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Congenital cardiac surgery and diaphragmatic paralysis: efficacy of diaphragm plication

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Abstract

Background Diaphragmatic paralysis due to phrenic nerve injury is a rare but potentially serious complication following congenital cardiac surgery, with reported incidences ranging from 0.28 to 5.6%. Early plication has been recommended for children diagnosed with diaphragmatic paralysis, especially those requiring prolonged respiratory support after cardiopulmonary bypass. It is important to note that late plication may jeopardize the results of successful surgical plication due to diaphragm atrophy.

Results This retrospective study was performed between May 2020 and June 2023. Patients who could not be weaned from the ventilator and underwent diaphragmatic plication in symptomatic patients after phrenic nerve injury were included in the study. During the study period, 22 (3.5%) of 624 pediatric patients operated for congenital heart disease required diaphragm plication. The mean age of the patients ranged from 1 month to 13 years with a mean of 23.88 ± 37.99 months. Fourteen (63.6%) of the patients were female, and 8 (36.4%) were male. Two of the patients (9%) were exited due to ventilator-associated pneumonia and sepsis.

Conclusions In conclusion, diaphragmatic paralysis after congenital heart surgery is a rare but important complication that can lead to serious respiratory and cardiac problems. Diaphragmatic plication has been shown to be an effective intervention for pediatric patients with the potential to improve pulmonary function, reduce dyspnea, and minimize complications associated with diaphragmatic paralysis. Careful consideration of patient selection, timing, and potential complications is crucial in optimizing the outcomes of this surgical intervention.

Keywords Diaphragm, Plication, Phrenic nerve palsy, Diaphragm paresis, Cardiac surgery

Background

Diaphragmatic paralysis (DP) due to phrenic nerve injury is a rare but potentially serious complication following congenital cardiac surgery, with reported incidences ranging from 0.28 to 5.6% [1]. The phrenic nerve may be damaged during thymus removal, resection of the pericardium, traumatic dissection of the nerve, and application of ice to the mediastinum. Rarely, it has been reported to be injured after chest tube insertion and jugular and subclavian central catheter insertion [2, 3]. Electrophysiologic evaluations of the phrenic nerve have been found to be useful in determining phrenic nerve

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injury following cardiothoracic operations and provide valuable information for diagnosis and treatment [4].

Treatment of diaphragmatic paralysis often involves diaphragmatic plication, a surgical procedure that has been shown to lead to good results in both pediatric and adult patients. Its aim is to reduce the amount of dysfunctional diaphragm not participating in inspiration in symptomatic patients. In addition, in both unilateral and bilateral cases of diaphragmatic paralysis, diaphragm plication has been reported to improve respiratory function and reduce dyspnea [5]. Hemidiaphragm plication after repair of congenital heart defects in children has been associated with quantitative return of diaphragm function over time, demonstrating its efficacy in this patient population. Early plication has been recommended especially for children diagnosed with diaphragm paralysis who require long-term respiratory support after cardiopulmonary bypass [3, 6]. It is important to remember that late plication may jeopardize the results of successful surgical plication due to diaphragm atrophy [4, 7].

We will share the results of patients in whom we performed diaphragm plication for paralysis of the phrenic nerve after pediatric cardiac surgery in our clinic.

Methods

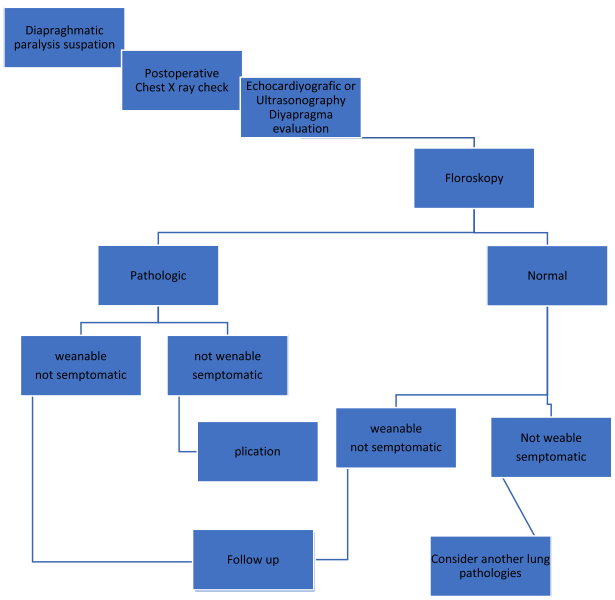
This retrospective study was conducted between May 2020 and June 2023. Patients who could not be weaned from the ventilator after the diagnosis of phrenic nerve injury and who underwent diaphragm plication applied to symptomatic patients were included in the study.

