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A case of the diaphragmatic flutter with an electromyographic study of the respiratory muscles

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A 28-year-old female was complained with dyspnea and involuntary rhythmic movements in her chest and upper abdomen. Diagnosis of the diaphragmatic flutter was established since high frequency intermittent discharges of the respiratory muscles superimposed on her ordinary respiratory activities. The origin of these abnormal discharges may be in the central nervous system and the psychosomatic factor is participating in the development of the diaphragmatic flutter.

Key words : diaphragmatic flutter, central nervous system, psychosomatic disorder, behavioral control of breathing

INTRODUCTION

Diaphragmatic flutter is a rare disorder characterized by rapid involuntary contractions of the diaphragm superimposed on ordinary respiratory excursions [1, 2]. An electromyographic study to show the abnormal electric discharges of the diaphragm and other respiratory muscles to make a definite diagnosis and to identify the origin of diaphragmatic flutter [3].

We describe a female patient with diaphragmatic flutter, who did not show any organic lesions and the electromyographic study suggests that the abnormal activities of the respiratory muscles may have originated from the central nervous system. Since the psychosomatic status played a significant role in the development of diaphragmatic flutter in this patient, this may be a disorder in the behavioral control of breathing.

CASE REPORT

A 28-year-old female, housewife, was referred to Tokai University Hospital because of dyspnea and involuntary movements in the epigastrium. She was in good health until she delivered her second baby. Thereafter, she was holding the problem in infant nursing and frequently complained of anorexia. Two months before a visit to our hospital, she suddenly felt involuntary motions in the epigastrium associated with dyspnea that then recurred on almost a weekly basis. She was referred to our hospital and at that time, she was poorly nourished with weighting 42 kg. High frequency bilateral oscillatory movements were observed in her chest and upper abdomen during both inspiration and expiration. Involuntary movements were not seen in her hands and legs. The attacks usually continued for 1 or 2 hours. She could speak and move her extremities normally during attacks. She did not complain of muscle weakness and numbness. Arterial blood gases on room air revealed pH 7.373, $PaCO_2$ 31.6 mm Hg, PaO_2 110 mm Hg and HCO_3^- 17.8 mmol/l. Blood cell counts were normal. Blood chemistry including electrolytes and creatine phosphokinase did not show any abnormalities. There were no abnormalities in her chest roentgenogram and MRI of the head and neck. Although we could not record her electroencephalogram we recorded

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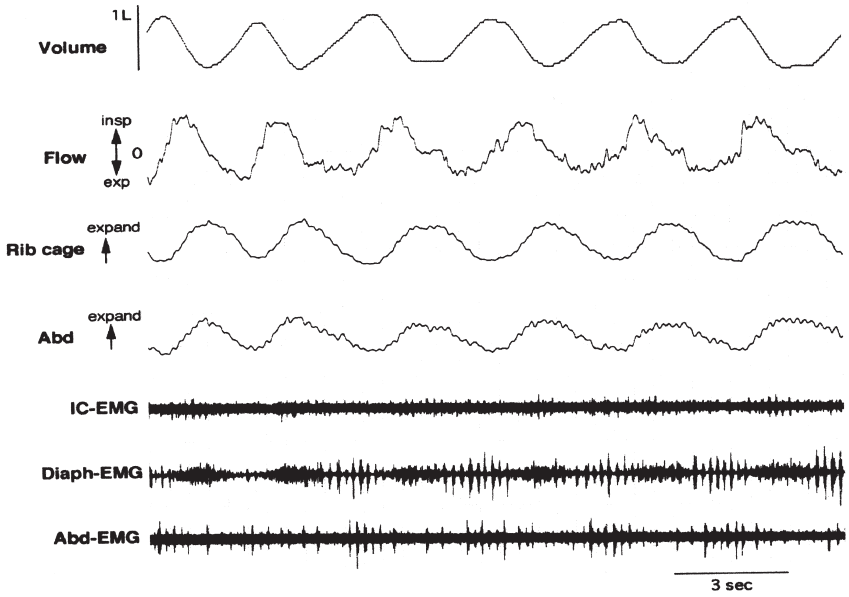

Fig. 1 A record during an attack. Volume; expiratory volume of respiration, Flow; respiratory flow, Rib cage; rib cage movement, Abd; abdominal movement, IC-EMG; intercostal electromyogram, Diaph-EMG; diaphragmatic electromyogram, Abd-EMG; abdominal electromyogram.

