A 4-yr-old boy presented with frequent snoring and mouth breathing, but no other symptoms often seen in children with obstructive sleep apnea (OSA) (e.g., observed episodes of apnea, daytime sleepiness, enuresis, and hyperactivity). His mother stated that the frequency and severity of his snoring varied from time to time. He did not frequently have colds, estimated at once or twice a year. He had normal physical and mental development (height 110 cm, body weight 19 kg, body mass index 15.7 kg/m²). He appeared to have a mild adenoid face (a slightly short upper lip with mandibular hypoplasia) and obvious tonsillar hypertrophy (grade 3+ tonsils with < 75% of space between pillars) and a stage II Friedman classification.

The patient underwent overnight polysomnography, and a radiograph of the lateral neck was obtained. Surprisingly, we found that the patient had unusually severe OSA based on the apnea-hypopnea index (AHI), which was as high as 102.5 events/h. During a recorded total sleep time of 7.75 h (supine position 64%, rapid eye movement (REM) sleep 25.1%), he had 713 obstructive apnea events (longest at 53 sec), 73 hypopnea events (longest at 55 sec) and 8 mixed apnea events (longest at 31 sec) but no central sleep apnea events. As demonstrated in Figure 1A, the patient experienced frequent and very severe reductions in blood oxygen saturation (SpO₂, lowest at 55%). Persistent, large-range variations in heart rate (70–140 beats/min) were associated with frequent OSA events and occurred throughout the night. The radiograph showed nasopharyngeal stenosis with tonsil and adenoid hypertrophy (adenoidal nasopharyngeal ratio (A/N): 0.92; normal range < 0.5–0.6).

The patient’s parents did not want to consider surgery and preferred medication as a first treatment. Therefore, montelukast (oral, 5 mg/day) was prescribed. After taking montelukast for 2 mo, the patient’s frequency and severity of snoring was reduced. Repeat polysomnogram showed a surprising reduction in the AHI to 2.4 events/h during a total sleep time of 8.19 h (supine position 34%, REM 19.4%). Overnight measurements of SpO₂ and heart rate stabilized to nearly normal levels (Figure 1B). The nasopharyngeal airway also appeared wider in the radiograph (A/N: reduced from 0.92 to 0.80).

**QUESTION:** When might montelukast be used to treat OSA in children?
Figure 1—Polysomnogram-detected changes in blood oxygen saturation, heart rate, body position, and sleep stage (left) and lateral neck radiograph (right).

(A) Before and (B) after taking montelukast for two months. AHI, apnea-hypopnea index; AI, arousal index; TST, total sleep time; Pos, body position; REM, rapid eye movement sleep; N1–3, Non-REM sleep 1–3. Arrows: A, adenoid; N, Nasopharyngeal.
ANSWER: Oral montelukast may be useful in the management of some severe OSA related to adenoid hypertrophy.

DISCUSSION

In this case, a 4-y-old boy showed unusually severe OSA with AHI 102.5 events/h and dramatic reductions in overnight SpO₂. For comparison, of 845 preschoolers with suspected OSA who had overnight PSG records in our center over the past 7 y, only 0.35% had an AHI greater than 100 events/h; 0.95%, 5.21%, 21.89%, and 71.60% had AHI in the ranges of 60–100, 30–60, 10–30, and less than 10 events/h, respectively. Based on the parents’ desire to avoid surgery in this case, montelukast was chosen as an alternative therapeutic intervention. As described previously, the patient experienced marked improvement in OSA as reflected by the outcomes of PSG and radiograph examination at the 2-mo follow-up visit.

Tonsil and adenoid hypertrophy is the most common underlying risk factor for the development of OSA, and the size of tonsils and adenoids are associated with the severity of OSA.1 Enlarged adenoids and tonsils primarily consist of hypertrophied lymphoid tissue, and evidence of inflammation has been documented in children with OSA. C-reactive protein, a systemic marker of inflammation, is increased in children with OSA.2 Moreover, the level of leukotrienes and their receptors are increased in the tonsils and adenoids of children with OSA.3 Montelukast, an oral leukotriene receptor antagonist, has been approved for preventive treatment of inflammation associated with asthma and allergic rhinitis in children older than 1 y. Oral montelukast has been used in children with mild to moderate OSA. Previous clinical studies,4–7 including double-blind and placebo-controlled trials, have shown that treatment with montelukast significantly reduced adenoid size and reduced respiratory-related sleep disturbances in children with mild to moderate OSA. An open study4 reported that obstructive apnea index was reduced from 3.0 to 2.0 and the A/N ratio was decreased from 0.76 to 0.56 after 16 w of montelukast therapy. Goldbart et al.6 showed significant improvement in the obstructive apnea index (3.9 ± 1.6 events/h before vs. 1.7 ± 1.0 events/h after treatment) and a decrease in A/N ratio (0.81 ± 0.004 before vs. 0.57 ± 0.004 after treatment) after 12 w of montelukast therapy. In another placebo-controlled trial,7 AHI was reduced from 9.2 to 4.2 after 16 w of montelukast therapy in children age 2 to 10 y. To our knowledge, the efficacy of montelukast in the treatment of severe OSA in children has not been documented. This is the first report we know of that shows dramatic improvement of severe OSA with the use of montelukast. Therefore, this case suggests that oral montelukast may be useful in the management of some severe OSA relating to adenoid hypertrophy, just as it has been documented to be useful for mild to moderate OSA in children, though the efficacy needs further evaluation by clinical trials.

During the initial examinations, the child did not have symptoms of upper respiratory tract infection, and his mother stated that he had experienced cold symptoms that lasted for a few days in the month prior to examination. Therefore, the severe OSA events were not likely due to upper respiratory tract inflammation. However, there have been reports of seasonal variability in pediatric OSA.8,9 This is more common in children younger than 5 y and occurs more often in borderline cases,9 which have been found to show more severe OSA during winter and spring.9 This child did not have allergic respiratory symptoms, but the initial PSG examination was indeed carried out during winter (January), and the second examination during spring (March). Thus, seasonal OSA might not be completely excluded in this case. As such, the use of oral montelukast in the management of seasonal OSA cannot be conclusively determined.

In addition, effects from time spent in the supine position may be present as well as the amount of REM sleep as indicated by differences on the assessment night after drug treatment (supine position 34%, REM 19.4%, concurrent REM and supine position 4.3%) compared to the diagnostic night (supine position 64%, REM 25.1%, concurrent REM and supine position 16.7%).

SLEEP MEDICINE PEARLS

Contrary to the recommended treatment guideline of using a continuous positive airway pressure machine to treat OSA in adults, adenotonsillectomy is recommended as the first treatment plan for children with OSA.10 The huge reduction in both AHI and A/N after treatment for 2 mo in this unusually severe case of childhood OSA suggests that oral montelukast may be useful in the management of some severe OSA related to adenoid hypertrophy. If successful, montelukast could preclude the need for surgical intervention in some cases.

CITATION


REFERENCES


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