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Virginity-sparing management of hematocolpos with imperforate hymen: case report and literature review

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Abstract

Imperforate hymen results from failure of the endoderm of the urogenital sinus to completely canalize and has an incidence of 0.01% to 0.05%. This sometimes presents as a pelvic mass that compresses the bladder causing acute urinary retention. A 13-year-old girl was referred to our department with a history of primary amenorrhea, cyclic lower abdominal pain, abdominal–pelvic mass, constipation and acute urinary retention. She had an ultrasonography misdiagnosis of a huge ovarian mass before referral to our unit. On examination, the vagina was bulging and compressing the rectum. Repeat abdominal ultrasonography confirmed the diagnosis of hematometrocolpos. She underwent X-shaped hymenotomy with a favorable outcome. Diagnosis of imperforate hymen requires high suspicion index. Virginity-sparing surgery constitutes a good treatment option for cultural and religious reasons.

Keywords

Imperforate hymen, pelvic mass, hematometrocolpos, adolescence, urinary retention

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Introduction

Imperforate hymen results from failure of the endoderm of the urogenital sinus to completely canalize[1] and has an incidence of 0.01% to 0.05%.[2,3] The presence of primary amenorrhea in an adolescent girl with cyclical abdominal pain, urinary retention, constipation and/or lower abdominal mass is suggestive of the condition.[4] Imperforate hymen could be confused with other obstructive malformations OHVIRA (obstructed hemivagina and ipsilateral renal agenesis) syndrome.[5,6] Hymenectomy after a cruciate, plus or X-shaped hymenotomy is the standard surgical treatment of imperforate hymen.[4,7] Late discovery of imperforate hymen may lead to pain, infection, hydronephrosis, endometriosis and infertility.[8,9] We are presenting a case of imperforate hymen managed at the Douala General Hospital, Cameroon.

Case report

A 13-year-old girl came to our unit with the complaint of primary amenorrhea, cyclic lower abdominal pain, pelvic mass, constipation and acute urinary retention (relieved by catheterization at another hospital 3 days earlier). She was self-medicating with anti-helminthic medication. She had an ultrasonography misdiagnosis of a huge ovarian mass but refused surgery before referral to our unit. Physical examination showed normal secondary sexual development (pubic hair and well-developed breast buds). There was a mobile, slightly tender mass arising from the pelvis and almost bordering the umbilicus, measuring 17 cm from the superior border of the symphysis pubis. On pelvic examination, the external genitalia was normal, and the hymen was closed (Figure 1). The hymen and vagina were bulging and compressing the rectum, and rectal examination revealed a mobile central pelvic mass. A repeat abdominal ultrasonography at our hospital was consistent with an ovoid mid-pelvic sonolucent mass extending from the area

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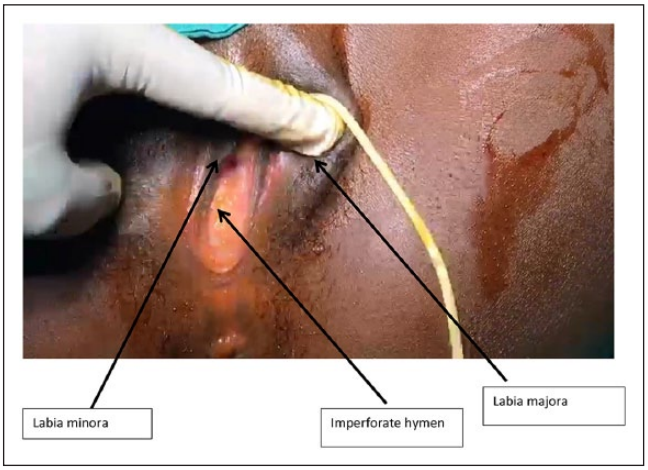

Figure 1. Image of vulva showing imperforate hymen.

of the visible upper vagina to the level just below the umbilicus (Figure 2). A separate uterus could not be seen. The bladder was not distended. The history, physical examination and ultrasonography findings were suggestive of imperforate hymen with hematometrocolpos. After signing the assent form by parents, she underwent X-shaped hymenotomy under general anesthesia. The hymen was thicker than normal and hymeneal leaflets were slightly trimmed with preservation of some tissue all round for virginity preservation. The edges were oversewn with absorbable interrupted sutures to prevent bleeding, enable better scarring and prevent hymen closure. About 700-mL old blood was removed (Figure 3) and the uterus was considerably reduced in size after the procedure. Both ovaries were of normal size on palpation. Normal micturition followed 2 h after removal of the Foley catheter. The patient was discharged from hospital 72 h later and her outcome was uneventful. She became asymptomatic and had regular menstrual cycles at 2 months of follow-up.

