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Case Report

CASE REPORT

Successful management of giant hydrocolpos in a limited-resource setting

Giulia Reggiani[1,*], Damiano Pizzol[2], Daniele Trevisanuto[3] and Mario Antunes[4]

[1]Dipartimento di Salute della Donna e del Bambino, Department of Woman's and Child's Health, Universita degli Studi di Padova, Padova, Italy, [2]Doctors with Africa, Doctors with Africa Mozambique, Mozambique, [3]Dipartimento di Salute della Donna e del Bambino, Dipartimento di Salute della Donna e del Bambino, Universita degli Studi di Padova, Padova, Italy, and [4]Department of Surgery, Central Hospital of Beira, Beira, Mozambique

*Correspondence address. Dipartimento di Salute della Donna e del Bambino, Department of Woman's and Child's Health, Universita degli Studi di Padova, Padova, Italy. Tel: +39 338 7851438; E-mail: giulia.reggiani.ped@gmail.com

Abstract

Abdominal distention and urinary retention are rare manifestations in newborns. The differential diagnosis of a female neonate presenting these signs, especially when combined, should include hydrocolpos due to imperforate hymen. The prognosis of imperforate hymen is generally good, although it can be associated with serious nephro-urologic and infectious complications. Early diagnosis and drainage of hydrocolpos allow prevention and/or improvement of these possible complications. In limited-resource settings, diagnostic imaging is more difficult to obtain, and, therefore, increased caution and an accurate physical exam with perineal inspection are essential. We report the case of a 8-day-old female neonate showing abdominal distention and urinary retention. She had a final diagnosis of imperforate hymen with giant hydrocolpos, complicated by obstructive uropathy and following urosepsis and bladder perforation.

INTRODUCTION

Imperforate hymen is the most common congenital malformation of the female genital tract. It is defined as a genital anomaly in which a layer of the epithelialized connective tissue that forms the hymen has no opening and completely obstructs the vaginal introits. The reported incidence of imperforate hymen is 0.0014 to 0.1% per year in full term newborns and it can manifest in the neonatal period as hydro(metro)colpos or at menarche as haematocolpos [1]. Hydrometrocolpos is defined as the accumulation of secretions within the endovaginal and endometrial canal and its reported incidence is ~0.006% per year in full term newborns [1]. Although the main cause is imperforate hymen, other causes as labial adhesions, transverse vaginal septum, vaginal atresia, vaginal a-genesis and malformations of cloaca can lead to this condition [2]. If only the vagina or the uterus is distended independently, it is called hydrocolpos or hydrometra, respectively. This condition occurs when uterine and cervical glands stimulated by maternal estrogen in utero or by withdrawal of hormones after birth, secrete mucus in the presence of distal vaginal obstruction [3]. The clinical manifestations include abdominal distention and symptoms due to compression of bladder, bowel or pelvic veins. The anomalies associated with hydrometrocolpos can be isolated, such as imperforate anus or persistent urogenital sinus, or part of a

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genetic syndrome like Bardet–Biedl, McKusick–Kaufman and Pallister–Hall [4].

We report the case of a 8-day-old girl who presented abdominal swelling and urinary retention due to isolated imperforate hymen with secondary hydrocolpos.

CASE REPORT

A 8-day-old female term neonate was transferred from a peripheral health center to the Beira Central Hospital due to important abdominal distention and vomiting episodes. The neonate was born at home and before resorting to conventional treatments, traditional unspecified treatments had been carried out. Her mother referred that she had never urinated since birth, but was regularly defecating.

Upon arrival at the pediatric emergency department, the newborn was in critical conditions, febrile, with significant abdominal distention and thoraco-abdominal respiration (Fig. 1A). Perineal inspection revealed a tense bulging membrane at vaginal introits, initially misinterpreted as vaginal prolapse (Fig. 1B). The urethral orifice was not visible, while the anus was normally positioned and perforated. No other abnormalities were found by a physical examination. A plain radiograph of the abdomen highlighted a homogenous soft tissue opacity that was occupying the suprapubic and mesogastric region, causing displacement of the other abdominal structures and elevation of the diaphragm. In the light of fever and septic appearance a broad-spectrum antimicrobial therapy was started. On the ninth day of life, abdominal ultrasound revealed a septet cystic mass with fluid-debris level occupying the mesogastric and hypogastric region, with compression of both kidneys and mild dilatation of renal pelvis bilaterally. Dislocation of the liver and of the spleen without ascites was also present. Anuria was confirmed and, since it was impossible to insert a urethral catheter, on the 10th day of life the urologist performed a suprapubic aspiration in order to resolve urinary retention. Successively, the results of the initial laboratory tests showed elevated creatinine levels (230 umol/L) and moderate hypernatriemia.

