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Case Report

Case Series Connection between both Phrenic Nerves

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Introduction

The diaphragm is the main respiratory muscle. Although it is a single muscle, each hemidiaphragm is innervated by a phrenic nerve. Therefore, there is a right phrenic nerve and a left phrenic nerve with different anatomical variations. This nerve comes from a branch of the cervical plexus, originating from the anterior motor roots of C3, C4 and C5, with occasional contribution of C2 or C6. Its main contribution comes from the anterior cervical motor root C4. There is a so-called accessory phrenic nerve, in 30% of cases the C5 fibers descend directly from the interior of the thorax, crossing in front of the internal mammary artery, before joining the nerve itself [1]. They appear on the lateral margin of the anterior scalene muscle, at the height of the upper edge of the thyroid cartilage [2], crossing it and reaching the internal part of the base of the neck where the intrathoracic route would begin.

The left phrenic nerve enters the thoracic cavity between the common carotid and subclavian arteries, crossing laterally to the internal mammary artery (IMA). During its descent, it is situated between the pericardium and the mediastinal pleura (Figure 1), passing in front of the pulmonary hilum region and branching out into three muscular branches (sternal, anterolateral and posterior or crural), innervating the left hemidiaphragm. The right phrenic nerve has a shorter and more vertical intrathoracic route, also entering laterally to the IMA, running through the pleural cavity on the lateral surface of the superior vena cava, crossing in front of the pulmonary hilum to reach the central tendon of the diaphragm, branching in the same way and innervating the ipsilateral hemidiaphragm.

Figure 1: a) Braided needles for stimulation and registration. 1b) Double headed arrow. Example of braided needles inserted in right hemidiaphragm.

IMA provides at least 73% of the blood supply to the nerve [3] through the pleural, pericardial and diaphragmatic vessels. Diaphragmatic dysfunction is one of the major problems that can occur during the postoperative period of cardiothoracic surgery, making it difficult or impossible to extubate patients, often having to resort to a tracheotomy with the consequent burden of associated comorbidity and prolonged hospital stay. In turn, respiratory parameters are also altered in unilateral diaphragmatic lesions with a decrease of e.g. maximum inspiratory pressure (MIP) up to approximately 60% and maximum transdiaphragmatic pressure around 40% [4].

During a descriptive pilot study to analyze the incidence of diaphragmatic dysfunction in patients undergoing lung transplantation with intraoperative phrenic nerve monitoring, due to a needle placement error, a potential for muscular action was recorded by stimulating the left phrenic nerve while the right hemidiaphragm was being recorded. After this event, the response of the contralateral diaphragm when stimulating both phrenic nerves was monitored to check whether this potential was a single or universal case and whether it was uni or bilateral.

Material and Methods

The 9 patients signed an informed consent form to be included in this study, describing in detail the examinations to be performed and their purpose. This study was approved by the hospital's CEIC (Clinical Research Ethics Committee) of the Puerta de Hierro University Hospital in Majadahonda in January 2018 in accordance with current legislation (Law on Biomedical Research of 3 July 2007), considering that its overall approach was acceptable from a methodological and ethical point of view and in compliance with the regulations on handling personal data.

In addition to intraoperative monitoring, three electromyographic studies were performed on the eight patients who underwent lung transplantation: one pre-transplant study and two post-transplant follow-up studies (one month and six months after the operation). The thymoma patient was only monitored intraoperatively during the thymoma resection in May/2020. These individuals had intraoperative phrenic nerve monitoring. The following is a description of the technique used.

MOTOR STIMULATION to obtain intraoperative CMAP. Concentric braided needles of 18 mm (sgm medical monitoring) (Figure 1a) were used to stimulate in the neck after the sternocleidomastoid muscle at the point where the best response in amplitude and latency was obtained. 18 mm needles (sgm medical monitoring) for men and 13 mm needles (sgm medical monitoring) for women were used as recording needles (Figure 1b) because of the different thickness of the intersex diaphragmatic musculature. The needles were placed in the muscle under direct vision by the surgeon close to the midline and with the greatest separation between them in order to obtain the greatest possible motor amplitude. The stimulation parameters were pulse widths of 400-500 msec and intensities between 80-120 mA to obtain the potential with the maximum amplitude. This stimulation was carried out under anesthetic conditions of continuous infusion of muscle relaxant at a dose of 0.1 mg/kg/hour. An 8-channel ISIS IOM express device was used to monitor the CMAP. A patient was included in the study who underwent a post-surgical study after resection of an epithelioid hemangioendothelioma block along with a subclavian vein segment and 2 cm of phrenic nerve at that level (Figures 2 and 3). This patient underwent a post-surgical study only with an electromyographic exploration of both hemidiaphragms and stimulation of both phrenic nerves to obtain the compound muscle action potential with needle recording in diaphragmatic musculature. Below we detail the technique performed.

