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Herbst Oral Appliance for Obstructive Sleep Apnea When Uvulopalatopharyngoplasty and Nasal CPAP Failed †

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구개수구개인두성형술 및 지속적 기도 양압 공급치료에 실패하였으나 Herbst 구강내 장치로 효과를 보인 폐쇄성 수면 무호흡 증후군 1예

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폐쇄성 수면 무호흡 증후군(obstructive sleep apnea syndrome)의 치료를 위하여 구개수구개인두성형술(uvulopalatopharyngoplasty)을 시행하였으나 효과가 없었고, 수술후 시행한 비강을 통한 지속적 기도 양압 공급치료(continuous positive airway pressure : CPAP)에서는 5cmH_2O의 비교적 낮은 압력에서 폐쇄성 수면 무호흡이 효과적으로 소실되었으나 공기의 압력이 입으로 분산(mouth air leak)되어 숙면을 취할 수 없다는 이유로 CPAP의 적용을 거부하는 환자에서 저자들이 제작한 Herbst 구강내 장치(oral appliance)를 장착함으로써 매우 좋은 치료 효과를 보였다. 환자에게 구강내 장치를 가정에서 규칙적으로 장착하도록 지시한 후 5개월간에 걸쳐 수면다원검사(polysomnography)와 수면설문지검사(sleep questionnaires)를 반복 실시함으로써 치료 효과와 부작용 발생 여부를 추적 관찰하였다. 폐쇄성 수면 무호흡과 여러 가지 임상 증상이 지속적으로 현저히 호전되었고, 구강내 장치의 장착으로 잠에서 깨어난 후 일시적인 측두하악관절의 불쾌감을 호소하였으나 약 1개월 이후에는 소실되었으며, 현재까지 파손된 구강내 장치의 수리와 재조정을 제외하고는 다른 특기할 문제점 없이 규칙적인 사용을 계속하고 있다. 따라서 구강내 장치는 적응증이 되는 환자에서 적합한 형태를 선택하여 적절히 사용할 경우에는 폐쇄성 수면 무호흡 증후군의 효과적인 치료법으로 이용될 수 있으며, 특히 구개수구개인두성형술의 시행에도 불구하고 치료 효과가 없는 환자에서 지속적 기도 양압 공급치료에 적응하지 못하는 경우에는 구강내 장치의 적용을 고려할 수 있을 것으로 생각한다.

Key words : Obstructive sleep apnea syndrome, Oral appliance, uvulopalatopharyngoplasty, Continuous positive airway pressure

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Obstructive sleep apnea syndrome (OSAS) is a common disorder[1]. Most studies suggest that OSAS is associated with a higher than expected morbidity and mortality as a consequence of systemic and pulmonary hypertension, cardiac arrhythmias, myocardial infarction, and stroke[2,3]. However, there are many controversies concerning the treatment of OSAS, and there is no universally accepted standard for effective treatment. Treatment options are varied and range from conservative measures such as weight loss, nasal continuous positive airway pressure (CPAP), and oral appliance(OA) to more invasive procedures such as uvulopalatopharyngoplasty (UPPP)[4]. Nasal CPAP is the most popular and reliable form of treatment. However, a significant number of patients in whom nasal CPAP is initiated are unable to tolerate the device used to provide it, and must discontinue treatment[5]. The therapeutic effects of limited surgical procedures such as UPPP to eliminate specific abnormalities in the upper airway are often unsatisfactory. Moreover, recent reports suggest that the compliance of nasal CPAP may be compromised by previous UPPP[6,7]. When UPPP and nasal CPAP all have failed, oral appliance(OA) therapy may be an option as an alternative therapeutic modality[8~11]. In this report, we present a 5-month follow-up of the efficacy of Herbst OA in a OSAS patient who has failed with UPPP as the primary treatment and was unable to tolerate the subsequent nasal CPAP because of mouth air leak.

CASE REPORT

A 30-year-old man was referred to the sleep disorders clinic of St. Paul's hospital at the Catholic University of Korea. Heavy snoring, repeated apneic episodes observed by bed partners, choking during sleep, and excessive daytime sleepiness were causes of referral. There was no history of smoking, alcohol drinking, cardiovascular diseases.

