

VISUOMOTOR INTEGRATION SKILLS IN CHILDREN AFFECTED BY OBSTRUCTIVE SLEEP APNEA SYNDROME: A CASE-CONTROL STUDY

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ABSTRACT

Introduction: Sleep related breathing disorders (SRBD) consist of frequent and repetitive episodes of pharyngeal obstruction during sleep, with consequent intermittent hypoxia, sleep architecture fragmentation, daytime sleepiness and/or behavioural problems and executive impairment in children. When untreated, SRBD and obstructive sleep apnea syndrome (OSA) mainly, may impact school performance, cognition, metabolism, and cardiovascular function.

Aim of the present study is assessing the visuomotor integration skills in children affected by OSA.

Materials and methods: 57 subjects affected by mild-to severe OSA, PSG diagnosed according to international diagnostic criteria, (31 males and 26 females) (mean age 10.8; SD \pm 2.49) and 83 healthy children (45 males and 38 females) (mean age 9.95; SD \pm 1.87; $p = 0.725$). All subjects underwent assessment of motor coordination skills with Movement-ABC tests and visual-motor integration ability with Visual Motor Integration (VMI) test.

Results: The subjects with OSA show a worse average performances in all items of Movement ABC ($p < 0.001$) respect of controls. Specifically, children with OSAS show significantly higher values of total points ($p < 0.001$), manual dexterity ($p < 0.001$), ball skills ($p < 0.001$) and balance ($p < 0.001$). Accordingly, the average centile in OSA children at the MABC-test is significantly reduced compared with controls ($p < 0.001$). (Table 1) On the other hand, the VMI test evaluation among children with OSAS shows worst result in total Visuo-Motor Integration ($p < 0.001$), and in Motor Coordination sub-item ($p < 0.001$) than controls. (Table 1).

Conclusion: Our results also support for children and adolescents the hypothesis that executive functioning deficits might be linked primarily to the degree of severity nocturnal hypoxemia rather than daytime sleepiness, although several other studies are needed.

Keywords: obstructive sleep apnea syndrome, OSA, visual-motor integration, visual-motor training skills.

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Introduction

Sleep related breathing disorders (SRBD) consist of frequent and repetitive episodes of pharyngeal obstruction during sleep, with consequent intermittent hypoxia, sleep architecture fragmentation, daytime sleepiness and/or behavioural problems and executive impairment in children⁽¹⁻⁵⁾.

Moreover, when untreated, SRBD may impact school performance, cognition⁽²⁾, metabolism, and cardiovascular function. In developmental age, the same alterations may be present in other pathological conditions such as migraine without aura⁽⁶⁻¹⁷⁾, pin-pointing once again the known close link between neurological impairment and sleep in children⁽¹⁸⁻²⁸⁾.

On the other hand, learning and neuropsychological ability seem to have a key role in quality of life and disease management in more neurological conditions in developmental age⁽²⁹⁻³¹⁾.

Specifically, some studies revealed a specific neuropsychological impairment both in adult⁽³²⁾ in children affected by obstructive sleep apnea syndrome (OSA)⁽²⁾. In 2013, Esposito et al⁽³³⁾ reported a significant impairment in visuo-motor and motor-coordination competencies in a sample of enuretic children highlighting the role of sleep troubles in neuropsychological impairment. In this light, scarce attention has been reserved on visuo-spatial and visuo-motor integration skills in children affected by other sleep disorders such as OSA.

Herein, the aim of the present study is assessing the visuomotor integration skills in children affected by OSA.

Materials and methods

Study population comprised 57 subjects affected by OSA, diagnosed according to international diagnostic criteria, (31 males and 26 females) (mean age 10.8; SD \pm 2.49) and 83 healthy children (45 males and 38 females) (mean age 9.95; SD \pm 1.87; $p = 0.725$). All subjects underwent assessment of motor coordination skills with Movement ABC tests and visual-motor integration ability with Visual Motor Integration (VMI) test.

