ORIGINAL RESEARCH

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Diaphragmatic paralysis after pediatric cardiac surgery: Associated implications and outcomes

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Abstract

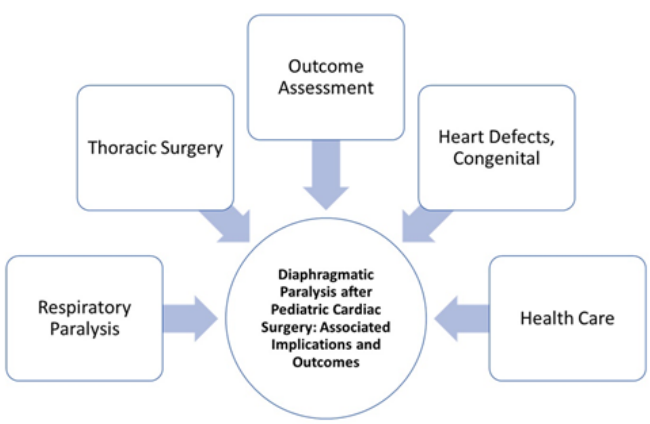
Objective: This study aimed to analyze the associated implications and outcomes of diaphragmatic paralysis following pediatric cardiac surgery at a center specializing in congenital heart diseases.

Methods: The eligibility criteria for this cross-sectional study included patients who underwent surgery between January 2020 and June 2021. We collected sociodemographic, anthropometric, cardiac diagnostic, surgical, diaphragmatic paralysis diagnostic, ventilatory, and temporal variables and those related to the outcome of diaphragmatic paralysis and hospital outcomes. We conducted descriptive and inferential (logistic regression model) statistical analysis.

Results: Among a total of 246 patients, a prevalence of 2.4% of diaphragmatic paralysis was estimated, predominantly in those with cyanotic congenital heart disease. Patients with extubation failure were 3.59 times more likely to have diaphragmatic paralysis ($p<0.05$). The need for non-invasive ventilation increased the patient's chance of being diagnosed with diaphragmatic paralysis by 14 times (OR=14.29, $p>0.05$). Unilateral diaphragm injury and the necessity for surgical treatment by plication were predominant. All patients with diaphragmatic paralysis were in the intensive care unit discharged.

Conclusion: Extubation failure and non-invasive support were associated with diaphragmatic paralysis. Outcomes included diaphragmatic plication and subsequent discharge from the intensive care unit.

Graphical abstract



Key words: Respiratory paralysis, thoracic surgery, heart defects, congenital; outcome assessment, healthcare

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Introduction

Diaphragmatic paralysis is a susceptible manifestation in the postoperative phase of congenital heart disease (1-3). Despite advances in surgical interventions, complications from injury to surrounding structures represent a potential risk (4). The phrenic nerve, due to its anatomical positioning, is vulnerable during heart surgery (5).

Stretching, crushing, amputation, or thermal injury due to hypothermia (contact with cold saline solution) or hyperthermia (contact with the cauterization plate) of the phrenic nerve are factors that induce diaphragmatic paralysis (6, 7). The asymmetry of thoracic excursions suggests its diagnosis (8, 9). Other symptoms include respiratory distress with tachypnea, paradoxical breathing, degree of cyanosis, shortness of breath, or increased work of breathing (8, 10). Difficulty weaning from ventilation (11) and recurrent lung infections are signs of suspected diaphragmatic paralysis (12).

For diagnostic confirmation, complementary exams are helpful (11). Elevation of the diaphragmatic hemi-dome on chest X-ray suggests diaphragmatic paralysis (11). Diaphragm ultrasound to assess mobility and range of motion is a non-invasive method at the bedside (11). Fluoroscopy with exposure to ionizing radiation is considered by many to be the gold standard for this diagnosis (11). In select cases, diaphragmatic plication surgery is the treatment of choice with good results (13).

The respiratory complication of diaphragmatic paralysis can result in delays in hospital recovery (2) and increased postoperative morbidity and mortality (14). Identifying methods to improve early recognition and treatment of diaphragmatic paralysis can contribute to better care for the pediatric population undergoing cardiac surgery, reducing hospitalization time and associated costs (15).

