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Evaluation of behavioral change after adenotonsillectomy for obstructive sleep apnea in children with autism spectrum disorder

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ABSTRACT

Background and objective: Obstructive sleep apnea (OSA) may affect daily cognitive functioning in children. The aims of our study were two-fold. The first aim was to detect, using the Child Behavior Checklist (CBCL), whether adenotonsillectomy (AT) for the treatment of OSA improved the behavior of children with autism spectrum disorder (ASD). The second aim was to identify characteristics for behavioral improvement following the treatment of OSA in these children with ASD.

Methods: The behaviors of ASD children aged 5–14 years diagnosed as having OSA (n = 30) were evaluated using CBCL before and after AT. CBCL evaluation of ASD children without OSA at two time points with the same interval served as a control (n = 24). We statistically examined the two groups. In addition, we conducted a paired *t*-test to assess changes in CBCL Tscores between the improved group and unchanged/deteriorated group to identify characteristics that may affect behavioral changes following OSA treatment.

Results: After AT, T-scores of the CBCL scales were significantly improved in the OSA group, but no change was observed in the control. A paired *t*-test revealed that the improved group had significantly higher scores on the CBCL pre-AT than the unchanged/deteriorated group in ASD children with OSA after OSA treatment.

Conclusions: Behavioral problems were significantly improved following AT in ASD children with OSA. Early detection and treatment of children with OSA is essential to prevent behavioral problems and to support mental development.

What this paper adds?

Pediatric obstructive sleep apnea (OSA) has been associated with deficits in behavior and emotion regulation, selection, sustained attention and scholastic performance. When comparing children's neurobehavioral functioning before and after adenotonsillectomy (AT), many studies have reported improvement of cognitive function, behavior and learning. However, no studies have been

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conducted to evaluate the effect of AT on the behavior of children in Japan. And there are no studies investigating the effect of AT on behavioral problems in children with autism spectrum disorder (ASD), although it was recently suggested that the frequency of OSA is higher in ASD than community samples. This study sought to determine whether AT for the treatment of OSA improved the behavior of children with ASD, and to identify the characteristics that may affect behavioral improvement in children with ASD after treatment of OSA by AT. We analyzed Child Behavior Checklist (CBCL) scores before and after AT in ASD children with OSA. After AT, Tscores of the CBCL scales were significantly improved in ASD children with OSA. We revealed that the improved group shown significantly higher scores on the CBCL pre-AT than the unchanged/deteriorated group in ASD children with OSA after OSA treatment. We believe that our study makes a significant contribution to the literature because, to the best of our knowledge, it is the first to report the efficacy of the treatment of OSA as a means of improving the behavior of children with ASD.

1. Introduction

In the third edition of the International Classification of Sleep Disorders (ICSD-3), the prevalence of pediatric obstructive sleep apnea (OSA) is estimated to be 1–4% (American Academy of Sleep Medicine, 2014). In children, OSA has been associated with not only daytime sleepiness but also metabolic problems, e.g., obesity (Bhattacharjee et al., 2010; Mitchell & Kelly, 2007; O'Brien, Sitha, & Bauer, 2006; Tauman, Gulliver, & Krishna, 2006), cardiovascular accidents (Marcus, 2001), and growth problems (Marcus, 2001). In addition, OSA may affect daily cognitive functioning in children and lead to attention problems, hyperactivity, impulsivity, or aggression (American Academy of Sleep Medicine, 2014).

Sleep-disordered breathing (SDB) ranges in severity from primary snoring, which is not associated with any gas exchange abnormalities or sleep disturbance as detected on conventional polysomnography (PSG), to OSA, which is characterized by not only snoring, but also apnea, intermittent hypoxia, hypercarbia, and/or repeated arousals from sleep (Gozal, 2001). A review of 61 studies, and others that focused on the relationship between childhood SDB and neurobehavioral functioning, reported that SDB was associated with deficits in such variables as behaviors, emotional regulation, scholastic performance, sustained or selective attention, alertness, and so on (Beebe, 2006; Blunden et al., 2000; Giordani et al., 2008; O'Brien et al., 2004; Zhao et al., 2008). Symptoms were not, however, affected by the severity of SDB (Blunden et al., 2000; Giordani et al., 2008; O'Brien et al., 2004; Zhao et al., 2008).

The diagnostic criteria, causal factors, and first-line treatment of childhood OSA are different from those of adults (Chervin et al., 2005). For example, the diagnostic criterion for the apnea-hypopnea index (AHI), which represents the number of apnea and hypopnea events per hour, is five in adult OSA, and only one in childhood OSA (American Academy of Sleep Medicine, 2014). In addition, childhood OSA is mainly caused by hypertrophy of the adenoid and/or palatine tonsils, therefore, the current standard treatment for childhood OSA is adenotonsillectomy (AT) rather than continuous positive airway pressure (CPAP), which is the first-line treatment for adult OSA (Marcus et al., 2012).

Many studies, including the randomized Childhood Adenotonsillectomy Trial (CHAT) (Marcus et al., 2013), have reported improvements in cognitive function, behavior, and learning after surgical intervention with AT (Ericsson, Lundeborg, & Hultcrantz, 2009; Friedman et al., 2003; Goldstein et al., 2002; Huang et al., 2007; Li et al., 2006; Malow et al., 2006; Marcus et al., 2013).

We previously reported the case of an obese child with severe OSA whose hyperactivity was improved after introducing CPAP for the treatment of OSA (Miyoshi et al., 2006). However, no studies have been conducted to evaluate the effect of OSA treatment on the behavior of children in Japan.

Several studies have reported an improvement in children's behavior after treatment of these symptoms (Tordjman et al., 2013; Cohen et al., 2014). Huang et al. (2007) reported that AT is more effective for behavioral improvement in children with attention deficit hyperactivity disorder (ADHD) and mild OSA than methylphenidate. Concerning autism spectrum disorder (ASD), it highly comorbid with sleep problems such as sleep-wake rhythm disturbances, insomnia, and parasomnia (Richdale & Schreck, 2009; Malow & McGrew, 2008). Furthermore, the severity of sleep problems in ASD has been found to be significantly correlated with behavioral problems (Hirata et al., 2016). Recently, we found that the prevalence of OSA is higher in ASD than community samples (Hirata et al., 2016). Thus, we hypothesized that treatment of OSA may also improve behaviors in ASD children with OSA. AT is a difficult procedure for ASD children, because such novel events may provoke in them a great deal of anxiety; this being in addition to the intrinsic risks of AT procedures, such as bleeding, throat pain, and respiratory complication (Baugh et al., 2011). If we can predict the efficacy of AT, we can avoid unnecessary surgery or recommend an age for surgical intervention. Unfortunately, there is no literature which characteristic is sensitive for behavioral and cognitive impairments followed sleep improvement. However, obesity and OSA severity did not an affect to improvement in cognitive function and behavior after surgical intervention (Marcus et al., 2013; Huang et al., 2007). Thus, we examined characteristic which is the most recommendable case for AT, i.e. age, sex, obesity, OSA severity, or behavioral problems.

The aims of our short-term retrospective study were two-fold. The first aim was to determine whether AT for the treatment of OSA improves the behavior of Japanese children with ASD using the Child Behavior Checklist (CBCL). The second aim was to identify characteristics for behavioral improvement following the treatment of OSA in these children with ASD.

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