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Original Article

Diaphragmatic Plication for Acquired Phrenic Nerve Injury after Congenital Cardiac Surgeries

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Abstract

Background: Phrenic nerve injury and diaphragmatic dysfunction are common after pediatric cardiac surgery leading to failure to wean from ventilatory support. Diaphragmatic plication is the standard management of diaphragmatic paralysis. The aim of this retrospective study is to review our experience with diaphragmatic plication and its effect on the operative outcome.

Methods: This retrospective cohort study included all patients who underwent diaphragmatic plication from June 2010 to June 2017. Seventy-six patients (2.87%) had unilateral diaphragmatic paralysis following 2646 congenital cardiac procedures. Sixty-four patients (2.4%) underwent diaphragmatic plication.

Results: The median age for the patients who had plication was 2.75 months (range 0.5- 36) and 3.7 months (range 0.66 to 123) for non-plicated patients. Thirty-six were males (56.25%), and the most common procedure associated with diaphragmatic plication was modified Blalock Taussig Shunt (n= 13; 20.3%). Left-sided diaphragmatic plication was performed in 44 patients (68.7%). The mean time between the primary surgery and diaphragm plication was 6.42±4.51 days. The mean ventilation period before plication was 4.93±3.71 days, and post plication ventilation median time was 2.11±1.82 days. Two patients (3.1%) required tracheostomy for prolonged respiratory insufficiency. One patient (1.6%) needed surgical revision, and two patients (3.1%) had their diaphragmatic plication during the initial surgery.

Conclusion: Diaphragmatic plication is an effective procedure in the management of postoperative diaphragmatic paralysis. We recommend early plication for patients with symptomatic diaphragmatic paralysis causing prolonged ventilation.

KEYWORDS

Diaphragmatic paralysis; Plication; Congenital cardiac surgery

Article History

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Introduction

Diaphragmatic muscle paralysis (DP) in the pediatric population has a dismal effect on respiration. This can be particularly important in children with univentricular physiology because respiratory mechanics play a vital role in hemodynamics and venous return [1 – 5]. The exact incidence of diaphragmatic paralysis after cardiac surgery in the pediatric age group is unknown. The reported prevalence varies from 0.3% in retrospective studies [6] to 12.8 % in prospective studies [7]. Diaphragmatic plication is the standard treatment of diaphragm paresis for children aged less than 1 year, yet the long-term outcome is unclear [8, 9].

Although spontaneous diaphragmatic recovery function has been observed, the time

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course and predictors of recovery are unknown [6, 9 – 12] Many authors have recommended early diaphragmatic plication for postoperative phrenic injury to decrease the mechanical ventilation time and length of hospital stay [3, 13 – 16]. The aim of this retrospective study is to review our experience with diaphragmatic plication and its effect on the operative outcome.

Patients and Methods:

Design and patients:

This retrospective cohort study included all patients who underwent diaphragmatic plication from June 2010 to June 2017 at King Faisal Specialist Hospital and Research Center, Jeddah, Saudi Arabia. The study was approved by the institutional review board (IRB), and the patients' consent was waived.

The diagnosis of diaphragmatic paralysis was suspected if the patient could not be weaned from mechanical ventilation four days after surgery without any apparent other reason or those who needed re-intubation after initial extubation. Additionally, the elevation of the diaphragm in the postoperative chest x-ray or paradoxical subcostal retraction during spontaneous ventilation is another clue for suspected diagnosis. Fluoroscopy study elucidating the paradoxical movement of the affected side confirmed the diagnosis in all cases, while the patient on spontaneous breathing.

Surgical technique:

Our technique for plication was performed in accordance with that described by Schwartz and Filler [14]. All our diaphragmatic plications were performed through a thoracotomy at the sixth or the seventh intercostal space. The level of posterolateral thoracotomy was guided by the height of the diaphragmatic copula on the chest x-ray. Multiple parallel rows of interrupted 2-0 pledget monofilament sutures were placed in an anterior-to-posterior orientation. The plication sutures were taken primarily in the membranous area of the diaphragm with the intention of bringing the diaphragm down to the point that is 1 to 2 interspaces below its usual level.

Data and outcomes:

We have reviewed the incidence of DP among our patients, demographic criteria, the affected side, and the associated surgical procedure, ventilation time before and after plication, ICU, and hospital stay.

Patients' confidentiality was maintained, removing any identifying information from the data set by the data controller before further usage and analysis. The data were coded into Alpha-numeric format for concealment with few designated persons having the coding key.

Statistical Analysis

Statistical analysis was performed with SPSS statistical program (SPSS 15 Inc., Chicago, IL, USA). The Shapiro-Wilk normality test was used to assess normal distribution. Continuous variables with normal distribution were reported as the mean ± the standard deviation. Continuous data without normal distribution were reported as the median with ranges. Categorical data were presented as number and/or frequency.

