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ORIGINAL SCIENTIFIC ARTICLE

Sleepiness, occlusion, dental arch and palatal dimensions in children attention deficit hyperactivity disorder (ADHD)

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Abstract

Aims This was to compare sleepiness, occlusion, dental arch and palatal dimensions between children with attention deficit hyperactivity disorders (ADHD) and healthy children (control group).

Methods 15 children with ADHD (10 boys, 5 girls, mean age 10.98 years) and 36 healthy age matched children (21 boys, 15 girls, mean age 10.60 years) were included. Intra-oral three-dimensional scans of the teeth and palate were performed to evaluate the occlusion, dental arch and palatal dimensions. Sleepiness was evaluated from the questionnaires. The differences between the two groups were analysed by Fisher's exact test and general linear models adjusted for age and gender.

Results The ADHD children had a significantly narrower dental arch at the gingival level of the canines ($p < 0.05$) and a tendency to increased prevalence of posterior cross-bite compared to the controls (13.3 vs. 0.0%, $p = 0.086$). The ADHD children snored significantly more ($p < 0.05$) and slept restlessly significantly more often ($p < 0.0005$) compared to the controls. The ADHD children had a tendency to sleep fewer hours during the night ($p = 0.066$) and felt inadequately rested in the morning ($p = 0.051$) compared to the controls.

Conclusion The results indicate that sleepiness and palatal width, especially the more anterior skeletal part of the palate, may be affected in children with ADHD. The results may prove valuable in the diagnosis and treatment planning of children with ADHD. Further studies are needed to investigate sleep and dental relations in children with ADHD.

Keywords Attention deficit hyperactivity disorder · Craniofacial abnormalities · Sleepiness · Child

Introduction

Attention deficit disorder (ADHD) is a common neurodevelopmental disorder with onset in childhood and often continuing into adulthood (Thapar and Cooper 2016) and is characterised by developmentally inappropriate levels of inattention, hyperactivity and impulsivity (American Psychiatric Association 2013). The symptoms must be present before the age of 12 and exist in different environments (American Psychiatric Association 2013). The symptoms of varying degrees of inattention, hyperactivity and impulsivity have great impact on the children's family life, relationship to peers, result of their education and future prospects in life (Davies 2014). The estimated prevalence of ADHD in children is 3.4% and the disorder is most common in boys (Willcutt 2012; Polanczyk et al. 2015). The aetiology is only partly understood and is multi-factorial (Gerlach et al. 2008). ADHD may be inherited and multiple genes as well as biological, environmental and psycho-social factors are considered in the aetiology (Gerlach et al. 2008; Thapar and Cooper 2016).

Children with ADHD show high co-morbidity of other neuro-psychiatric disorders such as autism spectrum disorders, depression and anxiety disorders, Tourettes syndrome, and obsessive–compulsive disorder (Gillberg et al. 2004). Other disorders frequently seen in children with ADHD are sleep-related respiratory disorders such as sleep-disordered breathing (SDB) and obstructive sleep apnea (OSA) (Huang et al. 2007; Sedky et al. 2014).

In children OSA is usually caused by excess of lymphoid tissue, airway inflammation or craniofacial deviations that result in decreased dimensions of the upper airways (Nespoli et al. 2013). The clinical characteristics of OSA in children differ from those seen in adults and may show a varied

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presentation (Sinha and Guilleminault 2010). The symptoms may include snoring and frequent arousals and hyperactivity and inattentiveness is seen in many children (Sinha and Guilleminault 2010). Since many children with ADHD suffer from OSA, it can be assumed that those with ADHD have a higher degree of upper airway obstruction than children without ADHD (Huang et al. 2007; Sedky et al. 2014).

In patients with OSA and/or upper airway obstruction an extended head posture and a low position of the tongue is often observed (Ricketts 1968; Solow et al. 1993). Previous studies have shown that head posture has an influence on craniofacial development and development of malocclusion (Solow and Tallgren 1976; Solow and Siersbaek-Nielsen 1986; Solow and Sonnesen 1998). The lowered position of the tongue also has an influence on craniofacial development, especially that of the palate and upper dental arch dimensions (Ricketts1968).

