electron density (Fig 2, bottom right, D). The cytoplasm in the center showed the usual cell organelles. Based on these morphologic findings and in view of the preceding antibiotic therapy and the absence of bacteria, we interpreted these structures as fungi.

**Development of the Case**

Since pathogens could not be clearly demonstrated, especially not in serologic investigations, we continued antibiotic treatment with 1 g of meropenem IV tid for 7 days and, subsequently, with an oral dose of 400 mg moxifloxacin qd for 2 weeks. Antiphlogistic therapy with a systemic corticosteroid and inhalation with fluticasone, as well as sufficient doses of analgesics, completed our treatment regimen. It led to defervescence 5 days after the patient’s admission to our hospital. The initially severe restriction of lung function became normal during clinical treatment (maximum vital capacity, 2.22 L vs 3.81 L), as did the inflammatory parameters in the blood. The patient could be discharged as cured approximately 4 weeks after his petroleum aspiration. Two months after his discharge from our hospital, radiography was performed, showing only small scarred residues of the previous inflammation (Fig 1, right, C).

**DISCUSSION**

Our clinical findings largely correspond to the described picture of hydrocarbon pneumonitis.1–4 Our patient showed the typical clinical symptoms, such as dyspnea, cough, hemoptysis, chest pain, and high temperature immediately after petroleum aspiration. Radiograph examination of the chest as well as CT 1 day after petroleum aspiration revealed lung consolidations and incipient formation of pneumatoceles, ie, well-defined cavity nodules.5 Conservative antiphlogistic and antiphlogistic therapy led to a rapid decrease of the described symptoms, accompanied by an obvious improvement of the patient’s general condition; however, a striking finding was the persistent high temperature and the increase of all inflammatory parameters in the blood despite the administration of broad-spectrum antibiotics. Using the two qualitatively and quantitatively best bronchoscopic methods (protected specimen brush and BAL), we were not able to demonstrate any pathogens, either microscopically or in culture, which would have confirmed the diagnosis of pseudoinfectious lung disease.6 In contrast, electron microscopic inspection of the BAL specimen revealed two interesting findings. One peculiarity was the occurrence of macrophages with lipid-containing inclusions that exhibited all morphologic signs of an activation.7 The occurrence of lipid vacuoles indicates an increased phagocytic activity of macrophages that leads to an increased release of cytokines by macrophages, which in turn can trigger an increased and prolonged inflammatory reaction. The second interesting finding refers to the ultrastructural demonstration of structures we interpreted as fungi. We believe that we were dealing with a fungus colonization and not with a pulmonary mycosis, especially since all other culture and serologic tests for fungi and fungus antigens yielded negative results. This one special case is, of course, insufficient to decide to what extent a secondary fungal colonization is characteristic of hydrocarbon pneumonitis, and whether or not it plays a role in the prolongation of this inflammatory reaction. It could, of course, also have occurred accidentally, especially after the preceding antibiotic therapy. Further investigations are necessary to be able to decide whether this is a diagnostic feature. Another question to be clarified is the specificity of the electron microscopic picture of this fire-eater’s lung lavage, especially in view of the diagnosis of aspiration of petroleum or other low-viscosity, volatile hydrocarbides, for example after occupational accidents.

**REFERENCES**


**Esophageal Foreign Bodies Causing Obstructive Sleep Apnea in a Patient With Sturge-Weber Syndrome**

Nathaniel F. Watson, MD; and Vinish Kapur, MD, MPH

We report the case of a severely mentally handicapped 30-year-old woman with Sturge-Weber syndrome who developed obstructive sleep apnea syndrome (OSA) following esophageal aspiration of two foreign bodies, which were discovered incidentally during a neck CT scan. Initial polysomnography findings revealed significant OSA with an apnea-hypopnea index (AHI) of 40.8 events per hour. Repeat polysomnography following endoscopic removal of the foreign bodies revealed marked improvement of her OSA with a decrease of AHI to 15.6 events per hour. Our report highlights the importance of considering foreign body aspiration as a cause for OSA in mentally handicapped patients.