The body mass index (BMI, kg/m^2) was 26.5, and blood pressure was 140/90mmHg. The examination of nose, oral cavity, pharynx and hypopharyx by the specialist of otolaryngology showed a large tongue, enlarged tonsils, a floppy uvula, and a redundant soft palate. Chest radiographs, pulmonary function tests and an ECG were normal. Laboratory values were within normal limits.

This patient underwent sleep questionnaires and full overnight polysomnography at baseline. The polysomnographic recording included modified electroencephalographic monitoring (C3/A2 & O2/A1), a bilateral electrooculogram, an ECG, and a submental and anterior tibialis electromyogram. Airflow was monitored by oral and nasal thermistors. Respiratory effort was monitored with chest-wall and abdominal piezoelectric bands. Continuous arterial oxygen saturation (SaO_2) was monitored by pulse oximetry. Snoring was monitored by snore microphone and digital sound level meter. The sleep recording was scored using the standard sleep staging manual of Rechtschaffen and Kales. An obstructive sleep apnea was defined as a cessation of airflow at least 10 sec, but with continued respiratory effort. A central sleep apnea was defined as a cessation of airflow at least 10 sec, during which no respiratory effort is evident. A mixed sleep apnea was defined as a combination of the two, during which a central respiratory pause precedes obstructed respiratory efforts. A hypopnea was defined as a greater than 50% decrease in the thermister tracing compared with baseline at least 10 sec in combination with an oxygen desaturation. Sleep efficiency was the total sleep time (TST) divided by the time from lights off to final awakening in

Table 1. Polysomnographic data before and after UPPP, nasal CPAP and Herbst oral appliance insertion*

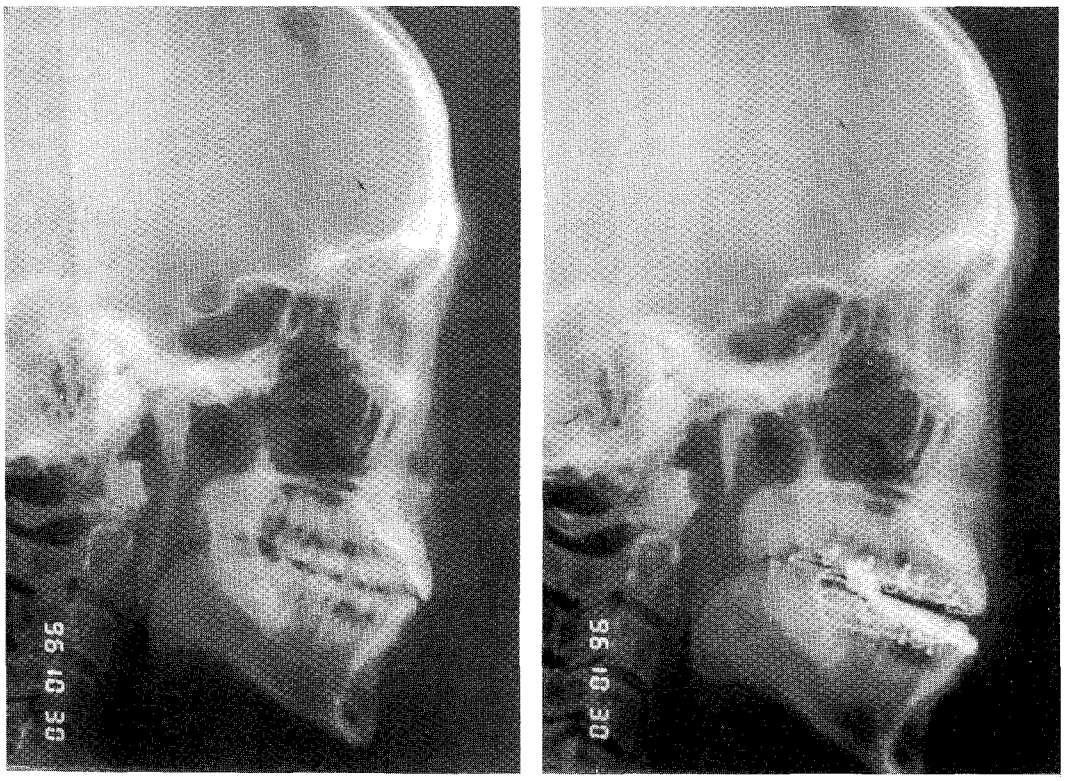
	Baseline	After UPPP	After nasal CPAP	Herbst oral appliance				
				Before	After 4-Day	After 1-Month	After 5-Month	
							Before	After
BMI(kg/m^2)	26.4	26.4	26.4	26.4	26.4	26.4	27.1	27.1
Snoring(dB)	90.1	77.5	65.0	78.2	75.1	70.2	7.89	69.2
AI, n/hour	31.5	38.9	0.1	47.7	1.2	0.4	63.2	0.3
AHI, n/hour	42.3	56.1	1.6	59.2	7.3	5.5	72.1	8.1
Prominent apnea type	OSA	OSA	–	OSA	–	–	OSA	–
Mean SaO_2, %	95	92	98	90	96.6	97.5	95.5	97.2
SaO_2 nadir, %	82	83	93	77	87	93	83	88
Total sleep time(TST), min	377.0	370.0	410.5	380.5	360.5	372.5	387.5	393.5
Stage 1&2 NREM(%TST)	73.1	77.9	50.0	60.1	51.0	68.4	74.0	58.2
Stage 3&4 NREM(%TST)	14.2	7.6	23.2	14.0	19.2	18.0	5.9	18.2
Stage REM(%TST)	14.3	9.9	23.8	18.1	26.2	12.8	15.3	21.6
Total apnea time(%TST)	21.5	22.2	0.1	28.9	0.5	0.2	42.2	0.1
Sleep efficiency, %	91.7	93.0	94.4	92.0	93.5	94.5	91.9	95.7

