Sleep Fragments

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Fluctuating Flow

Vibrato, Tremolo, or Upper Airway Oscillation

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In Brief

We describe a case of a 48-year-old woman with an incidental finding on a research polysomnogram, which posed an interesting differential diagnosis elucidated with the aid of advanced diagnostic methods. This case illustrates the utility of quantitative airflow assessment during sleep in the evaluation of involuntary upper airway phasic activity.

consciousness after trauma but had no other neurologic symptoms.

The polysomnogram revealed a sleep latency of 5 minutes, which included decreased slow wave and REM sleep, with a slightly decreased sleep efficiency (74.9%). The total, non-REM (NREM), and REM apnea-hypopnea index values were 6.2, 1.2, and 47 events per hour, respectively.

Similarly, the total, NREM, and REM arousal indices were 11.3, 12.7, and 26.1 events per hour, respectively. All of the respiratory events were obstructive. The minimum O₂ saturation was 84% during NREM sleep and 88% during REM sleep. Airflow tracing showed rhythmic phasic oscillations during wakefulness and sleep. Oscillations persisted during NREM and

Case Vignette

A 48-year-old African-American woman presented for an overnight polysomnography study as part of participation in a research protocol. Although the patient had no prior complaints, on the screening questionnaire she acknowledged loud snoring and mouth dryness upon awakening. Her screening physical examination was significant for narrowed lateral pharyngeal arch, Mallampati grade IV airway, and audible staccato breathing during expiration. The rest of her examination was unremarkable. She had a traumatic incident 16 years ago that required hospitalization for splenic repair. She admits that there was brief loss of

Figure 1. Two-breath cycles in wakefulness (Wake) and in non-REM (NREM) and REM sleep. Polysomnogram includes EEG, EKG, oxygen saturation, airflow, thoracic effort, abdominal effort, and leg EMG channels. Flow oscillations persist during wakefulness and sleep as well as during inspiration and expiration.

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Video 1. Three-dimensional dynamic MRI showing the sagittal view of the upper airway, extending from the nasopharynx to the glottic folds of the larynx. The cardiac rhythm (1.4 Hz) and upper airway movement frequency (2.2 Hz) are identical to the findings on the EKG and airflow channels of the polysomnogram.

REM sleep with similar frequency and amplitude and were only absent in apnea (Figure 1).

We noticed subtle quivers in the patient's speech during sustained phonation, and she subsequently had a dynamic MRI of the upper airway, obtained as part of research. The MRI did not show any aberrant upper airway tissue or mass; however, bilateral rhythmic movements of the palatal, pharyngeal, and laryngeal walls were noted. The MRI findings are shown in Video 1.

Question

Based on the clinical vignette, polysomnogram, and MRI findings, what is the diagnosis?

Discussion

Voice tremor is the phonatory manifestation of the essential tremor syndrome, characterized by involuntary skeletal muscle contraction that leads to oscillatory movement of laryngeal muscles at a rate of 4 to 10 Hz (1). There is wide disparity in symptom severity, with severe forms presenting as audible dysphonia and mild cases undetected or detected as mere incidental findings (2). In some cases, the muscles of the palate and pharynx are seen

to synchronously oscillate with the larynx (3). This kind of upper airway involvement can lead to rapid intermittent changes in airway resistance with continuous interruption in airflow (4).

Persistent Oscillations in Airflow Signal

In this patient, the airflow tracing from the polysomnograph showed oscillations at a rate of 2.2 Hz during inspiratory and expiratory phases (Figure 1). Airflow oscillations continued throughout the study, except during apneas, although rapid phasic bursts in chin EMG signal persisted at the same frequency as the fluctuations in flow. At first, we ruled out artifacts that could affect the airflow tracing, such as

humming (50–60 Hz), cardioballistic airflow (frequency most commonly <1.5 Hz), and channel cross-talking between ECG (1.33 Hz or 80 bpm) and EMG channels (Figure 2). Salas and colleagues described a case of idiopathic palatal myoclonus with demonstrable flow oscillations from polysomnography (7). Conditions in the differential, such as Shy-Drager syndrome and other extrapyramidal disorders, may also result in flow oscillations during wakefulness (8) but have not been shown to persist during sleep.

