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A rare cause of abdominal cyst in the neonatal period: Hydrometrocolpos

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The authors stated that the written consent was obtained from the patient presented with images in the study.

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Abstract

Hydrometrocolpos is a clinical condition that arises from vaginal outflow obstruction during fetal life and rarely manifests with an abdominal cystic mass in the neonatal period. In cases with delayed diagnosis, urinary complications, respiratory distress, and gastrointestinal obstructions may occur and thus a prompt diagnosis is of prime importance for the prevention of such complications via surgical drainage and repair. In the present study, we report on a neonatal hydrometrocolpos case manifesting as an abdominal cyst associated with intrauterine vaginal atresia and causing bilateral hydronephrosis, urosepsis, and respiratory distress.

Keywords: Hydrometrocolpos, Neonate, Cystic mass

Introduction

Hydrometrocolpos (HMC) is a clinical condition characterized by the expansion of the vagina and uterus due to the accumulation of cervicovaginal secretions produced by the uterine and cervical glands in the presence of vaginal outflow obstruction during fetal life [1]. This condition may occur in 1 in 5,000-10,000 live female births [2]. HMC may be caused by an imperforate hymen, vaginal or cervical atresia, and vaginal septum. Hydrocolpos associated with vaginal atresia is caused by the failure of canalization of the proximal vaginal plate and may manifest as an abdominal mass in the neonatal period or with other symptoms in later ages such as abdominal pain, pelvic mass, primary amenorrhea, and urinary dysfunction [3]. The mass effect of hydrocolpos on the adjacent organs may result in various complications including lower extremity edema, urinary retention, hydroureteronephrosis, urosepsis, endometriosis [4]. Prompt diagnosis and drainage of hydrocolpos are highly important for preventing these complications.

In the present study, we report on a neonate with an antenatal diagnosis of an abdominal cyst, who was admitted with bilateral hydronephrosis, urosepsis, respiratory distress, and an abdominal cyst. She was diagnosed with HMC associated with vaginal atresia, which is an extremely rare entity.

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Case presentation

A 27-day-old girl was referred to our clinic after being diagnosed with an abdominal cystic mass during the antenatal period. Physical examination revealed tachypnea and subcostal and intercostal retractions. An abdominal examination revealed marked distension and a well-circumscribed, stiff, mobile, palpable mass measuring 9x7 cm in diameter, extending from the pelvis to the umbilicus. Ultrasonography (USG) revealed a thick-walled, midline abdominopelvic mass measuring 10x7.5 cm with dense content and internal echoes. Additionally, the renal pelvicalyceal system was dilated bilaterally, and grade 2-3 hydronephrosis was detected on the left, and grade 3 hydronephrosis was detected on the right. The patient received continuous oxygen support due to respiratory distress caused by the mass effect. A nasogastric catheter was inserted due to abdominal distension. Antibiotic therapy was initiated according to culture-antibiogram results due to the detection of pyuria in urinalysis and bacterial growth in urine culture. An abdominal magnetic resonance imaging (MRI) scan confirmed the mass as a mesenteric cyst.

Intraoperative exploration showed an expanded cervix and uterus. The cervix had a diameter of 10x8 cm, was edematous, and anchored to the neighboring tissues (Figure 1). An incision made on the posterior cervix led to the discharge of a large amount of seromucinous fluid (Figure 2). After fluid aspiration, it was noticed that the catheter passed through the cervix could not be advanced to the vagina. Subsequently, due to the presence of vaginal atresia, a lumen was formed on the outer vaginal wall by making a cross-shaped incision with the aid of the vaginal bump established under the guidance of the catheter. The surgery was finalized by placing a 10 Fr catheter for drainage (Figure 3). An echocardiogram (ECHO) performed for the assessment of additional abnormalities indicated the presence of a partial pulmonary venous return anomaly (PAPVC). Following the surgery, the complaints of abdominal distension and respiratory distress regressed, and spontaneous urination occurred. The patient was discharged uneventfully.

The patient's family consented to participate in this study.