All subjects of both groups were recruited within the same urban area, all were Caucasian and homogeneous for socioeconomic status

Ethical approval from the local Research Ethics Center and informed parental consent were obtained.

Polysomnographic (PSG) Data

After reviewing and analyzing PSG data collected from inpatients children between January and June 2016 to establish the presence of OSA in the experimental group, OSA severity was determined according to the current guidelines specified by the American Academy of Sleep Medicine (AASM): mild OSA was defined by an obstructive apnea-hypopnea index (o AHI) of 1 to <5 events per hour; moderate OSA was defined as was defined as ≥ 5 to <10 events/hour, and severe OSA ≥ 10 events/hour.

Motor coordination skills evaluation

Movement assessment battery for children (M-ABC) The impairment of motor coordination performance relative to age expectations was determined

using the Movement Assessment Battery for Children (Movement-ABC). This test is frequently used in both clinical and research settings to assess children for motor coordination impairment and has high reliability and validity⁽³⁴⁾. In fact, it assesses fine and gross motor skills using three manual dexterity tasks, two ball skills tasks, and three balance tasks, each of which is scored on a five-point scale. The raw score of each item is then converted to a score scale ranging from zero to five. The higher score indicates a less-than-adequate performance.

Consequently, 0 reflects a complete success by the candidate on the task examined, while 5 reflects a failure in the execution of the task, in fact a failed (F), an inappropriate (I), or refused (R) performances are transformed into 5. The sum of the eight scores of items corresponds to the total score of disability, ranging between 0 and 40, where a lower score is a result of implementing the best move. The content of the items differs depending on the age of the child examined, with increasing difficulty according to age, so that the battery is made up of four different types of activities considered to be made in relation to age (4-6 years, 7-8 years, 9-10 years, and 11-12 years). Each subject was assessed individually in about 20-40 min⁽³⁴⁾. The total impairment score was calculated from these individual tasks and is used to generate a centile score compared to the standardization sample.

Visual-Motor Integration skills evaluation

The fine motor coordination and the visual-motor integration was assessed with the Beery Visual Motor Integration task⁽³⁵⁾, a paper-and pencil test where children have to imitate or copy up to 27 geometric forms with increasing complexity using paper and pencil. Copying errors were marked if they reflected problems in fine motor coordination, rather than a pure visuo-spatial problem. The task is specifically designed for children and takes about 10 min. The Beery VMI scores were standardized for age and gender using normative data for the Italian general population⁽³⁵⁾. The centile scores were used for diagnosing the visual-motor abnormalities in our sample, and a value $\leq 5^{\text{th}}$ was considered for visual motor integration impairment.

Statistical analysis

For comparison between the two groups (OSAS and controls) it was applied t- testing and Chi-square test, where appropriate. P values < 0.05 were considered statistically significant.

For statistical analysis it used the software STATISTICA (data analysis software system, version 6, StatSoft, Inc. (2001). For the comparison between the groups the t-test was applied. P values<0.05 were considered statistically significant.

Results

For the present study we selected only children affected by mild-to severe OSA (57 subjects). The two groups of subjects (57 vs 83 controls OSAS) were statistically comparable for age ($10.08 \pm 2:49$ vs. 9.95 ± 1.87 ; $p = 0.725$) and gender ($p = 0.878$).

The subjects with OSAS show a worse average performances in all items of Movement ABC ($p < 0.001$) respect of controls. Specifically, children with OSAS show significantly higher values of total points ($33,078 \pm 12,422$ vs. $7,906 \pm 2,514$; $p < 0.001$), manual dexterity (13.068 ± 2.419 vs 2.947 ± 2.526 , $P < 0.001$), ball skills (8.501 ± 1.914 vs 2.067 ± 1.839) and balance ($11,892 \pm 1.756$ vs 1.801 ± 1.044 , $p < 0.001$). Accordingly, the average centile in OSAS children at the MABC-test is significantly reduced compared with controls (9.445 ± 5.973 vs $45,590 \pm 20,789$; $p < 0.001$) (Table 1).

