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Case Report

First Successful Mechanical Splint for Obstructive Sleep Apnea With an Orally Administrable Pharyngeal Stenting Device

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We report the case of obstructive sleep apnea in a 19-year-old, otherwise healthy male presenting with persistent daytime sleepiness and nonrestorative sleep after velo- and uvuloplasty. An individually tailored prototype of an orally inserted pharyngeal stenting device was proposed in the framework of a first clinical feasibility trial. The noninvasive, easily self-administered device is mounted on a simple inferior dental guard. Baseline total apnea-hypopnea index (AHI) was 15.5 and 24.4 per hour of rapid eye movement (REM) sleep. With the device, total AHI dropped to 6.7 per hour (56.8% reduction) and 1.4 per hour of REM (94.3% reduction). Recorded sleep efficiency during treatment was excellent at 96.5%.

Key Words: pharyngeal stent, mechanical splint, Obstructive Sleep Apnea, non-surgical treatment.

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INTRODUCTION

We report the case of obstructive sleep apnea (OSA) in a 19-year-old, otherwise healthy and drug-free male patient with a body mass index of 23 kg/m^2, presenting with persistent daytime sleepiness and nonrestorative sleep sensations after velo- and uvuloplasty. The initial clinical evaluation also reported moderate macroglossia and loud snoring at anamnesis. A former sleep recording, performed in a private hospital, revealed an apnea-hypopnea index (AHI) of 14.4/hr, an oxygen desaturation index of 16.3/hr of sleep, and 37.4% of sleep time spent snoring. Given the patient's prior explicit refusal of continuous positive airway pressure (CPAP) treatment, mandibular advancement device, or additional surgical intervention, an individually tailored prototype of a mouth-administered, pharyngeal stenting device (patent pending) was proposed in the framework of a first clinical practicability trial (PhaStentOSA).

The device comprises an orally inserted functional pharyngeal stent mounted on a simple inferior dental guard (Fig. 1). This noninvasive device is easily self-administered and removable. By design, it does not exert any traction on the mandible, tongue, or temporomandibular joints and aims at providing a mechanical upper airway (UA) splint by limiting the retrolingual collapse between the tongue and the posterior wall of the pharynx (Fig. 1).

CASE REPORT

After informed consent and ethical committee approval, the patient was admitted for two consecutive polysomnographic (PSG) recordings (baseline and active treatment respectively), performed with identical procedure and settings, at the academic sleep lab of the Brugmann University Hospital.

On admission, the patient showed normal vital sign parameters. Thorough physical, neurological, and mental examination did also not show any abnormalities. Oral inspection exposed a class 2 Mallampati score and well-executed partial velo- and uvuloplasty. Further clinical evaluation was completed through structured self-report instruments assessing perceived sleep quality (Pittsburgh Sleep Quality Index [PSQI]), daytime sleepiness (Epworth Sleepiness Scale [ESS]), and fatigue (Fatigue Severity Scale). These symptom-scales mainly confirmed reported excessive daytime sleepiness (15/24 on the ESS), but only light nonrestorative sleep sensations with a PSQI level of 7 and moderate daytime fatigue (4.1 on the FSS). At

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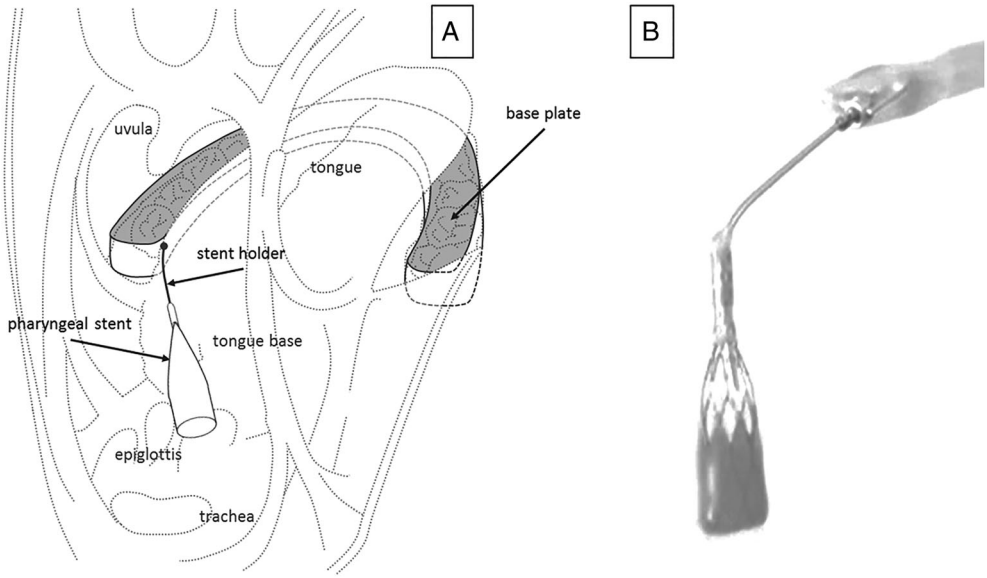

