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CASE REPORT PEER REVIEWED | OPEN ACCESS

Meconium pseudocyst without radiological calcification

Reshma Doodnath, Cidi Dubay, Roger Khan

ABSTRACT

Introduction: Meconium peritonitis is a rare disease which can be potentially fatal.

Case Report: We present the history of a full-term male infant born at 2945 g via lower segment cesarean section (LSCS). There was an antenatal diagnosis of an intra-abdominal cyst. At 27+6 weeks gestation, the ultrasound scan noted a thick-walled cystic mass superior to the bladder. Postnatally, an X-ray showed a paucity of gas in the right lower quadrant and an ultrasound scan confirmed a large central avascular cystic lesion, with posterior sediments noted. Laparotomy revealed extensive adhesions surrounding a meconium filled cyst. This was excised and a primary anastomosis performed. However, he failed to progress with feeds, having intermittent abdominal distension, and after a contrast study on day 18 post op which showed failure of contrast to pass the dilated proximal jejunum, he was taken back to the operating room and was found to have significant adhesions. Adhesiolysis was done and the anastomosis was patent and well healed. However, these episodes of abdominal distension continued, and he never progressed with feeds. On day 39 of life, there was severe metabolic acidosis refractory to resuscitation, leading to cardiac arrest.

Conclusion: There can be subtle presentations of meconium peritonitis in the newborn and it should be considered in any neonate with an intra-abdominal cyst.

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INTRODUCTION

Meconium peritonitis is a rare but potentially dangerous and sometimes lethal disease of the newborn. It is thought to be due to intrauterine perforation that can happen early or late in pregnancy and the incidence has been found to be about 1 in 35,000 newborn live births [1]. A variety of pathologies is thought to possibly cause this which includes stenosis, atresia, and volvulus to name a few. As a result of any of these pathologies, antenatal bowel perforation occurs and this results in sterile peritonitis, which in turn causes a chemical reaction that leads to a fibrotic reaction with intra-abdominal calcifications. This is a pathognomonic feature of meconium peritonitis which is seen on abdominal X-ray (AXR) [2] but these are not always present [3]. The most common cause of intra-abdominal calcifications is meconium peritonitis, and this can present in different ways that are determined by the underlying cause and at what stage in the pregnancy perforation occurs, in addition to whether sealing of the perforation happens on its own. Surgical intervention is usually warranted to treat the underlying cause and achieve intestinal continuity. Even though survival outcomes have improved over the years with improvements in antenatal diagnosis, neonatal intensive care and postoperative management, an overall mortality of about 11% is still estimated to exist [4]. This case report describes an infant with an intra-abdominal

cyst, with no evidence of intra-abdominal calcification, who unfortunately had a poor outcome.

CASE REPORT

A full-term male infant born at 2.945 kg was delivered via LSCS to a 29-year-old mother, gravida 1+0. There was an antenatal diagnosis of an intra-abdominal cyst. At 27 weeks gestation, the ultrasound scan (USS) described a 5.8 cm × 5.4 cm × 4.9 cm thick-walled cystic mass noted superior to the bladder with internal septations and debris that suggested a fetal intestinal cyst. There was no evidence of echogenic bowel, dilated loops, or ascites. Postnatally, an X-ray showed a paucity of gas in the right lower quadrant with no intra-abdominal calcification as seen in Figure 1 and no free air evident on a left lateral decubitus film as seen in Figure 2.

An USS confirmed a large central avascular cystic lesion with posterior sediments noted, with the largest portion being 4.1 cm in diameter, with uncertainty as to the origin of the cyst. There was good bowel peristalsis, and all the solid organs were normal in appearance. Laparotomy findings revealed a large meconium-filled cyst in the right abdomen with extensive adhesions around the entire cyst as seen in Figure 3, making dissection difficult.

Bowel proximal to the cyst was dilated and measured 20 mm and appeared to enter the cyst. The bowel leaving the cyst was collapsed patent jejunum as seen in Figure 4, and measured 8 mm. Patency of entire distal small bowel and all large bowel was checked. The cyst was excised in its entirety and a primary end-to-end anastomosis was done with interrupted 5/0 polydioxanone (PDS). No intra-abdominal calcifications were seen at laparotomy.

