



## Ultrasonographic postoperative evaluation of diaphragm function of patients with congenital heart defects

*Doğuştan kalp defektli hastalarda ameliyat sonrası ultrasonografi ile diyafram fonksiyonunun değerlendirilmesi*

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### ABSTRACT

**Background:** This study aims to investigate the role of ultrasonography in the postoperative evaluation of diaphragm function in patients with congenital heart defect.

**Methods:** This prospective study included a total of 360 patients (176 males, 184 females; mean age 2 years; range, 1 month to 8 years) who underwent congenital heart surgery and 44 patients (22 males, 22 females; mean age 1 years; range, 1 month to 4 years) who underwent diaphragm ultrasonography between September 2018 and March 2019. Ultrasonography was performed for the patients who had difficulty in weaning from mechanical ventilation or who were thought to have diaphragm dysfunction due to pathological findings on postoperative chest X-rays. The findings were interpreted as normal, paresis, or paralysis.

**Results:** Diaphragm dysfunction was demonstrated in 23 patients (6.3%), paralysis in 11 patients (3%), and paresis in 12 patients (3.3%). A median sternotomy was performed in 21 patients (91%), and seven of them (30%) were redo cases. Five patients (21%) had single ventricle physiology. Six patients (1.6%) needed an intervention due to diaphragm dysfunction. The interventional procedures were diaphragm plication in three patients (0.8%) and tracheotomy in three patients (0.8%). Three of these patients had a single ventricle and three had biventricular physiology. The median time after surgery for these procedures was 36 days. One patient (0.2%) died in the intensive care unit. The mean length of stay in the intensive care unit and hospital was 36±12 and 48±21 days, respectively.

**Conclusion:** Diaphragm dysfunction should be kept in mind in patients undergoing congenital heart surgery and in those who need prolonged intubation during the postoperative period. Ultrasonography is a non-invasive diagnostic tool which can be used to identify diaphragm dysfunction and the best course of management of this clinical condition.

**Keywords:** Congenital heart disease, diaphragm dysfunction, ultrasonography.

### ÖZ

**Amaç:** Bu çalışmada doğuştan kalp defektli hastalarda ultrasonografinin ameliyat sonrası diyafram fonksiyonunun değerlendirilmesindeki rolü araştırıldı.

**Çalışma planı:** Bu prospektif çalışmaya Eylül 2018 - Mart 2019 tarihleri arasında doğuştan kalp cerrahisi yapılan toplam 360 hasta (176 erkek, 184 kadın; ort. yaş 2 yıl; dağılım, 1 ay-8 yıl) ve diyafram ultrasonografisi yapılan 44 hasta (22 erkek, 22 kadın; ort. yaş 1 yıl; dağılım, 1 ay-4 yıl) alındı. Mekanik ventilatörden ayrılmada zorluk yaşayan veya ameliyat sonrası akciğer grafisinde patolojik bulgulara göre diyafram disfonksiyonu düşünülen hastalara ultrasonografi yapıldı. Bulgular normal, paresis veya paraliz şeklinde yorumlandı.

**Bulgular:** Yirmi üç hastada (%6.3) diyafram disfonksiyonu, 11 hastada (%3) paraliz ve 12 hastada (%3.3) paresis görüldü. Hastaların 21'ine (%91) median sternotomi yapıldı ve bunların yedisi (%30) yeniden ameliyat olgusu idi. Beş hastada (%21) tek ventrikül fizyolojisi vardı. Altı hastaya diyafram disfonksiyonu nedeniyle girişim yapıldı. Girişimsel işlemler üç hastada (%0.8) diyafram plikasyonu ve üç hastada (%0.8) trakeostomi idi. Bunların üçünde tek ventrikül ve üçünde çift ventrikül fizyolojisi vardı. Ameliyat sonrası bu işlemlerin medyan uygulanma zamanı 36 gün idi. Bir hasta (%0.2) yoğun bakım ünitesinde kaybedildi. Ortalama yoğun bakım ünitesi ve hastanede kalış süresi sırasıyla 36±12 ve 48±21 gün idi.

