

Laparoscopic Removal of Nonseparated Cavitated Horn in Unicornuate Uterus: Surgical Aspects and Long-Term follow-up

Massimo Candiani, M.D.¹, Nevio Ciappina, M.D.², Francesco Fedele, M.D.², Valentino Bergamini, M.D.³,
Alessandro Ferdinando Ruffolo, M.D.¹, Fabio Parazzini, M.D.²

1. Obstetrics and Gynecology Department, IRCCS San Raffaele University, Milan, Italy

2. Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, University of Milan, Milan, Italy

3. Obstetrics and Gynecology Division B, Azienda Ospedaliera Integrata, University of Verona, Verona, Italy

Abstract

Background: In unicornuate uterus cases, when the rudimentary horn is very close to the uterus and is firmly attached, laparoscopic surgery can be very challenging due to the danger of massive bleeding and the possibility of damaging the healthy hemiuterus. The aim of study is to verify if the laparoscopic resection of the horn site of hematometra, when solidly attached to the unicornuate uterus, is safe and effective.

Materials and Methods: This is a retrospective analysis of prospectively collected data in a tertiary referral centre. From 2005 to 2021, a total of 19 women were diagnosed with unicornuate uterus with cavitated noncommunicating horn (class II B). We reviewed the original documentations of the patients and created a database. The follow-up results were assessed by questionnaires answered by the patients. In all cases, the chosen treatment was: laparoscopic removal of the rudimentary horn together with the ipsilateral salpinx and reconstruction of the myometrium of the hemiuterus. Statistical Package for Social Science (SPSS) version 21.0 was used to perform data analysis. We decided to calculate continuous variables in terms of mean and standard deviation (SD) or as median and interquartile range (IQR), as appropriate. Instead, categorical variables were expressed in terms of percentage.

Results: Five patients (12-18 years old) with unicornuate uterus and rudimentary horn with hematometra and broadly connected to the hemiuterus were operated laparoscopically. The surgical procedure was successful in all cases. No major complications were recorded. Postoperative course was uneventful. In the follow-up in all cases dysmenorrhea and pelvic pain disappeared. Three patients sought to become pregnant and have children. They had in total 4 pregnancies with 2 abortions in the 1st trimester and two pregnancies with premature births at the 34th and 36th weeks. No serious gestational complications were recorded and the pregnancies ended with caesarean sections due to breech presentation.

Conclusion: Overall, for the rudimentary horn solidly attached to the unicornuate uterus, the laparoscopic resection of the horn site of hematometra seems to be safe and effective.

Keywords: Dysmenorrhea, Mullerian Anomalies, Unicornuate Uterus

Citation: Candiani M, Ciappina N, Fedele F, Bergamini V, Ferdinando Ruffolo A, Parazzini F. Laparoscopic removal of nonseparated cavitated horn in unicornuate uterus: surgical aspects and long-term follow-up. Int J Fertil Steril. 2023; 17(2): 145-150. doi: 10.22074/IJFS.2022.543827.1231.

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Introduction

The prevalence of unicornuate uterus is estimated close to 0.1% in the general female population, 0.5% in infertile patients, and 2% in those with a history of miscarriage (1). Its anatomical presentation is highly variable and is associated with the presence of a contralateral rudimentary horn in about 74-90% of cases (2). Approximately 25% of these horns are cavitated and not communicating with the hemiuterus (class IIB according to the American Fertility Society (AFS) classification of Müllerian anomalies) (3, 4). Such horns may be the site of cryptomenorrhea and require surgical removal to alleviate symptoms and to avoid severe complications as ectopic pregnancy or

extensive endometriosis. Currently, laparoscopic resection is considered the standard treatment to remove the Mullerian remnants and the results are very satisfactory when the rudimentary horn is lateralized and becomes independent of the hemiuterus (5, 6). Instead, when the rudimentary horn is very close and firmly attached, laparoscopic surgery can be more challenging due to the risks of bleeding and the possible injuries to the integrity of the healthy hemiuterus (7-9).

