CASE REPORT

Multidisciplinary Management of the Airway in a Trauma-Induced Brain Injury Patient

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ABSTRACT

Laryngomalacia occurs in some brain injury patients secondary to global muscle hypotonia. Surgical therapies for epiglottis prolapse have centered around removal or reshaping of the epiglottis. This approach has brought mixed success and frequent complications. We present a case that demonstrates successful nonsurgical treatment of a 33-year-old male brain injury patient with moderate obstructive sleep apnea that is believed to be a consequence of post-brain injury nocturnal epiglottis prolapse. The presence of a tracheostomy performed at the time of emergency surgery had become an emotional and physical barrier to our patient's recovery. The tracheostomy could only be reversed if the obstructive sleep apnea disorder could be managed in an alternative fashion. A titratable mandibular repositioning appliance was prescribed and its effectiveness was demonstrated with nasolaryngoscopy and polysomnography.

After initially fitting the oral appliance, a period of accommodation and gradual protrusive adjustments was allowed. Subsequent confirmation polysomnography demonstrated improvement, but not suitable resolution, of disordered breathing events. However, an additional 1.25-mm protrusive titration of the oral appliance during the course of the confirmation polysomnogram led to therapeutic success. The patient's tracheostomy was subsequently reversed with significant quality of life benefits.

KEYWORDS: Sleep apnea, oral appliance therapy, epiglottis prolapse, brain injury, laryngomalacia

Sleep and Breathing, volume 8, number 3, 2004. Address for correspondence and reprint requests: Steven C. Scherr, D.D.S., 21 Crossroads Dr., Ste. 240, Owings Mills, MD 21117. E-mail: thesmiledoc@comcast.net. [1]Franklin Square Hospital Sleep Disorders Center, Baltimore, Maryland; [2]private practice, Owings Mills, Maryland. 1520-9512,p;2004,08,03,165,170,ftx,en;sbr00268x.

The successful use of oral appliances to treat patients diagnosed with sleep-disordered breathing is well documented.[1,2] Several crossover studies[3–5] have compared oral appliance therapy to continuous positive airway pressure (CPAP). CPAP was nearly always effective. However, patient compliance was often unsatisfactory. The use of a mandibular repositioning device (MRD) was often, but not always, effective. Patients overwhelmingly preferred the oral appliance, and MRD compliance exceeded that of CPAP.

Smith[6] used magnetic resonance imaging to study the impact of an MRD on airway volume. He reported an oral appliance increased measured airway volume by 32%. Fransson and associates[7] and Liu and colleagues[8] each found a 10% increase in mean pharyngeal area using cephalometric measurements. Several mechanisms of action have been attributed to oral appliances. These include anterior posturing of the tongue away from the airway,[9] elevation and forward displacement of the hyoid bone,[10,11] and encouragement of mouth breathing in addition to nasal breathing.[12] The latter negates the need to press the tongue and soft palate together to seal the oral airway. In addition, appliance pull on the palatoglossus and palatopharyngeal muscles expands the velopharynx. Activation of the superior pharyngeal constrictor muscle reduces pharyngeal redundancy and improves airway patency.

The degree of mandibular protrusion is thought to impact treatment success.[13,14] As the mandible is progressively postured anteriorly, the airway is expanded. Some otherwise resistant patients significantly benefit when the MRD postures the mandible forward of their maximum active protrusion,[15] possibly by passively stretching the temporomandibular ligaments.[16] For others, overextension may narrow the airway laterally and restrict the cross-sectional airway.[17] As a result of individual anatomic and physiologic differences, the newer MRD appliances that afford easy titration during polysomnography present advantages that improve treatment success.[18] This is comparable to the way CPAP is titrated in the laboratory to optimize both its effectiveness and patient comfort.

We present a case report that demonstrates the benefit of active titration of a variable mandibular repositioning oral appliance during polysomnography. The patient had received a tracheostomy during emergency surgery following head trauma. He subsequently received an overnight polysomnogram (PSG) and was diagnosed with moderate obstructive sleep apnea. His otolaryngologist suspected acquired laryngomalacia, related to global muscle hypotonia, as a contributing factor.

A mandibular repositioning appliance was fabricated that was capable of easy titration in the sleep laboratory setting. After an initial period of accommodation and appliance adjustment, the patient presented to the Franklin Square Hospital Sleep Disorders Center for confirmation of oral appliance effectiveness and titration during polysomnography. A series of fine adjustments to mandibular posture was performed until the respiratory disturbance index was within safe limits. The tracheostomy was later reversed, bringing significant quality of life benefits.

