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Original Article

A four year follow-up of sleep and respiratory measures in elementary school-aged children with sleep disordered breathing

Anna Vlahandonis [a], Gillian M. Nixon [a,b], Margot J. Davey [a,b], Lisa M. Walter [a,1], Rosemary S.C. Horne [a,*,1]

[a] The Ritchie Centre, Monash Institute of Medical Research, Monash University, Melbourne, Australia
[b] Melbourne Children's Sleep Centre, Monash Children's Programme, Monash Medical Centre, Melbourne, Australia

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ABSTRACT

Objective: Little is known of the long-term prognosis of children treated for sleep disordered breathing (SDB) and even less of children with milder forms of SDB who remain untreated. We aimed to investigate the long-term sleep and respiratory outcomes of children with a range of SDB severities.
Methods: 41 children with SDB and 20 non snoring controls (mean age, 12.9 ± 0.2 y), underwent repeat overnight polysomnography (PSG) 4.0 ± 0.3 years after initial diagnosis. SDB severity, presence of snoring, sleep and respiratory parameters, sleep fragmentation index (SFI), the Pediatric Daytime Sleepiness Scale (PDSS), Sleep Disturbance Scale for Children (SDSC), and obstructive sleep apnea 18-item quality of life questionnaire were re assessed. Children with SDB were grouped into resolved (no snoring and obstructive apnea–hypopnea index [OAHI] <1) and unresolved (snoring or an OAHI ⩾1).
Results: At follow-up OAHI was reduced in both SDB groups ($p < 0.05$); however, 54% ($n = 22$) of children still continued to snore, having either persistent or new OSA ($n = 4$). In this unresolved group, sleep was significantly disrupted; % nonrapid eye movement stage 1 (NREM1) sleep and SFI were increased ($p < 0.05$), and total sleep time (TST) and sleep efficiency were decreased compared to the resolved and control groups ($p < 0.05$). Overall, 29% of children were treated, and of these, 67% had resolved SDB. SDB groups had higher PDSS, SDSC, and OSA-18 scores compared to controls at follow-up ($p < 0.01$).
Conclusions: Our study demonstrated that although SDB improved in the long-term, more than 50% of children had residual SDB (mostly primary snoring) and sleep disturbance. As even mild forms of SDB are known to have adverse cardiovascular, learning, and behavioral outcomes, which have implications for the health of these children.

1. Introduction

Snoring is the cardinal symptom of childhood sleep-disordered breathing (SDB), an umbrella term often used to encompass the range of severity of this disorder. SDB severity ranges from primary snoring (PS), which is not associated with any gas exchange abnormalities or sleep disturbance, to obstructive sleep apnea (OSA) which is associated with sleep fragmentation and repetitive hypoxia and hypercarbia. The most common cause of pediatric SDB is adenotonsillar hypertrophy. Although the etiology of SDB is different to that of adults, there is mounting evidence that children with SDB also have adverse health consequences. Our group has previously shown that all severities of SDB in elementary school-aged children are associated with elevated blood pressure (BP) [1], autonomic dysfunction [2], neurocognitive deficits [3], as well as behavioral and attention difficulties [4]. The finding that even children with PS, a condition once thought to be benign, also were subject to these consequences has significant implications, as the prevalence for PS is far greater than OSA. Up to 35% of children are estimated to have PS [5] compared to 2% to 3% of children who have OSA [6].

The first-line treatment for OSA in children is adenotonsillectomy (T&A); however, studies now show that this treatment is not as efficacious as once thought, with a number of children exhibiting residual or persistent SDB after treatment [7]. Furthermore, although a number of studies have assessed the effectiveness of T&A for the resolution of OSA [7–9], far fewer have examined outcomes in children who are not treated. Those studies that have assessed the natural history of SDB or the effects of treatment in children have been limited by variable or short follow-up periods (most ⩽6 mo) [10–13], or have not objectively assessed SDB severity using the gold standard of polysomnography (PSG) but rather have relied on parental report [11,13,14]. In addition, previous studies have included a wide age range that is skewed towards a younger population (mean ages, 6–10 y) [10–12,14,15]. Hence, little is known of the progression of SDB in

* Corresponding author. Address: The Ritchie Centre, Level 5, Monash Medical Centre, 246 Clayton Road, PO Box 5418 Clayton, Victoria 3168, Australia. Tel.: +61 3 95945100; fax: +61 3 95946811.
E-mail address: rosemary.horne@monash.edu (R.S.C. Horne).
[1] Denotes senior co-authorship.

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children as they get older and what impact it has on their sleep quality.

Sleep fragmentation has been found to be a common consequence of SDB in adults and has been shown to be a predictor of excessive daytime sleepiness [16]. There are a number of methods to quantify sleep fragmentation such as the identification of microarousals; however, this method is time consuming [17]. An alternative measure of sleep fragmentation is the sleep fragmentation index (SFI), which is defined as the number of stage shifts and awakenings divided by total time spent asleep [17]. This index, however, has rarely been assessed in children. Another method of assessing sleepiness is the sleep pressure score (SPS), which has been shown to be a sensitive measure in children.

The aim of our study was to assess both PSG and questionnaire-defined sleep and respiratory outcomes together with SFI in elementary school-aged children diagnosed with a range of severities of SDB over a long-term follow-up period of 4 years.

2. Methods

Approval for this project was granted by the Southern Health and Monash University Human Research Ethics Committees. Participation was voluntary and no monetary incentive was offered. Written informed consent was obtained from parents and verbal assent from the children on the night of the study.

