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Original Research Article

A retrospective study on the surgical management and outcome of congenital diaphragmatic hernia in neonates

Javaid Iqbal Gulshan[1]*, Sanjay Raina[2], A. Hamid Wani[1]

[1]Department of Surgery, [2]Department of Pediatric Surgery, Government Medical College, Jammu, Jammu and Kashmir, India

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***Correspondence:**
Dr. Javaid Iqbal Gulshan,
E-mail: djigq2222@gmail.com

ABSTRACT

Background: Congenital diaphragmatic hernia is a developmental disorder in the anatomy of the diaphragm, which can range from the presence of a thin sac to the frank absence of part of the diaphragm. It results in protrusion of abdominal contents in the chest. CDH once considered as a surgical emergency is no longer a valid dictum. But definitive management of CDH is the surgical correction of defect. Study the demographics, presentation, diagnosis, and surgical outcomes of congenital diaphragmatic hernia (CDH) repair without significant associated congenital anomalies at a tertiary care institute.
Methods: This was a retrospective study over 3 years from January 2021 to December 2023. Data was collected and noted from the medical records department. Patient's clinical data in terms of demography, presentation, radiology, preoperative condition, timing of surgery after admission, intra-operative findings and post-operative course (including postoperative complications, duration of NICU stay, the incidence of sepsis, and need of inotropes), etc.
Results: Most patients were male (85%). Among the total cohort, 70% were born outside the tertiary care institute and were referred from peripheral hospitals. Respiratory distress was the predominant complaint (85%). Congenital diaphragmatic hernia (CDH) was incidentally discovered in 5% of neonates. The majority of patients (70%) underwent surgical intervention between the 3rd postnatal day and up to one week. Left-sided herniation was observed in 90% of cases. An identifiable hernial sac was found in 18% of patients, with part or the entirety of the stomach being the most frequently encountered herniated viscera.
Conclusions: Congenital diaphragmatic hernia (CDH) presents as a significant congenital anomaly in the pediatric surgery domain. Hidden mortality in case of CDH leads to underreporting of the actual incidence of the condition. The success of surgical intervention depends on a myriad of factors, encompassing patient-specific attributes and surgical methodologies. However, instances devoid of overt congenital anomalies and featuring stable preoperative physiological parameters typically yield satisfactory surgical outcomes, with favorable results.

Keywords: Bochdalek hernia, Congenital diaphragmatic hernia

INTRODUCTION

Congenital diaphragmatic hernia is a congenital anomaly that results in protrusion of abdominal content into the chest cavity due to defective development of the diaphragm, which separates two cavities.[1,2] The reported incidence of (CDH) ranges from 1 in 2500 to 5000 infants.[3] Diagnosis of CDH is made either after birth when the infant presents with respiratory distress or prenatally, more new cases are now diagnosed prenatally.[4] Ultrasound is the standard imaging modality used for prenatal diagnosis of congenital anomalies including CDH but it can miss CDH in more than 25% of cases even after 24 weeks of gestation.[5] CDH develops due to failure of closure of the pericardial-peritoneal canal by underdeveloped pleuroperitoneal fold.[2,6]

