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Brief Clinical Report

Laparoscopic Management of a Noncommunicating Uterine Horn in a Patient with an Acute Abdomen

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Summary: A 13-year-old girl with a history of cloacal anomalies presented with acute abdominal pain. Abdominal ultrasound was not definitive, and vaginal probe ultrasound was precluded by the patient's stenotic vagina. Magnetic resonance imaging delineated a left hematometra and hematosalpinx as well as a more normal-appearing right hemiuterus. Operative laparoscopy was used to lyse the extensive pelvic adhesions in a patient with a history of an imperforate anus and to resect a left rudimentary uterine horn with outflow obstruction. A review of cases in the world literature reveals that operative laparoscopy can be used to treat these patients successfully. Key Words: Rudimentary uterine horn—Acute abdomen—Magnetic resonance image.

Failure of lateral fusion of the mullerian ducts during the 8th week of intrauterine development may result in two separate uterine bodies (1). Association of this with maldevelopment of one mullerian duct results in a unicornuate uterus or a unicornuate uterus with a small rudimentary horn attached. In one series, 11 of 13 patients with a unicornuate uterus had a rudimentary horn (2). Although an isolated unicornuate uterus is usually asymptomatic, reproduction is compromised by increased infertility, fetal wastage, and preterm labor (3). Association of a unicornuate uterus with a rudimentary horn, however, may result in surgical emergencies. A rudimentary horn pregnancy will usually develop with the signs and symptoms of an ectopic pregnancy. O'Leary and O'Leary reviewed a total of 327 cases of rudimentary horn pregnancy reported since Mauriceau's case in 1669 and estimated that there is no communication between the rudimentary horn and its more normal companion in about 90% of cases (4). Outflow obstruction can lead to presentation with an acute abdomen in the absence of pregnancy. When a rudimentary horn contains functional endometrium and does not communicate externally, severe dysmenorrhea will begin soon after menarche (2).

This report describes the laparoscopic treatment of a unicornuate uterus with noncommunicating rudimentary horn, a configuration categorized as class IIB by the American Fertility Society (AFS) classifications of mullerian anomalies (5). The previously reported five cases of laparoscopic treatment of mullerian remnants, including four class IIB anomalies, are reviewed. The case reported here is unique for the use of magnetic resonance imaging (MRI) in the diagnosis, the treatment of a pediatric patient, and the patient's associated developmental anomalies.

CASE REPORT

A 13-year-old girl, gravida 0, arrived at the emergency department 5 days after onset of her menses complaining of 5 days of severe backache and abdominal pain. A product of a normal term pregnancy remarkable for the presence of a single um-

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bilical artery, she had a history of an imperforate ...s requiring a colostomy shortly after birth, followed by an abdominal perineal pull-through procedure at 2 years of age. Operative findings at that time suggested a bicornate uterus and possible vaginal septum. An intravenous pyelogram revealed normal ureters; however, the urethral meatus was located high in the vagina. Menarche began 9 months before presentation, and the patient was in her third episode of menses on arrival at the emergency department. She was not sexually active and had no prior pelvic speculum exam. Serum beta human chorionic gonadotropin was negative. An abdominal ultrasound revealed an enlarged, poorly defined endometrial stripe. Bilaterally, serpiginous adnexal structures were noted, suggesting dilated fallopian tubes. Given the caliber of the patient's vagina, a vaginal probe ultrasound was not possible and an MRI was obtained, which suggested the existence of a small right-sided uterus and a larger left-sided dilated pelvic mass containing blood. Initial management consisted of pain control while examination under anesthesia and definitive surgery were planned.

Under general endotracheal anesthesia, the patient underwent operative videolaparoscopy. Ex...ination under anesthesia revealed a stenotic vag...nal canal. Bimanual examination revealed a firm 8-cm-diameter left pelvic mass associated with the vaginal apex. Vaginoscopy using a 5-mm laparoscope demonstrated a normal-appearing cervix on the right side of the vaginal apex. No cervical opening was seen on the left side. A small longitudinal septum measuring 2 mm was noted at the vaginal apex. Laparoscopy revealed extensive adhesions of the sigmoid colon to a left hematosalpinx and to a 10 × 6 × 6-cm dumbbell-shaped hematometra representing the left noncommunicating rudimentary uterine horn. Extensive adhesions of the cecum and vermiform appendix to the right adnexal area were also noted, as was a tortuous dilated right fallopian tube. After lysis of adhesions, a right-sided 5 × 2 × 1 cm hemiuterus was uncovered in close association with the peritoneum of the right pelvic side wall and was shown via manual examination to be contiguous with the cervix. Bilaterally, the ovaries were involved in adhesions but were otherwise normal with a functional cyst on the right.