Figure 2: Tumour resection specimen.

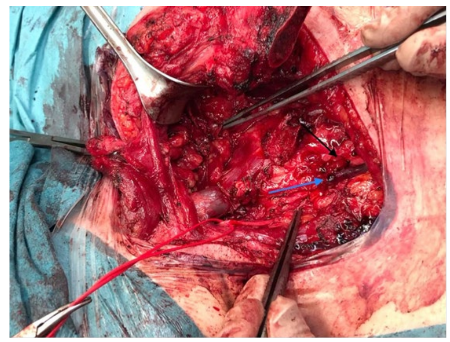

Figure 3: Hemangioendothelioma case. Surgical field after tumor en-bloc exeresis. Black arrow: distal end of phrenic nerve; Blue arrow: left mammary vein.

MOTOR STIMULATION to obtain CMAP and needle EMG in the post-surgical study. The electrocardiographic examination was performed with a 37 mm coaxial needle in both hemidiaphragms. Once the muscle was located, ipsilateral nerve stimulation was performed at neck level. Stimulation parameters were pulse width of 200 ms and intensities between 40-80 mA. A 10 channel Medelec Sinergy 2005 version 14.0 (VYASIS healthcare UK Ltd.) was used.

Statistical Analysis

We have not performed statistical analysis because of the small sample size and because the objective of this study is to reflect a casual finding.

Results

In the 9 patients monitored intraoperatively, a CMAP was obtained in both hemidiaphragms by stimulating both nerves and recording in both hemidiaphragms. The attached images of 2 patients (Figures 4 and 5) show examples of four CMAP: two when stimulating the left phrenic nerve (one recording in right hemidiaphragm and another recording in left hemidiaphragm) and two when stimulating the left phrenic nerve recording in left and right hemidiaphragm. There are two signals, one in green which corresponds to the initial basal signal at the beginning of the stimulation, and another in white which is the last signal obtained. Some of the CMAP obtained appear with inverted polarity with respect to the other potential recorded in the contralateral hemidiaphragm. This is due to differences in the input channel of the braided recording needles or the change in polarity when stimulating the neck level (negative or positive). Table 1 describes the parameters of conduction of the CMAP: distal latency and amplitude with the same stimulation parameters.

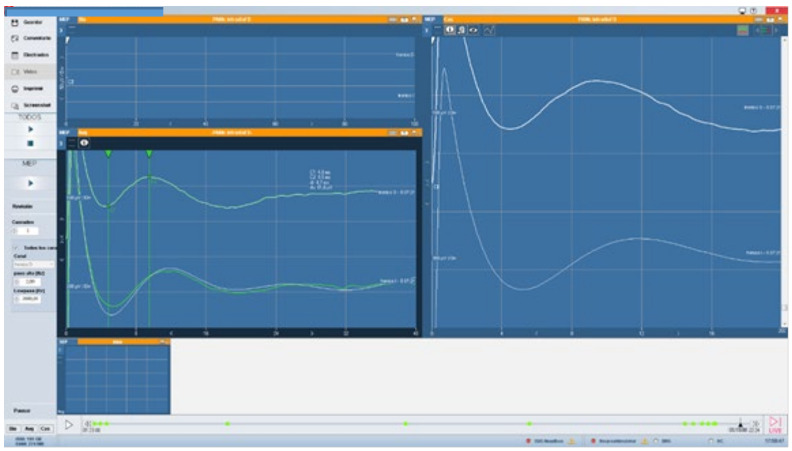

Figure 4: Example of RSRR and RSLR (First case).

In the patient in the post-surgical study during electromyographic examination of the left hemidiaphragm, the presence of motor unit potentials during inspiration was observed, suggesting diaphragmatic origin. By stimulating the left phrenic nerve and recording in the ipsilateral hemidiaphragm no compound muscle action potential was obtained. By stimulating the contralateral phrenic nerve, a low amplitude but reproducible compound muscle action potential was obtained (Figure 7). By stimulating the right phrenic nerve and recording in the ipsilateral diaphgram a CMAP of 1,2 mV was obtained.