Here we present the surgical aspects and a long-term follow-up in five patients with cavitated rudimentary horn solidly attached to unicornuate uterus, who underwent laparoscopic removal procedures. We hope that the results shown in this

Received: 28/November/2021, Revised: 07/May/2022, Accepted: 24/May/2022

*Corresponding Address: Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, University of Milan, Milan, Italy

Email: francesco.fedele@unimi.it



Royan Institute
International Journal of Fertility & Sterility

paper help all physicians who focus on adolescent gynecology, in order to gain more confidence with the treatment of this anomaly. The aim of study is to verify if the laparoscopic resection of the horn site of hematometra, when solidly attached to the unicornuate uterus, is safe and effective.

Materials and Methods

From 2005 to 2021 a total of 19 women (12-18 years old) were diagnosed with unicornuate uterus with cavitated noncommunicating horn (class IIA) and underwent laparoscopy. These cases were referred from 2005 to 2017 at Obstetrics and Gynaecology Division of IRCCS Ospedale Maggiore Policlinico, University of Milan, Italy; and from 2018 to 2021 to Obstetrics and Gynaecology Division of San Raffaele University, Milan, Italy. This study was approved by The Ethics Committee of I.R.C.C.S. S. Raffaele Hospital Milano. Protocol Code GARA, register number CE 73/INT/2021.

For all 19 patients we thoroughly reviewed the hospital records and created a database for the analysis of the documentations. Long-term results were assessed by questionnaires given to the patients. The questionnaire asked about dysmenorrhea, pelvic pain and pregnancy, including their number and results, complications, gestational age at delivery, and the mode of delivery. Reviewing the original documentations and the laparoscopic records we found that only five out of these patients had a broad connection of the rudimentary horn with hemiuterus, and these patients constituted the subjects of the present study. The remaining 14 patients had rudimentary horns lateralized and independent, attached to hemiuterus only by a fibrous band.

All five women came to our center because of severe dysmenorrhea and catamenial pain. One of these patients (17 years old) had already received a diagnostic laparoscopy and was taking hormonal treatment (continuous oral contraceptives) to avoid cyclic pain. Two patients, of 12 and 14 years old respectively, were referred to us in emergency conditions because of acute pelvic pain due to a large hematometra.

Preoperative work-up

First, a detailed medical history was obtained from the young patients and their families to determine pubertal development and onset, timing, and the nature of pelvic pain. Physical examination with perineal inspection and rectal/vaginal examination was performed to exclude other obstructing anomalies such imperforate hymen and obstructing vaginal septa. All patients underwent abdominal and transrectal/transvaginal ultrasound in order to define the characteristics of the uterus, the presence of hematosalpinx and hemoperitoneum. When the presence of a rudimentary uterine horn was found, in every case its relationship with the uterus was determined. Based on its shape, we deemed reasonable to apply the formula used to calculate the volume of an ellipsoid ($V=D1 \times D2 \times D3 \times 0.5223$) (10) to determine the size of the rudimentary horns. Magnetic resonance

imaging (MRI) was used when the ultrasound examination was inconclusive or unclear. Before the operation, a counselling session was conducted with each patient and their parents. The anatomical malformation, the therapeutic possibilities and the risks were illustrated in detail.

Surgery

In all cases the chosen treatment was: resection of the rudimentary horn together with the ipsilateral salpinx, reconstruction of the myometrium of the hemiuterus, peritoneization, and finally attachment of the round ligament of the horn removed to the hemiuterus. Laparoscopy was performed after menstruation, because the presence of a large hematometra facilitates identification and dissection of the rudimentary horn. At the beginning of the procedure, the appearance of the uterine shape and the morphology of the adnexa was assessed in the peritoneal cavity. Moreover, the presence of hemoperitoneum and endometrial localization were evaluated. The seat of the rudimentary horn and its relationship with the hemiuterus was evaluated, and those that were distant and independent were distinguished from the close and widely connected ones (Fig.1).

