CONSERVATIVE TREATMENT OF HERLYN-WERNER-WUNDERLICH SYNDROME: ANALYSIS AND LONG-TERM FOLLOW-UP OF 51 CASES

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ABSTRACT

Objective

The purpose of this study is to analyze the precise anatomical characteristics of a large group of patients with Herlyn-Werner-Wunderlich syndrome togheter with the long-term follow-up and the reproductive performance.

Study design

Retrospective analysis of prospectively collected data. In two tertiary medical centers, from 2008 to 2021, 51 patients with HWWS underwent surgery via the same technique. Presenting symptoms, preoperative investigations, operative management, and long-term follow-up with obstetric outcome were assessed.

Results

The surgical procedure was successfull in all cases with no major complications recorded. Postoperative course was uneventfull. The median follow-up was 6,5 years, with a range from 6 months to 13 years. After surgery, among 14 patients who sought pregnancies, 12 (85,71%) were successful. Of these 11 patients had a total of 22 pregnancies, resulting in spontaneous miscarriage in 27% (6/22), premature birth (<37 weeks) in 36% (8/22) and full-term birth in 36% (8/22). Overall the pregnancies demonstrated had a good course

CONCLUSIONS

Early diagnosis is essential in establishing prompt and correct surgical treatment.

Keywords: female genital anomalies, Müllerian anomalies, Herlyn-Werner-Wunderlich syndrome, OHVIRA syndrome

1. INTRODUCTION

Herlyn-Werner-Wunderlich syndrome (HWWS) is a rare congenital malformation that simultaneously involves the Mullerian and Wolffian ducts. Characterized by the presence of double uterus , obstructed hemivagina and ipsilateral renal agenesis it was first reported, in 1922, by Purslow (1). It is also known as OHVIRA syndrome (Obstructed HemiVagina and Ipsilateral Renal Agenesis).

The incidence of HWWS is estimated at around 0,1 – 3,8 % of Mullerian anomalies which in turn have an incidence estimated between 2-3% (2) of the female population. However, this syndrome is the most common obstructive Müllerian anomaly diagnosed in adolescents after menarche (3). Its very low reported incidence may be attributable to underdiagnosis related to poor awareness of this condition. Consequentely, patients have formerly undergone unnecessary interventions for misdiagnosed pelvic mass, leading to other gynecological complications such as abscess formation and endometriosis from retrograde menstrual flow. Series reports featuring numerous cases with adequate follow-up are lacking in the literature. Moreover, the presenting symptoms and signs are often misleading, preventing an immediate, precise understanding of the case. Thus, the treatments performed are often incongruous, with long-term consequences that are difficult to predict.

The aim of this paper is to analyze the precise anatomical characteristics of a large group of patients with HWWS togheter with long-term follow-up and reproductive performance. Our experience will thus help refine diagnostic criteria and unify treatment protocols.

2. MATHERIAL AND METHODS

From 2008 to 2021 all cases of HWWS referred to the senior author were studied and underwent surgery according to the same technique. These cases were treated from 2008 to 2017 at the State University of Milan, and from 2018 to 2021 at the University Vita Salute Ospedale San Raffaele, Milan.

A total of 59 women were diagnosed with HWWS and were surgically treated. For all 59 patients we revised the original documentations and create a database for the analysis of the data. Long-term results were assessed by questionnaire and obtained for 51 patients. The questionnaire concerned

dysmenorrhea, vaginal discharge, dyspareunia and pregnancy, including their number and results, gestational age at delivery, mode of delivery, and whether the pregnancy was ipsi- or contralateral to the obstructed hemivagina.

Eight patients had incomplete data or did not respond to the questionnaire. Of these cases we have all the reports of the operations, but in two the description of the surgery is not precise and 6 patients did not answer the questionnaire. These patients were excluded from the analysis and therefore our study concerns the remaining 51 patients.

The patients complained of various symptoms: dysmenorrhea , spotting , chronic pelvic pain, vaginal discharge, dyspareunia , fever and acute abdominal pain . Other less common but noteworthy symptoms include urinary retention by hematocolpic compression, recurrent urinary tract infections, and pyocolpos (secondary infection of the blood retained in the obstructed hemivagina).
The preoperative details are shown in Table 1.

