myocardial disease itself.

Although a spontaneous recovery of myocardial function is possible, it seems unlikely since our patient responded dramatically within the first few weeks of treatment with pindolol. Beta-adrenoceptor antagonists could improve myocardial function through multiple mechanisms. Beta-receptor density has been shown to increase on chronic exposure to propranolol. By restoring the number of receptors to normal, the adrenergic system would once again be able to respond by providing inotropic support during periods of stress. However, it is interesting and perhaps significant that our patient improved on pindolol, a beta-blocker which has been shown to reduce beta-receptor density. Beta-blockers may also improve cardiac function by protecting against cellular damage. It is clear that further work needs to be done to delineate the mechanisms by which beta-receptor antagonists may be useful in the treatment of dilated cardiomyopathy.

REFERENCES

The Irregularly Irregular Pattern of Respiratory Dyskinesia*
Samuel T. Kuna, M.D.;† and Rabia Awan, M.D.;‡

The respiratory pattern during wakefulness and sleep was characterized in a 70-year-old woman with respiratory dyskinesia. During wakefulness, both respiratory frequency and tidal volume exhibited an irregularly irregular pattern. In addition, wide fluctuations occurred in the position of the rib cage and abdomen at end expiration. A normal respiratory pattern appeared during non-REM and REM sleep.

An abnormal respiratory pattern has infrequently been associated with the oro-buccal and facial choreiform movements of tardive dyskinesia. These dyskinetic movements are reported to be absent during sleep. No physiologic recordings during wakefulness or sleep have documented the pattern of breathing in subjects with respiratory dyskinesia.

CASE REPORT

Over a 40-year period, this 70-year-old woman received many medications including several different neuroleptic drugs in the treatment of her psychiatric problems. In 1983, she developed involuntary oro-buccal and facial movements which were felt to be consistent with the diagnosis of tardive dyskinesia. At that time, no notation was made concerning an abnormal respiratory pattern. Upon evaluation in October, 1985, her unkept appearance was characteristic of frequent paroxysms of involuntary chewing movements usually associated with a rhythmic elevation of her eyebrows. Respiration was shallow and irregular with a rate of 60/min. Inspiration was marked by prominent anterior motion of the abdominal wall and visible flaring of the nostrils. Grunting was frequently present on expiration. Even though she spoke in short sentences, her breathing seemed to interfere with phonation. The irregularity of her respiratory pattern became much worse when she was agitated. The patient was cognizant of her involuntary movements but denied any difficulty breathing. Although voluntary control of limb movements was unimpaired, she was unable to voluntarily hold her breath or perform any coordinated respiratory maneuvers. Arterial blood gas sampling while breathing room air revealed: pH 7.45, Pco₂ 40 mm Hg, Pco₂ 40 mm Hg and HCO₃ 26 mEQL. Chest x-ray film, CBC and routine blood chemistry test results were all within normal limits.

The subject was studied at night while lying quietly in bed in a darkened room for two hours while awake and an additional two hours asleep. At the time of the study, she was not aware that her respiratory pattern was the focus of attention. Her movements were observed with a video monitor. The appearance of speech was confirmed by a one-way intercom. The following parameters were monitored: electroencephalogram (C3/A2), bilateral electrooculograms, rib cage and abdominal excursions and the electromyograms (EMG) of the frontalis and alae nasi muscles. Rib cage and abdominal excursions were measured by impedance plethysmography in DC mode (Respi-trace). Both rib cage and abdominal calibration factors were ar-

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bitarally set at 1. EMG of the frontalis muscle was obtained from two surface electrodes placed adjacent to one another 1 cm above the left eyebrow. Alae nasi EMG was also obtained from surface electrodes, these attached above each nostril. EMG signals were filtered (0.1 to 3 kHz) through a differential amplifier (Grass P15). All parameters were recorded on an electrostatic recorder (Gould ES 1000).

