

ACUTE ABDOMEN CAUSED BY HEMATOMETRA AND HEMATOSALPINX FOUR MONTHS FOLLOWING RIGHT OOPHORECTOMY IN A TEENAGER WITH RIGHT PERITUBAL ADHESIONS, UNICORNUATE UTERUS AND A RIGHT NONCOMMUNICATING RUDIMENTARY HORN

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Unicornuate uterus with a rudimentary horn is a rare uterine malformation resulting from the arrest of one of the müllerian ducts during embryologic development [1]. The incidence of this congenital uterine anomaly in the female population is estimated to be about 0.00001% [2]. An unicornuate uterus with a noncommunicating horn and functional endometrium constitute a distinct subgroup with clinical repercussion, because it can cause hematometra and endometriosis leading to progressive dysmenorrhea, lower abdominal pain, infertility, and obstetrical complications, such as rupture of pregnant rudimentary horn and ectopic pregnancy [1,3,4]. Unfortunately, this type of uterine anomaly is usually unrecognized until these complications develop. Here, we report a case of a unicornuate uterus with noncommunicating functional horn complicated with hematometra and hematosalpinx, in which severe dysmenorrhea and an acute abdominal pain developed after an oophorectomy. An iatrogenic etiology is discussed.

A 17-year-old nulligravida woman presented to our emergency department with severe right lower abdominal pain. Intermittent right lower abdominal pain was noted for about 3 weeks and became persistent. Progressive dysmenorrhea was also noted for about 4 months. Physical examination revealed diffuse tenderness of the lower abdomen with rebounding pain

and voluntary guarding of the right lower quadrant. Her leukocyte count was 11,000/cm³, the urinary pregnancy test was negative, and plain abdominal X-ray showed moderate bowel air accumulation in the right lower quadrant. According to her statement, she denied any systemic disease since childhood. However, she underwent a laparoscopic right oophorectomy at a local clinic 4 months previously because of an ovarian tumor, but she was not informed of any genital tract anomalies at that time. She had been suffering from dysmenorrhea since her menarche at the age of 13, but her symptom worsened after the surgical procedure. The appearance of her external genitalia was unremarkable, with normal pubic hair and an intact hymen. A transabdominal ultrasound scan suggested a bicornuate uterus with a homogeneous hypoechoic sand-like lesion at the right side of the cavity, consistent with hematometra (Figure 1). A computed tomography scan revealed an absent right kidney, a right adnexal cyst, and a bicornuate uterus with a right hematometra (Figure 2A). Pelvic examination under anesthesia showed a single cervix deviated to the left side without vaginal anomaly. Uterine sounding showed no connection between cervix and the right adnexal mass. The patient underwent laparotomy that demonstrated a left unicornuate uterus with cervix and a noncommunicating right rudimentary horn attached to the unicornuate uterus at the base, dilated by a large hematometra with chocolate-like content (Figures 2B, 2C and 3). Right hematosalpinx and multiple endometriotic implants were found in the peritoneum of the Douglas pouch. Removal of the rudimentary horn, ipsilateral

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salpingectomy, and electrocauterization of endometriosis were performed. The postoperative course was uneventful, and the patient was discharged on the third postoperative day. Histology was consistent with a noncommunicating rudimentary horn with a functional

endometrium, hematometra and hematosalpinx. Six months after the surgery, the patient continued to have regular menstrual cycles without dysmenorrhea.

Patients with a noncommunicating uterine horn and a functional endometrium are at risk for developing several complications, such as progressive dysmenorrhea, acute and chronic pelvic pain, infertility, tubal ectopic pregnancy, and rupture of the pregnant rudimentary horn [1,3,5,6]. If this uterine anomaly is unrecognized or misdiagnosed during a previous surgical procedure, an obstruction of the retrograde outflow of the rudimentary horn may lead to surgical complications.

To date, at least six cases of unicornuate uterus with noncommunicating functional horn complicated with dysmenorrhea and abdominal pain following pelvic operations, such as tubal sterilization (two cases), cesarean section (two cases) and unilateral adnexectomy (two cases), have been reported (Table) [7–11]. The reported intervals between the first surgical procedure and acute symptoms requiring immediate surgery varied from 5 months to 8 years. The Table shows that three of the six cases had unilateral renal agenesis and



Figure 1. Transabdominal ultrasound shows a 4.3 × 3.0 cm homogenous content inside the rudimentary horn that is consistent with hematometra (H).