As an association between ADHD and SDB/OSA, as well as between SDB/OSA and deviation in the craniofacial morphology including occlusion, has previously been shown, there may be an altered craniofacial morphology and occlusion in children with ADHD. Few studies have previously focused on orofacial characteristics of children with ADHD (Atmetlla et al. 2006; Sabuncuoglu 2013). The previous studies were a clinical evaluation of the craniofacial morphology and occlusion where a significant difference was found in craniofacial morphology between an ADHD group and a control group but no significant difference in sagittal molar relation, overjet or overbite. It is still unclear if space conditions, dental arch and palatal dimensions or occurrence of cross bite are affected in children with ADHD.

The aims of this study were to compare (1) sleepiness between children with ADHD and a healthy control group, (2) occurrence of malocclusions and space anomalies between children with ADHD and healthy controls and (3) dental arch and palatal dimensions between children with ADHD and healthy controls.

Materials and methods

The material included questionnaires of sleepiness and sleep quality, and intraoral scans of the teeth and palate obtained from children with ADHD and a control group of healthy children. The ADHD group comprised all children with ADHD verified by a psychiatrist, referred to two schools specialising in ADHD, Furesø ADHD and autism centre, Syvstjernen and Borupgårdskolen in the north of Zealand, Denmark in the period June 2015–January 2016, who met the inclusion criteria described below. The control group comprised all the healthy children who attended for a routine dental examination during autumn 2015 or winter 2016 at the community dental health service, Panum Institute, Copenhagen, and who met the inclusion criteria described below. The study followed the guidelines of the Helsinki Declaration and had been approved by the Scientific-Ethical Committee of Copenhagen (H-1-2014-130) and the Danish data protection agency (j.nr. 2015-57-0121).

The children with ADHD comprised 15 children: 5 girls (aged 9.05–12.50; mean 10.70 years) and 10 boys (aged 9.39–12.86; mean 11.13 years). The inclusion criteria were: (1) verified ADHD as primary diagnosis by a psychiatrist according to the Danish national guidelines for diagnosing and treating AHDH in children and adolescents (Sundhedsstyrelsen 2014), (2) age between 9 and 12 years, (3) no prior history of prolonged sucking habits, and (4) informed consent from parents/guardian. The children were well-regulated by ADHD related medicine. Two children showed co-morbidity as Asperger's syndrome and Myotonic dystrophia type II, and two children had an indication for orthodontic treatment.

The control group comprised 36 healthy children: 15 girls (aged 9.27–12.11; mean 10.74 years) and 21 boys (aged 9.21–12.41; mean 10.49 years). The inclusion criteria were: (1) age between 9 and 12 years, (2) no history of known diseases or syndromes, known sleep disturbances including snoring and mouth breathing, (3) no prior history of prolonged sucking habits, (4) no enlarged tonsils assessed clinically and (5) informed consent from parents/guardian. Four children had an indication for orthodontic treatment.

In both groups the intraoral scans of the children with indication for orthodontic treatment were taken prior to an orthodontic treatment. Before the analysis of the data, the study was blinded regarding groups.

Questionnaires

To evaluate sleepiness, the children and their parents were asked to complete two questionnaires, the Epworth sleepiness scale (ESS) and the Berlin Questionnaire (BQ) both translated into Danish and modified for children (Johns 1991; Netzer et al. 1999). The ESS comprised 8 questions concerning how likely the child would fall asleep in different settings and gave a score for sleepiness between 0 and 24, where the highest score was the most sleepiness (Table 1). The modified BQ comprised nine questions concerning snoring and the quality of sleep (Table 2). If a question was unanswered or misunderstood, the answer was excluded from the analysis of this particular question.

Intraoral scans

The teeth and the palate were scanned by a 3-dimensional transportable dental scanner (Lythos, Ormco

Table 1 Mean and standard deviations (SD) of age, ESS score, hours of sleep, occlusion and dental arch and palatal dimensions in the two groups

Variable	ADHD Group			Control Group			P
	N	Mean	SD	N	Mean	SD	
Age (years)	15	10.98	1.34	36	10.60	0.93	0.18
ESS score	15	3.6	3.54	35	3.37	3.14	0.86
Hours of sleep	15	9.03	0.85	36	9.57	0.79	0.066
Overjet (mm)	15	3.52	2.52	34	3.49	1.83	0.81
Overbite (mm)	15	2.71	1.44	35	3.01	1.60	0.24
Space (mm)	13	1.75	3.60	32	2.47	4.29	0.51
DistanceTG3 (mm)	9	24.84	2.26	22	25.90	1.75	0.038*
Distance TG6 (mm)	15	35.02	2.22	36	35.63	2.84	0.47
Distance TC3 (mm)	9	32.82	2.24	22	33.09	2.50	0.13
Distance TC6 (mm)	15	40.85	2.68	36	41.46	3.21	0.48
Height V3 (mm)	9	4.28	1.28	22	4.00	1.39	0.34
Height V6 (mm)	13	11.85	1.71	34	10.96	1.64	0.13

ADHD attention deficit hyperactivity disorder

*p < 0.05

Corporations, Glendore, California, USA). All scans were performed by the same person in either a class room setting. (ADHD group) or a dental clinic (control group). If any of the measuring points were not clearly visible because of the quality of the scan, or a missing tooth due to exfoliation, this particular measurement was excluded from the analysis.

