## OSAHS

# Behavioural disorders and parental stress in children suffering from obstructive sleep apnoea syndrome: a pre- and post-adenotonsillectomy confrontation

Disordini comportamentali e indice di stress genitoriale nei bambini affetti da Sindrome dell'apnea ostruttiva nel sonno: confronto pre e post adenotonsillectomia

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#### SUMMARY

The primary goal of the present study was to compare breathing difficulties resulting from OSAS to possible cognitive-behavioural problems of the child linked to their parents' emotional-relational aspects. There is strong evidence that sleep breathing disorders are associated with behavioural alterations, a tendency towards aggressiveness, weak school performance and a clear disorder in continuous and selective attention other than vigilance status. Not all patients suffering from OSA have cognitive and/or behavioural manifestations; furthermore, the degree of dysfunction that the patient may present does not seem to be associated with the seriousness of sleep breathing disorder (SBD). It is therefore likely that genetic susceptibility associated with particular environmental factors has a role in determining phenotypic manifestations which are unique for every single patient. Questionnaires were given to parents, one regarding executive functions and one regarding parental stress: Conners' Rating Scale Revised; Parenting Stress Index. All parents of children who suffer from moderate to severe OSA, with a McGill score of 3 to 4 and with no exclusion criteria are included in the study; behavioural and parental stress evaluation was made during hospitalisation and at 6 months after adenotonsillectomy. The results show that resolving OSAS led to important improvements in the competence and behavioural attitudes of the patient, as well as in relational and management difficulties by parents. The identification of such indicators could represent a support to surgical programming, even in nonsevere SBD. Future research will have the goal of identifying standardised risk indicators that can provide further indications for surgical treatment in children up to 5 years of age.

KEY WORDS: obstructive sleep apnea syndrome (OSAS), parental stress, excessive daily drowsiness, Conner's rating scale revised (CRS-R), parenting stress index, McGill score

#### RIASSUNTO

Scopo del presente lavoro è valutare la relazione esistente tra le difficoltà respiratorie legate all'OSAS ed i possibili disordini neurocomportamentali correlando questi ultimi al grado di stress genitoriale. Non tutti i pazienti affetti da OSAS presentano questo tipo di problematiche tuttavia i disturbi di comportamento non sembrano essere associati con la severità del SBD; è probabile che una suscettibilità genetica associata a particolari fattori ambientali abbia un ruolo determinante nello sviluppo fenotipico di queste manifestazioni. Sono stati utilizzati due questionari: Conners' Rating Scale Revised; Parenting Stress Index. Sono stati arruolati nello studio pazienti con OSAS di grado moderato-severo, con Mc Gill score 3 o 4 in cui non fossero presenti criteri di esclusione, la valutazione dei genitori è stata effettuata durante il ricovero e 6 mesi dopo l'intervento di adenotonsillectomia. I risultati ottenuti hanno dimostrato che risolvendo l'OSAS si ottengono miglioramenti nelle competenze e nel comportamento dei pazienti ed inoltre si riducono le difficoltà genitoriali. L'identificazione di tali indicatori può rappresentare un supporto all'indicazione chirurgica anche in casi di DRS non severo.

PAROLE CHIAVE: sindrome dell'apnea ostruttiva del sonno (OSAS), stress genitoriale, eccessiva sonnolenza diurna, questionario di Conner's rivisitato (CRS-R), punteggio di McGill

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# Introduction

In 1976, Guillelminault was the first to describe, in a sample of 8 patients, the association between sleep breathing disorders and difficulty in learning, presence of school problems and the appearance of clear mood alterations that were somehow connected to a more or less accentuated daily drowsiness <sup>1</sup>.

In the last 40 years, the literature has provided a concrete support to what was a mere observational evaluation. Today, we know that sleep breathing disorders are clinically expressed in a continuum which has its origins in primary snoring and which its highest expression sees in obstructive sleep apnoea syndrome (OSAS). Night ventilator dynamic in a patient suffering from obstructive sleep apnoea (OSA) results in totally compromised hypopnoeic and apnoeic events, secondary to a partial or total obstruction of the respiratory system, determining a de-structuring of sleep with significant alteration of normal patterns <sup>2</sup>.