**Sonuç:** Doğuştan kalp cerrahisi yapılan hastalarda ve ameliyat sonrası uzamış entübasyona ihtiyacı olan kişilerde diyafram disfonksiyonu akla getirilmelidir. Ultrasonografi, diyafram disfonksiyonunu saptamada ve klinik durumun en iyi şekilde yönetilmesinde kullanılabilen noninvaziv bir tanı aracıdır.

**Anahtar sözcükler:** Doğuştan kalp hastalığı, diyafram disfonksiyonu, ultrasonografi.

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The diaphragm is the main respiratory muscle in infants and children and aids roughly 75% of respiration. Pneumonia, collapse of the lungs, and atelectasis can develop in case of unilateral or bilateral diaphragm dysfunction in congenital heart surgery (CHS) patients. Morbidity and mortality can also increase as a result of difficulty in weaning from mechanical ventilation due to diaphragm dysfunction.<sup>[1,2]</sup>

The incidence of diaphragm dysfunction due to phrenic nerve injury during cardiothoracic operations has been reported as 0.3 to 12.8% in different studies. Injuries vary from paresis to paralysis, preventing standardized follow-up and management in these patients.<sup>[3]</sup> There are different diagnostic tools used to evaluate the diaphragmatic functions. Radiological imaging, fluoroscopic evaluation, and neural conduction studies are the primary methods to establish the diagnosis.<sup>[4]</sup>

Ultrasonography (USG) is being used increasingly in the cardiac intensive care unit (ICU). There are studies reporting that the evaluation of lung pathologies and diaphragm dysfunction with USG may be beneficial, particularly during the postoperative period, as USG can be performed easily and rapidly.<sup>[4-6]</sup>

In the present study, we aimed to evaluate the effects of USG in determining disturbances in diaphragm functions and management of this pathology in postoperative patients in our cardiac surgery center.

## PATIENTS AND METHODS

This prospective study was conducted at Istanbul Mehmet Akif Ersoy Thoracic and Cardiovascular Surgery Training and Research Hospital between September 2018 and March 2019. A total of 360 patients (176 males, 184 females; mean age 2 years; range, 1 month to 8 years) who underwent CHS and 44 patients (22 males, 22 females; mean age 1 years; range, 1 month to 4 years) who underwent diaphragm USG were included. *Inclusion criteria were as follows:* having uncontrolled excessive breathing efforts, low oxygen saturation, and hypoxemia (lower than expected saturations for cyanotic patients) with conventional or non-conventional positive pressure ventilation methods; having re-intubation within the first 24 hours of extubation; and having diaphragm elevation or atelectasis in the absence of abdominal swelling. Those who were older than 18 years or with a history of premature birth, congenital lung pathology, known neuromuscular disease, and suspected diaphragmatic dysfunction before the operation were excluded from the study. Routine chest X-rays were performed in each patient every morning

after the operation and during their ICU stay. An echocardiographic evaluation was performed on the day of planned extubation to visualize any residual hemodynamic defects, and diaphragm functions were evaluated with concurrent USG. A written informed consent was obtained from each patient. The study protocol was approved by the Istanbul Mehmet Akif Ersoy Thoracic and Cardiovascular Surgery Training and Research Hospital Ethics Committee. The study was conducted in accordance with the principles of the Declaration of Helsinki.