*dB=decibel(maximum sound level of snoring) ; OSA=obstructive sleep apnea

the morning. Apnea index (AI) and apnea-hypopnea index (AHI) were the number of events per hour of sleep.

This patient was diagnosed as OSAS with AI 32.6, AHI 46.6 (Table 1). Lateral cephalometric radiograph was taken, and the interpretation was followed using the rule of Riley and colleagues (Fig. 1) (Fig. 2)(Table. 2)[12]. The Müller's maneuver during the fiberoptic nasopharyngolaryngoscopy evaluation showed nomal position of soft palate and obstruction at the oropharyngeal level. UPPP was proposed by the otolaryngologist for the treatment of OSAS, and UPPP was done. Follow-up sleep questionnaires and full overnight polysomnography were performed at 3 months after UPPP. However, subjective symptoms and polysomnographic data were not improved at all (Table 1). So we attempted the application of nasal CPAP. On the first night trial for determination of optimal CPAP pressure, the polysomnographic data, including snoring, apnea, hypopnea, SaO_2 and sleep stage distribution, were markedly improved by pressure of 5 cm H_2O (Table 1). He purchased a device, and tried home nasal CPAP for about one month. However, he complained of mouth air leak and sleep disruption with nasal CPAP, and refused to use the device. So we had to try another therapeutic modality inevitably.

Herbst oral appliance, removable mandibular advancement device which holds the mandible forward 50% of the patient's maximal protrusive position, was prepared from the orthodontic laboratory (Fig. 3)[13]. After 3-night trial for adjustments, cephalometric radiograph after the appliance insertion was evaluated. The distance of posterior airway space



A B

Fig. 1. Cephalometric radiographs before (A) and after (B) Herbst oral appliance insertion.

increased from 1.5mm to 3.5mm after the insertion, and the distance from mandibular plane to hyoid bone decreased from 22mm to 17mm (Fig. 1, 2 and Table 2). In the polysomnographic studies before and after the appliance insertion, after 3-night trial for adjustments, AI decreased from 48.1 to 1.7, and AHI also decreased from 69.1 to 12.2 (Table 1). Follow-up overnight sleep study after 1-month regular use showed similar results, and repeated sleep studies, before and after the appliance insertion, after 5-month regular home use showed successful therapeutic effects (Table 1). Subjective symptoms also markedly improved with Herbst OA. The Herbst OA was well tolerated by the patient. There was no serious complications except mild temporomandibular joint discomfort after awakening in the morning during initial 1-month trial. He has been wearing the appliance during sleep on a regular and consistent basis without complaints.

