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Sleep-related breathing disorders (SRBD) and attention-deficit/hyperactivity disorder (ADHD) in children attending an orthodontic clinic: a pilot case-control study

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A R T I C L E I N F O

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A B S T R A C T

Introduction: : Attention deficit hyperactivity disorder (ADHD) is the most common neurodevelopmental disorder in children. Sleep-related breathing disorders (SRBD), often caused by craniofacial abnormalities narrowing the airway, may be associated with ADHD. Therefore, orthodontists involved in the diagnosis and the treatment of sleep disorders should also be concerned by ADHD. The primary objective of this study was to explore the association between SRBD and ADHD in children referred to an orthodontic clinic. The study also investigated cephalometric characteristics in these children.

Material and Methods: : This pilot case-control study included 22 children attending an orthodontic clinic: 11 ADHD children diagnosed by a neuropsychologist and 11 control children matched for age, gender, BMI, and ethnicity. Control subjects were free of ADHD according to DSM-V classification. Type 3 ventilator polygraphy and cephalometric radiographs were performed. Parents answered the pediatric sleep questionnaire regarding SRBD symptoms of their child (PSQ-SRBD). A severity score and a restricted version of it removing 6 questions on behaviour were calculated. Both scores range from 0 to 1 with high values indicative of SRBD.

Results: : ADHD children evidenced a higher PSQ-SRBD severity score than controls (0.50 ± 0.14 vs. 0.11 ± 0.11, mean difference 0.39 ± 0.16, $p < 0.0001$) and similarly for the restricted score (0.33 ± 0.17 vs. 0.058 ± 0.056, mean difference 0.37 ± 0.17, $p = 0.0003$). Ventilator polygraphy parameters although not significantly different between groups were always higher in ADHD children (e.g. ventilatory effort 17.3 ± 7.0 vs 14.5 ± 12.3%; sleep fragmentation 14.5 ± 4.8 vs 13.5 ± 7.3 micro-arousals/h). Cephalometric features were comparable but ADHD subjects showed more mandibular deviation to the left ($p = 0.032$).

Conclusion: : Children with ADHD were more likely to suffer from sleep-related breathing disorders according to the PSQ-SRBD severity score, although the polygraph results did not confirm this finding. As for craniofacial anomalies, they only showed more mandibular deviation to the left. ADHD is important in pediatric, ENT and orthodontic practice to manage treatment of children optimally.

1. Introduction

Attention-deficit/hyperactivity disorder (ADHD) is a specific neurodevelopmental disorder in children with an estimated worldwide prevalence of 5 % and a decrease of life expectancy in many cases.[1] ADHD is a chronic, heritable, and heterogeneous syndrome arising from

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Glossary of abbreviations: ADHD, Attention Deficit and Hyperactivity Disorder; AHI, Apnea and Hypopnea Index; BMI, Body Mass Index; DSM, Diagnostic and Statistical Manual of Mental Disorders; ES, Effect Size; Jawac, Jaw Activity; OSA, Obstructive Sleep Apnea; PSQ, Pediatric Sleep Questionnaire; RDI, Respiratory Disturbance Index; RERA, Respiratory Effort-Related Arousal; SRBD, sleep-related Breathing Disorder.