The USG assessment of the diaphragm and echocardiographic evaluation was performed by a single pediatric cardiac intensivist skilled in USG/echocardiographic examinations using a Vivid S5 (GE, Vivid S5, Norway) 6-MHz transducers. The inspiratory and expiratory changes, amplification heights, and movement of the diaphragm in the M-mode views at the subxiphoid region (in the transverse axis at 3 o'clock) and the right and left eighth and ninth intercostal spaces throughout the midaxillary line (in the sagittal axis at 12 o'clock) were used for evaluation of the right and left hemidiaphragms. The results were divided into the following three main categories: (i) Normal (diaphragm moves upward with a flexion wave toward the liver or caudally, the movement amplitude is greater than 4 mm, and the difference between the hemidiaphragm domes is less than 50%); (ii) Paresis (the movement amplitude toward the liver or spleen is less than 4 mm, or the difference between the hemidiaphragm domes is more than 50%); and (iii) Paralysis (lack of movement or paradoxical movement).<sup>[2]</sup>

For each case, a study form was completed including gender, age, weight, cardiac diagnosis, presence of genetic anomalies, risk category of the surgical procedure, follow-up in ICU, and the results of the USG evaluation. The type and diagnosis of congenital anomalies were also noted. Operative data included lesion and repair types by the Risk Adjustment for Congenital Heart Surgery-1 (RACHS-1) risk category<sup>[7]</sup> and Aristotle Complexity Score (ACC).<sup>[8]</sup>

### Statistical analysis

Statistical analysis was performed using the PASW for Windows version 17.0 software (SPSS Inc., Chicago, IL, USA). Continuous variables were expressed in mean  $\pm$  standard deviation (SD) or median (min-max) values, while categorical variables were expressed in number and frequency. Demographic features and perioperative variables were compared by the Mann-Whitney U and

chi-square tests. A *p* value of <0.05 was considered statistically significant.

## RESULTS

A total of 360 patients were included in the study. Demographic clinical characteristics and perioperative diagnosis of the patients are shown in Table 1.

Forty-four patients (12%) underwent a screening for diaphragmatic USG. The diaphragmatic USG indications were non-cardiac respiratory distress in 28/44 (64%), hemi-diaphragmatic elevation on chest X-ray in 20/44 (45%), the need for post-extubation positive pressure in 19/44 (43%), extubation failure in 10/44 (23%), and physical examination in 10/44 (23%) patients.

Diaphragmatic dysfunction was observed in 23/44 (52%) patients. The type of diaphragmatic dysfunction was paresis in 12 patients (bilateral paresis: n=1, right-sided paresis: n=4, left-sided paresis: n=7) and paralysis in 11 patients (bilateral paralysis: n=2, left-sided paralysis: n=6, and right-sided paralysis: n=3). Among the patients with diaphragmatic dysfunction, left-sided diaphragm dysfunction was observed in 70% of patients, and right-sided diaphragm dysfunction was observed in 43% of patients. Twenty-three patients were diagnosed with diaphragmatic dysfunction with

an incidence rate of 6.4% in our overall postoperative surgical cases.

Fluoroscopic evaluation (n=13) was performed in all patients with paralysis (n=11), in one patient with bilateral paresis (recurrent intubation) (n=1), and in one patient with right-sided paresis (recurrent intubation and physical examination findings) (n=1). Fluoroscopy was not performed in cases of paresis and normal diaphragmatic USG findings due to the rapid recovery of the patients' clinical conditions.

Fluoroscopic evaluation revealed diaphragm dysfunction in 10 patients, one with paralysis with bilateral paresthesia, and one with right-sided paresthesia according to the diaphragmatic USG evaluation. The fluoroscopic evaluation was normal in a patient with a left-sided paralysis as assessed by USG. This patient was one-month-old male infant who had arch reconstruction with median sternotomy. Recurrent atelectasis occurred at the left lung. Recovery was seen at serial USG scans and the patient was uneventfully discharged at the postoperative fourth week.

All patients who had diaphragmatic dysfunction were under three years of age with a mean age and mean weight of 9±2.7 months and 5±2.2 kg,

**Table 1. Demographic characteristics and perioperative diagnosis of patients**

	n	%	Median	Range
Diaphragmatic ultrasound number	44/360	12		
Gender				
Male	176	49		
Female	184	51		
Age (month)			4	0.1-96
Weight (kg)			5	2.5-52
RACHS-1				
Undefined	8	2.1		
Category 1	40	11.3		
Category 2	151	42.1		
Category 3	105	29.3		
Category 4	43	12.2		
Category 5	-	-		
Category 6	13	3.0		
Aristotle comprehensive complexity				
Level I	35	9.9		
Level II	103	28.6		
Level III	92	25.4		
Level IV	130	36.1		

RACHS: Risk adjusted classification for congenital heart surgery.