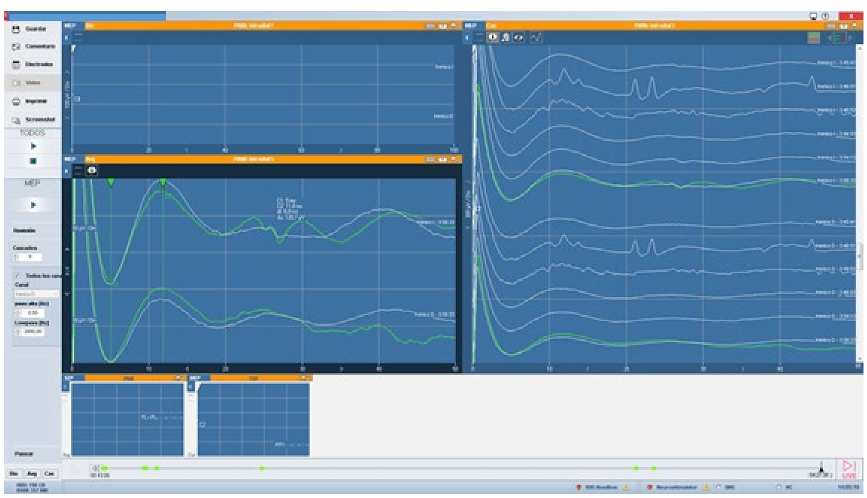

Figure 5: Example of SLRL and SLRR (Ninth case)

	RSRR latency	**RSLR latency**	**RSRR amplitude**	**RSLR amplitude**	**SLRL latency**	**SLRR latency**	**SLRL amplitude**	**SLRR amplitude**
First case	4,8 msg	5,2 msg	83 µV	195 µV	5 msg	5,3 msg	107 µV	782 µV
Second case	5,1 msg	5,1 msg	1,7 mV	406 uV	5,2 msg	5,2 msg	428 µV	614 µV
Third case	5,1 msg	5,1 msg	317 µV	1,1 mV	5,2 msg	5,2 msg	164 µV	753 µV
Fourth case	5,2 msg	5,2 msg	96,7 µV	70,2 µV	5,1 msg	4,9 msg	417 µV	253 µV
Fifth case	5,3 msg	4,9 msg	202 µV	202 µV	4,6 msg	4,9 msg	215 µV	373 µV
Sixth case	4,8 msg	4,8 msg	157 µV	565 µV	4,9 msg	4,9 msg	183 µV	865 µV
Seventh case	5 msg	5 msg	452 µV	35, 6 µV	5,4 msg	5,4 msg	239,2 µV	553 µV
Eighth case	5,2 msg	5,2 msg	282 µV	223 µV	5, 3msg	5,1 msg	248,7 µV	309,2 µV
Ninth case	4,9 msg	4,9 msg	534 µV	114 µV	5 msg	5 msg	120 µV	99 µV

RSRR: right phrenic nerve stimulus and recording in right hemidiaphragm; RSLR: right phrenic nerve stimulus and recording in left hemidiaphragm; SLRL: stimulation in the left phrenic nerve and recording in the left hemidifragm; SLRR: stimulation in the left phrenic nerve and recording in the right hemidifragm.

Table 1: Describes the parameters of conduction of the CMAP: distal latency and amplitude with the same stimulation parameters

In the immediate postoperative period after tumor resection, the patient presented clinical and radiological signs compatible with diaphragmatic dysfunction (dyspnea with effort and decubitus and elevation of the left hemidiaphragm in the chest X-ray). Three months after surgery, the respiratory clinic began to improve in particular the dyspnea in decubitus which makes us think that it is not due to the participation of the of the accesory breathing muscles.The attached images show the chest x-ray in the immediate postoperative (Figure 6). Also attached is an image of the action potential obtained by stimulating the right phrenic nerve (Figure 7).

Figure 6: Chest X-ray 3 days' post-surgery. Left hemidiaphragm elevation.

