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Case Report

Perforated Meckel's diverticulum with congenital diaphragmatic hernia

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ABSTRACT

Congenital diaphragmatic hernia (CDH) is a congenital anomaly in which there is herniation of abdominal contents into the thoracic cavity, this leads to pulmonary hypertension and pulmonary hypoplasia to a varying degree. Association of CDH with various extra-diaphragmatic malformations is known. We present a case of a CDH, in which the patient developed perforation of Meckel's diverticulum (MD) post-operatively. This is the first reported case of a patient with CDH developing perforation of a MD.

Keywords: Congenital diaphragmatic hernia, Perforated Meckel's diverticulum, Neonate, Neonatal surgery

INTRODUCTION

Congenital diaphragmatic hernia (CDH) has an incidence of about 1 in 2000-3000 new-borns.[1,2]

The most common associated extra-diaphragmatic malformation is cardiac-constituting around 63%. Others include renal, genital anomalies, neural tube defects, and chromosomal abnormalities.[2,3]

The incidence of MD is around 2% in the general population.[4]

There have been reports of MD being present in CDH patients. Perforated MD has also been reported in a limited number of neonates.[5] However, there have been no reported cases of a perforated MD in a patient with CDH.

CASE REPORT

The patient was a male born at 38 weeks of gestation through a normal vaginal delivery, with a birth weight of 3690 grams. Umbilical cord was found to be twisted twice around the neck. APGAR score of 9 and 8 was noted at 1 and 5 minutes. He was not maintaining adequate oxygen saturation, had chest retractions and received oxygen initially via nasal prongs followed by mechanical ventilation. Chest X ray revealed a left sided CDH. The patient was initially unstable and required high frequency oscillatory ventilation. After a period of stabilisation, he was taken for surgery.

He underwent laparotomy and repair of left CDH under general anaesthesia. Most of the small bowel, right colon, transverse colon and spleen were in the left side of the thoracic cavity. The contents were reduced. The diaphragmatic hernia was primarily repaired with interrupted Mersilk-0 sutures. Patient was shifted back to the neonatal intensive care unit in a stable condition.

After a period of conventional ventilation, patient developed desaturation and requited to be shifted back to high frequency oscillatory ventilation. He was noted to have free air in the abdomen on abdominal X-ray for which a drain was placed in the right lower quadrant, which drained clear fluid initially, followed by bile the following day.

Patient's condition stabilized and shifted back to conventional ventilation; he underwent exploratory laparotomy. There was eextensive meconium and bile contamination of the peritoneal cavity. A perforated MD 35 cm from ileo-cecal junction was noted. Rest of bowel was edematous but otherwise healthy. Stomach was healthy. Malrotation of the bowel was noted, duodenum was straight and Duodeno-Jejunal flexure was located on the right side of midline. The perforated MD was resected including an adjacent 2 cm of ileum on either side with end-to-end ileo-ileal anastomosis with interrupted non-absorbable sutures. An appendicectomy was performed. Abdomen was closed without any tension. A right internal jugular vein central line was inserted.

Histopathology of the resected MD specimen showed perforation and transmural neutrophilic inflammation with submucosa showing edema and congestion. Serosa showed mixed inflammatory infiltrates. There was no gastric/pancreatic tissue noted and no atypia/malignancy.

The patient continued to be on mechanical ventilation postoperatively with bile drainage from the nasogastric tube-for which an upper GI contrast study was done on post-operative day 20 (from the second surgery).

The upper GI contrast study showed intact anastomotic site with no leak and no features of bowel obstruction, with prolonged transit time. He developed fungal sepsis and was started on intravenous fluconazole and Amphotericin-B. He developed right pneumothorax for which intercostal drain was inserted. He continued to have sepsis for an extended period and succumbed to the prolonged sepsis.

