Neonatal hydrocolpos is a rare condition that involves fluid accumulation in the vagina. On diagnostic imaging, the dilated vagina, along with the compressed uterus, can simulate a mature cystic teratoma with a mural nodule. Herein, we report the case of a newborn girl with congenital hydrocolpos that was caused by an imperforate hymen; the hydrocolpos mimicking a mature cystic teratoma on abdominal ultrasonography and magnetic resonance imaging. Any newborn girl with a pelvic cystic mass should be suspected as having a congenital vaginal obstruction manifesting as hydrocolpos or hydrometrocolpos. Thorough examination of the external genitalia, as well as imaging of the uterus and vagina, enables correct diagnosis and optimal treatment.

Key Words: Hydrocolpos, Hydrometrocolpos, Imperforate hymen, Infant, Newborn

INTRODUCTION

When diagnosing cystic masses in the pelvic cavity of newborn girls, clinicians must differentiate among hydrocolpos or hydrometrocolpos, cystic teratoma, ovarian cyst, old ovarian torsion, megacystis, anterior sacral meningocele, anal atresia, gastrointestinal duplication cyst, and so on. Correct diagnosis is crucial, especially before abdominopelvic surgery, because each of these conditions has different therapeutic and prognostic implications. However, it is often a challenge. Herein, we report the case of a newborn girl with congenital hydrocolpos that mimicked mature cystic teratoma on abdominal ultrasonography (US) and magnetic resonance imaging (MRI).

CASE REPORT

A girl as the second of twins was born by cesarean section at 37+2 weeks gestation to a 37-year-old gravida 1 Korean mother in Chungnam National University Hospital, Daejeon, South Korea. The mother had no history of medical illness or medication before or during pregnancy, and had conceived by intrauterine insemination. Fetal ultrasonogram (US)
findings at 34+6 weeks gestation had revealed cystic, mixed-echoic mass near the fetal bladder. The size of the mass was 5.1×4.3 cm, and it grew slightly over time.

At birth, the girl weighed 2,470 g (50th percentile), measured 49 cm tall (50th percentile), and had a head circumference of 32 cm (10th-50th percentile). The Apgar scores were 8 and 10 at 1 and 5 minutes, respectively. She was clinically stable, and a general physical examination revealed no other abnormalities, such as abdominal distension, palpable mass, or vulvar bulge.

She had normal plain chest and abdominal radiography, but US evaluation demonstrated a well-marginated cystic mass and echogenic debris in the intraperitoneal space of the pelvis. The size of the mass was measured 6.2×3.8×8.9 cm. It was positioned retrovesically and had caused a hydroureteronephrosis by compressing the ureter of the right kidney. Furthermore, it was difficult to delineate the uterus or either ovary on the US. Attached to the inner wall of the cyst was a round, heterogeneous, echogenic nodular lesion that was 1.5 cm in diameter and had a fatty component (Figure 1). These findings were thought to indicate mature cystic teratoma. An enhanced MRI of the pelvis yielded similar findings, suggesting a mature cystic teratoma with mural nodule (Figure 2). Serum levels of lactate dehydrogenase, human chorionic gonadotropin β-subunit and α-fetoprotein were within the normal range for the girl's age.

We performed an exploratory laparotomy 7 days after birth to release pressure from the urinary collecting system. This surgery revealed a large hydrocolpos with imperforate hymen. Instead of removing the mass, we performed a tube vaginostomy, and the accumulated fluid was drained for 15 days.
A follow-up US at postoperative day 8 showed no residual fluid in the vagina, and there was marked improvement in hydroureteronephrosis. The renal structure was normal, and only mild bilateral hydroureteronephrosis remained.

At 25 days of age, she was discharged in a good general condition. At 2 months of age, the hydroureteronephrosis had completely resolved, and a follow-up US revealed a normal uterus and vagina. At 12 months of age, she had a patent hymen; repeated US confirmed that fluid had not reaccumulated in the uterus or vagina, and that the kidneys were normally sized without hydroureteronephrosis.

**DISCUSSION**

Neonatal hydrocolpos is a rare condition, with a prevalence of 1 in 16,000 newborn girls\(^4\). About 15% of abdominal masses in newborn girls are manifestations of hydrometrocolpos\(^5\); nonetheless, this too is a rare condition, with an incidence of 0.006% in newborns\(^6\).

