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Sleep, Behavioral, and Cognitive Complaints in Children with Neurofibromatosis Type 1: A Public Health Challenge

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Abstract

Background: Neurofibromatosis type I (NF1) is a genetic and neurodevelopmental condition impacting the peripheral nervous system. Predominant clinical features include "café au lait" spots, neurofibromas, and cognitive challenges. While some clinical reports suggest a link between NF1 and alterations in sleep, there is a lack of studies exploring this connection specifically in children. This becomes crucial, given that sleep significantly influences a child's neurobehavioral, neurocognitive, and psychosocial development. **Objective:** To describe sleep, cognitive, and behavioral complaints in Brazilian children diagnosed with Neurofibromatosis type 1. **Methods:** Parents of children within the age range from 1 to 12 years old answered an online questionnaire composed of 24 questions related to sleep habits, sleep patterns, and cognitive and behavioral complaints. **Results:** Data was collected from 41 children, who were categorized between preschoolers (32%) and school-aged (68%). 63% of the entire sample reported sleep complaints. 66% of the children slept less than 10 hours a day, 26% of them being preschoolers and 74% school-aged. 85% of the children used electronics devices at night, and more than half (54%) of the sample had more than 2 hours of daily screen time. 66% of the children had a late bedtime. Easy crying (31%) was the most reported behavioral complaint in preschoolers, and anxiety (32%) was more prevalent in school-aged children. Alterations in attention and focus (44%) was the most frequent cognitive issue in the entire sample. **Conclusions:** This is the first study carried out in Brazil that assesses sleep, behavioral and cognitive complaints, and sleep patterns in children with NF1. It was reported that children with NF1 have significant sleep complaints and insufficient sleep duration. Additionally, behavioral, and cognitive complaints were widely reported. We reinforce the relevance of sleep assessment during routine clinical examination of patients with NF1.

Keywords: Neurofibromatosis 1, Child, Sleep, Cognition, Behavior.

Introduction

Neurofibromatosis type I (NF1) is the most common autosomal dominant genetic disease, with a prevalence of 1:3000 live births, exhibiting almost complete penetrance and highly variable phenotypic expressivity (Souza et al., 2009). The diagnosis is usually made in childhood and is established by the identification of two or more of the following clinical criteria: more than six "*cafe au lait"* spots, axillary or inguinal freckling, two or more cutaneous neurofibromas or one plexiform neurofibroma, two iris hamartomas (Lisch nodules), optic nerve gliomas, skeletal dysplasia's, and history of a primary relative with the disease (Rodrigues et al., 2014). Since cognitive and behavioral issues are among the most frequent manifestations of the disease, NF1 has been recognized as a neurodevelopmental disorder. It is indicated that children with neurodevelopmental disorders have a high risk of developing sleep problems (Robinson-Shelton & Malow, 2016). Some studies showed a higher prevalence of sleep disorders in children with NF1 (Johnson et al., 2005), while others found a similar incidence in the general pediatric population (Pérez et al., 2015).

Previous research has shown that insufficient and inadequate sleep in children is linked to a series of adverse health outcomes, such as daytime fatigue (Lehmkuhl et al., 2008), delayed neuro psychomotor development (Smaldone et al., 2007), reduced cognitive performance (Gruber et al., 2014), compromised immune system (Smaldone et al., 2007), and obesity (Fatima et al., 2015). Additionally, persistent sleep problems in childhood can reduce the wellbeing of the family and increase the risk of development of cognitive and behavioral difficulties in adolescence and adulthood, such as anxiety disorders, depression, aggressive behavior, and attention struggles (Gregory et al., 2008). However, pediatric medical anamnesis usually does not include assessments on sleep patterns and habits (Carvalheiro et al., 2017). Diagnosis and treatment of sleep problems are essential to ensure healthy development. In Brazil, to the best of our knowledge, there are still no investigations on the prevalence of sleep problems in children with NF1. Considering the significant impact these problems can have on the lives of these children and their families, this study aims to describe sleep, cognitive and behavioral complaints in Brazilian children from 01 to 12 years old with NF1.

