

## PEDIATRIC SELECTIVE MUTISM AND SLEEP DISORDERS: A PILOT RESTROSPECTIVE CASE CONTROL-STUDY

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**ABSTRACT**

**Introduction:** Selective mutism (SM) is characterized by the persistent inability to speak in some specific contexts (i.e. school, home, social contexts) or within interaction with peer group and/or adults when a normal verbal linguistic competence may be expected, according to DSM5 criteria. The main sleep disorders in children with neurodevelopmental disorders are represented by difficulty in falling asleep at night (51%) and nocturnal awakenings (67%).

The main goal of the present study is verify the putative relationship between sleep disorders and selective mutism in children.

**Materials and methods:** 30 children were diagnosed with SM (13 males and 17 females) with mean age 9.47 (SD ± 1.29). The case-control group was composed by 30 typically developing children (TDC) (12 males and 18 females) (mean age 8.95; SD ± 2.01;  $p = 0.238$ ). All the subjects of both groups were recruited within the same urban area, Caucasian and homogeneous in socio-economic level. SDSC questionnaire was used in order to assess the sleep habits in both groups.

**Results:** The two groups (SM and TDC) were similar for age and gender. About the presence of reported sleep disorders, SM children show a significantly higher prevalence of reported sleep troubles than TDC subjects (Graph 1).

**Conclusion:** The present pilot retrospective case control-study has shown the presence of sleep disorders in children with SM. This new comorbidity has not been previously reported in clinical literature.

**Keywords:** Selective mutism, sleep disorders, Sleep Disturbance Scale for Children.

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**Introduction**

Selective mutism (SM) is characterized by the persistent inability to speak in some specific contexts (i.e. school, home, social contexts) or within interaction with peer group and/or adults when a normal verbal linguistic competence may

be expected, according to DSM5 criteria. Scientific reports about SM are scarce and tend to assess a prevalence ranging from 0.03% to 1%<sup>(1)</sup>.

Being a selective problem, it presents itself in an articulated way in different contexts, for example children affected by the disorder can communicate with gestures, nodding or shaking

their heads, tugging or pushing the interlocutor or, in some cases, with brief monosyllabic expressions or sometimes using altered tones of voice.

This disorder is not explained by a generic communication disorder and usually identified at the school onset between 2 and 5 years, although the diagnosis and early therapeutic intervention is usually between 6 and 8 years<sup>(1)</sup>.

Actually, SM has been classified within the anxiety disorders closely to externalizing disorders with a multifactorial etiology including genetic; temperamental; and environmental factors: parenting styles, school environment.

In general, different causative areas models for SM have been proposed such as psychodynamic (SM as result of unresolved intrapsychic conflicts), behavioral (SM as effect of maladaptive reinforcements patterns), and systemic area, maladaptive family dynamics.

A significant part of the limited literature on SM has investigated the role of familiarity and genetic predisposition.

In particular, some studies<sup>(2-4)</sup> have shown that families of children with SM present significantly more than psychopathological problems in the area of social withdrawal, social anxiety, avoidance of personality disorder or behavioral traits indicative of reduced sociality (shyness, solitary activities, reticence), indirectly suggesting a possible basic genetic component.

More recently, a study was carried out on families with anxiety spectrum and social withdrawal problems by analyzing the genetic material and in particular the contactin-associated protein-like 2-gene (CNTNAP2), and one of the polymorphisms was associated with SM<sup>(5)</sup>. These data, as well as further confirming the adequacy of the inclusion of SM among anxiety disorders, again support the contiguity with social anxiety disorder<sup>(6,7)</sup>.

However several studies have shown a relationship between stress, social behavior, emotion and cognition<sup>(8,9)</sup>.

However, other studies have not detected this trait and this poor coherence supports the fact that this dimension probably does not constitute a basic etiological factor but in a subpopulation of children it can be part of the multiplicity of behavioral manifestations that are consequently taken into consideration in the course of the treatment.

Sleep disorders are extremely common in children with neurodevelopmental disorders. About 25% of preschool children typically

have sleep-related problems, while the prevalence of sleep disorders in children with neurological disorders can be up to 80%<sup>(10; 11)</sup>.

The aetiology of sleep disorders in neurodevelopmental disorders can include: intrinsic neurological pathology, behavioral disorders, psychiatric disorders, drug side effects and disordered sleep breathing.

Regardless of the aetiology, sleep disorders can have a significant impact on both the quality of life of patients and caregivers.

The main sleep disorders in children with neurodevelopmental disorders are represented by difficulty in falling asleep at night (51%) and nocturnal awakenings (67%).

The early identification and treatment of sleep-related disorders can significantly improve cognitive function or delay the progression of underlying diseases in some patients with neurodevelopmental disorders, especially given that cognitive disorders have already been traced back to pre-school age<sup>(12)</sup>.

The main goal of the present study is assessing the putative relationship between sleep disorders and selective mutism in children.

## Materials and methods

### *Ethic statement*

This was a pilot retrospective case control-study conducted to identify the clinical comorbidity of presence of sleep disorders among children affected by SM. Parents were not required to give informed consent to the study, because the analysis used anonymous data that were obtained after they agreed to evaluation for their children by written consent. The study was performed in accordance with the Declaration of Helsinki criteria.

### *Exclusion criteria*

We considered as exclusion criteria obesity and overweight, neurological diseases (i.e. epileptic syndromes, primary headaches, neuromuscular disorders), psychiatric disorders (i.e. autism spectrum disorders, ADHD, psychosis), cognitive disability (Intelligent Quotient <70), malocclusal problems, psychotropic drugs treatment<sup>(13-67)</sup>.