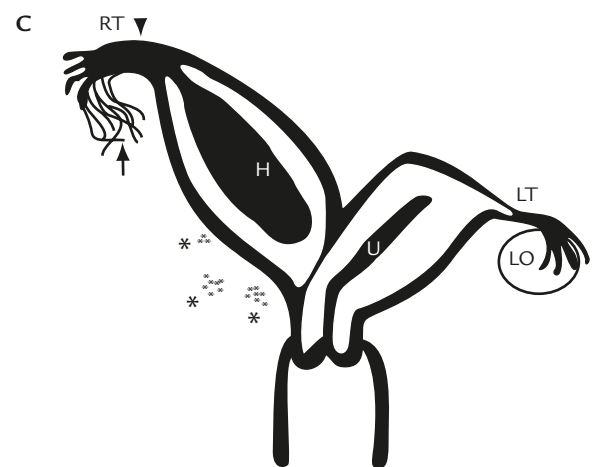
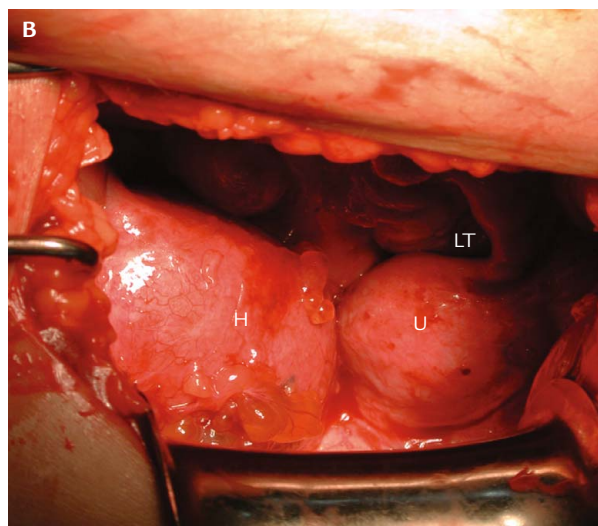


Figure 2. (A) Computed tomography scan reveals two uterine cavities with fluid accumulation in the right side. (B) Gross image of the right rudimentary horn and left unicornuate uterus during laparotomy. (C) An illustration of the uterine anomaly with right hematosalpinx (arrowhead), right peritubal adhesions (arrow) and multiple endometriosis foci over the peritoneal surface (*). H=hematometra; LO=right ovary; LT=left tube; RT=right tube; U=unicornuate uterus.

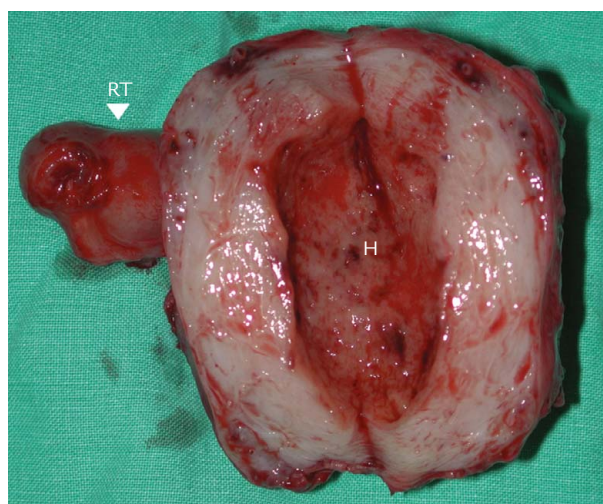


Figure 3. Gross appearance of the right rudimentary horn (H) and right tube with hematosalpinx (RT).

all, except one, were associated with endometriosis. All cases had a good outcome after removal of the rudimentary horn. Variation of intervals has been noted to be dependent on the degree of obstruction of the endometrial activity within the rudimentary horn [6]. Müllerian duct anomalies are usually associated with renal aberrations, and the presence of unilateral renal agenesis will predict an ipsilateral müllerian anomaly in more than half of the cases [6,12]. Noncommunicating functional horns have been noted to be associated with endometriosis as a result of the retrograde menstruation [1,3,5]. In the present case, there were a right rudimentary horn with hematometra, right hematosalpinx, right renal agenesis and pelvic endometriosis, and severe dysmenorrhea and an acute abdomen developed 4 months after the initial operation. The short interval of 4 months between the development of hematometra and the previous oophorectomy in our case was likely because of multiple iatrogenic factors such as severe postoperative adhesions around the right adnexa and the complete obstruction to the retrograde menstruation.

