Case Report

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Severe Obstructive Sleep Apnea Alleviated by Oral Appliance in a Three-Year-Old Boy

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Key Words
Oral appliance · Obstructive sleep apnea · Fraenkel appliance

Abstract
This clinical report describes a 3.5-year-old boy suffering from chronic daytime fatigue, accumulated snoring and dramatically appearing apnea during sleep. Oxycardiorespirography revealed a breathing pattern similar to repetitive obstructive apnea and an oxygen saturation periodically dropping to 80%. During tidal breathing, fiberoptic bronchoscopy showed aspiration of the aryepiglottic folds and the epiglottis during inspiration. Adenotonsillar hypertrophy was excluded. Due to the acknowledged side effects from various surgical approaches and nasal continuous positive airway pressure, a removable, functional Fraenkel II oral appliance was applied during sleep. Clinical assessment demonstrated resolution of the main respiratory symptoms, and oxycardiorespirography revealed a fundamental reduction in periodic obstructive apnea and desaturation. In conclusion, we consider the use of an oral functional appliance for severe obstructive sleep apnea in children to be a valuable alternative to other treatment methods.

Introduction

Obstructive sleep apnea (OSA) is a sleep disorder in which repeated reductions or cessations in airflow occur. The disorder may vary in its severity and is often associated with other physiologic symptoms [1, 2]. Today, the cut-off point selected for normal breathing in children is not accepted. Reports usually define OSA in children as an apnea-hypopnea index (events/hour) anything above 1/h or more and a respiratory disturbance index of at least 10/h on polysomnograph readings [3]. OSA in children is usually caused by anatomic anomalies occurring with constriction of the upper airway and/or hypotonia of the muscles in the pharyngeal ring and tongue [4]. The International Classification of Sleep Disorders [5] defines OSA as repetitive episodes of upper airway obstruction that occur during sleep, usually associated with a reduction in blood oxygen saturation. Girls and boys show a prevalence of 2% for OSA; for snoring, it is between 3 and 12% [6]. The soft tissue of the aryepiglottic folds and the epiglottis in young children are more susceptible to collapsing into the airway and less resistant to submucosal edema in comparison with adults. These characteristics are believed to predispose the child towards OSA.

Adenotonsillectomy is often considered the treatment of choice for pediatric OSA, mainly in children with persistence of a narrow upper airway [1]. Especially if the

