J. Obstet. Gynaecol. Res. Vol. 31, No. 3: 242–246, June 2005

Case of iatrogenic dysmenorrhea in non-communicating rudimentary uterine horn and its laparoscopic resection

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Abstract

A case of non-communicating rudimentary uterine horn is presented and is a characteristic example of the way that surgical treatment can exacerbate dysmenorrhea by blocking retrograde menstruation. There is always some risk of missing the true diagnosis in cases of uterine abnormalities, even by direct inspection, but intraoperative laparoscopic ultrasonography can be an invaluable tool in defining anatomy. As laparoscopic resection has become the standard procedure for the treatment of unicornuate uterus, laparoscopic ultrasonography can be a useful tool for providing anatomical information to the operating surgeon about the patient.

Key words: laparoscopic ultrasonography, laparoscopy, rudimentary uterine horn, unicornuate uterus.

Introduction

Unicornuate uterus with rudimentary horns, a rare uterine anomaly, is caused by the arrested development of one of the Müllerian ducts. The American Fertility Society (AFS) has classified this anomaly into four subgroups: rudimentary horn with cavity communicating with a unicornuate uterus (IIa); rudimentary horn with cavity non-communicating (IIb); with no cavity (IIc); and without horn (IId).[1] Of these, type IIb is the most common, and the most clinically significant, because it is associated frequently with progressive severe dysmenorrhea, hematometra, pyometra, and endometriosis attributed to retrograde menstruation.[2,3]

This report describes the diagnosis and treatment of a patient with rudimentary uterine horn in which previous adnexectomy triggered her dysmenorrhea and hematometra. Laparoscopic resection of the horn relieved her symptoms completely. The surgery was carried out with the assistance of laparoscopic ultrasonography, which is also discussed.

Case Report

A 32-year-old, Japanese woman, gravida 1, para 1, presented with a chief complaint of progressive severe dysmenorrhea for 3 years. She experienced menarche at the age of 12 years and had never had trouble during menstruation. Three years before she presented, a left ovarian endometrioma was found incidentally, and cystectomy was scheduled at another hospital. According to the referring letter from that hospital, a bicornuate uterus and left severe hydrosalpinx were unexpected operative findings, and left adnexectomy was carried out rather than cystectomy. Adhesiolysis was carried out as well, because of massive endometriosis with a half-closed cul-de-sac. The patient started to experience progressive worsening of her dysmenorrhea postoperatively and was placed on gonadotropin-releasing

Received: October 18 2004.
Accepted: February 21 2005.
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hormone agonist (GnRHa) for 6 months with transient pain relief. However, when her regular menstrual cycle returned, so did the dysmenorrhea. The pain was even worse than it had been before and she was again placed on GnRHa. After 6 months of GnRHa therapy, she was referred to our hospital for a second opinion.

Speculum examination at the first visit showed a single cervix with no vaginal anomaly. Bimanual examination showed a uterus deviated to the right, with a tender mass on the left side. Transvaginal ultrasound examination showed an enlarged left cornua measuring 5.5 × 5.7 cm in diameter, its cavity was filled with both low- and high-level echoes, consistent with hematometra (Fig. 1). Hysterosalpingography showed a unicornuate uterus deviated to the right side, with normal spill of dye from the ipsilateral tube (Fig. 2). Magnetic resonance imaging (MRI) delineated a left hematometra as well as normal-appearing right hemiuterus. These findings suggested a unicornuate uterus with a left-sided rudimentary uterine horn, rather than bicornuate uterus. Intravenous pyelography was carried out, showing the absence of a left kidney and ureter. The right ureter was slightly dilated, assumedly secondary to extrinsic compression.