The study aimed to analyze the associated implications and outcomes of diaphragmatic paralysis after pediatric cardiac surgery in a center specializing in congenital and acquired heart diseases.

Methods

Study design and population

The study employed an observational, retrospective, cross-sectional design. We conducted the research within the hospital setting of the Children's Postoperative sector of the National Institute of Cardiology, a federal hospital of the Ministry of Health recognized as a reference center for congenital and acquired heart disease treatment. Study participants included patients between 0 to 18 years of age who underwent pediatric cardiac surgery from January 2020 to June 2021. Exclusion criteria included individuals without a defined hospital outcome (discharge from the Children's Postoperative sector, transfer to another hospital, or death) at the end of the study follow-up.

Sample size calculation, considering a prevalence of diaphragmatic paralysis of 5.5% (3) with a margin of error of 5% and a confidence level of 95%, estimated that at least 80 patients should be included in the study.

We selected records from a specific Microsoft Excel database (version 365) that stored medical record information for all patients throughout their hospitalization. This database facilitated the identification and comparison of participants with or without a diagnosis of diaphragmatic paralysis comprising 246 patients.

Informed consent for procedures and surgery was obtained from patients` parents or guardians. The Research Ethics Committee of this institution approved the study under number 66022422.0.0000.5272.

Diagnosis of diaphragmatic paralysis

At the time, the multidisciplinary team suspected diaphragmatic paralysis, phrenic nerve injury, in patients with elevation of hemidiaphragm on chest X-ray, paradoxical thoracic movement, recurrent or persistent atelectasis, difficulty in weaning of mechanical or non-invasive ventilation, and those who failed extubation (16).

Collected data

Variables collected, categorized into blocks, included: sociodemographic (age and sex), anthropometric (weight), cardiac diagnoses (type and categorization of congenital heart disease), surgical (type and presence of previous surgery), diagnosis of diaphragmatic paralysis (presence of paralysis and, if applicable, unilateral or bilateral injury), ventilatory (ventilatory status at admission in the immediate postoperative period and, if applicable, need for mechanical ventilation, extubation failure, and need for non-invasive ventilation), outcome of diaphragmatic paralysis (conservative or surgical treatment), temporal (length of hospital stay and, if applicable, time on mechanical ventilation and non-invasive ventilation), and hospital outcome (discharge from the Children's Postoperative sector, transfer to another hospital, or death).

Statistical analysis

Statistical analysis was performed using the R statistical package, version 3.3.4, and STATA 13.0 and reported the 95% confidence level and confidence intervals.

In the descriptive and exploratory statistical analysis, we employed absolute (n) and relative frequencies (%) for categorical variables and measures of central tendency (mean and median) and dispersion (standard deviation) for continuous variables. In comparing patients with versus patients without diaphragmatic paralysis, we applied the Chi-square test and t-test.

In the inferential analysis, we adopted a logistic regression model for the outcome used, diaphragmatic paralysis diagnosis, and adjusting or stratifying the analysis for potential confounding variables. The explanatory variables were represented by the characteristics of study patients with p-value <0.05, according to the chi-square test. Generalized Linear Models were used to estimate the effects, a class of statistical models that offer great flexibility in probability distributions, depending on the type of response variable.

Results

A total of 246 patients met the eligibility criteria. The prevalence of diaphragmatic paralysis was 2.4% (6 out 246). The majority were male, undergoing corrective heart surgery without prior procedures. Only one patient underwent palliative surgery (Blalock-Taussig shunt). The primary diagnosis of pulmonary atresia with interventricular communication (PA with VSD, cyanotic congenital heart disease) stood out, followed by Tetralogy of Fallot (ToF, cyanotic congenital heart disease), transposition of the great arteries (TGA, parallel circulation), total atrioventricular septal defect (AVSD, pulmonary hyperflow), and aortic coarctation (systemic hypoflow), each with the same proportion. An institutional surgeon's team performed pediatric cardiac surgeries. It was not possible to identify surgeon distribution in the patients with diaphragmatic paralysis versus those without diaphragmatic paralysis.