The initial evaluation of the patient, in addition to a meticulous collection of the history (beginning, cyclicality, location, irradiation of pelvic pain, vaginal bleeding or purulent), was directed to assess signs of appropriate or delayed puberty and to reveal other abdominal or systemic pathologies. When it was not possible to perform a complete vaginal examination, a rectal examination proved very useful. Patients underwent the following instrumental investigations: abdominal and transvaginal / transrectal ultrasound (all), MRI (if not performed before sending) and hysterosalpingography (HSG) (in selected cases).

The abdominal ultrasound was aimed at highlighting the characteristics of the uterus, the presence of hematocolpos and / or hematometra, as well as the presence of menstrual or inflammatory fluid in the peritoneum, and provide information on the state of the urinary system. In transvaginal/transrectal ultrasound the sonographer (with least 5-25 years experience in gynaecological imaging) specified the characteristics of the uterus , didelphic vs septate (4). The sonographer then, sought to clarify whether the hemivagina was related to one of the three types traced by Rock et al. (5) : 1. without any communication with the contralateral Müllerian axis; 2. complete imperforate and with isthmic communication between the two hemiuteri; 3. with incomplete obstruction and no communication between the two hemiuteri (Figure 1).

The MRI was consulted to provide more detailed information for surgical strategies (Figure 2), while HSG was used only to document communications between the obstructed and the contralateral side (Figure 3).

In all cases the chosen treatment was resection of the obstructed vaginal septum with drainage of retained collections. At the time of surgery, a diagnostic laparoscopy was performed (Figure 4).

The endoscopic examination of the abdomen and of the pelvis included assessment of the uterine external profile, fallopian tubes and ovarian morphology,

presence of endometriotic foci and inflammatory localizations. Retroperitoneal masses representing pelvic displacement of kidney and general pelvic anatomy were also systematically investigated. At vaginal time firstly the obstructing vaginal septum was localized which in large retentions is evident, in other cases it is identified via the transvaginal ultrasound. Using a monopolar needle the blind vaginal cavity was penetrated, the retained material (dense liquid, frequently hematic, sometimes mixed with mucus or purulence) was drained and the obstructive septum totally removed. Repeated irrigations were performed, and the ipsilateral uterine cervix was identified and its patency ascertained. The free margins of hemivagina then were sutured with multiple interrupted 3-0 Monocryl stitches thus achieving complete marsupialization (Figure 5). All patients received perioperatively broad-spectrum antibiotics (cephalosporins of the latest available generation).

3. RESULTS

The surgical procedure was successfull in all cases**.** No major complications were recorded. Mean (± SD) operating time was 78 ±30 minutes, while mean (± SD) intraoperative blood loss was 125 ± 70 ml . Postoperative course was uneventfull and mean (± SD) hospital stay was 2,3 ± 1,5 days.
Intraoperative findings showed that the uterus was didelphic in 40/51 cases (78,4%), complete septate in 9/51 (17,6%) and bicornuate in 2/51 (3,9%). In one case the surgical finding revealed a cervical dysgenesis in the hemiuterus ipsilateral to the vaginal obstruction of a septate uterus. Cervicoplasty (hysteroscopic drilling of obliterated endocervix) was also performed in this case to restore the communication of the hemiuterus with the normal vagina. Vaginal obstruction was right-sided in 34/51 cases (66,7%) and left-sided in 17/51 cases (33,3%), communicant with the contralateral hemivagina in 10/51 (19,6%). In eight cases (15,7%) there was interystmic communication between the two hemiuteri. Ipsilateral renal agenesis was diagnosed in 49/51 patients (96,1%) while in the remainder two an ipsilateral dysplastic hypoplastic kidney was found. In all 51 cases the contralateral kidney was hypertrophic. In one patient ureteral remnant opened in the obstructed hemivagina was found. In one case, surgical finding revealed a cervical dysgenesis in the hemiuterus ipsilateral to the vaginal obstruction of a didelphic uterus. Cervicoplasty was also performed in this case to restore the communication of the hemiuterus with the normal vagina . In three patients fallopian tube ipsilateral to obstruction was found occluded with adhesion of the fimbriae. Ovaries were bilaterally present in all patients. Endometriosis was found in nine cases (17,6%) : in seven peritoneal, in two on the ovary (cystic).