Evaluation and Diagnosis

A three-dimensional dynamic MRI of the upper airway performed during wakefulness at 18 frames per second, as part of research,

Figure 2. Twenty-second epoch displaying the EKG and airflow channel. The frequency of the cardiac rhythm (1.4 Hz) is shown to be slower than the flow oscillations (2.2 Hz).

Figure 3. Forty-five-second epoch showing an apneic event–related oxygen desaturation during REM sleep. The EMG and airflow channels are duplicated to show the persistence of inspiratory phase high-frequency (>30 Hz) activity after low-frequency filtering. Low-frequency oscillations (<3 Hz) occur in inspiratory and expiratory limbs of the flow signal and continue in the EMG channel during apnea.

showed involuntary elevation of the soft palate, tongue movements anteriorly, and adduction of the palato-pharyngo-laryngeal walls. The upper airway structures demonstrated rhythmic contraction during rest and continuous phonation, at the same frequency (2.2 Hz) as seen in the polysomnograph airflow signal. In the clinical setting, laryngoscopy would be the first procedure in the workup for subtle upper airway closure; however, three-dimensional MRI provided the ability to visualize the entire airway simultaneously.

We believe the EMG bursting signals throughout the polysomnograph represent one or a combination of any of the mouth floor and tongue muscles because rhythmic movements of hyoid bone anteriorization (genioglossus), tongue twitching (genioglossus), and tongue elevation (mylohyoid) (5) were clearly visualized on the MRI at rest (Video 1). In the polysomnogram, we measured airflow using a full face mask and a pneumotachograph that could quantify

the airflow insufficiency associated with airway closure oscillation. Airflow measurements revealed an 80 to 90% decrease in inspired and expired airflow rate during oscillations. We suspect that this may interact with upper airway collapse and sleep-disordered breathing, although we do not know if this is intrinsically related to the patient's sleep apnea *per se*. Airflow oscillations occurred throughout the night except during complete upper airway obstruction (Figure 3), suggesting that oscillations take place at or below the site of obstruction. Persistence of the low-frequency EMG spikes (Figure 3) suggests that the underlying muscle contractions continue during apnea.

Conclusions

Semiquantitative instrumentation, such as nasal cannula or thermistors, may have inadequate sensitivity for routinely

characterizing airflow oscillations as described. Measurement of airflow during sleep could indeed become a neurotopological biomarker and a valuable tool in the evaluation of involuntary upper airway phasic activity, providing physicians with a quantitative and qualitative means for characterizing the presence and intensity of abnormal muscle rhythmicity (6).

The current volunteer would benefit from a complete neurologic and ear, nose, and throat assessment to further characterize the research findings.

Answer

Essential upper airway muscles' tremor with mild obstructive sleep apnea.

This patient's polysomnogram demonstrated snoring, mild apnea with a total apnea–hypopnea index of 6.2, and persistent flow oscillations in wakefulness and sleep. The amplitude and frequency of oscillations are preserved in NREM and REM sleep. A rhythmic pattern persisted in inspiration, expiration, and flow-limited breathing but was absent in apnea. An involuntary rhythmic elevation of the soft palate, concentric medialization of the pharyngeal wall, elevation of the hyoid bone, laryngeal complex, and twitching of the tongue were simultaneously seen on the real-time dynamic MRI at 18 frames per second.

Together these examination findings are consistent with an essential tremor syndrome; in this case, upper airway muscles tremor with mild sleep-disordered breathing. However, a very close differential that has been considered is palato-pharyngo-laryngeal myoclonus. ■

Author disclosures are available with the text of this article at www.atsjournals.org.

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