Aetiopathogenesis of CDH is multifactorial but the exact cause and mechanism are not clear.[7-12] Multiple factors are likely to play a role in etiopathogenesis genetic, environmental nutritional, etc.[8] Low retinol and retinol-binding protein levels in new-borns have been strongly associated with CDH.[9] Anatomically CDH can classified as posterolateral, anterior, and central defects in the diaphragm.[11] The diaphragm is made of costal and crural muscle domains, CDH is due to abnormal development of the costal diaphragm.[12] The majority of diaphragmatic hernia (i.e.) more than 90% are posterolateral, with left-sided being 85%, right-sided 13%, and 2% bilateral.[4] Abdominal contents in the chest cause mass effects resulting in pulmonary hypoplasia on the same side of the lung.[13] According to the dual hit hypothesis, there is an initial insult during organogenesis resulting in bilateral hypoplasia, and mass effect due to herniation causes hypoplasia in the latter stage.[14] Prognostic factors include polyhydramnios, position of the stomach, liver, side of the defect, diagnosis during the early antenatal period, left ventricular hypoplasia, lung-to-head ratio, etc.[15] CDH is considered a syndrome in which there is varying degrees of pulmonary hypoplasia, lung immaturity, left ventricular hypoplasia, and persistent pulmonary hypertension.[16] Infants with respiratory distress are managed by supportive measures depending upon the severity of respiratory distress, if an infant is not able to maintain the required respiratory parameters on nasal oxygen, intubation is considered with an intermittent mandatory rate of 20-40 breath per minute, positive end-expiratory pressure of 5cm H_2O.[17] High peak inspiratory pressures above 28cm H_2O should be avoided as it can cause lung injury and deep sedation should be used in intubated patients.[18] Pulmonary hypertension is associated with higher mortality and morbidity in CDH patients, hypoplasia results in a decrease in a number of pulmonary vessels and vascular bed overall size.[19,20] Infants with complex CDH have higher mortality compared to those with isolated CDH.[21] Pulmonary hypertension is managed by inhalational nitric oxide, milrinone, sildenafil, prostaglandin I_2, and prostaglandin E_1.[22] Surfactant deficiency due to prematurity or hypoplasia can be treated with surfactants.[23] If an infant is not able to maintain preductal saturation >85% or post ductal saturation >70%, in all available modes of ventilation, ECMO is used as lifesaving, in those patients with gestational age >34 weeks weight >2 kg and without any lethal anomalies [7,24] Surgical repair once considered emergent should be done after the physiological stabilization of the infant, open surgery is preferred over minimal invasive.[18,25] Eventration of the diaphragm can be repaired by plication, in the case of a hernia with a sac, excision with patch repair can be done similarly for a hernia with a true defect.[24]

Our objective of this study was to study patients' demography, clinical profile and assess surgical outcomes of diaphragmatic hernia repair and post-operative expectant complications.

METHODS

This retrospective study collected data from January 2021 to December 2023 from the hospital records department and analyzed it using MS Excel. The study included a total of 20 patients who underwent surgery at the pediatric surgery department of the Government Medical College and associated hospitals in Jammu.

Inclusion and exclusion criteria

Inclusion criteria were all patients diagnosed with congenital diaphragmatic defects requiring surgical intervention. The exclusion criteria were patients with major congenital anomalies directly affecting survival. Patients requiring invasive ventilation. Neonates' candidates for extracorporeal membrane oxygenation.

According to the records, all patients underwent surgery following initial resuscitation and stabilization, with the exclusion of any significant congenital anomalies. The majority of diagnoses were made postnatally due to the unavailability of antenatal fetal scans for most cases. After obtaining proper consent from the parents of patient and arranging one-unit cross-matched packed red blood cells, patients were shifted to the operating table. All cases were done under general anesthesia and preoperative dosage of antibiotics was given in each case. After patients were intubated by an anesthetist, per urethral catheter was placed under all aseptic conditions, electrocautery lead was properly placed on the lower back, avoiding touching any bony prominences, desired position of patient on the operating table was done, by placing sterile sheet roll under the lower thorax. Patients were wrapped in sterile cotton on limbs and head to avoid hypothermia. 5% betadine was used to paint the chest and abdomen avoiding excessive spills to avoid electrocautery accidents. Draping was done with sterile sheets. Transabdominal approach was used in all cases, left subcostal incision with the number 10 blade was made incising skin, muscles were cut by electro cautery, all layers were opened one by one till peritoneum. After breaching the peritoneum and reaching the abdominal cavity, site and size of the hernia in the diaphragm was identified. A thorough examination of the abdominal cavity was done. Hernial contents were identified and reduction was done slowly but with gentle traction towards the pelvis after decompression of the stomach through the nasogastric tube. Herniated contents were placed in abdominal cavity after gentle reduction. Edges of diaphragmatic defect were identified and held by nontraumatic forceps and ease of approximation was checked, edges were sutured with nonabsorbable monofilament suture. In cases of sac without complete absence of part of the diaphragm, excision of part of the sac was done followed by approximation of edges was done. A prosthetic mesh patch was not used in any case as remnant diaphragmatic edges were adequate for approximation. Intercostal chest drains were used in all cases, 12 French suction catheters were used with added

