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CASE SERIES

Hernia of the cord with patent omphalomesenteric duct and ileal prolapse in two preterm neonates: case series

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ABSTRACT

Hernia of the cord (HOC) is a rare condition that results from failure of the viscera to return to the abdominal cavity by the 10th week of fetal development. It sometimes presents together with a patent omphalomesenteric duct (POMD), another rare condition, which occurs earlier in fetal development. A proportion of POMD cases may also have ileal prolapse (IP) through the POMD lumen. Neonates diagnosed with the combination of these rare clinical conditions require immediate surgical intervention to resect the POMD and non-viable bowel segments, reduce the hernia, and repair the umbilical defect. In this case series, we report two neonates diagnosed with HOC with POMD and IP. One patient who had complete IP was not fit for immediate surgery and died of respiratory distress and sepsis. The other patient had a successful surgical correction of the congenital defects, but succumbed to sepsis postoperatively. Treatment of patients with this combination of clinical conditions should focus mainly on both surgical correction, and infection control and management.

Keywords. omphalocele, congenital umbilical cord hernia, vitelline duct anomalies, neonatal sepsis

INTRODUCTION

Hernia of the cord (HOC), a small omphalocele with less than 1.5 cm umbilical defect, affects 0.025% of all neonates. Embryologically, it results from failure of the viscera to return to the abdominal cavity by the 10th week of fetal development, leaving behind an intact omphalocele sac.[1] Patent omphalomesenteric duct (POMD), on the other hand, occurs earlier in fetal development. The duct remains patent because the vitelline or omphalomesenteric duct fails to obliterate and supposedly terminate the communication between the primitive gut and the yolk sac by the 7th week of development.[1] POMD affects 0.1% of all neonates[2] and belongs to a spectrum of vitelline duct anomalies that also includes Meckel's diverticulum, Meckel's band, umbilical polyps, and umbilical cysts. Since it is continuous with the small bowel, POMD will present externally in the abdomen as a mucosal opening, with fecal discharge, in the umbilicus. About 11.8% of all POMD cases are associated with HOC. Additionally, 5.9% of POMDs may also present with ileal prolapse (IP) through the POMD lumen.[2] The prolapse results from postnatal increase in intra-abdominal pressure, usually upon coughing secondary to a pulmonary infection or a congenital pulmonary pathology.[3] Clinical information regarding the simultaneous occurrence of HOC, POMD, and IP in a single patient is very limited.[2]

The diagnosis of HOC with POMD and IP is usually established clinically after a careful physical examination of the patient. A fistulogram is usually done to demonstrate continuity of the POMD with the ileum and to facilitate surgical planning. Because these clinical conditions can occur with other congenital anomalies, such as cardiac and musculoskeletal conditions,[1] doing other congenital work-ups—including a babygram and a two-dimensional echocardiogram (2D echo)—may be necessary. Management requires treatment of all components (HOC, POMD, and IP) in a single procedure through an umbilical exploration. Typically, the surgical procedure involves reduction of the IP, resection of the POMD up to its base within the ileum and anastomotic repair of the remaining ileum, repair of the HOC, and umbilicoplasty.[3] The prognosis of HOC with

IN ESSENCE

Patent omphalomesenteric duct (POMD) and hernia of the cord (HOC) are rare congenital anomalies that occur on the 7th and 10th week of fetal development, respectively.

In this case series, two female neonates were diagnosed with HOC with POMD and ileal prolapse at birth.

One patient was unfit for surgery and died of respiratory failure and sepsis 10 days after diagnosis. The other patient had a successful corrective surgery, but died of sepsis post-operatively.

POMD and IP is usually not favorable, and the outcomes of patients having the condition are usually affected by the promptness of diagnosis and intervention, and the presence of concomitant congenital anomalies and complications.[4]

In this case series, we present two patients who both had HOC with POMD and IP. They were referred within the same week to our Pediatric Surgery Service for assessment and management. They had similar presentations in the beginning, but their comorbidities and the rest of their subsequent clinical courses differed.

Patient A

CLINICAL FEATURES

Patient A, a female neonate, was referred immediately after birth to the Pediatric Surgery Service for co-management due to an umbilical mass. The patient was delivered in our institution via repeat Cesarean section, with APGAR scores of 5, 6, and 8 at 1, 5, and 10 minutes, respectively, and a Ballard score of 33 weeks. At birth, the patient had signs of respiratory distress, including cyanosis, alar flaring, and both subcostal and intercostal retractions, which prompted the Pediatrics service to intubate the patient.

The patient's mother was 39 years old, gravida 2, para 2 after the patient's birth. Her first delivery was by Cesarean section for cephalopelvic disproportion. Antenatal care had been adequate, and she had no maternal illnesses, physical trauma, or exposure to any harmful substances during her second pregnancy. A prenatal ultrasonography performed at 30 weeks age of gestation (AOG) did not reveal any remarkable findings. Reverse transmission - polymerase chain reaction (RT-PCR) test for COVID-19, a routine test for all in-patients in our institution at the time of the mother's admission for delivery, revealed that the mother was negative for SARS-CoV-2 infection. The patient's family history was unremarkable.