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disturbances in brain functioning defined by impairing two or more symptoms of inattention and/or hyperactivity-impulsivity and cannot be better explained by another condition.[2] It begins before the age of 12 years. Behavioural, cognitive, emotional, social, and academic disorders are a disability for the child and a burden for parents and society.[3,4] The pathogenesis can result from the interaction between a genetic predisposition and environmental triggers.[1,4] Symptoms may be exacerbated by comorbidities or mimicked by other conditions. Thus, sleep disorders are frequently reported in children with ADHD, with an estimated prevalence ranging between 25 and 50 %.[5,6] Among sleep disorders, the obstructive sleep apnea (OSA) syndrome affects approximately 1 to 5 % of children. It can be associated with behavioural and neurocognitive problems, learning difficulties, growth retardation, cardiovascular and metabolic diseases.[7,8] Children affected by sleep-related breathing disorders (SRBD) may suffer from ADHD due to sleep fragmentation and hypoxia.[9] The first cause of airway obstruction in children is adenotonsillar hypertrophy, with a peak incidence between 2 and 8 years of age.[10] Obstruction can also result from structural narrowing of the pharyngeal cavity, usually caused by fat accumulation in the neck and underdevelopment of the dental arches. Obstructive sleep disorders can also be caused by narrowness and retrusion of the jaws. This can be assessed by a cephalometric examination which provides numerical measurements of angles and distances of the child's craniofacial morphology. Few studies have examined the complex interrelations between malocclusions, SRBD and ADHD, although several authors have suggested potential associations.[11–16] Therefore, the primary objective of this pilot study was to explore the presence of sleep-related breathing disorders in ADHD children. Secondarily, the study aimed to characterize craniofacial features in ADHD children undergoing cephalometry.

2. Materials & methods

2.1. Study patients

This pilot case-control study was conducted in the Department of orthodontics and dentofacial orthopedics, University hospital of Liege (Liege, Belgium). Twenty-two children (11 ADHD cases and 11 matched controls) were enrolled between October 2019 and January 2021. ADHD patients were diagnosed and referred by a neuropsychologist using the Diagnostic and Statistical Manual of Mental Disorders (DSM)-V international diagnostic criteria.[2] Controls were matched on age (± 12 months), gender, body mass index (BMI, ± 0.5 kg/m^2) and ethnicity. ADHD children were 6 −12 years old and naïve to medication for ADHD (methylphenidate or any other type of pharmacotherapy for ADHD). Controls were individuals free of ADHD according to DSM-V classification and referred to the orthodontic clinic. Exclusion criteria for both groups, were: (1) previous orthodontic treatment; (2) child or parent unable to understand instructions or to give informed consent; (3) craniofacial or neuromuscular syndrome; (4) history of trauma affecting jaw development. The study was approved by the Ethics Committee of the University Hospital (B7072020000077). The study was explained to the parents and patients, and informed consent was obtained.

2.2. Sleep variables

Parents were asked to complete the Pediatric Sleep Questionnaire about Sleep-Related Breathing Disorders (PSQ-SRBD) symptoms of their child. The PSQ-SRBD consists of 22 items about sleep, breathing and behaviour. A global severity score was calculated by dividing the number of symptoms (positive responses) by the total number of positive and negative answers, excluding questions that were unanswered. The severity score ranges between 0 and 1 and any score above 0.33 (one-third-of positive responses) strongly suggests SRBD.[10] A "restricted" severity score was also computed from the PSQ-SRBD by excluding the six behavioural items ("does not listen", "difficulty organizing", "easily distracted", "restless", "active like a battery", and "interrupts") concerning predominantly signs of inattention and hyperactivity.

Besides the PSQ-SRBD, all the subjects underwent a validated type 3 respiratory polygraphy (Somnolter® Nomics, Liege, Belgium) at home, including sensors for airflow, respiratory effort, heart rate, oximetry, and body position. Sleep breathing diagnosis was based on analysis of mandibular movements using non-invasive movement sensors placed on the chin and forehead.[11,12] Although the sensors were installed by a qualified healthcare technician to ensure proper recording, recordings of oximetry signals had to be discarded due to the difficulty of maintaining the oxygen saturation sensor on the children's fingers overnight. The following parameters were recorded: total recording time (h); total sleeping time (h); validity of signals; Apnea Hypopnea Index (AHI); Respiratory Disturbance Index (RDI) defined as the number of respiratory events; apnea, hypopnea, and respiratory effort-related arousals (RERA) per hour of sleep; respiratory effort and sleep fragmentation index defined as the number of micro-arousals per hour of sleep. The Jaw Activity (Jawac) algorithm detects mouth opening from as little as 0.3 mm of mandibular lowering and serves as a marker of respiratory effort and vigilance during sleep. It can distinguish between phases of sleep and wakefulness, including micro-awakenings often associated with respiratory events such as apnea. The Jawac sensor detects micro-arousals by neuro-muscular measurement. The total sleep time measured by the Jawac has shown strong correlation with sleeping time obtained by polysomnography ($r = 0.93$) .[17] The sleep assessment was conducted without the investigator knowing which group the study participants belonged to.