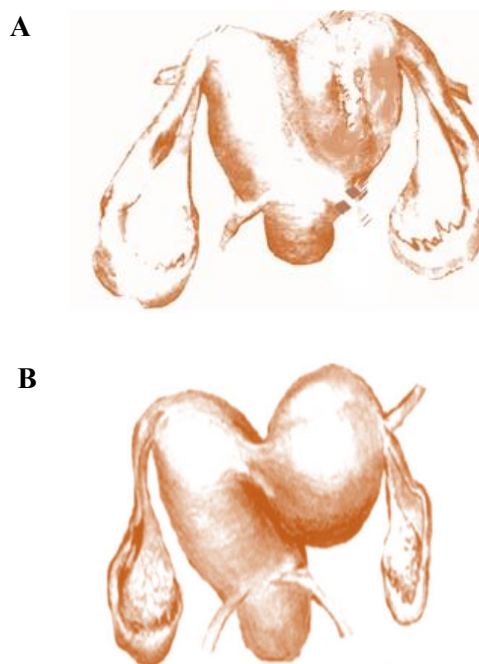


Fig.1: Unicornuate uterus with rudimentary cavitated horn. **A.** The horn is firmly attached to the hemiuterus and **B.** The horn is minimally connected by a fibrous band.

To confirm the side of the rudimentary horn and identify its boundaries, two preliminary procedures are typically performed: i. Dye test with a diluted solution of methylene blue, and ii. Hysteroscopic transillumination of the hemiuterus with simultaneous switching off of the laparoscopic light for the spatial identification of its endometrial cavity. With the aid of these two procedures, the boundaries of the horn to be removed with respect to the hemiuterus were outlined using bipolar forceps. The presence of endometriosis was classified according to

AFS guidelines (11) and treated appropriately.

The first steps of the surgery were: coagulation and separation of the round ligament, the isthmic portion of the tuba and the utero-ovarian ligament ipsilateral to the rudimentary horn. Transection of the round ligament opened up both anterior and posterior leaves of the broad ligament and exposed the vascular pedicle of the rudimentary horn and, below, ureter, which is kept well away from the operative field. Then, applying an

appropriate traction, the rudimentary horn is definitively separated from the hemiuterus by monopolar coagulation. Finally the mesosalpinx was cauterized and cut, allowing the removal of the tube. Once dissection was completed, the bloody area, in which the hemiuterus was connected with the rudimentary horn, was closed with one layer of interrupted sutures similar to laparoscopic myomectomy. The round ligament of the rudimentary horn was then attached to the hemiuterus, in order to prevent its lateralization (Fig.2).