Gross pathological examination revealed a tan dusky cyst that measured 6.1 cm × 3.6 cm × 2.7 cm, which was connected to a dilated segment of small bowel proximally and a collapsed segment of small bowel distally. Green watery stool was noted within the cyst which had a greenish tinge mucosal lining. Histology revealed the absence of a mucosal lining with replacement by macrophages. The cyst wall was composed of fibrotic submucosa and organized smooth muscle, which was continuous with the normal intestine, which showed necrosis and attenuation in some areas. There were numerous calcified deposits and multi-nucleated giant cells and hemosiderin-laden macrophages within the cyst wall. Overall features were consistent with a meconium pseudocyst.

However, he failed to progress with feeds, having intermittent abdominal distension, and after a contrast study on day 18 postoperatively which showed failure of contrast to pass the dilated proximal jejunum, he was taken back to the operating room and was found to have significant adhesions. Adhesiolysis was done and the anastomosis was found to be patent and well healed. However, these episodes of abdominal distension continued, and he never progressed with feeds. On day 39 of life, he had severe metabolic acidosis, had a cardiac arrest, and was unable to be resuscitated.

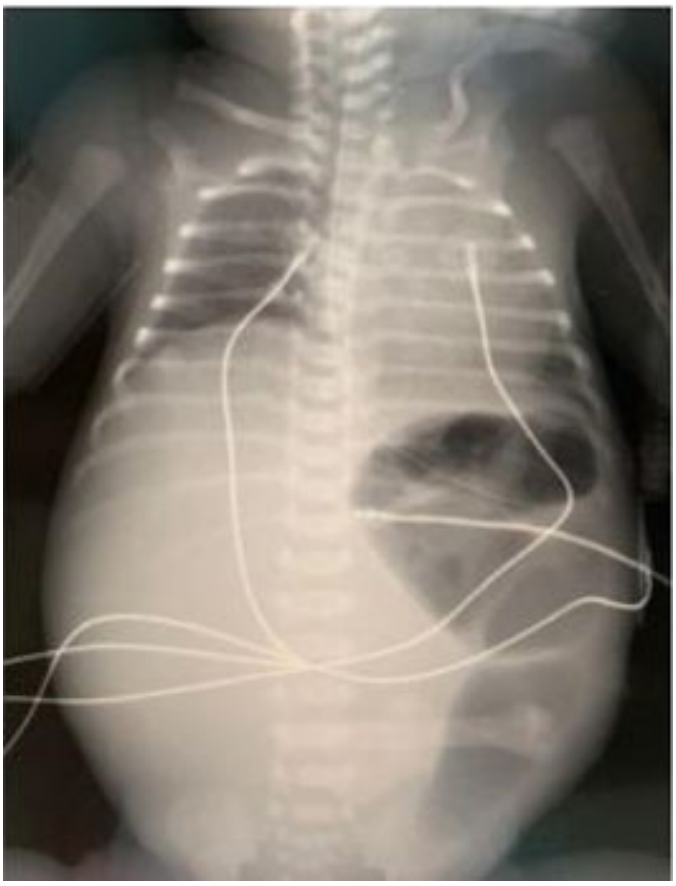

Figure 1: Supine abdominal X-ray demonstrating a right-sided paucity of gas with no calcification.

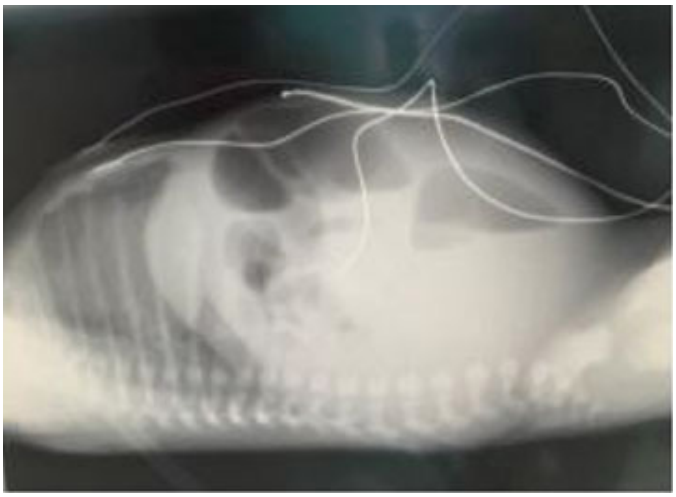

Figure 2: Lateral decubitus film with no calcification and no free air.

