Commentary

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Identifying Improvements in Daytime Sleepiness Following Adenotonsillectomy in Children with Non-Severe Obstructive Sleep Apnea and Narcolepsy

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Introduction

Daytime sleepiness is defined as the inability to stay awake during major wake periods of the day. Narcolepsy is a disorder characterized by excessive daytime sleepiness in which patients repeatedly lapse into sleep during typical wake time. Narcolepsy may occur with or without cataplexy, a sudden loss of muscle tone provoked by strong emotion. Narcoleptic patients may also experience sleep paralysis, hypnagogic hallucinations, and nocturnal sleep disruption. Naps in narcoleptic patients are typically short in duration with the patient initially feeling refreshed upon awakening. The typical age of onset of narcolepsy is 15–25 years old, but the diagnosis can be made in children as young as 5 years of age. While narcolepsy with cataplexy affects 0.02%–0.18% of adults, the prevalence of this disorder in children is unknown. The diagnosis of narcolepsy with cataplexy in children can be made clinically but is typically confirmed by full-night polysomnography followed by Multiple Sleep Latency Test (MSLT). Treatment involves behavioral modifications such as scheduled naps and medical therapy with wake promoting agents such as stimulants and sodium oxybate.

Prevalence of OSA in Pediatric Narcoleptic Patients

Increased frequencies of other sleep disorders, most commonly Obstructive Sleep Apnea (OSA) and Rapid Eye Movement (REM) sleep behavioral disorder; have been reported in patients with narcolepsy. In a recent study, co-morbid OSA occurred in over 25% of adult narcoleptic patients. Obesity, which is common among narcoleptic patients, may contribute to the high prevalence of OSA in this population. Research has demonstrated improvements in daytime sleepiness in narcoleptic adults receiving Continuous Positive Airway Pressure (CPAP) therapy for OSA. The prevalence of OSA in pediatric narcoleptic patients is unknown. There is limited data regarding outcomes in children with OSA and narcolepsy. Pediatric OSA affects 2 to 3% of healthy school aged children. OSA in children is characterized by intermittent episodes of upper airway collapse during sleep. The pathophysiology of pediatric OSA is multifactorial, but the two main risk factors include adenotonsillar hypertrophy and obesity. This disease entity has been linked to metabolic changes and cardiovascular sequelae. Furthermore, there is a growing body of literature demonstrating the negative impact of OSA on Quality of Life (QOL) and cognitive function in children. Pediatric OSA has been associated with behavior problems, daytime sleepiness, hyperactivity, and poor school performance. Polysomnography is currently the gold standard for the diagnosis of pediatric OSA. The severity of OSA is categorized according to the obstructive Apnea Hypopnea Index (AHI) on PSG. According to the most commonly utilized system, an AHI between 1 and 10 indicates non-severe OSA, while an AHI greater than 10 is diagnostic of severe OSA. Adenotonsillectomy is the primary treatment for pediatric OSA and results in improvements in PSG parameters, behavior, and QOL measures. The impact of OSA on daytime sleepiness in narcoleptic patients is still being investigated. No published research has assessed outcomes in children with OSA and narcolepsy. The primary objective of this study is to assess for improvement in daytime sleepiness following adenotonsillectomy in children with a history of narcolepsy and non-severe OSA.

Prevalence of Non-Severe OSA in Pediatric Patients with Narcolepsy

We performed a retrospective chart review at a tertiary care children's hospital over a 15 year period from 2000 to 2015. Approval from the Eastern Virginia Medical School and the Children's Hospital of The King's Daughters Institutional Review Board was obtained. Children who underwent both adenotonsillectomy and MSLT were identified by searching billing records using Current Procedural Terminology (CPT). A total of 68 patients were initially identified that underwent adenotonsillectomy and MSLT during the study period. After chart review, 38 children were excluded because they did not meet the criteria for narcolepsy. Fourteen children underwent adenotonsillectomy prior to diagnosis of narcolepsy and were thus excluded. An additional 3 children had significant comorbid medical conditions including DiGeorge syndrome, chromosomal

deletion, and severe traumatic brain injury and were not eligible. Excessive daytime sleepiness is one of the primary symptoms of patients presenting with narcolepsy. The diagnosis of narcolepsy may be confounded by the presence of co-morbid OSA. In adult narcoleptics, the co-existence of OSA and narcolepsy is high (25%), while the exact prevalence of OSA in narcoleptic children is unknown. Furthermore, no prior study has examined treatment outcomes in children with narcolepsy and OSA. Adenotonsillectomy may improve daytime sleepiness in children with narcolepsy and non-severe OSA. Future research is needed to determine the prevalence of non-severe OSA in pediatric patients with narcolepsy and to establish standardized treatment protocols for this population.