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A Case of Hypoglossal Nerve Stimulator-Resistant Obstructive Sleep Apnea Cured With the Addition of a Chin Strap

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INTRODUCTION
Obstructive sleep apnea (OSA) affects 13% of men and 6% of women in North America. The gold standard treatment is continuous positive airway pressure (CPAP). However, tolerance of CPAP is low due to the social factors of wearing the device as well as discomfort from the air pressure. In 2014, the Food and Drug Administration (FDA) approved the first hypoglossal nerve stimulation (HGNS) therapy for use in adult patients who failed CPAP, with apnea-hypopnea index (AHI) of 15 to 65 events/hour, and who do not have complete concentric collapse of the velopharynx. In posttrial clinical use, this therapy is found to be well tolerated, with improvement of AHI <15 events/hour in 68% to 95%. Use of HGNS results in improvement of sleep apnea by protruding the tongue, improving base of tongue and palatal collapse, and opening the airway. However, there is a population of patients who do not respond to HGNS. Although some of the predictors of poor therapy response have been identified, such as body mass index (BMI) >32 kg/m² and complete circumferential velopharyngeal collapse noted on drug-induced sleep endoscopy (DISE), the other predictors of poor outcomes have not yet been defined. How to further treat these patients has remained an open question in sleep science. We report a case of a patient who has failed CPAP treatment, did not achieve adequate management of OSA with multiple surgeries, and initially did not respond sufficiently to HGNS therapy. The patient subsequently improved with the addition of a chin strap to nighttime regimen.

CASE REPORT
A 43-year-old nonobese male with history of tonsillectomy, gastrosophageal reflux disease, and OSA first presented to the otolaryngology department with complaints of continued daytime sleepiness despite multiple interventions. The patient reported continued symptoms with CPAP use and was unable to tolerate the machine. After a trial of oral appliances, which did not improve the patient’s sleep apnea, the patient was first referred to otolaryngology for further management, at which time overall AHI was 29 events/hour, with desaturations to 88%. The patient’s exam at that time demonstrated redundancy of the uvula and a slight underbite. The patient subsequently underwent maxillomandibular advancement, uvulopalatopharyngoplasty, and hyoid suspension. Postoperative polysomnogram (PSG) demonstrated AHI of 18 events/hour overall and 30 events/hour while supine. However, in the lateral position, AHI was now 4 events/hour. Given the patient’s normal AHI in the lateral position, the patient was recommended to trial supine avoidance and return should he be unable to tolerate the change.

Three years later, the patient presented to the senior author (M.V.S.) reporting inability to tolerate lateral sleeping and with continued daytime sleepiness. The patient was referred for PSG, which demonstrated...
overall AHI of 21 events/hour, supine AHI of 43 events/hour, and lateral AHI of 13 events/hour, along with four paroxysmal leg movements per hour. Given the patient’s continued OSA despite multiple interventions, the patient was recommended to undergo DISE to evaluate level of obstruction. During DISE, the patient was found to have complete anterior–posterior collapse of the velopharynx. There was no significant collapse of the oropharynx; however, complete anterior posterior collapse of the tongue and complete collapse of the epiglottis was identified. Given the patient’s lack of circumferential velopharyngeal collapse, the patient’s appropriate BMI of 21 kg/m$^2$, and demonstrated failure of other methods, HGNS therapy was recommended, and the patient underwent the procedure without complications.

After appropriate postoperative waiting period to allow the surgical sites to heal, the device was activated and titrated for effect with PSG. The initial postoperative PSG with device optimization recommended settings of 0.7 volts at the default electrode configuration of $[-0+]$, although fully effective setting could not be tolerated. The patient initially reported partial improvement with improved snoring but continued to report daytime sleepiness and discomfort with the required voltage settings. The patient underwent multiple titrations without improvement. At that point, home sleep test (HST) was performed and demonstrated a supine AHI of 27 events/hour and a nonsupine AHI of eight events/hour. Given the patient’s continued OSA despite the HGNS therapy, the decision was made to evaluate efficacy of the treatment under DISE. Without the stimulator, the results of DISE demonstrated complete anterior–posterior collapse of the velopharynx, epiglottis, and tongue (Fig. 1A). With the stimulator on at the original settings, base of tongue, and epiglottis collapse was demonstrated, although partially improved with mouth closure. When the stimulator was set to 0.9 volts at an alternative electrode configuration $[-o-]$, partial relief of the airway obstruction due to epiglottis collapse was demonstrated (Fig 1B). However, when the patient’s mouth was closed, the airway opening was significantly improved, and the optimal settings were noted to be at $-0-0.8$ volts (Fig. 1C). Based on improved airway obstruction with mouth closure, the patient was recommended to use a chin strap as well as HGNS. With these measures, the AHI was found to be within normal limits at four events/hour on follow-up PSG, with the AHI of four events/hour in nonsupine position and three events/hour in supine. The patient reported improved daytime sleepiness and snoring symptoms, although the patient has occasional jaw soreness due to the chin strap usage.