Discussion

Imperforate hymen is the most common obstructive congenital abnormality of the female genital tract with an incidence between 0.01% and 0.05% in newborns.[2,3] The effect of maternal estrogen secretion either in the prenatal or postnatal period may cause mucus secretion by the cervical glands.[10] Imperforate hymen, transverse vaginal septum and vaginal atresia with or without persistence of a urogenital sinus of cloaca are the common causes of secretory hydrometrocolpos.[11] This condition could rarely be diagnosed in the antenatal period during third trimester ultrasonography.[12] Perforation of the hymen commonly occurs during fetal life or in the perinatal period,[13] but there are reports of spontaneous rupture of the hymen in the adolescent period.[14]

The clinical presentation of imperforate hymen is varied ranging from an incidental finding, midline lower abdominal mass with or without protruding hymen, urinary retention, urinary tract infection, acute renal failure, constipation, acute abdomen with paralytic ileus, primary amenorrhea/cyclical abdominal pain and respiratory distress.[15] Most cases are diagnosed after menarche because of accumulation of blood in the vagina (hematocolpos) and the uterus (hematometra). Urological complications have been reported in more than 50% of cases presenting with complex congenital vaginal malformation either in neonates or during puberty. Hydrometrocolpos and hematometrocolpos cause acute urinary retention from urethral compression/urethral angulation.[15] The main presentation in the index case was cyclical lower abdominal pain, constipation and acute urinary retention.

The differential diagnosis of imperforate hymen is in the context of other obstructive malformations.

During embryonic development, two Mullerian ducts fuse and form a single uterovaginal canal around 8–10 weeks, while the vagina is developed from the uterovaginal canal sinovaginal bulb.[16] Complete patent vagina is formed around the fifth month of embryonic development. The vagina has six different developmental phases, while hymen development has dual origin from sinovaginal bulb and urogenital sinus or from isolated origin of urogenital sinus.[13] It separates the vagina from urogenital cavity and vestibule. Birth defects can occur during development, differentiation, migration, fusion and canalization.[1] Vaginal atresia with hydrometrocolpos may be a part of complex syndromic anomalies which include McKusick–Kaufman syndrome (post-axial polydactyly, hydrometrocolpos from vaginal obstruction and cardiac malformations).[17] Imperforate hymen is a local fusion anomaly with defective resorption of Mullerian septum, and it is a sporadic event. However, the development of mesonephric and paramesonephric ducts and urogenital sinus structures is interdependent, and their paracrine actions also have a role in hymen development. Therefore, hymen anomalies may present with concomitant anomalies of the above structures.[13] Mullerian malformations result from defective fusion of the Mullerian ducts during development of the female reproductive system. The least common form of these malformations is the Herlyn–Werner–Wunderlich (HWW) syndrome.[18] The association of uterus didelphis with OHVIRA (obstructed hemivagina and ipsilateral renal agenesis) syndrome is a rare congenital anomaly constituting about 0.16% to 10% of Mullerian duct anomalies.[18] It is most commonly diagnosed in adolescence due to pelvic and abdominal pain, worsening dysmenorrhea, pelvic mass and an ipsilateral renal agenesis. It can rarely be found in neonates or adults with primary infertility, pyometra, urinary retention and ischiorectal swelling. This condition is rare; therefore, diagnosis may be delayed thereby resulting in complications like endometriosis, pyocolpos and infertility.[19]

Imperforate hymen and OHVIRA syndrome have similar clinical presentation: diagnosis in the adolescent period, although it could also be done in utero or early infancy in cases of hydrometrocolpos. Both conditions may cause urinary obstruction and constipation like in the index case.