METHODS

The male patient treated was 33 years old when he sustained serious trauma to the right side of his head from a work-related injury. He was not expected to survive the resultant cerebral vascular accident. The patient was rushed to Maryland General Hospital for emergency surgery. A tracheostomy was performed in an effort to keep him alive. Over the next 2 years, the patient markedly improved with the energetic assistance of talented therapists and a wonderfully supportive family. He continued to experience significant weakness in the left extremities, the left facial and masticatory muscles, and the left side of the tongue.

The patient learned to speak again. However, his speech pattern was somewhat slurred and labored. He attributed ongoing speech frustration to the tracheostomy and a transitional removable upper partial denture that replaced a maxillary

incisor lost during emergency intubation. He was eager to have the tracheostomy reversed. An overnight PSG was completed at Franklin Square Hospital Center. At the time of the sleep study, he had a body mass index of 33.1 kg/m^2. An average of 21.6 disordered breathing events (DBEs) per hour with moderate-severe snoring were observed. DBEs were associated with electroencephalogram arousals and oxyhemoglobin desaturations to a nadir of 84%. A diagnosis of moderate obstructive sleep apnea was made. The tracheotomy could not be safely removed until alternative measures were confirmed to control sleep-disordered breathing. The tracheostomy orifice continued to be plugged during the day and left open at night.

The patient preferred to use an oral dilator appliance for the management of sleep apnea. This was considered to be less restrictive than CPAP and more conservative than surgical alternatives. A Thornton Adjustable Positioner, or TAP, (Airway Laboratory, Dallas, Texas, USA) was selected because it may be easily adjusted during a PSG by either the patient or a nonprofessional caregiver. This is important, because most sleep laboratory technicians are not comfortable manipulating oral appliances. As a result, we developed a protocol that provided for adjustments without the need for direct technician contact with the appliance. In addition, the TAP appliance is advantageous because it is titratable in 0.25-mm increments over a wide range of mandibular advancements. This creates the opportunity to dilate the airway to be both maximally effective and comfortable for the patient.

The patient's TAP appliance was inserted, and firm, stable seating was confirmed. The appliance was temporarily set to the patient's maximum active mandibular protrusion position, and the excess extension of the threaded hook was cut off flush with the end of the adjustment knob. This created a reference position from which to gauge future adjustments. The appliance was then reset to 70% of his maximum active protrusion. For safety reasons, the patient was asked to continue unplugging the tracheostomy during sleep.

With the help of a caregiver, the patient advanced his mandibular posture in 0.25- to 0.50-mm nightly increments. He was unable to return in 2 weeks as recommended. When he did return after 5 weeks, the mandibular posture was 1.1 cm anterior to the maximum active protrusion reference. The patient reported mild lower front teeth sensitivity for the first 30 minutes after appliance removal. He considered this minor and had no other complaints. He was asked to discontinue further appliance adjustments.

RESULTS

An outpatient awake nasolaryngoscopic examination was performed with and without the TAP appliance in place as shown in Figure 1. When the TAP appliance was worn, the epiglottis took on a more vertical and anterior posture with cross-sectional expansion of the superior laryngeal orifice.

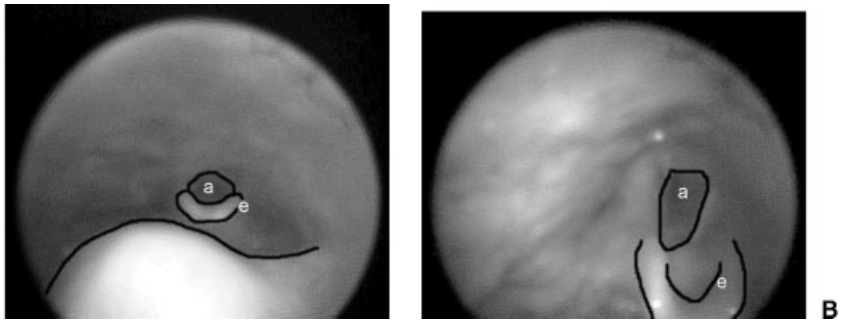

Figure 1 (A) No oral appliance. The epiglottis has a horizontal posture. (B) With oral appliance. The epiglottis has a vertical posture and the laryngeal orifice is expanded. a, laryngeal airway; e, epiglottis.