On the other hand, the VMI test evaluation among children with OSAS shows worst result in total Visuo-Motor Integration ($13,819 \pm 8,061$ vs. $56,431 \pm 11,018$; $p < 0.001$), and in Motor Coordination sub-item ($18,623 \pm 7,439$ vs $52,820 \pm 15,788$; $p < 0.001$) than controls (Table 1).

| | OSA N=57 | Controls N=83 | P |
|--------------------------------------|----------------|------------------|--------|
| M-ABC Dexterity | 13.068± 2.419 | 2.947±2.526 | <0.001 |
| M-ABC Ball skills | 8.501±1.914 | 2.067±1.839 | <0.001 |
| M-ABC Balance | 11.892± 1.756 | 1.801±1.044 | <0.001 |
| M-ABC Total score | 33.078± 12.422 | 7.906±2.514 | <0.001 |
| M-ABC Centile | 9.445± 5.973 | 45.590±20.789 | <0.001 |
| Visual Motor Integration Total Score | 13.819±8.061 | 56.431±11.018 | <0.001 |
| VMI Visual Task | 46.891±21.712 | 51.703±22.186 | 0.206 |
| VMI Motor Task | 18.623±7.439 | 52.820±15.788 | <0.001 |

Table 1: shows the comparison of performance at the Movement-ABC and the VMI tests evaluation in children with obstructive sleep apnea syndrome (OSA) and controls. For the comparison between the groups the t-Test was applied. P values <0.05 were considered statistically significant.

Discussion

OSA in children can cause significant consequences, affecting the CNS, the cardiovascular systems, metabolic, and somatic growth, resulting a general reduction in daytime life activities and in quality of life⁽³⁶⁾. Actually, the pathogenetic mechanism involved in OSA consequences seems to be the intermittent hypoxia, related consequent hypercapnia, repeated intrathoracic pressure swings, and frequent arousal. These mechanisms can contribute to the oxidative stress and pro-inflammatory status, particularly in obese children^(37;38).

According to the research hypothesis, a significant difference has been reported in visuomotor-coordinative skills in subjects with OSAS than in control subjects. The scores of the balance of skill, dexterity and skill with the ball have shown that children with OSA have visuomotor skills really lower than normal, indicating and emphasizing once again the importance of precocious diagnosis. In order to manage pediatric OSA in the better manner, medical, surgical, orthodontic and pharmacological interventions are disposable, but in this light also rehabilitative treatment such as behavioural/psychomotor therapy may be included in therapeutic management⁽³⁹⁾.

In general, children affected by OSA appears to be dominated by the slow pace of development concepts which, however, do not seem to be related to degree of severity of the respiratory disorder. Several studies have focused on the role of prefrontal areas as possible relay zones between OSA and cognitive impairment, linked to reported reduction in frontal-parietal regions and hippocampal gray matter by MRI studies⁽⁴⁰⁾.

Our results, therefore, suggest the presence of a close relationship between nocturnal breathing troubles and prefrontal cortex areas activity, underlined by the particular efficiency reduction in subjects presenting a higher AHI.

Our results also support for children and adolescents the hypothesis that executive functioning deficits might be linked primarily to the degree of severity nocturnal hypoxemia rather than daytime sleepiness, although several other studies are needed.