Correlating with the preoperative examinations both the ultrasound and the MRI had successfully identified the uterine anomalies and the vaginal obstruction but had totally failed in the identification of intervaginal and interystmic

communications. For the latter, HSG has proven very useful in cases where it was performed.

MRI had correctly identified the ureteral remnant found in an imperforate hemivagina and another that had escaped the surgical finding.

Peritoneal endometriosis was always treated with excision or bipolar coagulation. Endometriotic cysts on the ovary were removed by stripping. In four cases the presence of inflammatory fluid and fibrin were found in the Douglas cavity and on the anterior border of the liver. In these cases, a sample was always taken for bacteriological and cultural examination for aerobic and anaerobic germs followed by repeated washes with warm physiological solution as is customary in peritonitis. Of the three cases of tubal occlusion in one a terminal salpingoplasty was performed, in the other two a salpingectomy.

The removed vaginal septa were referred for histological examination. The side of the normal vagina was always characterized by Malpighian epithelium, while the side of the imperforate hemivagina was a columnar epithelium with glandular crypts of Mullerian type. In cases where there was a communication between the two hemivaginae the columnar epithelium presented zonal processes of squamous metaplasia.

FOLLOW-UP

Our median follow-up was 6,5 years, with a range of 6 months to 13 years. The first checkup after surgery was done after the first menstruation. In all cases signs and related symptoms of obstruction (hematocolpos, pyocolpos) were completely resolved. Dysmenorrhea improved in 92% of cases and it remained so in the following months and years. After six months, abdominal pain reappeared in the young patient in whom salpingoplasty was performed. The ultrasound showed that a sactosalpinx had reformed. A second laparoscopy with salpingectomy was performed and this time and the pain disappeared. Eighteen months after the operation, one of the two patients in whom the cervicoplasty was performed began to have worsening dysmenorrhea again. An MRI revealed the formation of cervicometra. A second laparoscopy was performed with removal of the compromised hemiuterus. An operated patient, with the onset of sexual activity, developed severe dyspareunia which then disappeared spontaneously after two years. In the months following the operation and for years (in the patients followed for a longer time) all the operated patients complained of more or less abundant, clear vaginal discharge, without burning or itching. These losses tended to slowly decrease over time. In cases in which it was possible to carry out a serial colposcopic control over time (32 patients), it was not possible to demonstrate a complete transformation of the Müllerian epithelium into squamous epithelium, as reported by Rock (5) and the Schiller test

always remained negative. This data could explain the persistent presence of vaginal discharge.

Analysis of reproductive outcome in patients wanting children showed that before our surgical repair none had been pregnant. After surgery performed in our facility, of the 14 patients who sought pregnancy, 12 (85,71%) were successful. Of these 11 patients had a total of 22 pregnancies resulting in spontaneous abortion in 27% (6/22), premature birth (<37 weeks) in 36% (8/22) and in full-term birth in 36% (8/22). Most of the pregnancies were implanted in the hemiuterus contralateral to the imperforate hemivagina (75%). 14 patients out of 16 had a caesarean section: 9 for breech presentation, 1 for placenta previa, and 4 for maternal request. The pregnancies overall had a good course but there were four complications: two growth delays, one placenta previa, and one case of gestational diabetes. The 12th patient is currently pregnant at the 14th week. It is a spontaneous twin pregnancy that arose after one year of research in a patient with a complete septate uterus who had previously undergone treatment for vaginal obstruction and after 4 years the lysis of the uterine septum.

4. DISCUSSION

This study has certain limitations : it is retrospective and the sample size is relatively small. However we describe one of the few larger series in the literature that highlights the great heterogeneity of this disorder, the importance of the use of laparoscopy and the need for a therapy tailored to the specific characteristics of each case.

Our experience confirms that in HWWS conservative therapy (removal of the obstructing vaginal septum and laparoscopic control) is practicable in all cases and is safe. Only in patients with cervical atresia hemihysterectomy should be considered due to high risk of reobstruction. It allows a clear definition of the concomitant anomalies, is resolutive from the symptomatological point of view and allows a good preservation of the reproductive capacity.