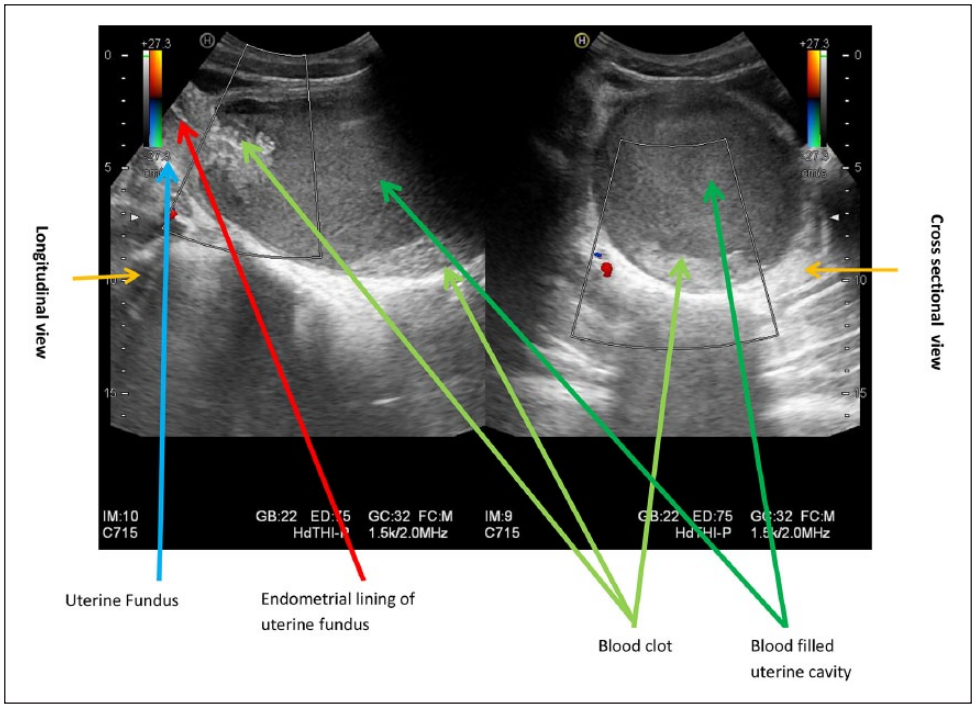

Figure 2. Ultrasonographic image of imperforate hymen.

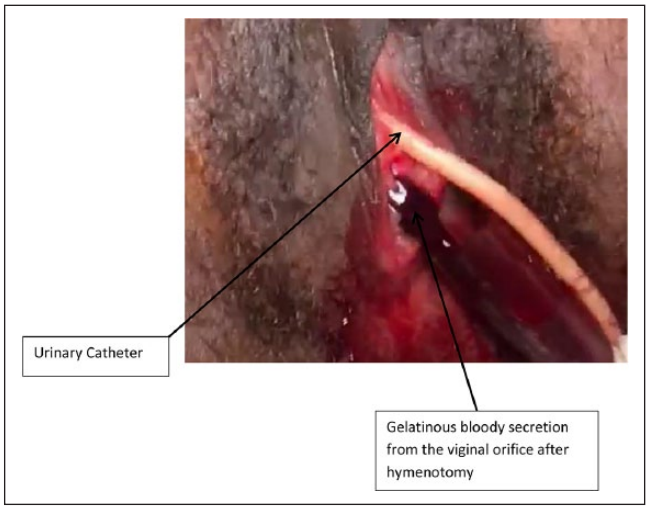

Figure 3. Vulva after hymenotomy showing dark bloody secretions.

