

CASE REPORTS

Tongue Stabilizing Device-Emergent Central Sleep Apnea: A Case Report

Waled M. Alshhrani, BDS, MS^{1,2}; Yuuya Kohzuka, DDS, PhD³; Kentaro Okuno, DDS, PhD⁴; John A. Fleetham, MD⁵; Fernanda R. Almeida, DDS, MSc, PhD¹

¹Department of Oral Health Sciences, Faculty of Dentistry, University of British Columbia, Vancouver, British Columbia, Canada; ²Department of Prosthetic Dental Sciences, College of Dentistry, King Saud University, Riyadh, Saudi Arabia; ³Department of Perioperative Medicine, Division of Anesthesiology, Showa University School of Dentistry, Tokyo, Japan; ⁴Department of Geriatric Dentistry, Osaka Dental University, Japan; ⁵Division of Respiratory Medicine, Department of Medicine, University of British Columbia, Vancouver, British Columbia, Canada;

Treatment-emergent central sleep apnea is a phenomenon that has been reported after many obstructive sleep apnea treatment modalities. We present a case of demonstrating treatment-emergent central sleep apnea while using the tongue stabilizing device therapy. This case adds to the evidence that showed the effect of the supine position on the severity of central sleep apnea and shows the advantage of polysomnography follow-up after oral appliance therapy for central apnea assessment.

Keywords: obstructive sleep apnea, oral appliance, sleep position, tongue stabilizing device, treatment-emergent central sleep apnea

Citation: Alshhrani WM, Kohzuka Y, Okuno K, Fleetham JA, Almeida FR. Tongue stabilizing device-emergent central sleep apnea: a case report. *J Clin Sleep Med.* 2019;15(4):659–662.

INTRODUCTION

Treatment-emergent central sleep apnea (TECSA) is the development of central events after treatment of obstructive events. It is called “complex sleep apnea” by some and is recognized in the International Classification of Sleep Disorders, Third Edition as TECSA.¹ It has been reported after obstructive sleep apnea (OSA) treatment modalities such as PAP therapy,² mandibular advancement device,^{3–5} and surgeries.⁶ However, this report adds to the evidence that non-CPAP treatment might be associated with increased central sleep apnea, and, to the best of our knowledge, the tongue stabilizing device (TSD) emergent central sleep apnea has not been reported.

REPORT OF CASE

A 70-year-old male was diagnosed with severe OSA with an apnea-hypopnea index (AHI) of 49.6 events/h on October 2013 based on an overnight polysomnography (PSG) study. His past medical history includes diabetes mellitus type 2, hypertension, coronary artery disease, depression, latent tuberculosis, non-Hodgkin's lymphoma, and mild hypercalcemia. His daily medications include Metformin, Cozaar, Crestor, Citalopram, and Aspirin. He complained of loud snoring, and his Epworth Sleepiness Scale was 9/24. A positive airway pressure (PAP) trial was provided on January 2014 with both a nasal mask and pillows mask. On February 2014, he was followed up for a post oximetry and PAP was set at 10 cm H₂O pressure without a CPAP titration study. Two months later, he stopped using PAP therapy and remained without treatment until March 2017.

In 2017, the patient was referred to the University of British Columbia (UBC) Sleep Apnea Dental Research

team. As part of a research trial, new baseline with a level 3 monitor was recorded at home and the patient was then fitted with a TSD to hold the tongue in a protruded position and decrease the collapsibility of the upper airway during sleep (Aveo-TSD, Innovative Health Technologies, New Zealand). After 2 months of treatment, he received a repeat level 3 at-home study for follow-up. Based on level 3 studies, the respiratory event index (REI) decreased from 49.6 events/h to 7.2 events/h with the TSD, as shown in **Table 1**. Thus, the patient was referred back to his sleep physician for further assessment.

In 2018, as part of a research trial, a split-night study was conducted at the Leon Judah Blackmore Centre for Sleep Disorders at the UBC Hospital for which the results are shown in **Table 1**. It showed, on the first half (with no treatment), moderate sleep apnea with an AHI of 25.5 events/h (including an obstructive AHI of 22.2 and a central AHI of 1.1). The second half started at 3:04 AM (with TSD) and showed a significant increase in the OSA severity to an AHI of 71.1 events/h (including an obstructive AHI of 23.0 events/h and a central AHI of 40.4 events/h).