Bilateral diaphragmatic paralysis occurred in only one, while the majority with unilateral diaphragm injury experienced it on the left side. The bilateral paralysis patient had a primary diagnosis of ToF. In the child-adolescent paralysis group, the diagnosed with PA VSD and TGA underwent Rastelli surgery.

Mechanical ventilation was required for all patients with diaphragmatic paralysis immediately postoperatively. Of these, 83% needed non-invasive ventilation (NIV) and faced extubation failure. Diaphragm plication surgery occurred in 67% of cases, all involving infants, and was recommended based on the patient's respiratory condition after an observation time.

The overall study sample was mainly infants, male sex, undergoing corrective cardiac surgery, coming from the surgical center intubated and, consequently, requiring mechanical ventilation, and having the hospital outcome the discharge from the intensive care unit (ICU) to the ward (Table 1).

Compared to patients without diaphragmatic paralysis, those with diaphragm injury had higher average weight and age, along with a longer average duration of mechanical ventilation postoperatively and during their ICU stay, though differences did not reach significance level ($p>0.05$) (Table 1). The study observed similar patterns in NIV mode postoperatively, overall hospitalization period, length of stay in the postoperative period, and ICU. Only one patient with diaphragmatic paralysis used non-invasive support preoperatively. None of the patients with diaphragmatic paralysis died (Table 1).

The most prevalent diagnoses in the group without diaphragmatic paralysis were, respectively, VSD, ToF, total AVSD, hypoplastic left heart syndrome, and atrial septal defect (ASD). In almost 4.5% of these patients, the diagnosis was TGA, double outlet right ventricle (DORV), and PA with VSD. Between 2% to 4% were diagnosed with ASD and VSD, common atrium with single ventricle, coarctation of the aorta, truncus arteriosus, tricuspid atresia with interventricular communication, partial AVSD, total anomalous pulmonary venous return, and dilated cardiomyopathy. In the 1% range were patent ductus arteriosus, double inlet left ventricle, partial anomalous pulmonary venous return, aortic stenosis, pulmonary stenosis, cardiac tumor, and PA. Among the less prevalent diagnoses were anomalous left coronary artery, tricuspid atresia with pulmonary stenosis, intermediate AVSD, restrictive cardiomyopathy, mitral stenosis, interrupted aortic arch, and Ebstein's anomaly.

The study revealed significant differences in ventilation status – more patients with diaphragmatic paralysis needed NIV ($p<0.05$) and experienced extubation failure ($p<0.05$ for both), the duration of NIV postoperatively and ICU stay were significantly longer ($p<0.05$ for both) than in patients without diaphragmatic paralysis.

Table 1. Clinical and perioperative variables of study patients

Variables	Sample (n=246)	Diaphragmatic Paralysis (n=6)	Without Diaphragmatic Paralysis (n=240)
Male sex, n(%)	132 (53.87)	4 (66.6)	128 (53.3)
Weight, kg	14.37 (14.83) (8.2)	18.81(21.22) (8)	14.26 (14.68) (8.2)
Age, months	42.37 (56.28) (12)	51.83(71.90) (10)	42.14 (56.01) (12)
Age groups, n(%)			
Neonate	27 (11.02)	0 (0)	27 (11.25)
Infant	133 (54.29)	4 (66.6)	129 (53.75)
Child	57 (23.26)	1 (16.6)	56 (23.3)
Adolescent	28 (11.43)	1 (16.6)	27 (11.25)
Type of surgery, n(%)			
Corrective	194 (78.86)	5 (83.3)	189 (78.75)
Palliative	53 (21.54)	1 (16.6)	52 (21.66)
Previous surgery	28 (11.38)	1 (16.6)	26 (10.8)