It is now acknowledged that non-treated OSA can lead to important consequences for the child's health, starting from a normal postural and ponderal growth to the presence of important cardiovascular alterations, which can lead to systemic arterial and pulmonary hypertension. Furthermore, there is strong evidence that sleep breathing disorders are associated with behavioural alterations, a tendency towards aggressiveness, weak school performance and a clear disorder in continuous and selective attention other than vigilance status <sup>3</sup>. Neuro-behavioural manifestations can be explained by intermittent hypoxic phenomena associated with the de-structuring of normal sleep stages, and animal models provide the plausibility that OSA contributes to neuro-cognitive deficits; in fact, experiments on young mice subjected to intermittent hypoxia during sleep highlight similar difficulties in conditioned learning <sup>4</sup>.

MRI and spectroscopy scans have demonstrated that apnoeic patients suffer from an alteration of metabolites in the left hippocampus and right frontal cortex <sup>5</sup>; in the prefontal cortex, OSA-mediated hypoxic damage might play a role in the development of this cerebral area, with irreversible consequences even if apnoeic condition is resolved <sup>6</sup>.

Many questions are yet to be answered. It is known that – in its initial stage – fragmented sleep is less frequent in a paediatric age; in particular, the microstructure of sleep in EEG tracings (A1 phase and A1 index of the CAP, Cycling Alternating Pattern) is modified, thus suggesting the nonrestorative nature of non-REM sleep in these patients <sup>7</sup>.

Oxidative stress can also play a role by increasing the loss of neuronal elements through direct damage as well as through activation of secondary inflammatory processes and reparative events <sup>7</sup>.

Last, but not least, it is opportune to consider that not all patients suffering from OSA have cognitive and/or behavioural manifestations; furthermore, the degree of dysfunction that the patient may present does not seem to be associated with the severity of sleep breathing disorder (SBD). It is, therefore, likely that genetic susceptibility associated with particular environmental factors has a role in determining phenotypic manifestations that are unique for every single patient <sup>8</sup>.

Recent studies have confirmed the importance that fragmented sleep has on the genesis of attentive problems, whereas intermittent hypoxaemia, by acting mainly on a state of inflammation of the vascular area, is responsible for alterations in the hippocampus and pre-frontal region, mainly modifying executive functions <sup>9</sup>.

As there are no sufficient studies that deal with the child's behaviour and parental stress in the presence of paediatric OSAS, we carried out a study to evaluate both these aspects in a group of patients treated in our paediatric hospital.

It is now acknowledged in the literature that excessive daily drowsiness in the paediatric population suffering from SBD is present in 40-50% of cases.

Our purpose is to confirm these findings, evaluating performance of the child to the family context and trying to understand how this factor is influenced by the patient's health and how it represents a conditioning "environmental factor".

## **Materials and methods**

#### Procedure

All parents of children who suffer from moderate to severe OSA, with a McGill score of 3 to 4 (McGill score is a pulsossimetric score which consent a diagnosis of OSAS without performing a regular polysomnography and indicate priority for surgical intervention) and with no exclusion criteria (association with other genetic syndromes, cognitive retardation, or diagnosed language disorders under treatment, certified ADHD, mild night breathing disorders, previously ORL surgery) at our centre for Sleep Breathing Disorders of the ORL Operative Unite over a period from September 2014 to February 2016 were invited to participate in the study. Parents who took part in the research signed an informed consent approved by the Institutional Review Board.

Behavioural and parental stress evaluation was made during hospitalisation and at 6 months after adenotonsillectomy, in line with the international literature <sup>12-14</sup> to verify the absence of rehabilitation and/or medical therapies (methylphenidate and psycho-stimulant generally). Post-surgery questionnaires were filled-in during a routine check-up at 6 months after surgery.