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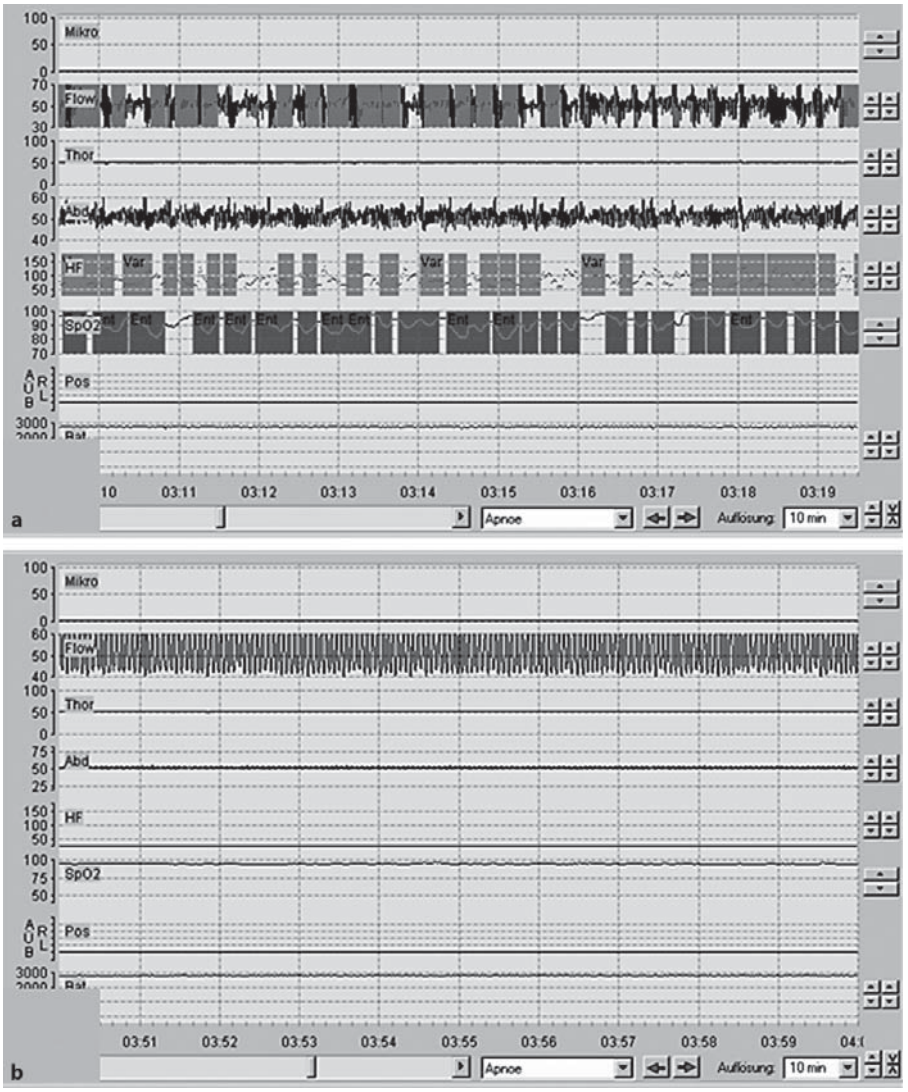

Fig. 1. Oxycardiorespirography (Poly-MESAM) before (**a**) and after (**b**) oral jaw positioning.

adenoids and tonsils are not enlarged, nasal continuous positive airway pressure is an effective and safe treatment for OSA, but it is limited by parents' and patients' acceptance [7]. Therefore, orthodontic treatment is a valid but often overlooked alternative [1, 8].

Case Report

We report on a 3.5-year-old Caucasian boy who presented with snoring and frequent apneic episodes during the night. He was admitted to hospital after such apneic episodes had seriously worsened during the previous 4 months, appearing dramatic and life-threatening to the parents, with the child's frequent futile breathing efforts and night sweats causing great distress. The boy was chronically tired, restarted napping and slept late in the morning. His ability to concentrate deteriorated. He had had an uncomplicated otitis media 5 weeks earlier and had undergone an adenotomy 2 years ago. The parents reported that he had snored and had occasional nocturnal breathing problems during infancy and early childhood, but his symptoms had never been thus serious before.

Initial laboratory tests were inconspicuous, including blood gases. Except for having small submandibular lymph nodes and only slightly hyperplastic tonsils, the boy was found healthy on physical examination. However, we observed periodic, accumulated snoring with an open mouth and apnea during sleep. Oxycardiorespirography (Poly-MESAM) with videotaping revealed a flow pattern similar to repetitive obstructive apnea and an oxygen saturation periodically dropping to 80% for more than 70% of the entire sleep recording (fig. 1a). Ruling out heart failure, a cardiac ultrasound was performed without any abnormal finding. However, fiberoptic pharyngolaryngoscopy showed evidence of complete aspiration of aryepiglottic folds and epiglottis during inspiration.

