Silent Upper Airway Resistance Syndrome*

Prevalence in a Mixed Military Population

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Study objectives: The upper airway resistance syndrome (UARS) is a recently described form of sleep-disordered breathing in which transient increases in upper airway resistance result in repetitive EEG arousals. UARS is not associated with apnea or diminished airflow, although snoring and excessive daytime somnolence (EDS) are common. This report describes a subset of patients with UARS diagnosed by polysomnography who do not manifest snoring, which we define as silent upper airway resistance syndrome (SUARS).

Design: A retrospective review of all polysomnographies performed at our sleep disorders center during 2000.

Setting: Sleep disorders center of a large, academic, military hospital.

Patients: Our center serves military personnel, military retirees, and their dependent families.

Interventions: Esophageal manometry during polysomnography was routinely performed on patients with hypersomnolence (Epworth sleepiness scale > 10) who demonstrated a total arousal index ≥ 10/h and a respiratory disturbance index of < 5/h on prior polysomnography. UARS was definitely diagnosed in patients who demonstrated repetitive increased upper airway resistance (IUAR) associated with brief EEG arousals followed by normalization of esophageal pressure (Pes). IUAR was defined by a pattern of crescendo negative inspiratory Pes of ≤ −12 cm H₂O.

Results: During calendar year 2000, we performed 724 polysomnographies in 527 patients. Obstructive sleep apnea was diagnosed in 383 patients (72.6%), and 44 patients (8.4%) were found to have UARS. In four patients with UARS (0.8% of total and 9.1% of UARS), snoring was not reported by history or observed during polysomnography, and SUARS was ultimately diagnosed.

Conclusions: UARS may occur in the absence of clinically significant snoring and may be an occult cause of EDS. We report a prevalence of SUARS of 9% among UARS patients and nearly 1% of all patients studied for hypersomnolence by polysomnography.

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Key words: esophageal manometry; excessive daytime sleepiness; hypersomnolence; increased upper airway resistance; obstructive sleep apnea; sleep-disordered breathing; upper airway resistance syndrome

Abbreviations: BMI = body mass index; EDS = excessive daytime sleepiness; ESS = Epworth sleepiness scale; OSAS = obstructive sleep apnea syndrome; Pes = esophageal pressure; RDI = respiratory disturbance index; RERA = respiratory effort-related arousal; SUARS = silent upper airway resistance syndrome; TAI = total arousal index; UARS = upper airway resistance syndrome

Upper airway resistance syndrome (UARS) is a recently described form of sleep-disordered breathing that may result in excessive daytime sleepiness (EDS).1–4 UARS is defined by repetitive increases in upper airway resistance (IUAR) associated with brief EEG arousals.1,5 The diagnosis requires demonstration of IUAR in a crescendo pattern of negative inspiratory pressures on esophageal manometry.5 We used a negative inspiratory pressure of ≤ −12 cm H₂O in scoring these studies. The resulting arousals are followed by normalization of Pes.1,4–6 However, esophageal manometry is not

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commonly employed, making UARS difficult to diagnose definitively. Clinicians often make the diagnosis presumptively based on the presence of crescendo snoring associated with respiratory effort-related arousals (RERAs). Limited use of esophageal manometry may underdiagnose UARS and may lead to misclassification of a patient’s hypersonolence. This misdiagnosis may, in turn, result in the inappropriate use of stimulants as a potential treatment of EDS. These therapies may be ineffective or may mask the underlying sleep disorder.

Although difficult to diagnose, UARS is suggested by EDS associated with snoring in patients who do not demonstrate apneic or hypopneic respiratory events on polysomnography. However, patients may manifest sleep-disordered arousals consistent with UARS even in the absence of snoring, which we define as silent UARS (SUARS). The purpose of this article is to report the occurrence and prevalence of SUARS in our population.

**Materials and Methods**

We retrospectively reviewed all patients who underwent polysomnography for the evaluation of EDS at our sleep disorders center during 2000. Our center serves active duty military personnel, military retirees, and their dependent family members.

