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CASE REPORT

A Rare Association of Small Bowel Atresia and In-Utero Midgut Volvulus Presenting as Fetal Meconium Pseudocyst

Jih Huei Tan*[1], Henry Chor Lip Tan[1], Zi Qin Ng[2], Najua Ramli[1], V Muthualhagi A/P Vellusamy[1]

[1]Sultanah Aminah Hospital, Malaysia ,[2]Department of General Surgery, St John of God Public and Private Hospitals, 1.Clayton Street, Midland, 6056, Australia

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ABSTRACT

Fetal meconium peritonitis (MP) is a rare occurrence. Neonate that survives this rare condition is likely to form a meconium pseudocyst. Hereby, we present a case of a 33-year-old pregnant woman that presented with premature rupture of membranes at 34 weeks of gestation. Antenatal ultrasonography at 33 weeks revealed abnormal dilated bowel within the viable fetus. An emergency Cesarean section was performed and a baby boy was delivered at 34 weeks. Physical examination of the baby at birth revealed a palpable central abdominal mass. Lower gastrointestinal contrast imaging revealed a non-opacified dilated proximal small bowel and opacified collapsed distal small bowels and colon. A laparotomy was carried out and revealed MP with pseudocyst formation due to a midgut volvulus. Interestingly, meconium contamination was confined due to associated atretic small bowel which occurred secondary to the volvulus.

Key words: Meconium peritonitis; Pseudocyst; Midgut volvulus

INTRODUCTION

Midgut volvulus commonly manifests during the 1st month of life (60% of cases). Its occurrence during the prenatal period is rare with the majority of such pregnancy that is spontaneously aborted or undergoes in utero death [1]. Meconium peritonitis (MP) is a rare entity, which is often diagnosed by antenatal ultrasound [2]. They may form meconium pseudocyst at birth [3]. Herein, we report on a rare case of a premature baby boy presenting with an abdominal cystic mass. Antenatal ultrasound revealed dilated bowels without any ascites or calcification. Laparotomy revealed a meconium pseudocyst containing a segment of dilated gangrenous small bowel separated from the dilated proximal small bowel and collapsed distal small bowel.

CASE REPORT

A healthy 33-year-old gravida two pregnant woman underwent a transabdominal ultrasound scan in an obstetric clinic at 33 weeks. The fetal sonogram showed dilated bowel loops but no ascites, calcification, or pseudocyst was detected. The baby was delivered through emergency cesarean section at 34 weeks due to premature rupture of membranes.

A 2.400 g male neonate was delivered with a good Apgar score of 9 at birth. On clinical examination, there was no gross dysmorphic features. A cystic mass measuring 3 cm × 3 cm was palpable over the right side of the abdomen. The nasogastric tube aspirate drained approximately 60 mL of greenish fluid within the first 3 h of life.

Abdominal X-ray depicted dilated proximal jejunal loops and distal small bowel and colon shadow were not visualized (Figure 1) contrast enema revealed a patent distal small bowel; colon and rectum which were opacified by contrast.

An upper transverse laparotomy was performed and revealed a midgut volvulus with meconium pseudocyst formation onto the inferior liver surface. The volvulus segment of small bowel (15 cm) was autoamputated from the atretic ends of proximal and distal small bowel (Figure 3). Adhesions between the small bowels were released and necrotic volvulus segment of small bowel was resected. Proximal and distal small

Correspondence*: Jih Huei Tan, Sultanah Aminah Hospital, Malaysia
E-mail: huei_87@hotmail.com

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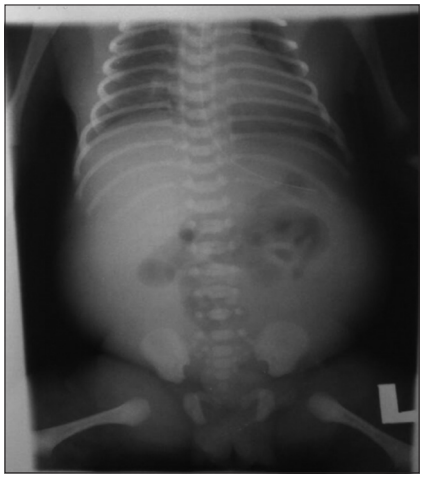

Figure 1: Plain abdominal radiograph showing dilated proximal small bowels

bowels were placed over the right lower abdomen as a double-barrelled stoma. Postoperatively, the patient with remaining 35 cm of short bowel developed short bowel syndrome with high output stoma losses. Total parenteral nutrition was instituted. Reversal of stoma was done 6 weeks post-surgery, and the baby has made a steady recovery on the last follow-up 6 months post-surgery.