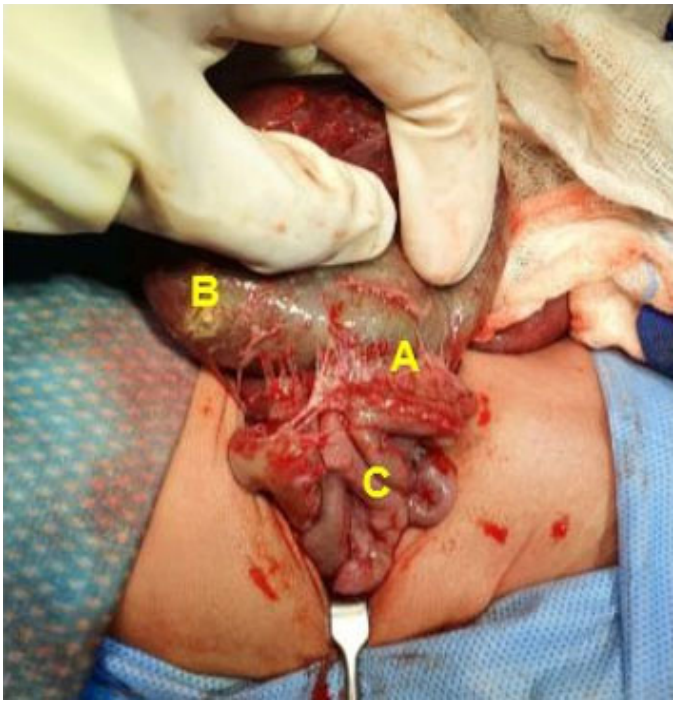

Figure 3: Intraoperative picture showing extensive adhesions (A) between the entire cystic structure (B) and the collapsed small bowel (C).

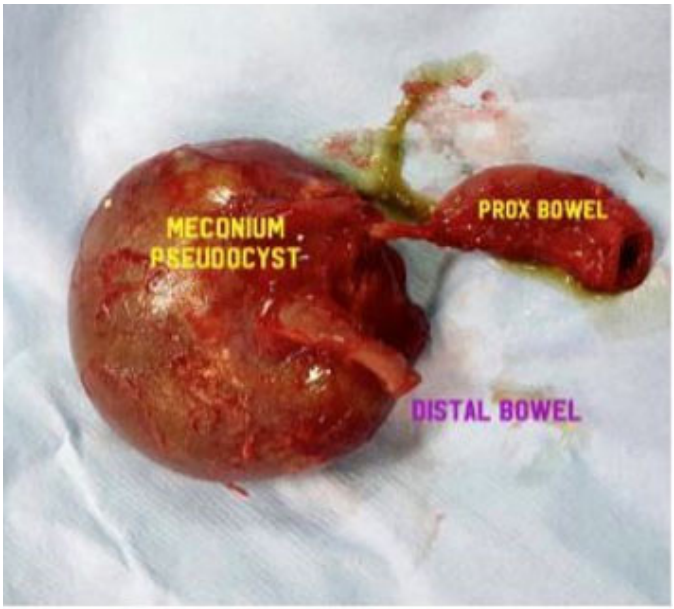

Figure 4: Cystic structure between proximal and distal bowel.

DISCUSSION

Many different pathologies can give rise to an antenatally visualized intra-abdominal cyst in a fetus which includes intestinal mesenteric cysts and pseudocysts, duplication cysts, choledochal cysts, congenital cysts of the pancreas, renal cysts, obstructive uropathy, urachal cysts, ovarian cysts, ureteroceles, and tumors like cystic *Sacroccocygeal teraroma*s [3]. Meconium peritonitis is a sterile, chemical peritonitis which results from in-utero fetal bowel perforation and occurs in approximately 1 in 10,000 live births, with severe cases leading to serious morbidity and mortality. The perforation is usually in the distal small bowel proximal to an obstruction, and the cause of the obstruction can vary, with pathologies ranging from atresia, stenosis, volvulus, internal bowel hernia, Meckel's diverticulum, meconium ileus, and peritoneal bands, with the commonest of these being intestinal atresia and meconium ileus. As a result of the body's reaction to contain the meconium collection extruded into the peritoneal cavity, adhesions form between loops of intestine and the omentum, and this creates a cystic mass that can be seen on ultrasound. In the case of a sealed perforation, there may sometimes be a solid non-cystic mass with calcium deposits which seal off the perforation [5]. This may sometimes cause a diagnostic dilemma as the formation of this apparently solid abdominal mass occurs, making an accurate diagnosis between an abdominal tumor and meconium collection somewhat difficult.

