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CASE REPORTS

DISE-PAP: a method for troubleshooting residual AHI elevation despite positive pressure therapy

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Despite excellent positive airway pressure (PAP) adherence, a subset of patients with obstructive sleep apnea experience residual elevation of the apnea-hypopnea index (AHI). Drug-induced sleep endoscopy during PAP application provides an opportunity to examine the anatomic effect of PAP therapy on the upper airway and to troubleshoot refractory residual AHI elevation. We present a patient who demonstrated persistent moderate-severe AHI elevation during titration polysomnogram and subsequent data download reports despite numerous mask refits, chin strap, positional modifications, and multiple pressure and mode adjustments in both the clinic and sleep laboratory settings. Drug-induced sleep endoscopy was performed with the flexible endoscope passed through the PAP circuit into the upper airway. Jaw laxity and associated mandibular retrusion at sleep onset was found to result in a complete fixed tongue base obstruction that PAP therapy, delivered via the patient's oronasal interface, was unable to overcome. Various strategies to overcome these obstacles are discussed.

Keywords: refractory obstructive sleep apnea, drug induced sleep endoscopy, positive airway pressure therapy

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INTRODUCTION

Strategies to improve positive airway pressure (PAP) therapy effectiveness most commonly target patient adherence, including measures to increase comfort, reduce device-related side effects, and overcome psychosocial barriers.[1] We occasionally encounter "PAP failure" patients who struggle, not due to inadequate adherence, but rather to inadequate PAP efficacy. In this subset, patient motivation and adherence are high, with good self-reported and objective therapy use; however, residual apnea-hypopnea index (AHI) elevation and associated persistent symptoms remain problematic. For many, efficacy is improved with therapy modifications such as mask, pressure, and/or mode changes. For others, patient modifications such as positional therapy, weight loss, lowering nasal resistance, or upper airway surgery may improve outcomes.

Despite these troubleshooting options, some patients continue to experience refractory AHI elevation. Reiter et al studied a population of 217 individuals and found that on manual AHI scoring, 32.3% had residual AHI > 5 events/h, 23% had an AHI > 10 events/h, and 7.8% had persistent AHI > 15 events/h.[2] The anatomic basis for residual obstructive respiratory events on PAP therapy is not completely understood and likely varies from patient to patient. Drug-induced sleep endoscopy (DISE) is a widely used diagnostic tool for evaluating upper airway anatomy and patterns of collapse under conditions that mimic an obstructive sleep-disordered breathing state. It is most commonly employed preoperatively in patients who have failed medical therapy and are considering upper airway reconstructive procedures or neurostimulation therapy. Additionally, DISE may be used to troubleshoot the medical management of PAP-adherent patients who struggle with residual AHI elevation.

REPORT OF CASE

A 58-year-old female with a history of moderate-severe obstructive sleep apnea (OSA) and glucose intolerance presented for an evaluation of persistent symptoms of unrefreshing sleep, daytime sleepiness, and cognitive dysfunction despite continuous (CPAP) and bilevel (BPAP) positive airway pressure therapy for eleven years. She had previously declined a mandibular repositioning device due to financial barriers and concern for dental side effects. The incoming BPAP prescription from an outside sleep laboratory included an inspiratory pressure (IPAP) of 13 cm H_2O, an expiratory pressure (EPAP) of 8 cm H_2O, and an oronasal interface (full face mask). Her initial titration study, performed when her body mass index (BMI) was 34.5 kg/m^2, reported normalization of AHI at those settings. Symptoms persisted however despite excellent patient-reported accommodation and use with no significant mask- or pressure-related adverse side effects. Objective data monitoring software corroborated her reports with > 99% of nights used and > 7 hours of average nightly use without significant mask leak. However, for years, the estimated residual obstructive AHI was consistently between 15 and 30 events/h. She had no findings of or risk factors for central sleep apnea. Otolaryngologic examination was notable for mandibular deficiency, tongue ridging, modified Mallampati III, and a decreased BMI of 30.4 kg/m^2. No significant nasal obstruction was identified.

Over the following year, she subsequently underwent multiple troubleshooting attempts in our sleep medicine clinic to improve PAP efficacy: (1) mask refits to a nasal pillows and nasal mask interfaces, both with and without a chin strap, (2) positional therapy devices including a cervical pillow to maintain neck extension, and (3) conversion to an autotitrating bilevel mode (minimum EPAP 8 cm H_2O, maximum IPAP 20 cm H_2O, maximum pressure support of 8). Despite these changes and ongoing excellent adherence, self-reported and objective OSA outcome measures regressed with worsened symptoms and further increase in AHI to over 30 events/h.

