ORIGINAL RESEARCH PAPER

Neonatology

KEY WORDS: Neonatal Pyometra ; Pyocolpos ; Imperforate hymen in children ; Haematometra and Haematocolpos in children

# CASE SERIES: NEONATAL PYOMETRA AND PYOCOLPOS: A CLINICAL CHALLENGE

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**ABSTRACT**

Haematometra and hematocolpos secondary to congenital vaginal obstruction has been commonly reported in literature, especially at the onset of puberty. But presentation of neonatal or congenital pyometra and pyocolpos is uncommon and unclear about its origin. The grossly distended genital tract may cause pressure symptoms on urinary obstruction and the enlargement of the bladder may hinder palpation of the vaginal cyst. Pyuria will be a further complication in cases of secondary hydroureter and hydroneprosis due to pyometra. Posterior pressure may also cause rectal obstruction. In this communication we present our experience with three cases of neonatal pyometrocolpos along with review of the pertinent literature.

**INTRODUCTION:-**

We all know the common term haematometra / haematocolpos , when vagina with or without uterus becomes distended with menstrual blood due to obstruction[1]. But when the same picture is seen in neonates, this is usually due to the distension caused by cervical & vaginal secretions. Different causes like Imperforate hymen ,congenital vaginal agenesis, transverse vaginal septum – high / low, or urogenital sinus / cloaca can lead to this condition. Due to maternal hormonal changes, the fetus produces lot of mucoid secretions, which get collected causing significant distension of these organs[2].

Godfrey described this clinical presentation in very early period of 1856[3]. Very less literature was available since then until Mahoney & chamberlain published elaborated description about the same in 1940[4].

Commonly it may present with a bulging hymen (imperforate hymen) or may present with abdominal mass , may be with pressure symptoms like urinary retention, dysuria or constipation. Parents usually come in a distressing situation if the presentation is like respiratory distress. Neonatal Clinical examination has its own limitations & routine ultrasonography can not really detect the cause of such swellings. Timely evaluation by the experienced person , & correct diagnosis helps in proper management considering the long term affection of her further sexual & reproductive life.

The reason of publishing our case series is to make everyone aware about varied presentations of this clinical entity. Till now very few cases of neonatal pyometrocolpos have been reported all over the world with unclear opinions about source of bacterial colonisation and exact patho-physiology for the same. Depending upon the presentation and size of swelling confirming with radiological investigations, approach for surgical intervention (trans abdominal or vaginal ) is decided.

The management needs consideration of two conditions : first, the collection which needs immediate drainage to relieve the pressure symptoms & also to treat infections in case of pyometra . Second, the best possible treatment to treat the obstruction in some part of vagina to avoid recurrence , as well as due consideration is needed for future sexual & reproductive functions. Due to these complexities, inappropriate treatment due to misdiagnosis / delayed diagnosis are very common , thus warranting further illustration in literature / text books .

**Case Reports :-**

**Case 1:-**

5 months old female child brought by parents with complaints of excess cry while passing urine, increased lower abdominal distension and excessive watering from right side eye since birth. Antenatal screening was not done and it was normal delivery. Due to poor compliance of parents, bay was brought only when started presenting with severe problems .On clinical examination, baby was healthy with normal cry tone activity. Right side upper eyelid deformity - coloboma (Fig.1) was present. On abdominal examination Single ,oval shaped, firm, non-tender, smooth surfaced lump palpable in lower abdomen in midline extending from pubic symphysis to lower border of umbilicus. On genital examination, external appearance was normal with no any evidence of bulged membrane or cyst but no vaginal orifice seen. On hematological invetsigations, no abnormal variations noted. Ultrasonography of abdomen-pelvis ( Fig.2 & Fig.3 ) revealed two cystic swellings, One huge oval shaped cystic swellings noted in pelvis anterior to rectum and another cystic swelling in abdomen above the lower cystic swelling smaller in size. Urinary Bladder was shifted upwards with stretched out urethra. On Computerised tomography scan with intravenous contrast ( Fig.4 ) there was evidence of two large loculated swellings in pelvis anterior to rectum arising due to collection in vagina and uterus suspicious of haematometra - haematocolpos. On right side ovary could not be visualised. Urinary Bladder was not visualised . Rest Ultrasonography findings confirmed. Patient was planned for Ultrasound guided pig tail catheter insertion under local anasthesia but as distended bladder was coming in between making procedure difficult , exploratory laparotomy was planned. Under general anaesthesia, on diagnostic cysto-genitoscopy there was normal urethral meatus with normal urinary bladder but no vaginal orifice seen. Urinary bladder catheterised with foley's catheter No 8. Exploratory laparotomy performed with Pfannenstiel incision, urinary bladder was lifted up from cystic swelling seen posterior to bladder (Fig.5 & 6). Around 190 ml pus drained out from swelling with small incision with foley's catheter no 10 kept in situ for postoperative drainage of the cavity . After complete decompression the uterus and vagina could be identified properly. Pus was sent for culture and sensitivity. Wound closed in layers. On post operative day 10 , contrast study with urograffine 76% dye through drain in situ was performed. Size of cavity was reduced with well defined cervical imprint seen but no flow per vagina confirming vaginal atresia(Fig.7). Drain removed after dye study and patient was kept on postoperative follow up. On postoperative three months ultrasonography abdomen and pelvis screening revealed normal anatomy. Culture reports of pus reported E Coli infection. Reconstructive vaginoplasty is planned in subsequent sittings.