Methods

Participants

From April to May of 2019, we recruited parents of children with an NF1 diagnosis through a cross-sectional study was conducted with volunteers of the "Associação Mineira de Apoio às Pessoas com Neurofibromatose" website and support groups of the Neurofibromatosis Reference Center, Brazil. The inclusion criteria were (1) being the parent of a child from 01 to 12 years of age diagnosed with NF1 and (2) agreeing to participate in the study. The exclusion criteria were not agreeing to participate in the study and not finishing the complete questionnaire.

Procedure

Through an online questionnaire, we informed the participants about the research objectives and obtained their informed consent. The instrument was answered anonymously at home, with a duration of approximately 40 minutes. This study obtained approval by the Research Ethics Committee of the Federal University of Minas Gerais under the registration ETIC 570/07.

Instrument

There are no validated questionnaires in Brazil related to this subject in this population. Therefore, we developed an online questionnaire for parents that aimed to assess sleep

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complaints, patterns, and habits of children with a diagnosis of NF1. The instrument had 24 questions that collected information concerning the daily life of the family and the child, sleep habits and sleep patterns, and complaints related to the child's sleep, behavior, and cognitive functioning.

Statistical Analysis

We performed statistical analysis using IBM SPSS (Statistical Package for the Social Sciences) on its 26.0 version. Categorical variables were described as frequencies, and continuous variables were described as mean. The data were submitted to an exploratory and inferential descriptive analysis, with a significance level of 5%. Continuous variables were tested using the t-Student test. Categorical data were tested with Pearson's chi-square association test and Fisher's exact test.

Results

Our final sample included parents of 41 children. Of this sample, 28 (68%) children were within the age range from 06 to 12 years old, and 13 (32%) were within 01 to 05 years of age.

Table 1 presents data related to sleep patterns by age range. Overall, the children have a late bedtime (mainly children aged 01 to 05 years old - p=0.043*) and a sleep duration shorter than 10 hours per night, with children aged 01 to 05 years sleeping 40 min more on average (p=0.042).

Table 1.

Sleep patterns of participants by age range

	All (n=41)	1-5 years (n= 13)	6-12 years (n= 28)	Z	P-value
Sleep patterns					
Average bedtime	22:21pm ±1h21[a]	22:25pm ±1h33[a]	21:49pm ±1h17[a]	2,09	0,043*(1)
Bedtime earlier than 10 p.m. n (%)	14 (34%)	3 (23%)	11 (39%)	0,44	0,480(2)
Bedtime at 10 p.m. or later n (%)	27 (66%)	10 (77%)	17 (61%)		
Average sleep duration	09:45 ±1:38[a]	10:11 ±1:27 [a]	09:33±1:42[a]	2,10	0,042*(1)
Sleep duration shorter than 10h n (%)	27 (66%)	7 (54%)	20 (71%)	0,56	0,300(3)
10h or more of sleep duration n (%)	14 (34%)	6 (46%)	8 (29%)		

Note. [a]mean ±SD. *Significant at the level 5%; (1) t-student test (2)fisher's test (3)Chi-squared test

Table 2 presents data related to sleep disturbances by age range. Most children from the age range from 06 to 12 years old presented sleep complaints. There was a statistically significant difference for the complaint of parasomnia for children aged 1 to 5 years (p=0,008).

Table 2.

Sleep disturbances of participants by age range

	All (n=41)	1-5 years (n= 13)	6-12 years (n= 28)	X^2	P-value
Resistance to initiate sleep				0,98	0,310(3)
Child falls asleep alone in own bed n (%)	19(46%)	8 (61%)	11(39%)		
Child needs parent in the room to fall asleep n (%)	22(54%)	5 (39%)	17(61%)		
Resistance Frequency				8,71	0,030* (2)
Never resists bedtime n (%)	3(7%)	2(15%)	1(4%)		
Rarely resists bedtime n (%)	10(24%)	1(8%)	9(32%)		
Sometimes resists bedtime n (%)	15(37%)	8(66%)	7(25%)		
Always resists bedtime n (%)	13(32%)	2(15%)	11(39%)		
Sleep fragmentation				0,00	1,000(2)
< 1h awake n(%)	37(90%)	12(92%)	25(89%)		
≥ 1h awake n(%)	4(10%)	1(8%)	3(11%)		
Sleep complaints					
Refers sleep complaints n(%)	26(63%)	9(69%)	17(61%)	0,03	0,860(2)
Complaints related to insomnia n(%)	13(32%)	4(31%)	9(32%)	0,00	1,000(3)
Complaints related to parasomnia	6(15%)	5(38%)	1(8%)	6,08	0,008*(2)
Unspecified sleep complaint n(%)	7(17%)	4(31%)	3(11%)	0,00	1,000(2)