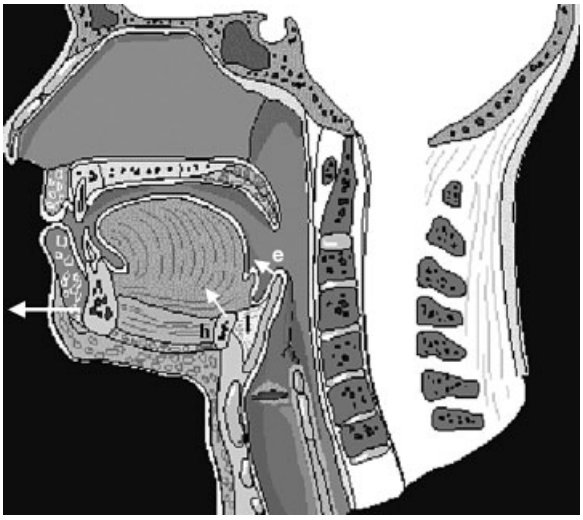

Figure 2 Mandibular protrusion leads to elevation and anterior displacement of the hyoid bone. In turn, the hyoepiglottic ligament seems to lift the epiglottis and open the airway. h, hyoid bone; l, hyoepiglottic ligament; e, epiglottis. (Adapted from ADSM Educational CD-ROM, Chicago, IL; 2001. Reprinted with permission of the Academy of Dental Sleep Medicine.)

This may be related to elevation and anterior displacement of the hyoid bone during oral appliance therapy (Fig. 2). The hyoepiglottic ligament may be responsible for the improved epiglottis position observed.

A PSG was performed at Franklin Square Hospital Center with the TAP appliance in use and the tracheostomy plugged. The lowest blood oxygen saturation (SpO_2) improved to 93% and the apnea hypopnea index (AHI) improved marginally to 17.4 DBEs per hour, when compared with the earlier diagnostic PSG (SpO_2 of 84, AHI of 21.6). Additional total protrusive appliance adjustments of 1.25 mm were made during the study, which further lowered the AHI to 7.1 DBEs per hour.

DISCUSSION

Brain injury is the leading cause of death from blunt trauma.[19] Cerebral hemorrhage, and the neurologic deficit that results, leads to reduced muscle and ligament tone. Epiglottis prolapse occurs in some brain injury patients[20] secondary to global muscle hypotonia.[21,22] Hyoepiglottic ligament laxity promotes posterior displacement of the epiglottis. An upper airway obstruction may subsequently be induced by aspiration of the flaccid epiglottis during inspiration. Surgical therapies for epiglottis prolapse have centered around removal or reshaping of the epiglottis. This approach has brought mixed success and frequent complications. The case presented demonstrates successful nonsurgical treatment of post-brain injury nocturnal epiglottis prolapse and concomitant obstructive sleep apnea. As a result, the tracheostomy was successfully reversed and the patient has continued to make ongoing progress in his recovery.

The patient was extremely compliant, using the oral appliance 7 nights per week for 6 to 8 hours per night. He considers the appliance to be very comfortable and much preferable to the tracheostomy. Nasopharyngeal laryngoscopy confirmed expansion of the airway while awake. Polysomnography with the oral appliance in place demonstrated significant improvement compared with the baseline study. However, the titrated position of the oral appliance that was clinically determined prior to polysomnography was only partially successful because significant disordered breathing events continued to occur during rapid eye movement sleep. Additional small incremental advances in mandibular posture resulted in further reduction in the respiratory disturbance index. The total additional mandibular advancement was 1.25 mm over the course of the entire PSG.

It is interesting to note that the final position of the oral appliance postured the patient's mandible anterior to his maximum active protrusion position as measured on the day of appliance insertion. This may be the result of gradual accommodation of the horizontal fibers of the posterior temporalis to overcome initial muscle splinting. The masseters, medial pterygoids, and vertical fibers of the anterior temporalis direct a superior force on the mandibular condyle against the slope of the articular eminence.

This may similarly restrict mandibular advancement at initial appliance placement. It is also possible that the temporomandibular joint ligaments may have become stretched by the orthopedic forces developed during the course of oral appliance therapy. The patient has exhibited no temporomandibular joint or muscular symptoms, and no joint sounds have been detected at the time of this writing. He continues to be monitored on an ongoing basis.

This case demonstrates the benefit of titrating a variable oral appliance in the sleep laboratory. Many patients previously considered to be partial responders have the potential to improve further with small additional alterations in mandibular posture. Even when the patient is unable to tolerate sufficient mandibular advancement for complete success, partial opening of the airway may permit the simultaneous use of nasal or oral CPAP at reduced, more tolerable pressures than required with CPAP alone. Research is needed to confirm these findings in a larger patient population and to determine the optimal PSG protocol for appliance titration.

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