ORIGINAL ARTICLE

Frequency and treatment of diaphragmatic paralysis after cardiac surgery in pediatric patients

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Background. *Due to phrenic nerve injury after cardiac surgery diaphragmatic paralysis is a serious respiratory complication and leads to a longer hospital stay and even death. They display respiratory distress and delayed extubation. The diagnosis is carried out with a chest X-ray and confirmed with diaphragmatic mobility by fluoroscopy or ultrasound.* ***Objective.*** *To analyze the frequency of diaphragmatic paralysis (DP) in patients with a cardiac surgery history and to evaluate the clinical evolution and treatment.* ***Material.*** *A descriptive, retrospective, observational study of patients with a diagnosis of DP following cardiac surgery from January 2000 to December 2015 was performed.* ***Results.*** *A total of 4,154 heart surgeries were performed, with 39 cases (0.96%) of DP confirmed by fluoroscopy. The most affected groups have been infants with 59%, and weight less than 10 kg with 64%. Concerning the type of heart disease underwent surgery, those with univentricular physiology were the most frequent ones (33%). The most frequent palliative surgeries associated with DP were Blalock-Taussig shunt (35%). The most affected side was the right one (56.4%). Diaphragm plication was performed in 25 patients by thoracoscopy in 84% of them.* ***Conclusions.*** *DP is a rare complication after cardiac surgery, which should be suspected when respiratory support cannot be decreased, with no cardiac or pulmonary cause to justify it. Complications such as recurrent respiratory infections or ventilatory troubles may occur. Early diagnosis and treatment would cut down the number of complications as well as the in-hospital stay and economic costs.*

Key words: *Cardiac surgery; Congenital heart disease; Diaphragmatic paralysis; Diaphragmatic plication.*

Antecedentes. *Debido a la lesión del nervio frénico después de la cirugía cardíaca, la parálisis diafragmática es una complicación respiratoria grave y conduce a una estancia hospitalaria más prolongada e incluso a la muerte. Los pacientes presentan dificultad respiratoria y retraso en la extubación. El diagnóstico se realiza con una radiografía de tórax y se confirma con movilidad diafragmática mediante fluoroscopia o ecografía.* ***Objetivo.*** *Analizar la frecuencia de parálisis diafragmática (PD) en pacientes con antecedentes de cirugía cardíaca y evaluar la evolución clínica y el tratamiento.* ***Material.*** *Se realizó un estudio descriptivo, retrospectivo, observacional de pacientes con diagnóstico de PD tras cirugía cardiaca desde enero de 2000 hasta diciembre de 2015.* ***Resultados.*** *Se realizaron un total de 4,154 cirugías cardíacas, con 39 casos (0.96%) de PD confirmados por fluoroscopia. Los grupos más afectados fueron los lactantes con un 59%, y los de peso inferior a 10 kg con un 64%. En cuanto al tipo de cardiopatía intervenida, las de fisiología univentricular fueron las más frecuentes (33%). Las cirugías paliativas más frecuentes asociadas a la PD fueron la fístula Blalock-Taussig (35%). El lado más afectado fue el derecho (56.4%). La plicatura del diafragma se realizó en 25 pacientes por toracoscopia en el 84% de ellos.* ***Conclusiones.*** *La PD es una complicación poco frecuente tras la cirugía cardiaca, que debe sospecharse cuando no se puede disminuir el soporte respiratorio, sin causa cardiaca o pulmonar que lo justifique. Pueden ocurrir complicaciones tales como infecciones respiratorias recurrentes o problemas ventilatorios. El diagnóstico y tratamiento precoces reducirían el número de complicaciones así como la estancia hospitalaria y los costos económicos.*

Palabras clave: *Cirugía cardiaca; Cardiopatía congénita; Parálisis diafragmática; Plicatura diafragmática.*

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Due to phrenic nerve injury after cardiac surgery diaphragmatic paralysis (DP) is a serious respiratory complication resulting in respiratory failure, pulmonary infections, prolonged hospital stay, and even death, especially at a younger age.

Phrenic nerve injury is a complication described after cardiac surgery. The mechanism may be due to section, traction or elongation of the nerve, ischemia due to manipula-

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tion of the internal mammary artery, electrocautery, local hypothermia at the pericardial level or even placement of thoracic drainage [1]. There is a wide myriad of signs and symptoms going from dyspnea, recurrent cases of pneumonia, chronic bronchitis and chest pain to being asymptomatic or a radiological finding without other alterations [2].

