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Case Report

Isolated Congenital Hydrocolpos with Imperforate Hymen - Two Case Reports with Literature Review

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Abstract

Introduction: Hydrocolpos is congenital cystic distension of vagina due to outlet obstruction. It presents as cystic swelling at introitus. A large hydrocolpos produces mass effects on pelvic organs leading to complications. It is potentially associated with other congenital anomalies and may present with clinical syndrome. Incidence is 1 in 16,000 to 30,000 births. Antenatal detection is possible with ultrasound anomaly scan and foetal magnetic resonance imaging scan. Prompt recognition and management at birth is essential to prevent complications. We are presenting two cases of isolated hydrocolpos without any complications.

Methods: Two cases of hydrocolpos were seen at our out patient clinic on different days recently. Both cases were examined and noted with case details.

Results: We had seen two out born cases of hydrocolpos at our out patient department and presenting both cases as clinical case reports with literature review.

Conclusion: Early recognition of hydrocolpos at birth is essential for further evaluation, early management and prevention of complications.

Keywords: Hydrocolpos; Newborn; Congenital

Case 1

Term female neonatal infant brought by attendants for clinical opinion for a cystic swelling at introitus, protruding through the vagina. Baby delivered by caesarean section at term. Baby's Apgar scores at 1,5 minutes and later were normal. No adverse events were recorded during antenatal, intranatal and postnatal period. Antenatal ultrasound anomaly scan was normal. Baby passed urine and meconium within 12 hours after birth. Baby was on breast feeds. Baby weighed 2.2 kg on day 1 after birth, small for gestational age, pulse rate 118/minute, respiratory rate 42/minute, SpO2 97% in room air, CRFT 2 seconds and RBG 58 mg/dl. A cystic reddish pink swelling was noticed and protruding out of vagina. Surface was smooth, tense, thin walled with visible capillaries. Swelling was non-tender and non-pulsatile. Ultrasound scan of abdomen revealed urinary bladder filled with urine. No other pelvic masses were noticed. Both kidneys and ureters were normal. The cyst was spontaneously ruptured on day two with discharge of clear, mucinous fluid from the ruptured cyst at mother's cot side at maternity care centre. Remnants of ruptured cyst were seen at vagina.

Case 2

One day old female baby brought by attendants for consultation for a swelling at introitus and protruding through vagina. Baby delivered at term by LSCS to a second-gravida mother at a healthcare facility. Pregnancy was uneventful. Antenatal anomaly scan was normal. Parents were non-consanguineous young couple. Baby's Apgar scores were normal at 1 and 5 minutes. Baby was on mixed feeds. Baby passed urine and meconium within 12 hours after delivery. Baby's axillary temperature was 35.8°C, pulse rate 132 per minute, SpO2 98% and respiratory rate 46 per minute.

A tense cystic swelling measuring approximately 1.8 × 1.5 inches, pink in colour, smooth surfaced with visible blood vessels - capillaries, non-tender, non-pulsatile, translucent, thin layered swelling protruding through vagina. Ultrasound scan of abdomen revealed mildly distended urinary bladder filled with urine, no

other pelvic mass noticed, both kidneys and ureters were normal in size and shape. The cyst was incised and drained by surgeon at maternity care centre. The contents of drained cyst were clear and mucinous fluid.

Figure 1: Cystic swelling at introitus of vagina (Hydrocolpos) of case 1 and 2.

Discussion

Our both cases of hydrocolpos were small cystic swellings with no mass effect on surrounding structures. Both babies passed urine and stools within 24hrs after delivery. Both cases were isolated anomalies. Case one spontaneously ruptured on day two at mother's bedside. Case two was surgically incised. Both cases were isolated anomalies due to imperforate hymen. Both babies were normal on follow up at two weeks of age.

Literature Review

Hydrocolpos is an abnormal, congenital, cystic distension of vagina presenting externally at introitus due to outlet obstruction usually by imperforate hymen. It is filled with mucinous fluid secreted by the endocervical glands under maternal hormonal influence of oestrogen during intrauterine life. Early description of Hydrocolpos was given by Godefroy in 1856. Incidence is 1 in 16,000 [1] to 30,000 births [2].

Development of vagina and hymen - early genital tract develops during third week of gestation. Urorectal septum seperates rectum from urogenital sinus during 7th week of gestation. Mullerian ducts grow downwards upto urogenital sinus forming uterovaginal canal in 9th week. Mullerian ducts fuse and form uterovaginal canal by 12th week. The upper part of vagina is formed from fusion of Mullerian ducts and lower part of vagina is developed from sinovaginal bulbs of urogenital sinus. Hymen is formed from proliferation of sinovaginal bulbs and separates the vaginal lumen from the urogenital sinus [3]. The central portion of hymen perforates and communicates with introitus before birth. Failed canalisation results in imperforate hymen. After birth, the hymenal opening widens by approximately 1mm per year.

Mechanisms involved in the formation of hydrocolpos - possibly include dual factors 1. Failure of canalization of hymen or outlet obstruction due to atresia of vagina. 2. Copious endocervical secretion of mucinous fluid under the influence of maternal hormones - oestrogen [2]. Delayed presentation of hydrocolpos or hematocolpose in older female children can be explained by the action of progesterone in inhibiting the oestrogen mediated endocercervical secretion. Progesterone can be secreted by the placenta or foetal adrenal cortex. This delayed presentation can be suspected in an adolescent female child presenting with suprapubic pain with secondary sexual features but not attained menorchae [4].

Associated anomalies and complications of hydrocolpos - include cloacal dysgenesis, persistent urogenital sinus, uterine agenesis, renal agenesis, intestinal/rectal atresia or obstruction. A large hydrocolpos may compress and produce mass effect on the surrounding structures in the pelvis or abdomen of the foetus or newborn like urinary bladder with urine retention with or without rupture, ureters leading to obstructive hydronephrosis/hydroureter and subsequent renal failure if unrecognized, sepsis [5] and hematometrocolpos. Hence, prompt early recognition of hydrocolpos at birth is essential to plan the treatment and prevent complications. Undrained hydrocolpos may be infected and lead to abscess formation (Pyocolpos) and may involve uterus (Pyometrocolpos). Small isolated hydrocolpos may not produce complications if recognized and incised to drain at birth.

Hydrocolpos with syndromic presentation - several syndromes may be associated with hydrocolpos. 1. McKusick - Kaufman syndrome (vaginal atresia, urogenital sinus, postaxial polydactyly [6,7]. Ellis Van Creveld Syndrome - Hydrocolpos, polydactyly, congenital cardiac defects, short stature, teeth and oral frenulum abnormalities), 3. Herlyn-Werner-Wunderlich syndrome -malformed uterus, sinked vagina and renal agenesis, 4. MURCS syndrome - absent vagina and uterine, renal and vertibral anomalies, 5. Mayer - Rokitansky - Kuster - Hauser syndrome and 6. Bardet-Biedl syndrome.

Conclusion

Early recognition of hydrocolpos at birth is essential for further

evaluation and management for the following reasons - a) to find out the cause of hydrocolpos b) to estimate the magnitude of mass effect on surrounding structures in pelvis and its early management to prevent complications c) parental counselling for prognosis - immediate and late (especially if other anomalies co-exist).

Consent

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