Occlusion, space anomalies, dental arch and palatal dimensions were assessed on the intraoral scans of the teeth. The data from the scans were analysed using O3DM (OrthoLab, Poznan, Poland) and OnyxCeph 3TM (Image Instruments, Chemnitz, Germany). The following registrations and measurements were made:

- Stage of dental development according to Björk et al. (1964).
- Occlusion (Björk et al. 1964): sagittal molar occlusion, overjet, overbite, and cross-bite/scissor-bite.
- Space anomalies: for children in the permanent dentition according to Proffit et al. (2007) and for children in the mixed dentition according to Tanaka and Johnston (1974).
- Transversal dimensions of the maxilla (Lione et al. 2014) (Fig. 1):
    - TG3: distance between the centres of the dento-gingival junctions of the primary or permanent canines.
    - TG6: distance between the centres of the dento-gingival junctions of the permanent first molars.
    - TC3: distance between the cusp tips of the primary or permanent canines.
    - TC6: distance between the mesio-lingual cusp tips of the permanent first molars.
- Palatal height (Lione et al. 2014) (Fig. 1):
    - V3: the perpendicular to TG3 in the mid-sagittal plane.
    - V6: the perpendicular to TG6 in the mid-sagittal plane.

Reliability of the method

The intra-observer reliability was determined by repeating all measurements on 25 randomly selected scans after 10 days. The reliability for the sagittal and transversal molar occlusion and dental stage showed very good agreement ($\kappa = 0.92–1.0$) (Landis and Koch 1977). For the overjet, overbite, space conditions, dental arch and palatal dimension no systematic error was found. The method error ranged from 0.014 to 0.64 mm and Houston reliability coefficient from 0.79 to 1.0 (Dahlberg 1940; Houston 1983).

Statistical methods

The statistical analyses were performed using SAS (v 9.4, Cary, NC, USA). The results of the tests were considered to be significant at $p < 0.05$.

The continuous data were tested by Q–Q plots on the residuals and were found to be normally distributed.

The categorical data (gender, snoring, results from BQ, dental stage and sagittal and transversal molar relationships) were tested by Fisher's exact test. The continuous data (age, ESS score, hours of sleep, overjet, overbite, space conditions, dental arch- and palatal dimensions) were tested by a general linear model adjusted for age and gender.

Table 2 Results of the Berlin questionnaire (BQ) in the two groups

Variable	ADHD group						Control group						P
	N	Never	1–2 times a month	1–2 times a week	3–4 times a week	Almost every night	N	Never	1–2 times a month	1–2 times a week	3–4 times a week	Almost every night	
How often was snoring reported	14	11	1	1	1	0	36	36	0	0	0	0	0.019*
Has anyone noticed that you have stopped breathing during sleep?	14	14	0	0	0	0	36	36	0	0	0	0	–
Have you experienced awakenings with gasping for breath or feeling that you're suffocating?	14	14	0	0	0	0	36	36	0	0	0	0	–
How often do you feel inadequately rested?	14	2	4	3	5	0	35	14	12	7	2	0	0.051
How often do you feel abnormally or extremely tired in the day?	13	3	8	1	0	1	36	17	16	2	1	0	0.27
Are you restless in your sleep resulting in curled up bed sheets?	14	5	0	3	0	6	35	25	6	0	1	3	0.0005***

ADHD attention deficit hyperactivity disorder

*$p < 0.05$, ***$p < 0.001$

Results

The results are presented in Tables 1 and 2. There were no significant differences between age, gender and stage of dental development between the two groups. The children with ADHD had a significantly narrower palate at the gingival level of the canines compared to the control children ($p < 0.05$, Table 1). There was a tendency to a higher frequency of cross-bite in the ADHD group (13.33 vs. 0.00%; $p = 0.086$). There were no significant differences between the two groups in the occurrence of sagittal molar occlusion, overjet, overbite, space anomalies, palatal height, or dental arch dimensions measured at the level of the first maxillary molars (Table 1). The analysis of the questionnaires showed that children in the ADHD group snored more ($p < 0.05$) and slept more restlessly ($p < 0.0005$) compared to the children in the control group (Table 2). The children in the ADHD group had a tendency to sleep fewer hours during the night and felt inadequately rested in the morning compared to the controls (Table 2). There was no significant difference in how tired the children felt during the day or in ESS score (Table 1, 2).