**Table 2. Demographic characteristics of patients undergoing operation due to diaphragmatic paralysis**

Case	Diagnosis	Age (day)	Fluoroscopy result	Diaphragmatic ultrasound diagnosis	Interventional procedure	Intervention time (day)	Outcome
1	TGA	210	Paradox	Bilateral paralysis	Plication*	32	Discharged
2	TAPVC	60	Paradox	Left paralysis	Plication†	40	Discharged
3	DILV	80	Paradox	Bilateral paralysis	Plication†	36	Discharged
4	AVSD	180	Akinesia	Right paresis	Tracheostomy	34	Discharged
5	VSD-arcus hypoplasia	40	Paradox	Left paresis	Tracheostomy	42	Discharged
6	LAI-mitral atresia	45	Akinesia	Bilateral paresis	Tracheostomy	40	Died

TGA: Transposition of the great arteries; TAPVC: Total anomalous pulmonary venous connection; DILV: Double inlet left ventricle; AVSD: Atrioventricular septal defect; VSD: Ventricular septal defect; LAI: Left atrial isomerism; \* Bilateral; † Unilateral-left sided.

respectively. The surgical procedure was performed via median sternotomy in 21/23 (91%) of the patients. Thirty percent of the patients underwent at least one open heart surgery previously. Eighteen (79%) of the patients had biventricular physiology, and seven of 23 patients (21%) had univentricular physiology.

The diagnosis of cardiac pathology and operation type were as follows: Aortic arch repair was performed due to aortic arch hypoplasia + ventricular septal defect or aortic coarctation in seven patients, interrupted aortic arch in two patients, arterial switch operation + aortic arch repair due to Taussig-Bing anomaly in two patients, total surgical repair due to atrioventricular septal defect (AVSD) in two patients, total surgical repair due to tetralogy of Fallot (ToF) in four patients, arterial switch operation due to transposition of the great arteries in two patients, primary sutureless surgical repair due to total anomalous pulmonary venous connection in two patients, single ventricle palliation procedures (Glenn operation: n=2, Fontan operation: n=1) in three patients, and left atrial aneurysmectomy due to left atrial aneurysm in one patient. Eleven (47%) of the patients who had diaphragmatic dysfunction underwent aortic arch repair.

Six patients (1.6% of all cases) needed surgical interventions due to diaphragmatic dysfunction within a median of 36 (range, 32 to 44) days after open heart surgery. Fluoroscopy showed different degrees of diaphragmatic motion defects in all patients. Half of the patients had single ventricle and the remaining three had biventricular physiology. Our approach was case oriented in our study. Three patients underwent diaphragmatic plication (one patient bilateral, two patients unilateral), whereas the other three patients underwent tracheostomy. Table 2 summarizes the

characteristics of the patients who underwent additional surgical interventions.

One patient died in the ICU. The mean duration of the ICU and hospital stays was 36±12 days and 48±21 days, respectively.

## DISCUSSION

In the present study, we evaluated the role of USG evaluation in the diagnosis of diaphragm dysfunction after CHS. Our results demonstrated that USG was a useful diagnostic modality which could detect different diaphragmatic movement pathologies. Although diaphragm dysfunction was observed after a variety of surgical procedures, its incidence was significantly higher after aortic arch surgery. To the best of our knowledge, this subject has been previously examined in a very limited number of studies.

Phrenic nerve damage is a recognized complication of cardiac surgeries and leads to diaphragm dysfunction. It can be induced by amputation, crushing, stretching, or thermal nerve injury. The reported incidence of diaphragmatic dysfunction has shown variation according to the symptomatic status of the patient. Previous studies have found that the incidence ranges between 4.1 and 12.8%.<sup>[2,9,10]</sup> In our study, the incidence was 6.4% which is consistent with previous studies.