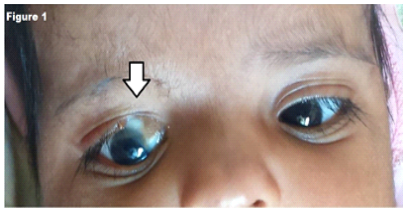


**Figure 1 :- Coloboma on Right side upper eyelid**

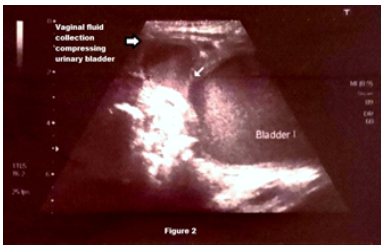


**Figure 2:- Ultra-sonographic evaluation**

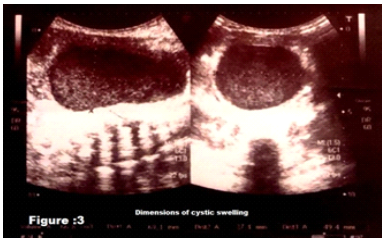


**Figure 3:- Ultra- sonographic dimensions**

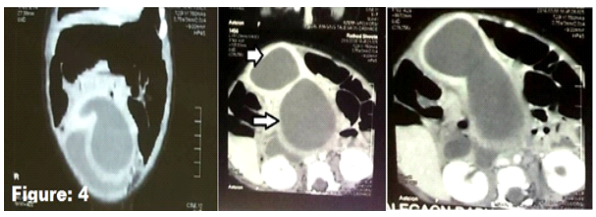


**Figure 4:- Computed tomographic scan evaluation showing extend of cyst.**

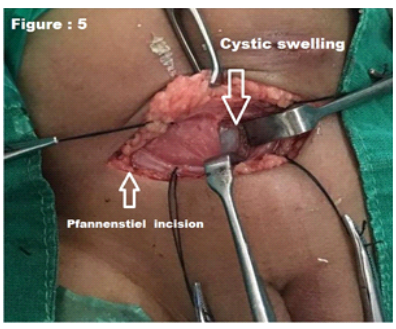


**Figure 5 :- Intra-operative findings of vaginal fluid**

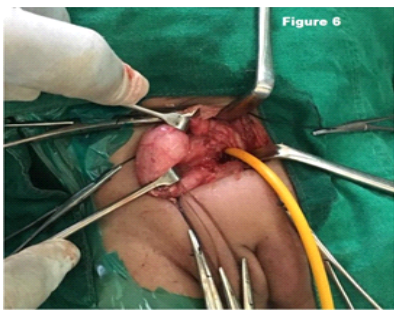


**Figure 6 :- After inserting indwelling intravaginal catheter draining cystic swelling '''**

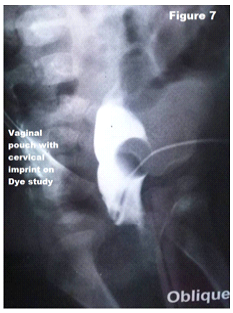


**Figure 7:- Post-operative Dye Study showing delineated Vaginal pouch with Cervical imprint .**

**Case 2**

A normal delivered full term newborn baby referred immediately after birth for prominent cystic bulge seen at genital region[Fig.8]. Antenatal fetal screening sonography till 28 weeks of gestation was normal study. On local examination large cystic bulge at genital region seen with no separate urethral meatus and vaginal orifice seen. Ultrasound screening of abdomen and pelvis revealed huge loculated cystic swelling in pelvis extending up to abdomen with absent right side kidney and ovary. Magnetic resonance imaging with intravenous contrast study showed a single huge loculated swelling arising from fluid collection in vagina and uterus with absent right side ovary and kidney suspecting haematometra - haematocolpos secondary to imperforate vaginal membrane [Fig.9]. Under general anaesthesia, around 180 ml pus drained out with cruciate incision taken on prominent site of bulge with foley's catheter no 10 kept in situ for postoperative drainage. After complete decompression of the cavity , urethral meatus is seen normally. On culture report of pus drained from cavity showed E coli growth. Post operatively drain was removed on day 10. Ultrasound screening showed no residual collection. Patient was kept on vaginal dilatation with hegar's dilator. On postoperative one year follow up till date ,baby is doing well with no any fresh complaints .