Also, we found there was an increase in the total central events in supine position from 2 events without TSD to 73 events with using TSD; in non-supine position, there was an increase from 1 event without using TSD to 24 events with using TSD. The AHI in rapid eye movement (REM) sleep and AHI in total sleep time (TST) were found comparable between both halves of the split study.

The patient started positional therapy and was referred back to the Sleep Apnea Dental Clinic for the evaluation of mandibular advancement therapy. The patient's medications and/or medical conditions have not changed over the course of his treatments.

Table 1—Summary of sleep study reports.

	Study 1	Study 2	Study 3	Study 4	
Date	October 2013	March 2017	May 2017	January 2018	
Description	Overnight PSG Baseline	Level 3 Baseline	Level 3 With TSD	Split-night PSG Without TSD With TSD	
Total recording time, minutes	424.0	394.4	389.2	184.1	166.0
TST, minutes	347.0	NR	NR	162.5	148.5
TST in supine, minutes	216.4	169.6	57.6	65.7	120.4
REM sleep time, minutes (%TST)	46.5 (13.4)	NR	NR	4.5 (2.8)	11 (7.4)
Number of REM periods	3	NR	NR	1	1
Total number of PLM episodes	55	NR	NR	7	39
Apnea and Hypopnea Analysis					
Apnea-hypopnea index, events/h	14.7	NR	NR	25.5	71.1
Respiratory event index, events/h	NR	49.6	7.2	NR	NR
Obstructive apnea index, events/h (count)	0.9 (5)	23.4 (154)	1.9 (12)	8.1 (22)	11.7 (29)
Obstructive hypopnea index, events/h (count)	13.1 (76)	26.2 (172)	5.4 (35)	14.0 (38)	11.3 (28)
Central apnea index, events/h (count)	0.7 (4)	NR	NR	1.1 (3)	39.2 (97)
Central hypopnea index, events/h (count)	0.0 (0)	NR	NR	0.0 (0)	1.2 (3)
Mixed apnea index, events/h (count)	0.0 (0)	NR	NR	2.2 (6)	7.7 (19)
Arousal index, events/h	17.8	NR	NR	17.7	46.5
Events by Body Position and Sleep Stage					
Supine apnea-hypopnea index, events/h	18.58	43.2	10.4	22.2	55.8
Total events in supine	67	122	10	60	138
Total events in non-supine	18	204	36	9	38
Total central apnea and hypopnea events in supine	NR	NR	NR	2	73
Total central apnea and hypopnea events in non-supine	NR	NR	NR	1	24
Total events in REM sleep	20	NR	NR	5	6
Total events in NREM sleep	65	NR	NR	65	170
Oximetry Analysis and Snoring Volume					
ODI, events/h	14.7	49	7.1	NR	NR
Time > 90% SpO ₂ , minutes (%TST)	1.0 (0.3)	23.6 (6.0)	0.0	4.2	32.6
Mean SpO ₂ , %	94.0	93.4	94.9	93.5	91.4
Min SpO ₂ , %	81.0	81.0	91.0	84.0	80.0

NR = not recorded, ODI = oxygen desaturation index, PLM = periodic limb movement, REM = rapid eye movement, TSD = tongue stabilizing device, TST = total sleep time.

DISCUSSION

The TECSA is a demonstration of predominately OSA followed by significant resolution of the obstructive apnea and emergence or persistence of central sleep apnea (not caused by another identifiable comorbidity) during PSG.¹