On the 13th day of life, after clinical stabilization, the newborn was submitted to surgical intervention under general anesthesia. Skin was incised till peritoneum and, due to evidence of previous bladder perforation (Fig. 1C), urine was aspirated and cystorrhaphy was performed. The pelvic cyst (hydrocolpos) was then mobilized and drained by vaginal way through incision of the imperforate hymen. A urinary catheter was placed and the abdominal wall synthesis was performed. No intraoperative complication occurred.

Since the second post-operative day the newborn restarted regular breastfeeding with appropriate weight gain. On the fourth post-operative day she had fever associated to leucocytosis and we started the second line antibiotics, with good clinical response.

The following post-operative course was regular. The neonate presented adequate diuresis and normalization of creatinine levels. Ten days after the surgical intervention an abdominal ultrasound control showed a remaining moderate right pyelectasis without other pathological findings. The urinary catheter was removed after 2 weeks. On the 17th post-operative day the infant was discharged at 1 month of age.

DISCUSSION

We described a case of imperforate hymen presenting as neonatal giant hydrocolpos, complicated by obstructive uropathy due to mass effect and subsequent urosepsis and bladder perforation.

Hydro(metro)colpos is a rare cause of neonatal abdominal distension. In case of a newborn presenting abdominal swelling with identification of a cystic mass the differential diagnosis includes ovarian cyst, cystic renal masses, enteric duplication cyst, mesenteric cyst, meconium pseudocyst, choledochal cyst, adrenal cyst, splenic cyst, urachal cyst, anterior sacral meningocele, intra-abdominal cystic variety of sacrococcygeal teratoma and chylous ascites [2].

Urinary retention is rare in children, especially in newborns [5]; its association with abdominal distention in a female neonate should raise the suspect of hydrocolpos, whose most frequent cause is represented by imperforate hymen. Perineal inspection reveals the presence of a cystic mass at the vaginal introits and suggests the diagnosis, that can be confirmed trough abdominal ultrasound and magnetic resonance imaging, if needed to rule out complex genitourinary malformations [6]. The diagnosis of congenital hydrocolpos is also possible prenatally by means of ultrasonography and eventual fetal MRI [7]. In our case, the diagnosis was made at a late stage due to the lack of prenatal screening and the consultation of traditional healers as first choice. This delay led to the development of complications and, thus, to more difficult management and lower chance of good outcome.

The management of imperforate hymen depends on the age of presentation and on the severity of the condition. In

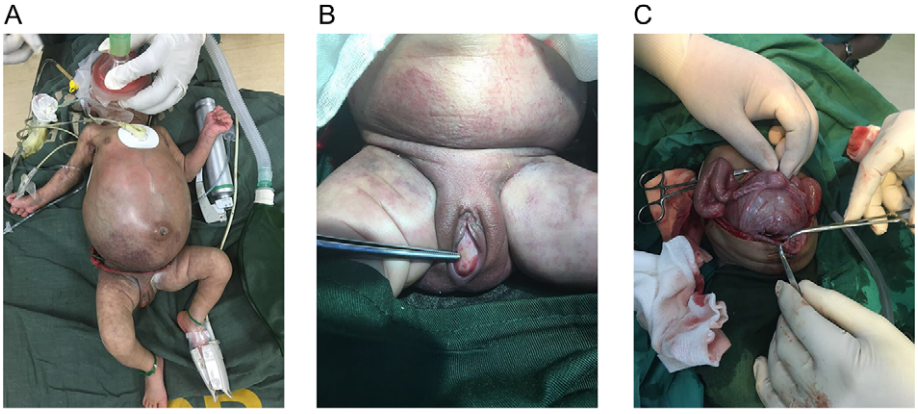

Figure 1: (**A**) Important abdominal distention in a female newborn. (**B**) Tense bulging membrane at vaginal introitus. (**C**) Intraoperatory image showing giant hydrocolpos and bladder perforation.

asymptomatic cases a conservative approach can be considered, but symptomatic patients require drainage of the secondary hydrocolpos. The drainage can solve obstructive acute kidney injury, avoiding chronic renal damage, and improve hydronephrosis, as was seen in our case and reported in the literature [8]. The surgical treatment can be made by simple hymenotomy or hymenectomy, while laparotomy is indicated for the treatment of abdominal complications [2]. In this case a laparotomy was necessary due to the bladder breakage and in order to exclude other complications. It is not clear when and how the bladder broke; this could have happened during the puncture procedure. If this hypothesis were true, we should take into account the resulting urine leak into the abdominal cavity and its consequences.