Results

Seventy-six patients (2.87%) had unilateral diaphragmatic paralysis following 2646 congenital cardiac procedures from June 2010 to June 2017. Out of those, sixty-four patients (2.4%) underwent diaphragmatic plication. Thirty-six of the patients were males (56.25%), and the median age for plicated patients was 2.75 months (range 0.5- 36) at the time of diagnosis. This is compared with a median age of 3.7 months (range 0.66 to 123) for non-plicated patients. Thirty-six patients were males (56.25%).

Single Ventricle physiology was the most common among the patients who underwent plication (n= 37, 57.8%) with modified Blalock Taussig Shunt (BTS) was the most common procedure associated with DP whether it was done alone or as part of Norwood procedure. On the other hand, all patients with non-plicated diaphragm underwent biventricular repair, and the most common procedure was atrioventricular canal repair. The diagnoses and operations resulting in phrenic nerve injury are listed in (Table 1)

Table 1: The diagnosis and surgical procedure in patients with postoperative diaphragmatic paralysis. Data are presented as number and percent.

Procedure	Total Number of procedures	DP cases
PA banding	189	1 (0.5%)
Blalock-Taussig shunt	264	13 (4.9%)
Norwood with BT shunt	72	4 (5.5%)
Norwood with Sano modification	66	3 (4.5%)
Norwood with Glenn	7	0
Bidirectional Glenn	243	9 (3.7%)
Fontan	179	7 (3.9%)
VSD closure	333	1 (0.3%)
VSD closure, RV to PA conduit	49	1 (2%)
VSD closure, RVOT resection	54	1 (1.85%)
VSD closure, RV to PA conduit, unifocalization	13	2 (15%)
VSD closure, RVOT patch augmentation	203	2 (0.98%)
ASD/VSD closure	102	1 (0.98%)
Aortic arch reconstruction	158	4 (2.5%)
Coarctation repair	247	2 (0.8%)
Arterial switch	295	7 (2.37%)
TAPVR repair	55	2 (3.6%)
Truncus arteriosus repair with RV -PA conduit	64	2 (3.1%)
Unifocalization, unilateral	31	2 (6.45%)
Ross-Konno	23	0
Total cases	2646	64 (2.41%)

DP: diaphragmatic paralysis; PA: Pulmonary artery; BT: Blalock-Taussig; RV: Right Ventricle, RVOT: Right Ventricular Outflow Tract, VSD: Ventricular Septal Defect, ASD: Atrial septal Defect, TAPVR: Total Anomalous Pulmonary Venous Return

Left-sided DP, which needed plication, was reported in 44 patients (68.7%), and right-sided plication was performed in 20 patients (31.25%). The mean time between the primary surgery and diaphragm plication was 6.42±4.51 days. The mean ventilation period before plication was 4.93±3.71 days, and post plication ventilation median time was 2.11±1.82 days. Two patients (3.1%) required tracheostomy for prolonged respiratory insufficiency.

One patient (1.6%) needed revision of the DP after suture disruption, and two patients (3.1%) had their diaphragmatic plication during the initial surgery when diaphragmatic paralysis was anticipated during the operation. (Table 2)

Table 2: Postoperative outcomes after diaphragmatic plication. Data are presented as median, range, mean, and standard deviation.

Parameter	Median (range)	mean± SD
Median age at plication (months)	2.7 (range 0.5-36)	3.53±2.96
Time to do plication (days)	13 (range 3-27)	6.42±4.51
CPAP pre-plication (days)	6 (range 3-11)	4.24±2.45
Ventilation pre-plication (days)	6 (range 2-55)	4.93±3.71
Ventilation post plication (days)	4 (range 1-14)	2.11±1.82
ICU stay (days)	31 (range 8-65)	16.68±7.54
Hospital stay (days)	36 (range 11-80)	19.41±11.22

ICU: intensive care unit; CPAP: continuous positive airway pressure

Discussion

The reported incidence of diaphragmatic paralysis after cardiac surgery in the pediatric age group varies from 0.3% in retrospective studies to 12.8% in prospective researches [6, 7]. The rate of diaphragmatic paralysis required plication in our study was 2.4 %. The higher prevalence noted in prospective versus retrospective studies has several explanations, including increased surveillance in prospective studies to detect asymptomatic cases by routinely performing percutaneous phrenic nerve stimulation to confirm the diagnosis of abnormal phrenic nerve latency [17]. Many children with abnormal phrenic nerve latency do not have clinical symptoms, which are often the criteria prompting investigation and detection in retrospective series and some prospective studies [5].