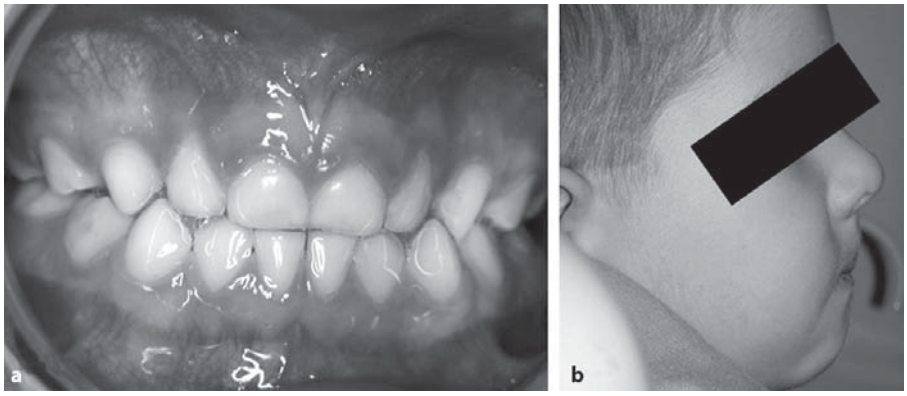

Fig. 2. a Full deciduous dentition with a skeletal and dental class I relationship and no malocclusion in the vertical and transversal planes. **b** No mandibular malposition.

As a consequence of limitations in the parents' acceptance of nasal continuous positive airway pressure and surgical treatment, an orthodontic evaluation was initiated. The child possessed full deciduous dentition with a skeletal and dental class I relationship and no malocclusion in the vertical and transversal planes, and no skeletal abnormalities; nose, soft and hard palate were normal and the airway space sufficient (fig. 2). The patient was fitted with a Fraenkel II regulator worn during sleep to prevent the mandible from dropping open and backwards; the appliance is shown in figure 3. The characteristics of the Fraenkel regulator are its buccal shields and lip pads that stabilize the device in the vestibulum oris. The appliance is anchored to the maxillary dental arch with the palatinal bow and canine claps in a positive manner. The palatinal bow stimulates the tongue toward proper appliance position. The acrylic lingual pad and the lingual wire maintain the mandible in its anterior-posterior relationship. The bite opening of the appliance is constructed to hold the mandible in minimal protrusion with a vertical opening of about 5 mm.

The patient tolerated the oral appliance from the very beginning. To improve treatment compliance, he was encouraged to wear the device for 1 week during the day. After an adaptation period of 1 week, he wore the appliance continuously every night.

While the device was being worn during the night, snoring, apneic episodes and daytime tiredness disappeared and the boy stopped napping in the morning. Oxycardiorespirography (Poly-MESAM) 3 months later showed regular breathing for 80% of the night. The child presented three episodes with sleep-disordered breathing with a lowest oxygen saturation of 88% (fig. 1b). The parents reported that sweating during the night diminished completely and the child was more active and explorative. Despite the fact that some few residual drops in oxygen saturation were noted during sleep, the child presented important daytime clinical improvement.

The compliance, tolerance and side effects of the appliance were assessed monthly by the orthodontist. Due to skeletal growth, the appliance was remade and readjusted after 9 months. And after 14 months of usage, there was no temporomandibular joint discomfort, nor any dental or skeletal changes. The results in the control sleep study remained stable (table 1).

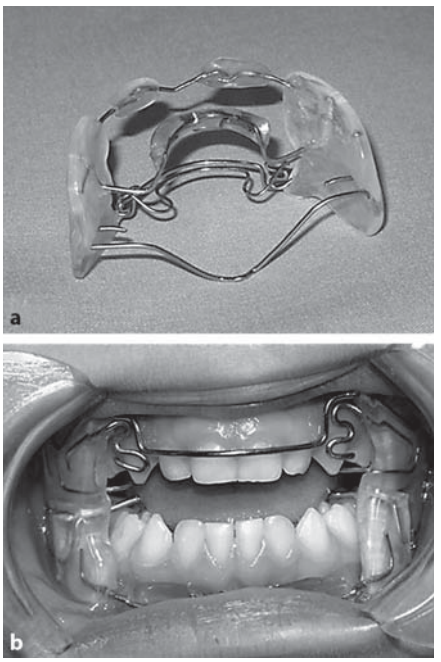

Fig. 3. a Fraenkel II type regulator inducing the therapeutic effect by advancing the mandible and the tongue to enlarge and stabilize the upper pharyngeal airway. **b** Fraenkel II type regulator positioned in the patient.