The main differences between both conditions are that imperforate hymen patients do not exteriorize the menstrual flow and have normal uteri (indentation depth < 10 mm) according to CUME (Congenital Uterine Malformation by Experts) criteria,[20] while OHVIRA syndrome patients exteriorize their menses, though, some is retained in the hemiuterus with the septum. Furthermore, OHVIRA syndrome patients have abnormal uterus (indentation depth > 10 mm) according to CUME criteria.[20]

The diagnosis of both conditions is based on good clinical examination and magnetic resonance imaging (MRI). MRI has a high sensitivity compared to ultrasonography or computed tomography (CT) scan in the diagnosis of Mullerian malformations.[21] The index case did not seek medical attention early, but there was misdiagnosis in the first health facility that was corrected later. Some hospitals in Cameroon like in other low-income countries lack qualified or certified radiologists and therefore depend on sonographers without appropriate training.[22,23]

The treatment of imperforate hymen and most Mullerian anomalies is surgical. Virginity-sparing techniques and endoscopic techniques have been described (Table 1). In recent times, a new hymen-sparing management of a blind hemivagina in OHVIRA syndrome with the use of transrectal ultrasound (TRUS)-guided vaginoscopic septoplasty supported by pre- and postoperative diagnostics with the use of three-dimensional saline-solution infusion contrast sonovaginocervicography (3D-SVC) with virtual speculoscopy has been described. This new technique provides good visualization with favorable outcomes. There is need to evaluate the validity of this technique with a larger number of cases managed with the technique.[45]

In most cultures worldwide, there is still that strong desire for virginity before marriage.[47] We preserved a rim of hymenal tissue during incision and suturing in order to provide suitable ground for defloration during coitus. Apart from acting as a barrier to infections during the prepubertal period, no other exact functions of the hymen are known.

Table 1: Literature review and management of women with haematometrocolpos (imperforate hymen and OHVIRA syndrome).

Study (reference)	Description of study	Type of Malformation	Intervention
Yogendra Sanghvi et al.[24]	Case report: 2 cases	Herlyn-Werner-Wunderlich syndrome (HWW)	Endoscopic resection of obstructing septum
Hamidi and Haidary[25]	Case report: 1 case	Late presentation MR imaging, Surgical treatment of Herlyn-Werner-Wunderlich syndrome (HWW)	
Pereira et al.[26]	Case report: 1 case	Herlyn-Werner-Wunderlich syndrome (HWW)	Hydrodissection between hemivagina septum and surgical resection of septum
Kapczuk et al.[27]	Case report: 22 cases	Obstructive Müllerian Anomalies in Menstruating Adolescent Girls	Different methods
Gungor et al.[5]	Case report: 1 case	(OHVIRA syndrome) HWW presenting as acute abdomen	Unilateral hysterectomy and vaginal septum resection
Koticha[28]	Commentary	HWW (OHVIRA) and Zinner syndrome ZS (OSVIRA)	
Ludwin et al.[20]	Case report	HWW (OHVIRA)	Endoscopy: TRUS guided vaginoscopic septoplasty
Agarwal et al.[29]	Case report: 1 case	OHVIRA syndrome in post-cesarean period	
Kamio et al.[30]	Case report: 1 case	OHVIRA with septic shock	
Cetin et al.[31]	Case report: 1 case	Imperforate hymen	Annular hymenotomy with electrocautery
Basaran et al.[32]	Case report: 2 cases	Imperforate hymen	Vertical hymenotomy
Buchan et al.[33]	Case report: 1 case	Hematocolpos	Surgery
Saleh et al.[34]	Case report: 1 case	Hematometrocolpos with imperforate hymen disguised as abdominal pain	
Noviello et al.[35]	Case report: case series 6 cases	Herlyn-Werner-Wunderlich	One-stage surgical treatment; in one case second look required
El Saman et al.[36]	Case report: 1 case	Segmental vaginal aplasia	Dual-Force Vaginoplasty
Yavuz et al.[37]	Case report: 13 cases	HWW syndrome	Not described
Xu et al.[38]	Case report: 1 case	Herlyn-Werner-Wunderlich syndrome (HWWS)	Hysteroscopic resection of oblique septum
Zivkovic et al.[6]	Case report: 1 case	OHVIRA syndrome	
Lakhi et al.[39]	Case report: 1 case	Hematoureter due to endometriosis with hematocolpos and septum	Laparoscopy
Bakacak et al.[40]	Case report: 1 case	Hematometrocolpos due to dysfunctional uterine bleeding following progestin use	Not described
Bhoil et al.[41]	Case report: 1 case	Herlyn-Werner-Wunderlich syndrome	Surgical drainage
Aranke et al.[42]	Case report	Haematometrocolpos and acute pelvic pain associated with cyclic uterine bleeding: OHVIRA syndrome	Not described
Sleiman et al.[43]	Case report: 1 case	OHVIRA syndrome with haematosalpinx and pyocolpos	Not described
Al Ghafri et al.[44]	Case report: 1 case	OHVIRA syndrome	Not described
Ludwin et al.[45]	Case series: 4 cases	Blind hemivagina as a component of OHVIRA syndrome and varying level and features of obstruction including: 1) hemihydrocolpos; 2) hemihematocolpos; 3) "old blood" deposits in small hemivagina; and 4) narrow hymenal opening	Transrectal ultrasound (TRUS)–guided vaginoscopic septoplasty supported by pre- and postoperative diagnostics with the use of a novel ultrasound technique: 3-dimensional saline-solution infusion contrast sonovaginocervicography (3D-SVC) with virtual speculoscopy.

