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Case Report

Surgery Section

Thoracic Occupation of All Abdominal Organs in a Neonate: Intraoperative Surprise of Diaphragmatic Eventration

SOMYA GOEL[1], THAVENDRA DIHARE[2], RAJU K SHINDE[3], KHUSBU VAIDYA[4], ASHISH JIVANI[5]

ABSTRACT

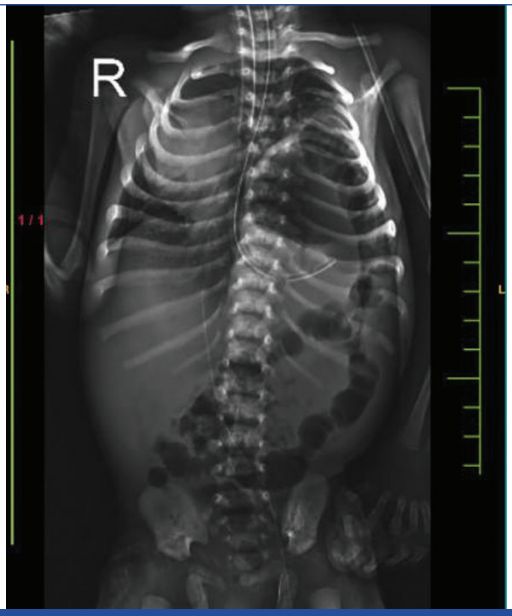
Congenital Diaphragmatic Hernia (CDH) is commonly diagnosed at birth or in the early days of life. It is commonly observed on the left side, with a male predominance. It can be diagnosed during prenatal scans using common diagnostic modalities such as ultrasound. Diaphragmatic eventration is an abnormal elevation of the diaphragm caused by muscle paralysis, thinning, and/or weakness without any disruption or continuity in the diaphragm. Severe congenital diaphragmatic eventration can mimic CDH and present a diagnostic challenge. Foetal Magnetic Resonance Imaging (MRI) can help differentiate congenital diaphragmatic eventration from CDH. This is a case of a nine-day-old male neonate, presented to the emergency department with respiratory distress, and was subjected to radiological investigations, which showed left-sided CDH, Bochdalek type. Intraoperatively, a large diaphragmatic eventration was found. The neonate underwent a successful Bochdalek congenital diaphragmatic repair and was discharged on postoperative day 30. A follow-up at 1.5 months showed the infant recovering well postoperatively with no new complications. This case highlights the imaging-operative discordance. This neonate with acute respiratory distress, imaging suggestive of Bochdalek CDH, and an intraoperative finding of extensive eventration (rather than a full-thickness posterolateral defect) is rarely detailed. The successful management of this case was evident by an uneventful postoperative recovery.

Keywords: Bochdaleck type hernia, Congenital conditions, Hernia in paediatric patients, Late diagnosis, Respiratory distress

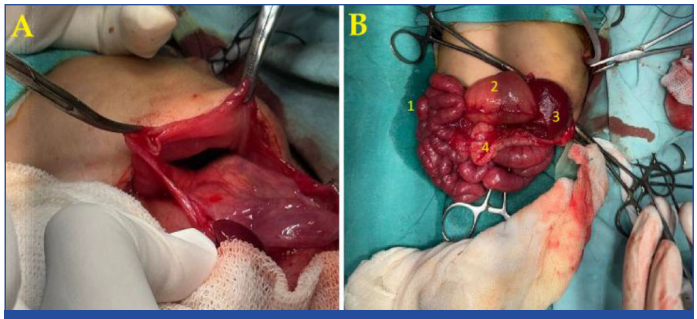
CASE REPORT

A neonate (9 days) with 37 weeks of gestation was delivered via vaginal delivery to a primigravida mother. The neonate cried immediately after birth, and there were no signs of respiratory distress. The rest of the medical history was non-significant. On the 9th Day Of Life (DOL), the child had a fever, with complaints of cough and difficulty breathing, for which the primary care centre referred the patient to our tertiary care centre. On DOL 10, radiological examination by X-ray showed an abnormal bowel gas pattern within the thoracic cavity, especially on the left side, displacing the lung margin. The diaphragmatic outline was poorly defined on the left side, and the abdomen was relatively gas-free. These findings were consistent with Congenital Diaphragmatic Hernia (CDH), likely a Bochdalek-type left-sided hernia [Table/Fig-1].