Discussion

In this case, severe OSA was related to aspiration of the aryepiglottis during inspiration. Application of an oral functional jaw-positioning appliance markedly improved the patient's condition.

Table 1. Sleep recording parameter: diagnostic recording without the appliance, with the appliance in situ after 3 months and with the appliance in situ after 14 months of treatment

	Diagnostic recording	With the appliance after 3 months	With the appliance after 14 months
Recording time, h	8	9	8
RDI, h	66	2	2
Desaturation index, h	61	3	4
Mean Sa_{O_2}, %	89	95	98
Minimum Sa_{O_2}, %	80	88	89
Sa_{O_2} artifacts of recording time, %	1.28	0.34	0.68

RDI = Respiratory disturbance index; Sa_{O_2} = mean oxygen saturation.

As the obstruction involves anatomical changes, the orthodontic approach is an appropriate alternative treatment [9, 10]. Callus distraction in this condition is mostly performed in children with syndromes like Goldenhar syndrome, if there is an additional skeletal deformation.

Oral protrusive appliances are an established therapeutic option for treating obstructive sleep-disordered breathing in adults under defined conditions [8, 11]. Most protrusive appliances prevent the pharynx from collapsing through anterior and vertical displacement of the mandible, resulting in an open and stabilized pharyngeal airway. One study has indicated that oral protrusive appliance had a beneficial effect in about 50% of the studied children presenting with skeletal class II, deep bite or crossbite malocclusion and OSA [12]. Our patient had no clinical dental or mandibular malposition, but while sleeping, the mandible dropped downwards and backwards. A lateral cephalogram has no clinical relevance at the age of 3.5 years and was not taken. The therapeutic aim of oral appliance treatment was to stabilize the mandible during sleep and to improve the tongue position to enlarge the retropharnyngeal space. Furthermore, an increase in vertical dimensions and a change of the occlusal plane inclination by this therapy have been described [13].

A requirement of an effective mandibular protrusion device in treating obstructive sleep-disordered breathing is the firm anchorage on both dental arches to hold the mandible in a more anterior position. Furthermore, the appliance should allow for adjustment of the protrusion and lower jaw movement [14]. In children with deciduous or mixed dentition, firm appliance anchorage cannot be achieved. A protrusion of the mandible in the growing child can result in unintended, amplified stimulation of mandibular condylar growth [15]; thus, conventional oral protrusive devices cannot be applied in children to treat sleep apnea.

As opposed to monobloc appliances, e.g., an activator or bionator, the Fraenkel regulator has been well-accepted by young children under clinical observation. The acrylic shields are positioned in the vestibulum oris and do not influence the oral cavity nor restrict the space or the tongue. Common side effects are temporomandibular joint discomfort, dental or skeletal deformations, which can be reduced by being worn only during sleep. This treatment should not be considered in children with neurological disorders.

We have followed our patient for 14 months to date, and we have not observed abnormal dental or skeletal changes. However, we cannot predict how long the patient may be able to use the device or if improvement in the sleep-disordered breathing may be seen with developmental growth. Especially the issue of adenotonsillectomy should be reconsidered later to possibly further improve the result and further normalize the sleep study. We must emphasize that patient cooperation is mandatory for application of this oral appliance. Monitoring is also vital to assure continued effectiveness and to prevent unfavorable side effects, such as dehiscence of the lower incisors, dental malocclusion and mandibular growth [16].

In conclusion, the applied Fraenkel II type regulator proved effective in the treatment of obstructive sleep-disordered breathing in this specific clinical situation in a child with deciduous dentition and no mandibular malposition, but the parents' and child's compliance are very important. Yet, it is unknown how long the appliance should be worn and whether his improved respiratory situation will continue once the appliance is removed.

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