A fundamental role is played by the timeliness of the diagnosis. In our cases, all reported by basic gynecologists or small hospitals, the diagnosis was straightforward: our institutes are reference centers for adolescent gynecology. However, for some practitioners the diagnosis is made difficult, not only by the rarity of the syndrome, but also by the fact that vaginal exploration in young adolescents can be impractical or not very indicative. The diagnosis of HWWS is made by investigating the etiology of dysmenorrhea and abdominal pain that occurs shortly after menarche. In fact, while a hemivagina is obstructed, the contralateral is patent and therefore the patients have normal menstrual bleeding. Other causes of

abdominal pain must be excluded in female pediatric patients such as ovarian diseases, acute appendicitis (6), and primary dysmenorrhea acute pelvic inflammation (PID). In primary dysmenorrhea the pain is accompanied by more severe cramps and is always localized on the median line. In HWWS the pain, despite the diversity of the anatomical pictures, is unilateral, late and persists for days following the flow. The real differential element, however, is the physical examination which, in "essential" dysmenorrhea, is by definition negative. In the communicating varieties or with incomplete vaginal obstruction, the presence of mucopurulent vaginal discharge and sometimes fever, can suggest an acute pelvic inflammation (PID). Notably, HWWS lacks first of all the anamnestic elements justifying an inflammatory process and there is never an impairment of the general physical state and of the blood chemistry. Furthermore, in PIDs, any adnexal swellings are almost always bilateral. Ovarian cysts and neoplasms represent the most frequent diagnosis when facing a pelvic mass in a regularly menstruating adolescent and often motivate urgent laparoscopic or laparotomy surgery. In these cases, instrumental examinations and the finding of renal agenesis can be highly indicative. The incidence of unilateral renal agenesis is 1 in 1100 while between 25% and 50% of affected women have associated genital abnormalities (7). Unicornuate uterus with a noncommunicating but functional horn may also have an essentially identical clinical presentation, including (frequently) ipsilateral renal agenesis (8). In these patients, however, the diagnosis should become clear when paravaginal mass is not found.

To be effective over time, excision of the obstructing vaginal septum must be extensive enough with precautions taken against injury to the urethra, bladder, and rectum (3). In fact, the simple incision of the septum or its only partial removal almost always expose the development of hematocolpos or pyocolpos after a spontaneous closure 9). Wang J et al. (10) reported restenosis of vaginal septum in five of 61 patients; all five patients were completely cured after a second or third surgery. However, even too large an excision can bring complications. In fact, analyzing the profound dyspareunia that arose in our patient after surgery, we saw, in the description of the surgery and in the histology of the surgical piece, that the removal of the septum had left exposed large bloody vaginal areas that were not adequately marsupialized. The painful symptomatology in the relationships was therefore to be attributed to the exposure of nerve endings that vaginal reconstruction had re-protected over time

In the presence of pyocolpos, many authors (11, 12) , due to the danger of sepsis, first perform a limited resection of the septum, postponing the definitive surgery to a few months later. In our experience, however, we have not encountered complications, when immediately performing the definitive intervention thereby allowing ample drainage of the purulent material.

Another procedure not to be recommended is to place the patient in an emergency under hormonal treatment (continuous oral contraceptives or GnRH-agonists) to avoid cyclic pain before surgery. This pre-treatment could compromise the execution of the surgery and the outcome of the septal resection, making it less evident or hiding the retention (13). Instead, it would be useful to plan the

intervention in the acute phase, when the distension of the obstructing septum is more evident. Tong et al. (14) believe that when postponement of surgery is required for young adolescent girls, GnRH-agonists may be a good option for maintaining amenorrhea, especially in cases complicated by cervical atresia.