References

- 1) Santamaria F, Esposito M, Montella S, Cantone E, Mollica C, De Stefano S, Mirra V, Carotenuto M. *Sleep disordered breathing and airway disease in primary ciliary dyskinesia*. *Respirology*. 2014 May; 19(4): 570-5. doi: 10.1111/resp.12273.
- 2) Esposito M, Antinolfi L, Gallai B, Parisi L, Roccella M, Marotta R, Lavano SM, Mazzotta G, Precenzano F, Carotenuto M. *Executive dysfunction in children affected by obstructive sleep apnea syndrome: an observational study*. *Neuropsychiatr Dis Treat*. 2013; 9: 1087-94. doi: 10.2147/NDT.S47287.
- 3) Carotenuto M, Gimigliano F, Fiordelisi G, Ruberto M, Esposito M. *Positional abnormalities during sleep in children affected by obstructive sleep apnea syndrome: the putative role of kinetic muscular chains*. *Med Hypotheses*. 2013 Aug; 81(2): 306-8. doi: 10.1016/j.mehy.2013.04.023.
- 4) Carotenuto M, Esposito M, Parisi L, Gallai B, Marotta R, Pascotto A, Roccella M. *Depressive symptoms and childhood sleep apnea syndrome*. *Neuropsychiatr Dis Treat*. 2012; 8: 369-73. doi: 10.2147/NDT.S35974.
- 5) Carotenuto M, Esposito M, Pascotto A. *Facial patterns and primary nocturnal enuresis in children*. *Sleep Breath*. 2011 May; 15(2): 221-7. doi:10.1007/s11325-010-0388-6.
- 6) Verrotti A, Carotenuto M, Altieri L, Parisi P, Tozzi E, Belcastro V, Esposito M, Guastaferrro N, Ciuti A, Mohn A, Chiarelli F, Agostinelli S. *Migraine and obesity: metabolic parameters and response to a weight loss programme*. *Pediatr Obes*. 2015 Jun; 10(3): 220-5. doi: 10.1111/ijpo.245
- 7) Esposito M, Parisi L, Gallai B, Marotta R, Di Dona A, Lavano SM, Roccella M, Carotenuto M. *Attachment styles in children affected by migraine without aura*. *Neuropsychiatr Dis Treat*. 2013; 9: 1513-9. doi: 10.2147/NDT.S52716. PMID: PMC3794987.
- 8) Esposito M, Roccella M, Gallai B, Parisi L, Lavano SM, Marotta R, Carotenuto M. *Maternal personality profile of children affected by migraine*. *Neuropsychiatr Dis Treat*. 2013; 9: 1351-8. doi: 10.2147/NDT.S51554.
- 9) Esposito M, Marotta R, Gallai B, Parisi L, Patriciello G, Lavano SM, Mazzotta G, Roccella M, Carotenuto M. *Temperamental characteristics in childhood migraine without aura: a multicenter study*. *Neuropsychiatr Dis Treat*. 2013; 9: 1187-92. doi:10.2147/NDT.S50458.
- 10) Esposito M, Gallai B, Parisi L, Castaldo L, Marotta R, Lavano SM, Mazzotta G, Roccella M, Carotenuto M. *Self-concept evaluation and migraine without aura in childhood*. *Neuropsychiatr Dis Treat*. 2013; 9: 1061-6. doi: 10.2147/NDT.S49364.
- 11) Esposito M, Parisi P, Miano S, Carotenuto M. *Migraine and periodic limb movement disorders in sleep in children: a preliminary case-control study*. *J Headache Pain*. 2013 Jul 1; 14: 57. doi: 10.1186/1129-2377-14-57.
- 12) Esposito M, Roccella M, Parisi L, Gallai B, Carotenuto M. *Hypersomnia in children affected by migraine without aura: a questionnaire-based case-control study*. *Neuropsychiatr Dis Treat*. 2013; 9: 289-94. doi: 10.2147/NDT.S42182.