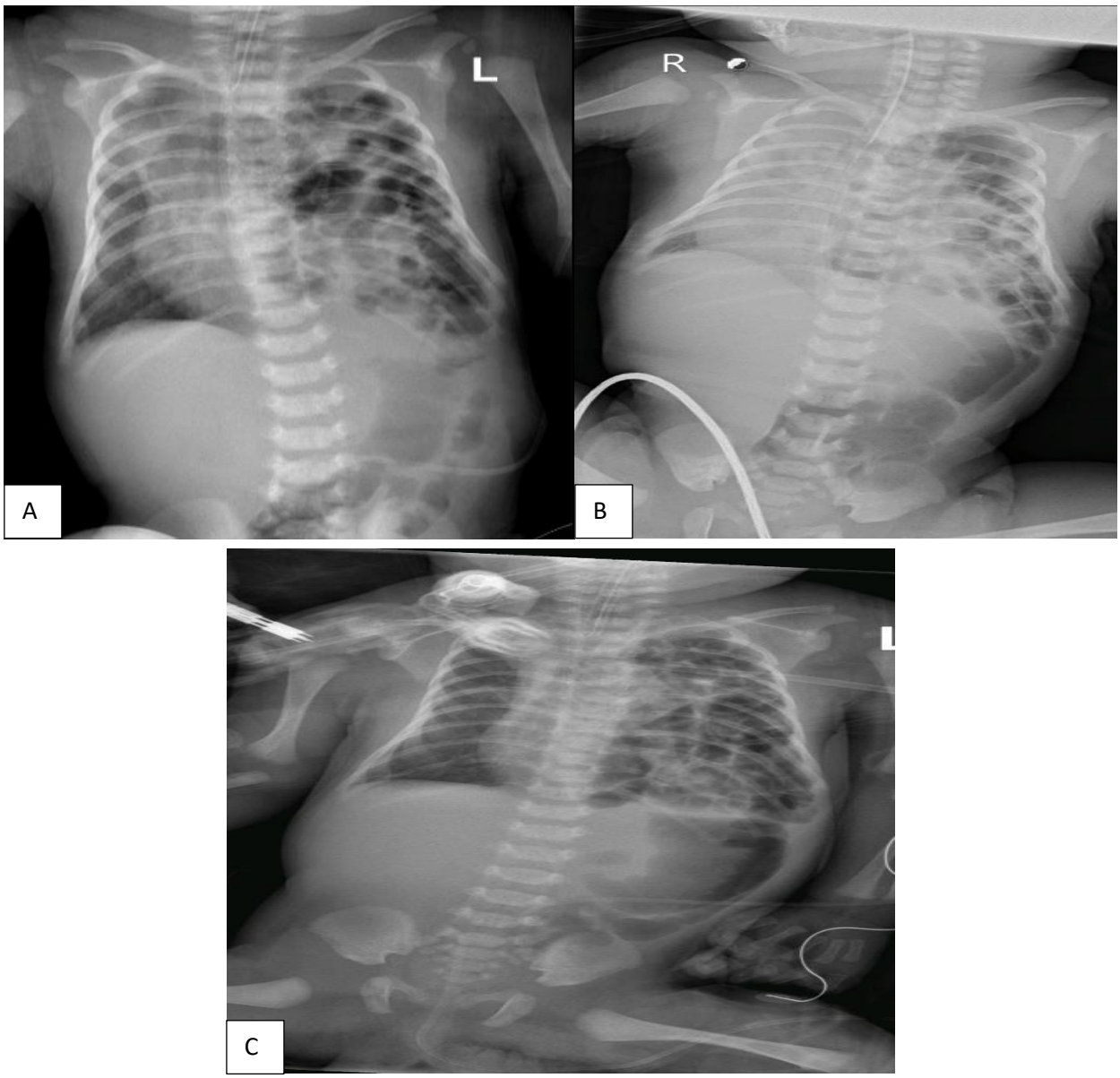

Figure 1 (A-C): Pre-operative x-rays of the patient showing bowel loops in the left side of the thoracic cavity with right sided mediastinal shift. The stomach is seen within the abdominal cavity.