We did the physical examination while the patient was placed in a radiant warmer and hooked to a mechanical ventilator in assist-control mode. Body temperature of the patient was 36.6°C, and heart rate was within the range of 135 to 147 beats per minute. The patient weighed 1.9 kilograms (kg), measured 44 centimeters (cm) in length, and had a head circumference of 30 cm and chest circumference of 27 cm, all adequate for gestational age (AGA). We did not hear any cardiac murmurs, and breath sounds were clear. There was a round, soft mass measuring 3×3×3 cm protruding from the patient's umbilicus, with a round base measuring 1.5 cm in diameter. The mass contained part of the umbilical cord and some bowel loops, which could be seen through a translucent sac that covers most of the surface area of the mass. A tubular

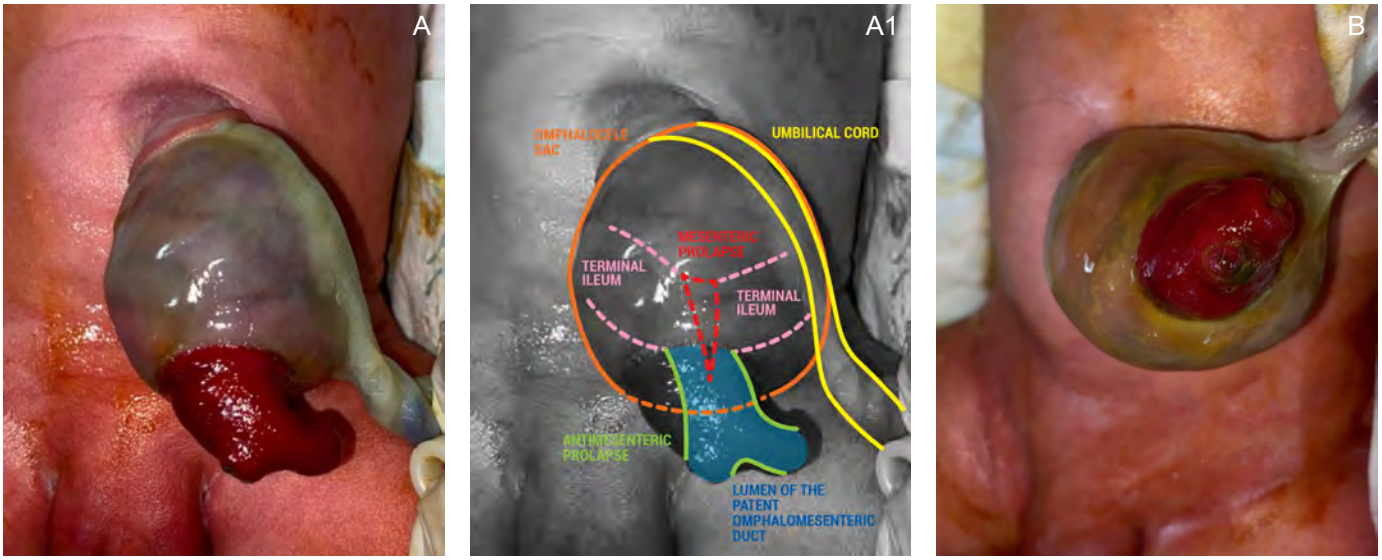

Figure 1 An omphalocele sac, 3x3x3 cm in size, containing part of the umbilical cord and some small bowel segments protruding into it. The patent omphalomesenteric duct (POMD), which protrudes from the omphalocele sac caudally, measures 1.5 cm in length and 1.5 cm in diameter (A). The duct contains two narrow mucosal openings with meconium output (B). This diagram over photo of the anatomy of the hernia of the cord (HOC) and POMD is based on the patient's fistulogram findings (A1).