Fig. 1. Schematic presentation of the removable pharyngeal stenting device prototype.

baseline, the patient presented with a total AHI of 15.5 and 24.4 per hour of rapid eye movement (REM) sleep, but no nocturnal hypoxemia (Table I). During the second PSG, with the treatment device in place, residual AHI dropped to 6.7/hr (56.8% reduction) and to 1.4 per hour of REM sleep (94.3% reduction). The device was well tolerated and kept in situ during the PSG without interruptions or complications. Sensation at morning arousal was reportedly refreshing by the patient. Recorded sleep efficiency under active treatment, with respect to sleep maintenance, was excellent at 96.5%. Sleep architecture, with respect to sleep-stage proportions and sleep fragmentation appeared to be numerically similar between both nights (Table I). Visual videographic inspection indicated a reduction of supine position with the device in place, but no conclusions about a positional effect could be made yet, as valid respiratory parameters for each position under active treatment were unavailable due to calibration errors.

DISCUSSION

These results illustrate for the first time the practicality and therapeutic efficacy with a unique, orally inserted, noninvasive prototype of a self-administered and removable pharyngeal stenting device providing an effective mechanical retrolingual UA splint.

Although the standard treatment for OSA remains positive airway pressure in general, and CPAP in particular,[1] mid- or long-term treatment adherence may present with various drawbacks despite its efficiency. Least-sufficient compliance rates (usage of > 4 hours per 24 hours) vary widely (from 83% to only 46%) across patient samples in follow-up studies.[2,3] Mandibular advancement devices (MADs) are often considered as an alternative for the management of OSA. A recent meta-analysis indicated that MADs can be comparable to CPAP in mild disease and can be an appropriate treatment for CPAP-intolerant OSA patients.[4] UA surgery may also be performed in OSA patients,[5] and up to 28 different surgical treatment options have been identified.[6] However, different outcomes and a large variability of overall success rates do not yet add up to clear-cut guidelines or evidence-based algorithmic alternatives in comparison to CPAP.[1] Moreover, surgery is mostly irreversible and may even imply further or partial worsening of related nocturnal respiratory events in some cases (i.e., snoring, mucosal atrophy). Aiming at providing a mechanical UA splint for OSA with removable stenting

TABLE I.
Sleep Variables.

	Baseline	Stenting Device
Sleep efficiency (%)	94.8	96.5
N1 (%)	10.8	10.7
N2 (%)	43.8	49.3
N3 (%)	26.2	20.8
REM (%)	19.2	19.1
ArI (events/hr)	11.0	12.9
AHI (events/hr)	15.5	6.7
AHI_{REM} (events/hr)	24.4	1.4
CAI (events/hr)	1.0	0.7
ODI (events/hr)	3.3	2.0
Mean SaO_2 (%)	95	96
Minimum SaO_2 (%)	91	93
Snoring (%)	5.2	6.7

Sleep efficiency (index) = (total sleep time/sleep period time)*100 in percent (%). Non-REM sleep stages 1 (N1), 2 (N2), and slow wave sleep (N3) in percent of total sleep time.

AHI = apnea-hypopnea index; AHI_{REM} = AHI per hour of REM sleep; ArI = microarousal index; CAI = central apnea index; ODI = oxygen desaturation (drop of 3%) index; REM = rapid eye movement sleep; SaO_2 = oxygen saturation.