A psychologist handed out the questionnaires and explained them to parents, who then filled them in autonomously in the waiting room. The PSI questionnaire was completed by mothers and not fathers.

In order to avoid missing data, the psychologist checked that all questionnaires were properly completed.

The study was carried out in compliance with the Helsinki declaration.

#### Observation questionnaires

Questionnaires were given to parents, one regarding executive functions and one regarding parental stress. During the same evaluation meeting, children underwent non-structured qualitative behavioural observation.

*Conners' Rating Scale Revised: (CRS-R: C. Keith Conners)*<sup>10</sup>: this is a structured questionnaire composed of 80 questions which investigated problematic or psycho-pathological behaviours during developmental age, from 3 to 17. There are three versions of the questionnaire, one for parents (CPRS-R), one for teachers (CTRS-R) and a self-report for adolescents from 12 to 17 (YSR-R).

There are 14 areas of investigation: 1) Oppositivity 2) Cognitive disorders 3) Hyperactivity-Impulsiveness 4) Anxiety-Shyness 5) Perfectionism 6) Social Issues 7) Psychosomatic traits 8) ADHD index 9) CGI – Restlessness/ Impulsiveness 10) Emotional Instability 11) CGI – Total 12) DSM IV – Lack of Attention 13) DSM IV – Hyperactivity/Impulsiveness 14) DSM IV-Total.

The questionnaire includes 0-4 multiple choice. In our study, we used the parental version.

*Parenting Stress Index (Richard Abidin)*<sup>11</sup>: this is a multiple choice questionnaire with a score between 0 and 4, composed of 36 questions, which investigates parental stress meant as discrepancy perceived by parents among the resources available and the need to maintain a parental role.

Specifically, it investigates four areas: 1) Parental distress; 2) Dysfunctional child-parent interaction; 3) Difficult child; 4) Defensive response.

#### Statistical analysis of data

Categorical data are transcribed as numbers and percentage, while continuous data are transcribed as average and  $\pm$  SD. As these are ordinal data, statistical analysis was elaborated with Wilcoxon Test for paired data. A value of p < 0.05 was considered significant, and a value of p < 0.01 as even more significant. Statistical elaboration was carried out with STATA 14 statistical software.

#### 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 (Ospedale Pediatrico Bambino Gesù Ethical Committee) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

### **Results**

Overall, 50 children of age ranging from 28 months to 82 months were found eligible (68% boys) (average: 44;  $\pm$  SD:10.3).

Abandonment rate was 0%. All children chosen, in fact, underwent evaluation before surgery and at 6 months after surgery.

Mothers who filled in the questionnaire were asked about their educational and economic level; 18% had secondary school diploma, a little over half had a high school diploma (52%), while the remaining 30% had a degree. Their socioeconomical level was similar to their educational level, i.e. 20% had a low level, a little more than half (54%) had medium level and about one-quarter of mothers had a high social economic level (26%) (Tab. I).

As shown in Table II by Conners' scales, the most significant items, with p < 0.01, were: oppositivity (p = 0.0008), hyperactivity (p = 0.001), perfectionism (p = 0.003), restlessness/impulsiveness (p = 0.0003), emotional instability (p = 0.0005) and general difficult behaviour (p = 0.0008).

In particular, there was an increase in 24% of children with standard oppositivity post-surgery, compared to presurgery evaluation (from 62% of sample to 86%); in the hyperactivity subscale, there was an increase in 20% of

**Table I.** Socio-demographic characteristics of sample (N = 50).

	Ν	(%)
Gender		
Male	34	(68)
Pre-surgery chronological age, months		
(average, $\pm$ SD)	44	(± 10,3)
Post-surgery chronological age, months		
[(average, $\pm$ DS)	50	(± 10,3)
Education		
Secondary School Diploma	9	(18)
High School Diploma	26	(52)
Degree	15	(30)
Socio-economic level		
Low	10	(20)
Medium	27	(54)
High	13	(26)

Table II. Pre-post-surgery confrontation, results of Conners' test (entire sample).