DISCUSSION

The contributing causes that lead to OSAS are multifactorial, and the management of OSAS is complex. Medical and surgical approaches have been developed for airway maintenance during sleep, and the majority of patients can be effectively treated by at least one of the available modalities. However, the

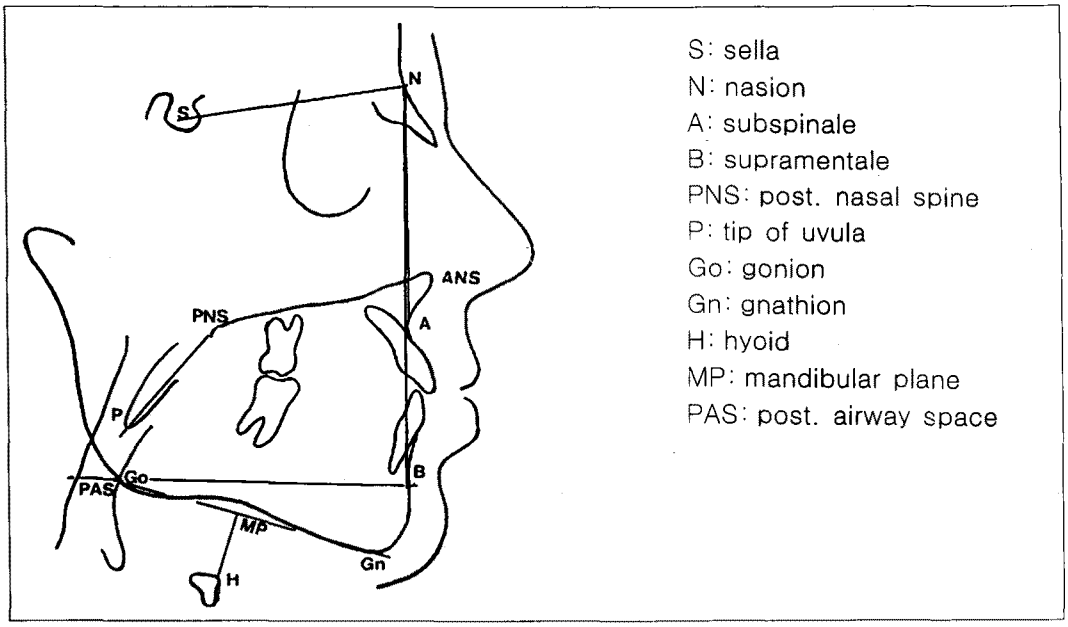
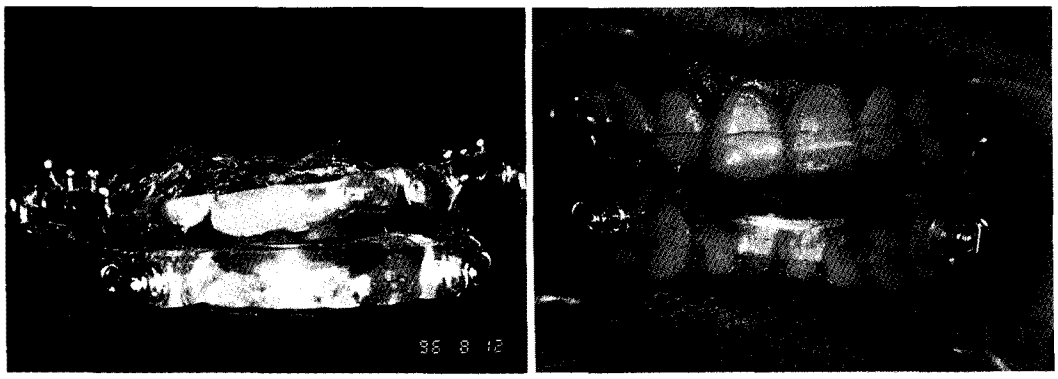


Fig. 2. The cephalometric landmarks.



A B

Fig. 3. The Herbst oral appliance before (A) and after (B) insertion.

choice of rational therapeutic modality for individual patients is essential for clinicians.