Table 1. Clinical and perioperative variables of study patients (continued from page 378)			
Variables	**Sample (n=246)**	**Diaphragmatic Paralysis (n=6)**	**Without Diaphragmatic Paralysis (n=240)**
Immediate postoperative ventilatory status, n(%)			
Orotracheal intubation	220 (89.43)	100 (6)	220 (91.66)
Oxygen therapy	9 (3.66)	0 (0)	9 (3.75)
Room air	17 (6.91)	0 (0)	17 (7.08)
Need for MV	229 (93.09)	100 (6)	223 (92.91)
Need for NIV*	73 (29.67)	83.3 (5)	68 (28.3)
Extubation failure*	32 (14.68)	83.3 (5)	27 (11.25)
Duration of MV, days			
Preoperative	1.88 (5.24) (1)	0.83 (0.41) (1)	1.91 (5.31) (1)
Postoperative	5.83 (12.06) (2)	12.5 (10.93) (10)	5.68 (12.06) (2)
Total ICU stay	7.04 (14.18) (2)	12.5 (10.93) (10)	6.91 (14.24) (2)
Duration of NIV, days			
Preoperative	2.5 (0)	-	2.63 (3.83) (0)
Postoperative*	4.95 (6.24) (3)	10.4 (9.65) (8)	4.59 (5.87) (3)
Total ICU stay*	5.67 (7.41) (3)	12.4 (12.5) (8)	5.23 (6.86) (3)
Mode of NIV, n(%)			
CPAP	32 (43.84)	2 (33.3)	30 (12.5)
Bi-level	41 (56.16)	3 (50.0)	38 (15.8)
Length of hospital stay, days			
Preoperative	2.61 (9.16) (0)	0.16 (0.41) (0)	2.67 (9.26) (0)
Postoperative	11.76 (21.34) (5)	27.5 (25.68) (14)	11.36 (21.14) (5)
Total ICU stay	14.18 (25.59) (6)	27.66 (26.01) (14)	13.85 (25.55) (5)
Hospital outcome, n(%)			
Discharge to ward	216 (87.80)	6 (100)	210 (87.5)
Transfer	9 (3.66)	0 (0)	9 (3.75)
Death	21 (8.54)	0 (0)	21 (8.75)

Data are presented n(%) and mean (SD) (median), * - p< 0.05.
CPAP - continuous positive airway pressure, ICU - intensive care unit; kg – kilogram, MV - mechanical ventilation, NIV - non-invasive ventilation, SD – standard deviation

The logistic model with log-link function provided the odds ratio used as a measure of association between the response variable (diagnosis of diaphragmatic paralysis) and the explanatory variables (extubation failure and need for NIV).

Patients with extubation failure were more than three times more likely to have diaphragmatic paralysis (OR=3.59; $p<0.05$). The need for NIV was a clinically relevant implication. These patients with diaphragmatic paralysis were 14 times more likely to require NIV (Table 2).

Table 2. Logistic regression model for the diagnostic outcome of diaphragmatic paralysis			
Diaphragmatic Paralysis			
Adjusted Variables	**OR**	**95% CI**	**p**
Extubation failure	3.59	1.41-5.77	0.001
Need for NIV	14.29	-	0.995

CI – confidence interval, NIV - non-invasive ventilation, OR - odds ratio

Discussion

In this observational and cross-sectional study, we highlighted the associated implications and outcomes of diaphragmatic paralysis after pediatric cardiac surgery in a single center. Diaphragm dysfunction after cardiac surgery is a complication that leads to delays in recovery and increased postoperative morbidity and mortality (15), correlating with the data found in our study. The average total length of stay in the ICU was almost twice as long in patients diagnosed with diaphragmatic paralysis.

The estimated prevalence of diaphragmatic paralysis (2.44%) can be considered low, consistent with other studies. Foster et al. (15) reported a 2.2% diaphragm dysfunction, while Dagan et al. (17) found a dysfunction of 0.3% . Lemmer et al. (7) and Akbariasbagh et al. (3) reported higher values of 4.1% and 5.5%, respectively. It is important to note that different study methods and approaches can impact the reported values.

In the literature, associated procedures such as bidirectional Glenn cardiac surgeries, Fontan, systemic shunts for pulmonary arteries, TOF repair, and VSD repair have been linked to higher percentages of diaphragmatic paralysis (2). According to Akay et al. (12), ToF correction was the most common surgery associated with diaphragmatic paralysis, followed by Blalock-Taussig shunt, ventricular septum correction, and TGA. In our study, 50% of patients diagnosed with diaphragmatic paralysis underwent TOF and TGA corrective surgeries, and Blalock-Taussig shunt palliative surgery.