All patients were studied by attended overnight polysomnography in our sleep laboratory using a 16-channel montage (SensorMedics α Somnostar System; SensorMedics; Yorba Linda, CA). Polysomnography consisted of continuous recordings of central and occipital EEGs, bilateral electro-oculograms, submental and bilateral tibial electromyograms, and ECG. Nasal and oral airflow was measured by thermistor or pressure transducer. Tracheal sounds were monitored using an acoustic microphone. Thoracic and abdominal excursions were measured using inductance plethysmography. Continuous oxygen saturation was assessed using noninvasive pulse oximetry. Body positioning was performed using a multiport Gaeltc catheter (Gaeltc Ltd; Hackensack, NJ). RERAs were scored when IUAR was followed by an EEG arousal. IUAR was established by the development of a negative inspiratory pressure of ≤ −12 cm H2O preceding an arousal. Episodes of IUAR were followed by normalization of esophageal pressure (Pes).

The diagnosis of UARS required a RERA index ≥ 5/h and an RDI ≤ 5/h.

Data were analyzed using repeated measures with analysis of variance for the crossover design. The two groups, SUARS and UARS, were compared and analyzed using independent-sample t test, and the level of significance was set at p < 0.05. Statistical analysis was carried out using software (SPSS for Windows version 11.0; SPSS; Chicago, IL).

**Results**

During calendar year 2000, 527 patients underwent a total of 724 polysomnographies. Of those studied, obstructive sleep apnea was diagnosed in 353 patients (72.6%), and 44 patients (8.4%) were found to have UARS.

Among those with a diagnosis of UARS, snoring was not reported by history or observed during polysomnography in four patients. SUARS was ultimately diagnosed in these individuals. Among those with SUARS, three were men and the average age was 34.8 ± 6 years. None met criteria for obesity by body mass index (BMI), and all four patients reported EDS. Further patient demographics and polysomnography results are depicted in Tables 1, 2. These demographics and clinical variables were compared to 20 consecutive patients with UARS diagnosed using polysomnography (Table 2). No significant differences between these two groups were identified.

The overall prevalence of UARS was 8.4% of those patients studied in our center in calendar year 2000. SUARS was seen in 9.1% of UARS patients and 0.8% of all patients studied using polysomnography for EDS.

**Discussion**

UARS can occur in the absence of snoring and may be an occult etiology for EDS. Sleep-disordered breathing without clinically apparent snoring has been previously reported in the literature. In a

<table>
<thead>
<tr>
<th>Age, yr</th>
<th>Gender</th>
<th>BMI</th>
<th>ESS</th>
<th>TAI</th>
<th>RDI</th>
<th>RERA Index</th>
<th>Pes Nadir, cm H2O</th>
</tr>
</thead>
<tbody>
<tr>
<td>41</td>
<td>Male</td>
<td>24.5</td>
<td>11</td>
<td>29.3</td>
<td>0.6</td>
<td>40.5</td>
<td>−31</td>
</tr>
<tr>
<td>28</td>
<td>Male</td>
<td>27.7</td>
<td>18</td>
<td>26.4</td>
<td>3.8</td>
<td>23.8</td>
<td>−45</td>
</tr>
<tr>
<td>40</td>
<td>Female</td>
<td>26.4</td>
<td>23</td>
<td>27.2</td>
<td>0.2</td>
<td>21.3</td>
<td>−32</td>
</tr>
<tr>
<td>30</td>
<td>Male</td>
<td>22.0</td>
<td>13</td>
<td>34.2</td>
<td>2.4</td>
<td>29.5</td>
<td>−25</td>
</tr>
</tbody>
</table>
study of sleep-disordered breathing among postmenopausal women, Guilleminault and colleagues\textsuperscript{10} noted that over one fourth of patients with a diagnosis of UARS did not report snoring. However, SUARS as a clinical entity has not been fully described.

Failure to employ esophageal manometry in the assessment of unexplained arousals may lead to missed diagnoses in patients with UARS. Attributing unexplained EDS to other etiologies, such as idiopathic hypersomnolence or periodic limb movement disorder, may result in unnecessary medical therapy and failure to reverse the patient’s underlying sleep-disordered breathing.\textsuperscript{3} Although common, snoring is not essential to the diagnosis of UARS, as evident in our experience. An overreliance on snoring as a screening tool in the evaluation or fundamental factor for the diagnosis may lend itself to diagnostic errors.