Discussion

The aim of this study was to compare sleep, occlusion, dental arch and palatal dimensions between children with ADHD and healthy children. Few other studies have focused on orofacial characteristics of children with ADHD (Atmetlla et al. 2006; Sabuncuoglu 2013) whereas more studies have looked at sleepiness in children with ADHD (Sadeh et al. 2006; Cortese et al. 2009; Galland et al. 2011; Wiebe et al. 2013; Herman 2015). Thus, it is still unclear if occlusion, space conditions or dental arch and palatal dimensions are affected in children with ADHD.

The ADHD sample comprised children between 9 and 12 years who attended two schools who specialised in ADHD in the period June 2015–January 2016 and who were accepted to participate in the study. The sample may be small but it included all children in a particular period of time with ADHD as their primary diagnosis. Two of the children showed co-morbidity as Asperger's syndrome and Myotonic dystrophia type II, which is in agreement with previous studies reporting that ADHD is associated with high co-morbidity of other syndromes (Gillberg et al. 2004). Asperger's syndrome may not have any influence on the results because of the different aetiologies. Myotonic dystrophia is known to affect muscle strength and therefore could have an influence on the craniofacial development (Kiliaridis et al. 1989). The child in the present study suffered from type II of the disorder, which is a mild form that progresses

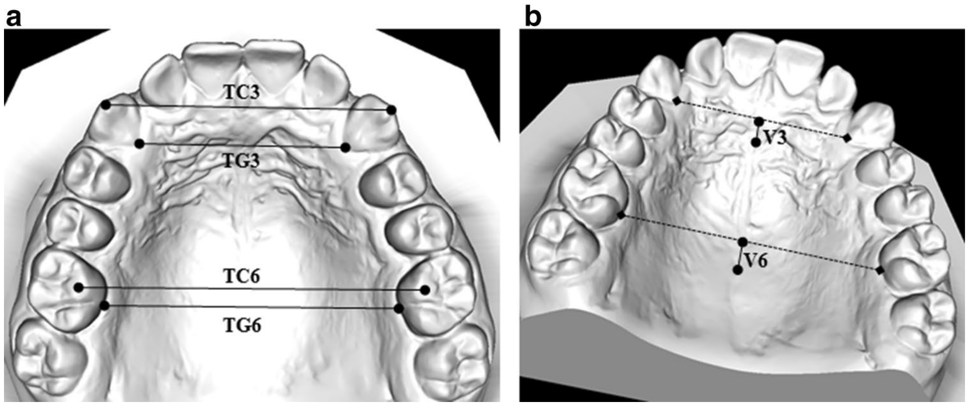

Fig. 1 Illustration of the dental arch and palatal analyses on the intraoral scans (Lione et al. 2014). **a** TG3: distance between the centres of the dento-gingival junctions of the primary or permanent canines, TG6: distance between the centres of the dento-gingival junctions of the permanent first molars, TC3: distance between the cusp tips of the primary or permanent canines, TC6: distance between the mesio-lingual cusp tips of the permanent first molars. **b** V3: the perpendicular to TG3 in the mid-sagittal plane, V6: the perpendicular to TG6 in the midsagittal plane

slowly and rarely affects the facial muscles (Ashizawa and Sarkar 2011). Therefore it may not affect the results of the present study.

Two of the children with ADHD and four of the children in the control group had an indication for orthodontic treatment. The prevalence of children with indication for orthodontic treatment was similar in the two groups and represents the prevalence in a Danish child population in general (Helm 1970).