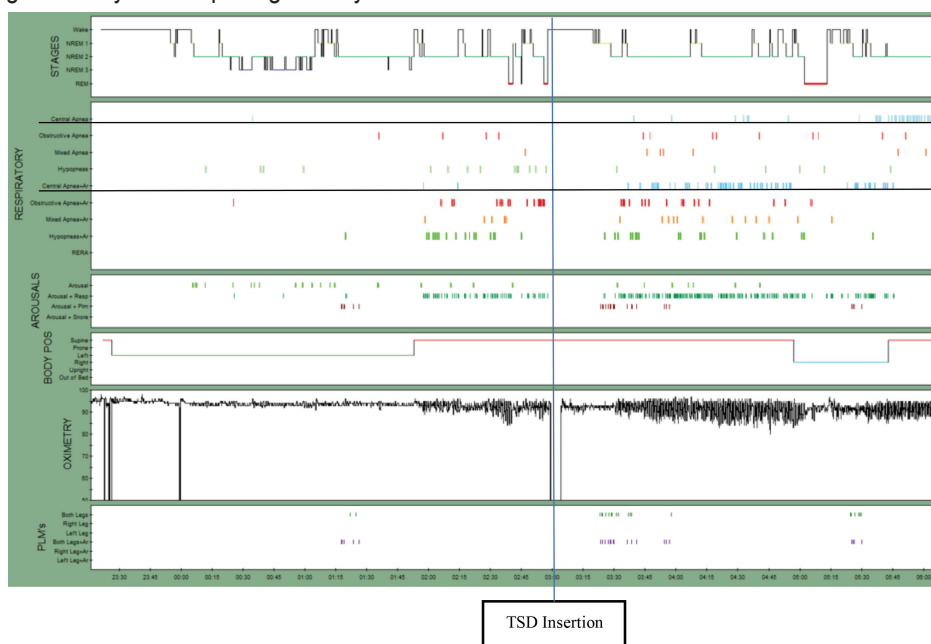
In this case report, we found the REI significantly decreased in the short term follow-up assessment with a level 3 sleep study (from 49.6 events/h without TSD to 7.2 events/h with TSD), but in the long-term split-night study, we found the obstructive apnea slightly increased with TSD (from 8.1 events/h to 11.7 events/h). In addition, there was a significant increase in the central apneas and hypopneas in supine and non-supine positions. It showed that the supine position while using TSD were associated with a significant increase in the central apnea index independent of sleep-stage effects or TST, as shown in **Figure 1** and **Figure 2**. Also, we found TST in supine position was longer (45.7 minutes) in the second half. This could explain the increase in the total central events in supine position

from 2 to 73 events consistent with other studies that reported the influence of the supine position on the central sleep apnea severity.⁷⁻⁹ However, it was found that the total central events in the non-supine position also increased from 1 to 24 events. In this patient, the development of central events was most likely a result of using TSD, since the AHI in REM sleep and TST were found comparable between both halves of the split study. Although some other non-diagnosed comorbidity could also have been present. Our case also emphasizes the great value of PSG follow-up after oral appliance therapy for central apnea assessment.

ABBREVIATIONS

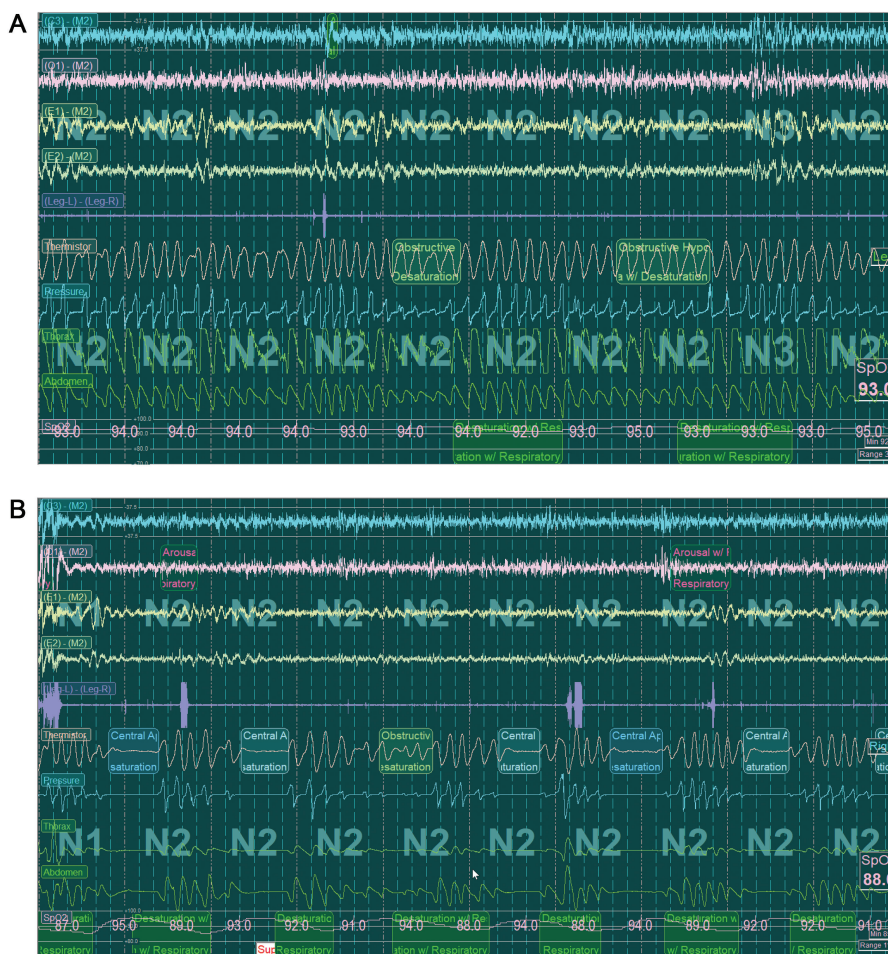
AHI, apnea-hypopnea index
OSA, obstructive sleep apnea
PAP, positive airway pressure
PSG, polysomnography