The postoperative respiratory circuit was connected to a Servo 900C anesthesia ventilator (Siemens Erlangen, Germany). Patients were extubated in the intensive care unit when adequate spontaneous respiration and airway reflexes were observed. The extubation decision was based on the following criteria: response to simple commands, oropharyngeal temperature more than 36.5 °C, hemodynamic stability, and the absence of uncontrollable arrhythmias. In addition, in arterial blood gas analyses, pH was required to be higher than 7.30, PaO2 to be higher than 60 mmHg with FiO2 less than 50%, and PaCO2 to be lower than 45 mmHg. Early mobilization, postural drainage, and respiratory exercises were performed by our expert respiratory physiotherapists or experienced nurses during MV weaning in the postoperative period.

Patient management

Routine chest radiography was performed every morning postoperatively and during their stay in the intensive care unit. Early postoperative regular and intermittent echocardiographic evaluation was performed if necessary. Failure to wean from mechanical ventilation, excessive respiratory effort, low oxygen saturation and hypoxemia (lower saturations than expected for cyanotic patients); reintubation within the first 24 hours after extubation; and diaphragmatic elevation or atelectasis in the absence of abdominal distension. Diaphragmatic paralysis diagnosis was suspected because of diaphragmatic elevation on chest X-ray or paradoxical movement of the epigastrium during spontaneous ventilation. One rib elevation of the right hemidiaphragm on chest X-ray was considered as a normal finding, and two rib elevations were considered as pathologic elevation. Left hemidiaphragm higher than right was considered as a pathological finding. Echocardiographic evaluation of patients with suspected diaphragmatic paralysis was performed by an expert pediatric cardiologist. Inspiratory and expiratory changes, amplification heights, and movement of the diaphragm in M-mode views in the subxiphoid region and the right and left eighth and ninth intercostal spaces along the midaxillary line were used. In all cases, DP was confirmed by ultrasound and/or fluoroscopy during spontaneous breathing. All patients underwent preoperative X-rays, and none of them showed any signs of diaphragmatic paralysis such as elevated diaphragm preoperatively. All patients scheduled for diaphragmatic plication underwent fluoroscopy to clarify the diagnosis (Algorithm 1).

Algorithm 1. The follow-up algorithm we apply in our clinic



For each case, sex, age, weight, cardiac diagnosis, the presence of genetic anomaly, risk category of the surgical procedure, intensive care unit follow-up, and

outcomes were collected from the chart. The type and diagnosis of congenital anomalies were also noted. Operative data were classified according to the Risk Adjustment for Congenital Heart Surgery-1 (RACHS-1) risk category [8]. Patients older than 18 years, those with a history of preterm birth, those with congenital pulmonary pathology, and those with known neuromuscular disease were excluded from the study.

Surgical procedure

In all patients, diaphragmatic plication was performed through a lateral thoracotomy through the sixth or seventh intercostal space via a thoracic approach. The central plication technique was used. Plication of the diaphragm was performed with a series of nonabsorbable longitudinal U-sutures placed from back to front, supported at both ends by a Teflon felt. Suturing was performed with a minimum of four sutures. A thoracic tube was routinely placed. The surgical goal was to achieve diaphragm plication to provide maximum possible lung capacity.

The study was performed retrospectively with the permission of the hospital management and in accordance with the Declaration of Helsinki and ethical guidelines.

NCSS (Number Cruncher Statistical System) 2007 (Kaysville, UT, USA) program was used for statistical analysis. Descriptive statistical methods (mean, standard deviation, median, frequency, ratio, minimum, maximum) were used to evaluate the study data. Significance was evaluated at $p < 0.01$ and $p < 0.05$ levels.