The laparoscopic resection of the noncommunicating rudimentary horn was carried out with the purpose of relieving the patient's symptoms. During the operation, the left rudimentary uterine horn was found to be attached to the right uterus, at its lowest part (Fig. 3). As the uterus and horn were both atrophic, most likely because of the long-term effect of GnRHa, it was not easy to identify both the atrophic rudimentary horn and a distinct plane between the corpus and the rudimentary horn. Additionally, there was the possibility that the subtype was not IIb (noncommunicating) but IIa (communicating), and that the communication was simply blocked by acquired adhesion between the horn-cavity and the uterine cavity. Thus, laparoscopic ultrasonography was used to find the correct plane, and to confirm the preoperative diagnosis, which showed that the uterine cavity and the horn-cavity were clearly separated by myometrium of the rudimentary horn (Fig. 4). Laparoscopic ultrasonography was carried out using 10-mm diameter, 7.5-MHz steerable transducers (Aloka, Tokyo, Japan; Fig. 5). During the procedure, the pelvis was filled with saline to obtain optimal imaging. The ultrasound probe was introduced through 12 mm trocar and the uterus and the rudimentary horn examined. With the confirmation that the type of anomaly was type IIb, the round ligament was grasped and transected, opening up both the anterior and posterior leaves of the broad ligament. The broad ligament surrounding the rudimentary horn was dissected downward. As the rudimentary horn was dissected using an ultrasonic scalpel, the main blood supply coursing below the horn was identified and it was easily coagulated by bipolar cautery. The rudimentary horn appeared to be attached to the unicornuate uterus by a band of fibrous tissue and was separated with little difficulty. A 3–4-cm

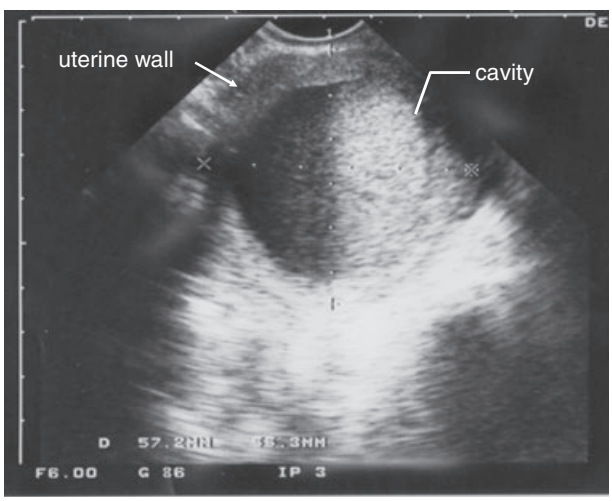

Figure 1 Transvaginal ultrasonogram of the left rudimentary horn. The cavity of the horn is filled with hypo- and hyperechoic content, suggesting hematometra. The size of the hematometra was 57 × 55 mm.

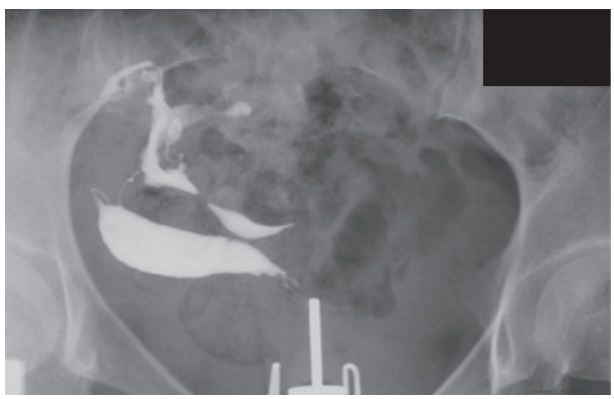

Figure 2 Hysterosalpingography. The uterine corpus was deviated to the right side with normal spill of dye from the ipsilateral tube, suggesting a unicornuate uterus with a left-sided rudimentary uterine horn.

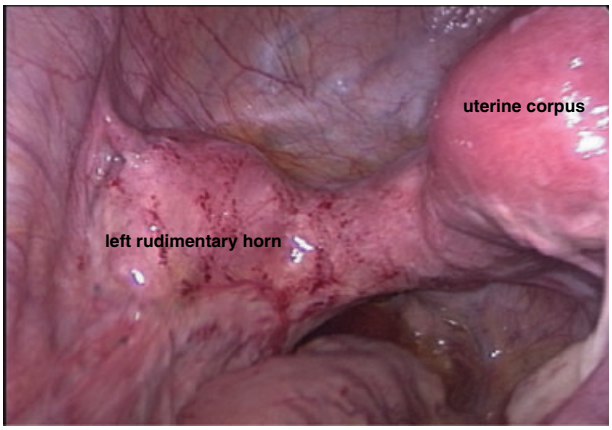

Figure 3 Laparoscopic view of the unicornuate uterus with left rudimentary horn. The uterine corpus is deviated towards the right.

Figure 5 Laparoscopic ultrasound probe.

