Long-term functional evaluation of diaphragmatic motility after repair of congenital diaphragmatic hernia

Francesco Arena¹,*, Carmelo Romeo³, Maria Pia Calabrò⁴, Pietro Antonuccio¹, Salvatore Arena¹, Giuseppe Romeo¹

¹Unit of Pediatric Surgery, Department of Medical and Surgical Pediatric Sciences, University of Messina, Italy
²Unit of Pediatric Cardiology, Department of Medical and Surgical Pediatric Sciences, University of Messina, Italy

Abstract
Background/Purpose: A few studies have taken into account the diaphragmatic function in patients successfully treated for congenital diaphragmatic hernia (CDH). Monodimensional sonography has been reported to be useful in assessing the diaphragmatic motility. Aim of the present study was to investigate, in a long-term follow-up, the diaphragmatic function after CDH repair.

Methods: Ten patients, with a mean age of 16 (5-26) years, were enrolled. All had had a left diaphragmatic hernia repaired, but no one received a patch. Ten subjects of matched age were used as controls. The diaphragmatic excursions appear, at M-mode sonography, as a sinusoid; the amplitude of the curve on the vertical axis measured the movement in centimeters. Chest x-ray and spirometry were also performed in CDH patients.

Results: A reduced diaphragmatic motility on the left (treated) side was recorded. The amplitude of the contraction was significantly reduced when compared with the contralateral side (1.19 ± 0.2 cm vs 2.33 ± 0.9 cm; P = .017) and was also significantly reduced in comparison with the motion of the left side of controls (1.19 ± 0.2 cm vs 1.83 ± 0.4 cm; P = .01). There was no difference in the amplitude of contraction between the left and right sides of control patients and between the right side of CDH patients and the controls. Spirometry was normal in all patients but one, who had a slight reduction of ventilation on the left side.

Conclusion: M-mode sonography appears as a very useful tool in quantitative evaluation of diaphragmatic movements and should be extensively used during follow-up of patients after CDH repair. Motility of the repaired diaphragmatic is reduced, even after a long period, but this does not affect the respiratory function in patients who survived CDH repair.

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Congenital diaphragmatic hernia (CDH) is one of the most serious neonatal emergencies. Mortality is still high, despite the newly developed perioperative therapeutic strategies, and is mainly related with the degree of pulmonary hypoplasia.

A few studies have been focused on the incidence of long-term problems in patients successfully treated for CDH [1,2]. Attention has been paid particularly to postoperative impairment of respiratory function or to gastroesophageal reflux, whereas diaphragmatic function after operation has been scarcely investigated [3,4].
Aim of the present research was to assess, with M-mode sonography, the diaphragmatic motility after CDH repair.

1. Material and methods

Forty patients with CDH were treated at the Paediatric Surgery Unit of University of Messina (Italy) from 1972 to 1997. The male/female ratio was 1:1.2 (18 male patients, 22 female patients). In 36 patients, the defect was on the left side and in 4 patients on the right. CDH was diagnosed during the neonatal period between the first and sixth days of life; no one had a prenatal diagnosis. Surgical repair was performed through a subcostal or a midline abdominal incision. The overall survival rate was 55%. Sixteen patients were followed up; 10 of them, with a mean age of 16 (5 to 26) years, agreed to enter the study. All had a left diaphragmatic hernia repaired, but no one received a patch. After informed consent, patients underwent chest x-ray, spirometry, and diaphragmatic ultrasonographic examination.

The echocardiogram study was performed in the supine position, using an Aloka SSD 2200 machine (Aloka Co. Ltd, Tokyo, Japan) and a multifrequency probe (5-2.5 MHz). All patients were examined in the fasting state, after a 15-minute bed rest. No one was assuming drugs potentially able to affect diaphragmatic motion. To rule out a structural heart disease, each patient had been previously submitted to clinical cardiac evaluation including electrocardiogram and echocardiogram. Ten subjects of matched age (15.2, range 6-30 years), free from cardiovascular disease, agreed to have the diaphragm studied during a routine echocardiography performed for the evaluation of an “innocent” murmur and were used as controls.

All ultrasonographic examinations have been performed by the same investigator (M.P.C.). Using a subcostal approach, the diaphragm was first identified through a bidualimensional view, also on the basis of its respiratory movements and higher echogenicity with respect to surrounding tissues. The ultrasonic beam was directed in such a way to visualize the maximal excursion of the diaphragm, and an M-mode tracing was recorded during normal breathing; a further recording was obtained during forced respiratory movements. The left hemidiaphragm was examined first, and then the right one. The M-mode diaphragmatic ultrasonogram shows a sinusoidal undulating shape, where during inspiration, the muscle echocardiogram is displaced toward the transducer and moves away from it during expiration. The diaphragmatic excursion was measured in centimeters as the distance between the lower and the higher point of the curve (Fig. 1). Measurements were performed during the examination, directly on the screen, using electronic calipers. In any study, 3 values, obtained at 1-minute interval from each other, were averaged.