	Pre		Post		P value*
	N	(%)	Ν	(%)	
Opposition					
Average	31	(62)	43	(86)	0.0008
Borderline	14	(28)	4	(8)	
Pathological	5	(10)	3	(6)	
Inattention/ Cognitive disorders					
Average	40	(80)	42	(84)	0.2674
Borderline	6	(12)	5	(10)	
Pathological	4	(8)	3	(6)	
Hyperactivity					
Average	32	(64)	42	(84)	0.0014
Borderline	9	(18)	7	(14)	
Pathological	9	(18)	1	(2)	
Anxiety/Shyness					
Average	42	(84)	43	(86)	0.40
Borderline	4	(8)	5	(10)	
Pathological	4	(8)	2	(4)	
Perfectionism					
Average	32	(64)	42	(84)	0.003
Borderline	10	(20)	6	(12)	
Pathological	8	(16)	2	(4)	
Social issues					
Average	48	(96)	50	(100)	0.16
Borderline	1	(2)	0	(-)	
Pathological	1	(2)	0	(-)	
Psycho-somatic disorders					
Average	37	(74)	44	(88)	0.02
Borderline	8	(16)	5	(10)	
Pathological	5	(10)	1	(2)	
ADHD					
Average	34	(68)	40	(80)	0.038
Borderline	10	(20)	6	(12)	
Pathological	6	(12)	4	(8)	
Restlesness/impulsiveness					
Average	29	(58)	40	(80)	0.0003
Borderline	11	(22)	9	(18)	
Pathological	10	(20)	1	(2)	
Emotional instability					
Average	33	(66)	45	(90)	0.0005
Borderline	12	(24)	2	(4)	
Pathological	5	(10)	3	(6)	
General behavioural issues					
Average	32	(64)	42	(84)	0.0008
Borderline	10	(20)	7	(14)	
Pathological	8	(16)	1	(2)	

	Pre		Post		P value*
	Ν	(%)	N	(%)	
DSM-IV inattention					
Average	38	(76)	43	(86)	0.02
Borderline	8	(16)	4	(8)	
Pathological	4	(8)	3	(6)	
DSM-IV hyperactivity/impulsiveness					
Average	37	(74)	44	(88)	0.01
Borderline	9	(18)	6	(12)	
Pathological	4	(8)	0	(-)	
DSM-IV ADHD combined					
Average	37	(74)	42	(84)	0.05
Borderline	9	(18)	6	(12)	
Pathological	4	(8)	2	(4)	

Table II. Pre-post-surgery confrontation, results of Conners' test (entire sample) (follows).

\* Wilcoxon test for paired data

children in the standard (pre-post-surgery: 64% vs. 84%); during follow-up, children with problems of perfectionism were statistically decreased (pathological pre-post: 16% vs. 4%); almost all children with pathological restlessness/ impulsivity went back to standard/borderline (from 20% to 2%). A similar outcome resulted in sub-scale "general difficult behaviour" (from 16% to 2%); lastly, emotional instability also improved, ranging from 66% to 90% of children. Sub-scales with p < 0.05 and p ≥ 0.01 were as follows: psychosomatic problems (p = 0.02), ADHD, DSM-IV inattention (p = 0.02) and hyperactivity/impulsiveness (p = 0.01) (Tab. II). The DSM-IV ADHD combined parameter resulted in borderline statistical significance (p = 0.05) (Tab. II).

More specifically, children with psychosomatic problems decreased from 10% to 2%, with an increase of 12% in children with no ADHD (from 68% to 80% post-surgery). A similar increase was also seen for subscale DSM-IV inattention, with a decrease of 10% at 6 months. Lastly, no pathological result was found for subscale DSM-IV hyperactivity/impulsiveness in post-surgery (8% of pathological children at the beginning vs. 0% in post-surgery).

Table III shows the results obtained with the PSI questionnaire on parental stress, in this case related to mothers. The area with p < 0.01 was on total stress, ranging from 62% of parents to 78% in post-surgery.