DISCUSSION

MP is a rare condition which is often diagnosed prenatal period with the advances of antenatal imaging. It is a sterile inflammation with the meconium contamination, a condition where the results have been reported to improve in recent times [4]. The incidence is 1 in 30,000 live births [2]. The causes of the MP reported were small bowel atresia, stenosis, volvulus, intussusception, Meckel's diverticulum, meconium ileus, imperforate anus, or peritoneal bands [5].

In fetus with MP due to midgut volvulus, the perforated intestinal loops leaks meconium. In minority of cases, the omentum or surrounding intestinal loops plug off the perforation [2]. However, in the present case, the perforated intestinal segment that had undergone volvulus was isolated from proximal and distal small bowel due to secondary atresia. The presence of intestinal atresia led to limited meconium contamination and the formation of meconium pseudocyst. This explained the baby having a good Apgar score at birth and remained relatively well during the neonatal period.

An antenatal fetal ultrasound imaging aids in early diagnosis by detecting fetal ascites, intra-abdominal calcifications, echogenic bowel, dilated bowel loops, pseudocyst formation, and/or polyhydramnios [2]. In this case, the only ultrasound finding was the dilated bowel loops. Ascites and calcification were not seen. This may be due to the walled off meconium contamination by surrounding omentum and small bowels or missed finding by the sonographer. The presence of small bowel atresia limits the extent of meconium contamination which lessened the chance of seeing a calcification on ultrasound. Ascites was also not present in few other reports of fetal midgut volvulus. In the current case, pseudocyst could not be detected may be due to poor window secondary to the dilated bowel shadow.

Mortality rates of MP were reported to have reduced from 40% to <10% in a recent series [2]. The risk factors of mortality include low birth weight and the presence of sepsis [2,6]. In our case, the patient thrived well at the neonatal period as expected with a good birth weight of 2.4 kg and limited sepsis due contained meconium contamination.

However, there were few learning points of the case. Accurate prenatal ultrasound by trained personnel is essential to look for all features of MP. When doubt exists, contrast enema is an useful alternative imaging to diagnose MP secondary to either atresia, volvulus or malrotation, and exclude other pathology such as Hirschsprung disease. Early identification of this entity potentially carries good outcome to the newborn with current health-care.

CONCLUSION

MP secondary to intrauterine midgut volvulus is a rare neonatal surgical emergency that has a good prognosis with present day health-care.

Author's Contribution

All authors contributed equally in concept, design, literature review, drafting the manuscript, and approval of the final manuscript.

Consent Statement

Authors declared that they have taken informed written consent, for publication of this report along with clinical photographs/material, from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient however it cannot be guaranteed.

REFERENCES

1. Molvarec A, Bábinszki A, Kovács K, Tóth F, Szalay J. Intrauterine intestinal obstruction due to fetal midgut volvulus: A report of two cases. Fetal Diagn Ther 2007;22:38-40.

2. Ping LM, Rajadurai VS, Saffari SE, Chandran S. Meconium peritonitis: Correlation of antenatal diagnosis and postnatal outcome - an institutional experience over 10 years. Fetal Diagn Ther 2017;42:57-62.

3. Keskin U, Karasahin KE, Ozturk M, Atabek C, Demirbağ S, Ergün A, *et al.* Prenatal diagnosis of the acute meconium peritonitis secondary to ileum volvulus perforation: A case report. Fetal Pediatr Pathol 2015;34:9-13.

4. Uchida K, Koike Y, Matsushita K, Nagano Y, Hashimoto K, Otake K, *et al.* Meconium peritonitis: Prenatal diagnosis of a rare entity and postnatal management. Intractable Rare Dis Res 2015;4:93-7.

5. Konje JC, de Chazal R, MacFadyen U, Taylor DJ. Antenatal diagnosis and management of meconium peritonitis: A case report and review of the literature. Ultrasound Obstet Gynecol 1995;6:66-9.

6. Chan KW, Lee KH, Wong HY, Tsui SY, Wong YS, Pang KY, et al. Cystic meconium peritonitis with jejunoileal atresia: Is it associated with unfavorable outcome? World J Clin Pediatr 2017;6:40-4.