Table 1. (Continued)

Study (reference)	Description of study	Type of Malformation	Intervention
Fidele et al.[46]	Retrospective study of 87 cases	Didelphys uterus with obstructed hemivagina Septate uterus with obstructed hemivagina Bicornuate bicollis uterus with obstructed hemivagina Didelphys uterus with monolateral cervical atresia Bicornuate uterus with septate cervix and obstructed hemivagina	Vaginal excision of obstructive septum Vaginal excision of obstructive septum and Hysteroscopic metroplasty Vaginal excision of obstructive septum Laparoscopically assisted cervico-vaginal opening (cervicoplasty) Vaginal excision of obstructive septum
Temizkan et al.[47]	Case report: 2 cases	Virginity sparing surgery for imperforate hymen	Small central circular hymenotomy, forming an intact annular hymen
Yakıştıran et al.[48]	Case report: 1 case	True management of OHVIRA syndrome	Vaginoscopy and hysteroscopy (unipolar needle electrode) without hymenotomy

HWWS: Herlyn-Werner-Wunderlich syndrome; OHVIRA: obstructed hemivagina ipsilateral renal anomaly.

Conclusion

Early diagnosis of imperforate hymen in adolescent girls is associated with good prognosis; therefore, a thorough examination of the neonate at birth is of paramount importance. Virginity-sparing surgery constitutes a good treatment option because of cultural and religious reasons.

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Ethical approval

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Informed consent

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References

1. Langman J and Wilson DB. Embryology and congenital malformations of the female genital tract. In: RJ Kurman (ed.) *Pathology of the female genital tract*. New York: Springer, 1982, pp. 1–12. http://link.springer.com/chapter/10.1007/978-1-4757-1767-9_1 (accessed 7 August 2016).
2. Lui CT, Chan TWT, Fung HT, et al. A retrospective study on imperforate hymen and haematometrocolpos in a regional hospital. *Hong Kong J Emerg Med* 2010; 17: 435–440.
3. Mwenda AS. Imperforate Hymen-a rare cause of acute abdominal pain and tenesmus: case report and review of the literature. *Pan Afr Med J* 2013; 15: 28.
4. Liang CC, Chang SD and Soong YK. Long-term follow-up of women who underwent surgical correction for imperforate hymen. *Arch Gynecol Obstet* 2003; 269(1): 5–8.
5. Gungor Ugurlucan F, Bastu E, Gulsen G, et al. OHVIRA syndrome presenting with acute abdomen: a case report and review of the literature. *Clin Imaging* 2014; 38(3): 357–359.
6. Zivkovic K, Prka M, Zivkovic N, et al. Unusual case of OHVIRA syndrome with a single uterus, unrecognized before labor and followed by an intrapartal rupture of obstructed hemivagina. *Arch Gynecol Obstet* 2014; 290(5): 855–858.
7. Coppola L. Unique case of imperforate hymen. *J Pediatr Adolesc Gynecol* 2016; 29(1): e1–e3.
8. Lardenoije C, Aardenburg R and Mertens H. Reminder of important clinical lesson: Imperforate hymen: a cause of abdominal pain in female adolescents. *BMJ Case Rep* 2009; 2009: bcr08.2008.0722.