Early removal of the rudimentary horn and its ipsilateral tube is recommended upon diagnosis to avoid future complications and to relieve symptoms [5,6,9]. However, diagnosis of a unicornuate uterus with a noncommunicating functional horn can be difficult because of the inexperience of the surgeons, the lack of a complete work-up or a small surgical field [7,10]. Jayasinghe et al [6] suggested that thorough preoperative work-ups including detailed history taking, complete pelvic examination and complementary imaging studies, such as ultrasonography, intravenous pyelography, computerized tomography and magnetic resonance

Table. Reported cases with unicornuate uterus, noncommunicating functional horn, dysmenorrhea, and abdominal pain associated with previous surgical procedures

Author	Age (yr)	Müllerian duct abnormalities	Previous surgery	Interval	Symptom	Associated endometriosis	Unilateral renal agenesis	Outcome following removal of rudimentary horn
Robischon et al [7]	27	Unicornuate uterus + right noncommunicating horn	Tubal sterilization	5 mo	Dysmenorrhea	No	No	Good
Chang et al [8]	30	Unicornuate uterus + right noncommunicating horn	Cesarean section	8 mo	Dysmenorrhea and progressive abdominal pain	Yes	Yes	Good
Kriplani et al [9]	24	Unicornuate uterus + right noncommunicating horn	Cesarean section	2 yr	Dysmenorrhea and progressive abdominal pain	Yes	No	Good
Lee et al [10]	39	Unicornuate uterus + left noncommunicating horn	Tubal sterilization	6 yr	Dysmenorrhea	Yes	No	Good
Tanaka et al [11]	32	Unicornuate uterus + left noncommunicating horn	Left adnexectomy	3 yr	Dysmenorrhea	Yes	Yes	Good
Present case	17	Unicornuate uterus + right noncommunicating horn	Right oophorectomy	4 mo	Dysmenorrhea and progressive abdominal pain	Yes	Yes	Good

imaging, are mandatory. In doubtful cases, laparoscopy in combination with hysteroscopy, a tubal dye study, and an intraoperative ultrasonography are useful [7,11,13].

In summary, we present a case of acute abdomen caused by hematometra and hematosalpinx 4 months following a right oophorectomy in a teenager with severe right peritubal adhesions, a unicornuate uterus and a right noncommunicating rudimentary horn. We emphasize the importance of early recognition and intervention of the congenital uterine anomaly.

References

1. Liu MM. Unicornuate uterus with rudimentary horn. *Int J Gynaecol Obstet* 1994;44:149–53.
2. Atmaca R, Germen AT, Burak F, Kafkasli A. Acute abdomen in a case with noncommunicating rudimentary horn and unicornuate uterus. *JSL* 2005;9:235–7.
3. Heinonen P. Unicornuate uterus and rudimentary horn. *Fertil Steril* 1997;68:224–30.
4. von Eye Corleta H, Villodre LC, Reis R, Capp E. Conservative treatment for a non-communicating rudimentary horn. *Acta Obstet Gynecol Scand* 2001;80:668.
5. Chakravarti S, Chin K. Rudimentary uterine horn: management of a diagnostic enigma. *Acta Obstet Gynecol Scand* 2003;82:1153–4.
6. Jayasinghe Y, Rane A, Stalewski H, Grover S. The presentation and early diagnosis of the rudimentary uterine horn. *Obstet Gynecol* 2005;105:1456–67.
7. Robischon K, Baram D, Phipps W. Presentation of a müllerian anomaly with outflow obstruction after tubal ligation. *Fertil Steril* 1996;65:866–8.
8. Chang CY, Chang SY, Changchien CC, Lui CC, Huang HW. Hematometra of the rudimentary horn of a unicornuate uterus resulting from cesarean section. *Am J Obstet Gynecol* 2001;185:1263–4.
9. Kriplani A, Agarwal N. Hysteroscopic and laparoscopic guided miniaccess hemihysterectomy for non-communicating uterine horn. *Arch Gynecol Obstet* 2001;265:162–4.
10. Lee CY, Cheng WC, Hwang JL. Delayed severe dysmenorrhea after tubal ligation in a patient with rudimentary horn: a case report. *Taiwan J Obstet Gynecol* 2005;44:87–90.
11. Tanaka Y, Asada H, Uchida H, Maruyama T, Kuji N, Sueoka K, Yoshimura Y. Case of iatrogenic dysmenorrhea in non-communicating rudimentary uterine horn and its laparoscopic resection. *J Obstet Gynaecol Res* 2005;31:242–6.
12. Goluda M, St Gabrys M, Ujec M, Jedryka M, Goluda C. Bicornuate rudimentary uterine horns with functioning endometrium and complete cervical-vaginal agenesis coexisting with ovarian endometriosis: a case report. *Fertil Steril* 2006;86:462.e9–11.
13. Takeuchi H, Sato Y, Shimanuki H, Kikuchi I, Kumakiri J, Kitade M, Kinoshita K. Accurate preoperative diagnosis and laparoscopic removal of the cavitated non-communicated uterine horn for obstructive Müllerian anomalies. *J Obstet Gynaecol Res* 2006;32:74–9.