

## Severe Obstructive Sleep Apnea Syndrome in a Toddler

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We report the case of a 3-year-old boy who had experienced intense snoring, frequent awakenings, intense respiratory effort during sleep, and delayed growth starting at the age of 15 months. He underwent adenoidectomy at 18 months. Symptoms initially improved but reappeared 3 months after surgery. He underwent a second adenoidectomy, this time with tonsillectomy, but there was no significant clinical improvement. Polysomnography revealed severe sleep apnea-hypopnea with an apnea-hypopnea index of 45. Continuous positive airway pressure improved sleep quality, although some symptoms, mainly snoring, persisted. A third adenoidectomy was necessary to normalize his breathing pattern during sleep.

**Key words:** *Obstructive sleep apnea syndrome. Infancy. Adenoidectomy.*

### Introduction

Sleep apnea-hypopnea syndrome (SAHS) manifests itself differently in children and adults. In children, the symptoms include snoring, restless sleep, breathing difficulty, hyperactivity, and behavioral changes.<sup>1</sup> Polysomnographic diagnostic criteria are also different in adults and children, since the latter usually suffer from obstructive hypoventilation, rather than from complete apneas and hypopneas.<sup>2</sup> The most frequent cause of SAHS in children is anatomical obstruction of the upper airway—mainly due to enlarged adenoids and tonsils—and adenotonsillectomy is the treatment of choice.

We present the case of a 3-year-old boy diagnosed with clinically severe SAHS and an unusual polysomnogram

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### Síndrome de apnea obstructiva del sueño infantil de presentación grave

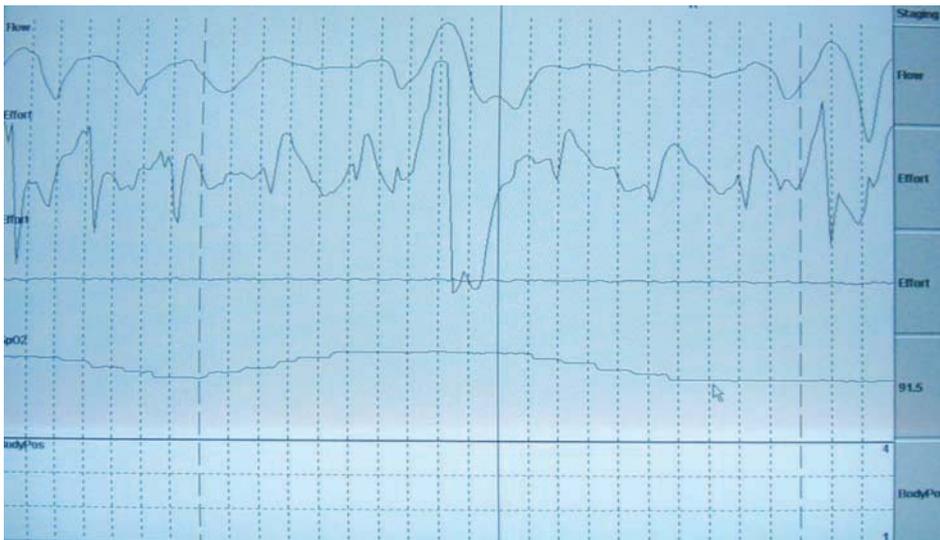
Describimos el caso de un paciente varón de 3 años de edad, que desde los 15 meses presentaba un cuadro clínico consistente en ronquidos intensos, despertares frecuentes, esfuerzo respiratorio intenso durante el sueño y retraso del desarrollo pondoestatural. Se le practicó una adenoidectomía a los 18 meses de edad, con mejoría inicial de los síntomas, que sin embargo reaparecieron a los 3 meses de la cirugía. Se le realizó una segunda adenoidectomía, acompañada esta vez de amigdalectomía, sin mejoría significativa de la clínica. Se le efectuó una polisomnografía, que resultó diagnóstica de síndrome de apneas-hipopneas durante el sueño de carácter grave, con un índice de apneas-hipopneas de 45. Tras comenzar tratamiento con presión positiva continua de la vía aérea mejoró la calidad del sueño, pero persistieron algunos de los síntomas, fundamentalmente el ronquido. Para normalizar su patrón respiratorio durante el sueño requirió, finalmente, una tercera adenoidectomía.

