomy, left ovarian suspension, and pelvic adhesiolysis".	108	Cure

Statistical Significance

The incidences of various operations differed significantly across different types and subtypes ($P < 0.001$). Similarly, clinically significant differences were observed in cure rates among these types and subtypes, which also demonstrated statistically significant variations ($P = 0.013$).

Discussion

In our study, a one-stage surgical procedure was successfully performed in 56 patients (82.4%). Appropriate clinical management is essential for achieving optimal surgical outcomes. The primary objective in managing patients with UGTOIRA syndrome is to alleviate symptoms while preserving fertility, a goal that can only be attained through proper clinical management. Based on our clinical experience and supported by the relevant literature,[6,13–15] the optimal clinical management entails precise early diagnosis, individualized surgical intervention, and systematic postoperative follow-up—all crucial for the early detection and treatment of recurrent obstruction.

Why Some Cases of the UGTOIRA Syndrome Cannot Be Cured by a One-Stage Surgery

From the surgeon's perspective, there are two main reasons why UGTOIRA syndrome cannot be cured through one-stage surgery. Firstly, as indicated by the surgical histories described in Table 2, the surgeon was not a genitourinary specialist and lacked sufficient knowledge of UGTOIRA syndrome, resulting in inaccurate preoperative and intraoperative diagnoses. Consequently, only operations for related complications were performed without addressing the fundamental problem of unilateral genital tract obstruction. Secondly, as outlined in Table 3, while the preoperative and intraoperative diagnoses were correct, there were incomplete surgical interventions to relieve the obstruction such as inadequate resection of the OVS, insufficient cervical dilatation, and failure to remove a severely dysplastic unilateral cervix.

Complete Removal of Unilateral Genital Tract Obstruction

Surgical intervention represents the most efficacious therapeutic approach for ameliorating clinical symptoms and preserving fertility in patients diagnosed with UGTOIRA syndrome. For patients classified as Type I and II, maximal resection of the OVS constitutes the optimal surgical strategy. In contrast, Type II-b patients necessitate concurrent incision of the ipsilateral obliterated cervical os coupled with cervical reconstruction. The optimal timing for surgical intervention in these cases is approximately during the menstrual period, as this physiological state enhances the visualization and tactile identification of markedly distended hematocolpos, thereby facilitating more precise surgical resection.[11] The greater the vertical distance between the lower margin of the OVS and the external vaginal orifice, the reduced accumulation of blood within the posterior compartment of the OVS. This anatomical configuration correlates with an increased incidence of ipsilateral cervical hypoplasia, consequently rendering complete surgical excision of the OVS more technically challenging. In such cases, concurrent cervical canal dilation and remodeling may prove clinically necessary. Type II-a is defined by a superiorly positioned OVS accompanied by an ipsilateral hypoplastic but patent cervix, whereas Type II-b features a similarly elevated OVS associated with ipsilateral cervical orifice atresia. The majority of patients achieve complete resolution following resection of the OVS. In our study, the overall cure rate for Type I was 100% (53/53), whereas Type II demonstrated a cure rate of 57.1% (4/7). Patients classified as Type III, characterized by unilateral cervical orifice atresia with an otherwise normal vagina, require hysteroscopic cervical plasty. The cure rate for Type III was 100% (3/3). For Types IV and V, surgical management via laparoscopic or open unilateral hysterectomy with salpingectomy is recommended. The corresponding cure rates for Types IV and V were 100% (3/3 and 2/2), respectively.

Early Surgery Reduces Complications

Early surgical intervention can mitigate the risk of potential genital tract infections and obstetric-gynecological complications.[14] Some studies have demonstrated that patients with UGTOIRA frequently achieve favorable clinical outcomes and improved fertility rates following early surgical intervention.[4,16–20] Han et al reported that 13% of patients underwent surgical intervention prior to puberty, with no postoperative complications observed in any pediatric cases.[21] In our study, the median age at operation was 15 (12–23) years, with ages ranging from 5 to 52 years. The median time interval between diagnosis and surgery was 0 (0,0) months, with intervals ranging from 0 to 12 months.

Surgical Treatment of Complications

Resection of OVS should be the first step in treatment, and surgery plays an important role in the management of endometriosis and pelvic adhesions.[22] During preoperative evaluations, encompassing both gynecological assessments and imaging studies, it is advisable to maintain a high index of suspicion for potential complications. When indicated, laparoscopic exploration should be performed, followed by surgical intervention for any identified pathological findings. In our study, 26.5% (18/68) of the patients underwent surgery for complications including endometriosis, pelvic adhesions, and pelvic inflammation.