2.3. Cephalometric variables

For each patient, a lateral radiograph and a postero-anterior radiograph were taken using the Planmeca Promax 2D®. Thirty-eight cephalometric variables (17 for the postero-anterior analysis and 21 for the lateral one) were collected. All cephalometric measures were performed twice. The postero-anterior cephalometric landmarks came from Ricketts'analysis.[8] The lateral cephalometric landmarks were like those of the study of Katyal et al.[9]

2.4. Statistical analysis

A power calculation based on the comparison of the PSQ-SRBD severity score (primary outcome measure) between cases and controls showed that with at least 20 patients (10 cases and 10 matched controls), an effect size (ES) of at least 1.0 could be evidenced with a power of 80 % and a significance level of 5 %, assuming a "Normal" distribution and a two-sided paired *t*-test. Quantitative data were summarized by mean and SD whereas frequency tables were used for categorical data. Means of cases and controls were compared using the paired *t*-test and proportions by the McNemar (Fisher exact) paired test. Results were considered significant at the 5 % critical level ($p < 0.05$). The Benjamini-Hochberg method was applied to account for multiple comparisons. Statistical calculations were performed using SAS Version 9.4 and graphics using R Version 4.1.1.

3. Results

3.1. Demographic characteristics

Demographic characteristics of ADHD cases and matched controls are displayed in Table 1. The 11 ADHD patients (2 girls and 9 boys) were 8.8 ± 1.5 years old (range: 6.4 - 11.2 years) and their mean BMI was 15.8 ± 2.2 kg/m^2. As for ethnicity, most children were Caucasian (81.8 %).

3.2. Sleep variables

ADHD patients expressed significantly more positive responses than

Table 1
Characteristics of cases (ADHD patients) and matched controls.

Variable	Category	Cases($N = 11$)	Controls ($N = 11$)
Age (years)		8.8 ± 1.5	8.8 ± 1.4
Gender	Boy	9	9
	Girl	2	2
Ethnicity	Caucasian	9	9
	North African	1	1
	Mixed	1	1
BMI (kg/m^2)		15.8 ± 2.2	16.3 ± 1.3

controls for several PSQ-SRBD questionnaire items, specifically: A5 “breathed heavily “(73 % vs. 9 %, $p = 0.016$), B1 “tired in the morning” (82 % vs. 18 %, $p = 0.016$), C3 “did not listen” (82 % vs. 18 %, $p = 0.016$), C5 “had organizational difficulties” (91 % vs. 27 %, $p = 0.039$), C10 “were restless” (91 % vs. 27 %, $p = 0.016$), and C14 “agitated” (91 % vs. 18 %, $p = 0.0078$). All other items were comparable between the two groups. As a result (Table 2), a highly significant difference was observed between ADHD cases and controls for the global PSQ-SRBD severity score (0.50 ± 0.14 vs. 0.11 ± 0.11, mean difference 0.39 ± 0.16, ES=2.4, $p < 0.0001$) and similarly for the proportion of severity scores ≥ 0.33 (90 % vs. 0 %, $p = 0.0020$). The restricted PSQ-SRBD severity scores also differed (0.33 ± 0.17 vs. 0.058 ± 0.056, mean difference 0.37 ± 0.17, ES=2.2, $p = 0.0003$). All three p-values remained significant after adjusting for multiple comparisons. By contrast, the Jawac ventilatory polygraphy did not evidence differences between cases and controls, but mean values were higher in cases than in controls for all relevant parameters (Table 2). Ventilatory effort averaged 17.3 ± 7.3 % in the ADHD group and 14.5 ± 12.3 % in the control group. RDI was 2.6 ± 1.32 for ADHD children and 1.74 ± 2.0 for controls, AHI was 0.17 ± 0.51 and 0.01 ± 0.63 events/h, respectively, while sleep fragmentation was 14.5 ± 4.8 and 13.5 ± 7.3, respectively. Of note, ADHD patients slept on average 9.2 ± 1.2 h and controls 8.6 ± 1.2 h but overall, 13 (59 %) children had <9 h of sleep, the recommended sleeping time.[13]