9. Posner JC and Spandorfer PR. Early detection of imperforate hymen prevents morbidity from delays in diagnosis. *Pediatrics* 2005; 115(4): 1008–1012.
10. Yildirim G, Gungorduk K, Aslan H, et al. Prenatal diagnosis of imperforate hymen with hydrometrocolpos. *Arch Gynecol Obstet* 2008; 278(5): 483.
11. Cerrah Celayir A, Kurt G, Sahin C, et al. Spectrum of etiologies causing hydrometrocolpos. *J Neonatal Surg* 2013; 2(1): 5.
12. Garcia Rodriguez R, Perez Gonzalez J, Garcia Delgado R, et al. Fetal hydrometrocolpos and congenital imperforate hymen: prenatal and postnatal imaging features. *J Clin Ultrasound* 2018; 46(8): 549–552.
13. Hegazy AA and Al-Rukban MO. Hymen: facts and conceptions. *Health* 2012; 3: 109–115.
14. Ben Hamouda H, Ghanmi S, Soua H, et al. [Spontaneous rupture of the imperforate hymen in two newborns]. *Arch Pediatr* 2016; 23(3): 275–278.
15. Ramareddy RS, Kumar A and Alladi A. Imperforate hymen: varied presentation, new associations, and management. *J Indian Assoc Pediatr Surg* 2017; 22(4): 207–210.
16. Sajjad Y. Development of the genital ducts and external genitalia in the early human embryo. *J Obstet Gynaecol Res* 2010; 36(5): 929–937.
17. Halim A, Afzal T, Fatima S, et al. A newborn with rare McKusick syndrome. *J Coll Physicians Surg Pak* 2018; 28(6): S140–S142.
18. Cox D and Ching BH. Herlyn-Werner-Wunderlich syndrome: a rare presentation with pyocolpos. *J Radiol Case Rep* 2012; 6(3): 9–15.
19. Siu Uribe A, Vargas Cruz V, Murcia Pascual FJ, et al. [Clinical characteristics and complications in patients with OHVIRA (obstructed hemivagina and ipsilateral renal anomaly) syndrome]. *Cir Pediatr* 2019; 32(1): 11–16.
20. Ludwin A, Martins WP, Nastri CO, et al. Congenital Uterine Malformation by Experts (CUME): better criteria for distinguishing between normal/arcuate and septate uterus. *Ultrasound Obstet Gynecol* 2018; 51(1): 101–109.
21. Olpin JD, Moeni A, Willmore RJ, et al. MR imaging of Müllerian fusion anomalies. *Magn Reson Imaging Clin N Am* 2017; 25: 563–575.
22. Sheth SS and Sonkawde R. Uterine septum misdiagnosed on hysterosalpingogram. *Int J Gynaecol Obstet* 2000; 69(3): 261–263.
23. Yoshida A, Murabayashi N, Shiozaki T, et al. Case of mature cystic teratoma of the greater omentum misdiagnosed as ovarian cyst. *J Obstet Gynaecol Res* 2005; 31(5): 399–403.
24. Sanghvi Y, Shastri P, Mane SB, et al. Prepubertal presentation of Herlyn-Werner-Wunderlich syndrome: a case report. *J Pediatr Surg* 2011; 46(6): 1277–1280.
25. Hamidi H and Haidary N. Late presentation, MR imaging features and surgical treatment of Herlyn-Werner-Wunderlich syndrome (classification 2.2); a case report. *BMC Womens Health* 2018; 18(1): 161.
26. Pereira N, Anderson SH, Verrecchio ES, et al. Hemivaginal septum resection in a patient with a rare variant of Herlyn-Werner-Wunderlich syndrome. *J Minim Invasive Gynecol* 2014; 21(6): 1113–1117.
27. Kapczuk K, Friebe Z, Iwaniec K, et al. Obstructive Müllerian anomalies in menstruating adolescent girls: a report of 22 Cases. *J Pediatr Adolesc Gynecol* 2018; 31: 252–257.
28. Koticha RB. OHVIRA and OSVIRA syndrome. *Indian J Radiol Imaging* 2018; 28(3): 375–376.
29. Agarwal A, Agarwal S and Sharma A. OHVIRA syndrome in post-cesarean period: an exclusive clinical scenario managed by two-staged operative procedure. *Taiwan J Obstet Gynecol* 2018; 57(4): 613–615.