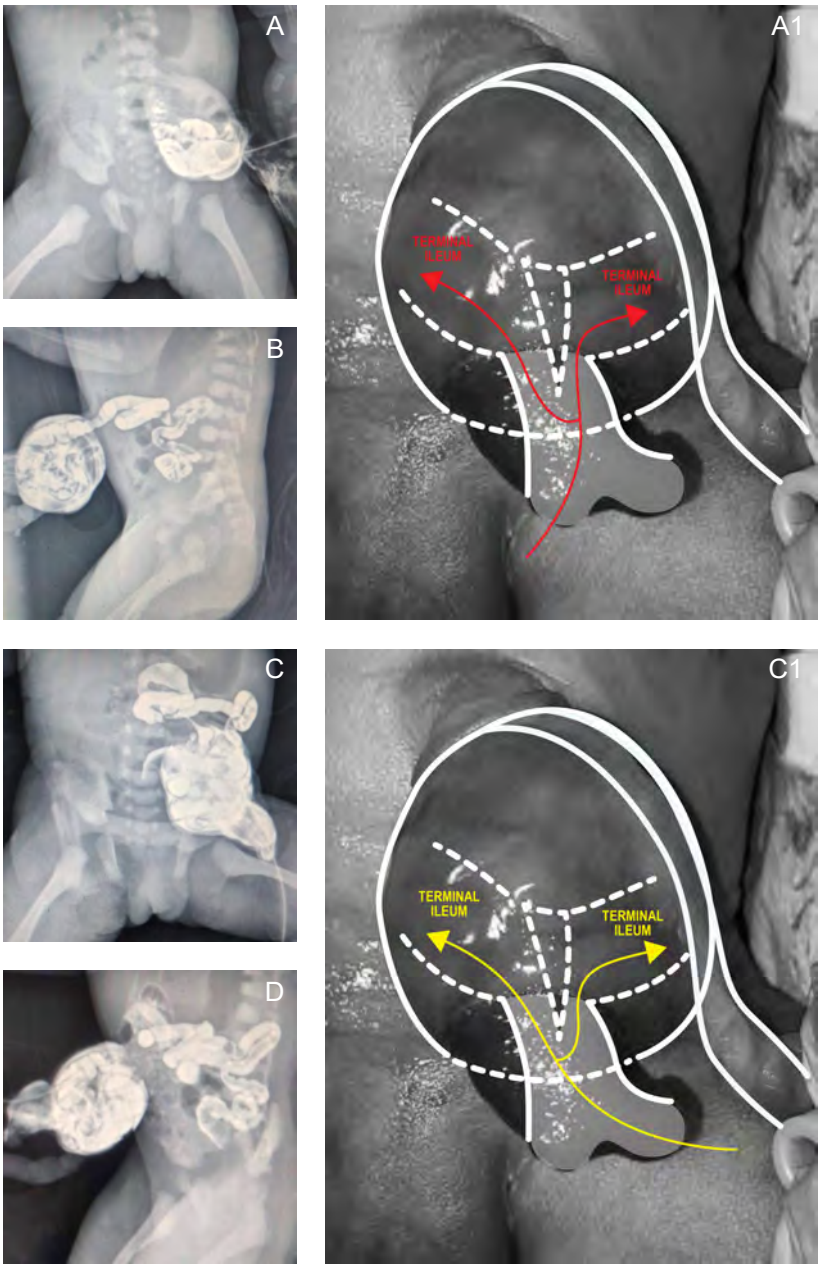

Figure 2 Fistulogram AP (A & C) and lateral (B & D) views and diagrams over photos (A1 &C1), showing continuity of the two mucosal openings with the lumen of the terminal ileum. When a water-soluble contrast medium was injected into the opening on the left, the dye filled the terminal ileum within the abdomen (A, A1 & B). Further, when contrast was injected into the opening on the right, the dye flowed into the same terminal ileum (C, C1, & D). The diagrams suggest that the two passages of the mucosal openings share a common lumen, possibly within the POMD.

mucosal outpouching, measuring 1.5 cm in length and 1cm in diameter, protruded from the caudal aspect of the round mass. The outpouching had a Y-shaped bifurcation, and each of the two tips had a mucosal opening with meconium discharge (Figure 1). The abdomen was soft, not distended, and not tender. The rest of the physical examination findings were unremarkable.

We managed the patient as having HOC with POMD and IP. The umbilical defect with bowel loops inside a sac is consistent with the clinical features of HOC. The tubular outpouching denotes the presence of a POMD. The protruding mucosal lining of the tube indicates that ileal prolapse through the POMD has occurred, and the meconium discharge from the two mucosal openings suggest bowel continuity.

Patient A

DIAGNOSTIC APPROACHES

We did a fistulogram, which revealed that the two mucosal openings shared a common lumen at the level of the POMD and were both continuous with the lumen of the terminal ileum within the abdomen (Figure 2). These findings support our initial impression of a POMD with complete IP, presenting clinically as a single-tubular mucosal protrusion with a Y-shaped bifurcation. Mechanical ventilation allowed temporary stabilization of the patient, however, subsequent arterial blood gas values pointed to respiratory acidosis. Initial babygram revealed reticular densities in both inner lung zones, suggestive of respiratory distress syndrome, probably secondary to surfactant deficiency disease. The babygram also revealed increased soft tissue density and bowel loops within the left lower hemiabdomen, corresponding to the HOC seen clinically, and the absence of rectal vault gas. Only 10 bilateral ribs could be appreciated, and there were suspicious vertebral body anomalies at T11, T12, and L1 found in the babygram. Another babygram done on the 9th day of life revealed hazy densities in both inner to middle lung zones—supporting the previous impression of a surfactant deficiency disease—a magnified heart, and a paucity of rectal vault gas. To rule out concomitant cardiac anomalies associated with 47% of omphalocele defects, a 2D echo was requested,[1] which revealed a 0.31-cm patent foramen ovale with left to right shunting, a large (0.5 to 0.6 cm) perimembranous type ventricular septal defect with left to right shunting (maximum gradient of 20 mmHg), and enlargement of the left atrium and left ventricle. The patient tested negative for SARS-CoV-2 infection by routine RT-PCR at birth. On the patient's 3rd day of life, complete blood count revealed a hemoglobin count of 175 g/L, leukocyte count of 11.62 x10^3/uL, and thrombocytopenia. Blood

chemistry revealed hypocalcemia and hyperkalemia.