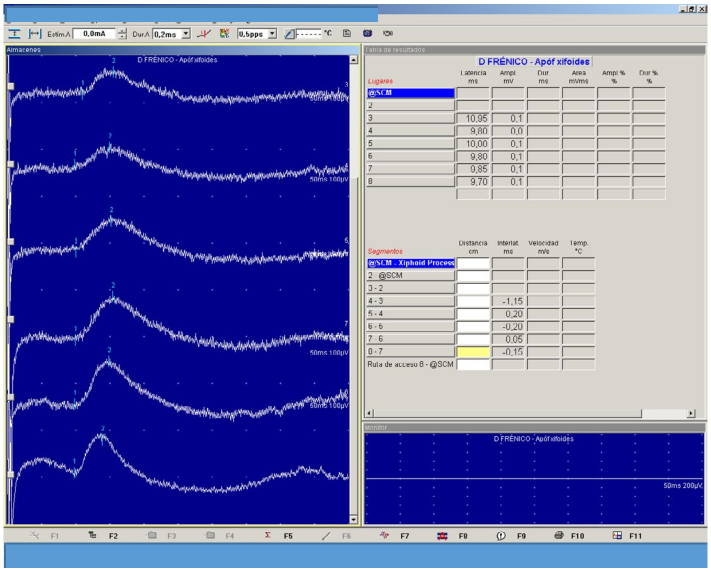

Figure 7: CMAP Post-Surgical Vascular Tumor Study. CMAP stimulating right phrenic nerve recording left hemidifragm.

Discussion

In the 9 patients who were monitored intraoperatively the similarity of the values obtained in the distal latencies between RSRR (right phrenic nerve stimulus and recording in right hemidiaphragm) and RSLR (right phrenic nerve stimulus and recording in left hemidiaphragm) and SLRL (stimulation in the left phrenic nerve and recording in the left hemidifragm) and SLRR (stimulation in the left phrenic nerve and recording in the right hemidifragm) and the technique used (needles are placed in the muscle under direct vision) suggest the presence of a connection between both nerves. The fact that these responses are reproducible and constant would also incline us to think so. Another possible explanation is that the phrenic nerve partially innervates the contralateral diaphragm directly without connecting with the contralateral phrenic nerve through any of its branches, although the similarity of the distal latency values obtained, which are even the same in some cases, makes us incline towards the first of the options (Sur connection). If it were a direct innervation, we would expect a delay in latency in all of them, although we cannot rule out this possibility completely.

As for the amplitude of the responses, this is a less stable parameter than the previous one, since, in this type of surgery, fluctuations in their values are frequent, due to traction and compression during the surgical manipulation of neighboring structures. In addition, depending on the placement of the needles and the separation between them, the amplitude values may be higher or lower. The values obtained in amplitude and latency are within the range of possible values that can be obtained in electromyographic studies performed in the laboratory by stimulating the phrenic nerve and recording in the ipsilateral hemidiaphragm in ambulatory patients [5].

Initially, although the latency values obtained were very similar and even the same in many cases, there was also reasonable doubt that it was a connection at a central level through the bulb (crossed phenomenon) [6], already described, that was responsible for the recording of this compound muscular action potential in the contralateral hemidiaphragm. After the post-surgical study of the patient with the vascular tumour we are inclined to think that there is a peripheral connection, we do not know if it is unique or in addition to the existing central level, at an intrathoracic level not previously known and which I have called the Sur connection. The clinical improvement presented by the patient would also support this hypothesis. This fact would open up the possibility of using this connection therapeutically in diaphragmatic dysfunctions due to phrenic nerve neuropathy, and even avoid in some cases the need for nerve reconstruction or diaphragmatic plication in cases where it is necessary to section or remove the nerve [7,8].

We assume that by stimulating the nerve unharmed we could somehow rehabilitate or achieve a relatively functioning contralateral hemidiaphragm that would palliate the negative effects at the respiratory level of severe diaphragmatic paralysis and the risks derived from diaphragmatic plication. We do not know if stimulation of this connection or partial innervation would improve diaphragmatic function and what type of technique would be ideal, but the improvement in the clinical situation gives us relative hope that it was one of these hypotheses that has improved the patient's symptoms. In order to determine this, studies in patients with this type of sequelae will be necessary. This work has limitations, it is a series of cases with an important selection (patients on waiting lists for lung transplants mostly), the number of patients is not very large and all records have been made by a single researcher. With respect to the intra-surgical technique for obtaining the potential for action due to the anesthetic conditions and the fact that the nerve is in the depth the stimulation parameters have a high pulse width and intensity. Given the proximity between the contralateral nerve and the point of stimulation, it is possible that they were stimulated by continuity. For this reason, the outpatient post-surgical study is very important, in which the stimulation parameters are much lower and in this case the left phrenic nerve cannot be stimulated by continuity [9-16].

