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Case Report

A Comprehensive and Multidisciplinary Approach to Obstructive Sleep Apnea: Case Examples

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Abstract

Positive airway pressure (PAP) therapy is well established as the standard of care for most patients with obstructive sleep apnea (OSA), however, up to 50% are not successfully treated with this approach and their further care remains a challenge. The value of engaging a multidisciplinary team in the diagnosis and management of OSA has been well recognized for over 30 years; however, the difficulty has been translating this principle into clinical practice. We present a series of selected patients with a diagnosis of OSA who initially failed PAP therapy but were successfully treated using a multidisciplinary and evidence-based approach that is reproducible at other sleep centers.

Key Words: obstructive sleep apnea, sleep endoscopy, positive airway pressure

List of Abbreviations

Adaptive servo ventilation (ASV)

Apnea-hypopnea index (AHI)

Cognitive behavioral therapy for insomnia (CBT-I)

Drug induced sleep endoscopy (DISE)

Hypoglossal nerve stimulator (HGNS)

Obstructive sleep apnea (OSA)

Polysomnogram (PSG)

Positive airway pressure (PAP)

Uvulopalatopharyngoplasty (UPPP)

Introduction

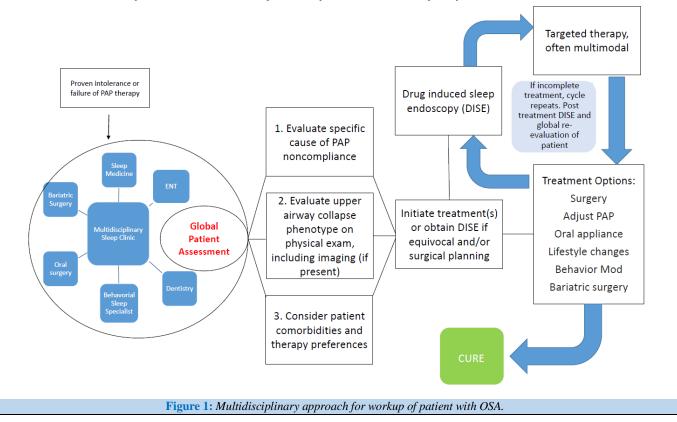
Despite several decades of advancement in our understanding of the pathophysiologic mechanisms, sequelae, and treatment of obstructive sleep apnea (OSA), there remain substantial challenges to effective management. OSA is recognized as a complex chronic condition with far reaching health consequences. Attention to this disease is imperative now as its prevalence continues to rise, with many estimates suggesting prevalence as high as 49.7% in the general population¹. Positive airway pressure (PAP) therapy is well established as the standard of care for most

patients with OSA. Unfortunately, only about half of patients prescribed PAP are successfully treated or adherent to treatment [1]. PAP nonadherence remains challenging to address, and management is often limited to troubleshooting issues with the PAP device or interface, rather than examining underlying factors that may contribute to non-use. Alternative surgical and non-surgical therapies may be considered, but the ultimate success of any therapy depends on several factors, including patient acceptance and the suitability of the patient's functional anatomy. Without a foundational understanding of the upper airway, treatment options may be offered in a haphazard or trial-and-error fashion. This approach can result in suboptimal selection of sleep surgery candidates, which may in part account for lower success rates of certain sleep surgery procedures for OSA. For example, when Fujita and colleagues first described the uvulopalatopharyngoplasty (UPPP) in 1964, they were not able to identify specific sleep or respiratory parameters that differentiated a successful response to surgery. Further research in proceeding years has led to a more global understanding of upper airway dynamics and patientspecific factors contributing to OSA, leading to lower failure rates with PAP alternatives [2].

Multidisciplinary approach to successful treatment of patients with OSA- Case Examples

The value of engaging a multidisciplinary team in the diagnosis and management of OSA is well recognized [3]. However, the challenge has been translating this principle into clinical practice since clinicians have the tendency to work in "silos" rather than engaging outside their area of expertise. Our experience has been that the workup to determine optimal treatment is much more individualized and heterogeneous than traditionally practiced. We herein present a series of selected patients with OSA who failed PAP therapy and who were treated successfully using a multidisciplinary and evidence-based approach that can be reproduced at other sleep centers.