According to Chan and colleagues [18], phrenic nerve paralysis is frequently under-diagnosed. If the child is asymptomatic, the diagnosis may be missed, while if the child is ventilated, the diagnosis may not be considered, or it is difficult to be confirmed in the ICU setting.

Phrenic nerve injury is less well tolerated in infants and small children than in older children [9, 11, 12, 14, 16]. Several factors contribute to this, including underdeveloped intercostal musculature, greater chest wall compliance, horizontal rib cage orientation, and mediastinal hypermobility [3]. Moreover, the recumbent placement position reduces the vital capacity and facilitates the retention of secretions and obstructive bronchial debris, particularly with the small caliber of the infant bronchial tree. The marked cranial displacement of the flaccid diaphragm in the supine position and the contralateral mediastinal shift result in a significant reduction of the functional residual capacity. This results in alveolar collapse and formation of atelectasis, which will compromise arterial oxygen saturation and increase pulmonary vascular resistance [4].

In our series, infants who underwent DP were younger, with a median age of 2.75 months (range 0.5- 36) at the time of diagnosis. These data are comparable with Baker and colleagues [9] with a median age of 6.81 months (range 0.27–63 months) and that of Van Onna and colleagues [3]. Tonz and colleagues [4] studied the clinical outcomes of 25 patients with phrenic nerve injury after pediatric cardiac surgery, noting that no patient older than 2 years required plication, whereas 7 of 9 patients aged less than 1 year required a procedure. The median age of Non-plicated patients in our study was 3.7 months.

Phrenic nerve injury is more common with the creation or takedown modified Blalock-Taussig shunt, systemic venous to pulmonary artery connection, pulmonary arterioplasty, and arterial switch procedures [4 – 7, 11 – 13]. In our study, Blalock–Taussig shunts were the most common procedure associated with DP, whether it was done alone or as part of the Norwood procedure.

Different mechanisms of phrenic nerve injury in pediatric cardiac surgery have been described. The use of cold solutions within the pericardium has been associated with a hypothermic phrenic injury [19]. Internal jugular vein cannulation is also identified as a possible causative factor [3]. Harvesting large pericardium patch for subsequent intraoperative repair may result in injury [16]. Previous cardiothoracic operations have been reported to increase the risk [6, 11]. The higher risk related to repeated operations is most likely associated with technical difficulties in dissection caused by fibrous adhesions surrounding the phrenic nerve and the use of electrocautery in the direct vicinity of the phrenic nerve [4, 16].

Direct percutaneous stimulation of the phrenic nerve probably provides the most accurate diagnosis and can be applied to patients on mechanical ventilation [17, 20]. We did not use this method as we believe the technique is painful, especially in infants, and the presence of jugular venous cannula can make direct stimulation technically difficult. On the other hand, the ultrasound approach is easy, involves no patient discomfort, and is readily repeatable [7]. In our study, most of our patients either had ultrasound examinations and/or fluoroscopy.

Optimal management of phrenic nerve paralysis in children post-cardiac surgery remains controversial, while some authors [7, 11, 21] advocated an anticipatory approach with long-term ventilatory support.

On the other hand, Tonz and colleagues [4] promoted plication of the diaphragm as earlier extubation reduces the hazards of prolonged mechanical ventilation, the potential risk of ventilator-associated pneumonia, and the increased length of stay [3]. The procedure is safe with low morbidity and does not interfere with the return of normal function [4]. Timing of plication has been a matter of debate, although most authors [4, 6, 7, 12, 14, 16] argue that it is better to withhold diaphragmatic plication for 2 to 3 weeks in anticipation of potential spontaneous recovery of phrenic nerve function. The mean time between the primary surgery and DP in our group was 6.42±4.51 days.

Anatomically, it has been expected the right phrenic nerve to be more vulnerable, and this has been suggested by other authors [12]. In our study, the majority of phrenic nerve injuries requiring diaphragmatic plication occurred on the left side with a ratio near 2 to 1 in favor of a left side. This high incidence of left-sided injuries may be related to extensive thymic resections in primary cases or the mobilization required for arch reconstructions. On the other hand, left side injury may be related to the combined mechanical compressive effects of the heart and the diaphragm. We could extubate our patients with a mean time interval of 2.11±1.82 days after plication; these results were comparable with other reports [3, 22].

Study limitations

The major limitation of the study is the retrospective nature. Many patients with diaphragmatic paralysis may be passed undiagnosed because of the lack of symptoms. Another limitation is the single-center experience, and generalization of the results may be not possible.

Conclusion

Diaphragmatic plication is an effective procedure in the management of postoperative diaphragmatic paralysis. We recommend early plication for patients with symptomatic diaphragmatic paralysis causing prolonged ventilation. Randomized clinical trials are needed to determine the role and optimal timing of diaphragmatic plication.

Conflict of interest: Authors declare no conflict of interest.

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