**Key words:** autotitrating continuous positive airway pressure; foreign bodies; mental retardation; obstructive sleep apnea; polysomnography; Sturge-Weber syndrome

**Abbreviations:** AHI = apnea-hypopnea index; GERD = gastroesophageal reflux disease; OSA = obstructive sleep apnea

**Selected Reports**
Obstructive sleep apnea (OSA) results from repetitive episodes of partial or complete pharyngeal collapse during sleep. Obesity (ie, body mass index, > 28), age, male sex, positive family history, and alcohol ingestion are major risk factors for this disorder. Developmental delay, craniofacial abnormalities, and neurologic diseases also can be associated with sleep-disordered breathing. OSA frequently results in excessive daytime sleepiness and can cause hypertension. Our patient was deaf, mute, and blind due to Sturge-Weber syndrome and thus was unable to provide any medical history. To the best of our knowledge, this is the first report of reversible secondary OSA resulting from esophageal foreign body aspiration.

**Case Report**

A 30-year-old, severely developmentally delayed woman with Sturge-Weber syndrome was brought in for evaluation due to the subacute onset of nighttime coughing, choking, snoring, and lethargy. Respiratory “rattling” sounds were heard, and increased drooling and dysphagia of solids and liquids were noted. Her medical history was significant for a left hemiparesis, cleft palate, drooling and dysphagia of solids and liquids were noted. Her medical history was significant for a left hemiparesis, cleft palate, and seizures. The initial assessment revealed a profoundly mentally handicapped lethargic woman slumped in her wheelchair with a BP of 91/46 mm Hg, a heart rate of 68 beats/min, a respiratory rate of 14 breaths/min, pulse oximetry of 98% on room air, and a temperature of 37.0°C. Examination revealed sialorrhea, facial dysmorphic features with a right cheek hemangioma, deafness, blindness, rales in the bilateral lung bases, and a right hemiparesis with increased tone.

A barium swallow examination revealed a mild delay in the initiation of swallowing but no clear obstruction or aspiration. Overnight polysomnography revealed significant OSA with an apnea-hypopnea index (AHI) of 40.8 events per hour, pulse oximetry nadir of 83%, and a desaturation index of 6.9 (Table 1, Fig 1). Treatment with an autotitrating, nasal, continuous positive airway pressure machine (Tranquility auto-CPAP; Respironics Inc; Murrysville, PA) resolved the patient’s sleep-related choking syndrome. Sleep-related abnormal swallowing syndrome results in sleep disruption due to muscular incoordination; and asynchronous pharyngeal muscles; pooling of secretions in the upper airway. Our patient had significant secretions in the upper airway. The sleep disruption and daytime symptoms.

Eighteen months later, the patient underwent cervical spine CT following a traumatic fall down stairs. Foreign bodies were incidentally found lodged in the proximal esophagus near the cricoid cartilage (Fig 2). Two plastic stars 5 cm in diameter (from tip to tip) were removed by endoscopy (Fig 3). Repeat polysomnography revealed marked improvement in her OSA with a decrease in her AHI to 15.6 events per hour and an increase in her pulse oximetry nadir to 94%. Her desaturation index normalized to 0.0 (Table 1, Fig 1). Following the discontinuation of therapy with continuous positive airway pressure, the patient remained alert during the day with a stable seizure disorder and reduced sialorrhea and dysphagia.

**Discussion**

Sturge-Weber syndrome is a neurocutaneous disorder manifested by a cutaneous vascular port-wine nevus of the face, contralateral hemiparesis and hemiatrophy, glaucoma, seizures, and mental retardation. The resulting muteness, blindness, and deafness in our patient prevented her from reporting the foreign body aspiration to caregivers, thus allowing her nighttime respiratory difficulties to be misidentified as idiopathic OSA. The foreign bodies were found incidentally during CT scanning of the neck. Other potential diagnostic imaging modalities include chest radiograph or lateral soft-tissue plain films of the neck. Neither of these methods are as effective as CT scanning in revealing aspirated foreign bodies.