3.3. *Cephalometric variables*

Cephalometric features of cases and controls are described in Table 3. No significant difference was found between the two groups for lateral cephalometric features. Similarly, on postero-anterior cephalometric analysis, the two groups were comparable, except for mandibular symmetry where a mean mandibular deviation to the left was observed in ADHD patients compared to controls (0.88 ± 1.32 vs. −0.52 ± 0.95, mean difference 1.40 ± 1.87, ES=0.75, $p = 0.032$). The latter p-value did not resist adjustment for multiple testing. The effect sizes of ADHD on anterior cephalometric features are displayed in Fig. 1A and on postero-anterior cephalometric features in Fig. 1B Noteworthy, in ADHD subjects, the anterior maxillo-mandibular height represented by the ANS-Me distance was increased (ES=0.48), the oropharynx was sagittally narrower (ES=−0.54), the width of the face was transversely narrower (ES=−0.54), the right maxillo-mandibular width was increased (ES=0.46), the distance between the left first molar and the jugal plane was reduced (ES=−0.46), the inferior intermolar width was increased (ES=0.41), the inferior intercanine distance was reduced (ES=−0.57), and, the angle between the Frankfurt plane and the orbital plane was increased (ES=0.43). The first three parameters are the most relevant. Although not reaching statistical significance, they globally display narrowness and vertical elongation with posterior compression of the face. The other parameters show a greater maxillary asymmetry in ADHD subjects.

Table 2
PSQ-SRBD scores and sleep data among cases (ADHD) and matched controls.

Variable	Cases ($N = 11$)	Controls ($N = 11$)	P-value
PSQ-SRBD			
Total severity score	0.50 ± 0.14	0.11 ± 0.11	<0.0001*
Number (%) of scores > 0.33	10 (90)	0 (0)	0.0020*
Restricted score	0.33 ± 0.17	0.058 ± 0.056	0.0003*
Respiratory polygraphy			
Total analysis time (h)	10.1 ± 1.3	9.6 ± 1.1	0.23
Sleep time (h)	9.2 ± 1.3	8.6 ± 1.2	0.23
Sleep time/analysis time (%)	90.8 ± 2.9	89.3 ± 7.1	0.57
Validity of Jawac signals (%)	95.9 ± 7.1	94.7 ± 7.1	0.73
Validity of thoracic belt (%)	99.4 ± 1.8	97.6 ± 2.3	0.11
Validity of abdominal belt (%)	95.6 ± 7.6	97.1 ± 3.9	0.61
Mean validity of the 2 belts (%)	97.6 ± 4.0	97.4 ± 3.0	0.92
AHI (number/h)	0.17 ± 0.51	0.01 ± 0.03	0.32
RDI (number/h)	2.6 ± 1.3	1.7 ± 2.0	0.20
Ventilatory effort (%)	17.3 ± 7.3	14.5 ± 12.3	0.47
Sleep fragmentation (number/h)	14.5 ± 4.8	13.5 ± 7.3	0.74

* Significant even after adjusting for multiple comparisons.

Table 3
Cephalometric characteristics on lateral and postero-anterior radiographs of cases (ADHD) and matched controls.