Using CO_2 laser and hydrodissection, meticulous adhesiolysis was used to define further the markedly abnormal pelvic anatomy. The left infundibu...)pelvic ligament was identified. The left mesosalpinx and isthmic portion of the left fallopian tube were then serially coagulated using bipolar electrocautery and incised, allowing removal of the tube. The left hematometra was then incised and drained. The left round ligament was then coagulated and transected, facilitating development of a bladder flap by both sharp and blunt dissection, followed by coagulation and cutting of the left utero-ovarian ligament, broad ligament, and uterine artery. Given the extensive adhesions of the rectum to the lower uterine segment of the left hemiuterus, the decision was made to excise the hemiuterus at the level of the lower uterine segment. Exploration of this cavity revealed no communication to the vagina. Following ablation of the epithelium lining the cavity using bipolar coagulation, the cuff of the lower uterine segment was closed with interrupted 4-0 polyglactin (Vicryl) sutures. The specimen was cut in several parts and removed from the abdomen through the infraumbilical incision. Finally, sigmoidoscopy with pneumoinflation was performed, confirming an intact rectum and sigmoid colon.

Pathology reports confirmed the presumed diagnosis of mullerian dysgenesis with both tubal and endometrial tissues identified. Postoperatively the patient contracted an enterococcal urinary tract infection and was treated with ampicillin. She went home 3 days after surgery and began a regimen of intramuscular Depo-Provera (150 mg every 3 months). She continues to be free of pelvic pain 4 months after surgery.

DISCUSSION

The management of this case illustrates various techniques that are useful for the treatment of symptomatic mullerian dysgenesis. The utility of MRI in the evaluation of suspected mullerian anomalies is well documented (6). In this patient, in whom abdominal ultrasound was nondiagnostic and vaginal probe ultrasound was precluded by a stenotic vagina, preoperative MRI defined a small right-sided hemiuterus and an enlarged left hematometra (Fig. 1).

Operative laparoscopy is rapidly becoming the state-of-the-art treatment of symptomatic noncommunicating rudimentary uterine horns. A review of the world literature reveals five reports of laparoscopic removal of mullerian remnants between 1990 and 1995 (7–11). Four of these cases involved unilateral removal of a rudimentary horn, and one case

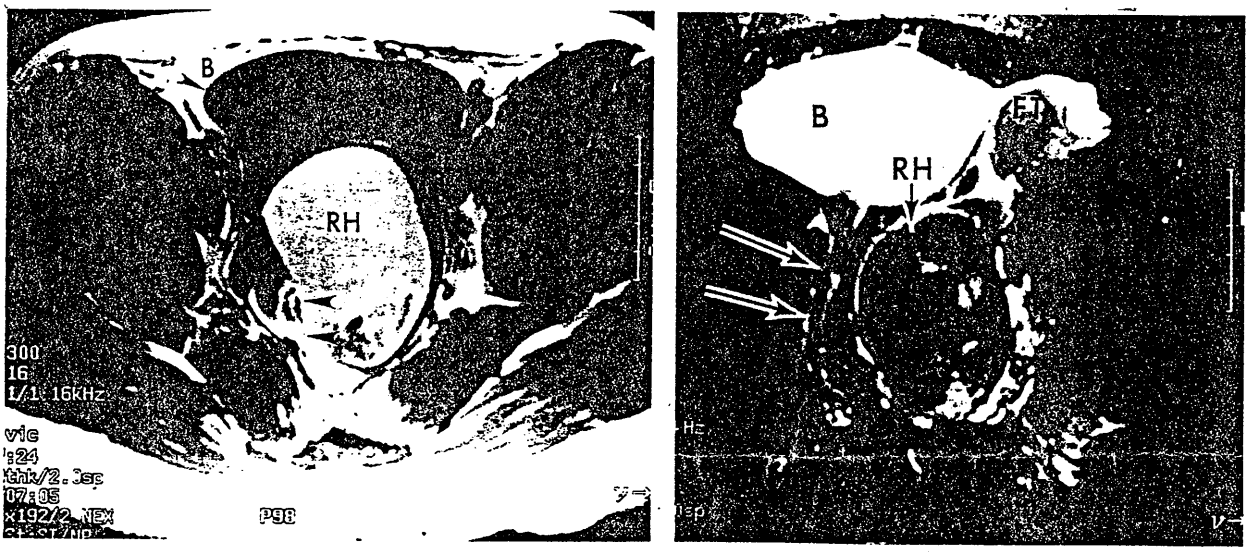

FIG. 1. Magnetic resonance images of the pelvis identify an obstructed left rudimentary horn (RH) and a smaller, thin right hemiuterus (arrows). Bladder (B) and fallopian tube (FT) are also shown.

involved removal of bilateral remnants in a patient without a vagina. Table 1 summarizes the cases.