- 13) Esposito M, Pascotto A, Gallai B, Parisi L, Roccella M, Marotta R, Lavano SM, Gritti A, Mazzotta G, Carotenuto M. *Can headache impair intellectual abilities in children? An observational study*. *Neuropsychiatr Dis Treat*. 2012; 8: 509-13. doi:10.2147/NDT.S36863.
- 14) Esposito M, Verrotti A, Gimigliano F, Ruberto M, Agostinelli S, Scuccimarra G, Pascotto A, Carotenuto M. *Motor coordination impairment and migraine in children: a new comorbidity?* *Eur J Pediatr*. 2012 Nov; 171(11):1599-604. doi:10.1007/s00431-012-1759-8
- 15) Balottin U, Fusar Poli P, Termine C, Molteni S, Galli F. *Psychopathological symptoms in child and adolescent migraine and tension-type headache: a meta-analysis*. *Cephalalgia*. 2013 Jan; 33(2): 112-22. doi: 10.1177/0333102412468386.
- 16) Balottin U, Termine C. *Recommendations for the management of migraine in paediatric patients*. *Expert Opin Pharmacother*. 2007 Apr; 8(6): 731-44. Review.
- 17) Balottin U, Chiappedi M, Rossi M, Termine C, Nappi G. *Childhood and adolescent migraine: a neuropsychiatric disorder?* *Med Hypotheses*. 2011 Jun; 76(6): 778-81. doi: 10.1016/j.mehy.2011.02.016.
- 18) Carotenuto M, Esposito M, Cortese S, Laino D, Verrotti A. *Children with developmental dyslexia showed greater sleep disturbances than controls, including problems initiating and maintaining sleep*. *Acta Paediatr*. 2016 Sep; 105(9):1079-82. doi: 10.1111/apa.13472.
- 19) Carotenuto M, Parisi P, Esposito M, Cortese S, Elia M. *Sleep alterations in children with refractory epileptic encephalopathies: a polysomnographic study*. *Epilepsy Behav*. 2014 Jun; 35: 50-3. doi: 10.1016/j.yebeh.2014.03.009.
- 20) Esposito M, Parisi P, Miano S, Carotenuto M. *Migraine and periodic limb movement disorders in sleep in children: a preliminary case-control study*. *J Headache Pain*. 2013 Jul 1; 14: 57. doi: 10.1186/1129-2377-14-57.
- 21) Esposito M, Gallai B, Parisi L, Roccella M, Marotta R, Lavano SM, Mazzotta G, Carotenuto M. *Primary nocturnal enuresis as a risk factor for sleep disorders: an observational questionnaire-based multicenter study*. *Neuropsychiatr Dis Treat*. 2013; 9: 437-43. doi: 10.2147/NDT.S43673.
- 22) Esposito M, Carotenuto M. *Intellectual disabilities and power spectra analysis during sleep: a new perspective on borderline intellectual functioning*. *J Intellect Disabil Res*. 2014 May; 58(5): 421-9. doi: 10.1111/jir.12036.
- 23) Carotenuto M, Esposito M, D'Aniello A, Ripa CD, Precenzano F, Pascotto A, Bravaccio C, Elia M. *Polysomnographic findings in Rett syndrome: a case-control study*. *Sleep Breath*. 2013 Mar; 17(1): 93-8. doi: 10.1007/s11325-012-0654-x.
- 24) Carotenuto M, Esposito M, Precenzano F, Castaldo L, Roccella M. *Cosleeping in childhood migraine*. *Minerva Pediatr*. 2011 Apr; 63(2): 105-9.
- 25) Esposito M, Carotenuto M, Roccella M. *Primary nocturnal enuresis and learning disability*. *Minerva Pediatr*. 2011 Apr; 63(2): 99-104.
- 26) Esposito M, Carotenuto M. *Borderline intellectual functioning and sleep: the role of cyclic alternating pattern*. *Neurosci Lett*. 2010 Nov 19; 485(2): 89-93.