Note. *Significant at the level 5%; (1) t-student test (2)fisher's test (3)Chi-squared test

Table 3 presents the results of the sleep habits assessed. Most parents referred that the children had an adequate sleep environment based on the variables of temperature, lighting, noise, and mattress quality. Regular physical activities were referred by 63% of the sample. Most children (85%) used some kind of electronic device, mostly at nighttime. There were no statistically significant results when comparing groups.

Table 3.

Sleep habits of participants by age range

	All (n=41)	1-5 years (n= 13)	6-12 years (n= 28)	X^2	P-value
Sleep environment quality					
Good bedroom temperature n(%)	41(100%)	13(100%)	28(100%)		-
No bedroom noise n(%)	41(100%)	13(100%)	28(100%)		-
Good bedroom lighting n(%)	41(100%)	13(100%)	28(100%)		-
Good mattress quality n(%)	40(98%)	13(100%)	27(96)		-
Use of electronic devices					
At night n(%)	35(85%)	11(85%)	24(86%)	0,00	1,000(2)
Up to 2h of daily screen time n(%)	19(46%)	4(31%)	15(54%)	1,05	0,200(3)
Over 2h of daily screen time n(%)	22(54%)	9(69%)	13(46%)	1,05	0,200(3)

Note. *Significant at the level 5%; (1) t-student test (2)fisher's test (3)Chi-squared test

Table 4 presents cognitive and behavioral complaints by age range. Overall, anxiety was the most referred behavioral complaint, and disturbances in attention and focus was the most frequent cognitive issue, mainly for children aged 06 to 12 years, but there was no statistically significant difference.

Table 4.

Behavioral and cognitive complaints of participants by age range

	All (n=41)	1-5 years (n= 13)	6-12 years (n= 28)	X^2	P-value
Behavioral complaints				8,16	0,200(2)
Anxiety n(%)	10(24%)	1(8%)	9(32%)		
Easy crying n(%)	7(17%)	4(31%)	3(11%)		
Bad mood n(%)	7(17%)	1(8%)	6(21%)		
Aggressiveness n(%)	4(10%)	1(8%)	3(11%)		
Low sociability n(%)	3(7%)	1(8%)	2(7%)		
Sadness n(%)	1(2%)	0(0%)	1(4%)		
Other n(%)	9(22%)	5(38%)	4(14%)		
Cognitive complaints				11,815	0,014 (2)
Attention and focus n(%)	18(44%)	4(31%)	14(50%)		
Academic problems n(%)	7(17%)	0(0)	7(25%)		
Learning n(%)	5(7%)	1(8%)	4(14%)		
Memory n(%)	1(2%)	0(0%)	1(4%)		
Other n(%)	12(29%)	8(62%)	4(14%)		

Note. *Significant at the level 5%; (1) t-student test (2)fisher's test (3)Chi-squared test

Discussion

To the best of our knowledge, this is the first study carried out in Brazil that assesses sleep complaints and sleep patterns in children with NF1. In Brazil, there are no organizations in different regions, apart from the Minas Gerais Association for Support to People with Neurofibromatosis (Associação Mineira de Apoio às Pessoas com Neurofibromatose), for adequate support for this disorder focused on children and adolescents. In this direction, we carried out this survey to learn more about sleep complaints and sleep patterns in children with NF1 in Brazil.

The exact age of each child was not collected, we opted to categorize them into two groups: preschoolers (01- 05 years) and school-aged (06-12 years). We took into account that during these two periods children share similar sleep and environmental variations that influence sleep patterns and habits, such as development of autonomy, changes in parental control and academic demands (Carvalheiro et al., 2017; Belísio et al., 2010).