Said et al. have described three different types of meconium peritonitis based on the ultrasound findings which have been described. These features vary depending on the stage of development in which the bowel perforation occurs and the severity of the inflammatory reaction induced by the meconium that has entered the peritoneal cavity [6]. The most frequent has been found to be the fibroadhesive type and is characterized by an intense fibroblastic reaction. This results in the formation of fibrotic membranes which can be quite adherent to the intestinal wall and they sometimes cover the perforation. On ultrasound imaging, the presence of diffuse punctiforme hyper-echogenic lesions around the peritoneal cavity can sometimes be seen and intra-abdominal calcifications are not usually observed. Some other characteristic findings include ascites, hydramnios, or bowel loop dilatation seen on antenatal imaging. The perforation often seals spontaneously and may not be easily seen.

The second most common type is the cystic type which results in a pseudocyst, and this is formed by a meconium collection that is surrounded by fibrotic membranes. This can be demonstrated sonographically by a large cystic structure filled with meconium that is lined by a thick membrane, and multiple calcium deposits may be seen [7]. It has been found that the cystic type usually forms because of prenatal volvulus with perforation.

The last category is the generalized type, and this occurs as a result of a peri-natal perforation with meconium spread throughout the abdominal cavity.

It is reported in a recent review of 12 patients with meconium peritonitis, there was intra-abdominal calcification in 60% of cases with cystic fibrosis, and in 100% of the cases without cystic fibrosis and it is postulated that the low concentration of pancreatic enzymes found in 80% of patients with cystic fibrosis may be necessary

for the calcifications to occur [8]. Calcification may take weeks to be visible on ultrasound. There was no evidence of cystic fibrosis in this reported case.

There has been one study by Zangheri et al. which has demonstrated the sonographic relationship with the post-natal course that occurs in meconium peritonitis, where 69 patients with meconium peritonitis were divided into four grades according to ultrasound features [9]. These were:

Grade 0—isolated intra-abdominal calcifications

Grade 1—intra-abdominal calcifications and ascites or pseudocyst or bowel dilatation

Grade 2—two associated findings

Grade 3—all sonographic features.

It was found that there was an increasing need for neonatal surgery with higher grades of the sonographic classification [9].

There are very few cases in the literature of infants with meconium peritonitis and pseudocyst formation without calcification, as this is usually a classic sign in a meconium pseudocyst in a neonate, accompanied by ascites and bowel dilatation. Valladares et al. reported a case of a giant pseudocyst secondary to volvulus of the ileum and perforation, with no intra-abdominal calcification seen on X-ray [3]. That infant also had a turbulent course, having to have a second laparotomy because of bowel obstruction, had short bowel syndrome, and died at seven months of age. Our case was of a very similar presentation, with only an intra-abdominal cyst noted on antenatal ultrasound, with no ascites of bowel dilatation, and severe adhesions forming postoperatively, with abnormal function of the intestine.

CONCLUSION

Meconium peritonitis and pseudocyst formation is a rare neonatal condition and can cause significant morbidity and mortality. When an echogenic intra-abdominal mass is observed on an antenatal scan, meconium pseudocyst should be considered in the differential diagnosis. The antenatal appearance is usually accompanied by signs of bowel obstruction, such as polyhydramnios and bowel dilatation, and ascites may or may not be present. However, it is important to note this case and recognize the occasional absence of the typical ultrasound features one will expect with a meconium pseudocyst. Thus, it may present without these features and should be considered in the differential diagnosis and workup of a neonate with an abdominal mass. This will ensure delivery in an appropriate center with neonatal and pediatric surgical expertise, where prompt and appropriate treatment can be facilitated as soon as possible after birth.

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Author Contributions

Reshma Doodnath – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Cidi Dubay – Conception of the work, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Roger Khan – Conception of the work, Acquisition of data, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

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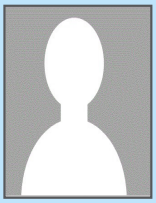

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