side eyes to avoid blockage of water seal bag was attached movement of the column was checked, drain was fixed with nonabsorbable suture to the chest wall, to avoid accidental removal or displacement. Intraoperative expansion of the lung and degree of hypoplasia were assessed. Air leak and suture security of the suture line of the diaphragm were confirmed by using saline and manual tidal volume. The abdominal incision was closed with an absorbable suture in layers and the skin was closed with a subcuticular absorbable suture. The antiseptic dressing was applied on the suture line. Intraoperative findings were documented in intraoperative notes. None of the patients was extubated in the immediate postoperative period. Post-operative care was done in the neonatal intensive care unit by pediatricians and pediatric surgeons. Most of the patients were extubated on the 3rd to 4th postoperative day

RESULTS

Analysis of data from a sample of 20 operated patients showed that 85% (17 patients) were male, with the remaining 15% (3 patients) being female (Table 1). The majority of patients, specifically 70% (14 neonates), were referred from peripheral hospitals. Antenatal fetal ultrasonography (USG) data was available for only four patients diagnosed with congenital diaphragmatic hernia (CDH), with no evidence of other significant congenital anomalies directly affecting survival (Table 1).

Table 1: Characteristics and findings of neonates.

Characteristics	Findings	Number	Percentage
Gender	Male	17	85
	Female	3	15
Birth	Inborn	6	30
	Outborn	14	70
Diagnosis	Antenatal	4	20
	Postnatal	16	80
Side of hernia	Right	2	10
	Left	18	90
	Mid sternal	0	0
Mode of delivery	LSCS	2	10
	NVD (hospital)	14	70
	NVD (home)	4	20
Signs/presentation	Respiratory distress	17	85
	Feed refusal/vomiting	2	10
	Incidental	1	5
Ventilation mode	NIV (nasal prongs)	13	65
	Intubation	7	35
Maturity	>37 weeks	20	100
Birth weight	2.1 kg-3 kg	16	80
	3 kg-3.2 kg	4	20
Timing of surgery	Within 3rd postnatal	6	30
	4th day to 1st week	14	70
Bacteriological cultures (Blood/ETT)	Klebsiella	8	40
	Pseudomonas	6	30
	Multiple organisms	6	30
	Enterobacter	4	20
Discharge	Within 14 days post op.	6	30
	Within 3 weeks	10	50
	>3 weeks	2	10

The mode of delivery included lower segment cesarean section (LSCS) in 10% (2 cases), normal vaginal delivery at the hospital in 70% (14 cases) of patients, and home delivery in 20% of cases. The presenting complaints were respiratory distress in 85% (17 patients), refusal to feed, vomiting, and lethargy in 10% (2 cases), and it was an incidental finding during a medical check-up in (1 case) (Table 1). All patients had a birth weight exceeding 2.1 kilograms, with a range observed between 2.1 kilograms and 3.2 kilograms (Table 1). Of the patient cohort, 65% (13 individuals) successfully maintained adequate oxygen saturation levels when utilizing non-invasive ventilation (NIV) with nasal prongs. Conversely, 35% (7 patients) required intubation for ventilatory support (Table 1).