The questionnaires used in the study were both validated and widely used (Netzer et al. 1999; Johns 2000). To be used in this study both had to be translated to Danish and adapted to children. No validated Danish version of ESS exists, but the one used in the present study is similar to the validated Norwegian version (Beiske et al. 2009) and Danish and Norwegian languages are very similar. Recently an English version of ESS for children was published (Janssen et al. 2007). This version was not available when the present study was initiated and therefore it was not possible to use the recent version in the present study. Furthermore, it is important to emphasise that the questionnaires cannot be used in diagnostics of OSA/SDB and are not a substitute for sleep evaluation by polysomnography (Margallo et al. 2014; Khaledi-Paveh et al. 2016). The aim of the present study was not to evaluate OSA/SDB but to record tiredness and subjective sleep quality by questionnaires.

In the present study snoring occurred significantly more often in children with ADHD compared to the control group. This is in agreement with a previous study by Galland et al. (2011). In order to exclude children with sleep-related respiratory disorders in the control group, snoring was one of the exclusion criteria of that group. This may have affected the result. The finding in the present study of significantly more restless sleep in the ADHD group, a tendency to sleep fewer hours per night and to feel inadequately rested was also in agreement with previous studies (Sadeh et al. 2006; Cortese et al. 2009; Herman 2015). Consideration may be given to whether the differences found in sleepiness between the two groups were caused by ADHD or SDB/OSA and this may further emphasise the importance of the differential diagnosis between ADHD and SDB/OSA in children. No significant difference in daytime sleepiness or in the ESS score was found between the two groups in the present research which is in agreement with a previous study (Wiebe et al. 2013). This finding may be expected as the most common symptom of poor sleep in children is hyperactivity and not excessive daytime sleepiness as in adults (Sinha and Guilleminault 2010).

No significant differences were found in the occurrence of malocclusion and space anomalies between the two groups, which is in agreement with Atmetlla et al. (2006). Previously, associations have been shown between ADHD and SDB/OSA as well as between SDB/OSA and deviation in the craniofacial morphology including occlusion (Solow and Tallgren 1976; Solow and Siersbaek-Nielsen 1986; Solow and Sonnesen 1998; Sinha and Guilleminault 2010). The children in the present study were not diagnosed by polysomnography to detect SDB/OSA but it has previously been shown that SDB/OSA occurred frequently in children with ADHD (Huang et al. 2007; Sedky et al. 2014). Therefore a significant difference in malocclusion, and space anomalies between the two groups, may be expected. In the present study a tendency to an increased occurrence of posterior cross-bites was found in the children with ADHD which may support the previously demonstrated associations.

In the present study the palatal width at the gingival level anteriorly was significantly narrower in the children with ADHD compared to the healthy children. This has not previously been reported in the literature. Unfortunately, it was not possible to measure the distance between the canines in all subjects because of exfoliation of primary teeth. There was no significant difference in the width of the palate posteriorly or the height of the palate between the two groups. Previously, an extended head posture has been found in relation to obstruction of the upper airway which leads to a low tongue position (Ricketts 1968), which may result in a narrow palate and cross-bite (Ricketts 1968). Previously, it has been shown that SDB/OSA occurred frequently in children with ADHD due to obstruction of the upper airways (Huang et al. 2007; Sedky et al. 2014). Therefore a narrower palate may be expected in children with ADHD as found in the present study. However, the palate was only significantly narrower at the gingival level representing the skeletal width of the palate. An explanation why the narrow palate was only found at the gingival level and not at the cusp tip level in the present study may be due to the dento-alveolar compensatory mechanism (Solow 1980). The teeth may have compensated for the narrow palate and therefore no significant differences at the cusp tip level and only a tendency to an increased occurrence in cross-bite in children with ADHD was shown herein.

Studies have shown that rapid maxillary expansion can decrease the nasal airway resistance in children with upper airway obstruction (Timms 1986; White et al. 1989). The decreased nasal airway resistance may improve the quality of sleep (Pirelli et al. 2004). In the present study it was shown that sleepiness and the width of the palate, especially the skeletal part anteriorly, may be affected in children with ADHD. The results of the present study may lead to increased focus on early diagnosis and expansion of the maxilla in children with ADHD symptoms and emphasises the importance of the differential diagnosis between ADHD and SDB/OSA in children.

Conclusion

Sleepiness and palatal width, especially the more skeletal part anteriorly of the palate, may be affected in children with ADHD. The results may prove valuable in the diagnosis and treatment planning of children with ADHD. Further studies are needed to investigate sleep and dental relations in ADHD children.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The study was approved by the Scientific-Ethical Committee of Copenhagen (H-1-2014-130) and the Danish data protection agency (j.nr. 2015-57-0121).

Informed consent Oral and written informed consent was given by the children's parents/guardian.

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