Whether to Perform Strassman Metroplasty and (or) Uterine Septum Resection

The decision to perform Strassman metroplasty and/or uterine septum resection during the operation to relieve obstruction depends on the patient's age and marital status. For young unmarried patients and adolescents, we suggest that only surgery to relieve the obstruction should be performed. They can then decide whether to undergo Strassman metroplasty and/or uterine septum resection after marriage based on their pregnancy situation. The incidence of pregnancy in a complete bicorporeal uterus after OVS resection has been reported to be equivalent to that in a normal uterus.[23] Currently, there is no comparative research on pregnancy outcomes between Strassman metroplasty for a complete bicorporeal uterus and expectant management following oblique septectomy. A large-sample cohort study demonstrated that septum resection does not yield superior pregnancy outcomes compared to expectant management in women with a septate uterus.[24] However, existing evidence indicates that hysteroscopic resection of a uterine septum may improve reproductive outcomes in women experiencing recurrent pregnancy loss associated with this anatomical anomaly.[25] Therefore, if the patient has a normal pregnancy history, Strassman metroplasty is not necessary. If the patient has a history of recurrent abortion or preterm birth, simultaneous consideration of Strassman metroplasty and septum resection may be made when performing an operation to relieve obstruction. In our study, a 17-year-old patient with Type III UGTOIRA underwent cervical plasty, Strassman metroplasty, and septum resection for a bicorporeal septate uterus, septate cervix, left obliterated cervical orifice with fistula on the cervix septum, and normal vagina. The postoperative recovery was positive and the uterus became normal. Another patient underwent uterine septum resection due to infertility.

Whether to Perform Ipsilateral Residual Ureterocele Resection

A prior study documented a case of bicorporeal septate uterus with cervical septum and OVS accompanied by ipsilateral renal hypoplasia, wherein a single ectopic ureter drained into the posterior compartment of the OVS. The cavity posterior to the oblique septum contained a mixed hematourinoid fluid. The patient underwent single-stage surgical management, involving complete resection of the OVS followed by laparoscopic excision of the hypoplastic kidney and its associated ectopic ureter, with favorable clinical outcomes.[26] Another study documented that among 21 patients presenting with varying degrees of menstrual obstruction (ranging from cervical to introital level, including 11 cases of obstructed vaginal septum), six exhibited a distended ectopic ureteral orifice opening into the gynecologic tract, which was filled with blood. Following resolution of the obstruction, the hemorrhagic manifestations in the ureteral remnant resolved completely. The investigators posited that this hemorrhagic phenomenon resulted from retrograde blood flow induced by elevated intraluminal pressure during genital tract obstruction.[27] After resecting the OVS without excising the ipsilateral residual ureterocele with an ectopic opening into the space behind the OVS, there is a risk of urine leakage into the vagina and retrograde infection in the residual ureterocele. Therefore, we believe that excision of the ipsilateral residual ureterocele with an ectopic opening into the space behind the OVS should be performed after complete resection of the OVS in a one-stage surgery. The successful completion of this procedure necessitates collaborative efforts between the urologist and gynecologist. In our study, four patients (5.9%) presented with an ipsilateral dysplastic kidney situated in the pelvic cavity, accompanied by a dilated and dysplastic ureter. Among these cases, three were associated with ectopic ureteral orifices or fistulous tracts extending into the posterior compartment of the OVS. These three patients underwent urological intervention, comprising excision of the ipsilateral dysplastic pelvic kidney and resection of the associated ureteral remnant cyst.

Strengths, Limitations, and Future Research

This study provides a comprehensive yet concise description of the surgical protocols employed in the one-stage management of 56 patients with various subtypes of UGTOIRA syndrome. Detailed surgical approaches are presented for six patients with prior surgical history and six patients who did not achieve clinical cure following one-stage surgery. Furthermore, we summarize key successful strategies, analyze contributing factors to treatment failure, and derive valuable clinical insights. These cases offer meaningful guidance for urogynecologists, pediatric surgeons, imaging specialists, and patients involved in the management of UGTOIRA syndrome.

Our study has several limitations. The study was a retrospective, single-center study. The number of cases was limited, the follow-up period was short, and pregnancy outcomes were not assessed because the median age of the study

participants was 15 (12–23) years. The median follow - up period was 12 (10–51) months. As a result, most of the cases had no plans for pregnancy, and some were juveniles.

In future research, a multicenter study with a larger sample size is necessary to validate the efficacy of each clinical management approach. Moreover, an extended follow - up of the study cohort is essential, with a particular focus on assessing long - term pregnancy outcomes.

Conclusions

UGTOIRA syndrome can be categorized into five types according to the location of the obstruction. Distinct surgical plans ought to be formulated for each type of UGTOIRA syndrome, also taking into account the treatment of complications. Systematic postoperative follow - up is advantageous for the early detection and treatment of recurrent obstructions. Appropriate management can result in optimal clinical outcomes and successful pregnancies. A professional team with extensive clinical experience serves as the guarantee for the favorable clinical outcome of this rare disease.

Data Sharing Statement

The data that supports the findings of this study, including any relevant details needed to reproduce the published results, are available from the corresponding author upon reasonable request. The data set used in the current study will be made available upon request from Ling Zhang via Email at zhangling0709@tjh.tjmu.edu.cn.

Ethics Approval and Consent to Participate

This study was conducted in accordance with the declaration of Helsinki. It received approval from the Medical Ethics Committee of Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology (approval number: TJ-IRB20220917). This is a retrospective and descriptive analysis based on prospectively collected data for routine clinical services. The Medical Ethics Committee of Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology, granted a waiver of informed consent for this retrospective case study on the grounds that the retrospective observational study met all of the following conditions: 1. The research holds significant social value; 2. The risks to subjects are no greater than minimal risk; 3. Waiving informed consent or certain elements of informed consent does not adversely affect the rights and health of the subjects; 4. The privacy, confidentiality, and anonymity of the subjects are guaranteed; 5. Relevant information will be provided to the subjects during follow-up after their participation in the study.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

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