On the other hand, older children can compensate for the loss of diaphragmatic function and usually undergo few or no symptoms. Therefore, diagnosis is difficult. It should be suspected when paradoxical movement of the diaphragm or elevation of a hemidiaphragm is observed on chest X-ray; however, it requires confirmation of mobility testing by ultrasound and/or fluoroscopy during spontaneous breathing. Diaphragmatic plication is the treatment of choice with excellent results [3,4].

MATERIAL

From January 2000 to December 2015, all pediatric patients undergoing cardiac surgery for congenital or acquired heart disease and who developed DP were consecutively included. A total of 4,154 cardiac surgery patients were operated on and 39 cases of DP were found, which corresponds to 0.39%. Data were obtained from the internal cardiovascular surgery files cross-checked with clinical and radiological records.

The diagnosis was made after clinical suspicion of respiratory failure with no other apparent cause, chest X-ray with an elevation of the hemidiaphragm of more than 2 intercostal spaces and confirmed by fluoroscopy where the paradoxical movement was evidenced.

Surgical treatment was indicated for patients with DP, who also presented respiratory distress and the impossibility of weaning from mechanical ventilation. Respiratory distress was defined by the following criteria: tachypnea, oxygen dependence or CO2 retention. The most commonly used surgical approach was minimally invasive thoracoscopy; however, open thoracotomies were also performed in some cases.

Statistics. Descriptive statistics were used, including measures of central tendency and dispersion reported as average, standard deviation, and percentages. Inference statistic SPSS was used for data analysis. X2 test with a confidence interval of 95% and a margin of error of 0.05 ($p<0.05$) were included.

RESULTS

During the period from January 01, 2000 to December 31, 2015, a total of 4,154 heart surgeries were reported, finding 39 cases of DP confirmed by fluoroscopy which represents a frequency of 0.93%. Patients weighing less than 10kg were the most affected with 25 (64.1%) cases, and patients weighing more than 10kg were 14, representing 35.9%. The most affected gender was male with a total of 20 cases (51%). As per the type of heart disease, those of univentricular physiology were the most frequent ones with 13 (33.3%), followed by Tetralogy of Fallot with 5 (12.8%) (**Table 1**), most of which were cyanogenic with 34 (87.2%) cases, while the acyanogenic ones.

Table 1. Type of congenital heart disease.

CARDIOPATHY	FREQUENCY (%)
APCIV	4 (10.3)
APSI	1 (2.6)
AT	2 (5.1)
CANAL	1 (2.6)
CATVP	4 (10.3)
VSD	3 (7.7)
COMPLEX	3 (7.7)
PS	2 (5.1)
FALLOT	5 (12.8)
TGV	1 (2.6)
TRUNK	2 (5.1)
UNI	11 (28.2)
Total	39 (100)

APCIV- Pulmonary atresia with ventricular septal defect, APSI- Pulmonary atresia with an integral septum, AT- tricuspid atresia, CANAL- Atrioventricular canal, CATVP- Total anomalous pulmonary vein connection, VSD- Ventricular septal defect, COMPLEX- this group includes.- Double left ventricular inflow tract, Double atrio-ventricular and ventriculo-atrial discordance, PE- Pulmonary stenosis, FALLOT- Tetralogy of Fallot, TGV- Transposition of great vessels, TRUNK- Truncus arteriosus, UNI- pathologies with univentricular physiology, includes.- Visceral heterotaxy, tricuspid atresia with atrioventricular discordance, Hypoplastic right ventricle, Single ventricle, Tricuspid atresia with pulmonary hypoplasia

Most of the surgeries were palliative procedures (22 patients= 56.4%); Out of them, the more frequent was the systemic-pulmonary shunt (Blalock-Taussig) which was performed in 14 (35.9%), followed by Glenn surgery in 6 (15.4%), and Fontan operation in 2 patients (5.1%). Total correction was performed in 17 cases (43.6%).

Of the 39 patients, the most affected side was the right one in 22 cases (56.4%) while the left side was in 17 patients (43.6%). It should be noted that no bilateral paralysis cases were observed.

All patients had a diagnosis of diaphragmatic paralysis confirmed by fluoroscopy study; 25 patients (64.1%) during

Table 2. Reintubated patients

ITEM	FREQUENCY (%)
No	25 (64.1)
Yes	14 (35.9)
Total	39 (100)

Table 3. Days of mechanical ventilation.

