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Kraaijenga, J.V.S.

Citation for published version (APA):
Kraaijenga, J. V. S. (2017). Diaphragmatic electromyography monitoring in preterm infants

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CHAPTER 3

Diagnosis of hemidiaphragmatic paresis in a preterm infant with transcutaneous electromyography: a case report

J.V. Kraaijenga
G.J. Hutten
F.H. de Jongh
A.H. van Kaam

ABSTRACT

Transcutaneous electromyography of the diaphragm (dEMG) is a non-invasive and easy applicable tool to measure the electrical activity of the diaphragm. dEMG monitoring has recently been introduced in the neonatal intensive care unit as a novel cardiorespiratory monitor providing direct information on the diaphragmatic breathing activity. We report a preterm infant with suspected paresis of the right diaphragm measured with transcutaneous dEMG, which showed a clear reduction in the electrical activity of the right-sided diaphragm. In conclusion, dEMG provides valuable information on regional diaphragmatic activity, which can assist the clinician in diagnosing hemidiaphragmatic paresis.

KEY WORDS
Diaphragm; Paralysis; Diaphragm function; Premature neonates; Electromyography

Established Facts
- Diaphragmatic paresis is diagnosed by chest X-ray and ultrasound, but these diagnostic tools have their limitations.

Novel Insights
- Transcutaneous diaphragmatic EMG (dEMG) provides separate and simultaneous information on right and left electrical diaphragmatic activity.
- dEMG could be an alternative and more direct method of diagnosing (hemi) diaphragmatic paresis in newborn infants.
INTRODUCTION

The diaphragm is the most important inspiratory muscle in preterm infants and generates, assisted by the intercostal muscles, the required force to maintain tidal breathing.\(^1\) Diaphragmatic paresis is a condition that is characterized by muscular weakness of the diaphragm with phrenic nerve injury following traumatic delivery as the most common cause.\(^1,2\) Diaphragmatic paresis is usually unilateral, predominantly affecting the right side. Unilateral diaphragmatic paresis can lead to respiratory distress symptoms such as tachypnea, cyanosis, a weak cry, or apnea resulting in a risk for mechanical ventilation.\(^2\)

The most common used tools to diagnose diaphragmatic paresis are an anteroposterior chest X-ray (CXR) and fluoroscopy, usually showing an elevated (hemi)diaphragm.\(^1,2\) More recently ultrasound of the chest has been used to diagnose diaphragmatic paresis by detecting the absence of diaphragmatic movements during spontaneous breathing.\(^2,3\) However, these diagnostic tools have their limitations, such as exposure to radiation, not applicable at the bedside, or false-negative results due to paradoxical movements of the affected hemidiaphragm during tidal breathing governed by the healthy hemidiaphragm.\(^1,2\)

Measuring the (regional) electrical activity of the diaphragm using transcutaneous electromyography (dEMG) could be an alternative and more direct method of diagnosing (hemi)diaphragmatic paresis in newborn infants. This report describes, for the first time, dEMG recordings in a preterm infant with suspected unilateral paresis of the diaphragm.

Case report

The index patient was a male preterm infant, born at 28 weeks of gestation by caesarean section with a birth weight of 1020 gram. Because of respiratory distress and increasing oxygen need, he was intubated and started on high-frequency ventilation. The first CXR showed signs of respiratory distress syndrome for which he was treated with two doses of exogenous surfactant. No asymmetry of the left and right diaphragm was noticed. The patient was transferred back to nasal continuous positive airway pressure on day 3 of life but still showed signs of severe respiratory distress. The second CXR demonstrated an elevated diaphragm on the right side resulting in a reduced lung volume compared to the left lung (Figure 1). Ultrasound of the diaphragm did show movement of the right diaphragm during spontaneous breathing, but this seemed to be less pronounced than the left side. Based on these results congenital right-sided diaphragmatic paresis was diagnosed. The patient was managed on nCPAP until the age of 8 weeks without showing any clinical and radiological improvement. For this reason a surgical plication of the right diaphragm was performed after which the patient could be weaned from respiratory support and was discharged home at the age of 4 months.
Transcutaneous dEMG measurement
Transcutaneous dEMG for measuring electrical activity of the diaphragm is currently being evaluated for clinical use in our unit. This allowed us to apply dEMG in the index patient after obtaining written informed consent from both parents. The measurement was performed on day 6 of life at which time the infant was supported by a nasal continuous positive airway pressure of 6 cmH\textsubscript{2}O and a fraction of inspired oxygen of 0.25. The patient was placed in supine position with the head in midline and diaphragmatic activity was measured for 30 min. Two electrodes were bilaterally placed at the costoabdominal margin in the nipple line (frontal diaphragm) and two bilaterally at the back at the same height (dorsal diaphragm). The common electrode was placed at height of the sternum. Diaphragmatic activity at right and left side was derived from the sum signal of the frontal and dorsal electrodes at each side. The dEMG electrodes were connected to a portable 16-channel digital reference amplifier (Dipha-16, Inbiolab BV, Groningen, Netherlands) and pre-processed dEMG data were send wireless to the front-end of the Dipha-16 system at a portable computer. Details on dEMG measurements have been described elsewhere. Data processing and analysis were performed using the software package Polybench (version 1.25.2, Applied Biosignals, Weener, Germany).