Prior studies have revealed a number of factors that contribute to sleep disordered breathing including the following: hypotonic pharyngeal muscles; pooling of secretions in the upper airway due to muscular incoordination; and asynchronous medullary neuronal output to the upper airway, diaphragm, and chest wall muscles. Esophageal aspiration of a foreign body is a mechanism for the accumulation of secretions in the upper airway. Our patient had significant sialorrhea, as well as dysphagia, suggesting that excessive secretions were present and may have caused her obstructive events. In addition, gastroesophageal reflux disease (GERD) is a common occurrence in patients with OSA as well as developmental delay. GERD can precipitate cortical arousals worsening sleep fragmentation. In this manner, GERD could have exacerbated the patient’s sleep disruption and daytime symptoms.

Esophageal aspiration of a foreign body could result in a number of sleep disorders. The sleep disruption and choking observed during her episodes are consistent with sleep-related choking syndrome. Sleep-related abnormal swallowing syndrome results in sleep disruption due to inadequate swallowing of saliva, causing aspiration, choking, and coughing. Sleep-related laryngospasm was suggested by the respiratory rattling sounds that the mother noted during sleep. Her inability to communicate prevented us from evaluating the subjective aspects of these disorders. Ultimately, the patient met the diagnostic criteria for OSA, which superceded these other diagnoses.

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**Table 1—Polysomnography Results Before and After Foreign Body Removal**

<table>
<thead>
<tr>
<th>Variables</th>
<th>Before Foreign Body Removal</th>
<th>After Foreign Body Removal</th>
</tr>
</thead>
<tbody>
<tr>
<td>AHI, events/h</td>
<td>40.8</td>
<td>15.6</td>
</tr>
<tr>
<td>Total apneas, No.</td>
<td>77</td>
<td>14</td>
</tr>
<tr>
<td>Total hypopneas, No.</td>
<td>188</td>
<td>56</td>
</tr>
<tr>
<td>Pulse oximetry nadir, %</td>
<td>83</td>
<td>94</td>
</tr>
<tr>
<td>Desaturation index, No./h</td>
<td>6.9</td>
<td>0.0</td>
</tr>
<tr>
<td>TRT pulse oximetry &lt; 90%, %</td>
<td>0.8</td>
<td>0.0</td>
</tr>
<tr>
<td>Sleep efficiency, % TST</td>
<td>82.7</td>
<td>98.5</td>
</tr>
</tbody>
</table>

*TRT = total recording time; TST = total sleep time.*

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CHEST / 124 / 1 / JULY, 2003 401
The diagnosis of OSA requires the presence of excessive daytime sleepiness, witnessed apneas, and snoring. All of these characteristics were observed by the caregiver. Polysomnography confirmed our diagnosis by revealing significant OSA. Typically, decreased upper airway tone, pharyngeal fat deposition, and nasopharyngeal, hypopharyngeal, and oropharyngeal obstructions cause OSA in adults. In mentally handicapped individuals, secondary causes of OSA, such as esophageal aspiration of a foreign body, also should be considered. Barium swallow evaluations and CT scanning are useful diagnostic options, although the barium swallow can give false-negative results. Endoscopy is the diagnostic and treatment modality

**CONCLUSION**

The diagnosis of OSA requires the presence of excessive daytime sleepiness, witnessed apneas, and snoring. All of these characteristics were observed by the caregiver. Polysomnography confirmed our diagnosis by revealing significant OSA. Typically, decreased upper airway tone, pharyngeal fat deposition, and nasopharyngeal, hypopharyngeal, and oropharyngeal obstructions cause OSA in adults. In mentally handicapped individuals, secondary causes of OSA, such as esophageal aspiration of a foreign body, also should be considered. Barium swallow evaluations and CT scanning are useful diagnostic options, although the barium swallow can give false-negative results. Endoscopy is the diagnostic and treatment modality.
of choice, and should be considered in any mentally handicapped patient with OSA who has an acute or subacute onset of the disorder as well as dysphagia, odynophagia, sialorrhea, or weight loss. A high index of suspicion is necessary to make this diagnosis successfully.

REFERENCES

Figure 2. Axial CT scan of the cervical spine revealing the foreign bodies anterior to the spine and posterior to the trachea. Compression of the airway is not present.

Figure 3. Endoscopic pictures of the two plastic stars (top) that were removed as well as the proximal esophageal region (bottom) where they were embedded. Note the residual outline of the stars in the esophageal mucosa.