Precision is important when defining the characteristics of the malformed uterus (didelphys vs septate or bicornuate) because if faced with a didelphic or bicornuate uterus, the case is simply resolved with the excision of the obstructing vaginal septum. On the other hand, if faced with a septate uterus (17.6% in this study) the case is not completely resolved and, to improve reproductive performance a hysteroscopic lysis of the uterine septum must be planned before the patient seeks pregnancy. In the presence of a complete septate uterus with distinct and separate cervices we perform only the resection of the septum between the two uterine cavities, leaving the cervical septum intact. In case of very small or very close cervices, we also perform the resection of the cervical septum. In both cases, we find no contraindications to a subsequent vaginal birth . We always avoid performing hysteroscopic surgery in conjunction with the excision of the vaginal septum to avoid contamination of the upper genital tract and because the hysteroscopic surgery must always be performed after hormonal preparation of the endometrial mucosa (4). MRI provides overall images useful for the intervention and is also the only method that highlights the ureteral remnants (two cases in the present study). These ureteral remnants are ipsilateral to the imperforate hemivagina, may be in communication with it, but have no symptomatological significance and must not be excised. Only the HSG can preoperatively reveal the presence and location of any communications between the two hemiuteri. To avoid the invasiveness of HSG, one can intraoperatively cannulate one of the two cervices and inject methylene blue which, in case of interystmic communication, will emerge from from the contralateral cervix. Given the frequent microscopic size of these communications, intraoperative hysteroscopy did not prove useful for this purpose. The diagnosis of these communications is essential to understand the symptoms but does not change the therapeutic plan.

For this malformative association, while single case reports or very small series abound, homogeneous and numerically rich case series are rare in the literature. The most significant studies are reported in Table 2.

Several case reports recently appeared in the literature on resection of the obstructing vaginal septum using an operative hysteroscope (21, 22, 23, 24, 25.), especially in virgin girls to maintain hymen integrity. These interventions are purpirtedly easy to perform, effective, and safe. However even if they resolve the obstructing symptoms presently , due to their partiality, they expose the young patient to the risk of re-occlusion and the certainty of abundant vaginal discharge which in the total removal of the septum are much less.

Even fewer studies have investigated long-term follow-up to provide data on fertility and outcomes of pregnancies in females with this rare malformation. The

results are summarized in Table 3. The high frequency of cesarean section reflects the high incidence of fetal breech presentation associated with uterine malformation . Concomitant unilateral renal agenesis may predispose to preeclampsia indicating meticulous prenatal care (18).

5. CONCLUSIONS

It is imperative that pediatricians and gynecologists develop strong awareness of this malformative association and have a lot of clinical suspicion when encountering adolescent girls with menstrual discomfort and unilateral kidney agenesis. Early diagnosis is essential to establishing prompt and appropriate surgical treatment that in turn will help prevent potentially severe complications.

Disclosure of interests

The Authors report no conflict of interests

Contribution to authorship

M.C., P.V. and L.F. are responsible for idea and data collection. CFC and S.S organized the follow-up of the patients. F.F. contributed to manuscript preparation. All named authors approved final version.

Details of ethics approval

This study was approved by The Ethics Committe of I.R.C.C.S. S.Raffaele Hospital Milano. Protocol Code GARA, register number CE 73/INT/2021.

REFERENCES

1. Purslow CE. A case of unilateral haematocolpos, haematometra and haematosalpinx. J Obstet Gynaecol Br Emp 1922; 29:643.

2. Burgis J. Obstructive Müllerian anomalies: case report, diagnosis,

and management. Am J Obstet Gynecol 2001; 185:338–344.

3. Kapczuk K, Friebe Z, Iwaniec K, Kędzia W. Obstructive Müllerian Anomalies in Menstruating Adolescent Girls: A Report of 22 Cases. J Pediatr Adolesc Gynecol. 2018 Jun;31(3):252-2573.

4. Fedele L, Bianchi S. Hysteroscopic metroplasty for septate uterus. Obstet Gynecol Clin North Am. 1995 Sep;22(3):473-89. PMID: 8524532.

5. Rock, J.A. and Jones, H.W.J. The double uterus associated with an obstructed shemivagina and ipsilateral renal agenesis. Am. J. Obstet. Gynecol., 1980138, 339–342.

6. Sharma P, Hegde R, Kulkarni A, Soin P, Kochar P, Rotem E (2020)

Imaging right lower quadrant pain: not always appendicitis. Clin

Imaging. 2020; 63:65–82.

7. Jayasinghe Y, Rane A, Stalewski H et al: The presentation and early diagnosis of the rudimentary uterine horn. Obstet Gynecol 2005;105:1456.