A multidisciplinary and individualized approach can identify the root causes of PAP failure, which may not be evident in the sleep laboratory or sleep clinic. This includes behavioral health evaluation of PAP tolerance and sleep habits, upper airway evaluation with drug induced sleep endoscopy (DISE), and identifying candidacy for alternative treatments such as oral appliances or sleep surgery [4]. A multidisciplinary sleep center offers tailored therapy to patients who do not respond to initial PAP therapy while considering a wide spectrum of both nonsurgical and surgical treatment options. In the following section, we present challenging cases of patients with OSA who directly benefited from a multidisciplinary evaluation and treatment.



For patients proven to have persistent intolerance to PAP therapy drug induced sleep endoscopy (DISE) is used to visualize the anatomical level of dynamic obstruction and subsequently decipher the correct approach on a broad treatment algorithm. Surgical options include traditional approaches such as tonsillectomy, uvulopalatopharyngoplasty, as well as newer treatments such as hypoglossal nerve stimulator. After each treatment or series of treatment a patient is assessed for cure. If incomplete treatment then DISE can be used to reassess, or patient can revisit sleep clinic.

Case #1 – OSA treated with hypoglossal nerve stimulator (HGNS) and cognitive behavioral therapy

72-year-old woman presented with a history of chronic insomnia and severe OSA. Her apnea-hypopnea index (AHI) was 31. PAP titration study confirmed improvement with CPAP, which she was unable to tolerate due to claustrophobia and pressure intolerance despite empiric pressure adjustments and trials of different masks. Subsequent DISE revealed an isolated base of tongue obstruction as the cause of her OSA. She underwent implantation of HGNS. Although this was an effective treatment for her anatomical obstruction, demonstrated by follow-up endoscopy and polysomnogram (PSG), she described anxiety and fear of frequent awakenings after HGNS placement, resulting in minimal use of the implant. In addition, her longstanding insomnia persisted, with average sleep latencies of 120 minutes, further limiting effective use of the implant. After multidisciplinary evaluation she was referred for cognitive behavioral therapy for insomnia (CBT-I), to address sleep onset insomnia. Treatment also incorporated relaxation training and graduated HGNS stimulation exposure to reduce anticipatory anxiety about the sensation of stimulator activation. The patient would activate the device at the lowest tolerable stimulation setting for 10 minutes once a day while engaged in a relaxing, pleasant activity. Once able to tolerate therapeutic levels of stimulation for 60 daytime minutes, she was able to activate overnight use without complaint. In this case, incorporating behavioral sleep medicine in the patient's treatment resulted in dramatic improvements in therapy adherence, insomnia symptoms, sleep quality, and treatment of her OSA over the course of eight weeks. At the conclusion of therapy, her residual AHI using HGNS was 4.7, using therapy on average 37 hours per week.

Case #2 – Complex sleep apnea treated with adaptive servo ventilation (ASV) and positional therapy

81-year-old woman presented with severe complex sleep disordered breathing in the context of obesity and diastolic heart failure. Initial (PSG) revealed severe mixed obstructive and central sleep apnea (AHI 127) that did not respond adequately to PAP due to persistence of central apneas. An estimated 25% of her central apneas were associated with Cheyne-Stokes respiration with a cycle length of 50 seconds. Subsequent titration using bilevel spontaneous-timed PAP and adaptive servo-ventilation revealed ongoing central apneas which triggered the bilevel's backup rate, although application of pressure resulted in an airflow waveform only in the mask, without chest or abdominal movement, suggesting some upper airway collapse which was not overcome by high inspiratory pressures. She received a 30-day trial of acetazolamide to address central apnea [5], without significant improvement. She then underwent a PAP-assisted DISE, which included a flexible endoscopy through her CPAP mask while under propofol sedation. This revealed lower pharyngeal sidewall collapse and base of tongue obstruction that was induced by the facemask itself. Her mouth breathing was exacerbated by overly tight mask straps that were necessary to prevent mask air leakage but displaced her jaw posteriorly, inducing severe base of tongue (BOT) collapse, which could not be overcome with PAP therapy. During DISE, the effect of a neck extension pillow with a chin strap was found to induce a beneficial effect and help eliminate BOT obstruction. These two simple adjunct therapies in combination with ASV therapy were successful in improving her OSA to a residual AHI of 4.8. She subsequently used ASV at home with a nasal mask, chinstrap, and neck extension pillow, with device downloads showing ongoing normalized AHI and adherent usage with symptomatic improvement.