**DISCUSSION**

The definition of success in HGNS has become an open question in the years since the first device was approved by the FDA. Previous measures of success were entirely based on objective measures such as the AHI. Based on the Sher et al. study in 1996, the definition of success has been AHI decrease $\geq 50\%$ and AHI $< 20$ events/hour. However, postoperative HGNS patients often report significant clinical improvement despite not meeting the above definition of successful OSA treatment. Using the Sher et al. criteria, success of HGNS in the STAR trial was 66%. In our population, we have implanted and analyzed outcomes for 29 patients to date. Out of the 29 patients, 15 (51.7%) have achieved an AHI $< 5$ events/hour and 24 (82.8%) have achieved an AHI $< 15$ events/hour prior to performing advanced therapy optimization for nonresponders. Previous studies have found that patients with high BMI and concentric velopharyngeal collapse have poor outcomes with HGNS, and for this reason are not considered to be good candidates for the therapy. However, the characteristics of the 30% of patients who are currently undergoing implantation and not seeing sufficient AHI improvement remain unknown. Advanced titration with DISE can be a very important measure for these patients in terms of workup or further steps in the treatment algorithm. Our patient would have fallen into the category of patients who did not benefit from HGNS and will have undergone an ultimately unsuccessful surgery. With the use of advanced titration with DISE, as well as trial of...
adjunctive measures, the patient’s residual obstruction was identified along with potential treatment options.

Heiser describes a similar case of a patient who after implantation had good initial therapy response. However, follow-up HST after 6 months demonstrated recurrence of obstruction with HGNS at the original settings. The patient underwent DISE, which demonstrated obstruction at the level of the epiglottis. In his situation, advanced titration of the stimulator by changing from a bipolar to a monopolar setting alleviated the obstruction. Heiser postulates that the monopolar setting allows for stimulation of the C1 branch of the hypoglossal nerve in the situations when it is not included in the cuff due to its low anatomic position. In our patient’s situation, changing the settings was not sufficient to improve airway obstruction, and it was the additional manipulation of closing his mouth that reduced collapsibility of the oropharynx.

Traditional oral appliances for sleep apnea include mandibular advancement splints (MAS) and tongue-retaining devices. These devices have mixed outcomes depending on site of obstruction. Mandibular advancement splints directly increase the retrolingual anteroposterior distance and stabilizes the airway. Additionally, the muscles have a synergistic response such that in responders the lateral airway size is also increased. Tongue-retaining devices result in the best resolution of retrolingual collapse by pulling the tongue anteriorly with a suction mechanism, counteracting the effects of gravity while the patient is supine. The chinstrap is a more controversial device, which has been shown to be successful in individual case reports for people with retrolingual supraglottic collapse. However, a randomized control trial found no efficacy in OSA or snoring in the general OSA population, possibly due to the elevation of the tongue base secondary to submental pressure from the device. Lee et al. describe the use of a MAS style oral appliance in a patient who did not achieve sufficient response to HGNS. They describe a good response with AHI of 3.5 down from 11.6 events/hour when the HGNS and MAS were used together as compared to the HGNS alone. All of these appliances are associated with jaw discomfort, and chin straps in particular when combined with HGNS can cause further discomfort by preventing the tongue from protruding during stimulation.

The success of all treatments for OSA, including HGNS is predicated on our as yet incomplete understanding of the neuromuscular anatomy of the tongue, palate, and oropharynx. For this patient, the HGNS provided tongue base advancement to reduce retrolingual collapse with the increased genioglossal muscle activation from the alternative electrode activation to reduce epiglottic collapse. In addition, the mouth closure with the chin strip reduces overall upper airway collapsibility, which in conjunction with the HGNS reduces both retrolingual and retropalatal collapse. Each patient’s type of dysfunction and requirements for intervention vary; it is the responsibility of the physician to continue diagnostic measures after apparent failures, such as with advanced titration with DISE. Furthermore, the use of multiple interventions to address OSA may make the difference between a cure and failure for a significant number of patients.

CONCLUSION

The use of postoperative DISE in patients who do not respond to HGNS can provide important information regarding therapeutic options. In this particular patient’s case, the snoring was resolved and AHI was normalized with combination therapy of HGNS and chin strap device.

BIBLIOGRAPHY