Results

During the study period, 22 (3.5%) of 624 pediatric patients operated for congenital heart disease required diaphragm plication. The mean age of the patients ranged from 1 month to 13 years with a mean of 23.88 ± 37.99 months. Fourteen (63.6%) of the patients were female, and 8 (36.4%) were male. The weight of the patients ranged between 3.2 and 60 kg with a mean of 10.75 ± 12.71 kg (median 7.85). Right-side plication was performed in 9 (40.9%) and left diaphragm plication in 13 (59.1%) patients.

The duration of intubation ranged from 6 h to 56 days with a mean of 25.77 ± 12.86 days. LOS ranged from 2 to 84 days with a mean of 37.18 ± 20.29 days. Hospital stay ranged from 8 to 128 days with a mean of 50.13 ± 26.17 days. On preoperative scopy, 12 patients (54.5%) had paradox movement of the diaphragm, and 10 patients (45.5%) had akinesia of the diaphragm.

Five (22.7%) patients required tracheostomy. Two (9%) of the patients died due to ventilator-associated pneumonia and sepsis. Nine (40.9%) single ventricle and 13 (59.1%) double ventricle repairs were performed. Four patients developed diaphragm paralysis after 2nd cardiac surgery, and one patient developed diaphragm paralysis during Glenn operation after PDA stenting before cardiac surgery.

Primary cases included VSD in 4 patients (18.2%), pulmonary banding in 1 patient (4.5%), unifocalization in 2 patients (9%), arterial switch operation (ASO) in 3 patients (13.6%) (Fig. 1), Glenn in 3 patients (13.6%), central shunt in 1 patient (4.5%), supracardiac TAPVD repair in 1 patient (4.5%), Fontan in 1 patient (4.5%), AV valve repair with Glenn in 1 patient (4.5%), and HLHS norwood stage 1 repair in 1 patient (4.5%). In redo cases, 1 patient (4.5%) had PVR after TOF, 1 patient (4.5%) had conduit exchange after unifocalization (Fig. 2), and 2 patients (9%) had postoperative diaphragm paralysis due to neopulmonary artery stenosis surgical repair after ASO.

Discussion

In a report sharing data from the STS congenital heart surgery database, they provided comprehensive data on the incidence and treatment of DP in pediatric patients undergoing heart surgery. They reported that pediatric

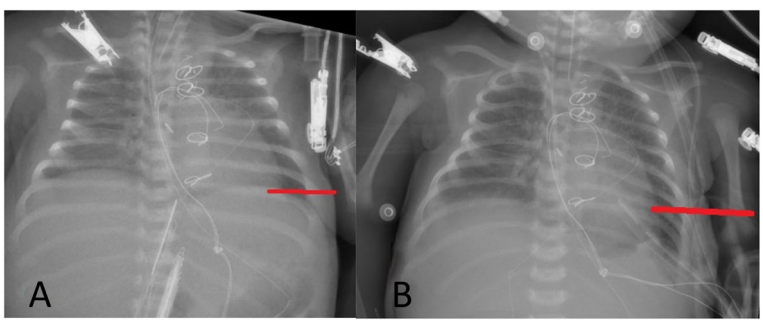


Fig. 1 **A** Pre-plication chest radiograph of a patient operated for transposition of the great artery. **B** Post-plication chest radiograph of a patient operated for transposition of the great artery

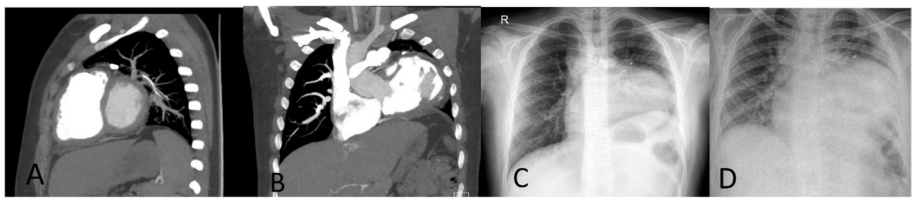


Fig. 2 **A**, **B** CT scan of a 13-year-old patient who underwent complete correction of VSD PA atresia at the age of 1 year before diaphragm plication. **C** Chest film of the same patient. **D** Chest film of the patient after plication

patients with DP had worse outcomes after cardiac surgery compared to patients without phrenic nerve injury. Plication was not associated with decreased mortality, morbidity, or shorter hospital stay, and in many categories, plication was associated with worse outcomes [9]. We shared the results of patients in whom we performed diaphragm plication in our clinical practice.