ITEM	1-14 days, n (%)	> 15 days, n (%)	TOTAL, n(%)
No plication	12 (30.8)	2 (5.1)	14 (35.9)
Plication	19 (48.7)	6 (15.4)	25 (64.1)
Total	31 (79.5)	8 (20.5)	39 (100)

the first hospitalization and 14 (35.9%) in a second hospitalization or in the outpatient clinic. The average number of days to establish the diagnosis following cardiovascular surgery was 30 ± 2 days.

The mean number of days requiring mechanical ventilation following cardiac surgery was 0-1 day in 12 patients (30.7%). However, two patients had 33 and 41 days of mechanical ventilation, and 14 patients required reintubation following airway progress (**Table 2**).

Table 3 shows the days of mechanical ventilation between children with diaphragmatic plication and those who did not require it. A p-value of 0.68 was obtained by Fisher's exact test, so there is no difference between the two groups.

Diaphragm plication was performed in 25 patients. The most frequent approach was thoracoscopy in 21 cases (84%) (**Fig. 1**). Eighteen of them (70%) were extubated after surgery. Hospital discharge was at 9.2 ± 8.4 days.

Table 4 shows the differences in days of in-hospital stay between patients who required diaphragmatic plication and those who did not. Using the Chi^2 test, a p-value of 0.98 was obtained. There was no significant difference.

Of the 25 patients underwent diaphragmatic plication, 8 of them were underwent surgery during the first hospitalization, and 17 during a second one. Of the latter ones, the diagnosis was confirmed in 10 cases after their first hospital discharge. Among the causes occurred in the 17 patients and for which they had to be readmitted for diaphragmatic plication were the following: recurrent pneumonia, respiratory distress syndrome, pleural effusion and atelectasis.

DISCUSSION

Congenital heart disease occurs in 2-4% of live births. At early age, corrective or palliative cardiac surgery has high risks and can cause such complications as diaphragmatic paralysis. In fact, phrenic nerve injury is currently the most important cause of diaphragmatic paralysis in children. In this study, the frequency of DP (0.93%) is comparable with other retrospective studies showing a frequency between 0.3% and 12.8% [1,4,5].

The age range most frequently found in patients with diaphragmatic paralysis was the younger infant group, which is consistent with the statement that infants and young children tolerate less diaphragmatic paralysis than older children, as infants depend primarily on diaphragmatic contraction for adequate gas exchange. Its intercostal muscles are weak, and the rib cage has a more horizontal orientation. In addition, the mediastinal structures show greater contralateral mobility on inspiration. The paralyzed diaphragm paradoxically moves with negative intrapleural pressures, which decreases the functional residual capacity, increasing the chance of atelectasis [6]. Similar to other studies, the disease was more prevalent in males.

The most frequent heart diseases were those with univentricular physiology, followed by Tetralogy of Fallot. Most of them have a cyanogenic heart disease history (87.2%) as Akbariasbagh et al. showed in their study, with a frequency of 68% for cyanogenic heart disease [6]. Most were palliative procedures, the most frequent being the systemic-pulmonary fistula (modified Blalock Taussig), which was performed in 14 patients (35.9%). However, the total correction was performed in 17 patients (43.6%), which is similar to the results of other series. The results are due to the fact that corrective surgeries of the cardiac defect require open heart surgery; consequently, structural changes are made to repair the present defect as opposed to palliative surgeries, but especially the proximity of the phrenic nerve to the dissected structures, increasing the risk of phrenic nerve damage. Previous cardiac surgery is another factor that increases the risk of diaphragmatic paralysis. In our study, 8 patients had had previous surgery [7]. In this study the most affected side was the right one in 22 cases (56.4%).

As reported in the literature, the main associated complications were infectious and ventilatory problems such as recurrent atelectasis and pleural effusion. We observed that there is no significant difference in reduction of complications in patients with diaphragmatic plication and those who did not require it. However, the group that underwent plication had the highest number of complications. Diaphragmatic plication is the treatment of choice for diaphragmatic paralysis, especially in children < 1 year of age. However, there is still controversy as to the best time to perform it. Some au-

Table 4. Days of in-hospital stay.