Figure 1: An expanded cervix and uterus

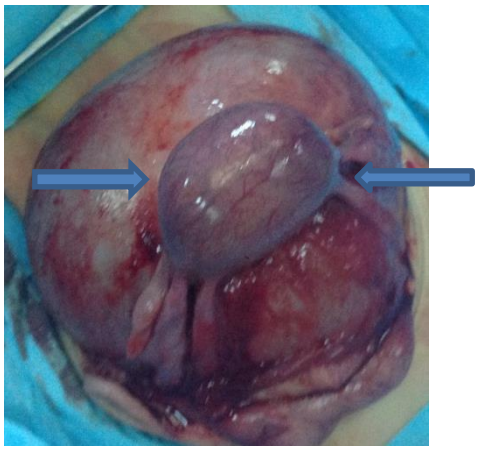


Figure 2: An incision made on the posterior cervix

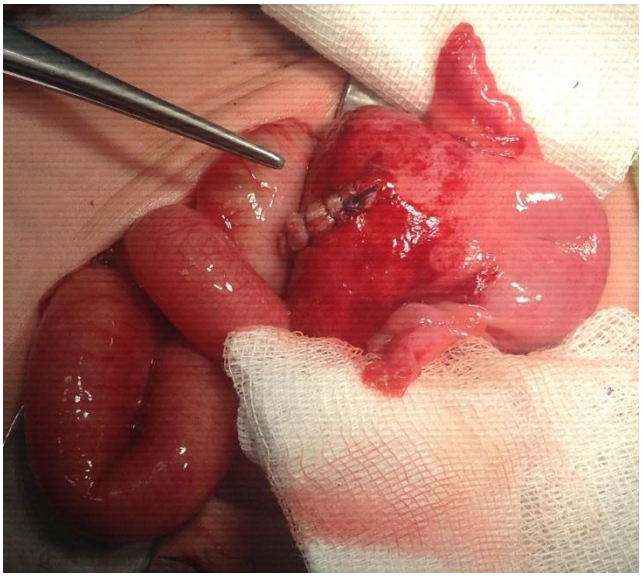
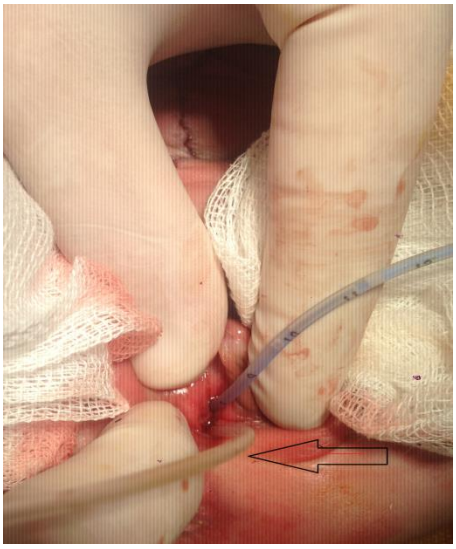


Figure 3: The catheter used for drainage



Discussion

Hydrocolpos is defined as the accumulation of secretions in the vagina while HMC refers to the accumulation of secretions both in the vagina and uterus [5]. HMC is mostly caused by imperforate hymen and rarely caused by other conditions including labial adhesions, transverse vaginal septum, vaginal atresia, vaginal agenesis, and cloacal malformations [6]. The HMC arising from vaginal atresia is an extremely rare neonatal condition commonly presenting with an abdominopelvic mass [3]. The mass often occurs as a result of increased mucus secretion by the uterine and cervical glands stimulated by maternal estradiol in the presence of vaginal outlet obstruction [5]. In the case presented, the Müllerian ducts did not merge with the lower section of the vagina arising from the urogenital sinus. The uterine cavity was normal, and functional endometrial tissue was present.

Common physical examination findings of HMC include a pelvic mass and abdominal distension [6]. In cases with delayed diagnosis, urinary retention may lead to serious nephrological and infectious complications as well as life-threatening conditions such as respiratory distress, gastrointestinal obstructions, and cardiac comorbidities. A prompt diagnosis is of prime importance for the prevention of

such complications via surgical drainage and repair and the early diagnosis of additional anomalies [3]. In the case presented, an abdominopelvic cystic mass localized in the posterior bladder wall was detected on USG during the antenatal follow-up. Due to delayed presentation, the patient had developed grade 2-3 hydronephrosis on the left and grade 3 hydronephrosis on the right kidney, urosepsis due to urinary retention, and respiratory distress secondary to mass compression.

Hydrometrocolpos (HMC) can be diagnosed during the neonatal or pubertal period depending on the underlying etiology. Moreover, most cases reported in the literature are stillbirth and are diagnosed by autopsy. Currently, HMC can be diagnosed by USG during the antenatal period as well [1]. The differential diagnosis of HMC often includes a mesenteric cyst, enteric duplication cyst, ovarian cyst, ovarian tumors, rectal duplication, and bladder duplication [7]. In the case presented, the cyst was initially diagnosed as a mesenteric cyst on MRI and then a diagnosis of HMC was made intraoperatively. These findings implicate that the diagnosis of this extremely rare congenital malformation requires a high index of suspicion and awareness.

Although HMC typically presents as an isolated entity, it can be accompanied by urogenital/anogenital malformations and cardiac anomalies or can manifest as part of a syndrome [8]. In the case presented, no additional anomaly other than PAPVC was detected and thus a syndromic diagnosis was not assigned.

Vaginal atresia and the resultant HMC should be considered in the differential diagnosis of female infants with a cystic mass in the intrauterine period. Early diagnosis and intervention are of prime importance for the prevention of complications.

References

1. Khanna K, Sharma S, Gupta DK. Hydrometrocolpos etiology and management: past beckons the present. Pediatr Surg Int. 2018;34(3):249-61.
2. Ayaz UY, Dilli A, Api A. Ultrasonographic diagnosis of congenital hydrometrocolpos in prenatal and newborn period: a case report. Medical ultrasonography. 2011;13:234-6.
3. Nazir Z, Rizvi RM, Qureshi RN, Khan ZS, Khan Z. Congenital vajinal obstructions: varied presentation and outcome. Pediatr Surg Int. 2006;22(9):749-53
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. 10.1136/bcr-2013-202231.
5. Okoro PE, Obiorah C, Enyindah CE. Experience with neonatal hydrometrocolpos in the Niger Delta area of Nigeria: Upsurge or increased recognition? Afr J Paediatr Surg. 2016;13(4):161–5.
6. Koc M, Akyol M. Imperofrate hymen: A rare cause of urinary retention: a case report. Journal of Inonu University Medical Faculty. 2008;15:125-7.
7. Warne S, Chitty LS, Wilcox DT. Prenatal diagnosis of cloacal anomalies. BJU Int. 2002;89:78–81.
8. Sharma D, Murki S, Pratap OT, Irfan G, Kolar GA. Case of hydrometrocolpos and polydactyly. Clin Med Insights Pediatr. 2015;14,9:7-11.

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