Case #3 – OSA managed with multi-level sleep surgery for multi-levels upper airway obstruction

60-year-old woman presented with a history of severe OSA (AHI 52) and PAP intolerance despite trials of multiple masks and pressure delivery modes. DISE revealed obstruction around the palate and base of tongue and therefore multilevel surgery was recommended. Her palate obstruction was addressed with expansion sphincter pharyngoplasty, and post-operative AHI was reduced only marginally to 46. Follow-up DISE confirmed that her retropalatal obstruction had resolved and that she had persistent base of tongue obstruction. She then underwent HGNS surgery, and post-treatment residual AHI was reduced to 0 with low therapeutic settings on the neurostimulator and high adherence rates of above 50 hours per week.

Case #4 – Morbid obesity with severe OSA treated with multiple upper airway surgeries and weight loss

47-year-old man presented with a history of severe OSA (AHI 124) and morbid obesity (body mass index 44) who was intolerant of PAP therapy due to nasal congestion and pulling his mask off overnight. Patient had been seen at other tertiary sleep centers with no alternative therapy options offered except CPAP adjustment or bilevel therapy given the severity of his OSA. He presented with worsening of symptoms and in light of his morbid obesity and very unfavorable upper airway anatomy underwent a tracheostomy. His post-operative AHI with a tracheostomy improved to 8, but he had persistent hypoxic events (SpO2 nadir 80%) from distal tracheal collapse related to his obesity. He then underwent bariatric surgery due to residual OSA symptoms and desire for tracheostomy decannulation. After losing 60 pounds post-operatively, the

patient accidentally decannulated himself and the tracheostomy site closed. Following bariatric surgery, his residual AHI without tracheostomy was 48. DISE revealed severe multi-level complete concentric collapse at the velopharynx, oropharynx, and hypopharynx despite a significant weight loss, which was suspected to be due to a hypoplastic maxilla and mandible. A maxillomandibular advancement (MMA) procedure was performed, after which his AHI improved from 48 to 27.5. DISE was repeated after MMA and showed a residual base of tongue obstruction aggravated by mouth opening, inducing base of tongue obstruction responsible for his persistent elevated AHI. To address this, he underwent HGNS surgery and his residual therapeutic AHI improved to 3.8.

Discussion

Positive airway pressure remains the mainstay of therapy for patients with OSA, but high rates of treatment failure due to nonadherence necessitate an exploration of alternative or adjunctive therapies. Although many instances of PAP nonadherence are easily addressed via pressure adjustment, trials of different mask interfaces, adjustment of humidification, and other comfort features, many patients remain unable to use PAP. Selection of effective treatments requires an understanding of the reasons for PAP failure, the patient's upper airway anatomy, and access to a broad range of specialists across the fields of dentistry, otolaryngology, oromaxillofacial surgery, behavioral health, and bariatric surgery. Moving from a traditional model of working in "silos" to a multidisciplinary and multi-specialty approach can significantly improve patient care, particularly when complex or multi-modal therapies are required.

At our institution, patients who fail PAP therapy are seen in our multispecialty sleep clinic, which includes providers in the areas of sleep medicine, otolaryngology, oral surgery, dentistry, and behavioral sleep medicine. Patients may have shared appointments with multiple providers or are arranged for follow up with specific providers when necessary. Additionally, patients are discussed at a multidisciplinary case conference. There is also close collaboration with our weight management and bariatric surgery programs. The multidisciplinary sleep team will not only take into consideration the specific cause of PAP nonadherence, but will also consider the upper airway collapse phenotype as noted on physical examination, DISE and imaging (e.g. cone beam computed tomography). Furthermore, patients' specific comorbidities and therapy preferences are factored into the treatment approach.