Our results showed that most of the children in the sample presented late sleep onset, prolonged night awakenings, bedtime resistance, and short sleep

duration. The International Classification of Sleep Disorders (ICSD-3, 2014) considers these complaints as symptoms of pediatric insomnia. Thus, the complaint of pediatric insomnia in our sample follows the rate found in the typical population (Mindell & Moore, 2014). Our findings also corroborate a study (Licis et al., 2013) that reported alterations in sleep onset and maintenance in American children with NF1.

Compared to a recent population-based study on the sleep quality of Brazilian children, which found a 25.5% prevalence of sleep disorders, we found a 63% frequency of sleep complaints among Brazilian children with NF1 (Almeida & Nunes, 2019). In a similar direction, an American study found a higher frequency of sleep problems among children with NF1 compared to their unaffected siblings, with significantly higher scores on the sleep disturbance scale for children questionnaire (Licis et al., 2013).

Using a different approach, an English study found no differences in the frequency of sleep disturbances between children with NF1 and the general population (except for parasomnias, especially sleepwalking and night terrors), but in a subgroup analysis it reported that changes in behavior (sleeping problems) conduct, emotional, or hyperactivity) were significantly higher among children with NF1 and frequent sleep problems compared to those with NF1 without frequent sleep problems (Johnson et al., 2005). This finding raises the question of the possible common neurobiological origin between behavioral and sleep symptoms in neurodevelopmental disorders, (Almondes, 2018) which needs further investigation. Another study in a Spanish hospital with 95 children with NF1 also did not find a higher frequency of sleep problems, compared to control, but in those with NF1 and developmental disorders (ADHD or mental retardation) there were higher frequencies of sleep problems, although statistically non-significant differences (Maraña-Perez et al., 2015). It is observed with these data that, although sleep disorders are often associated with NF1, the literature is not uniform in establishing a correlation. At least in part, such differences may be due to the non-use of standardized methods to assess sleep habits and problems in these studies.

We identified that 66% of the children in the entire sample have bedtime at 10 pm or later. This group is composed by 77% of all preschool children and 61% of all school- age children. The average bedtime of the entire sample was 22:21pm. However, the most worrying data was that children aged 01 to 05 years (preschoolers) slept even later compared to the school group. Literature data shows that late bedtime in children is associated with higher sleep latency and less total sleep duration (Smaldone et al., 2007; McLaughlin Crabtree et al., 2005; Mindell et al., 2009). It has also show that a bedtime after 10 pm is associated with childhood obesity (Olds et al., 2011).

Concerning sleep duration, we found that more than half of our total sample (66%) slept less than 10 hours a day, 26% being preschoolers and 74% school-aged. It is recommended that preschool children should sleep 10 to 14 hours daily, including naps (Hirshkowitz et al., 2015). School-aged children usually do not nap, but it is recommended that they should sleep 9 to 12 hours daily (Hirshkowitz et al., 2015). Getting an adequate amount of sleep is essential for children's healthy growth and development (Gruber et al., 2014). This discussion is supported by the statistically significant data found that children aged 01 to 05 years sleeping 40 min more on average of the sleep duration. That is, the data seems to suggest that the children needed to sleep more, because they slept later.

We identified that most of the children (54%) in the study needed a parent in the room to fall asleep, being this group mostly composed by school-age children (77%). Furthermore, 37% of the children in

the sample sometimes resist going to bed and 32% always resist. It is suggested that parental presence during the child's bedtime can influence bedtime resistance (Tikotzky & Shaashua, 2012), nocturnal awakenings and make it difficult to fall asleep independently since the child can associate the sleep onset with the presence of the parents (Sadeh et al., 2010). Bedtime resistance is also associated with poor sleep hygiene practices (Wilson et al., 2015).

Sleep hygiene consists of a series of behavioral and environmental habits aimed to promote sleep quality and is an essential component of the treatment of sleep disorders (Irish et al., 2015). To assess the quality of children's sleep hygiene, we inquired the parents about some sleep habits, such as quality of the physical sleep environment, practice of physical exercise, and use of electronic devices (Galland & Mitchell, 2011).