Variable	Cases ($N = 11$)	Controls ($N = 11$)	P-value
Lateral cephalometry			
SN (mm)	66.0 ± 3.6	64.7 ± 4.0	0.50
Ba-SN (°)	132.5 ± 4.1	130.9 ± 4.0	0.39
SNA (°)	80.4 ± 3.7	81.1 ± 2 .7	0.64
SNB (°)	75.7 ± 3.6	77.3 ± 3.2	0.30
ANB (°)	4.8 ± 2.0	3.8 ± 1.5	0.25
Wits (mm)	1.0 ± 3.8	0.3 ± 1.4	0.58
Co-A (mm)	78.3 ± 5.0	78.4 ± 3.4	0.95
Co-Gn (mm)	100.5 ± 7.4	101.0 ± 4.9	0.86
Co-A/Go-Gn (mm)	1.8 ± 1.4	1.5 ± 0.3	0.48
SN-PP (°)	6.8 ± 2.1	7.1 ± 3.4	0.76
FMA (°)	22.9 ± 4.9	21.5 ± 4.2	0.57
PP-MP (°)	26.4 ± 3.5	26.0 ± 8.7	0.90
Co-Go (mm)	47.7 ± 3.7	48.8 ± 5.2	0.67
ANS-Me (mm)	59.5 ± 5.7	57.7 ± 3.6	0.14
PP-U1 (°)	109.0 ± 9.7	107.7 ± 7.2	0.48
MP-L1 (°)	92.8 ± 10.1	94.4 ± 7.7	0.66
OP-FP (°)	9.7 ± 3.6	8.6 ± 3.4	0.51
AIS (mm)	11.1 ± 3.2	13.3 ± 3.4	0.10
PNS-AD1 (mm)	18.9 ± 4.1	19.8 ± 3.8	0.53
AD1-SP (mm)	10.8 ± 3.2	11.0 ± 3.0	0.92
MP-H (mm)	10.8 ± 4.9	9.8 ± 6.6	0.69
Postero-anterior cephalometry			
Facial width (mm)	112 ± 8.8	116 ± 4.5	0.10
Maxillary width (mm)	60.5 ± 4.4	61.5 ± 4.1	0.61
Mandibular width (mm)	77.5 ± 6.5	76.3 ± 4.2	0.51
Mandibular symmetry (°)	0.88 ± 1.3	−0.52 ± 0.95	0032*
Mx-Md relation right (°)	10.7 ± 2.3	9.4 ± 2.0	0.16
Mx-Md relation left (°)	10.7 ± 3.2	9.8 ± 2.4	0.33
B6/Jugal plane (mm)	6.5 ± 3.0	7.6 ± 1.5	0.27
6B/Jugal plane (mm)	6.0 ± 3.0	7.4 ± 1.5	0.16
Upper IM width (mm)	55.8 ± 3.4	56.2 ± 2.5	0.84
Lower IM width (mm)	55.0 ± 4.3	53.2 ± 2.7	0.20
Lower IC width (mm)	25.2 ± 2.9	27.2 ± 2.6	0.088
Molar relation right (mm)	0.58 ± 1.9	0.91 ± 1.0	0.49
Molar relation left (mm)	0.20 ± 2.0	0.85 ± 1.0	0.29
ZR-ZL/FP (°)	0.65 ± 1.3	0.05 ± 0.91	0.18
Maxillary plane/FP (°)	0.49 ± 1.4	−0.06 ± 1.2	0.30
Occlusal plane/FP (°)	0.48 ± 1.8	−0.13 ± 1.3	0.42
Mandibular plane/FP (°)	0.01 ± 1.3	−0.19 ± 0.56	0.65

IM intermolar; IC intercanine.

* No longer significant after adjusting for multiple comparisons.

4. Discussion

This study aimed to investigate the relationship between SRBD and ADHD in children attending an orthodontic clinic. It also strived to search for possible cephalometric anomalies in ADHD. Since this was a pilot study on a limited number of children, results should be interpreted cautiously.