Conclusion

With these 10 cases we believe we can infer the presence of a peripheral connection between both nerves or a partially inervation of each hemidiaphragm from the contralateral phrenic nerve not previously known. The real existence of this connection or partial innervation should be the subject of further studies trying to add imaging tests to prove it. If this finding is confirmed, new therapeutic possibilities open up for cases of diaphragmatic dysfunction due to phrenic nerve injury.

Acknowledgment

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References

1. Larkin FC. (1889) Accessory phrenic nerve. J. Anat. Physiol 23: 340.
2. Fell SC. (1998) Surgical anatomy of the diaphragm and the phrenic nerve. Chest Surg Clin N Am 8: 281-294.
3. O'Brien JW, Johnson SH, VanSteyn SJ, et al. (1991) Effects of internal mammary artery dissection on phrenic nerve perfusion and function. Ann Thorac Surg 52: 182-188.
4. Laroche CM, Mier AK, Moxham J, Green M. (1988) Diaphragm strength in patients with recent hemidiaphragm paralysis. Thorax 43: 170-174.
5. Resman-Gasperc A, Podnar S. (2008) Phrenic nerve conduction studies: technical aspects and normative data. Muscle nerve 37: 36-41.
6. Zaki Ghali M G. (2017) The crossed phrenic phenomenon. Neural Regen Res. 12: 845-864.
7. Celik S, Celik M, Aydemir B, Tunckaya C, Okay T, et al. (2010) Long-term results of diaphragmatic plication in adults with unilateral diaphragm paralysis. J Cardiothorac Surg. 5: 111.
8. Macchiarini P, Dartevelle P, Chapelier M, Lenot B, Cerrina J, et al. (1993) Technique for Resecting Primary and Metastatic Nonbronchogenic Tumors of the Thoracic Outlet. Ann Thorac Surg 55: 611-618.
9. Studera SM, Levyb RD, McNeilc K, Orensd J B. (2005) Resultados clínicos del Trasplante de pulmón: revisión sobre supervivencia, función del órgano trasplantado, aspectos fisiológicos, calidad de vida relacionada con la salud y rentabilidad. Eur Respir J 6: 40-53.
10. Sheridan PH, Cheriyan A, Doud J, Dornseif S E, Montoya A, et al. (1995) Incidence of Phrenic Neuropathy after Isolated Lung Transplantation. J HEART LUNG TRANSPLANT 14: 684-691.
11. Kline DG, Hudson AR. (1995) Nerve Injuries: Operative Results for Major Nerve Injuries, Entrapments, and Tumors. 611.
12. Gardnera I H, Dohertyb G M, and McAnenyb D. (2016) Intraoperative nerve monitoring during thyroid surgery. Curr Opin Endocrinol Diabetes Obes 23: 394-399.
13. de Danschutter S J , Schreinemakers J M , Smit L H, van der Laan L, Nuytinck H K. (2015) Thyroid surgery and the usefulness of intraoperative neuromonitoring, a single center study. J Invest Surg. 28: 86-94.
14. Franceschi F, Koutbi L, Gitenay E, Hourdain J, Maille B, et al. (2015) Electromyographic Monitoring for Prevention ofPhrenic Nerve Palsy in Second-Generation Cryoballoon Procedures. Circulation: Arrhythmia and Electrophysiology 8: 303-307.
15. Kai H, Xixia L, Miaoyun L, Qinchang C, Xinzhi P, Dingyuan L. (2017) Intraoperative nerve monitoring reduces recurrent laryngeal nerve injury in geriatric patients undergoing thyroid surgery. Acta Otolaryngol. 137: 1275-1280.
16. Alberto Pérez de Vargas, Francisco Isidro Mesas, Anwar Saab, Edwin Ebrat Mancilla, Marta vaquero Martinez, Luis Lopez Pajaro, Andrés Varela de Ugarte, David Gómez de Antonio.(2021) Intraoperative Monitoring using NAP (Neurogenic Action Potential). Neurol Neurosurg Curr Res. Volume 2 Issue 1