- doi:10.1016/j.neulet.2010.08.062.
- 27) Carotenuto M, Esposito M, Pascotto A. *Migraine and enuresis in children: An unusual correlation?* Med Hypotheses. 2010 Jul; 75(1): 120-2. doi:10.1016/j.mehy.2010.02.004.
- 28) Cavanna AE, David K, Bandera V, Termine C, Balottin U, Schrag A, Selai C. *Health-related quality of life in Gilles de la Tourette syndrome: a decade of research.* Behav Neurol. 2013; 27(1): 83-93. doi: 10.3233/BEN-120296. Review.
- 29) Calderoni S, Muratori F, Leggero C, Narzisi A, Apicella F, Balottin U, Carigi T, Maestro S, Fabbro F, Urgesi C. *Neuropsychological functioning in children and adolescents with restrictive-type anorexia nervosa: an in-depth investigation with NEPSY-II.* J Clin Exp Neuropsychol. 2013; 35(2): 167-79. doi:10.1080/13803395.2012.760536
- 30) Borgatti R, Piccinelli P, Montiroso R, Donati G, Rampani A, Molteni L, Tofani A, Nicoli F, Zucca C, Bresolin N, Balottin U. *Study of attentional processes in children with idiopathic epilepsy by Conners' Continuous Performance Test.* J Child Neurol. 2004 Jul; 19(7): 509-15.
- 31) Piccinelli P, Beghi E, Borgatti R, Ferri M, Giordano L, Romeo A, Termine C, Viri M, Zucca C, Balottin U. *Neuropsychological and behavioural aspects in children and adolescents with idiopathic epilepsy at diagnosis and after 12 months of treatment.* Seizure. 2010 Nov; 19(9): 540-6. doi:10.1016/j.seizure.2010.07.014
- 32) Salorio CF, White DA, Piccirillo J, et al. *Learning, memory, and executive control in individuals with obstructive sleep apnea syndrome.* J Clin Exp Neuropsychol 2002; 24:1, 93-100.
- 33) Esposito M, Gallai B, Parisi L, Roccella M, Marotta R, Lavano SM, Mazzotta G, Patriciello G, Precenzano F, Carotenuto M. *Visuomotor competencies and primary monosymptomatic nocturnal enuresis in prepubertal aged children.* Neuropsychiatr Dis Treat. 2013;9:921-6. doi: 10.2147/NDT.S46772.
- 34) Henderson SE, Sugden DA (1992) *Movement assessment battery for children manual.* The Psychological Corporation Ltd, London
- 35) Beery KE, Beery NA (2004) *The Beery-Buktenica developmental test of visual-motor integration, vol 5.* NCS: Pearson Inc, Minneapolis, MN
- 36) Blechner M, Williamson AA. *Consequences of Obstructive Sleep Apnea in Children.* Curr Probl Pediatr Adolesc Health Care. 2016 Jan; 46(1): 19-26. doi: 10.1016/j.cppeds.2015.10.007
- 37) Gozal D, Kheirandish-Gozal L, Bhattacharjee R, Kim J. *C-reactive protein and obstructive sleep apnea syndrome in children.* Front Biosci (Elite Ed). 2012 Jun 1; 4: 2410-22.
- 38) Carotenuto M, Santoro N, Grandone A, Santoro E, Pascotto C, Pascotto A, Perrone L, del Giudice EM. *The insulin gene variable number of tandem repeats (INS VNTR) genotype and sleep disordered breathing in childhood obesity.* J Endocrinol Invest. 2009 Oct; 32(9): 752-5. doi: 10.3275/6398
- 39) Esposito M, Gimigliano F, Ruberto M, Marotta R, Gallai B, Parisi L, Lavano SM, Mazzotta G, Roccella M, Carotenuto M. *Psychomotor approach in children affected by nonretentive fecal soiling (FNRFs): a new rehabilitative purpose.* Neuropsychiatr Dis Treat. 2013; 9: 1433-41. doi: 10.2147/NDT.S51257
- 40) Tummala S, Roy B, Park B, Kang DW, Woo MA, Harper RM, Kumar R. *Associations between brain white matter integrity and disease severity in obstructive sleep apnea.* J Neurosci Res. 2016 Jun 18. doi: 10.1002/jnr.23788

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