Patient ages ranged from 13 to 30 years. The case we report is remarkable for the diagnosis and treatment of a patient at age 13 years; in the other reported cases of AFS class IIB anomalies the mean age is more than 28 years. Although severe dysmenorrhea begins soon after menarche, cyclic menstruation from the communicating hemiuterus occurs and cryptomenorrhea may not be considered, thus delaying the diagnosis. With a patent fallopian tube connected to the rudimentary uterus, retrograde menstruation may lead rapidly to pelvic endometriosis (2). Thus, diagnosis and treatment as soon as possible are important to avoid compromising fertility. Four of the six cases reviewed in Table 1 reported either endometriosis or severe adhesions at laparoscopy. Of the two remaining cases, one was pretreated with a gonadotropin releasing hormone agonist for 6 months, and the other involved removing bilateral remnants without evidence of endometrial tissue on pathologic evaluation.

The case we report is also notable for the patient's other developmental anomalies: single umbilical artery and imperforate anus. The association of congenital anomalies in infants with one umbili-

TABLE 1. *Reported cases of laparoscopic removal of mullerian remnants (1990–1995)*

Author	Year	Age (yr)	Symptoms	Preop Rx	IVP	Endometriosis	Anomaly	Comments
Canis (7)	1990	30	Dysmen, Dyspar	GnRH agonist (3 mo)	Normal	Present	AFS class IIB	Horn removed via 2-cm incision
Yeko (8)	1992	18	Dysmen	None	Normal	Not present	Absent vagina, bilateral mullerian remnants	No uterine arteries present, no endometrium identified
Nezhat (9)	1994	28	Dysmen	Danazol (6 mo)	Bilateral sponge kidneys	Present	AFS class IIB	Hysteroscopy aided diagnosis
Mais (10)	1994	26	Dysmen, pelvic pain	GnRH agonist (6 mo)	Normal	Not present	AFS class IIB	Endoscopic stapler used
Schattman (11)	1995	29	Dysmen, infertility	None	Normal	Present	AFS class IIB	Standard technique
Present case	1995	13	Dysmen	None	Vaginal urethra	Not present, extensive adhesions noted	AFS class IIB, associated imperforate anus	MRI and vaginoscopy aided diagnosis

Rx, treatment; IVP, intravenous pyelogram; AFS, American Fertility Society; GnRH, gonadotropin releasing hormone; MRI, magnetic resonance imaging; dysmen, dysmenorrhea; dyspar, dyspareunia.

cal artery missing is well characterized and estimated at 30% (12). Most anorectal malformations result from abnormal development of the urorectal septum, resulting in incomplete separation of the cloaca. Imperforate anus results from failure of the anal membrane to perforate at the end of the 8th week of development (1). Although mullerian dysgenesis is not commonly associated with anorectal anomalies, both anomalies result from maldevelopment during the 8th week.

Ultrasound was used to image the pelvis in all cases. Only in the case reported herein was MRI used. Although all patients had intravenous pyelograms demonstrating normal ureters, the association of urinary tract anomalies with mullerian dysgenesis is well known (2,12). Bilateral mild medullary sponge kidneys were reported in one case (9), whereas in the case described here, a high vaginal urethral meatus was noted. Preoperative imaging of the ureters allows safer resection.

The surgical technique is similar to a completely laparoscopic hysterectomy. Variations in surgical technique included removal of the horn through a small suprapubic incision (7), concurrent use of hysteroscopy (9), and use of an endoscopic stapler (10). All surgeries were effective in relieving severe dysmenorrhea. No major complications were reported.

Regardless of the therapeutic surgical approach, diagnostic laparoscopy is useful in verifying the diagnosis in cases of mullerian dysgenesis. The advantages of laparoscopic resection over laparotomy include reduced postoperative adhesion formation, hospital stay, postoperative pain, and recovery time. The case reported here and the previously successful applications of minimally invasive surgical techniques demonstrate that with appropriately trained surgeons these techniques are becoming the standard treatment of symptomatic noncommunicating rudimentary uterine horns.

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