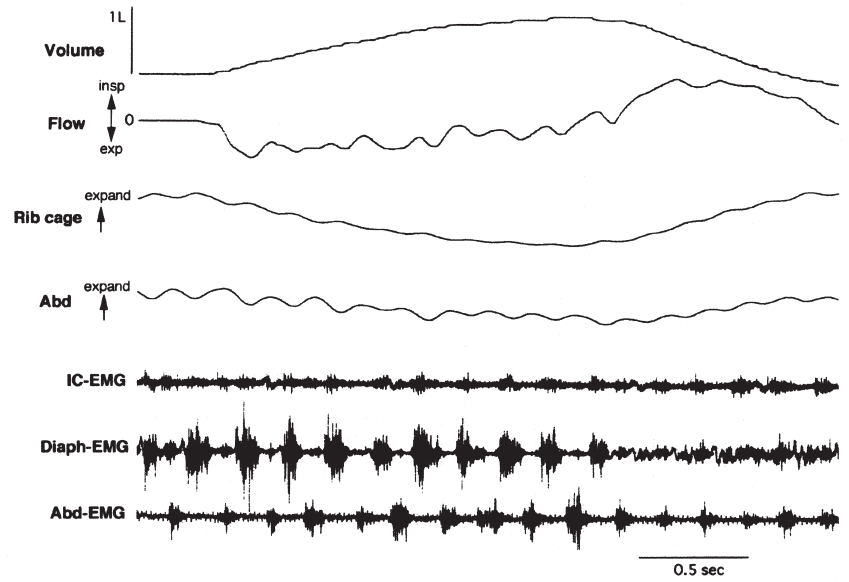

Fig. 2 Time scale of Fig. 1 is magnified and one breath is shown. Abbreviations are the same as in Fig. 1.

the respiratory flow and tidal volume via tight fitting nasal mask using a hot-wire flowmeter (RF2, Minato, Osaka), thoracoabdominal motions by inductiveplethysmography (Respitrace, Ambulatory Monitoring, NY) and electromyograms (EMGs) of the respiratory muscles. EMGs of the intercostal muscle at right second intercostal space, diaphragm at right 6 th intercostal space and abdominal muscle at right lower quadrant were recorded by bipolar surface electrodes while the patient was supine.

Figure 1 shows the record during an attack. In the rib cage and abdominal mo-

tions there were fine rhythmic movements (~280/min) superimposed on the motion of her own respiration. These small fluctuations were also observed on the trace of respiratory flow. In diaphragmatic EMG, bursts synchronizing with the high frequency motions were superimposed on "ramp-shaped" her own respiratory activities. Such intermittent bursts were also observed in the intercostal and abdominal EMGs. These intermittent bursts in the EMGs of the respiratory muscles decreased during inspiration in frequency and amplitude. Diagnosis of diaphragmatic flutter was made because of the rapid oscillatory activities superimposed on her own respiratory activities in the respiratory muscle EMGs [1]. In Fig. 2, the time scale of the record in Fig. 1 was magnified. The bursts in the diaphragmatic and intercostal EMGs developed as reciprocal to those in the abdominal EMG.

The flutter never occurred during sleep. She could not stop her flutter voluntarily. Her symptoms sometimes deteriorated after emotional events such as crying or missing her children. The flutter was suppressed by intravenous administration of either 10 mg of diazepam or 250 mg of diphenylhydantoin. However, she preferred not to take medication because she was worried about adverse effects. We conscientiously explained to her the nature of diaphragmatic flutter, and then the frequency of attacks gradually decreased. Six months after leaving hospital, her symptom of the diaphragmatic flutter has improved. However, she newly developed the symptoms of irritable colon syndrome.