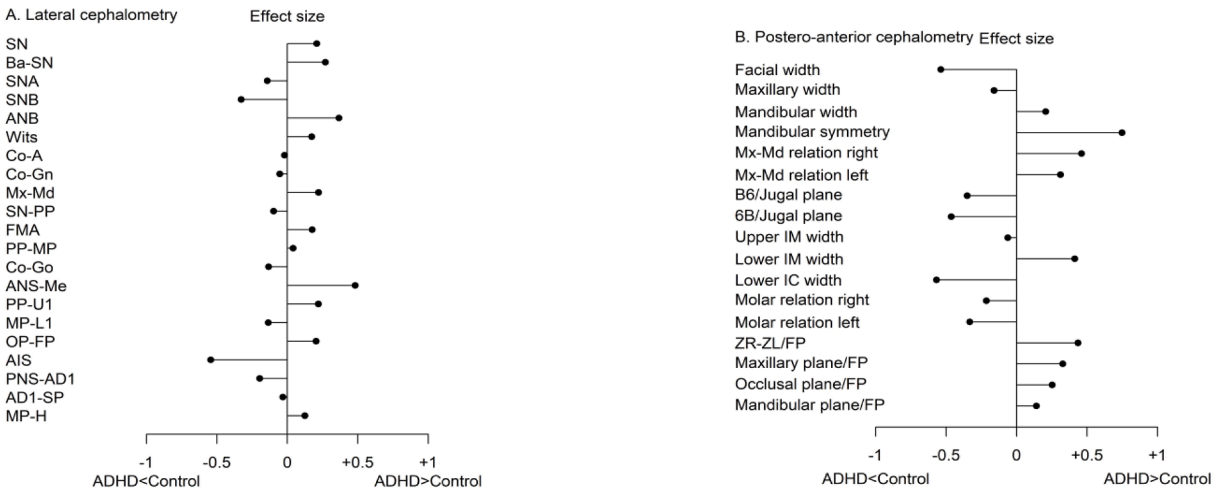

Fig. 1. Effect size (ES) of ADHD with respect to matched controls on features of lateral cephalometry (A) and on features of postero-anterior cephalometry (B). None of the ES was statistically significant after adjusting for multiple comparisons.

4.1. ADHD demographics

Although ADHD is more common in boys than in girls, the gender ratio of the study was far from the 2:1 ratio observed in the general pediatric population.[2] The most likely hypothesis is that girls with ADHD are currently underdiagnosed for two reasons. First, they more often present with the subtype “predominantly inattentive” than boys and unveil less frequently hyperactivity. Secondly, girls seem to hide their symptoms by adopting a more subdued and adaptive profile. Caucasian ethnicity was the most represented in this sample because it is the most common ethnicity in Belgium (about 67.3 %).[30,31] Some studies suggest that the distribution of ADHD varies according to ethnic origin. This could be explained by a different genetic predisposition to ADHD and other comorbidities (e.g., Asians would have higher rates of autism and language disorders and Caucasians more anxiety disorders). According to the Belgian National Health Interview Survey and the National Survey of Children's Health, the prevalence of ADHD is higher among Africans than Caucasians, while the US National Health and Nutrition Examination Survey showed a higher prevalence of ADHD among Caucasian children (12.65 %) aged 12–15 years than among Hispanics (7.11 %) and Africans (7.69 %).[32,33] Disparities seem to be explained partially by socio-cultural differences that may lead parents to consult less easily.

4.2. ADHD and SRDB

Based on the sleep-related pediatric questionnaire PSQ-SRBD, the study showed that ADHD patients scored higher than controls not only on the global severity scale but also on the restricted scale when excluding behavioural items. The proportions of positive responses in the ADHD group were notably greater than in the control group for several specific items of the questionnaire, respectively “breathed heavily”, “tired in the morning”, “did not listen”, “had organizational difficulties”, “were restless”, and “agitated”. Only the study by Andersson and Sonnesen considered sleep-related breathing disorders while focussing on malocclusions in ADHD children. Using a parental questionnaire, these authors found that ADHD patients had significantly less restful sleep, tended to sleep fewer hours per night, and did not feel rested in the morning.[16] However, they noted that there was still a risk of false positives in the case group and false negatives in the control group because parents of ADHD patients were interested in the study in the hope of finding an explanation for their child's difficulties.[18] Therefore, they may have been more likely to answer the questions in the affirmative way. In contrast, parents in the control group probably tended to consider that their child's sleep was normal and answered negatively in case of doubt. This response bias may have amplified the differences between the groups, even though these differences are still very large.