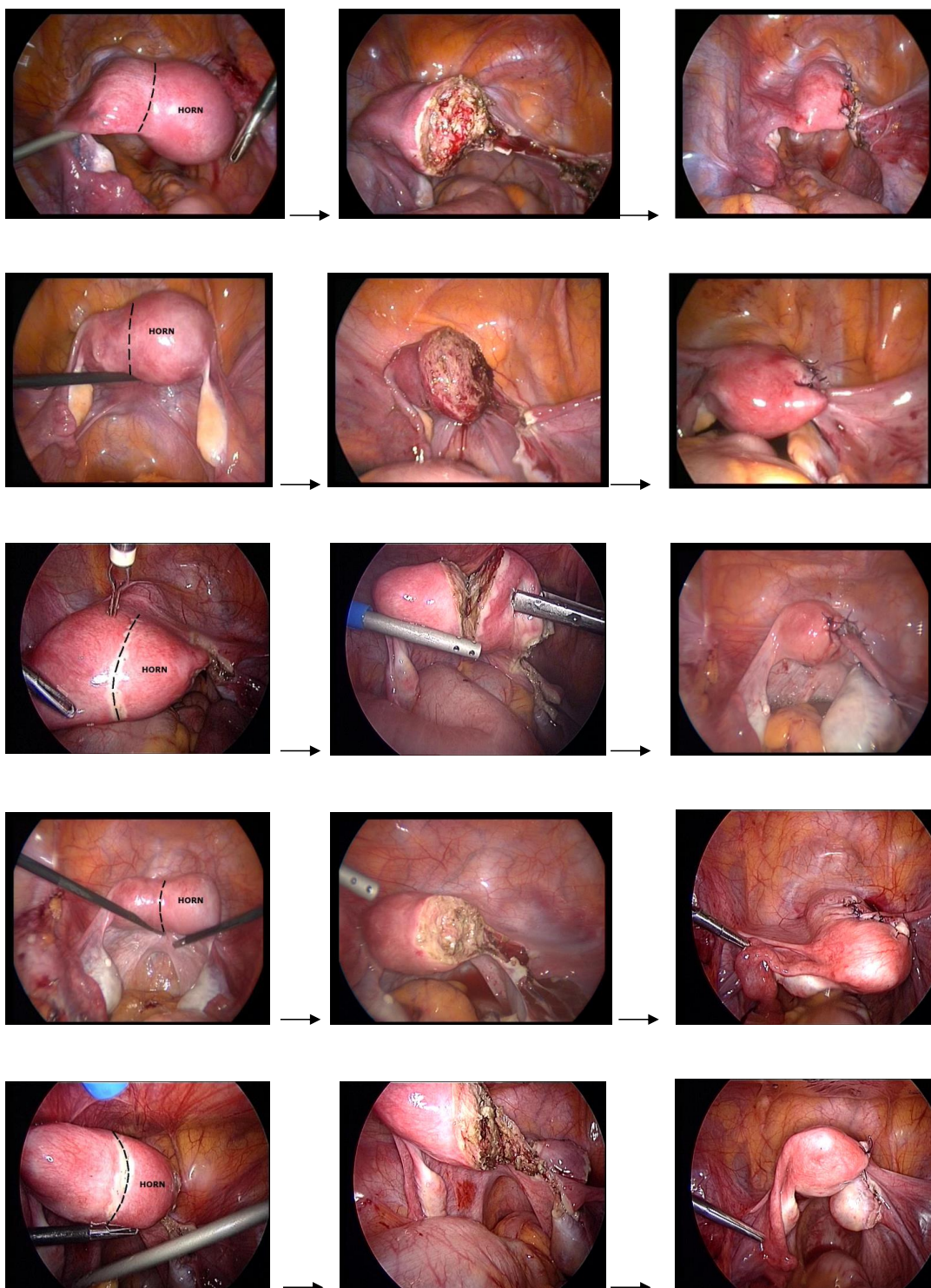


Fig.2: Sugical steps in the five cases.

The excised rudimentary horns were removed by morcellation after opening the horn and checking its contents. Colpotomy or enlargement of the suprapubic trocar site may be alternative methods (6).

Statistical analysis

We utilized Statistical Package for Social Science (SPSS, IBM, USA) version 21.0 to analyze data. We decided to calculate continuous variables in terms of mean and standard deviation (SD) or as median and interquartile range (IQR), as appropriate. Instead, categorical variables were expressed in terms of percentage.

Results

Characteristics of the 5 patients with unicornuate uterus with non-separated uterine horns are reported in Table 1. The hemiuterus cavity was never opened during the removal of the rudimentary horn. In two cases (2/5=40%) it was necessary to detach the bladder fold to expose the vascular pedicle well and to be able to coagulate it. The surgical procedure was successful in all cases. No major complications were recorded. Mean operating time was 48 (SD ± 25) minutes, while mean intraoperative blood loss was 110 (SD ± 30) ml. Postoperative course was uneventful and the mean hospital stay duration was 2.5 (SD ± 1.5) days. Our median follow-up was 8.5 (IQR 3-14) years. The first checkup after surgery was done after the first menstruation. In all cases, the signs and related symptoms of obstruction (dysmenorrhea and pelvic pain) were completely resolved as a result of the procedure. Three patients (3/5; 60%) married and had children. The first case had a miscarriage at 11 weeks of pregnancy. The second one had a miscarriage at 7 weeks of pregnancy, followed by a pregnancy up to 34 weeks, while the third woman had an uneventful pregnancy up to 36 weeks. Both deliveries (2/2; 100%) were caesarean due to breech presentation of the fetus.