Significant improvement was also seen in the parental distress subscale (p = 0.1), with a 16% increase in mothers with normal values post-surgery compared to baseline (from 58% to 74%) (Tab. III).

Lastly, there are no significant differences between pre- and post-surgery in the other subscales (Tab. III).

## Discussion

For a long time, the scientific literature has given importance to the link between sleep disorders and behaviour, focusing on the child's functional abilities in terms of attention, restlessness, memory and learning ability <sup>12,13,15,16</sup>. Parental stress related to child care for children suffering from SBD were not seen.

The present study confirmed the previous literature data regarding functional competence in children suffering from SBD improve significantly after adenotonsillectomy. The innovative part of our study is having observed the emotional behaviour by parents towards the problems of apnoeic patients together with children's executive functions. Breathing disorders result in parental stress and preoccupation, so much so that children negatively influence their sleep-wake rhythm, thus altering their parental competence. This parental dysfunctionality acts as environmental phenomenon which negatively influences already critical behavioural attitudes.

Behavioural improvements mainly focus on three areas: Oppositivity, Hyperactivity, Perfectionism, Restlessness/ impulsiveness, Emotional Instability and General Difficult Behaviour.

From the anamnesis of parents pre- and post-surgery, there was evidence that children had a net improvement in oppositive-provocatory behaviours, which in school age can lead to learning disorders.

Qualitative analysis of results is also very interesting, as parents report that their children are "easier to deal with in shopping centres" (Item 13 CRS), less "perfectionist" (Item 5 CRS), more "active" and less "tired" or "they move as in slow motion" (Item 73 CRS).

Table III. Pre-post PSI test confrontation.

	Pre		Post		P value*
	N	(%)	N	(%)	
Parental distress					
Average	29	(58)	37	(74)	0.01
Borderline	17	(34)	10	(20)	
Pathological	4	(8)	3	(6)	
Parent/ disfunctional child interaction					
Average	38	(76)	40	(80)	0.542
Borderline	10	(20)	9	(18)	
Pathological	2	(4)	1	(2)	
Difficult child					
Average	25	(50)	29	(58)	0.122
Borderline	16	(32)	15	(30)	
Pathological	9	(18)	6	(12)	
Total stress					
Average	31	(62)	39	(78)	0.004
Borderline	12	(24)	7	(14)	
Pathological	7	(14)	4	(8)	
Defensive response					
Average	28	(56)	32	(64)	0.132
Borderline	17	(34)	14	(28)	
Pathological	5	(10)	4	(8)	

\* Wilcoxon test for paired data

As for parental behaviour, the perception of their children improved, especially the idea that their child was "more agitated", and the fact that he/she had a tendency to cry more than other kids (Item 25 PSI) was dramatically decreased, and the same thing can be said with the feeling that their children woke up "in a bad mood" (Item 26 PSI), or that sleep and food rhythms were more difficult to manage (Item 31 PSI).

Food-wise, many parents referred that lack of appetite – a typical occurrence – changed into a suitable food behaviour.

For what concerns criticalities, we observed that non-Italian parents had problems filling-in the questionnaire; furthermore, during the first test, the lack of awareness of children's behaviours changed during post-surgery test, which led to a clearer precision in filling-in the questionnaire.

An advantage was that evaluations were not "operatormediated", thus avoiding a possible influence in the test result.

## Conclusions

The primary goal of the present study was to compare breathing difficulties resulting from OSAS to possible cognitive-behavioural problems of the child linking them to their parents' emotional-relational aspect.

The results obtained demonstrated that resolving OSAS brings about important improvements in the competence and behavioural attitude of the patient, in addition to relational and management difficulties by parents.

The identification of such indicators could represent a support to surgical programming, even in non-severe SBD.

Future research needs to identify standardised risk indicators that can provide further indications on surgical treatment in children up to 5 years of age.

It would be interesting to monitor whether behavioural and relational improvements are maintained in the family for periods longer than 6 months after surgery, and if the areas that did not undergo changes in the first 6 months improve after a longer time span.

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