The child was further subjected to a High-Resolution Computed Tomography (HRCT) scan of the thorax, which showed a left-sided diaphragmatic hernial defect measuring 2 cm, with herniation of splenic parenchyma and bowel loops, an elevated left hemidiaphragm, and a mediastinal shift towards the right. Left lung collapse was noted, which was suggestive of CDH. The child was intubated and taken up for emergency diaphragmatic repair on DOL 13. The patient was induced under standard anaesthetic conditions. A 4-centimetre incision was made below the left coastal margin. Intraoperatively, it was found that major abdominal organs had eventrated via a large non-functioning sac formed by the diaphragm into the left thoracic cavity with no defect. Due to the sac's size, redundancy, and fragility, plication was not feasible; excision was required, thereby creating an iatrogenic defect in the diaphragm. Major abdominal organs were found to have eventrated into the thoracic cavity, such as the stomach, small bowel and entire large bowel, and the spleen, leaving only the liver and gall bladder in the abdomen [Table/Fig-2]. All the contents of eventration, such as the spleen, stomach, large and small bowel, were reduced, and the sac was excised. The left lung appeared normal, with no evidence of fibrosis. An Intercostal Drain (ICD) was placed in the left thoracic cavity for complete lung expansion. The defect was sutured with Ethibond 2-0 after repositioning of the abdominal contents. The closure was done in layers, and the procedure was uneventful [Table/Fig-3]. The patient was transferred to the Paediatric Intensive Care Unit (PICU) in an intubated state after haemodynamic parameters stabilised. A 4-centimetre incision was taken below the left costal margin, and a blunt dissection was carried out. The sac was excised, and primary repair was completed after abdominal contents (spleen, stomach, large and small bowel) were repositioned. The layers were closed in accordance with standard procedure. The patient was shifted to the pediatric ICU in an intubated state. The procedure was completed uneventfully, and a postoperative X-ray confirmed a successful procedure [Table/Fig-4].



[Table/Fig-1]: Chest X-ray showing abnormal bowel gas pattern within the thoracic cavity, especially on the left side, displacing the lung margin.



[Table/Fig-2]: Intraoperative image showing major abdominal organs were found to have eventrated into the thoracic cavity, such as the stomach, small bowel, and entire large bowel, spleen (1-small intestine; 2-stomach; 3-spleen; 4-transverse colon).

[Table/Fig-3]: Immediate postoperative image.

[Table/Fig-4]: Postoperative X-ray confirming successful procedure.

The baby developed persistent pulmonary hypertension on postoperative day 2, and further blood investigations were prompted, which showed Total Leukocyte Count (TLC) of 46000/microliter of blood. The patient was transfused with two units of packed red blood cells for management. A blood culture report was sent, which was suggestive of Klebsiella pneumoniae sensitive to colistin. Antibiotic Colistin was administered at 25000 IU/kg/day, divided into two doses, intravenously, for seven days, as per pediatric guidelines. A repeat blood test showed parameters within normal limits. The child was discharged after 15 days with stable vitals. The child was advised a follow-up at 1.5 months and Vitamin D3 (1 mL OD) till one year of age, with a regular vaccination schedule and exclusive breastfeeding until the first six months of life.