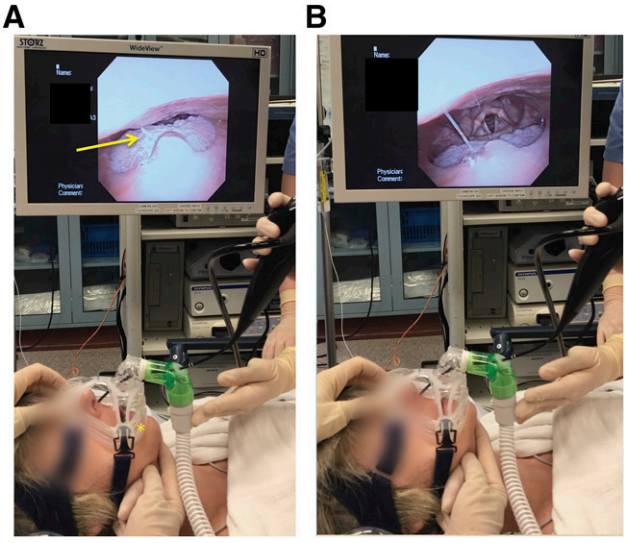
She then presented to our institution's sleep laboratory for an overnight split study, both to evaluate for other non-OSA causes of her persistent symptoms and to retitrate her PAP therapy. The diagnostic portion confirmed moderate obstructive sleep apnea only (AHI 28.4; no central apnea or nonrespiratory sleep pathology). The titration portion was challenging with persistent obstructive respiratory events and sleep fragmentation despite increases in pressure. At the final prescribed pressure (IPAP 22 cm H_2O, EPAP 18 cm H_2O), the residual AHI was 11.1 events/h and respiratory disturbance index was 25.3 events/h.

Given the multiple unsuccessful efforts in the clinic and sleep laboratory, she was offered DISE to investigate the anatomic basis of her residual AHI elevation. DISE was performed with a gradual propofol infusion, using clinical signs of sleep-disordered breathing, oximetry, and bispectral analysis monitoring, according to previously published guidelines.[2] The endoscopic examination was performed both without (baseline) and with (treatment) PAP therapy. At baseline, the patient demonstrated multilevel collapse with a primary anterior-posterior tongue base collapse pattern and a secondary effect of the tongue base on the distal soft palate (velum) and epiglottis. Significant jaw laxity and mouth breathing were observed with inferior and posterior displacement of mandible (2 cm interincisor vertical opening and 1 cm posterior retrusion). No intranasal pathology or other obstructing airway lesions (eg lymphoid hyperplasia, laryngeal stenosis) were identified.

The patient's home BPAP machine (13 cm H_2O/8 cm H_2O) was brought into the field, applied to the patient, and turned on. Her preferred oronasal mask was fitted securely in place, and a bronchoscopy adapter was connected to the BPAP circuit. The flexible endoscope was then inserted through the bronchoscopy adapter into the mask and in through the nose to the pharynx and upper airway. Repeat DISE with the BPAP in place demonstrated persistent tongue base collapse (**Figure 1A**). The oronasal mask itself appeared to further displace the mandible posteriorly. The baseline jaw laxity, in combination with the posterior displacement of the mandible from the mask and headgear as well as posterior displacement of the tongue and soft palate due to positive pressure via the patient's open oral airway, resulted in a persistent fixed obstruction of the tongue base against the posterior pharyngeal wall.

With the BPAP still in place, the mandible was then brought up manually into normal occlusion with a simple chin lift. This immediately resulted in resolution of the residual tongue base collapse and obstructive periodic breathing (**Figure 1B**). The video findings were reviewed with the patient after the procedure to guide treatment decision-making. Options discussed included (1) exploring additional PAP mask and chin strap devices with more effective mandibular stabilization potential, (2) revisiting the oral appliance therapy (OAT) option—either (a) as sole therapy or (b) as combination therapy with PAP to help ensure jaw/mouth closure, or (3) hypoglossal neurostimulation therapy to address the tongue base collapse with an alternative mechanism. She was referred to sleep dentistry in an effort to attempt PAP-OAT combination given the favorable mechanism of action seen during DISE-PAP. Unfortunately, she was unable to tolerate the OAT due to discomfort, so the efficacy of such combination therapy could not be assessed. She then completed informed consent for hypoglossal nerve stimulation therapy and is pending authorization and implant surgery scheduling at the time of manuscript submission.