Patient A

THERAPEUTIC APPROACHES

We immediately prepared the patient for an umbilical exploration, reduction of IP, resection of the POMD up to its base within the ileum and anastomotic repair of the remaining ileum, repair of the HOC, and umbilicoplasty. However, the Neonatology Service did not clear the patient for the planned procedures within the succeeding days because the patient had recurrent desaturations, thrombocytopenia, and metabolic abnormalities.

We adjusted the mechanical ventilator settings to reduce desaturation episodes. We also modified the components and rate of the intravenous (IV) fluids to correct the electrolyte imbalance. Noting the thrombocytopenia and recurrent desaturations, we considered sepsis, requested a blood culture study, and initially started the patient on ampicillin and gentamicin. To cover for possible Pseudomonas infection, the Neonatology Service decided to shift the antibiotics to cefotaxime and amikacin the following day. As prophylaxis for invasive candidiasis infection among high-risk patients at the neonatal intensive care unit (NICU), we started the patient on fluconazole on the 2nd day of life. On the same day, since the patient showed no clinical improvement, cefotaxime was shifted to meropenem. To correct thrombocytopenia, we transfused platelet concentrate and fresh frozen plasma. On the 8th day of life, the patient's blood culture studies revealed a growth of Serratia marcescens, so we revised our antibiotic coverage again by discontinuing vancomycin and starting colistin, ciprofloxacin, and linezolid.

We kept the HOC sac on daily dressing using sponges moistened with plain normal saline solution to prevent dessication. For stronger prophylaxis against local pathogen infection and fungal invasion in the HOC sac, we started the patient on vancomycin on the 4th day of life and later shifted fluconazole to amphotericin B.

Patient A

OUTCOME

Patient A had been hypoxic since birth, the respiratory distress worsened despite mechanical ventilation adjustments, the sepsis progressed, and the patient's clinical status deteriorated despite antibiotics, platelet transfusion, and IV fluid regulation. Patient A was unable to achieve a clinically-stable state for the contemplated procedures and succumbed to respiratory distress and sepsis on her 12th day of life.

Patient B

CLINICAL FEATURES

Patient B, a female neonate, was born with an umbilical mass and was immediately referred to the Pediatric Surgery Service for co-management. The patient was delivered preterm via normal spontaneous delivery in our institution, with APGAR scores of 8, 9, 9, and 9 at 1, 5, and 10 minutes, respectively, and a Ballard score of 33 weeks.

The patient's mother was a 23-year-old primigravid, with adequate antenatal care, no maternal illnesses, no trauma, and no exposure to harmful substances during her pregnancy. A prenatal ultrasonography done at 30 weeks AOG did not reveal any remarkable findings. The mother tested negative for SARS-CoV-2 infection by routine RT-PCR just before delivery. The patient's family history was unremarkable.

On physical examination, the patient had a heart rate of 126 beats per minute, a respiratory rate of 56 cycles per minute, and a temperature of 36.4°C. The patient weighed 1.92 kg, measured 44 cm in length, and had a head circumference of 28 cm and a chest circumference of 29 cm, all AGA. We did not hear any cardiac murmurs, and breath sounds were clear.

A 3x3x3 cm mass protruded from a 1.5-cm-diameter base on the umbilicus. Most of the mass was covered by a translucent sac, through which part of the umbilical cord and several bowel loops could be seen. There were two 1-cm-diameter mucosal openings, with meconium output, protruding from the superior aspect of the mass. The abdomen was soft, not distended, and not tender. The rest of the physical examination findings were unremarkable (Figure 3).

Our initial diagnosis for the patient's condition was HOC with POMD and IP. The umbilical defect with bowel loops encased in a sac is consistent with HOC. Meconium discharge from the two openings suggests bowel continuity and the presence of a POMD. The mucosal protrusions indicate IP through the POMD.