Left-sided diaphragmatic hernia was observed in 90% (18 patients), while right-sided diaphragmatic hernia was present in 10% (Table 1). Surgical intervention occurred between the 4th and 15th post-natal days, with 50% (10 patients) undergoing the procedure within the first week of birth and the remaining 50% after the first week (Table 1). The hernial contents within the chest were documented, indicating spleen involvement in 60%, small bowel in 75%, either part or the entirety of the stomach in 80%, colon in 50%, left lobe of the liver in 20%, the entire liver in 10%, and a hernial sac in 18%. Additionally, all cases exhibited varying degrees of pulmonary hypoplasia.

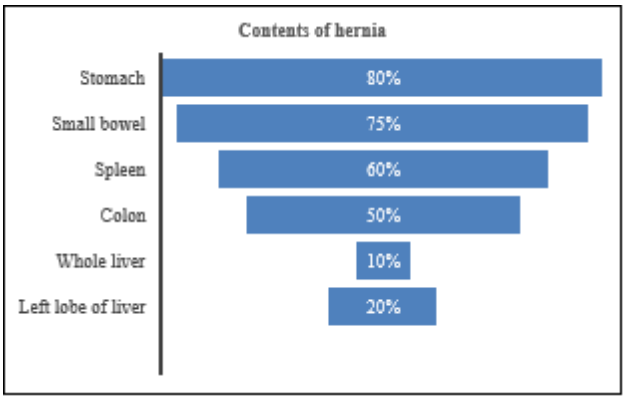

Figure 1: Percentage of herniated abdominal organs in the chest.

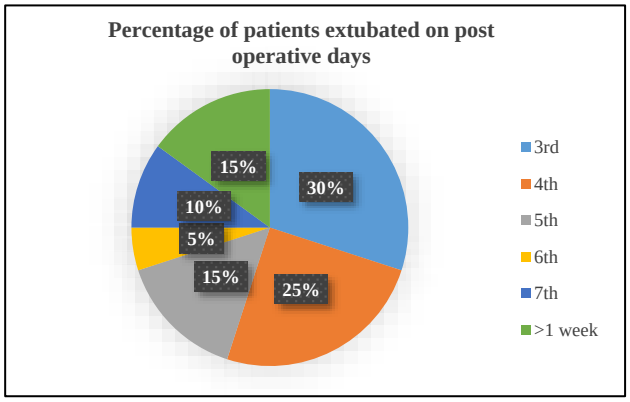

Figure 2: Percentage of patient's and post-operative day of extubation.

Blood cultures revealed *Klebsiella* in 40% of patients, multiple organisms in 30%, *Pseudomonas* in 20%, and Enterobacter in 10%. During the postoperative period, ionotropic support for more than 72 hours was required for 11 patients (55%). Thrombophlebitis at intravenous cannula sites affected 70% or 14 patients. Other complications included postoperative sepsis in 40% (8 patients), pulmonary hemorrhage in 15% (3 patients), ICCT blockage and pneumothorax in 20% (4 patients), and cellulitis in 15% (3 patients) (Figure 2). Overall mortality rate was 10% major causes were severe sepsis with shock in one patient and pulmonary hemorrhage in second patient. Patient with septic complication expired in the second week of post-operative while patients with severe pulmonary hemorrhage died on 4th post-operative day.

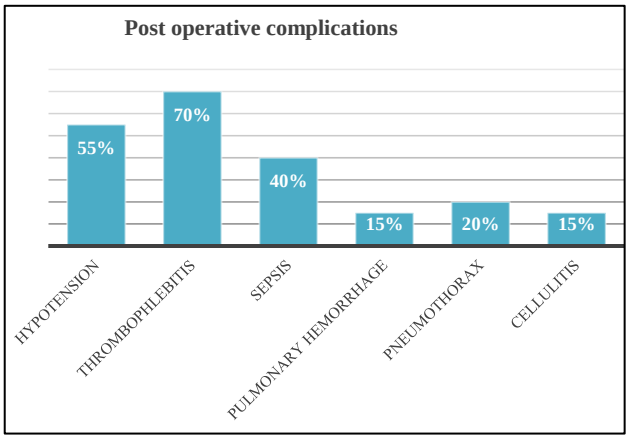

Figure 3: Post-operative complications.