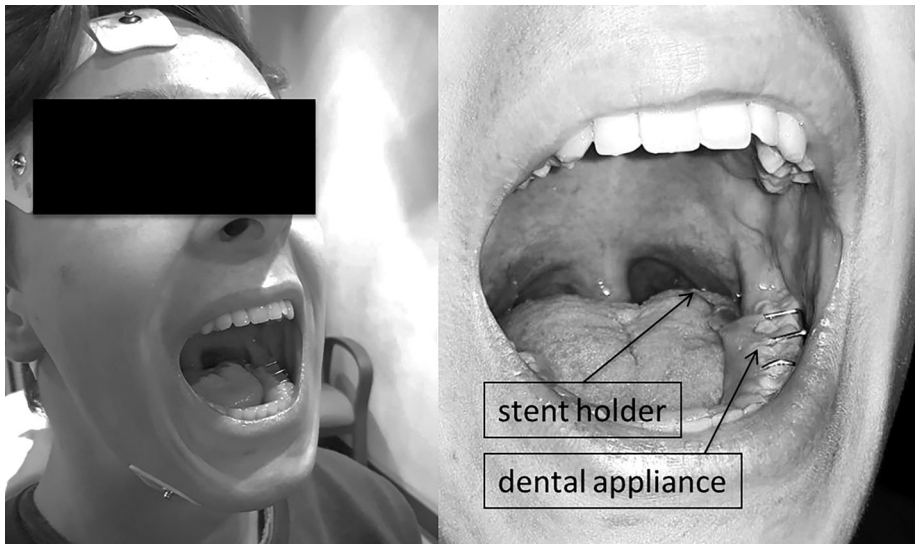

Fig. 2. Photographic illustration of the patient wearing the device.

devices has been tried previously by means of nasal stents/trumpets, with variable effectiveness and patient tolerance.[7] Accurate positioning is often difficult, introduction and removal are not convenient, and nasal trauma can regularly be observed. These nasopharyngeal devices must be introduced through the patient's nostril and carefully guided through the nasal cavity, passing the nasal turbinate and the nasopharynx, before crossing the velum. However, for potential effectiveness on UA obstruction, the distal tip should also theoretically reach an area within 1 cm of the epiglottis.[8] Conversely, only few nasally administered devices actually target the oropharynx.[9,10] A recent case series of eight patients treated with such a device reported a mean AHI reduction of 38.7% (AHI = 31.1 ± 12.0 vs. 19 ± 12.0) with the stent, in comparison to a mean reduction of 73.6% under CPAP (AHI = 8.2 ± 11.9).[10] Sufficiently long nasal trumpets are usually not used in general practice[7] or research due to limited tolerability.[9]

The studied device here may be suitable for patients who show adequate tolerance to oral appliance and gag reflex. It combines a mechanical splint directly at the oropharynx, maximizing potential therapeutic efficacy on OSA, as well as the comfort and ease of the application of the MAD, without its constraints of temporomandibular traction (much like a simple dental guard). This original approach of pharyngeal stenting allowed the patient to speak and breathe normally while awake. The device is unnoticeable from the outside when worn (Fig. 2), and presents with no restriction for mouth opening or swallowing. As per accepted protocol, the reported results are limited to the therapeutic outcome during a single PSG. Ambulatory follow-up data are not available. Despite the observed excellent sleep efficiency (with the device in place) and the reported high tolerance (confirmed by videographic recording) during this first night, claims about treatment-effect maintenance cannot be made yet. Furthermore, long-term compliance and potential side effects remain to be investigated. The device's main limitation may, however, reside in the overcoming of possible gag reflex during the initial application of the device. In the present case, three 20-minute ambulatory habituation sessions (aiming at behaviorally taming gag reflexes) prior to admission in the sleep lab appeared to be sufficient. Patient motivation and preference for a discreet device of rather small size is of particular importance to overcome this limitation, given the well-known psychological impact on the gag reflex. An additional limitation of this specific mechanical approach may be related to the anatomical and local restraint of the device. This stent mainly limits UA collapse with respect to lingual and retrolingual involvement. Hence, despite showing a clinically significant treatment effect here, future recommendations may require investigations to gain individual insights about different UA obstruction sites for particular OSA patients.

CONCLUSION

These initial results on sleep-related respiratory events along with apparently high patient tolerance during an initial night will lead to further research and development of such an oral appliance in subsequent clinical trials. This first report showed the achievability of mechanical splinting with an orally administered, pharyngeal stenting device, which may represent a novel option for the treatment of OSA.

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