DISCUSSION

Diaphragmatic flutter is a rare disorder characterized by rapid involuntary contractions of the diaphragm. It was firstly described in 1723 by Leeuwenhoek, who was personally affected with this disorder [4]. In the present case, high frequency intermittent contractions of the diaphragm superimposed on ordinary respiratory rhythm, i.e., "dual-rhythms" in the respiratory muscles, ensured a diagnosis of diaphragmatic flutter [1, 3].

EMG was measured by surface electrodes rather than esophageal electrodes in the present case. The signal from the surface electrodes may contain both intercostal and diaphragmatic signal. However, the location we attached the electrodes measuring diaphragmatic EMG was used with standard fashion and it is known that the signal from the surface electrodes and esophageal electrodes do not have much difference in the qualitative data [5]. We thought that surface electrode which did not require the placement of a nasoesophageal catheter is more acceptable to our patient who was extremely nervous.

Possible etiology of this disorder includes abnormal excitation of the central nervous system including cerebrum and the brainstem, direct irritation of the phrenic nerve and irritation of the diaphragm itself [2]. EMG analysis in the present case revealed that high frequency bursts were also observed in the intercostal and abdominal muscles indicating that the etiology of these abnormal activities of the respiratory muscles is above the spinal motorneurons. The high frequency activities in the abdominal muscle developed during both inspiratory and expiratory phase of ordinary respiration. This finding suggested that the origin of the abnormal activities may be independent to the "respiratory center" in the pontomedulla because abdominal muscles are principally expiratory muscles. However, we can not determined if the exact origin of the diaphragmatic flutter was the cerebrum or brainstem. Some reports use the term of respiratory myoclonus instead of diaphragmatic flutter [1, 6], but these two different disease names represent the same disease. We thought that respiratory myoclonus may be suitable instead of generally termed diaphragmatic flutter in this case since abnormal activities were recorded not only diaphragm but also other respiratory muscles.

The commands from the cerebrum regulate the respiratory muscles via corticospinal tract. This pathway is also thought to operate the respiratory muscles during playing wind instruments, speaking and singing [7]. It is also known that the changes in mental activities such as attention, anxiety and indignation also influence the pattern and depth of breathing via the forebrain [8]. These pathways are thought to mediate the behavioral control of respiratory muscles in humans. In the present case, suppression of the diaphragmatic flutter by intravenous administration of diazepam may indicate that these pathways are involved in generating diaphragmatic flutter, since this drug mainly acts on the cerebrum [9]. This speculation is

also supported by the observations that the attacks did not occur during sleep and that emotional events such as crying and missing her children led to a deterioration of her symptoms.

Although we tried to record her electroencephalogram during diaphragmatic flutter, the permission from the patient could not be got. She was afraid of being diagnosed as a mental disease. A normal MRI does not exclude the possibility of an organic disorder of the central nervous system. However, abnormal movement was only observed in the upper abdomen and thoracic wall. She could speak and move her extremities during diaphragmatic flutter. Furthermore, when we contacted her after leaving hospital for six months, she newly developed the symptoms of irritable colon syndrome instead of diaphragmatic flutter. These findings may support that the organic disease of her central nervous system is unlikely. She also had psychosomatic symptoms such as anorexia and her symptoms deteriorated after emotional events. The detail cause of her unstable mental condition was unknown. However, her symptom of the diaphragmatic flutter appeared with start of infant care. It is suggested that the pressure of child nursing will be participating in the appearance of her condition. Other flutter patients who did not show any organic lesions were young and they also had such psychosomatic symptoms [10]. We speculated that the psychosomatic status may play a significant rule in the development of the diaphragmatic flutter as like as hyperventilation syndrome and there may be a disorder in the behavioral control of breathing in the present case.

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