In order to reduce diaphragmatic nerve paralysis, it is important to pay attention to routine rules in surgery. During thymus removal, especially the left lobe seems to be closely associated with the diaphragmatic nerve in most children, and we should be very careful in this part. It is important not to use cautery during SVC release and cannulation and to visualize the nerve in interventions close to the phrenic nerve such as aortic arch reconstruction and unifocalization surgery [8–10]. Diaphragmatic paralysis following pediatric congenital heart surgery was rare (0.28 to 5.6% of all patients). As expected, it occurred more frequently following cardiac reoperations and is reported to occur more frequently in neonates. Diaphragmatic paralysis was associated with worse outcomes in all age and procedure groups [7, 9]. In our clinical practice, 22 of 624 patients (3.5%) required diaphragm plication. Since it is not possible to routinely evaluate asymptomatic patients and those who died before postoperative extubation, we believe that the incidence of diaphragm paralysis is much higher.

Stone et al. suggested that diaphragmatic recovery without plication is rare, and that plication is a permanent intervention and therefore should be used aggressively. They have a positive impression of plication and recommend aggressive plication practices and argue that this leads to rapid extubation and shorter hospitalizations [10, 11]. In our series (CT and chest X-ray images are shown in Fig. 2), we shared the results of a patient who underwent unifocalization at the age of 1 year and left diaphragm plication at the age of 13 years due to low saturation and left lung hypoplasia. Low saturation is a common finding after congenital cardiac surgery, and diaphragm paralysis is a rare cause of this finding. We believe that diaphragm evaluation should be performed carefully in the cardiologic follow-up of patients who have undergone cardiac surgery. Diaphragm paralysis may be overlooked when making decisions regarding complex cardiac pathologies of patients.

Adverse subdiaphragmatic venous hemodynamics combined with lack of inspiratory flow augmentation and low transhepatic gradient may be responsible for chronic ascites in patients with phrenic nerve palsy after Fontan surgery. Phrenic nerve palsy is considered a major risk factor for inadequate Fontan hemodynamics due to obstruction of passive venous blood flow. Fontan patients with phrenic nerve palsy have a longer hospital stay and a higher incidence of prolonged pleural effusion and chronic ascites than patients without paralysis. Early diaphragm plication may be appropriate to optimize Fontan circulation in these patients. A critical review of Fontan completion in patients with diaphragm paralysis has been advocated [11]. In our case series, 9 (40.9%) of the patients were single ventricle candidates. Single ventricle patients constitute a risky group for diaphragm paralysis requiring more than one operation, and inspiratory examination contributes significantly to passive venous flow in these patients. Single ventricle patients require careful evaluation for diaphragm paralysis in the decision to operate.

Many children with phrenic nerve injury have no clinical symptoms; these are often the criteria that lead to investigation and identification in retrospective series and some prospective series. In clinical situations, DP is usually first considered when clinical signs or breathing difficulties are present. Asymptomatic cases are usually first suspected when hemidiaphragmatic elevation is noted on a subsequent chest X-ray [12]. In our clinic, we do not routinely perform diaphragmatic USG and fluoroscopy, although we perform daily chest X-rays in all patients during the postoperative intensive care period. We perform further investigations in patients with suspected elevation clinically and on X-ray. We think that prospective studies with routine screening will give the best results on prevalence and risk factors.