To compare the amount of electrical activity of the left and right hemidiaphragm, we calculated the mean dEMG amplitude (µV) at both sides by subtracting the lowest electrical activity from the maximal electrical activity of each breath over a 60-second period. Following this initial measurement we performed two dEMG recordings to assess changes in dEMG activity over time.

Figure 1. CXR of the index patient showing an elevated right diaphragm resulting in a reduced lung volume compared to the left lung.
RESULTS

dEMG measurement was performed without any difficulties and was well tolerated by the index patient. Figure 2 shows a representative 60-second recording of the averaged dEMG signals across the right and left diaphragm. It clearly shows a strongly reduced electrical activity of the right diaphragm (amplitude 0.8 µV) compared to the left side (amplitude 1.57 µV). This difference in electrical activity between the right and left side was observed over the whole 30-min measurement. These findings did not change over time.

Figure 2. Tracing of the dEMG signal showing the averaged electrical activity (μV) of the right (R dEMG) and left (L dEMG) diaphragm of the index patient. The time window is 60 sec. Diaphragmatic activity at the right side (amplitude 0.8 μV) is strongly reduced compared to the left side (amplitude 1.57 μV), corresponding with the abnormalities observed on the CXR.

DISCUSSION

To our knowledge, this is the first report describing transcutaneous dEMG in a preterm infant with suspected hemidiaphragmatic paresis. This report illustrates that dEMG monitoring can provide separate information on the electrical activity of the left and right diaphragm during spontaneous breathing and that this information can be used to complete the diagnosis of (hemi)diaphragmatic paresis.

CXR is the most commonly used tool to diagnose hemidiaphragm paresis, typically showing an elevated diaphragmatic contour on the affected side. However, the use of CXR also has limitations. First, it repeatedly exposes the patient to radiation, because most often multiple CXR are needed to assess if there is spontaneous resolution of the paresis over time. Second, CXR may lead to both false-positive and false-negative results. A study in adults showed that the prevalence of an elevated unilateral hemidiaphragm on the CXR was 64%, while the actual percentage of unilateral paralysis, as judged by phrenic nerve stimulation, was only 24%. As also illustrated by our index patient, false-negative results may be present during mechanical ventilation, when the use of high(er) mean airway pressures may attenuate the elevation of the diaphragm on the affected site. Chest fluoroscopy is not often used in (preterm) infants because it cannot be applied at the bedside and requires a relatively high radiation dose.

In some centers, ultrasound of the diaphragm is used as an alternative or additional diagnostic tool for diagnosing diaphragmatic paresis. Although its main advantage is not
exposing patients to radiation, it also has several limitations. First, diaphragmatic ultrasound requires special expertise, which may not always be available in every center. Second, it does not allow for simultaneous visualization of both hemidiaphragms, which may make it difficult to assess differences in movement in case the affected side is not completely paralytic. Finally, ultrasound might lead to a false-negative result in case of paradoxical movement of the affected hemidiaphragm, i.e. upward moving during inspiration and downward movement during expiration.

In general, most infants with diaphragmatic paresis after birth trauma recover spontaneously within 6-12 months and treatment is mostly supportive. Surgical plication is performed if severe respiratory distress persists or if the infant requires prolonged mechanical ventilation.

As illustrated by this case report, dEMG has the potential to overcome all of the aforementioned shortcomings of CXR, chest fluoroscopy and ultrasound. It is easy to use at the bedside, is radiation-free, and provides direct, online and continuous information on the electrical activity of the left and right diaphragm. It was interesting to observe that the right diaphragm in this index patient did show some detectable electrical activity during inspiration and expiration. There may be several explanations for this finding. First, this might reflect some residual activity in the affected diaphragm. Second, this electrical activity may originate from other inspiratory muscles such as the left diaphragm and the intercostal muscles (e.g. crosstalk). This finding emphasizes the importance of visualizing the diaphragmatic activity on both sides simultaneously during dEMG. It is important to emphasize that reduced electrical activity of the diaphragm as measured by dEMG is not pathognomonic for diaphragmatic paresis and may have several other causes.

In conclusion, this report shows that transcutaneous dEMG monitoring can provide additional information on the electrical activity of the right and left diaphragm, which makes dEMG an ideal candidate for diagnosing hemidiaphragmatic paresis in neonates.
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