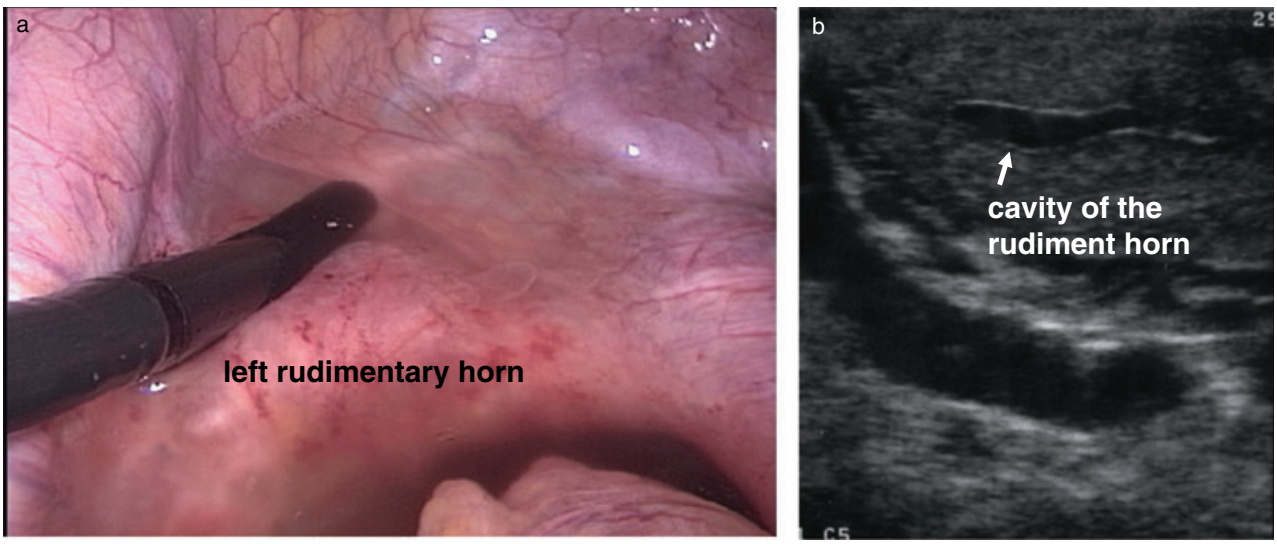

Figure 4 Laparoscopic ultrasonogram. (a) The probe is attached onto the rudimentary horn. Note that water fills the pouch of Douglas. (b) Ultrasonographic image of the rudimentary horn.

incision was made in the posterior vaginal wall, and the specimen was removed through the incision *en block*. On inspection of the whole pelvis, no endometriotic lesion was observed and the extent of adhesions was very minor. Blood loss was minimal. There was no perioperative complication, and the patient was discharged on the fourth postoperative day. The pathology report on the surgical specimen confirmed fragments of adenomyosis and normal endometrial lining. At the present time, 6 months after surgery, the patient reports complete resolution of dysmenorrhea.

Discussion

The true incidence of unicornuate uterus is hard to determine because so many patients with this anomaly go unrecognized because they never develop any obstetric or gynecologic symptoms. The incidence has variously been reported to be between 0.2 and 10%.[4,5] Approximately 90% of unicornuate uteruses with a rudimentary horn are non-communicating with a cavity (type IIb). Because concomitant urinary tract anomalies are present in 60% of cases of rudimentary uterine

horn, preoperative evaluation of the urinary tract is a prerequisite and care must always be taken to dissect and identify the ureters before resection.[6,7]

The uterine anomaly of this patient was initially diagnosed as a bicornuate uterus at the time of laparotomy, and adnexectomy caused retrograde obstruction of menstruation. We believe that her dysmenorrhea and hematometra were the consequence of this obstruction, as communicating horns usually contain a number of functional endometrial glands.[8] The correct diagnosis and resection of the rudimentary horn at the patient's initial surgery should of prevented all of her symptoms. A similar case was once reported in India, in which a unicornuate uterus with a rudimentary horn was missed during the initial procedure, and unilateral salpingectomy caused severe hematometra and dysmenorrhea.[9] Despite the presence of severe endometriosis observed during the initial laparotomy, we found no endometriomas or adhesions. This resolution is most likely another consequence of tubal blockage.

