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Clinical Letter

BiPAP as a Possible Treatment in Patients With Diaphragmatic Flutter

To the Director,

We present a 38-year-old woman with a history of relapsing-remitting multiple sclerosis, diagnosed one year earlier, under treatment with natalizumab (300 mg IV monthly). She consulted for episodes of sudden chest tightness and dyspnea for two weeks, presenting three to four episodes per day. Two in the early morning and two in the late afternoon, lasting approximately 5–8 min. During the crises, rhythmic involuntary movements were observed in the trunk, predominantly on the right. Magnetic resonance imaging, immunological/hormonal/microbiological study, endoscopy, electroencephalogram and laryngoscopy were performed; the results were normal. Electromyography showed grouped activity at 4 Hz in the right hemidiaphragm, coinciding with the clinical episode. And videofluoroscopy (Video 1) showed repetitive movements of the right hemidiaphragm. With movements of the left hemidiaphragm secondary to the right contractions. The diagnosis of diaphragmatic flutter (DF) was established.

The pain produced by the contractions was refractory to several lines of analgesic treatment including: fentanyl, metamizol, baclofen, cannabidiol, paroxetine, lorazepam, methadone, and gabapentin. In addition to pulsed radiofrequency and botulinum toxin, administration of both phrenics. The possibility of phrenic nerve block was raised, but the patient rejected this possibility. Therefore, mechanical ventilation (MV) was started with a Trilogy EVO device (Philips, Murrsville, PA, USA) with two settings: programming one, with preventive mode, which she used in the afternoon and during sleep. And programming two, with therapeutic modality, which he used during the episodes and the first hour after the episodes. This reduced the intensity and the number of crises to two per day. This made it possible to reduce the dose of analgesics. She only needed fentanyl (100 mg PO) and diazepam (5 mg PO) during the episodes. Clinical stability was achieved and discharge was decided, maintaining MV, presenting to date only one crisis in the mornings.

DF can be secondary to alterations of the central and peripheral nervous system, such as encephalitis and irritation of the phrenic nerve. Although in this patient, there was no multiple sclerosis-related brainstem involvement on magnetic resonance. The response obtained with MV could be explained by several mechanisms. By influence of EPAP, by affecting expiratory time.^{1,2} Or by alteration of the Hering–Breuer reflex. After initiating MV, repetitive stimulation of the respiratory center would cause habituation and desensitization of this reflex.^{2,3} Studies indicate

that, under these conditions, pontine stimulation would reduce the amplitude of the phrenic discharge by >50%.^{2,3}

On the other hand, the respiratory frequency is variable. This is because the respiratory musculature synchronizes its activity to 33–50% of that of the DF.⁴ This variability in respiratory rate can alter gas exchange due to variations in tidal volume, and cases of hypercapnic and/or hypoxemic respiratory failure have been reported.^{4,5} Although, with contraction frequencies ≥ 4 Hz, adequate gas exchange can be maintained, as was the case here. Therefore, the use of MV in this pathology could reduce diaphragmatic contractions by allowing deeper inspirations and reducing expiratory times.

Confidentiality of data

The authors declare that they have followed their institution's protocols on the publication of patient data.

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Contribution of each author

DELP and FJCS provided the possibility of mechanical ventilation in this patient. FJCS and SIBA were responsible for the patient's follow-up. SIBA is responsible for the preparation of this document. The preparation of this document was supervised by DELP and FJCS.

Conflict of interests

The authors state that they have no conflict of interests.

Use of artificial intelligence to generate texts

The authors declare that they have not used any type of generative artificial intelligence in the writing of this manuscript or for the creation of figures, graphs, tables or their corresponding captions or legends.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at [doi:10.1016/j.arbres.2024.06.011](https://doi.org/10.1016/j.arbres.2024.06.011).

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