Hydrocolpos refers to an accumulation of fluid in the vagina; if a large amount of fluid is accumulated in the uterine cavity, hydrometrocolpos can also develop. Both are caused by a combination of stimulated secretory glands and vaginal obstruction\(^5\). As well as increased secretion by cervical mucus glands are believed to be secondary to maternal estrogen stimulation or very sensitive estrogen receptors reactivity, any vaginal obstructions such as imperforate hymen, complete transverse vaginal septum, or partial vaginal agenesis may result in a pelvic mass.\(^6\) These processes can occur in isolation, or in combination with other congenital genitourinary anomalies, such as persistent urogenital sinus or cloacal dysgenesis. In addition, vaginal atresia can be associated with disorders like McKusick-Kaufman syndrome and Bardet-Biedl syndrome, and vaginal septum can occur alongside syndromes involving Mullerian aplasia\(^7\). Our patient had no congenital anomalies or syndromes, and there was no family history of urogenital disorder.

In one study involving 26 patients with congenital vaginal obstructions, six were newborn infants who had presented 1–25 days (mean 10 days) after birth. Symptoms such as abdominal mass, neonatal sepsis, urinary retention and respiratory distress were common presentations in those newborns. One of them had been referred after an exploratory laparotomy for an over-sensible ovarian cyst\(^8\).

Imperforate hymen is the most common obstructive anomaly of the female genital tract, occurring in 0.014%–0.1% of full-term births\(^9,10\). The condition can be diagnosed via physical examination of the genitalia; specifically, bulging occurs in the membrane of the vaginal introitus. However, vulvar bulge is not present in all cases of imperforate hymen, and it is commonly missed\(^11\).

Furthermore, in uncomplicated imperforate hymen, very little mucus accumulates in the vagina. Many patients with the disorder are asymptomatic until menarche, and the condition becomes clinically apparent only after puberty. Conversely, the over-distended vagina and or uterus may compress the adjacent organs, causing occasional abdominal pain, intestinal or urethral obstruction, hydroureteronephrosis, bladder perforation, and venous stasis in the lower extremities\(^12,14\). Another report concerns patients who are symptomatic during the neonatal period\(^13\): in newborns with complications, the imperforate hymen must be evaluated and treated soon after birth. In our case, the hydrocolpos was large enough to cause hydroureteronephrosis, and the accumulated fluid needed to be drained during the newborn period. We performed a transvaginal drainage via a tube vaginostomy. However, to prevent reaccumulation, trans-abdominal drainage of hydrocolpos via an indwelling tube is preferred over transvaginal drainage, especially in cases of hydrocolpos that involve more complicated genitourinary anomalies.\(^15\)

To diagnose pelvic cystic masses, clinicians must differentiate between benign and malignant ovarian neoplasms, sacral tumors, and even anterior meningocele as well as anomalies of the urinary or gastrointestinal tracts.\(^14\) Teratomas can occur in many locations at birth, such as the sacrococcygeal area, neck, retroperitoneum, pelvis, and so on.\(^17\) Mature cystic teratomas are encapsulated tumor composed of mature tissue or organ components. On US, mature cystic teratomas usually appear as a cystic mass with some mural hyperechoic components, Rokitansky nodules, or protuberances.\(^18,19\) In may cases, definitive diagnosis of a mature cystic teratoma is quite difficult. In fact, several pelvic disorders manifest apparent mural nodules and are therefore often wrongly identified as cystic teratomas.\(^20\)

In our own case, US revealed a nodular structure that was thought to be the mural component of a mature cystic teratoma. However, it transpired that the anomaly was a compressed uterus in the immediate vicinity of a cystic dilatation of the
vagina. Numerous reports have stated that undiagnosed or misdiagnosed imperforate hymen is associated with significant morbidity and mortality, and that it can prompt unnecessary abdominal exploration or laparotomy, or even hysterectomies. Moreover, abdominal distension, and bowel perforations associated with the condition can cause respiratory failure.

8,12,14,11,20

In conclusion, although hydrocolpos and hydrometrocolpos are uncommon conditions in newborns, clinicians should suspect them when treating patients with an abdominopelvic mass. In addition to careful examination of the external genitalia, imaging studies should be carried out to identify the uterus and vagina, distinguish hydrocolpos from hydrometrocolpos, and differentiate other causes of cystic mass, thus avoiding any erroneous therapy.

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