**Palabras clave:** *Síndrome de apnea obstructiva del sueño. Infancia. Adenoidectomía.*

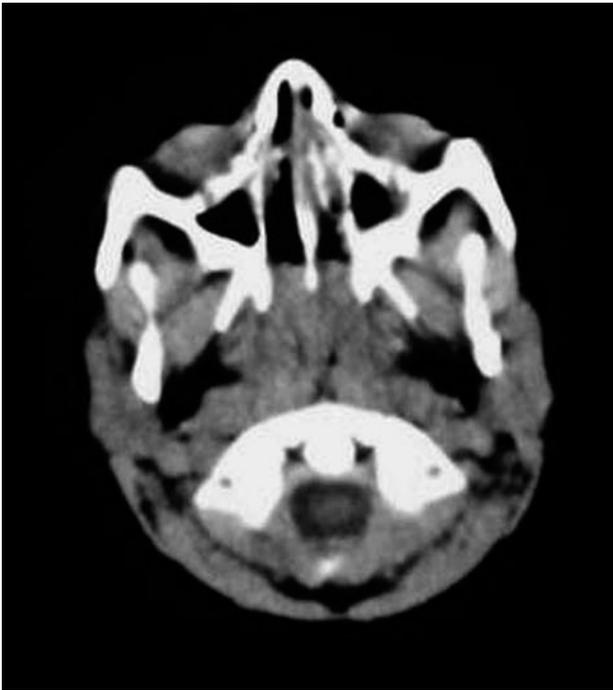
giving evidence of a pattern of complete apneas and very frequent hypopneas. The obstruction resolved with 3 operations carried out over a 2-year period, during which time the patient also required therapy with continuous positive airway pressure (CPAP).

### Case Description

The patient was a 3-year-old boy with no family history of interest, except that his father was obese and snored. The only remarkable feature of his personal medical history was an admission to hospital when he was 18 months old for pneumonia and bronchial asthma. Skin tests at that time were negative for the usual allergens. When he was 15 months old, his parents became aware of a clinical picture of intense snoring, very restless sleep with frequent awakenings, and use of the accessory respiratory muscles, with considerable retraction and paradoxical breathing. He underwent tests at the Ear, Nose, and Throat (ENT) Department, where he was diagnosed with adenoid hypertrophy. The first adenoidectomy at 18 months was followed by a clear improvement in symptoms. Three months after surgery, his



**Figure 1.** Nighttime polysomnogram (neurophysiological variables not recorded). The nasal flow channel shows hypopneas and apneas, with visible oximetric repercussion in the oximetry channel.



**Figure 2.** Computed tomography of the cavum, which reveals hypertrophy of the adenoid tissue of the cavum in relation to the child's age. Parapharyngeal fat is normal.

symptoms reappeared and intensified progressively until they became as evident as before surgery. A new ENT examination revealed recurrence of the adenoid lymph tissue hypertrophy, as well as enlarged tonsils. Therefore, he underwent tonsillectomy and a second adenoidectomy. However, his clinical situation did not improve after surgery, and he continued to suffer from intense snoring, frequent apneas observed by the parents, marked intercostal retraction, and paradoxical breathing during sleep. The patient was referred to the Sleep Unit of the Respiratory Diseases Medical–Surgical Department. Physical examination revealed delayed growth (10th percentile), adenoid facies with a high arched palate and marked nasal obstruction accompanied by mouth breathing and abundant nasal and pharyngeal secretions.