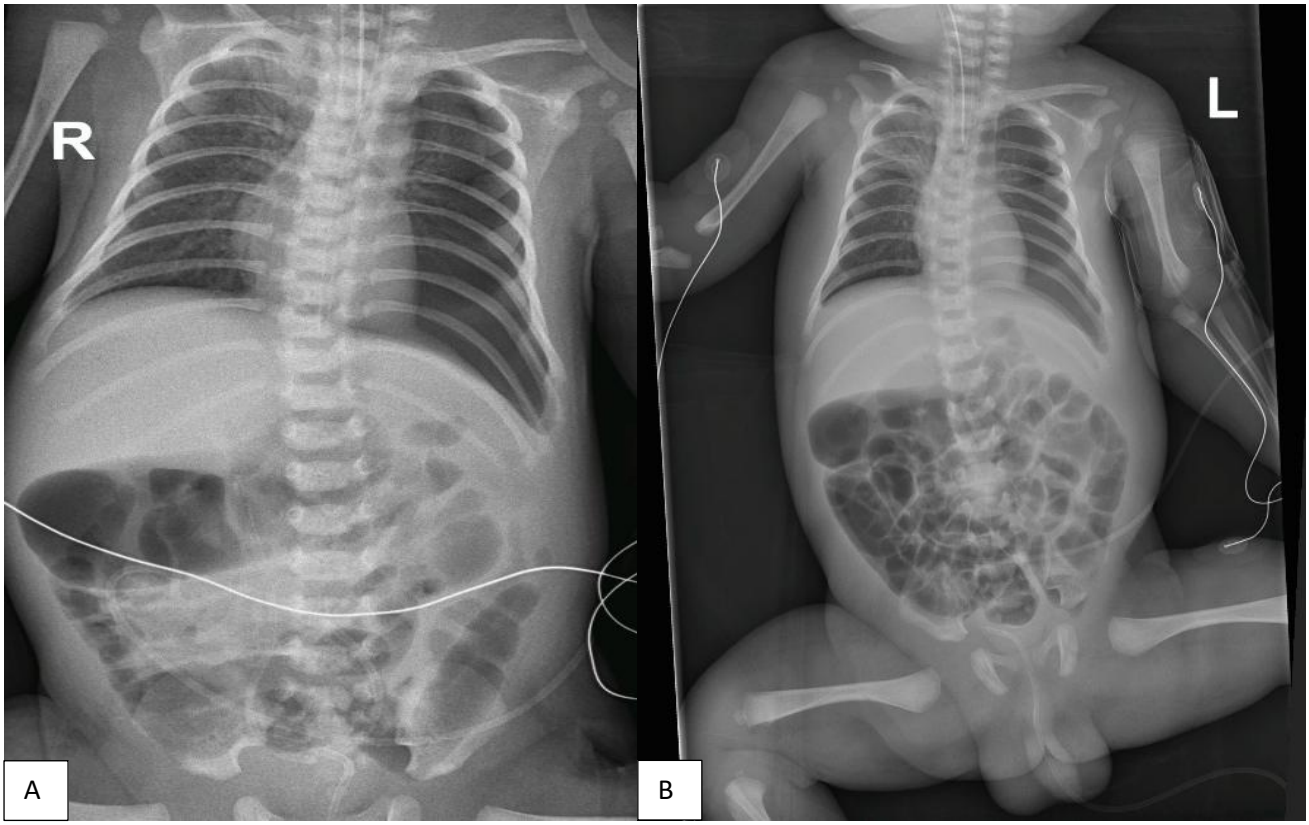

Figure 2 (A and B): Post-operative X-rays following the left CDH repair showing the left diaphragm intact and abdominal contents within the abdominal cavity.