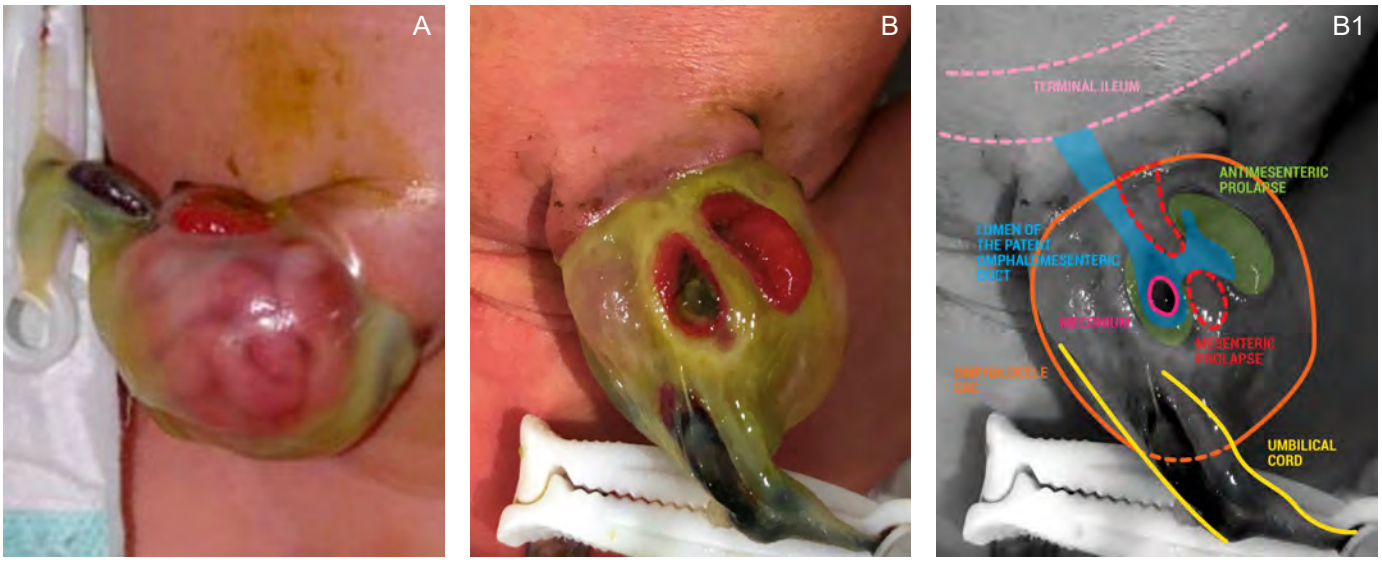

Figure 3 A 3x3x3 cm omphalocele sac protruding from a 1.5-cm-diameter base. The sac contains part of the umbilical cord and several bowel loops (A). Two mucosal openings, measuring 1 cm in diameter, with meconium output are seen protruding from the sac, superiorly (B). This diagram over photo of the anatomy of the HOC and POMD is based on the patient's fistulogram findings (B1).

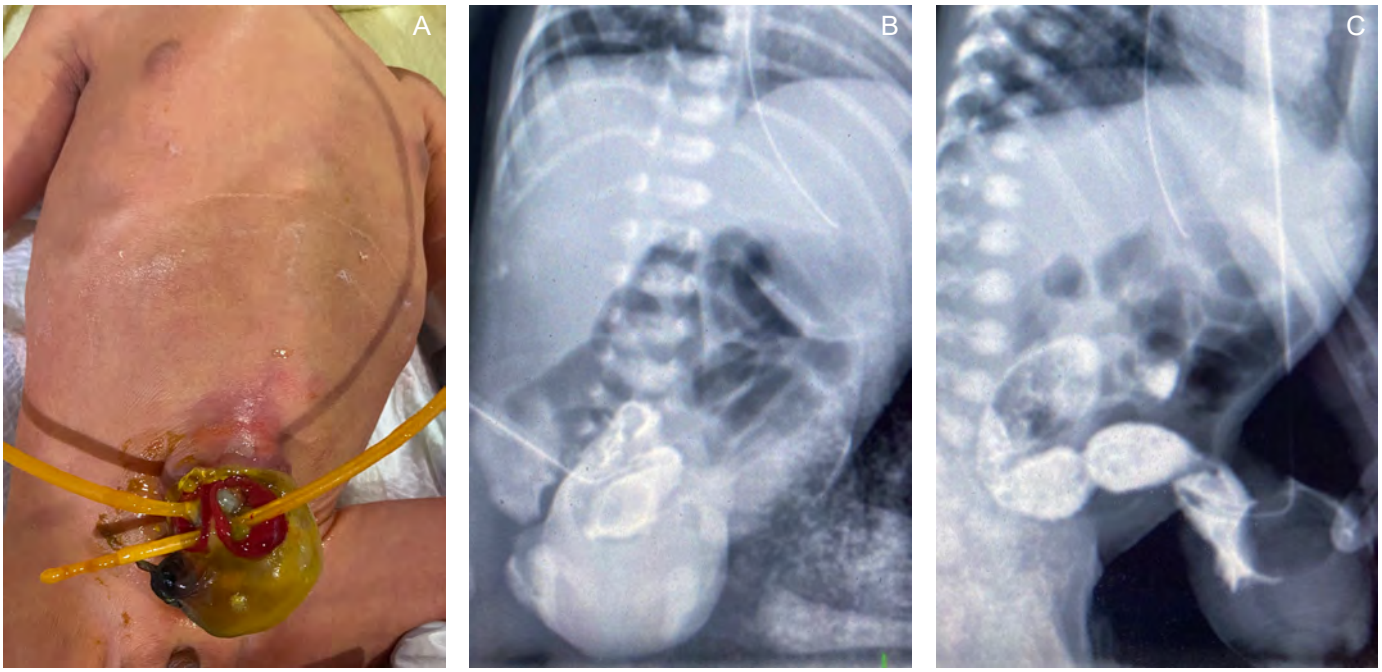

Figure 4 Cannulation of the mucosal openings that protrude from the omphalocele sac, showing a catheter inserted into the left opening. The catheter inserted into the right opening exits through the left opening, demonstrating a common lumen for the two openings within the POMD. The fistulogram, in AP (B) and lateral (C) views, shows continuity of the common lumen with the terminal ileum.