The remainder of the physical examination by system was normal. A nocturnal polysomnogram was recorded in the sleep laboratory. The neurophysiological parameters were not recorded in order to improve tolerance to the test, which gave abnormal readings throughout the recording (Figure 1). The breathing pattern, which was monitored with a thermistor, was made up of complete apneas (reduced oronasal flow  $\geq 80\%$  compared with baseline for at least 5 seconds) and frequent hypopneas (reduced oronasal flow  $\geq 50\%$  associated with desaturation and lasting for at least 5 seconds). The total number of respiratory events was very high, with an apnea–hypopnea index (AHI) of 45. A polygraph test was carried out with CPAP, and it was observed that with 6 cm H<sub>2</sub>O and a face mask the breathing and oximetry pattern improved, although it did not return to normal values completely, with a residual AHI of 5.5. (A face mask was used because the patient could not tolerate the nasal mask.) The clinical situation improved considerably during the following months, although the parents reported that the child continued to snore with CPAP and he occasionally presented paradoxical breathing and retraction. A second nocturnal polysomnogram was recorded and he was observed to need higher pressure (8 cm H<sub>2</sub>O), which once again improved the breathing pattern. However, he continued to experience episodes of hypoventilation and apnea that did not improve by increasing the pressure or using an auto-CPAP device. After 6 months with CPAP, the child had improved: he had gained weight and the quality of his sleep was better, although the nasal symptoms of obstruction and abundant secretions persisted, as did the pattern of daytime mouth breathing. Therefore, computed tomography of the upper airway was performed, and this showed a large adenoid mass that blocked most of the cavum and reached the posterior edge of the choanae (Figure 2).

Shortly afterwards, the patient underwent a third adenoidectomy, which clearly improved his clinical situation. After this third intervention, CPAP was withdrawn and, some months later, he had gained weight, moving from the 10th to the 50th percentile. Despite the clinical improvement and owing to the severity of the previously diagnosed SAHS, a nocturnal polysomnogram was recorded 3 months after the adenoidectomy and the AHI remained high (12.3), although nasal obstruction was more intense than usual because the patient happened to have a bad cold affecting the upper airways. The boy currently has check-ups at the ENT Department of our pediatric hospital to monitor and treat his nasal symptoms, and in our Sleep Unit to monitor the long-term outcome of his disease.

## Discussion

This case is interesting for 3 reasons: the severity of its clinical and polysomnographic presentation, the fact that 3 adenoidectomies were necessary to resolve it, and the continued residual obstruction in the polysomnogram, despite the considerable clinical improvement after the last operation.

SAHS is highly prevalent in children and affects 1.5% to 3% of the infant population.<sup>3</sup> The main symptoms are nocturnal—snoring, restless sleep, and difficulty breathing, which parents usually describe as “labored.” This child’s condition had become particularly severe, with frequent apneas and extreme breathing difficulty before 2 years of age, when adenoid hypertrophy usually begins to appear. Most children with SAHS due to adenoid hypertrophy clearly improve after surgery,<sup>4</sup> and the response to the first adenoidectomy in this case was remission of symptoms, as expected. However, these reappeared early owing to the persistent anatomical obstruction of the airway caused by new adenoid tissue growth and the accompanying enlarged tonsils. Even though a relapse of hypertrophic adenoids after adenoidectomy is very unusual with the currently used surgical technique, the failure of the second operation makes this case rare, since the persistence of the upper airway obstruction after 2 technically correct adenoidectomies and a tonsillectomy is exceptional. Although the development of the maxillofacial bone structure is not complete until age 11,<sup>5</sup> this child was developing marked adenoid features at a young age, reflecting the severity and persistence of the upper airway obstruction.

The diagnostic test of choice in a child with SAHS is polysomnography. Despite the fact that some methodological aspects remain unresolved and the diagnostic criteria may be different,<sup>6</sup> it is generally accepted that SAHS in children is characterized by partial upper airway obstruction, which leads to a pattern of persistent hypoventilation rather than of the repeated complete apneas that characterize adult SAHS.<sup>2,7</sup> Furthermore, in children, respiratory events are shorter than in adults,<sup>8</sup> so that obstructive episodes of any duration that can lead to desaturation are considered

abnormal and an AHI  $\geq 3$  is considered abnormal.<sup>7</sup> Nevertheless, this boy’s polysomnogram showed uncommonly severe alterations, with a very high number of complete apneas and hypopneas, such as those that would define severe adult SAHS. The patient could not tolerate nasal ventilation and needed a face mask because there was still obstruction due to a large adenoid mass; therefore, the fact that the upper airway caliber could only be partially opened with positive pressure would explain why the CPAP was not completely effective according to the polysomnogram. In addition to the adenoid features, which can lead to a reduction in upper airway caliber as in this case, functional factors that contribute to obstruction could also cause the persistence of a high AHI after 3 operations. Such functional factors, if persistent, can lead to the development of SAHS years later, especially after the onset of puberty.<sup>9</sup>

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