Figure 1—A scoring summary of the split-night study.



TSD inserted at 3:00 AM after which there was an increase in central apneas and central apneas with related arousals.

Figure 2—Two examples of 300-second epochs from the split-night polysomnogram.



The tests revealed changing (A) obstructive events during the first half of the night to (B) central events during the second half (B); both are in non-spine position.

REI, respiratory event index
 REM, rapid eye movement
 TECSA, treatment-emergent central sleep apnea
 TSD, tongue stabilizing device
 TST, total sleep time

REFERENCES

1. Sateia MJ. International classification of sleep disorders-third edition highlights and modifications. *Chest*. 2014;146(5):1387–1394.
2. Aurora RN, Chowdhuri S, Ramar K, et al. The treatment of central sleep apnea syndromes in adults: practice parameters with an evidence-based literature review and meta-analyses. *Sleep*. 2012;35(1):17–40.
3. Mohan A, Henderson J, Mador MJ. Mandibular advancement device-emergent central sleep apnea can resolve spontaneously: a case report. *J Clin Sleep Med*. 2016;12(1):137–138.
4. Kuźniar TJ, Kovačević-Ristanović R, Freedom T. Complex sleep apnea unmasked by the use of a mandibular advancement device. *Sleep Breath*. 2011;15:249–252.
5. Avidan AY. The development of central sleep apnea with an oral appliance. *Sleep Med*. 2006;7:187–188.
6. Corcoran S, Mysliwiec V, Niven AS, Fallah D. Development of central sleep apnea after maxillofacial surgery for obstructive sleep apnea. *J Clin Sleep Med*. 2009;(5):151–153.

7. Sahlin C, Svanborg E, Stenlund H, et al. Cheyne-Stokes respiration and supine dependency. *Eur Respir J*. 2005;25:829–833.
8. Oksenberg A, Khamaysi I, Silverberg DS, Tarasiuk A. Association of body position with severity of apneic events in patients with severe nonpositional obstructive sleep apnea. *Chest*. 2000;118:1018–1024.
9. Szollosi I, Roebuck T, Thompson B, Naughton MT. Lateral sleeping position reduces severity of central sleep apnea / Cheyne-Stokes respiration. *Sleep*. 2006;29(8):1045–1051.

SUBMISSION & CORRESPONDENCE INFORMATION

Submitted for publication July 16, 2018

Submitted in final revised form October 19, 2018

Accepted for publication October 30, 2018

Address correspondence to: Waled M. Alshhrani, BDS, MS, Department of Oral Health Sciences, Faculty of Dentistry, The University of British Columbia, 2199 Wesbrook Mall, Vancouver, B.C., Canada V6T 1Z3; Email: walshhrani@ksu.edu.sa

DISCLOSURE STATEMENT

All the authors have read the manuscript and have approved this submission. The authors report no conflicts of interest.