Generalizations about patient characteristics and other variables that may predict this syndrome are difficult to conclude given the limited number of described cases. Patients with UARS, and likely SUARS, tend to be younger and are less likely to be obese. In our population, no demographic, clinical, or polysomnographic differences between patients with UARS and SUARS were identified, further supporting that SUARS is merely a unique presentation of UARS and not an independent entity. As with obstructive sleep apnea syndrome (OSAS), this disorder is not limited to older, overweight individuals. Both UARS and OSAS result from an increased resistance to airflow in the upper airways resulting in arousals and sleep fragmentation resulting in EDS. Additionally, both have been described in a wide diversity of age, race, and body habitus. However, whether this syndrome is a distinct entity or merely a continuum of OSAS remains controversial.\textsuperscript{4,11–13} Despite this controversy, the prevalence of both disorders is likely underrecognized due to a lack of typical features, which further supports the routine use of Pes monitoring in those patients with unexplained EDS. In our reported experience, UARS is common and was seen in >8% of patients studied. While we do not recommend the use of esophageal manometry in the initial evaluation of EDS or suspected sleep-disordered breathing, we do recommend its use in hypersomnolent patients with unexplained and frequent arousals on polysomnography, even in the absence of snoring.

Use of esophageal manometry is currently the “gold standard” in the diagnosis of UARS, although other, less invasive diagnostic methods are used.\textsuperscript{2,4,5} Esophageal catheters are minimally invasive, safe, and induce only minimal changes in sleep architecture.\textsuperscript{14} Their use should not be avoided when clinically indicated.

Treatment of UARS and SUARS is similar to that of OSAS, with continuous positive airway pressure being the most widely used therapeutic option with proven efficacy.\textsuperscript{1,2,4} As with OSAS, oral appliances and surgical procedures have been used with effective outcomes.\textsuperscript{15–18}

Currently, UARS and SUARS are not recognized in the International Guidelines of Sleep Disorders Revised-Diagnostic and Coding Manual.\textsuperscript{19} This fact, along with a limited understanding of these disorders and their prevalence, is likely contributing to their underrecognition. Wider use of esophageal manometry, further reporting of other diagnostic tools, and treatment outcomes related to UARS promise to advance both our understanding of the disease and its prevalence. In patients with hypersomnolence and unexplained arousals, a high index of suspicion for UARS, and SUARS in the absence of snoring, may lead to increased recognition of these disorders.

### References


### Table 2—Patient Demographics and Polysomnography Results\textsuperscript{*}

<table>
<thead>
<tr>
<th>Variables</th>
<th>SUARS</th>
<th>UARS</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, yr</td>
<td>34.8 ± 5.8</td>
<td>35.6 ± 8.2</td>
<td>0.72</td>
</tr>
<tr>
<td>Male gender, %</td>
<td>75</td>
<td>70</td>
<td>0.22</td>
</tr>
<tr>
<td>BMI</td>
<td>25.1 ± 1.9</td>
<td>24.8 ± 2.2</td>
<td>0.41</td>
</tr>
<tr>
<td>ESS</td>
<td>16.3 ± 4.3</td>
<td>14.9 ± 3.1</td>
<td>0.24</td>
</tr>
<tr>
<td>TAL events/h</td>
<td>29.3 ± 2.5</td>
<td>29.6 ± 6.7</td>
<td>0.83</td>
</tr>
<tr>
<td>RDI, events/h</td>
<td>1.8 ± 1.4</td>
<td>2.2 ± 2.4</td>
<td>0.64</td>
</tr>
<tr>
<td>RERA index, events/h</td>
<td>28.8 ± 6.2</td>
<td>32.2 ± 4.3</td>
<td>0.23</td>
</tr>
<tr>
<td>Esophageal manometry</td>
<td>-33.3 ± 5.9</td>
<td>-30.3 ± 3.9</td>
<td>0.38</td>
</tr>
</tbody>
</table>

*Data are presented as average ± SD unless otherwise indicated.
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