2.1. Participants and study design

Our study was a prospective follow-up study of children who were originally studied between July 2004 and December 2008 when they were aged 7 to 13 years. At the baseline study, 155 children 116 of whom had been referred for assessment of suspected SDB and 39 non snoring control children who were recruited from the community were studied. The effect of SDB at initial diagnosis on BP [1,18], cardiovascular control [2], behavior and neurocognition [3,4], and sleep quality [19] have been previously published from this cohort. The current study began in 2009 and aimed to follow-up children who had a BP recording at baseline after a time frame of four years. Therefore, children without an adequate BP recording at baseline or those originally studied in 2004 were excluded, leaving 135 children ($n = 99$, SDB; $n = 36$, control) eligible to participate. Of those 135 children, 16 were not able to be contacted. The remaining 119 children were invited to participate in our study and initial contact was made via letter and then followed up with a telephone call to ascertain willingness to participate. Those who agreed to participate underwent repeat overnight PSG together with completion of a number of questionnaires to assess their quality of life and sleep habits and quality. Socioeconomic status (SES) was determined for each child according to the Australian Bureau of Statistics Socio-Economic Indexes for Areas 2006, which is based on postal code [20]. Treatment type (if any) received for SDB after initial diagnosis was documented via parental report on the night of the follow-up study. Treatment was not standardized as part of the study and reflected the clinical decision of the treating physician and parents.

2.2. Telephone questionnaire

Parents of participants who declined to take part in the follow-up study were asked to complete a short telephone questionnaire. This questionnaire included questions pertaining to the type of treatment their child received after the initial sleep study (if any), current snoring, and work of breathing at night. Reasons for refusal to participate in the study also were documented.

2.3. Protocol

Prior to the PSG study anthropometric measures including height and weight were recorded and body mass index (BMI) was calculated. BMI was converted to BMI z scores [21]. Participants underwent routine overnight pediatric PSG in the Melbourne Children's Sleep Centre using a commercially available PSG system (either Series S or Series E Sleep System, Compumedics, Melbourne, Australia at baseline, and Series E Sleep System at follow-up). The children went to bed once lead application was complete and were awakened at 6:00 AM. The following signals were recorded: electroencephalography (Cz, C4–A1, C3–A2, O2–A1, O1–A2), left and right electrooculogram, submental electromyogram (EMG), left and right anterior tibialis muscle and EMG, and electrocardiogram. Oxygen saturation (SpO_2) was measured using Masimo Radical®SET (Masimo Corp., CA, USA) at baseline and the Bitmos GmbH, (Dusseldorf, Germany) at follow-up, both of which use Masimo signal extraction technology for signal processing. All oximeters were set to a 2-second averaging time. Transcutaneous carbon dioxide was measured at baseline using a TCM4/40 (Radiometer, Copenhagen, Denmark) and at follow-up using a TINA TCM3 (Radiometer, Copenhagen, Denmark). Respiratory parameters recorded included thoracic and abdominal respiratory inductance plethysmography (Pro-Tech zRIP™ Effort Sensor, Pro-Tech Services Inc., Mukilteo, WA, USA), nasal pressure (Salter Style®, Salter Labs, Arvin, CA, USA) and oronasal airflow (Sandman®BreathSensor™, Child Airflow Thermistor, Tyco Healthcare, UK). In addition, BP was continuously and noninvasively measured using a Finometer™ (Finapres Medical Systems, Amsterdam, Netherlands) as previously described [1]; however, results are not presented here.

2.4. PSG data analysis and groups

Because the initial PSG studies had been conducted before the implementation of the American Academy of Sleep Medicine new scoring rules [22], all PSG studies were sleep staged using Rechtshaffen and Kales criteria [23] by experienced pediatric sleep technologists into wake after sleep onset (WASO), non rapid eye movement sleep stages 1, 2, 3 and 4 (NREM1-4), and rapid eye movement (REM) sleep. Sleep technologists were blinded to previous SDB severity or diagnostic group. Respiratory events were scored if they were $\geqslant 2$ respiratory cycles in duration, and the classification of apneas and hypopneas was based on the American Academy of Sleep Medicine criteria [24] with minor modifications in accordance to clinical practice at the time of the baseline study. Arousals were scored as either cortical arousals as defined by the American Sleep Disorders Association [25] or as sub cortical activations [26]. Sub cortical activations were scored when $\geqslant 2$ of the following were present, an increase in EMG or an increase in heart rate or a body movement. Respiratory events were only analyzed during periods without body movement. An obstructive apnea was defined as a decrease in flow to <10% of baseline in flow signal in the presence of continued or increased respiratory effort. An obstructive hypopnea was scored when a clear reduction from baseline in flow signal occurred in the presence of respiratory effort (with paradox or phase shift) and was associated with snoring or noisy breathing at event termination in conjunction with an arousal, awakening, or $\geqslant 3\%$ SpO_2 desaturation.

Sleep parameters recorded and calculated included, time in bed ([TIB], the time from lights out until the end of the study), sleep latency (the period from lights out until sleep onset), REM latency (the period from sleep onset to the onset of the first REM period), sleep period (the amount of time from sleep onset until the end of the study), total sleep time ([TST], the sleep period excluding any periods of wake), sleep efficiency (the ratio of TST to TIB), % NREM sleep (percentage of TST in NREM sleep), % REM sleep (percentage