Our study demonstrated that extubation failure and need for NIV are implications associated with diaphragmatic paralysis.

Diaphragm paralysis causes respiratory discomfort due to the paradoxical movement of the affected diaphragm and the contralateral displacement of the mediastinum, which can also present atelectasis, pneumonia, and difficulty in ventilatory weaning (13). This justifies the higher number of extubation failures in the group of patients with paralysis compared to those without, as well as the longer duration of mechanical ventilation in the postoperative phase.

The need for NIV is common in patients with diaphragmatic paralysis, as demonstrated in our study. Non-invasive CPAP (18) and bilevel-positive airway pressure ventilation are commonly used as non-invasive supports (19). The conservative approach involves maintaining prolonged invasive or non-invasive ventilatory support while awaiting recovery (11). However, injuries associated with prolonged mechanical ventilation may occur, increasing the risks of atelectasis, pneumonia, bronchospasms, or chronic lung disease (11).

Physiotherapeutic treatment is considered a beneficial adjuvant measure in diaphragmatic paralysis. It provides essential exercises for respiratory muscles, drainage of accumulated chronic secretions, and prevents possible chest deformities. Therapeutic positioning in lateral decubitus contralateral to the side of diaphragmatic paralysis can be used for lung expansion (11).

Surgical treatment involves diaphragmatic plication. Indications include the inability to wean from mechanical or non-invasive ventilation or when respiratory difficulty persists in the first month of the patient's life (11). Despite being considered effective, controversies remain regarding the optimal timing for plication (12). The timing varies across different studies, ranging from early to late (16). However, spontaneous recovery is very atypical (12). Early diaphragmatic plication reduces the duration of mechanical ventilation, limiting associated morbidity and mortality, especially severe pulmonary infections and ICU stay (12, 20, 21).

In our study, the percentage of plication evidenced (67%) was higher than that of Akbariasbagh et al. (3) (52%) and occurred in all infant patients, possibly because respiratory mechanics depend more on the function of the diaphragm. Diaphragmatic plication does not interfere with the return of normal function. This information aligns with our study, where all patients with diaphragmatic paralysis who underwent plication were discharged from the ICU.

Study limitations

Study limitations include the single-center context, the small number of patients diagnosed with diaphragmatic paralysis, and unanalyzed potential contributing factors to phrenic nerve injury, such as surgical techniques, approaches, and total surgery time.

Conclusion

The study indicated that extubation failure and non-invasive support were implications associated with diaphragmatic paralysis. Its outcomes included diaphragmatic plication and, subsequently, discharge from the intensive care unit.

It is expected that by elucidating the implications associated with diaphragmatic paralysis and analyzing the outcomes, the study can contribute to even more assertive decision-making and, in the future, optimize outcomes such as reducing the length of hospital stay.

Ethics: A written informed consent for all procedrues was obtained from the patients parents or guardians. The Research Ethics Committee of this institution approved the study under number 66022422.0.0000.5272.

Peer-review: External and internal

Conflict of interest: None to declare

Authorship: R.S.T., T.P.A., K.A.K., and L.S.L. equally contributed to the study and preparation for manuscript, and fulfilled authorship criteria.

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Statement on A.I.-assisted technologies use: We declare that we did not use AI-assisted technologies in preparation of this manuscript