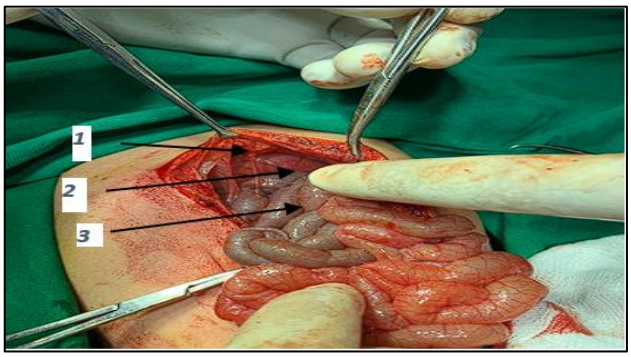

Figure 4: 1) Anterior edge of the diaphragm held with Allis forceps; 2) defect in the diaphragm; 3) small bowel pulled back into the abdomen.

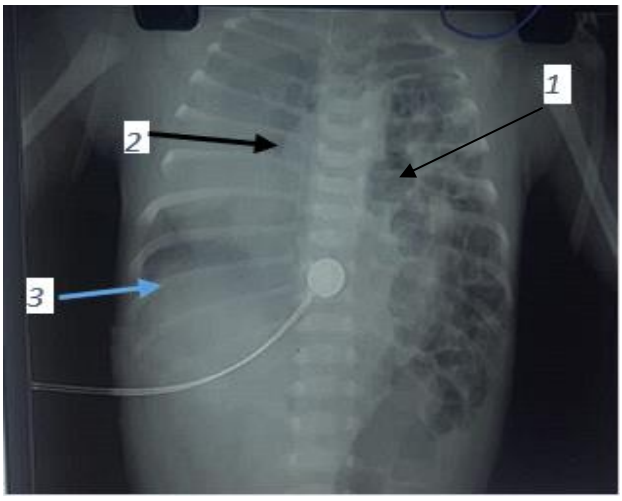

Figure 5: Chest X-ray of the neonate with CDH, 1) showing bowel loops in the left side of the thoracic cavity; 2) deviation of mediastinum towards the right side; and 3) right costo-phrenic angle.

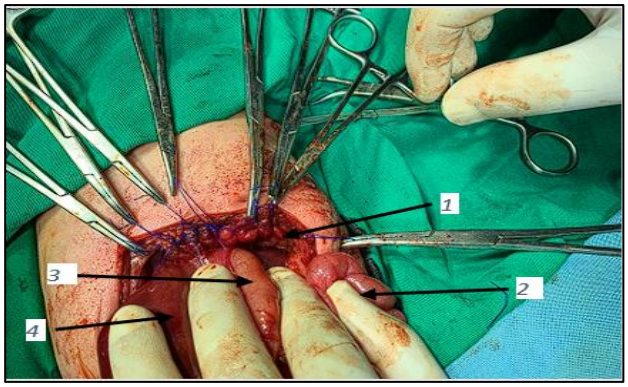

Figure 6: 1) Approximated edges of diaphragm with non-absorbable interrupted sutures; 2) small bowel; 3) stomach; and 4) liver in abdominal cavity.

DISCUSSION

Patients (neonates) with congenital diaphragmatic hernia (CDH) not only exhibit a diaphragmatic defect impacting survival, but also present with a diverse array of additional anatomical anomalies and varying physiological statuses at birth. Preoperative optimization further differs among patients and across health centers with distinct infrastructures. This variability poses challenges in establishing a standardized protocol of care and comparing overall survival and mortality rates. In this retrospective analysis, the survival rate was 90%, with a corresponding mortality rate of 10%. Our study specifically focused on patients without significant gross congenital anomalies and those who did not require aggressive initial resuscitation. The optimal timing of surgery remains a subject of debate, with conflicting opinions favouring both early intervention and advocating for a delayed surgical approach.[25-27]