Her respiratory pattern during wakefulness exhibited a marked irregularity in both the frequency and amplitude of rib cage and abdominal excursions (Fig 1). Over a 10-min period, mean respiratory frequency was 68.6 ± 1.2 (SEM) breaths/min. Paradoxic motion of the rib cage and abdomen was common. End expiratory position of the rib cage and abdomen showed considerable variation. Paroxysmal dyskinetic movements of the facial muscles were detected by the rhythmic activation of the frontalis muscle. Changes in the EEG and EOG during spasmic movements are felt to represent activity from facial and scalp muscles. Alae nasi EMG exhibited phasic inspiratory activity except during the facial dyskinesia, when its activity mirrored that of the frontalis muscle. During paroxysms of rhythmic frontalis muscle activation, respiratory rate decreased markedly.

The above findings during wakefulness were reconfirmed on a separate day when the subject was studied in a sitting position. At that time, the briefly wore a face mask connected to a low resistance Hans Rudolph valve. A pneumotachograph with a pressure transducer (Validyne) was attached to the inspiratory side of the valve. The flow signal was integrated over time to obtain tidal volume. The face mask caused the patient to become more agitated. Over a 5-min period, respiratory frequency was 85.4 ± 3.7 breaths/min. Inspiratory time (Ti), expiratory time, (Te), and tidal volume (Vt) varied widely (Table 1).

During sleep, dyskinetic activation of the frontalis and alae nasi muscles was absent and respiratory-related activity of the alae nasi muscle was very rare. The patient remained in the same position between the awake and sleep states. In all stages of sleep, the rib cage and abdomen moved synchronously and end expiratory volume remained relatively constant (Fig 2, 3). In non-REM sleep, the respiratory pattern became uniform and regular (Fig 2). Compared to the wakeful state, tidal volumes were larger and respiratory frequency was decreased. Mean frequency over a 10-min period was 15.3 ± 0.1 breaths/min. Respiratory pattern in REM sleep did not resemble that during wakefulness. Rather, it showed a variability similar to that seen in normal subjects (Fig 3). Mean respiratory frequency over a 10-min period of REM sleep was 15.1 ± 0.5 breaths/min. REM sleep was subdivided into phasic and tonic periods based on the respective presence or absence of rapid eye movements. Respiratory frequency was slightly higher in phasic compared to tonic REM (Fig 3). The tonic period was associated with larger tidal volumes. On awakening, the erratic respiratory pattern and facial dyskinesia resumed immediately.

**Discussion**

Although respiratory dyskinesia has been described in ten other subjects, no physiologic recordings are available to better characterize this phenomenon. The individual reported here had a rapid, shallow respiratory pattern during REM sleep. In phasic REM sleep, the hyperventilation and eye movements are increased, whereas the ventilation is more regular in tonic REM. This patient also exhibited rapid eye movements, with a regular respiratory pattern during REM sleep. The presence of rapid eye movements at the beginning and end of the recording indicate phasic period of REM sleep. Absence of rapid eye movements in middle of recording indicates tonic period.

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**Table 1—Respiratory Parameters during Wakefulness (60 Breaths)**

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Mean ± SEM</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>T₁ (sec)</td>
<td>0.24 ± 0.01</td>
<td>0.1-0.55</td>
</tr>
<tr>
<td>Tₑ (sec)</td>
<td>0.37 ± 0.02</td>
<td>0.15-1.2</td>
</tr>
<tr>
<td>Vₗ (ml)</td>
<td>143 ± 10</td>
<td>50-340</td>
</tr>
</tbody>
</table>

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**Figure 1.** Recording obtained on supine subject during wakefulness. Tracings are, from top to bottom: electroencephalogram (EEG), bilateral electrocorticograms (EOG), alae nasi electromyogram (EMG), frontalis EMG and rib cage and abdominal excursions. A paroxysm of involuntary dyskinetic facial movement is indicated by rhythmic activation of frontalis muscle.