References

1. Vidhyadhari BS, Madavi K. Influence of proprioceptive neuromuscular facilitation techniques on diaphragm muscle activity and pulmonary function in subjects with Guillain-Barre syndrome. Indian J Physiother Occup Ther 2015; 9: 24-8.
2. Talwar S, Agarwala S, Mittal CM, Choudhary SK, Airan B. Diaphragmatic palsy after cardiac surgical procedures in patients with congenital heart. Ann Pediatr Cardiol 2010; 3: 50–7.
3. Akbariasbagh P, Mirzaghayan MR, Akbariasbagh N, Shariat M, Ebrahim B. Risk factors for post-cardiac surgery diaphragmatic paralysis in children with congenital heart disease. J Tehran Heart Cent 2015; 10: 134-9.
4. Healy F, Hanna BD, Zinman R. Pulmonary complications of congenital heart disease. Paediatr Respir Rev 2012; 13: 10-5.
5. Hosokawa T, Shibuki S, Tanami Y, Sato Y, Ko Y, Nomura K, Oguma E. Fluorographic findings of diaphragmatic paralysis with spontaneous recovery. Pediatr Int 2021; 63: 895-902.
6. Baker CJ, Boulom V, Reemtsen BL, Rollins RC, Starnes VA, Wells WJ. Hemidiaphragm plication after repair of congenital heart defects in children: quantitative return of diaphragm function over time. J Thorac Cardiovasc Surg 2008; 135: 56-61.
7. Lemmer J, Stiller B, Heise G, Hübler M, Alexi-Meskishvili V, Weng Y, et al. Postoperative phrenic nerve palsy: early clinical implications and management. Intensive Care Med 2006; 32: 1227-33.
8. Parker LA. Part 2: Birth trauma: injuries to the intraabdominal organs, peripheral nerves, and skeletal system. Adv Neonatal Care 2006; 6: 7-14.
9. Nakagama Y, Kaneko Y, Ono H. Reversible diaphragmatic paralysis caused by a malpositioned chest tube. Cardiol Young 2015; 25: 1382-4.
10. Yoshida K, Kawabata H. The prognostic value of concurrent phrenic nerve palsy in newborn babies with neonatal brachial plexus palsy. J Hand Surg 2015; 40: 1166-9.
11. Gerard-Castaing N, Perrin T, Ohlmann C, Mainguy C, Coutier L, Buchs C, Reix P. Diaphragmatic paralysis in young children: A literature review. Pediatr Pulmonol 2019; 54: 1367-73.
12. Akay TH, Ozkan S, Gultekin B, Uguz E, Varan B, Sezgin A, et al. Diaphragmatic paralysis after cardiac surgery in children: incidence, prognosis and surgical management. Pediatr Surg Int 2006; 22: 341-6.
13. Zhang YB, Wang X, Li SJ, Yang KM, Sheng XD, Yan J. Postoperative diaphragmatic paralysis after cardiac surgery in children: incidence, diagnosis and surgical management. Chin Med J (Engl) 2013; 126: 4083-7.
14. El Tantawy AE, Imam S, Shawky H, Salah T. Diaphragmatic nerve palsy after cardiac surgery in children in Egypt: outcome and debate in management. World J Pediatr Congenit Heart Surg 2013; 4: 19–23.
15. Foster CB, Cabrera AG, Bagdure D, Blackwelder W, Moffett BS, Holloway A, et al. Characteristics and outcomes of children with congenital heart disease needing diaphragm plication. Cardiol Young 2020; 30: 62-5.
16. Parmar D, Panchal J, Parmar N, Garg P, Mishra A, Surti J, et al. Early diagnosis of diaphragm palsy after pediatric cardiac surgery and outcome after diaphragm plication - A single-center experience. Ann Pediatr Cardiol 2021;14: 178-86.
17. Dagan O, Nimri R, Katz Y, Birk E, Vidne B. Bilateral diaphragm paralysis following cardiac surgery in children: 10-years' experience. Intensive Care Med 2006, 32: 1222-6.
18. Willis BC, Graham AS, Wetzel R, Newth CJL. Respiratory inductance plethysmography used to diagnose bilateral diaphragmatic paralysis: a case report. Pediatric Critical Care Med 2004;5: 399-402.
19. Tokuda Y, Matsumoto M, Sugita T, Nishizawa J. Nasal mask bilevel positive airway pressure ventilation for diaphragmatic paralysis after pediatric open-heart surgery. Pediatr Cardiol 2004; 25: 552-3.
20. Georgiev S, Konstantinov G, Latcheva A, Mitev P, Mitev I, Lazarov S. Phrenic nerve injury after paediatric heart surgery: Is aggressive plication of the diaphragm beneficial? Eur J Cardiothorac Surg 2013; 44: 808-12.
21. Floh AA, Zafurallah I, MacDonald C, Honjo O, Fan CS, Laussen PC. The advantage of early plication in children diagnosed with diaphragm paresis. J Thorac Cardiovasc Surg 2017;154:1715-21.