Our results showed that all the children had a good sleep environment, with satisfactory bed and levels of temperature, lighting, and noise. If inadequate, these elements could difficult sleep falling asleep and cause night waking, reducing total sleep duration (Bagley et al., 2015). Most children in our sample (63%) practiced physical exercise regularly. Literature data show a direct link between physical exercise and sleep quality, as it helps to regulate the homeostatic mechanisms and circadian rhythms of the body (Hosker et al., 2019). Thus, the practice of physical exercise is essential for children's cognitive and motor development and relates to a higher level of wellness (Bidzan-Bluma, & Lipowska, 2018). These were the two elements indicative of good sleep hygiene in the sample.

On the other hand, we identified a high nighttime use of electronic devices by the children (85%). And more than half of the children (54%) had more than 2 hours of daily screen time. Nighttime use of electronic devices can lead to psychological and physiological arousal and is consequently associated with bedtime resistance, late bedtime, anxiety, as well as alterations in circadian rhythm functioning, since nighttime exposure to the bright blue-enriched lights emitted by LED electronic device screens can suppress melatonin production and secretion (Aishworiya et al, 2018; Hale & Guan, 2015; West et al., 2011). It is recommended that children under 2 years old do not use electronic devices, and children over 2 years old do not exceed 2 hours of screen time per day. Literature data suggest excessive screen time can be a risk factor for childhood obesity and that even regular physical exercise does not compensate the potential harmful effects. Furthermore, excessive screen time is associated with cognitive and behavioral disturbances, such as language delay, academic problems, anxiety, and aggressive behavior (Domingues-Montanari, 2017). It is important to note that the children in our sample showed a late bedtime pattern, which can be explained in part by these high nighttime use of electronic devices data.

Behavioral and cognitive complaints were widely reported in the sample. Concerning behavioral complaints, easy crying (31%) was the most reported in preschoolers, while anxiety (32%) was more prevalent in school-aged children. Alterations in attention and focus (44%) were the most frequent cognitive issue, reported majorly in school-aged children (50%). A systematic review (Schwetye, & Gutmann, 2014) indicates that almost 80% of children with NF1 show behavioral and cognitive complaints. It is described that children with NF1 have a greater rate of behavioral problems, such as anxiety and depression, in comparison with the typical population. Attention and concentration struggles were more reported in children with NF1 than in their healthy siblings. It is noteworthy that sleep disturbances are predictor factors for cognitive and behavioral issues (Gregory et al., 2008).

As with most studies with children, we based our study on the parents' report,

and without objective measurements of sleep, such as actigraphy, our data is subject to recall bias. In addition, there was no survey on the frequency of sleep complaints during a specific period. Since we did not evaluate differences between weekdays and weekend sleep patterns, we could not characterize whether the children had a consistent bedtime routine.

As with all research on the Internet, the cohort in this study is inclined towards higher education and these results may not be representative of all families within Brazil and other countries. We did not assess the parents' educational level, so we expect that this sample mainly represent urban-based populations with Internet access. Future studies should evaluate populations with restricted internet access, as well as those in other socioeconomic contexts. Additionally, these studies should include additional measures and aim to deepen the comprehension of the association between sleep disorders, behavioral issues, and cognitive difficulties in Brazilian children with NF1 by comparing them with a control group.

Application of instruments to measure the investigated alterations, such as questionnaires and specific scales for the assessment of sleep disorders, may also provide a more precise description of the sleep complaints in children with NF1.

Conclusions

This study brings a significant contribution to the national scientific outlook, since investigating the sleep characteristics of children with NF1 is fundamental for the development of preventive and interventional measures. It was possible to verify that sleep complaints in our sample were frequent and that the sleep duration of these children was insufficient. Considering that some good sleep hygiene elements were found in the sample, the data may indicate that the reported sleep complaints are related both to the parents' attitudes and to NF1. Therefore, we reinforce the relevance of giving special attention to sleep assessment during clinical examination of patients with NF1, as well as performing appropriate interventions to improve the quality of life of these patients.

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