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Improvement of monosymptomatic enuresis after adenotonsillectomy in children with obstructive sleep apnea syndrome

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Aim: To investigate the prevalence of monosymptomatic enuresis (ME) in children diagnosed with obstructive sleep apnea syndrome (OSAS) and the rate of resolution or improvement in enuresis following adenotonsillectomy.

Materials and methods: We retrospectively reviewed the charts of 541 consecutive patients who underwent adenotonsillectomy for OSAS secondary to adenotonsillar hypertrophy between January 2005 and January 2009. All of the charts for patients between 5 and 18 years of age at the time of surgery (n = 398) were included in the study. After reviewing the charts, the families were contacted by telephone. Those patients who had shown preoperative symptoms of enuresis were questioned to determine whether there had been any change in their complaints postoperatively.

Results: Of the 398 patients whose records were reviewed, 98 were excluded from this study because of incomplete records. The incidence of ME in the study group (n = 300) prior to adenotonsillectomy was 30.7% (92 patients). Among these 92 patients, 64 (69.6%) were male and 28 (30.4%) were female (P = 0.001). The parents of 46 of these 92 patients agreed to allow their children to participate in the study. In 46 patients, 26 (56.5%) had complete resolution, 8 (17.4%) had a partial improvement, and 12 (26.1%) had no change in enuresis following adenotonsillectomy. Patients with OSAS had a 2.38-fold higher risk of ME (odds ratio 2.38, 95% confidence interval 1.60 to 3.53, P = 0.001).

Conclusion: Children with OSA symptoms have a high rate of monosymptomatic enuresis. Relief of OSA symptoms also resulted in the complete resolution or partial improvement of ME in more than two-thirds of patients. In the differential diagnosis of a child presenting with enuresis, OSAS should be kept in mind and, conversely, the presence of enuresis should be investigated in children presenting with OSA symptoms.

Key words: Monosymptomatic enuresis, obstructive sleep apnea, adenotonsillectomy

Introduction

According to The International Children's Continence Society, enuresis is intermittent incontinence while sleeping and it is a term applicable to children who are at least 5 years old. Enuresis in children without any other lower urinary tract symptoms (nocturia excluded) and without a history of bladder dysfunction is defined as monosymptomatic enuresis (ME) (1). Although the true incidence

of ME is unknown because of the likelihood of underreporting, the reported rate is 15% in 5 year olds (2). The pathogenesis of ME has been debated but the current consensus is that this condition is multifactorial. Etiological factors include a number of possible mechanisms, such as genetic factors, sleep arousal dysfunction, nocturnal polyuria, psychological components, altered diurnal ADH secretion, and maturational delay (3).

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Obstructive sleep apnea syndrome (OSAS) is a breathing disorder that occurs during sleep. It is characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction, both of which disrupt normal sleep patterns and normal ventilation during sleep (4). OSAS causes impairment of quality of life and it is also associated with an increased risk of diabetes mellitus and cardiovascular diseases (5,6). Most upper airway obstruction in children is caused by adenotonsillar hypertrophy (7). There have been several studies concerning the relationship between upper airway obstruction and ME voiding dysfunction (8-18). It is well established that adenotonsillectomy has excellent results in resolving the apneic symptoms and there is some evidence that the surgical treatment of OSAS is beneficial for children with ME/(12,14-16). The aim of the present study was to evaluate the prevalence of ME in children diagnosed with OSAS and the effect of surgery on ME.

Materials and methods

The study design and number of patients are summarized in Figure 1.

We retrospectively reviewed the charts of 541 consecutive patients who underwent adenotonsillectomy for OSAS secondary to adenotonsillar hypertrophy between January 2005

and January 2009. A total of 398 patients between the ages of 5 and 18 years at the time of surgery were included in the study. Each of these patients' charts was reviewed for the age and sex of the patient in addition to the presenting symptoms and indication of surgery. After reviewing the charts, the families were contacted by telephone. Informed consent for the interviews was obtained from parents. The local ethics committee approved the study protocol.