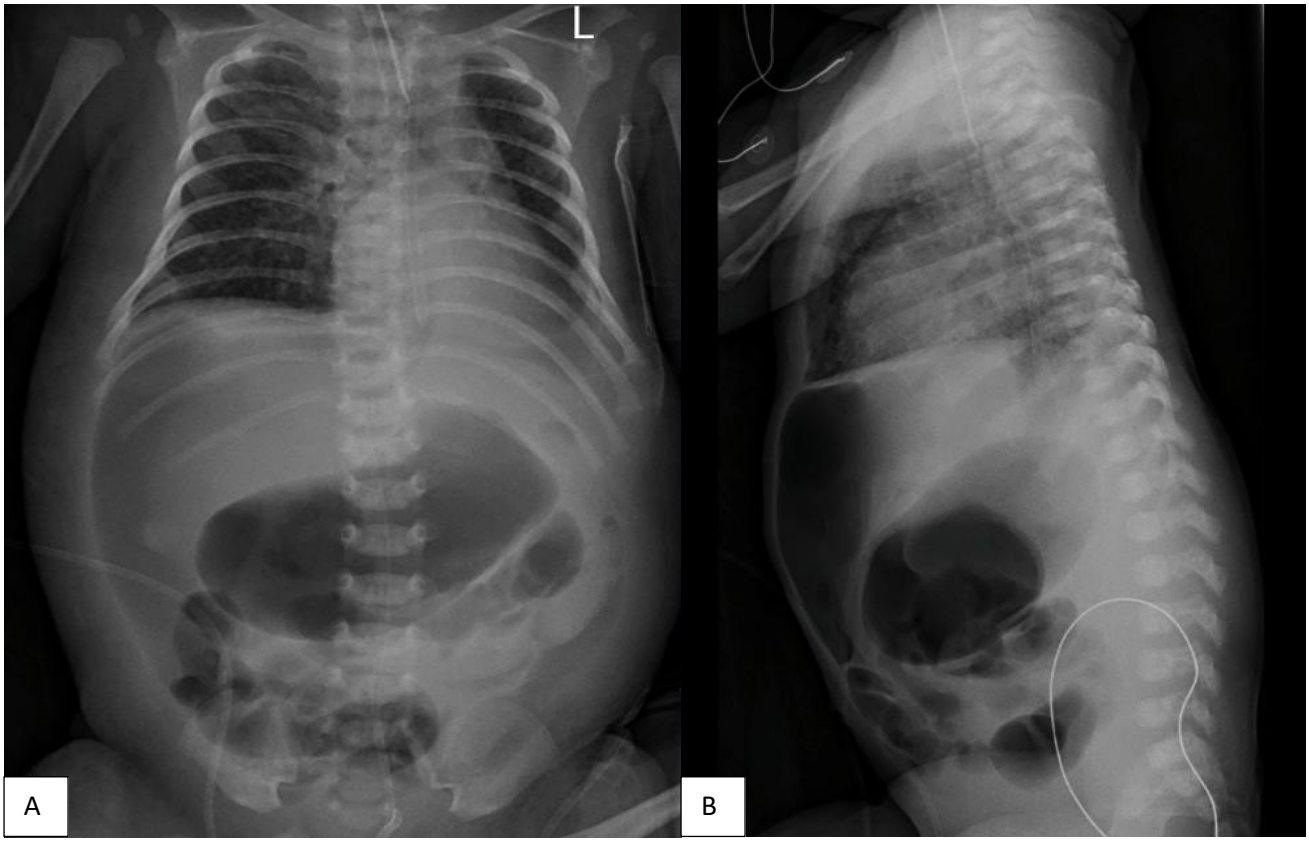

Figure 3 (A and B): Free air within the abdomen noted on post-operative day 5.