The prognosis of isolated imperforate hymen is generally good, but its presentation in the neonatal period with hydrocolpos can lead to complications associated with high morbidity and mortality [2, 9]. Despite the critical presentation, the lack of appropriate equipment and the occurrence of complications, thanks to appropriate management and accurate follow-up, the newborn was finally discharged in good conditions.

In conclusion, dealing with congenital hydrocolpos, early diagnosis, ideally in the prenatal period, and timely treatment should be the standard. However, in limited-resource settings, and considering the lack of prenatal screening, the paucity of experienced specialists and the strong belief in traditional healers, it is very difficult to achieve a prompt recognition of this condition [10]. In such a context, training of the involved health personnel towards a high index of caution could be a first step in order to prevent complications as urinary tract obstruction, renal failure, repeated urinary tract infections or hydrocolpos rupture with peritonitis. Afterwards it is obviously mandatory to strengthen the health system in terms of health workers ability and equipment and to reduce the gap between traditional and conventional medicine.

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CONFLICT OF INTEREST STATEMENT

No conflicts of interest.

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ETHICAL APPROVAL

No approval required.

CONSENT

The patient's mother gave informated consent for publication of the case report.

GUARANTOR

Giulia Reggiani.

REFERENCES

1. Vitale V, Cigliano B, Vallone G. Imperforate hymen causing congenital hydrometrocolpos. *J Ultrasound* 2013;**16**:37–9. DOI:10.1007/s40477-013-0009-x.
2. Khanna K, Sharma S, Gupta DK. Hydrometrocolpos etiology and management: past beckons the present. *Pediatr Surg Int* 2018;**34**:249–61. DOI:10.1007/s00383-017-4218-9.
3. Karteris E, Foster H, Karamouti M, Goumenou A. Congenital imperforate hymen with hydrocolpos and hydronephrosis associated with severe hydramnios and increase of maternal ovarian steroidogenic enzymes. *J Pediatr Adolesc Gynecol* 2010;**23**:136–41. DOI:10.1016/j.jpag.2009.10.002.
4. Nigam A, Kumar M, Gulati S. Fetal ascites and hydrometrocolpos due to persistent urogenital sinus and cloaca: a rare congenital anomaly and review of literature. *BMJ Case Rep* 2014; published online: 24 January 2014. DOI:10.1136/bcr-2013-202231.
5. Sharifiaghdas F, Abdi H, Pakmanesh H, Eslami N. Imperforate hymen and urinary retention in a newborn girl. *J Pediatr Adolesc Gynecol* 2009;**22**:49–51. DOI:10.1016/j.jpag.2008.07.018.
6. Nagaraj BR, Basavalingu D, Paramesh VM, Nagendra PDK. Radiological diagnosis of neonatal hydrometrocolpos—a case report. *J Clin Diagn Res* 2016;**10**:TD18–9. DOI:10.7860/JCDR/2016/18537.7510.
7. Nakajima E, Ishigouoka T, Yoshida T, Sato T, Miyamoto T, Shirai M, *et al*. Prenatal diagnosis of congenital imperforate hymen with hydrocolpos. *J Obstet Gynaecol (Lahore)* 2015;**35**:311–3. DOI:10.3109/01443615.2014.951608.
8. Ayaz UY, Dilli A, Api A. Ultrasonographic diagnosis of congenital hydrometrocolpos in prenatal and newborn period: a case report. *Med Ultrason* 2011;**13**:234–6.
9. Tilahun B, Woldegebriel F, Wolde Z, Tadele H. Hydrometrocolpos presentino as a huge abdominal swelling and obstructive uropathy in a 4 day old newborn: a dianostic challenge. *Ethiop J Health Sci* 2016;**26**:89–91. DOI:10.4314/ejhs.v26i1.15.
10. Okoro PE, Obiorah C, Enyindah CE. Experience with neonatal hydrometrocolpos in the Niger Delta area of Nigeria: upsurge or increased recognition? *Afr J Pediatr Surg* 2016;**13**:161–5. DOI:10.4103/0189-6725.194666.