In this report, we discuss the difficulties in treatment of OSAS. Surgical treatment such as UPPP, when it is effective, can cure the problem, so the patient does not require long-term therapy. However, the failure rate of surgical treatment is greater than that with nasal CPAP. So sufficient rationale and specific indications should be established before surgical procedures. In our case, there was no therapeutic effects of UPPP. We thought retrospectively the potential regions of obstruction in this case were not

Table 2. Cephalometric measurements before and after Herbst oral appliance insertion*

	Before	After
SNA(angle, °)	81	81
SNB(angle, °)	77	76
ANB(angle, °)	4	5
PNS-P(length, mm)	29	29
MP-H(length, mm)	22	17
PAS(length, mm)	1.5	3.5

SNA=angle measurement from sella to nasion to subspinale ; SNB=angle measurement from sella to nasion to supramentale ; ANB=difference from SNA and SNB ; PNS-P=distance from posterior nasal spine to tip of the soft palate ; MP-H=distance from hyoid to mandibular plane ; PAS=posterior airway space.
All measurements were performed using the rule of Riley and colleagues[11].

only pharyngeal level but also hypopharyngeal level.

Moreover, recent reports suggest that many patients with previous UPPP were noncompliant on nasal CPAP[6,7]. The primary treatment of this patient was UPPP. The patient was still effective with nasal CPAP and required much less pressure (5cm H_2O) to resolve the sleep-disordered breathing. However, the patient was noncompliant because of mouth air leak with nasal CPAP. We believed the mouth air leak was due to the loss of palatal sealing by previous UPPP, and we thought it could be important clinical problem if UPPP, as the primary treatment, did not resolve the sleep apnea.

Oral appliances of various designs have been proposed and studied in OSAS with the recent increasing interest in sleep apnea. The American Sleep Disorders Association has issued practice guidelines which state that OA therapy is indicated for simple snoring, for mild OSAS, and for moderate to severe OSAS if nasal CPAP is not accepted or if surgery is not appropriate[11]. However, the use of OA has been limited because of no studies compared OA to other therapeutic modality such as nasal CPAP or UPPP. Ferguson et al[14] have recently reported a crossover treatment trial comparing nasal CPAP to a mandibular advancing OA. According to their report, OA reduced AHI to satisfactory levels in 48% of patients.

In this case, Herbst OA, removable mandibular advancement device, was used as an alternative therapeutic modality for the OSAS patient who has failed with UPPP as the primary treatment and subsequent nasal CPAP. Herbst OA was very effective for the treatment of OSAS. Repeated overnight sleep studies, after 1-month and 5-month regular home use, showed similar successful therapeutic effects. The patient initially complained of mild temporomandibular joint discomfort after awakening in the morning, but such complaint disappeared after 1-month regular use. These results suggested that OA could be a successful alternative therapeutic modality for this patient.

Several reports suggest that various upper airway dimensions on cephalogram after OA insertion are increased by the advancement and downward rotation of madible[8,13,15], and hyoid bone position is also important in OSAS[13]. In this case, on cephalometric radiograph after Herbst OA insertion, the distance of posterior airway space was increased, and the distance from mandibular plane to hyoid bone was markedly decreased. These changes on cephalogram were considered as the action mechanism of Herbst OA in this patient.

In this case, we experienced the therapeutic dilemmas of OSAS. The choice of rational therapeutic modality for individual patients is often difficult prob-

lem. We thought good communication with other specialists (including pulmonologist, otolaryngologist, dentist, and other sleep specialists) should be established for effective treatment of OSAS.

SUMMARY

This report describes a 5-month follow-up of the efficacy of Herbst oral appliance(OA) in a obstructive sleep apnea syndrome patient who has failed with uvulopalatopharyngoplasty(UPPP) and was unable to tolerate to subsequent nasal continuous positive airway pressure(CPAP) because of mouth air leak. The obstructive sleep apnea and daytime performance were markedly improved by regular home use of OA, and the patient still continues to use OA without complications. It is suggested that OA can be a successful alternative therapeutic modality in patients who are unable to tolerate to nasal CPAP, especially after UPPP.

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