Patient B

DIAGNOSTIC APPROACHES

We did a fistulogram, which revealed that the two mucosal openings share a common lumen within the POMD, and that the common lumen is continuous with the terminal ileum within the abdomen (Figure 4). The initial babygram done during the first 6 hours of life showed shadows in the lower abdomen that correspond to the HOC, as well as the absence of rectal vault gas. To rule out other congenital anomalies, we requested a 2D echo, which revealed normal findings.The patient tested negative for SARS-CoV-2 infection by routine RT-PCR done in the NICU upon admission. Complete blood count series taken on the 2nd, 3rd, 7th, 10th and 11th days of life

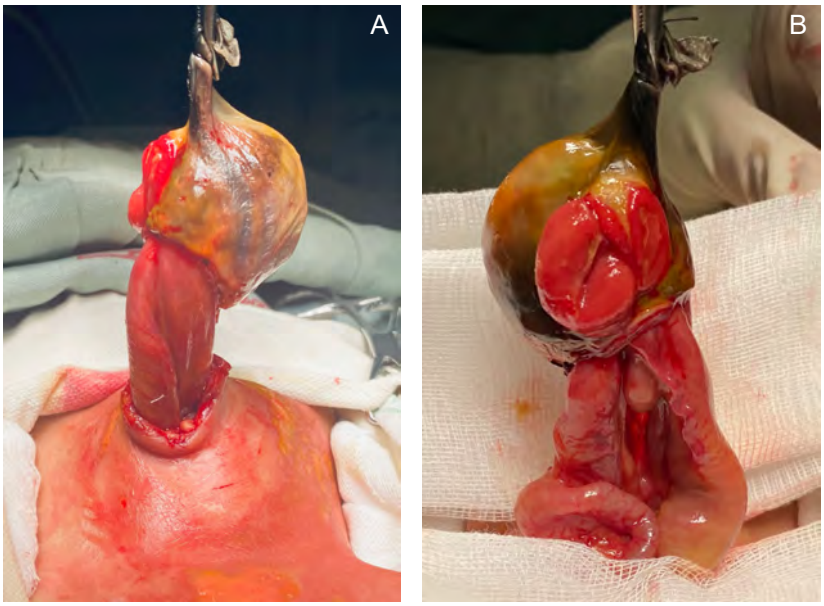

Figure 5 Intraoperative findings showing an omphalocele sac, dissected circumferentially at its base (A). This reveals a 1-cm-diameter umbilical defect consistent with HOC, and also shows a 2-cm-wide POMD with its antimesenteric border prolapsing to the outermost mucosal opening, thus dividing the POMD lumen into two 1-cm-wide external mucosal openings (B).

revealed an increasing trend in leukocyte count from 18.80 x10^3/uL to 27.72 x10^3/uL. Because of this trend, we considered a possible neonatal infection for Patient B.

Patient B

THERAPEUTIC APPROACHES

We requested for a blood culture study and initially started the patient on ampicillin and gentamicin. To cover for possible anaerobic infection, we started metronidazole on the patient's 2nd day of life. We kept the HOC sac moist using sponges soaked in plain normal saline solution. On the patient's 5th day of life, however, we noted a greenish discoloration on the surface of the HOC sac. We then decided to shift ampicillin to meropenem, and gentamicin to amikacin, to target possible *Pseudomonas* infection.

On the patient's 7th day of life, we performed umbilical exploration, reduction of IP, resection of POMD up to its base with anastomotic repair to its ileal origin, repair of the HOC, and umbilicoplasty. Intraoperatively, we released the HOC sac from its attachment in the umbilicus. We noted a 1-cm-wide fascial defect and a POMD that was attached to the sac. The antimesenteric border of the POMD prolapsed to the outermost mucosal opening, creating two 1-cm external mucosal openings that share the same lumen internally (Figure 5). This denotes a partial IP through the POMD. We extended the