While the SRBD questionnaire evidenced differences in sleep characteristics of ADHD children compared to controls, results obtained by the respiratory polygraphy did not evidence significant differences, although globally mean values of the relevant parameters were systematically more perturbated in cases than in controls. Based on these observations, the conclusion of a clear association between ADHD and sleep breathing disorders remains subject to debate. A full night polysomnography (PSG) applied to all study children might be recommended for future work but was clinically difficult to implement in this preliminary work. The present study showed that more than half of the children had <9 h of sleep, a duration lower than the recommended time 9–12 h by the American Academy of Sleep Medicine (AASM) for children aged 6 to 12.[19] A lack of sleep is likely to affect cognitive function and behaviour during developmental age, and increase the risk of accidents, injuries, hypertension, obesity, diabetes, and depression.[19] It should be recognized that sleep duration in this study was measured on a single night and therefore may not accurately reflect habitual sleep patterns.

Regarding the sleep polygraphy, continuous and generally increasing respiratory effort is indicative of an obstructive sleep disorder. There is evidence that sequences of subtle increases in ventilatory effort may be sufficient to play a role in SRBD observed in children. Although there is no consensus on the pathological threshold for ventilatory effort measured by Jawac in children, the correlation between abnormal oesophageal pressure increases and mandibular movements suggests that 10–20 % effort would be problematic in the polygraphy analysis.[20] Consequently, the children in this study had, on average, a high ventilatory effort that crossed the first threshold of abnormality, indicating that their breathing during sleep was laboured and probably oral type. None of the children in this study was apneic, the threshold for OSA in children being set at 1 obstructive event/hour.[19] Obstructive events are rare in children but it is debatable they would be more prone to prolonged partial obstructions. It should be remembered that some children may have breathing difficulties without reaching the AHI threshold required to identify OSA. Therefore, they should be investigated for high upper airway resistance syndrome on increased respiratory effort as evidenced by sleep fragmentation. There is no consensus about the pathological threshold for the RDI (proportion of sleep disturbance due to respiratory events). Experts determine a threshold, ranging from 1.5 to 3/h. On this basis, the frequency of respiratory events remained moderate, but slightly higher in the ADHD group. Sleep fragmentation was based on the number of micro arousals per hour of sleep and was identified by the Jawac sensor. In children, it is considered abnormal from 11 to 14 events/hour. The sleep of children in this study was, on average, fragmented and insufficiently restorative.

4.3. ADHD and cephalometry

The study fell short in finding significant differences between ADHD children and controls for cephalometric features, except for mandibular symmetry. The overall severity of craniofacial abnormalities was similar in both groups. More lateral crossbites in the ADHD group generally reflect maxillary narrowness usually caused by thumb or nipple sucking habits and breathing difficulties like oral ventilation. This is one of the malocclusions most often associated with the risk of developing SRBD.[21] Few studies have explored craniofacial dysmorphosis in ADHD subjects. In a case-control study comparing dental casts of 51 children, Andersson and Sonnesen found that ADHD subjects had a significantly narrower upper arch in the intercanine region, and that the prevalence of posterior crossbite tended to be higher in this group.[16] These findings are close to those obtained here. Hasanin et al. found a significantly narrower airway in ADHD subjects on CBCT of 87 children.[22] Effect size analysis showed that in our ADHD subjects the sagittal dimension of the oropharynx represented by the AIS distance was smaller in the ADHD group. Atmetlla et al. observed on 83 children that, despite no difference in occlusal relationships, the ADHD group was associated with a more ogival palate and a greater hypodevelopment of the middle third of the face.[23] Two recent studies have shown a higher prevalence of malocclusions in ADHD subjects using the dental aesthetic index (DAI) .[24, 25] Mota-Veloso et al. obtained a significantly higher score in the ADHD group (42 % higher prevalence of malocclusions) in their cross-sectional study of 633 children aged 7 to 12 years.[25]