In our study, a substantial majority of participants were male, comprising 85%, while females constituted the remaining 15%. This resulted in a male-to-female ratio of 5.7:1. In contrast, Kursheed et al and Crankson et al reported ratios of 1.5:1 and 1.8:1, respectively. Bhat et al, however, documented a ratio of 3:1 in their study.[29,30,32]

Most patients arriving at our center were referrals from peripheral hospitals, with 80% of these individuals lacking prenatal diagnoses or antenatal fetal scans upon presentation. In the study by Crankson et al, 56% of patients had received an antenatal diagnosis of congenital diaphragmatic hernia (CDH). Additionally, Teo et al reported that 83.3% of patients in their study sample had undergone antenatal fetal scans leading to a diagnosis of CDH.[30,31]

In our study, none of the neonates were below 37 weeks of gestation. Crankson et al reported that 33% of patients in their study were preterm, i.e., born before 37 weeks of gestation.[30]

In this study, 90% of neonates were delivered vaginally, while 10% were delivered via lower segment cesarean section (LSCS). Teo et al reported that in their study, 68.75% of cases were delivered vaginally, with the remaining delivered via LSCS.[31]

In our study, 30% of patients underwent surgery within the 3rd postnatal day, while the remaining 70% were operated on within the first week, with a mean operative time of 4.2 days. Similarly, Crankson et al reported a mean operative time of 4 days.[30]

In our study, patients with right congenital diaphragmatic hernia were successfully discharged. A study by Schaible et al reported a higher survival rate among patients with right-sided CDH when compared to those with left-sided CDH.[34]

Congenital diaphragmatic hernia (CDH) results in reduced lung capacities and hypoxia, leading in low oxygen saturation. In our study, 35% of patients required intubation, while the remaining patients received oxygen inhalation through nasal prongs. Sahoo et al reported that 97.3% of patients in their study needed intubation.[35]

Left-sided congenital diaphragmatic hernia (CDH) is the more prevalent type, with 90% of patients in our study presenting with left-sided CDH. Similarly, Colvin et al reported that 84% of patients in their study had left-sided CDH.[36]

Cardiac malformations represent the most common anomalies associated with congenital diaphragmatic hernia (CDH), carrying a poor prognosis, especially in severe cases.[26] In our study group, none of the patients exhibited significant anomalies or severe cardiac malformations that directly compromised survival in either the early neonatal stage or later. In contrast, Colvin et al reported that in their study sample, 47% of patients had major congenital anomalies.[36]

The most prevalent postoperative complications in our study included sepsis in 30% of patients and pneumothorax in 20% of patients. In a study by Colvin et al, sepsis was observed in 20% of patients, while pneumothorax occurred in 12% of patients.[36] Migliazza et al in their study reported a total incidence of post-operative pneumothorax in 38.3%.[37]

In our study, the majority of patients were discharged in the 3rd week post-operative, accounting for 40%, with 30% discharged in the 2nd post-operative week, Skari et al mentioned an average hospital stay of 21 days in their manuscript.[33] For patients who revisited the hospital on their follow-up, none of them had a recurrence, while a recurrence of 1.9% was mentioned by Bagłaj.[38] in their study. In the literature recurrence rate of 5-50% is mentioned by Crankson et al.[30]

The limitations of the study are sample size, short follow-up period, and the fact of hidden mortality in the case of congenital diaphragmatic hernia

CONCLUSION

Congenital diaphragmatic hernia (CDH) presents as a significant congenital anomaly in the pediatric surgery domain. Hidden mortality in case of CDH leads to underreporting of the actual incidence of the condition. The success of surgical intervention depends on a myriad of factors, encompassing patient-specific attributes and surgical methodologies. However, instances devoid of overt congenital anomalies and featuring stable preoperative physiological parameters typically yield satisfactory surgical outcomes, with favourable results.

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