The parents of each child were asked about the preoperative presence or absence of ME, any history of voiding dysfunction, and postoperative symptoms including the presence or absence of snoring, witnessed apnea, restless sleep, drooling, and mouth breathing. Only patients diagnosed with primary ME (as differentiated from those with secondary ME, who would have had a period of continence) were included in this study. Behavioral problems are uncommon in children with primary ME, especially those older than 10 years of age, whereas these problems are over 7 times more common in children with secondary ME (19). Patients between the ages of 5 and 18 years at the time of surgery were included in our study. In order to evaluate the relationship between OSAS and ME more precisely, we excluded children with secondary ME. Children with any other lower urinary tract (LUT) symptoms (increased/decreased voiding frequency, daytime incontinence, urgency, hesitancy, straining, a weak stream, intermittency, holding maneuvers, a feeling of incomplete emptying, post-micturition dribble, and genital or LUT pain) and those with a history of bladder dysfunction were not included in this study. The following questions were asked to the parents of patients who had preoperative symptoms of ME:

- 1. How frequently did your child wet the bed before surgery?
- 2. Did your child improve in his/her enuretic episodes? If the answer of this question was yes,
- 3. How frequently did your child wet the bed after surgery?

We evaluated the changes in symptoms within 3 months after surgery and categorized the patients into 3 groups:

- Group 1: Patients with complete resolution of ME.
- Group 2: Patients with partial improvement.

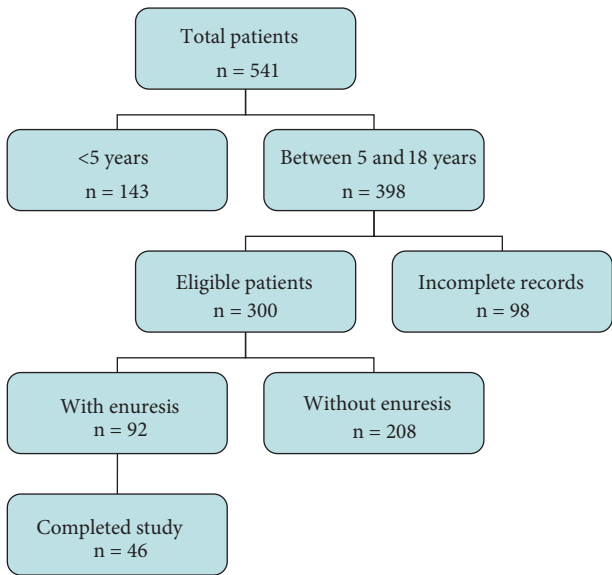


Figure 1. Study design and number of patients.

Group 3: Patients with no change in their complaints.

Partial improvement was defined as a minimum of 50% postoperative decrease in the frequency of bed wetting, as described previously (12). Children with other chronic medical conditions or those pharmacologically treated for ME were excluded. All data were collected between November 2008 and May 2009, such that postoperative data collection occurred anywhere between 4 months and 46 months postoperatively, with the average being 20 months postprocedure. A commercially available statistics software package (SPSS for Windows v. 12.0, Chicago, USA) was used to perform all statistical calculations. The chi-squared test was used to compare the prevalence of ME in our patients before and after surgery.

Results

Of the 398 patients, 98 were excluded from the study because of incomplete records. The remaining 300 (75.4%) patients had a mean (SD, range) age of 8.5 (3.05, 5-18) years. Of these patients, 166 (55.3%) were male and 134 (46.7%) were female. Twenty-six (8.7%) of the remaining 300 patients had a history of bladder dysfunction. The incidence of ME in the entire study group before adenotonsillectomy was 30.7% (92 patients). From this group, 64 (69.6%) of the patients were male and 28 (30.4%) were female. There was a statistically significant ($P = 0.001$) difference in the incidence of ME based on gender (male: 38.5%, female: 20.9%). The parents of 46 of these 92 patients agreed to allow their children to participate in the study. Past medical history revealed that none of these patients had any abnormal history or physical examination findings that may have been an underlying cause of ME, with the exception of upper airway obstruction. Of the 46 patients with preoperative ME, 26 (56.5%) had a complete resolution of enuresis episodes, 8 (17.4%) showed a partial improvement in the frequency of enuresis episodes, and 12 (26.1%) demonstrated no change in their complaints following adenotonsillectomy for OSAS (Table). The time of resolution/partial improvement of ME was 6.0 ± 2.28 (range 2-11) weeks. OSA symptoms were resolved in all of the patients postoperatively. The gender differences in patients in

Table. Enuresis after surgery.

	Patients n (%)
Preop enuresis	46 (100)
Postop status	
Resolved	26 (56.5)
Decreased	8 (17.4)
No change	12 (26.1)

each of the above mentioned groups are summarized in Figure 2. To determine whether the results related to ME were statistically significant, a chi-square test for equal proportions was performed. The chi-square value was found to be 18.961, resulting in $P < 0.001$. Patients with OSAS had a 2.38-fold higher risk of ME (odds ratio 2.38, 95% confidence interval 1.60 to 3.53, $P = 0.001$).