Neonates and infants have limited physiologic reserve. The mechanical disadvantage of increased chest wall compliance and reliance on the diaphragm as the main muscle of respiration limits ventilatory capacity. The diaphragm and intercostal muscles have fewer slow

contracting type l muscle fibers, and high oxidative fibers for sustained activity, and this contributes to early fatigue when the work of breathing is increased. Phrenic nerve injury is more difficult to tolerate in infants and young children than in older children due to the influence of patient age and weight on the clinical effect. Studies have shown that especially children under 6 months of age benefit a lot from diaphragmatic plication. Caution is therefore needed in these children, and early plication has been found to be associated with early extubation. Several factors contribute to making young children more vulnerable to respiratory complications due to loss of diaphragmatic function, including weak intercostal muscles, increased compliance of the chest wall, horizontal orientation of the rib cage, and increased mobility of the mediastinum. Infants prefer or are placed in the supine position, which reduces lung capacity and facilitates the retention of secretions and bronchial obstructive secretions due to the small caliber of the infant bronchial tree. Paradoxical movements of the diaphragm also contribute to this reduced pulmonary compliance [12]. In our series, the mean age of the patients ranged between 1 month and 13 years with a mean of 23.88 ± 37.99 months. The mean weight of the patients ranged from 3.2 to 60 kg with a mean of 10.75 ± 12.71 kg. In our series, the patient who was operated for transposition of the great artery (Fig. 1) had no respiratory effort after extubation, although he had muscle strength and limb movements. After USG and fluoroscopy performed on the patient who had repeated reintubations, left DP was detected, and the patient could be weaned after left diaphragm plication.

There is no consensus on when and how to intervene in cases of diaphragmatic dysfunction. Some authors have suggested that phrenic nerve injury should be allowed to heal spontaneously in 1 to 6 weeks, which should be allowed to pass with general supportive therapy [13]. In contrast, Talwar et al. recommend a plication procedure as soon as diaphragmatic dysfunction is diagnosed [5]. We use a patient-specific approach. And like Ozturk et al., we believe that age, body weight, concomitant cardiac disease, respiratory muscle strength, serial diaphragm USG, and duration of assisted ventilation can help in making the decision for tracheostomy and plication [14].

Surgery for diaphragmatic paralysis does not require removal of any part of the diaphragm; the aim should be to provide a taut and intact diaphragm that eliminates paradoxical movement. For unilateral involvement, transthoracic repair via a posterior-lateral thoracotomy through the seventh intercostal space is usually preferred. Diaphragmatic plication can be performed transthoracically or transabdominally, while thoracic plication can be performed open or thoracoscopically. Depending on the surgeon's preference, one of the two main techniques of plication, central or radial plication, can be applied with the help of nonabsorbable sutures with or without plagements. Care is taken to ensure that the sutures pass through the diaphragm muscles but not deep enough to damage the underlying organs such as the spleen and liver [6, 12]. In our own practice, we perform plication using thoracotomy with radial plaginated sutures.

Limitation

Our study is retrospective, and diaphragm USG and fluoroscopy are not routinely performed in every patient in our clinic, but diaphragm movements are evaluated during echocardiography. Further imaging is performed in patients with suspected diaphragmatic paralysis (DP) on thorax X-ray or symptomatic patients. Therefore, it is not possible to give a clear rate for diaphragmatic paralysis and hypokinesia in asymptomatic patients.

Conclusions

In conclusion, diaphragmatic paralysis after congenital heart surgery is a rare but important complication that can lead to serious respiratory and cardiac problems. Diaphragmatic plication has been shown to be an effective intervention for pediatric patients with the potential to improve pulmonary function, reduce dyspnea, and minimize complications associated with diaphragmatic paralysis. Careful consideration of patient selection, timing, and potential complications is crucial in optimizing the outcomes of this surgical intervention.

Abbreviations

DP	Diaphragmatic paralysis
CPB	Cardiopulmonary bypass
USG	Ultrasonography
AV valve	Atrioventricular valve
PA/IVS	Pulmonary atresia with intact ventricular septum
SVC	Superior vena cava

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Authors' contributions

Study design by EA, KAK, and SB; data collection by EA, MAY, FY, SH, and ZGKÖ; writing by EA, FY, and KAK; supervising by ARK and SB; and final approval by all authors. All authors have read and approved the manuscript.

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Availability of data and materials

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

Declarations

Ethics approval and consent to participate

The study protocol was approved by the hospital management. The study is retrospective, and consent to participate was waived by the IRB. The

committee's reference number is not applicable. The study was conducted in accordance with the principles of the Declaration of Helsinki.

Consent for publication
Specific research consent was not obtained and waived. Before surgery, written informed consent to publish this information was obtained from study participant's next of kin and/or parent/legal guardian.

Competing interests
The authors declare that they have no competing interests.

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