The diagnosis of uterine anomalies is sometimes very difficult and it can be overlooked even at the time of laparotomy.[10] MRI provides an excellent method of differentiating between different types of uterine anomaly. Although MRI is 96–100% accurate in classifying uterine anomaly, physicians should remain aware of the limitations of non-invasive diagnostic methods, such as MRI, transvaginal ultrasonography, and hysterosalpingography.[11] Laparoscopy provides additional information beyond radiologic tests of pelvic anatomy. In particular, differentiating type IIa from type IIb might be very difficult because the non-communication might be caused by fibrous adhesions acquired later in life, rather than myometrium.[9] If it is not type IIb but IIa, transcervical adhesiolysis is sufficient to alleviate symptoms. Thus, we would like to emphasize the importance of confirming the diagnosis before carrying out any definitive surgical procedure and also during the surgical procedure. The case presented here shows that laparoscopic ultrasonography is a valuable method for confirming diagnosis and providing anatomical information to the operating surgeon.

In addition to the variants defined in the classification system of the AFS, rudimentary horns can vary in another ways, that is, they might be part of the unicornuate uterus, or attached to it by a band of tissue.[3] Perrotin *et al*. reviewed 10 cases of laparoscopic removal of Müllerian remnants.[12] Of these, three cases were firmly attached to the unicornuate uterus, five were attached by a fibrous band, and two were not specified. If it is firmly attached, it might be very difficult to achieve the resection because there is no pedicle and the blood supply to the horn courses both lateral to the unicornuate uterus and right below the rudimentary horn. In contrast, if it is attached by a fibrous band, it is relatively easy to dissect the horn because the plane of dissection can be usually identified clearly. Although our case was the latter type, the plane of dissection was not easily identified because a thick broad ligament surrounded the rudimentary horn. Again, intraoperative laparoscopic ultrasonography was of great help in evaluating the relationship between unicornuate uterus and the rudimentary horn.

In conclusion, we present the case of laparoscopic resection of a rudimentary uterine horn in a patient with dysmenorrhea. Symptoms were exacerbated by antecedent adnexectomy, which blocked retrograde menstruation. Great care must be paid to accurately diagnose uterine anomaly and it should be noted that misdiagnosis can severely impact a patient's quality of life. Operative laparoscopy is rapidly becoming the standard treatment of symptomatic non-communicating rudimentary uterine horns,[12] and laparoscopic ultrasonography is a useful tool for providing anatomical information to the operating surgeon.

References

1. American Fertility Society. The American Fertility Society classifications of adnexal adhesions, distal tubal occlusion, tubal occlusion secondary to tubal ligation, tubal pregnancies, Müllerian anomalies and intrauterine adhesions. *Fertil Steril* 1988; **49**: 944–955.
2. Batioglu S, Zeyneloglu HB. Endoscopic management of a case of complete septate uterus with unilateral pyometra. *Gynecol Obstet Invest* 1999; **47**: 144–146.
3. Falcone T, Gidwani G, Paraiso M, Beverly C, Goldberg J. Anatomical variation in the rudimentary horns of a unicornuate uterus: Implications for laparoscopic surgery. *Hum Reprod* 1997; **12**: 263–265.
4. Raga F, Bauset C, Remohi J, Bonilla-Musoles F, Simon C, Pellicer A. Reproductive impact of congenital Müllerian anomalies. *Hum Reprod* 1997; **12**: 2277–2281.
5. Donderwinkel PF, Dorr JP, Willemsen WN. The unicornuate uterus: Clinical implications. *Eur J Obstet Gynecol Reprod Biol* 1992; **47**: 135–139.
6. Heinonen PK. Clinical implications of the unicornuate uterus with rudimentary horn. *Int J Gynaecol Obstet* 1983; **21**: 145–150.
7. Schattman GL, Grifo JA, Birnbaum S. Laparoscopic resection of a non-communicating rudimentary uterine horn. *J Reprod Med* 1995; **40**: 219–220.
8. Kadir RA, Hart J, Nagele F, O'Connor H, Magos AL. Laparoscopic excision of a non-communicating rudimentary uterine horn. *Br J Obstet Gynaecol* 1996; **103**: 371–372.

9. Kriplani A, Agarwal N. Hysteroscopic and laparoscopic guided miniaccess hemihysterectomy for non-communicating uterine horn. *Arch Gynecol Obstet* 2001; **265**: 162–164.
10. Dimitrova V, Nalbanski B. The echographic diagnosis of a rare congenital uterine anomaly. *Akush Ginekol Sofiia* 1997; **36**: 44–47.
11. Pui MH. Imaging diagnosis of congenital uterine malformation. *Comput Med Imaging Graph* 2004; **190**: 1669–1675.
12. Perrotin F, Bertrand J, Body G. Laparoscopic surgery of unicornuate uterus with rudimentary uterine horn. *Hum Reprod* 1999; **14**: 931–933.