Discussion

The prevalence of ME (30.7%) in our study group was more than the expected rate of ME in this age range (20) and this rate is similar to those published previously (12,14). Monosymptomatic enuresis is a common symptom in children complaining of OSA, and our data shows that the presence of OSAS plays a significant role in ME. Moreover, our data provided a high resolution rate of ME after adenotonsillectomy. It is natural that the best endpoint of adenotonsillectomy should be the complete resolution of enuresis. We achieved complete resolution in 56.5% of our patients. Additionally, another group of patients

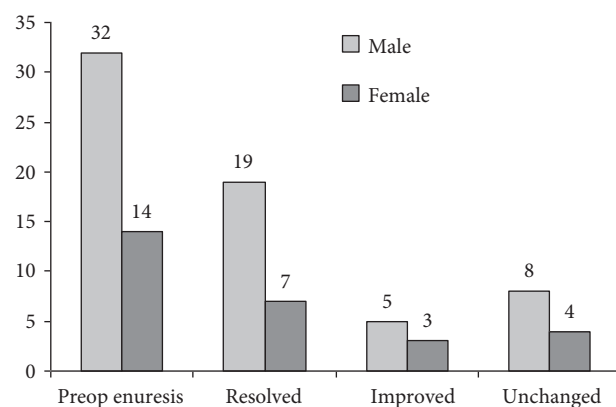


Figure 2. Gender differences in the improvement of enuresis postoperatively.

(17.4%) improved in the number of episodes of enuresis, as reflected by decrease. We think that this is also important. Altogether, 73.9% of our patients experienced either complete resolution (3/4) or partial improvement (1/4) in symptoms. Our data supports similar findings previously presented (12,14-16).

An important question is why OSAS may cause nocturnal incontinence. Although a clear relationship between OSAS and ME can be observed, the mechanism behind this relationship is unclear. Current hypotheses include increased bladder pressure due to increased respiratory efforts against an obstructed airway, insufficient arousal response, disruption in circadian ADH secretion, and increased levels of atrial natriuretic peptide resulting from upper airway obstruction (13,21-22). Another important question centers on whether acquired upper airway obstruction is responsible for inducing primary ME or simply aggravating it. Further prospective studies are needed to answer such questions.

Çinar et al. observed that the incidence of ME was 34.5% in children with OSAS and they also reported that 63% of their patients experienced the complete resolution of enuresis postoperatively (12). In another retrospective study, Basha et al. (14) demonstrated that 61.4% of their patients had total resolution while 22.8% had a significant decrease and 15.8% demonstrated no change in ME postoperatively. Weissbach et al. (16) reported that ME totally disappeared within 1 month in 41% of the 27 children examined while in a further 11% of children ME disappeared during the remaining follow-up time.

Yearly, the spontaneous resolution rate of ME in enuretic children aged 5-19 with a predominance of primary ME is around 15% (23,24). This rate is significantly lower than the results obtained after surgical treatment, as presented in our report. The spontaneous resolution of ME occurs at a relatively regular and steady rate throughout middle childhood.

The underlying cause is thought to be related to the delayed maturation of bladder mechanisms, portions of the central nervous system, or both (16).

Firoozi et al. reported that patients with OSAS had fewer voids per day and less daytime incontinence after adenotonsillectomy and they suggested that upper airway obstruction may also be involved in daytime urinary incontinence. Their explanation was that it might be spontaneous resolution with time or something as simple as the trauma of surgery/anesthesia, which may result in some change in the functional bladder capacity postoperatively, allowing for the improved storage of urine with a concomitant decrease in enuresis episodes (15).

We are aware that it would have been better to obtain data on every patient included in the study. Enuresis is an embarrassing situation for families in this country, however, and for this reason it is possible that some parents did not feel comfortable answering questions from the interviewer and did not participate in the study. In total, we were able to convince 46 patients to return for control evaluation. We are presenting only the data on this available group. Patients were assessed at different time periods after surgery. Recall bias might affect reliability and may consequently compromise the validity of data. These are the drawbacks of our study.

Conclusion

We have found a significant increase in ME prevalence in children with OSA symptoms and demonstrated that the relief of OSA symptoms also resulted in the complete resolution or partial improvement of ME in more than two-thirds of patients. In the differential diagnosis of children presenting with ME, OSA should be kept in mind and, conversely, the presence of ME should be investigated in children presenting with OSAS. Finally, further prospective studies are needed to explain the underlying mechanism of ME in children with OSAS.

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