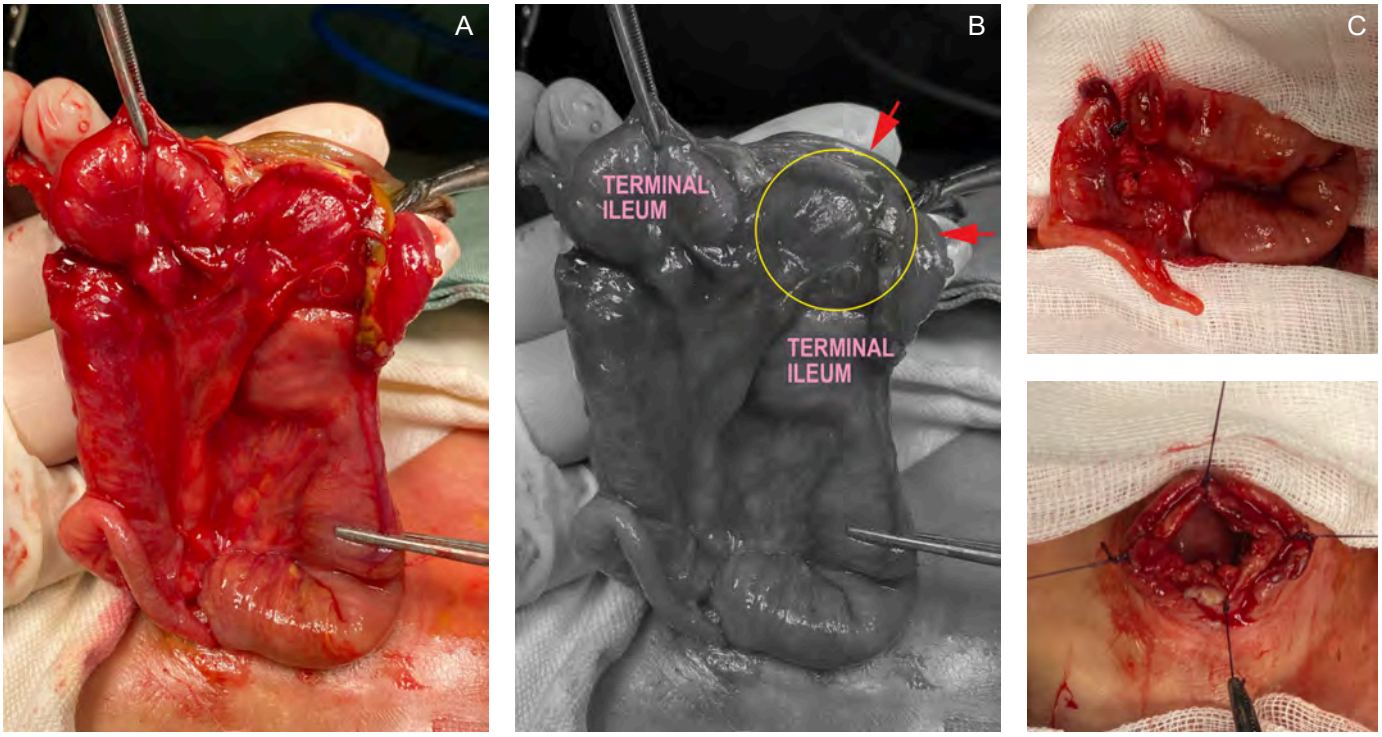

Figure 6 Excised omphalocele sac containing the herniated terminal ileal segments (A). The base of the POMD is attached to the terminal ileum. The shared lumen within the POMD (A1: yellow ring) ends with two mucosal openings at the tip of the POMD (A1: red arrows). After resection of the POMD, the remaining herniated terminal ileal segments (B) were anastomosed and reduced through the umbilical defect (C).

fascial opening laterally to 2 cm in order to exteriorize the underlying small bowel loops. As a result, we were able to fully visualize the prolapsed POMD, with its base in the terminal ileum and its location at 6 cm from the ileocecal valve. We then resected the part of the terminal ileum with the POMD, with a 1-cm margin on each side of the POMD base, and performed an end-to-end anastomosis of the ileal segments using Vicryl 4-0 sutures. We restored the anastomosed bowel intra-abdominally and closed the umbilical fascial defect using interrupted Vicryl 3-0 sutures (Figure 6). Finally, we performed an umbilicoplasty using subcuticular purse-string sutures using Monocryl 4-0 (Figure 7). The entire procedure was completed in 1.5 hours.

Patient B

OUTCOME

We initiated orogastric tube feeding after noting bowel movement and rectal output on the 4th postoperative day. We also started the patient on fluconazole as prophylaxis for invasive candidiasis among high-risk patients at the NICU. Cultures of blood, endotracheal aspirate, and eye discharge specimens taken at different times postoperatively showed growth of *Serratia marcescens, Pseudomonas aeruginosa,* and *Acinetobacter baumannii*, respectively. For better prevention of local pathogen infection on the incision site, we shifted amikacin to vancomycin and started ciprofloxacin on the 5th postoperative day.

On the 15th postoperative day, we noted that the patient had abdominal distention and absent bowel sounds upon auscultation. A series of postoperative babygrams, an abdominal X-ray with contrast study, and an abdominal ultrasound have all revealed diffuse bowel dilatation with no signs of mechanical bowel obstruction. Complete blood count revealed leukocytosis and thrombocytopenia, and blood chemistry revealed hypoalbuminemia, elevated blood urea nitrogen, elevated creatinine, and hyperkalemia. Based on the patient's increasing leukocyte count and thrombocytopenia, as well as the culture results indicating the presence of multiple opportunistic pathogens, we considered sepsis. Despite the antibiotics started earlier, babygram on the 23rd postoperative day showed bilateral pneumonia. We revised the antibiotic coverage on the 25th postoperative day, when endotracheal aspirate and eye discharge culture studies revealed *P. aeruginosa* and *A. baumannii*, respectively. We shifted ciprofloxacin and meropenem to cefepime and started colistin, shifted fluconazole to amphotericin B, and continued metronidazole.