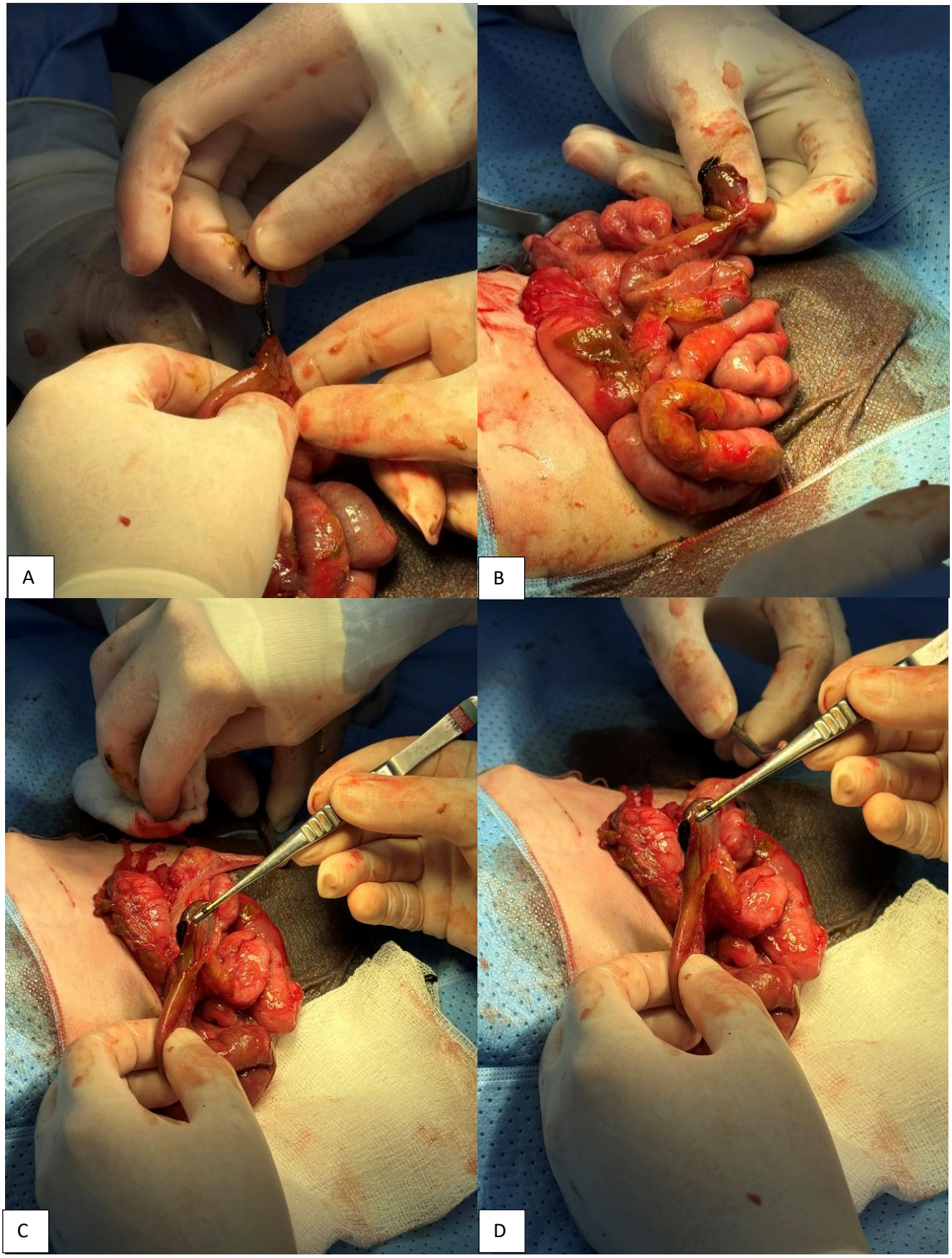

Figure 4 (A-D): Intra-operative images of the perforated MD.