All OSA cases presented in this series tried and failed PAP therapy for various reasons. The severity of their sleep apnea, body habitus, and upper airway anatomy responsible for upper airway obstruction, as noted on DISE, was different in each case, resulting in a different treatment recommendation for each patient. It is our observation that many patients who fail PAP therapy experience multilevel anatomical obstruction and require either multilevel surgery or multimodal therapy that combines surgery with other therapies. We have found that using DISE as part of the evaluation has great utility for identifying physiological factors contributing to intolerance, thus guiding selection of optimal surgical and nonsurgical treatments. DISE, first introduced in 1991 by Croft and Pringle [6], is an endoscopic evaluation of the dynamic airway under pharmacologic sedation. The endoscopic evaluation places an emphasis on structures or factors that may potentially be correctable to improve airway patency, such as the palate or base of tongue, as well as the effect that head and neck positional changes may directly have on these areas. DISE helps to identify appropriate surgical techniques and medical management options for patients, and it has also shown to predict outcome of mandibular advancement device treatment [7]. Sleep surgical procedures such as tonsillectomy, UPPP, expansion sphincter pharyngoplasty, maxillomandibular advancement, genioglossus advancement, hyoid suspension or HGNS are selected with a more

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targeted approach via DISE. In order to achieve consistently good outcomes, a multidisciplinary sleep center should have access to a sleep surgeon who is experienced with DISE and understands the indications and contraindications of the various existing sleep surgical procedures as well as the concept of multilevel upper airway collapse, which may require more than one sleep surgical procedure to be managed appropriately. A particularly novel application of DISE is to evaluate PAP non-responders with mask induced upper airway collapse, as presented in Case #2 and as reported recently by our colleagues [4].

Whether managed medically or surgically, we have also found that patients with OSA can benefit from inclusion of a behavioral expert on the team to facilitate adjustment and adherence to various treatment options. The first case presented here highlights the importance of addressing psychological components that may conflict with appropriate OSA treatment and adherence. This patient's underlying insomnia combined with her anxiety related to the anticipated sensation of HGNS caused inadequate use of the implant and further worsened her sleep onset insomnia. There is a high correlation between behavioral health and OSA; the rates of affective disorders such as depression or anxiety are as high as 63% among individuals with OSA [8]. Treating OSA has the potential to decrease affective symptoms through increased REM sleep and improved emotional memory processing [8]. However, in this case the patient's symptoms were in response to her sleep disorder and treatment, so a referral for behavioral health treatment was beneficial. Following a comprehensive behavioral sleep medicine evaluation to identify the factors perpetuating her sleep disturbance, the patient was treated with a combination of CBTI-I and graduated exposure therapy for adjustment to the hypoglossal nerves stimulator. The combination of both approaches targeted the factors maintaining her sleep disturbance (anticipation and anxiety about device activation, fear of not being able to fall asleep in time, lack of stimulus control, conditioned arousal, and poor sleep hygiene). While sleep hygiene education alone is not a sufficient treatment for chronic insomnia, it has been shown to improve sleep issues among individuals with OSA [9]. Thus patients with OSA may benefit from the inclusion of sleep hygiene education in their treatment.

Finally, given that the rate of obesity within OSA is as high as 80% [10], we have found great utility to the inclusion of weight management and bariatric providers as part of the multidisciplinary team. Patients with OSA are known to have increased base of tongue and parapharyngeal fat deposition, which can decrease both PAP and surgical effectiveness. Up to 20% reduction in AHI can be achieved with lowering body weight by 10% [10]. Unfortunately, patients with comorbid obesity and OSA are often not able to achieve and maintain a significant weight loss through lifestyle modifications alone. Therefore, we routinely refer patients for consultation to our weight management and bariatric surgery programs. Additionally, patients with persistent OSA and/or PAP intolerance after weight loss surgery are commonly better candidates for sleep surgery after weight loss [10].

Improving the quality of life for patients with OSA requires elimination of the upper airway obstruction while also addressing coexisting medical problems. PAP failure is common, and traditional treatment models that do not incorporate multimodal and multidisciplinary approaches to treat OSA are not optimal. Such patients benefit significantly from a multidisciplinary and patient-centered treatment approach with shared management between specialties. This approach should be adopted early on in the diagnosis and management of sleep apnea as it has proven to be beneficial to most patients with OSA who do not benefit from PAP therapy alone. The cases presented herein offer several examples of multidisciplinary collaboration at our Sleep Center that we believe can be reproduced at other centers to advance the care of patients with OSA.

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