Table 1: Characteristics of the 5 patients with unicornuate uterus with non-separated uterine horns

Characteristics	n=5
Age at presentation, median (IQR) (Y)	14.5 (12-18)
Interval between menarche and presentation, median (IQR) (Y)	2.7 (1-5)
Dysmenorrhea	5 (100)
Pelvic pain	5 (100)
Horn side	
Right	5 (100)
Left	0 (0)
Size of the rudimentary horns, median (IQR) (ml)	7.94 (39.7-13.6)
Mild Endometriosis	2 (40)
Hematosalpinx	3 (60)
Urinary Tract Anomaly (Horseshoe Kidney)	1 (20)

Data are presented as n (%). IQR; Interquartile range.

Discussion

Our experience confirms the findings of previous studies (12, 13) that even rudimentary horns closely

related to unicornuate uterus can be safely and effectively removed laparoscopically. In the past, many authors recommended the use of laparotomy in these cases, based on the danger of bleeding from the myometrium of the hemiuterus, and also because removal of a rudimentary horn very close and firmly attached to the hemiuterus leaves a larger myometrial defect on the latter, which must then be sutured (8, 14). In our cases the following were very useful: i. The preliminary marking of the myometrial section, ii. The use of a solid traction on the horn to be removed, iii. The meticulous coagulation of the myometrial arcuate arteries during dissection and iv. The reconstruction of the hemiuterus wall by means of solid interrupted stitches.

In similar cases, many researchers suggest a preoperative treatment with GnRH-a (14) or danazol (15) in order to reduce the size of the hematometra before surgery. We disagree with this pre-treatment, since it reduces the volume of the hematometra. All of our 5 cases were operated on during the first post-menstrual phase to facilitate identification of the rudimentary horn boundaries that are clearer when the cavity is distended by menstrual blood. The fallopian tube ipsilateral to the rudimentary horn has always been removed to prevent a cornual or tubal pregnancy, as the incidence of transperitoneal migration of gametes in humans is high (16). Nonetheless, instead of removing it at the same time of the uterine horn, we deemed more appropriate to perform it at the end of the surgery. In this way, the surgery results easier and more feasible.

This report is specifically notable because of the young age of the patients enrolled in the study. In the literature it is reported that some cases of functional rudimentary horns can become symptomatic in the third decade of life (17). In our cases, the timely recognition of the anatomical cause of dysmenorrhea also meant that endometriosis did not develop in severe stages.

All the rudimentary horns of our cases were located on the right side of the hemiuterus. The present series is too small to allow assessment of this anatomical variation. There is an unexplained tendency of the right-sided location of the rudimentary horn in the literature, with a frequency of 62-85.7% (6, 12, 18). The embryologic tendency of this dominance remains unexplained.

Among our case series, just one patient showed a kidney abnormality. The literature usually reports a higher association of renal abnormalities in patients with a unicornuate uterus (36%) (19). We do not know if the close connection of the rudimentary horns to the hemiuterus in our patient affected this scarce presence of urinary system anomaly.

Regarding fertility in the unicornuate uterus, the results of the main studies published in the literature are heterogenous, and the current knowledge depends on small retrospective series or case reports. They are however all characterized by an elevated frequency of abortions

and preterm births. The gestational capacity of our three patients whose desire was to have children seems to be in line with the fertility reports on patients with unicornuate uterus in general (20) and in particular with patients with the same characteristics reported by Pados et al. (11). Therefore, at least with regard to the two patients with live births, we can assume that the myometrial defect after surgical intervention did not decrease the integrity of the muscle mass of the hemiuterus.