Figure 1—Performance of DISE-PAP.



(A) DISE-PAP performed through a bronchoscopy adapter with the patient's home BPAP therapy applied through her oronasal interface. Note the open mouth-breathing posture (*) and residual tongue base collapse (arrow) on video-endoscopy. **(B)** Same DISE-PAP procedure with the addition of a simple chin lift to close the mouth. Note the significant improvement in the retrolingual portion of the airway on video-endoscopy. BPAP = bilevel positive airway pressure, DISE-PAP = drug-induced sleep endoscopy performed with a positive airway pressure device in place.

DISCUSSION

We present a patient who, despite excellent motivation and adherence to PAP, failed therapy due to persistent anatomical obstruction. The threshold for further investigation into cases of suboptimal PAP efficacy should be low. We have previously demonstrated how DISE may be used to evaluate incomplete oral appliance therapy response.[5] We propose that DISE, both with and without PAP therapy, similarly allows for a dynamic assessment of the upper airway to troubleshoot residual AHI elevation and provide an opportunity to further augment the effectiveness of PAP therapy with personalized treatment recommendations.

DISE-PAP has been studied in the literature as an alternative titration strategy[4,5] and to describe anatomic changes at various pressures.[6] To our knowledge, this report represents the first case in the literature describing DISE-PAP as a tool to evaluate and manage a patient with a residually-elevated AHI. Refractory OSA despite good adherence with PAP therapy is neither uncommon nor well understood, and it becomes difficult to offer solutions when the reasons for failure are unclear.[3] In our patient, the DISE-PAP procedure revealed an open mouth-breathing posture- and associated tongue base collapse- that were being exacerbated by the PAP oronasal interface, rather than improved by it. Several studies have shown higher residual AHI and lower adherence with oronasal masks than with nasal masks[4,5]; this case illustrates potential anatomic causes for these findings.

In select refractory situations, we propose that DISE-PAP may provide valuable information to assess PAP failures and may direct adjunctive medical or surgical treatment options with a personalized path to PAP success. Adjunctive solutions that could be identified during DISE-PAP include changes in jaw or neck position or targeting an obstructive upper airway lesion such as nasal valve collapse, intraluminal soft tissue (eg, neoplasm or lymphoid hyperplasia), or a floppy epiglottis—factors that are not commonly investigated during traditional sleep apnea testing or by PAP data download software. For our patient, DISE-PAP suggested favorable prognosis with combination PAP-OAT to ensure mouth closure and jaw stabilization. Hypoglossal neurostimulation therapy also appeared to be of potential benefit to activate the genioglossus muscle and anteriorly displace the tongue base.

It should be noted that DISE does have limitations, including variability in institutional technique and provider interpretation, and the findings should not be used to direct treatment in isolation. Rather, DISE should be viewed as an adjunctive diagnostic tool to evaluate upper airway locations and patterns of collapse, to assess the anatomic effectiveness of medical device therapies, and to investigate for alternative or adjunctive treatment strategies in cases of inadequate therapeutic response. We propose that DISE-PAP be considered for any patient in whom refractory obstruction appears to be precluding effective or comfortable use of their PAP therapy, especially in cases in which other strategies such as pressure/mode adjustments and mask refits have failed. Using DISE to visualize the anatomy while simulating home conditions, such as with PAP, postural devices, or OAT[6] in place, provides a tool by which conservative therapies could be adjusted and optimized, in addition to providing information for surgical planning should conservative therapies fail. Larger studies are warranted to explore the impact of DISE-PAP, standardize techniques, and determine which populations of OSA patients it would best serve.

CONCLUSIONS

Drug-induced sleep endoscopy during PAP application (DISE-PAP) represents an opportunity, under dynamic conditions, to examine the anatomic effect of PAP therapy on the upper airway and to troubleshoot incomplete treatment response. For patients with refractory residual AHI elevation despite excellent PAP adherence, DISE-PAP may provide clinically meaningful information to improve PAP outcomes.

ABBREVIATIONS

AHI, apnea-hypopnea index
BPAP, bilevel positive airway pressure
CPAP, continuous positive airway pressure
DISE, drug-induced sleep endoscopy
DISE-PAP, drug-induced sleep endoscopy performed with a positive airway pressure device in place
EPAP, expiratory pressure on positive airway therapy
IPAP, inspiratory Pressure on positive airway therapy
PAP, positive airway pressure
OSA, obstructive sleep apnea

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DISCLOSURE STATEMENT

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