Diaphragm paralysis can be observed after various CHS procedures. Akbariasbagh *et al.*<sup>[11]</sup> suggested that the Fontan procedure, arterial switch, and Blalock-Taussig shunt operation had the highest (16.6% in each) incidence for diaphragm paralysis. Akay *et al.*<sup>[12]</sup> also found the highest incidence rate of diaphragm paralysis after total correction surgery for ToF, Blalock-Taussig shunt, ventricular septal defect

closure, and pulmonary artery patch plasty. However, Hamadah et al.<sup>[2]</sup> reported the highest rate after arterial switch operations and aortic arch repair. Our findings are in consistent with the findings reported by Hamadah et al.<sup>[2]</sup>

In the literature, there are controversial reports about the affected side in previous studies. Akbariasbagh et al.<sup>[11]</sup> reported that the right diaphragm was more commonly affected, whereas Lemmer et al.<sup>[9]</sup> and Areola et al.<sup>[10]</sup> contradicted this finding. Our results are consistent with the Lemmer and Areolas' findings.<sup>[9,10]</sup>

It is well-known that redo surgeries increase the risk of phrenic nerve injury during the dissection of adhesions related to previous surgeries. Accordingly, the reported incidence of diaphragm dysfunction after redo surgery is between 9 and 49% in the literature.<sup>[2]</sup> In our study, 30% of diaphragm paralysis cases developed after redo surgeries.

Chest fluoroscopy, USG, and electromyography are among the most utilized diagnostic modalities in diaphragm dysfunction. In children with CHS, the use of electromyography is limited due to technical difficulties and difficulties accessing equipment and experienced staff.<sup>[4,13]</sup> Chest fluoroscopy has long been considered the gold standard for the diagnosis of diaphragm paralysis; however, it necessitates transporting the patient to the radiology theatre and resulting exposure to ionizing radiation. The benefits of utilizing USG in the emergency departments and ICUs have been supported by several studies. Due to the increased use of USG in critical care settings, bedside diaphragm USG is emerging as a simple, practical, and non-invasive method for quantifying diaphragmatic movement in a variety of settings.<sup>[6,14]</sup> In our study, we emphasized the importance of bedside USG in detecting diaphragmatic dysfunction.

There is no consensus on when or how to intervene in cases of diaphragmatic dysfunction,<sup>[15-17]</sup> Some authors have suggested that this period should be allowed to pass with general supportive treatment, considering that phrenic nerve damage may show spontaneous recovery within one to six weeks.<sup>[5,10]</sup> On the contrary, Talwar et al.<sup>[18]</sup> recommends a plication procedure as soon as significant diaphragmatic dysfunction is diagnosed. Hamadah et al.<sup>[2]</sup> proposes that plication should be performed, if successful weaning cannot be performed within two weeks in patients with a single ventricle physiology or in patients younger than four months old. Tracheostomy has been advocated in some reports in patients whose phrenic nerve recovery is significantly prolonged,

although it has also been associated with significant mortality.<sup>[1,3]</sup>

In our series, six patients required an interventional procedure. In all patients, at least four-week recovery period was allowed. In each patient, treatment plan was individualized. We believe that age, body weight, associated cardiac disease, the respiratory muscle strength, serial diaphragm USG, and duration of assisted ventilation may be helpful in decision making for tracheostomy and plication.

The main limitation of the present study is that it includes our clinical experience of a limited number of cases from a single center. Other limitations are that USG was unable to be compared with other diagnostic methods such as fluoroscopy and electromyography in patients with diaphragm dysfunction and inter-observer evaluation was unable to be performed.

In conclusion, patients undergoing prolonged intubation after cardiac surgery without any residual hemodynamic defects may suffer from diaphragm dysfunction. Ultrasonography, which is an easy and quick imaging technique, should be considered a useful method for detecting diaphragmatic dysfunction and managing associated clinical conditions.

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