Wu et al. conducted a study on children with SRBD and found that ADHD was significantly more common in children aged 4–5 years with severe adenoid hypertrophy, and in children aged 6–11 years with severe tonsillar hypertrophy. In addition, allergic rhinitis was positively associated with OSA and ADHD in the 4–5 age group. They also showed an increased incidence of ADHD which might be related to the long-term effects of OSA-induced hypoxia on brain development and function.[26, 27] Furthermore, the review of Urbano et al. cited a dozen articles that observed beneficial effects of adenotonsillectomy on cognitive function, ADHD symptoms, and quality of life.[4] Huang and Guilleminault showed better improvement of ADHD symptoms with the surgery than with methylphenidate in SRBD children.[28] Chervin et al. suggested that, even in the absence of OSA, there are potential benefits of adenotonsillectomy in children with ADHD symptoms.[29] It is therefore important to determine whether malocclusions can lead to the same conclusions. The history of surgical removal of tonsils and vegetations and their hypertrophic nature were not recorded in this study, although the PNS-AD1 and AD1-SP cephalometric measurements were done to register pharyngeal patency at the level of the adenoid mass.

To the best of our knowledge, this is the first study evaluating, although not conclusively, SRBD by ventilatory polygraphy in ADHD patients. Among cephalometric findings, only left mandibular deviation was significantly more common in children with ADHD. A correlation between malocclusions and signs of SRBD could not be formally established in this study.

4.4. Study limitations

The study was based on a small number of children ($N = 22$). However, this was only a pilot study and its case-control (matched) design provided more power than a conventional comparative study of same sample size. The power calculation was based on the PSQ-SRBD severity index (primary endpoint) and therefore study conclusions should primarily pertain to the sleep questionnaire. All other study variables (sleep and cephalometry) should be interpreted cautiously. Further, it is worth mentioning that recruiting 6–12 years old subjects with ADHD is not an easy task. This may be explained by the difficulty for neuropsychologists. to convince parents of ADHD children to participate.

The case-control design of the study may be questioned but it allows to account for potential confounders such as gender, age, race, and overweight. While some other confounders have been partly accounted for (e.g., drug treatment, sleep duration, presence of vegetations/tonsils, type of breathing), other have not (e.g., tongue position, perinatal history, social environment, genetic factors). Another major confounder is malocclusion which is likely to interact in the relationship between ADHD and SRBD. Malocclusion can be assessed by cephalometry. Given that all study children were admitted to orthodontics, they underwent lateral and postero-anterior cephalometry. Since results showed no major difference between the two groups, the two groups may be considered a posteriori comparable “on average” for cephalometry, hence mitigating its potential influence in the association between ADHD and SRBD.

The fact that control children who were recruited in the specific environment of an orthodontic department and not in the general population may limit generalizability of findings. However, since all children were referred to orthodontics and hence eligible to undergo ambulatory sleep polygraphy and lateral and postero-anterior cephalometry, the study was made possible.

The mismatch between the statistical significance (even after adjusting for multiple comparisons) of PSQ-SRBD severity indices and the lack of significance of sleep-related polygraphy parameters remains a shortcoming of the study. Of note, though, all relevant sleep polygraphy parameters were quantitatively more affected in ADHD children, a finding that needs to be confirmed on larger patient groups. A rigorous “gold standard” in-hospital (controlled) polysomnography (PSG) would have been preferable but difficult to justify ethically and clinically on presumably healthy control children. The Jawac method, however, has been shown a reliable substitute to PSG.

5. Conclusions

Children with ADHD were more likely to suffer from sleep-related breathing disorders according to the PSQ-SRBD severity score, although the polygraph results did not confirm this finding. As for cephalometry, no specific feature characterized ADHD children but a possible greater mandibular deviation to the left. Knowledge of ADHD in children is essential for pediatricians, ear-nose-throat specialists and orthodontists to tailor communication and treatment of ADHD children optimally.

CRediT authorship contribution statement

Marie Laurent: Writing – original draft, Visualization, Validation, Software, Resources, Methodology, Investigation, Data curation, Conceptualization. **Adelin Albert:** Writing – review & editing, Writing – original draft, Methodology, Formal analysis, Conceptualization. **Matthieu Thimmesch:** Writing – review & editing. **Anne-Lise Poirrier:** Writing – original draft, Conceptualization. **Annick Bruwier:** Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Software, Resources, Methodology, Formal analysis, Conceptualization.

Declaration of competing interest

I and all the authors of this article have no conflict of interest.

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