8. Stassart JP, Nagel TC, Prem KA, Phipps WR. Uterus didelphys, obstructed hemivagina, and ipsilateral renal agenesis: the University of Minnesota experience. Fertil Steril. 1992 Apr;57(4):756-61.

9. Morgan MA, Thurnau GR, Smith ML. Uterus didelphys with hematocolpos, ipsilateral renal agenesis and menses. A case report and literature review. J Reprod Med. 1987; 32:47–58.

10. Wang J, Zhu L, Lang J, Liu Z, Sun D, Leng J, Fan Q. Clinical characteristics and treatment of Herlyn-Werner-Wunderlich syndrome. Arch Gynecol Obstet. 2014 Nov;290(5):947-50.

11. Smith NA, Laufer MR. Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome: management and follow-up. Fertil Steril. 2007 Apr;87(4):918-22.

12. Gungor Ugurlucan F, Dural O, Yasa C, Kirpinar G, Akhan SE. Diagnosis, management, and outcome of obstructed hemivagina and ipsilateral renal agenesis (OHVIRA syndrome): Is there a correlation between MRI findings and outcome? Clin Imaging. 2020 Feb;59(2):172-178.

13. Zurawin RK, Dietrich JE, Heard MJ, Edwards CL. Didelphic uterus and obstructed hemivagina with renal agenesis: case report and review of the literature. J Pediatr Adolesc Gynecol. 2004 Apr;17(2):137-41.

14. Tong J, Zhu L, Lang J. Clinical characteristics of 70 patients with Herlyn-Werner-Wunderlich syndrome. Int J Gynaecol Obstet 2013; 121:173–175.

15. Candiani GB, Fedele L, Candiani M. Double uterus, blind hemivagina, and ipsilateral renal agenesis: 36 cases and long-term follow-up. Obstet Gynecol. 1997 Jul;90(1):26-32.

16. Haddad B, Barranger E, Paniel B (1999) Blind hemivagina: longtermfollow-up and reproductive performance in 42 cases. Hum Reprod. 1999 14(8):1962–1964

17. Gholoum S, Puligandla PS, Hui T, Su W, Quiros E, Laberge JM. Management and outcome of patients with combined vaginal septum, bifid uterus, and ipsilateral renal agenesis (Herlyn-Werner-Wunderlich syndrome). J Pediatr Surg 2006;41(5):987e92.

18. Heinonen P K. Pregnancies in women with uterine malformation, treated obstruction of hemivagina and ipsilateral renal agenesis. Arch Gynecol Obstet 2013; 287:975–978

19. Zhang H, Ning G, Fu C, Bao L, Guo Y. Herlyn-Werner-Wunderlich syndrome: diverse presentations and diagnosis on MRI. Clin Radiol. 2020 Jun;75(6):480.e17-480.e25.

20. Liu M, Zhang L, Xia Y, Huang X, Ye T, Zhang Y, Qi Z, Wang L, Lai X, Dai Q, Jiang Y. New Consideration of Herlyn-Werner-Wunderlich Syndrome Diagnosed by Ultrasound. J Ultrasound Med. 2021 Sep;40(9):1893-1900.

21. Cicinelli E, Romano F, Didonna T, et al: Resectoscopic treatment of uterus didelphys with unilateral imperforate vagina complicated by hematocolpos and hematometra: case report. Fertil Steril 1999; 72:553.

22. Cetinkaya SE, Kahraman K, Sonmezer M, Atabekoglu C. Hysteroscopic management of vaginal septum in a virginal patient with uterus didelphys and obstructed hemivagina.Fertil Steril 2011;96:e16–e18.

23. Sanghvi Y, Shastri P, Mane SB, Dhende NP. Prepubertal presentation of Herlyn-Werner-Wunderlich syndrome: a case report. J Pediatr Surg 2011;46:1277–1280.

24. Nassif J, Al Chami A, Abu-Musa A, Nassar AH, Kurdi A, Ghulmiyyah L. Vaginoscopic resection of vaginal septum. Surg Technol Int 2012;22:173–176.

25. Xu B, Xue M, Xu D. Hysteroscopic management of an oblique vaginal septum in a virgin girl with a rare variant of Herlyn-Werner-Wunderlich syndrome. J Minim Invasive Gynecol. 2015 Jan;22(1):7.