DISCUSSION

CDH arises from a developmental abnormality in the diaphragm, permitting the herniation of abdominal viscera into the thoracic cavity. Partial or complete diaphragmatic absence often leads to pulmonary hypoplasia. It is characterised by the protrusion of abdominal organs into the thoracic cavity, resulting in respiratory distress and fatal pulmonary hypertension [1,2]. Altered retinoid signalling has been implicated as a key etiological factor in CDH. Animal studies have shown that vitamin A deficiency in offspring increases the incidence of diaphragmatic defects, with herniation rates inversely proportional to vitamin A levels [3]. A prevalence of 2.6/10000 total live births has been reported with a mortality rate ranging from 33.8 to 45.1% [4]. Most of the cases of CDH are diagnosed in prenatal scans; if not, these neonates are born with acute clinical presentations with respiratory distress noted commonly [5]. However, cases are reporting the diagnosis later than one month of age. The diagnosis is often an incidental finding in asymptomatic infants. [4-6]. Late presentations have been observed across different age groups, including a 19-year-old [5].

Ultrasound can be used for the prenatal and postnatal diagnosis of CDH. However, advanced radiological modalities such as CT and MRI can be more helpful in differentiating the internal organs and the contents of the hernia [6,7]. Additionally, data parameters such as total lung volume and lung-to-head ratio are also being researched for their use as prognostic markers [2,7]. In this case, no prenatal scans were performed due to the patient's rural background. Also, there were no signs of distress at birth and the first clinical presentations were observed on 4th DOL initially as fever, cough and later by the findings of ultrasound and CT scans, which confirmed the diagnosis of CDH.

Diaphragmatic eventration refers to the elevation of an intact diaphragm resulting from the substitution of diaphragmatic muscle with fibroelastic tissue, either partly or entirely, constituting 5% of all diaphragmatic anomalies. It may be either congenital or acquired. This illness is exceedingly rare and commonly observed in neonates with clinical presentations such as cough and respiratory distress [8]. The contents of a CDH primarily determine the clinical implications for patients. Organs such as the stomach, spleen, liver, and kidneys are commonly noted as hernial contents in CDH of the Bochdalek type. The number and types of the organs can cause pulmonary dysplasia, respiratory distress, mediastinal shift, disrupted cardiac functionality and other gastrointestinal complications [9,10].

CDH can be managed by either laparoscopic surgery or open surgery. Cases with complicated presentations might require open surgery when laparoscopic management is not suitable [4]. Though most reported cases have used open surgical management with adhesiolysis, thoracotomy, diaphragmatic plication, restoration of anatomical structures, closure of the hernial defect, and other procedures as required [4-6].

A left-sided diaphragmatic defect is reported to be more common, with an incidence of 90% in the study group, compared to right-sided defects, which were also observed in this case [10]. Also, a male predominance is noted, with male-to-female ratios of 2:1 to 4:1 in the published literature [10,11]. A poor prognosis is associated with early diagnosis, attributed to the poor development of the lungs due to early displacement of the organs during pregnancy, as compared to those diagnosed at birth [11]. Though early and prompt diagnosis is crucial, attributed to the associated life-threatening complications.

CONCLUSION(S)

This case is a rare presentation of congenital diaphragmatic eventration in the early neonatal period, initially mimicking a left-

sided Bochdalek hernia on imaging. Prompt surgical intervention was crucial to the successful management of the condition. Despite postoperative complications such as pulmonary hypertension and sepsis due to Klebsiella pneumoniae, appropriate intensive care support and targeted antibiotic therapy led to complete recovery. This case highlights the importance of considering CDH as a differential diagnosis in early neonates.

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PARTICULARS OF CONTRIBUTORS:

1. Junior Resident, Department of Surgery, DMIHER, Sawangi, Wardha, Maharashtra, India.
2. Professor, Department of Surgery, DMIHER, Sawangi, Wardha, Maharashtra, India.
3. Professor and Head, Department of Surgery, DMIHER, Sawangi, Wardha, Maharashtra, India.
4. Junior Resident, Department of Surgery, DMIHER, Sawangi, Wardha, Maharashtra, India.
5. Junior Resident, Department of Surgery, DMIHER, Sawangi, Wardha, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Somya Goel,
Junior Resident, Department of Surgery, DMIHER, Sawangi, Wardha-442107, Maharashtra, India.
E-mail: somyagoel98@gmail.com

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