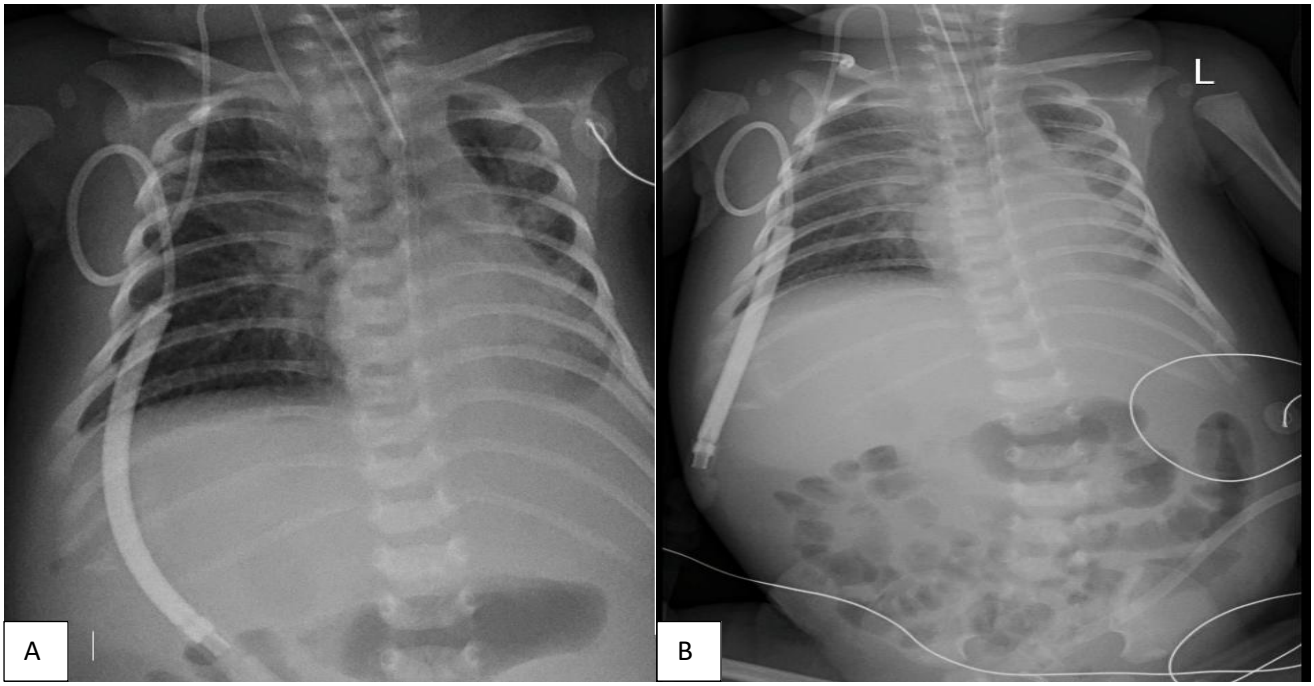

Figure 5 (A and B): Post-operative X-rays following the resection-anastomosis of perforated MD.

DISCUSSION

MD-remnant of the vitello-intestinal duct, occurs due to its incomplete obliteration at 7 weeks of gestation.[6] Its incidence is about 2% of the population, with a complication rate of around 4%.[4,7,8] Most of the patients with MD are asymptomatic.[9-11]

Symptomatic MD can present in the form of bleeding, diverticulitis, volvulus, intussusception or perforation.[9] The perforation is postulated to be a result of heterotropic mucosa or diverticulitis leading to erosion of its wall with resultant perforation.[4,9,12,13]

It has also been proposed that the perforation could be the result of separation of vitelline remnants from the abdominal wall.[14]

The histopathological analysis of the resected perforated MD in our patient showed transmural neutrophilic inflammation with the submucosa showing edema and congestion, serosa showing mixed inflammatory infiltrates, and without any heterotropic gastric or pancreatic mucosa.

Pneumoperitoneum as a presenting feature of symptomatic MD patients is said to be around 33.3%.[15]

CDH has been reported to have incidentally detected MD.[16,17] Mandhan et al reported a case of CDH with incidental MD and heterotropic pancreatic mucosa noted in the jejunum.[16] However, to the best of our knowledge, this is the first reported case of a CDH patient having developed a perforation in the MD in the immediate post-operative period.

There are reports of perforated MD in the neonatal age-with around 59 cases being reported 3027 manuscripts, and none of the perforated MD was in a case of CDH.[5] Liaqat et al reported 6 cases of perforated MD in neonates and performed a systematic review of literature which showed 84.7% of patients presenting in the first week of life.[5] In our case, the perforation occurred in the second week.

Treatment options of a perforated MD include surgical resection, MDtomy or creation of a stoma, depending on various factors such as the overall general condition, severity of peritoneal contamination and hemodynamic stability of the patient, and the condition of the adjacent bowel.[5,9,18] Our patient was hemodynamically stable with the rest of the bowel being healthy, and hence, resection of the MD with ileo-ileal anastomosis was undertaken.

The treatment of incidentally detected MD is controversial with morbidity being lower in patients who are asymptomatic compared to those with symptomatic MD.[18-21]

This is even more controversial in a patient undergoing repair of a CDH leaving the surgeon with a dilemma. It remains a topic of discussion and the risks versus benefits of resecting the MD, and converting a clean surgery into a clean-contaminated surgery, need to be considered.

CONCLUSION

Our patient is the first reported case of a perforated MD in a patient with CDH, who presented early in the post-operative period. The management of a perforated MD is resection-anastomosis, Meckel's diverticulectomy or

stoma creation. The management of incidental MD, especially in patients of CDH remains highly controversial.

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