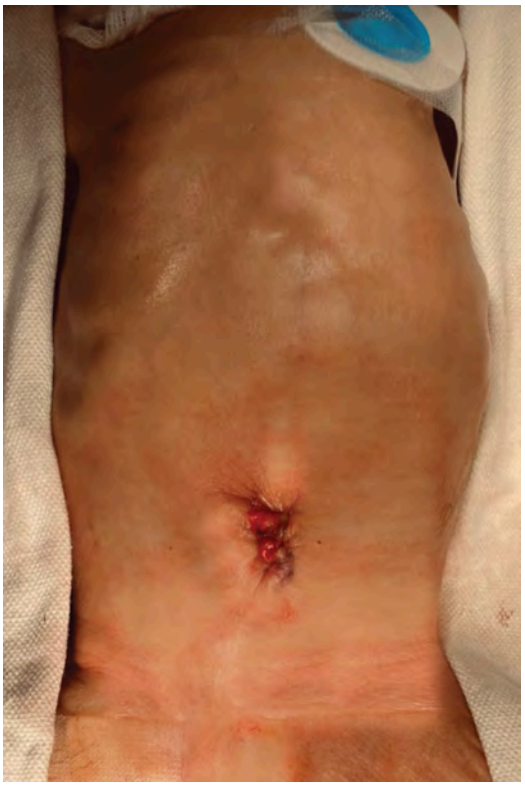

Figure 7 Completed abdominal wall closure and umbilicoplasty, using subcuticular purse-string Monocryl 4-0 sutures to reconstruct the umbilicus.

On the 27th postoperative day, the patient had regressing abdominal distention, but began to have episodes of bradycardia and desaturation, which required endotracheal intubation and mechanical ventilation. On the succeeding days, the patient's respiratory status worsened despite aggressive treatment. On the 30th postoperative day, the patient had cardiopulmonary arrest and subsequently died.

DISCUSSION

We diagnosed two patients with HOC with POMD, one with complete and the other with partial IP, and prepared each of them for a single surgical procedure to repair all component anomalies. The patient who had complete IP developed respiratory failure and sepsis, had not reached optimum clinical state for the contemplated procedure, and

subsequently died before surgery could be done. The other patient underwent corrective surgery successfully, but succumbed to progressing neonatal sepsis.

The simultaneous occurrence of HOC, POMD, and IP is very rare.[2] At present, very little is known about the pathophysiology, severity, management, and outcomes of these umbilical defects from the medical literature. The etiologies of HOC and POMD are commonly congenital, while an IP in a POMD is acquired. The prolapse results from an increase in intra-abdominal pressure after birth due to a congenital pulmonary pathology or a neonatal pulmonary infection. Suggested risk factors of IP include short distance of the POMD to the ileocecum, short POMD length, and wide-mouthed POMD base.[3] Remarkably, a POMD with IP usually presents with two mucosal openings sharing a common lumen towards its ileal origin, which is distinguishable from the single mucosal opening of a POMD without prolapse.[5 6] Maternal hyperthyroidism and its medical treatment (carbimazole and methimazole) have been separately associated with HOC and POMD.[5 6] However, risk factors for the simultaneous occurrence of HOC, POMD, and IP in a single patient have not yet been identified.

Both our patients and their mothers tested negative for SARS-CoV-2 during admission, and both mothers denied any previous illnesses suggestive of COVID-19. Based on these, there is a very low probability that COVID-19 caused the occurrence of the congenital umbilical defects in our patients. Even if both mothers had asymptomatic COVID-19 during gestation, there is presently no definitive evidence of a link between gestational COVID-19 and congenital anomalies.[7]

Clinical outcomes of combined HOC, POMD, and IP are highly dependent on the time of presentation, prompt recognition of diagnosis, presence of complications (prolapse, strangulation), and associated anomalies.[4] A review of multiple case reports from 1952 to 2019, with a total of 9 reported cases only, presented a 56% mortality within the first month of life. Among patients with the combined conditions, postoperative cause of death is usually anastomotic breakdown and leak.[8]

We were able to immediately recognize and diagnose the clinical conditions of our two patients. Neonatal infection got in the way of prompt surgery in one patient. While we were able to successfully carry out the proper surgical correction for HOC with POMD and partial IP on the other patient, bloodstream and pulmonary infections developed postoperatively. For both our patients, favorable clinical outcomes could have possibly been achieved with a more aggressive neonatal infection management.

In summary, we diagnosed two female neonates with combined HOC, POMD, and IP. One patient did not achieve a clinically-stable state for any corrective surgery and eventually died of respiratory distress and sepsis. The other patient had a successful corrective surgery for the clinical conditions, but eventually died of sepsis. HOC with POMD and IP can be corrected surgically, but infection and other comorbidities can influence the timing and success of the surgical correction, as well as the overall outcome. Apart from surgical correction, aggressive perioperative infection control and management should be ensured in the therapeutic approach to patients with this combination of clinical conditions.

Contributors
KPR, LLL, NMA, NFB and JSMJ contributed to the diagnostic and therapeutic care of the patient in this report. All of them acquired relevant patient data, and searched for and reviewed relevant medical literature used in this report. KPR wrote the original draft, performed the subsequent revisions. All approved the final version, and agreed to be accountable for all aspects of this report.

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Patient consent
Obtained

Reporting guideline used
CARE Checklist
(http://www.care-statement.org/downloads/CAREchecklist-English.pdf)

Article source
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None declared

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