Other approaches have been proposed in treatment of this malformative variant to resolve pelvic pain and avoid catastrophic potential obstetric consequences (1). The first one involves the hysteroscopic creation of a way of communication between the hemiuterus cavity and that of the rudimentary horn (21) and the second involves the reunification of the hemiuterus and the rudimentary horn by means of a metroplasty according to Strassmann (22), performed by laparoscopy or robotic surgery. The former creates a communication between the orthotopic endometrium and the the cavity of the rudimentary horn, while the latter comprehends an incision of the myometrium add the following reconstruction. The first option (21) solves the problem of catamenial pain as it connects the rudimentary horn with the hemiuterus, but is potentially disastrous from the reproductive point of view as it demolishes the integrity of the hemiuterus, without any repair, on the side of the rudimentary horn. The second option was reported in three recent papers. In the first article, Bhagavath et al. (22) report on two patients with unicornuate uterus and non-communicating cavitated rudimentary horn adhering to the hemiuterus, in which instead of the traditional resection of the rudimentary horn, a Strassmann metroplasty was performed using the assistance of a robot. The anatomical outcomes were favourable and the painful symptoms disappeared. There are no reports on reproductive follow-ups for these patients. The authors are aware of the potential risk of uterine rupture in the event of pregnancy, but believe it may be low considering the good reproductive outcomes of many myomectomies with extensive scars. The second one (23) is a video article that demonstrates the technical feasibility of laparoscopic metroplasty in a patient with a unicornuate uterus and rudimentary horn site of hematometra. The third one is reported by Zhang et al. (24) and is the most interesting experience. These authors also treated five patients with the same malformation (class II b, AFS classification) with Strassmann metroplasty, performed in one case by laparoscopy and in the remaining 4 using robot-assisted laparoscopy. After an average follow-time of 44 months all patients were relieved of the pain and experienced regular menstruation. Notably, at 2 years from the surgery, one patient conceived by *in vitro* fertilization (IVF) and embryo transfer. She gave birth to twins at 33 weeks and 5 days of pregnancy. She delivered through caesarean section; no postoperative complications were reported. Despite this last encouraging report, further data are required to understand whether the increase in the volume and muscle mass of the hemiuterus obtained with metroplasty can indeed improve the poor reproductive outcome in unicornuate uterus. For

now, the reunification procedure of the rudimentary horn at the hemiuterus has yet to be considered experimental and still requires much caution (25). It should be noted that in the past similar attempts performed in laparotomy (22) had not led to improved obstetrical outcomes and had caused problems of uterine rupture spontaneously or during labour. All these extreme attempts at conservative surgery do not take into account that in all cases, in which the removed uterine horns have been histologically examined, the presence of adenomyosis and hypotrophic endometrium has always been found (10). It is therefore logical that the conservation of these structures is of little use in increasing the potential implant surface of the uterus and may instead expose the recurrence of dysmenorrhea and placental pathology (insufficiency and accretism) in case of pregnancy. Therefore, the current standard therapy for non-communicating uterine horn in the unicornuate uterus remains to be laparoscopic horn resection; as this procedure is fast, efficient, and technically easy. Our experience shows that this is feasible and effective even when the rudimentary horn is firmly attached to the hemiuterus.

Conclusion

At the moment, in front of rudimentary horn solidly attached to the unicornuate uterus, the most appropriate therapeutic option seems to be laparoscopic resection of the horn site of hematometra. Following this procedure, the relief of dysmenorrhea and pelvic pain is persistent and fertility does not appear to be compromised. Our experience confirms the safety of laparoscopic procedure for removal of rudimentary horn. Although, a careful preoperative and intraoperative evaluation is essential for the success of such intervention.

Acknowledgments

There is no financial support and conflict of interest in this study.

Authors' Contributions

M.C., N.C.; Operated on the patients and collected the data. F.F.; Developed the concept and participated in writing the manuscript. V.B.; Participated in its design and revised it critically for important intellectual content. A.F.R.; Helped to draft the manuscript and participated in writing the manuscript. F.P.; Analysed the data and coordinated efforts. All authors have read and approved the final manuscript.

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