30. Kamio M, Nagata C, Sameshima H, et al. Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome with septic shock: a case report. *J Obstet Gynaecol Res* 2018; 44(7): 1326–1329.
31. Cetin C, Soysal C, Khatib G, et al. Annular hymenotomy for imperforate hymen. *J Obstet Gynaecol Res* 2016; 42(8): 1013–1015.
32. Basaran M, Usal D and Aydemir C. Hymen sparing surgery for imperforate hymen: case reports and review of literature. *J Pediatr Adolesc Gynecol* 2009; 22: e61–e64.
33. Buchan A, Merideth MA, Childs RW, et al. Novel management of vaginal chronic graft-versus-host disease causing haematometra and haematocolpos. *BMJ Case Rep* 2018; 2018. DOI: 10.1136/bcr-2017-222720.
34. Saleh R, Katzenbach G 3rd and Espinosa J. Hematometrocolpos disguised as abdominal pain. *J Emerg Med* 2017; 53(5): e97–e99.
35. Noviello C, Romano M, Nino F, et al. Clinical and radiological findings for early diagnosis of Herlyn-Werner-Wunderlich syndrome in pediatric age: experience of a single center. *Gynecol Endocrinol* 2018; 34(1): 56–58.
36. El Saman AM, Farag MA, Shazly SA, et al. Dual-force vaginoplasty for treatment of segmental vaginal aplasia. *Obstet Gynecol* 2017; 129(5): 854–859.
37. Yavuz A, Bora A, Kurdoglu M, et al. Herlyn-Werner-Wunderlich syndrome: merits of sonographic and magnetic resonance imaging for accurate diagnosis and patient management in 13 cases. *J Pediatr Adolesc Gynecol* 2015; 28(1): 47–52.
38. Xu B, Xue M and 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; 22(1): 7.
39. Lakhi N, Dun EC and Nezhat CH. Hematoureter due to endometriosis. *Fertil Steril* 2014; 101(6): e37.
40. Bakacak M, Avci F, Bostanci MS, et al. Management of hematometrocolpos due to dysfunctional uterine bleeding following progestin use: a case report. *North Clin Istanb* 2014; 1(1): 45–48.
41. Bhoil R, Ahluwalia A and Chauhan N. Herlyn Werner Wunderlich syndrome with hematocolpos: an unusual case report of full diagnostic approach and treatment. *Int J Fertil Steril* 2016; 10(1): 136–140.
42. Aranke M, Nguyen KL, Wagner RD, et al. Haematometrocolpos and acute pelvic pain associated with cyclic uterine bleeding: OHVIRA syndrome. *BMJ Case Rep* 2018; 2018. DOI: 10.1136/bcr-2017-223348.
43. Sleiman Z, Zreik T, Bitar R, et al. Uncommon presentations of an uncommon entity: OHVIRA syndrome with hematosalpinx and pyocolpos. *Facts Views Vis Obgyn* 2017; 9(3): 167–170.
44. Al Ghafri A, Fida A and Al-Gharras A. Obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) syndrome. *Oman Med J* 2018; 33: 69–71.
45. Ludwin A, Ludwin I, Bhagavath B, et al. Virginity-sparing management of blind hemivagina in obstructed hemivagina and ipsilateral renal anomaly syndrome. *Fertil Steril* 2018; 110(5): 976–978.

46. Fedele L, Motta F, Frontino G, et al. Double uterus with obstructed hemivagina and ipsilateral renal agenesis: pelvic anatomic variants in 87 cases. *Hum Reprod* 2013; 28(6): 1580–1583.
47. Temizkan O, Kucur SK, Agar S, et al. Virginity sparing surgery for imperforate hymen: report of two cases and review of literature. *J Turk Ger Gynecol Assoc* 2012; 13(4): 278.
48. Yakıştıran B, Şükür YE, Turgay B, et al. True management of Obstructed Hemi-vagina and Ipsilateral Renal Anomaly syndrome. *Turk J Obstet Gynecol*. 2016; 13(4): 208–211.