**Figure 2.** Recording obtained on subject during non-REM sleep. Tracings as in Figure 1.

**Figure 3.** Recording obtained on subject during REM sleep. Tracings as in Figure 1. Presence of rapid eye movements at the beginning and end of the recording indicate phasic period of REM sleep. Absence of rapid eye movements in middle of recording indicates tonic period.
wakefulness with an irregularly irregular rate and depth of respiration. In addition, the varying position of the rib cage and abdomen at end expiration suggests the presence of a variable and expiratory volume. This fluctuation may in part be explained by another prominent feature of her respiratory pattern during wakefulness: expiratory grunting. The grunting sounds appeared to be caused by contraction of anterior abdominal wall muscles accompanied by partial closure of her laryngeal airway. These active braced expirations could produce an inward movement of the abdomen in concert with an outward movement of the rib cage accounting for the paradox movements of rib cage and abdomen during wakefulness. During sleep, grunting was absent, end expiratory volume was relatively stable, and the rib cage and abdomen moved synchronously. The absence of involuntary movements associated with tardive dyskinesia during sleep has never been documented. During a two-hour sleep recording, this patient had no facial or respiratory dyskinesia.

The pathophysiologic basis for tardive dyskinesia and other movement disorders is felt to be an imbalance of facilitatory and inhibitory influences in nigrostriatal pathways. The substantia nigra and striatum are part of the extrapyramidal motor system which is felt to affect movement through efferent projections to the motor and premotor areas of the cortex. It has been suggested that the behavioral respiratory control system is involved in the generation of the irregular pattern of breathing in REM sleep of normal subjects. One might therefore have predicted that this subject’s respiratory pattern during REM sleep would resemble that during wakefulness. Instead, her respiratory pattern during REM sleep resembled that of a normal individual.

Within the nigrostriatal feedback loop, inhibitory dopaminergic neurons with cell bodies in the substantia nigra terminate on intrastrial interneurons which release acetylcholine, a facilitatory transmitter for the next limb of the loop, the striatonigral component. Antipsychotic drugs block dopamine receptors in the striatum. Following chronic neuroleptic treatment, intrastrial interneurons exhibit an enhanced sensitivity to dopamine. This imbalance within the nigrostriatal pathway is felt to result in the abnormal involuntary movements of tardive dyskinesia. In cats, the concentration of dopamine in the striatum and thalamus dramatically decreases during non-REM sleep. A similar occurrence in patients with tardive dyskinesia might restore balance within the nigrostriatal pathway, explaining the resolution of the movement disorder during non-REM sleep. Other experiments reveal an increase in acetylcholine release within the cat striatum during REM sleep. Such a mechanism could explain the absence of dyskinetic movements during REM sleep in this patient. Although the mechanisms explaining these phenomena are speculative, the clinical recognition of this chaotic respiratory pattern is important as it can be the first manifestation of tardive dyskinesia.

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REFERENCES


Use of Continuous-Wave Doppler Ultrasound to Evaluate and Manage Primary Pulmonary Hypertension*

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Doppler ultrasound was used to assess the response to serial drug testing in a patient with primary pulmonary hypertension. There was a close correlation between the pressure estimated by Doppler and the pulmonary arterial systolic pressure measured invasively (r = 0.98). Continuous-wave Doppler ultrasound, although not a substitute for initial Swan-Ganz catheterization, may obviate the need for repeat invasive procedures and permit closer monitoring of patients during treatment.

The diagnosis of primary pulmonary hypertension and an assessment of the initial and long-term response to therapy now require catheterization of the pulmonary artery. A reliable noninvasive technique that is sufficiently sensitive to detect the effects of therapy on pulmonary artery pressure would be an important addition to the management of this disorder.

Continuous-wave Doppler ultrasound can provide an accurate noninvasive estimate of the pulmonary arterial systolic pressure in patients with pulmonary hypertension, but thus far its use in monitoring sequential changes in

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