In both groups (patients and controls), the mean value and SD of left and right diaphragmatic movements recorded during normal and during forced breathing have been calculated; the statistical significance of differences between groups has been evaluated by means of Student t test for unpaired data.

2. Results

All patients had a normal life-style, and no one complained of respiratory symptoms. One male patient was practicing competitive sport, and another one was a saxophone player. Three women had multiple uneventful pregnancies, and 1 of these underwent cesarean section.

The chest x-ray revealed a reduction in the left lung volume in 2 cases and scoliosis with slight mediastinal displacement in a female patient who had 2 uncomplicated pregnancies followed by natural delivery. There was, in addition, 1 case of pectus carinatum (the saxophone player). The profile of the left diaphragmatic dome appeared normal in 7 cases, whereas it was flat and stretched in 1 patient, and in 2 cases appeared to be lower than the contralateral side.

Spirometry parameters, such as forced vital capacity, forced expiratory volume in 1 second, maximal mid-expiratory flow, were normal in all patients but one, who had a slight reduction of ventilation on the left side.
M-mode sonography disclosed, in patients with CDH, a reduced diaphragmatic motility on the left (treated) side: the contraction amplitude was significantly reduced when compared with the right side (1.19 ± 0.2 vs. 2.33 ± 0.9 cm; \( P = .017 \)). Moreover, the left diaphragm contraction amplitude of CDH patients was significantly reduced in comparison with the amplitude of the left side in controls (1.19 ± 0.2 vs. 1.83 ± 0.4 cm; \( P = .01 \)). There was, however, any difference in contraction amplitude between the left and the right side of controls (1.87 ± 0.4 vs. 2.03 ± 0.5 cm) as well as between the right side of CDH patients and the right side of controls (2.33 ± 0.9 vs. 2.03 ± 0.5 cm). When diaphragmatic motion was investigated during forced breathing, contraction amplitude in patients increased by 106.7% on the left side and by 105.3% (\( P = .007 \)) on the right side. Similar increases were observed in controls 98.5% and 97.8% on the right and left sides, respectively (\( P = .002 \)).

3. Discussion

The evaluation of diaphragmatic motility is very important in some clinical conditions, including repaired CDH. Until recently, however, the available diagnostic methods were either invasive, such as a radioscopy, or just provided an indirect evaluation of diaphragm motion, such as transdiaphragmatic pressure study or respiratory pletysmography. In 1991, Heyman et al [5] studied the effect of theophylline on diaphragmatic motility in neonates using M-mode sonography. More recently, B/M-mode sonography coupled to spirometry has been used to assess the diaphragmatic function [6-9]. M-mode sonography records in real time the diaphragmatic excursions as a sinusoid. The vertical axis of the curve measures the height of the movement in centimeters; the horizontal axis, the duration.

Our study shows that, several years after operation for CDH, the repaired diaphragm has a reduced motion, when compared with the contralateral or with the diaphragm of healthy controls. The lowest level of diaphragmatic excursion was observed in patients showing distortion of both the diaphragm and the thoracic cage, associated with severe thorax asymmetry in one case and with hypoplastic left lung in the other one. Patients with good pulmonary development, in contrast, had normal or near-normal diaphragmatic excursion. Similar data on diaphragmatic motion, evaluated with M-mode sonography in patients operated for CDH, have been reported by Fasching et al [4], who observed a 72% of normal findings in the affected side, when compared with the contralateral side.

Despite a reduced motility, the diaphragm of our patients operated for CDH maintains a good contractility during forced respiration, as demonstrated by motion amplitude increase by 106.7%. The increase was virtually identical to that observed in the contralateral unaffected side or in controls. These data help explaining the multiple pregnancies with uncomplicated natural delivery in 2 female patients, as well as the fact that one male patient is a professional athlete; and another one, a saxophone player. In all the above conditions, the diaphragm is forced to produce a very high effort.

In particular, labor and delivery represent a challenge for the diaphragm: Nava et al [10] suggested that this muscle fatigues significantly during the expulsive phase of labor, when intra-abdominal pressure may exceed 150 cm H2O. Rupture of the diaphragm during this acute fatigue has been occasionally reported in patients with repaired diaphragm for CDH [11]. In our patients, however, the overall respiratory function was normal, and the diaphragm succeeded in increasing its work despite the mild motion impairment present in basic conditions.

Similarly has been reported that CDH survivors can participate in competitive sports after an adequate period of training and periodic monitoring of maximum oxygen consumption [12,13].

In conclusion, M-mode sonography appears as a very useful and promising tool in quantitative evaluation of diaphragmatic movements and should be extensively used during follow-up of patients submitted to CDH repair. Our data suggest that, in patients who survived operation for CDH, the motility of the repaired diaphragm is reduced, but this does not affect significantly the respiratory function.

References