### *Population*

According with DSM-5 criteria, 30 children were diagnosed with SM (13 males and 17 females) with mean age 9.47 (SD ± 1.29). The case-control group was composed by 30 typ-

ically developing children (TDC) (12 males and 18 females) (mean age 8.95; SD  $\pm$  2.01;  $p = 0.238$ ). All the subjects of both groups were recruited within the same urban area, Caucasian and homogeneous in socio-economic level.

### **Sleep Disturbance Scale for Children (SDSC)**

To assess sleep habits, the mothers of all patients were given the Sleep Disturbance Scale for Children (SDSC) 9 a likert-like questionnaire consisting of 26 questions capable of identifying the presence of the most common sleep disorders in childhood. This test allows the identification of a global disturbance index (SDSC TOT) and six categories of sleep disorders: Sleep initiation and maintenance disorders (DIMS), sleep related breathing disorders (SBD), Arousal disorders (DA), Disorders of wake-sleep transition (DWST), disorders of excessive daytime sleepiness (DOES), nocturnal hyperhidrosis (SHY). In accordance with the scale validation criteria, scores respectively higher or equal to 71 for SDSC TOT, 17 for DIMS, 7 for SBD, 6 for DA, 14 for DWST, 13 for DOES, 7 for SHY were considered clinically relevant; for the individual items, on the other hand, frequency values  $\geq 3$ /week were considered significant<sup>(68)</sup>.

### **Statistic analysis**

T-Test and Chi-square analyses were performed when appropriated in order to compare the two groups (SM and TDC) for age, gender and SDSC scores.  $p$  values  $<0.05$  were considered as statistically significant. MedCalc online software was used for statistical analysis (<https://www.medcalc.org/calc/>).

### **Results**

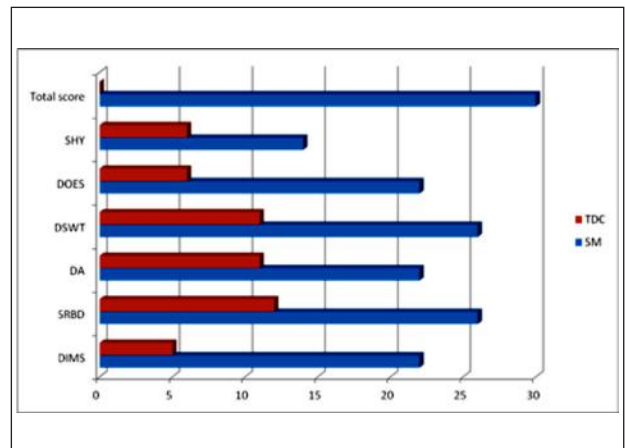
The two groups (SM and TDC) were similar for age ( $p = 0.238$ ) and gender ( $p = 1.00$ ). About the presence of reported sleep disorders, SM children show a significantly higher prevalence of reported sleep troubles than TDC subjects (Graph 1).

### **Discussion**

The present pilot retrospective case control-study has shown the presence of sleep disorders in children with SM. This new comorbidity has not been previously reported in clinical literature.

About the significant presence of sleep troubles in SM children, we can speculate that considering SM an anxiety disorder, it is easy to ex-

plain the sleep initiation and maintenance disorder (DIMS) since this finding is very frequent in children with internalizing disorders, while similar for the greater frequency of NREM sleep parasomnias.



**Graph 1:** Shows the percentage representation of sleep disorders between the two groups of children affected by selective mutism (SM) and among typical developing children (TDC), according to Sleep Disturbance Scale for Children (SDSC) reported by category: Difficulty in initiating and maintaining sleep (DIMS); Sleep breathing disorders (SBD); Disorders of arousal (DA); Sleep-wake transition disorders (SWTD); Disorders of excessive somnolence (DOES); Sleep hyperhidrosis (SHY); Total score.

More interesting, however, is the comorbidity with sleep related breathing disorders (SBD) which lends itself to some reflections. In fact, for a correct vocal expression it is necessary that the pneumo-phonetic apparatus may work perfectly in each functional aspects. SBD (snoring and sleep apnea) are frequent chronic diseases that can also alter bucco-facial anatomy and above all the neuromotor tone responsible for maintaining airway patency. The same neuromotor tone intervenes in the control of the passage of the air which is essential for a correct modulation of the tone of voice in the spoken common<sup>(69-71)</sup>.

In this light, it is also possible to speculate that even the respiratory disturbance may be the cause of SM considering the frequent report of a previous history of asthma, gastroesophageal reflux or laryngospasm in these children. These clinical conditions can stop in violent manner the voice emission. Since these children are fragile and emotionally sensitive, it could be a reason to decrease the frequency of vocalic emission until they are completely renounced. All these condition should be considered together, especially since a recent study reported a link between the disability condition of children with

obstructive sleep apnea syndrome and maternal stress that has a negative impact not only on psychological factors (self-esteem and locus of control) but also on memory performances of mothers<sup>(72)</sup>.

Finally, we cannot exclude in the pathogenesis of sleep disorders in SM children, the role of abnormal neurochemical pathways involving the orexin systems that may explain the vegetative alteration and hyperactivity<sup>(73-80)</sup>.

On the other hand, children affected by neurodevelopmental disorders must always be considered in its entirety while sleep disorders may be intended as a parental and familiar destabilizing factor and even recognition alone as a part of the whole could be a help and a strong motivation for the family to follow its directions.

In conclusion, the present study can be considered as innovative in the field of SM since nowadays there are no similar data reported.

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