ITEM	1-15 days, n (%)	16-30 days, n (%)	31-45 days, n(%)	46-60 days, n(%)	> 60 days, n (%)	TOTAL, n(%)
No plication	7 (17.9)	2 (5.1)	2 (5.1)	1 (2.6)	2 (5.1)	14 (35.9)
Plication	13 (33.3)	4 (10.3)	4 (10.3)	2 (5.1)	2 (5.1)	25 (64.1)
Total	20 (51.3)	6 (15.4)	6 (15.4)	3 (7.7)	4 (10.3)	39 (100)

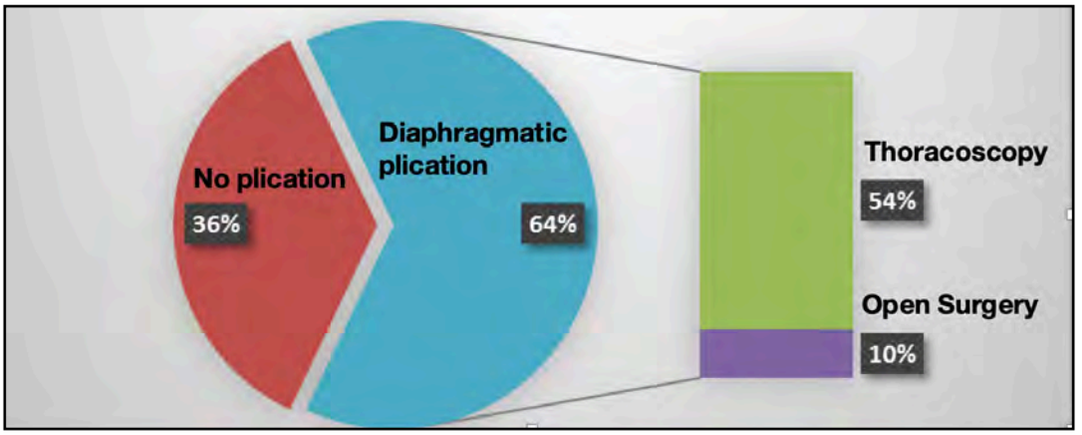

Figure 1. Patients who required diaphragmatic plication and the different type of approach.

thors suggest that plication should be performed as soon as paralysis is diagnosed, while others recommend waiting 1 to 6 weeks to get a spontaneous recovery. However, the success of late plication may be threatened by diaphragmatic atrophy. Many authors suggest that the decision for plication should be based on the patient respiratory status. In our study, the mean number of days from cardiac surgery to diaphragmatic plication was 30 ± 10 days (range, 17 to 165 days), longer than that reported in other series such as Bravo et al. with a mean of 3 days [8]. Regarding the impact of plication on mechanical ventilation time, there are reports describing a reduction after plication. In our study we did not find statistically significant differences in the time of mechanical ventilation between one group and another (due to the sample size), but we did observe that the patients who required plication were those who had more days of mechanical ventilation compared to the group that never underwent surgery. We also observed that after plication, the majority (70%) achieved immediate extubation after surgery and the rest only required mechanical ventilation for a maximum of 3 days. As for the days of hospital stay, the difference was not significant, observing that the patients who required plication were those who already had more days of in-hospital stay [9,10].

As a conclusion, the frequency of diaphragmatic paralysis in this study was low. However, it is comparable to previously reported in the literature. The above indicates that the incidence in our hospital is low or there is a significant number of undiagnosed cases or difficulties in diagnosis due to the presence of a non-specific clinical picture or confusion with another nosological entity.

DP is a relatively rare complication of cardiac surgery in infancy, which should be suspected when a patient respiratory support cannot be decreased without cardiac or pulmonary cause. After diagnosis, diaphragmatic plication should be considered if extubation is not possible within 2 weeks, especially if the patient is less than 1 year of age.

The long-term clinical evolution is generally favorable despite the persistence of paralysis, showing such complications as recurrent respiratory infections, mainly pneumonia or ventilatory problems, atelectasis, and pleural effusion. A very low mortality of 2.5% (one patient) was observed, which was due to causes not related to DP.

Although the differences in mechanical ventilation, days of in-hospital stay, and presence of complications were not statistically significant, we do observe that there is a trend for patients to have a greater number of complications, require more time on mechanical ventilation, and a longer hospital stay, which makes them candidates for plication.

Therefore, a timelier diagnosis and a more expeditious plication would probably further reduce the number of complications, which would result in a shorter hospital stay, faster recovery, and earlier hospital discharge.

The major limitation of the study was the lack of information contained in the records in addition to being an underdiagnosed entity due to the non-specific clinical manifestations of the patients that can be confused with other diagnoses, which may influence the low frequency observed.

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