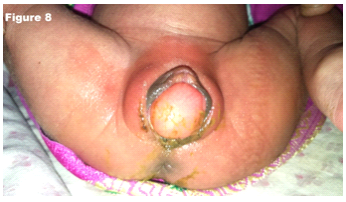


**Figure 8 :- Prominent cystic bulge seen at genital region**

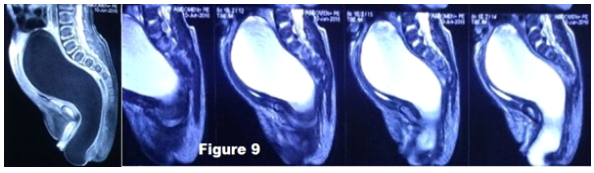


**Figure 9 :- Magnetic resonance imaging with intravenous contrast study.**

**Case 3**

A full term newborn baby delivered by lower segment caeserian section for obstructed labour referred for prominent vaginal bulge seen immediately after birth. On local examination there was prominent vaginal bulge with no urethral meatus seen. Ultrasound examination of Abdomen and pelvis revealed large amount of fluid collection in vagina with minimal intrauterine fluid collection USG suggestive of haematocolpos - haematometra. Rest uro-genital findings were normal. Under general anaesthesia around 70 ml frank pus was drained out with cruciate incision on bulge and foley's catheter no. 10 kept in situ for postoperative drainage [Fig.10]. On post operative day 10,ultrasound revealed no residual fluid collection. Fluid on culture showed E.Coli growth. Drain removed and patient was kept on vaginal dilatation with Hegar's dilator. On post operative 9 months follow up patient is doing well with no any fresh complaints.

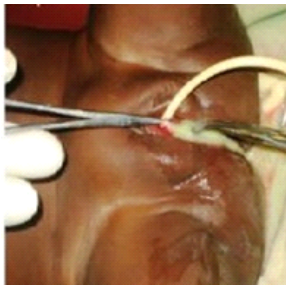

**Figure 10**

## DISCUSSION

Hyrocolpos or pyocolpos secondary to imperforate hymen is very uncommon disorder noticed in neonatal period , usually characterised by vaginal distension due to increased secretion by cervical mucus glands due to maternal hormone stimulation which gradually accumulates and may present as a pelvic mass[5]. Commonly hydrocolpos presents with compression effects on urinary system presenting with hydroureter and hydronephrosis ,ultimately renal damages. This can be prevented by draining fluid immediately after birth. Hammad and Upadhyay revealed in their studies that hydrometrocolpos was the cause in 23% of patients with infra-vesical obstruction and 39% of those cases presented at birth as a result of prenatal ultrasound diagnosis of pelvic pathology [6]. Such cases with prenatal diagnosis of pelvic mass should be labelled as high risk deliveries and should be promptly intervene by multidisciplinary approach immediately after birth. Tras-abdominal approach with indwelling catheter proved less recurrence with fluid re-accumulation and more preferred over trans-vaginal approach [7]. Prenatal ultra-sonography and MRI imaging provides additional anatomic details with excellent soft tissue contrast to determine the thickness of the transverse septum, length of the atresia, and the presence or absence of a cervix [8].

Our patients in this study did not have fetal MRI but it can be useful in further differentiating pelvic mass. Source of infection in case of hydrocolpos and haematometra with vaginal atresia turning into pyometra is not mentioned in literature yet though many theories have been proposed. In our study we could see growth of E.Coli growth in two cases. One case presented with no growth. Etiology for same is still under discussion.

## Conclusion

Diagnosis of pyometrocolpos in not difficult but it is important to diagnose cause of pyometrocolpos. MRI scan is helpful in prenatal and postnatal period to delineate exact anatomy of defect and planning management [9]. Main aim of surgical intervention is to create normal looking vaginal orifice fully separate from urethral orifice to prevent future chances of urinary tract infections, re-accumulation of fluid during menstrual cycle. Further studies required to find out source of infection causing pyometra presenting in perinatal and early neonatal period .

Imperforate hymen or membrane in the lower one third of the vagina leading to hydrocolpos is very common in which newborn may present with lower abdominal palpable swelling and bulged out membrane at vaginal orifice immediately after birth. There may be symptoms associated with compression effects on urinary bladder and other viscera like rectum, colon depending on size of swelling. As per literature mentions about the fluid getting accumulated in hyrocolpos and hydrometra is believed to be a secretion by cervical glands occurring under influence of maternal hormones. Maternal oestrogen may lead to shading of uterine cells and may start neonatal menstruation. In associated cases of imperforate hymen or any other congenital vaginal obstructions this may lead to hematometra and hematocolpos. But very few cases of neonatal pyometra and pyometra cases have been reported all over the world with unclear opinions about source of bacterial colonisation and exact patho-physiology for the same. Depending upon the presentation and size of swelling confirming with radiological investigations, approach for surgical intervention (trans abdominal or vaginal ) is decided. The transabdominal drainage of hydrocolpos with indwelling tube is more preferred than transvaginal drainage to prevent reaccumulation [10]. The drainage is performed by interventional radiologist transabdominally under US which enables real-time evaluation without radiation exposure [11]. In general, infants with hydrocolpos and urogenital sinus have increased risk of sepsis due to collection of urine in vaginal vault. There have been reported deaths due to sepsis associated with hydrocolpos [12].

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