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LETTER TO THE EDITOR

Comment on Horne et al. sleep and sleep disordered breathing in children with Down syndrome: Effects on behaviour, neurocognition and the cardiovascular system Sleep Med Rev 44 (2018) 1–11

To the Editor,

I have read with interest the comprehensive and informative review article by Horne and colleagues [1]. The authors have clearly emphasised the lack of studies regarding the diagnostic threshold required for the treatment of sleep-disordered breathing (SDB) in improving neurocognitive and cardiovascular sequelae in the paediatric population with Down syndrome (DS) [1]. In support of this timely review, it may be helpful to make the following additional points.

There have been few studies that have demonstrated the negative impact of SDB on cognition in children with DS [2–4]. Breslin et al. reported that the presence obstructive sleep apnoea (OSA) confirmed by polysomnography (PSG) in children with DS was associated with a verbal IQ that was 9 points lower than that of children without OSA [2]. Churchill et al. showed that children with DS have difficulty accomplishing activities of daily life when compared with typically developed peers and that children with DS and SDB were at increased risk for further impairment [3]. Also, attenuated sympathetic responses to SDB are indicative of autonomic dysfunction and may lead to cardiovascular complications in the setting of untreated OSA [4]. However, the limited indirect and direct evidence supports active management of SDB in patients with DS. Contributing factors, such as obesity, generalized hypotonia, and craniofacial abnormalities account for increased instability of the upper airway during sleep and increase the susceptibility to SDB in DS, and addressing only one of those factors is less likely to result in clinical improvement [5,6]. Although adenotonsillectomy is the most common first-line treatment for OSA in paediatric patients with DS, about 50% are left with moderate to severe residual OSA following treatment [6,7].

Continuous positive airway pressure (CPAP) is a non-invasive and effective option for patients who do not respond to adenotonsillectomy as well as for patients who are poor surgical candidates [8,9]. Although data on compliance with CPAP in children with DS and OSA is limited, a randomised controlled study of 28 individuals with DS showed significant reduction in daytime sleepiness and depression, and improved cognitive function following CPAP treatment [8]. However, poor compliance with CPAP in the paediatric patient population remains an issue [8].

Therefore, there is a need for other treatment options for a subset of DS children with moderate to severe residual OSA. Because children with DS may have multiple sites of upper airway obstruction, additional surgical interventions that may be considered include lingual tonsillectomy, turbinate reduction, uvulopalatopharyngoplasty (UPPP), and rapid maxillary expansion (RME) [10]. Drug-induced sleep endoscopy (DISE) can identify the site of upper airway obstruction and provides guidance for further treatment [11]. Depending on the level of obstruction, additional surgical procedures can then be performed concurrently, or at a later time, following discussion with family members. Recently, Best et al. reported the findings from a retrospective case series of children with DS who underwent PSG following adenotonsillectomy for SDB [12]. In a patient population with DS and OSA with a mean age of 4.8 years, 35.4% of patients required at least one further surgical procedure and 23.1% of patients underwent DISE, which is now part of the clinical management algorithm of persistent OSA in patients with DS following adenotonsillectomy for these authors [12]. RME is a treatment approach that can significantly increase the volume of the upper airway volume and provides long-term benefits in adolescents with isolated maxillary narrowing [10,13]. Upper airway hypoglossal nerve stimulation has been shown to be effective in OSA and is now approved by the U.S. Food and Drug Administration (FDA) for patients over the age of 18 years using specific patient selection criteria [14]. A recently published case series that included six adolescent patients with DS and severe OSA reported that a hypoglossal nerve stimulator implant was well tolerated and resulted in a 56–85% reduction in the apnoea-hypopnea index (AHI) at 6-month to 12-month follow-up and also improved patient quality of life [15]. Although the number of children with DS and OSA in this case series was small, these results hold promise for a potentially effective treatment modality for children with DS who are unable to tolerate CPAP [15].

Primary care providers need to be aware of CPAP and non-CPAP options available for children with DS with residual SDB. Clinical assessment by a sleep physician is valuable for this patient population with the decision for different treatment options individualised, to improve quality of life for children with DS. The review by Horne and colleagues provides a timely and helpful summary of points for clinical practice, and the recommendations provided as a research agenda for SDB in children with DS, particularly for the establishment of a threshold of severity of SDB warranting treatment, will be strongly supported by physicians working in sleep medicine [1].

Conflicts of interest

None.

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